Exploring Language Production, Comprehension and Naming in Adolescents with Fetal Alcohol Spectrum Disorders

By

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COMPULSORY DECLARATION

This work has not been previously submitted in whole, or in part, for the award of any degree. It is my own work. Each significant contribution to, and quotation in, this dissertation from the work, or works, of other people has been attributed, and has been cited and referenced.

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I, Claire Corbett, of 4 Klein Croonhoop, Molenvliet Rd, Observatory, do hereby declare that I empower the University of Cape Town to produce for the purpose of research either the whole or any portion of the contents of my dissertation in any manner whatsoever.

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LIST OF ABBREVIATIONS

ARBD:	Alcohol-related Birth Defect
ARND:	Alcohol-related Neurodevelopmental Deficit
BNT:	Boston Naming Test
CANTAB:	Cambridge Neuropsychological Test Automated Battery
CELF-R:	Clinical Evaluation of Language Fundamentals Revised
CIFASD-II:	Collaborative Initiative of Fetal Alcohol Spectrum Disorders – Second wave
CNS:	Central Nervous System
D-KEFS:	Delis Kaplan Executive Function System
DV:	Dependent Variable
FASD:	Fetal Alcohol Spectrum Disorders[1]
FAS:	Fetal Alcohol Syndrome
FSIQ:	Full Scale IQ
IQ:	Intelligence Quotient
IOM:	Institute Of Medicine
IV:	Independent Variable
NIAAA:	National Institute on Alcohol Abuse and Alcoholism
TOLD:	Test of Language Development
TROG-2:	Test of Reception of Grammar – Version 2
UCT:	University of Cape Town
USA:	United States of America
WISC-IV:	Wechler Intelligence Scale for Children- Fourth Edition
WISC-R:	Weschler Intelligence Scale for Children- Revised

[1] This is accepted English spelling used by the British Medical Association and British Medical Journal

ABSTRACT

Language impairments in young children with prenatal exposure to alcohol are well described. Cognitive abilities are known to change over the course of development in children with fetal alcohol spectrum disorders (FASD) but the development of language abilities of adolescents with FASD are less well determined. The aims of this study were i) to evaluate the domains of language production, comprehension and naming in children with moderate or heavy prenatal exposure to alcohol, and ii) to determine if these domains are a specific weakness in these children. The study compared 25 children with FASD to 25 typically developing non-alcohol exposed controls on four language measures; (1) the Clinical Evaluation of Language Fundamentals Revised; (2) the Test of Reception of Grammar- Version II; (3) the Cookie Jar Theft Picture test and; (4) the Boston Naming test. The FASD group had significantly lower IQ scores than the control group and intellectual functioning was covaried for using the WISC-IV Perceptual Reasoning IQ. Consistent with the language impairments demonstrated in younger children, the FASD group showed significant difficulties on all language domains. Impairments in language comprehension and naming were accounted for by perceptual reasoning IQ. Independent of non-verbal intelligence, language production in children with FASD was significantly weaker than in typically developing control children in this sample.

Keywords: Language; Adolescents; Fetal Alcohol Spectrum Disorders; Prenatal Alcohol Exposure; Intelligence Quotient; South Africa

CHAPTER 1
INTRODUCTION

1.1 Background to the thesis

Fetal Alcohol Syndrome (FAS) is known to be associated with numerous cognitive and behavioural deficits (Mattson & Riley, 1998). Despite decades of research there is still no firm conclusion as to whether these deficits represent global dysfunction or a unique neurobehavioural phenotype (Steinhausen, Willms, Metzke, Spohr, 2003).

Fetal Alcohol Spectrum Disorder[2] (FASD) is an umbrella term relating to the continuum of deficits caused by prenatal alcohol exposure (O'Leary, 2004). It is understood that as children with FASD develop their cognitive abilities may change. Assessments throughout the child's development are necessary to redefine our understanding of their strength and weaknesses.

Language is almost uniformly reported as deficient in children who are prenatally exposed to alcohol. In young children, difficulties in both comprehension and production of language, as well as naming have been reported. Not all studies have accounted for the association between language and intellectual dysfunction in these children. Of those studies that have, two concluded that deficits in language were accounted for by intellectual deficits and thus did not represent a specific strength or weakness in children prenatally exposed to alcohol (Becker, Warr-Leeper & Leeper, 1990; McGee, Bjorkquist, Riley & Mattson, 2009). Another study concluded that the language abilities of children with FAS were more impacted than what could be explained by general intellectual functioning (Hamilton, 1981).

Most language studies in children with prenatal alcohol exposure have examined young children between the ages of two and twelve years. One study extended the age range into the early twenties, but no explanation of the language ability of the older participants in comparison to the younger children was given (Iosub, Fuchs & Gromisch, 1981).

The language profile of adolescents with FASD warrants further exploration as it may be different to that of the younger children. Some changes in the language abilities in older children with FASD are reported in the literature. Two studies found changes in the language abilities of older children. Carney and Chermak (1991) found older children to have greater semantic difficulties in language comprehension and production while Coggins, Timler and

[2] The use of FASD in this study refers to all children diagnosed within the spectrum of disorders, whereas FAS refers to those with the primary diagnosis of Fetal Alcohol Syndrome. FASD and prenatal alcohol exposure have been used interchangeably unless otherwise indicated.

Olswang (2007) found greater difficulty in narrative production in older children. If changes in language ability in adolescents were known, early intervention and prevention programmes could be put in place.

1.2 Thesis

This study aimed to contribute to the literature on the language abilities in older children with FASD. Language comprehension, production and naming abilities of adolescents were explored. It was expected that these language domains would be similarly impaired as in younger children. In line with the literature on younger children, it was expected that the language abilities of children with FASD would be accounted for by their intellectual functioning.

1.2.1 Specific Aims

The specific aim of this study was to describe and compare domains of language comprehension, production and naming between adolescents with FASD and typically developing non-exposed controls.

To determine the aspects of language that may be deficient in adolescents with FASD three hypotheses were investigated:

1. Children with FASD would have significantly lower intellectual functioning than non-alcohol exposed controls.
2. Language comprehension, production and word naming would be significantly worse in children with FASD than non-exposed controls.
3. In children with FASD, deficits in language comprehension, production and word naming would be accounted for by deficits in intellectual functioning.

1.3 Value of this study

This study aims to contribute to the literature on the language functioning of adolescents with FASD. Concurrently, the study contributes to the growing body of research examining whether a specific neurobehavioural profile exists in children following prenatal alcohol exposure.

The understanding of language function in children with FASD is important because language ability is imperative for social and academic functioning and impacts on cognitive and behavioural difficulties which are specific problem areas in FASD. Language impairment reduces the ability to process and understand rules, as well as resulting in a reduced ability to use internal language skills to guide and control behaviour (Ford, Farah, Shera & Hurt, 2007).

In children with FASD, a behavioural problem is one of the most noticeable and socially destructive outcomes. Social deficits in children with prenatal alcohol exposure may become more pronounced in later years as social interactions become more complex, requiring a combination of understanding of language pragmatics, facial expressions and body language (Whaley, O'Connor & Gunderson, 2001). These social and behavioural problems may lead to more serious outcomes such as unemployment and incarceration (Fast & Conroy, 2009; Stattin & Klackenberg-Larsson, 1993).

FASD is a major public health problem in South Africa (Rosenthal, Christianson & Cordero, 2005). Special education services have been characterised by a lack of special education facilities catering for low socio-economic communities resulting in the majority of children with special education needs, including children with FASD being enrolled in the main-stream government school system (Engelbrecht & Forlin, 1998). Despite the implementation of inclusive education in South Africa in 2001 (Department of Education, 2001), educators in government mainstream schools indicate a need for training and resources and departmental, professional and parental support (O'Connor & Geiger, 2009). Class sizes are large and the public education system does not cater adequately for a range of problems experienced by children or teachers. The education medium is mainly verbal-based and generally there are few government learning and behavioural intervention programmes available or implemented. An understanding of the language abilities of adolescents with FASD could inform intervention programmes and supportive school teaching methods for children with FASD. An improvement in the language abilities in the children may also affect the behavioural outcomes for the children, so improving their longer term social outcomes.

CHAPTER 2
BACKGROUND INFORMATION

2.1 Overview and diagnosis of Fetal Alcohol Spectrum Disorder

Alcohol is a neurobehavioural teratogen that has deleterious effects on the developing embryo and foetus (Streissguth, 1997). The syndrome of deficits associated with prenatal alcohol exposure, termed Fetal Alcohol Syndrome, was formally described in 1973 (Jones & Smith, 1973). The spectrum of disorders (FASD) includes the following deficits:

1. craniofacial abnormalities,
2. growth retardation, and
3. central nervous system dysfunction, such as structural brain abnormalities and intellectual disability.

Fetal Alcohol Syndrome is at the most severe end of the primary diagnoses within the spectrum of disorders. Children are diagnosed with FAS when all three categories of anomalies are present to a specified extent. Partial FAS includes individuals with confirmed alcohol exposure as well as some facial characteristics, growth and central nervous system (CNS) deficits or behavioural and cognitive characteristics. Many children display only some of the diagnostic features and do not meet the criteria for diagnosis of FAS or partial FAS. These children are classified with either Alcohol-related Neurodevelopmental Deficits (ARND) or Alcohol-related Birth Defects (ARBD). ARND refers to children with minimal dysmorphology yet having neurobehavioural deficits, whereas ARBD refers to children that have some of the congenital physical abnormalities.

As the brain is constantly developing throughout gestation, prenatal alcohol exposure can affect the brain development at any stage during the pregnancy. Exposure to varying quantities of alcohol at particular points during gestation may explain why FASD manifests as a spectrum of disorders (Benton Gibbard, Wass & Clarke, 2003).

The direct brain damage due to prenatal alcohol exposure results in primary disabilities such as reduction in brain weight, disruption of many areas of brain development such as the hippocampus, corpus callosum and basal ganglia (Sowell, Thompson, Leonard, Welcome, Kan & Toga, 2004), as well as cognitive deficits. These cognitive deficits are addressed in section 2.3. The secondary disabilities that result following prenatal alcohol exposure are the behavioural and psychiatric results of brain damage such as mental health problems, disrupted school experience and a greater likelihood of incarceration (Streissguth, 1997).

2.2 Epidemiology of FASD

Epidemiological studies conducted on the prevalence of FASD in Wellington, a South African Western Cape community, report amongst the highest prevalence rates of FASD in the world (May et al., 2000; May et al., 2007; Viljoen et al., 2005). The first study in Wellington by May et al. in 2000 reported a prevalence rate for Fetal Alcohol Syndrome of 40.5 - 46.4 per 1000. The 2005 epidemiological study by Viljoen et al. included partial FAS and reported a prevalence rate of 65.2 - 74.2 per 1000. The most current reported prevalence rates in Wellington are 68.0 – 89.2 per 1000 for FAS and partial FAS. This prevalence rate for FAS and partial FAS has a greater range and a much higher maximum than that for FAS alone, which is expected as partial FAS is on the milder range of the spectrum (May et al., 2007).

These rates can be contrasted to the average rates for the developed world of 0.97 per 1000 (Abel & Sokol, 1991) and the United States of America (USA) estimates of 0.33 - 2.2 per 1000 (May & Gossage, 2001). The prevalence of FAS in Italy has more recently been reported as being much higher, viz. rates of 3.7 to 7.4 per 1000 (May et al., 2006). It was suggested that the higher rates in Italy were found due to the community sample method[3] of recruitment, which was also used in South Africa. In Italy the percentage of children with FAS was low (0.74%) and partial FAS higher (3.13%) (May et al., 2006). In comparison, the percentage of children with FAS in South Africa was higher (6.7%) with not as many partial FAS (2.2%) (May et al., 2007). This shows that the South African children tend to fall on the more severe end of the spectrum.

FAS is recognised as the leading cause of mental retardation in the USA (Abel & Sokol, 1986). FASD is one of the most common causes of learning disability in South Africa and, as such, is significant as an important and completely preventable cause of mental disability (Molteno & Lachman, 1996).

[3] In these community sample studies all first grade students from a large number of participating schools in the study site undergo dysmorphology screening. This method can be contrasted to other record based studies that investigate FAS prevalence amoung clients presenting at clinics.

A combination of factors is understood to partially account for the high prevalence rates in South Africa, specifically in at-risk coloured[4] communities (Viljoen et al., 2005). For example poor current and lifelong nutrition and small maternal body size of the mother may result in far greater impact of alcohol on the foetus. The social and economic despair of women, as well as cultural drinking norms and illegal bars, in combination with the legacy of the "dop[5]" system, and individual drinking patterns result in heavy episodic drinking that may increase the likelihood of FAS (May et al., 2007).

FASD has disastrous individual and social consequences (Mattson & Riley, 1998). This is particularly relevant in South Africa, which has very high FASD prevalence rates and a lack of adequate basic social infrastructure, such as education and health (Patel & Kleinman, 2003). The individual and social consequences are manifold. However, as FASD is a preventable condition, research is needed to better understand the effects in order to inform preventative measures and intervention.

2.3 Neurocognitive deficits in FASD

A major impact of FASD is on the central nervous system (CNS), including cognition and behaviour. A broad spectrum of neurobehavioural deficits is associated with FASD. The noticeable impact of alcohol exposure on cognitive deficits has been the focus of considerable research for three decades. It is still unclear as to whether there exists a unique pattern of cognitive behavioural functioning associated with FASD that would account for a behavioural phenotype, or alternatively, whether identifiable patterns of cognitive and behavioural functioning related to differential alcohol exposure, genetic and environmental factors may emerge. The majority of research supports the understanding that a generalised deficit in processing *complex* information is the essence of the cognitive-behavioural phenotype associated with FASD (Kodituwakku, 2007). As the cognitive and behavioural profile of children with FASD may also change over time, repeated neuropsychological assessments are required to refine an understanding of a child's strength and weaknesses throughout development to understand individual abilities and difficulties as well as inform

[4] Coloured refers to an ethnic group of mixed-race people in South Africa. Coloured people possess a mixed ancestry that includes sub-Saharan, European, Malaysian and Indonesian influences (Posel, 2001).

[5] The dop system refers to the historical practice of wine distribution as partial payment for labour by farmers. This practice has been outlawed for over four decades (May et al., 2007).

intervention programmes (Benton Gibbard et al., 2003).

2.3.1 Intellectual functioning in FASD

Children with prenatal alcohol exposure are well known to have deficits in intellectual functioning. The average IQ of children with FAS typically falls close to 70 (borderline range) but can range from mildly retarded (55-69) to borderline intellectual impairment (70-79) (Benton Gibbard et al., 2003; Becker et al., 1990; Carney & Chermak, 1991; Mattson & Riley, 1998, Shaywitz, Caparulo & Hodgson, 1981). The intellectual abilities of children with FASD correlate with the severity of the primary diagnosis within the FASD spectrum. Children with more severe dysmorphology were found to have lower intellectual functioning (Iosub, Fuchs, Bingol & Gromisch, 1981, Streissguth, 1997).

A longitudinal study by Streissguth (1976) compared the intellectual functioning of children with FAS to matched controls over seven years. For every child with FAS two control children were matched on maternal age, parity, education, sex, race, marital status, geographical region of delivery and occupation of the head of household. No statistically significant differences in IQ were reported at age four. But by seven years of age there were highly significant differences in IQ. The FAS group at age seven obtained a mean average IQ significantly lower than their IQ at age four. This decrease in performance may not be due to a deteriorative process but rather that children with FAS failed to make the age-appropriate gains expected, based on normative performances (Benton Gibbard et al., 2003). These results also support the understanding that children are seen to "grow into their deficits" (Anderson, Northam, Hendy & Wrennal, 2001).

Verbal IQ has been reported to be lower than performance IQ in children with FASD (Conry, 1990). However, a review by Mattson and Riley (1998) reported equivocal results for performance and verbal IQ. Therefore, whether a difference in performance and verbal IQ is present in FASD remains to be determined.

2.3.2 Other neurocognitive deficits in FASD

Attentional deficits are a pronounced characteristic of FASD. Children with prenatal alcohol exposure show similar deficits to those with attention deficit disorder (Nanson & Hiscock, 1990). Children with FASD have difficulties investing, organising and maintaining attention. These attentional difficulties impact upon their behaviour and they are often described as hyperactive and irritable (Mattson & Riley, 1998). Deficits in both visual and verbal memory and learning are associated with prenatal alcohol exposure. However, free recall and explicit memory are more impaired than recognition and implicit memory, which is consistent with the understanding that the processing of more complex information is deficient in FASD populations (Kodituwakku, 2007). Children with FASD also exhibit motor deficits in speed, precision and grip strength (Mattson, Riley, Gramling, Delis & Jones, 1998).

CHAPTER 3
LANGUAGE AND PRENATAL ALCOHOL EXPOSURE

3.1 Overview of language research in children prenatally exposed to alcohol

The first reported language development delays in children with prenatal alcohol exposure were noted by the research group headed by Lemoine in 1968 (Becker et al., 1990). The first large study on language abilities, specifically in comprehension and naming, in children with prenatal alcohol exposure was conducted by Iosub et al. (1981). Of 63 participants diagnosed with FAS, between 1 and 23 years of age, 80% of the participants were found to have language difficulties as measured on the Peabody Picture Vocabulary Test, Test of Auditory Comprehension of Language, the Token Test and the Illinois Test of Psycholinguistic Abilities. Unfortunately differences between young and older participants were not investigated, nor the contribution of intelligence to the language difficulties.

Few studies on language abilities in children with prenatal alcohol exposure have been conducted. Of these studies the results have been fairly consistent in reporting difficulties in childhood language comprehension and production. However as previously mentioned in section 1.1 the majority of research on language disabilities has been conducted on young children (ages 2 to 12 years),while the language functioning of adolescents with prenatal alcohol exposure has been under-reported. Only one published study on language abilities in children with FASD, which included adolescents, was found.

This chapter reviews the literature regarding the language abilities of children prenatally exposed to alcohol. In this review, the terms “language comprehension” and “receptive language” are used interchangeably to describe the understanding of the communication of others, be it spoken or written. Similarly the terms “language production” and “expressive language” are used interchangeably to describe communication through writing or speaking. Language comprehension and language production are typically used in the neuropsychological literature, whereas expressive and receptive language are used in speech therapy.

3.2 Evidence for language preservation in FASD

A study of 359 children at 1, 2 and 3 years of age reported intact expressive and receptive language for children who were prenatally exposed to low levels of alcohol (Greene, Ernhart, Martier, Sokol, and Ager, 1990). Receptive and expressive scales of the Sequenced Inventory of Communications Development and a 10 minute speech sample (as assessed by mean length utterance) at 2 years showed no significant language difficulties. However, Greene et al. (1990) concluded that if there was an effect of prenatal alcohol exposure (without FAS) on language functioning, the effect was exceptionally small. The quality of the care-giving environment was shown to be the primary determinant of language ability. The children in this study were exposed to very low levels of alcohol, namely 0.047 AA/day, the equivalent of 3 drinks a month.

3.3 Evidence for language impairment in FASD

Numerous studies have demonstrated difficulties in language production and comprehension following prenatal alcohol exposure. Various tests on a range of ages, mostly in younger children, have been used to assess language domains. Two of the studies reported the language impairments to be consistent with IQ, whereas others have found language impairments over and above that which can be accounted for by intellectual dysfunction (Hamilton, 1981; Becker et al., 1990; McGee, Bjorkquist, Riley & Mattson, 2009). Many of the studies did not account for intellectual functioning.

3.3.1 Language in children prenatally exposed to alcohol, but not diagnosed with FASD

The following studies reported overall language impairments of participants who had not been formally diagnosed within the spectrum of disorders. One study included two single cases of pre-school-aged children with confirmed high levels of prenatal alcohol exposure and facial and height anomalies, consistent with FAS, reported expressive and receptive language difficulties as measured by the Sequenced Inventory of Communication Development (Shaywitz et al., 1981).

- The first child aged 5 years 11 months had normal intellectual functioning with an IQ of 120, as measured by the non-verbally based Leiter International Scales of Performance. His language comprehension of single words and two stage commands was intact, but his overall receptive language score was at a much younger age level of 3 years 2 months. His overall expressive language score and spontaneous language sample was at a 2-year-old level and his utterances lacked fluency and intonation.

- The second child, aged 5 years and 4 months, was reported to be functioning intellectually at a normal level, for his age, as measured by the Weschler Preschool and Primary Scale of Intelligence block design subtest. This child's receptive language performance was similar to the child in the first case study with an overall receptive score at a 3 year 3 month age level. This child was unable to carry out 2-stage commands, and had significantly impaired syntactic development. This child's expressive language ability was also impaired with a score at a 2 year 5 month level and similarly showed severe syntactic deficits but better spontaneous language production.

Both children had language comprehension and production difficulties inconsistent with their normal level performance on non-verbal intellectual functioning tasks.

A larger study of 573 children with prenatal alcohol exposure reported difficulties in overall language functioning (Coggins et al., 2007). The children ranged between 6 years and 12 years 11 months of age. Of these children 63 were diagnosed with FAS and partial FAS; the remainder had confirmed prenatal alcohol exposure with static encephalopathy, neurobehavioural disorder or no central nervous system dysfunction.

The primary aim of this study was to examine a relationship between communicative performance and environmental risk. A range of standard language tests was used over a ten year period: the Clinical Evaluation of Language Fundamentals, Auditory Comprehension of Language as well as tests of language development, language competence and of word knowledge. The picture book *Bus Story* and *Frog*, *Where are you?*, were used to generate narrative discourses (for the children between 6 and 8-years-old and 8 to 12 years 11 months respectively). The Frog story narrative was deemed adequate if 8 of the 24 pictures were communicated unambiguously.

The overall language performance of the children, as measured by the standardised language tests, placed 31% in the mildly impaired range and 37% of the sample in the moderately-to-severely impaired range. Only 32% of the sample obtained scores within the normal range. Similarly, only 27% of the older participants produced adequate narrative discourses for the Frog story. The younger children performed much better on the Bus story with equal numbers of participants producing impaired and adequate discourses. Thus the older children had a much greater difficulty producing narratives than the younger children.

3.3.2 Language in children with FASD.

The studies of language in children with FASD can be separated into three groups, namely those studies that have found language impairments but have not accounted for intellectual functioning, studies that have found language impairments beyond that which general intellectual functioning could account for, and language impairments that are accounted for by the intellectual functioning of the child.

3.3.2.1 Language impairments without accounting for intellectual functioning

A study on American Indian children with FAS by Carney and Chermak (1991) found expressive and receptive language difficulties as measured on the Test of Language Development (TOLD-Primary and Intermediate). Ten children diagnosed with FAS with a mean age of 8 years 6 months were compared to a control group of 17 normal American Indian children with a mean age of 9 years 2 months. The children's ages ranged from 4 years to 12 years 11 months. The FAS group had a mean IQ score of 79 as measured by the WISC-R, and all FAS children were receiving special education services in public schools. The control group was not tested on intellectual functioning but was reported to be functioning at their grade level by their teachers.

In terms of language impairment, significantly poorer performances by the children with FAS were reported for all subtests of the TOLD-Primary, except word articulation. Also, significantly poorer performances of the older FAS group were reported for the sentence combining, word ordering and grammatical comprehension subtests of the TOLD-Intermediate. Carney and Chermak (1991) concluded that the younger FAS group showed global deficits on language comprehension and production whereas the older children showed difficulties in language comprehension and production but primarily in subtests requiring syntactic manipulation. However, these language differences may rather reflect differences in intellectual functioning between the groups.

3.3.2.2 Language impairments beyond that which intellectual functioning accounted for

Impairments in language were reported in 10 five-year-old children with FAS on a battery of syntactic, semantic and pragmatic tests (Hamilton, 1981). Differences in language ability were found in comparison to three groups:

1. normally developing five-year-old children;
2. three-year-old normally developing children matched on mean length utterance to the FAS group; and
3. five-year old children with Prader-Willi Syndrome whose performance IQ matched that of the FAS group.

It was reported that the FAS group developed language at a slower rate than the younger normally developing children who were matched on mean length utterance. The FAS group produced less complex and less grammatically complete language than the groups of normally developing children. They also had problems interpreting the pragmatic intent of language and as a result did not produce as appropriate responses to the dialogue as the younger normal children. The greatest variation of language performance was found in language production. In contrast, the FAS group performed better than the intellectually matched Prader-Willi group on expressive semantic measures. Hamilton thus concluded that the characteristics of Fetal Alcohol Syndrome and Prader-Willi syndrome may impact on the language abilities of the children beyond that which may be accounted for by general intellectual functioning (Hamilton, 1981).

3.3.2.3 Language impairments accounted for by intellectual functioning

A study of language comprehension and production of eight North American Indian children diagnosed with FAS was conducted by Becker et al. (1990). This study used a battery of tests similar to Iosub et al. (1981). The age of the FAS group ranged from 4 years 6 months to 9 years 6 months. Six of the FAS children were compared to six younger control children matched on non-verbal cognitive ability as measured by the Ravens Coloured Progressive Matrices. The age of the control group ranged from 3 years 6 months to 7 years. Two children with FAS were not compared to intelligence matched controls.

The language performance of the FAS group was compared to the younger control group, matched on intellectual functioning. The battery of tests included the Test of Auditory Comprehension of Language, Token Test, North-western Syntax Screening Test, Developmental Sentence Scoring procedure, Test of Psycholinguistic Abilities and naming on confrontations subtest of the Clinical Evaluations of Language fundamentals. It was found that the comprehension of morphological and syntactic forms for picture identification was poorer in the FAS group than their younger IQ-matched controls. The children with FAS also produced significantly fewer grammatically accurate and complete spontaneous sentences and had a poorer comprehension of verbal commands. These deficits were generally consistent with mental age suggesting slowed but normal development. There were no significant differences on naming ability between the groups.

Another study finding language abilities consistent with intellectual functioning examined 25 FASD children, between 3 and 5 years of age (McGee et al., 2009). These children were compared on the Clinical Evaluation of Language Fundamentals (Preschool version) to a control group of 26 non-exposed typically developing children. The control group was matched on age, sex, race and socio-economic status. Intellectual functioning was measured by the full scale IQ score from the WISC-IV. It was found that both groups had significantly better receptive than expressive language, but that the FASD group performed significantly worse than the control group on expressive and receptive language measures.

3.4 Language and FASD in South Africa

A South African study investigating the efficacy of a school language and literacy programme found significant language deficits, at baseline assessment, in children with FASD (Adnams et al., 2007). Forty 9-year-old children with FASD were assessed on the Ballard One-minute Addition and Subtraction Arithmetic Tests, Phonological Awareness and Early Literacy Test, and the University of Cape Town Reading and Spelling Tests prior to the intervention. Children with FASD were found to be significantly weaker in reading, spelling, addition, subtraction and phonological awareness than a control group of 25 non-exposed controls.

3.5 Neuroimaging and language

The relationship between brain structure and functioning, and the cognitive and behavioural consequences following prenatal alcohol exposure is not well understood. It has been established that there is a reduction in overall brain volume, CNS disorganisation and structural abnormalities of the corpus callosum, cerebellum, caudate and hippocampus in FASD (Norman, Crocker, Mattson & Riley, 2009).

It has also been established that a thinner cortex is associated with a more proficient performance on neuropsychological measures of verbal abilities (Sowell et al., 2004) and general intellectual functioning (Shaw, Greenstein & Lerch, 2006). Greater cortical thickness was reported following prenatal alcohol exposure in subjects between 8 and 25 years of age (Sowell et al., 2008). Cortical thickness, specifically in the temporal and parietal regions, has been noted following prenatal alcohol exposure (Sowell, Mattson, Thompson, Jernigan, Riley & Toga, 2001). Grey matter asymmetry is reported to be primarily altered at the conjunction of areas associated with language processing, Brodmann's areas 21 and 22 (Sowell, 2002).

It remains unknown if structural changes associated with prenatal alcohol exposure influence language processing. However, given that the areas associated with language processing have been shown to be abnormal following prenatal alcohol exposure it is likely that these physiological anomalies effect language processing. There is a need for the relationships between these cortical changes and the neuropsychological consequences to be explored.

3.6 Summary of the literature on language in children prenatally exposed to alcohol

The studies outlined above are consistent in reporting difficulties of language production and comprehension in children heavily exposed to alcohol prenatally. Some of the studies have concluded that the language abilities of children with FASD represent normal development, but at a slower rate, as the language difficulties experienced by the children are accounted for by their intellectual functioning.

The results have also shown that language functioning of children with prenatal alcohol exposure changes over time. Older children appear to have greater deficits in syntactic aspects of language rather than the global deficits found in younger children. Older children have been reported to have emerging difficulties in higher-level language skills, such as narrative discourse.

The studies discussed have had a number of limitations. The sample sizes have been small and the ages of participants varied across a wide range. In some studies no comparison groups were used and the effect of intellectual function on language ability was not investigated.

3.7 Rationale of the study

One of the manifold sequelae of prenatal alcohol exposure is language impairments. The literature on language impairments in young children with FASD has consistently found deficits in production and comprehension. The language abilities of adolescents with FASD have not been investigated. The prevalence of FASD in South Africa is the highest in the world. As such, research into the prevention of FASD and the deficits that follow prenatal alcohol exposure as well as intervention programmes that can address these issues are warranted. This study aims to contribute to the literature of language impairments in adolescents with FASD as well as to the understanding of whether language represents a core deficit in FASD.

CHAPTER 4
RESEARCH DESIGN AND METHODOLOGY

This is a comparative study of language production, comprehension and word naming in adolescents with FASD and normally developing controls. The project is a sub-study of a larger international study of the Collaborative Initiative of Fetal Alcohol Spectrum Disorders (CIFASD-II), funded by the NIAAA.

One of the CIFASD-II studies is investigating the neurobehavioural phenotype associated with FASD (CIFASD, 2010). This study is being conducted over six international sites, including South Africa, the University of Cape Town (UCT) being one of the collaborating institutions. The functional domains of executive functioning, working memory, verbal function and psychological symptomatology are being measured to determine if global deficits or a pattern of more specific strengths and weaknesses exists in children with FASD.

4.1 Participants

The participants in the present study are a convenience sample of 25 children diagnosed with FASD and 25 typically developing children with little or no exposure to alcohol prenatally. The children were recruited from the South African CIFASD-II neurobehavioural study participant cohort. This cohort was initially ascertained in an epidemiological study conducted in 2002 on grade 1 scholars in Wellington in the Western Cape Province (May et al., 2007).

Due to the difficulty in finding and recruiting children that met the inclusion criteria for participation in the present study, it was not possible to use random sampling. The children were between 14 years and 16 years and 11 months of age and were drawn from ten schools in Wellington: three secondary schools; six primary schools and one special-needs school. All participants came from a low socioeconomic background. Exclusion criteria for the subjects in this study included any head injuries, developmental and genetic disorders other than FASD; such as epilepsy, and hearing disorders. Information for the exclusion criteria and demographic data was obtained from the 2002 epidemiological study.

4.1.1 Diagnosis of FASD

The diagnosis of the children in the 2002 epidemiological study was carried out by a two-tier approach and active case ascertainment, as shown in Figure 4.1. In the initial tier 1 screening, Grade 1 children whose height, weight and occipito-frontal head circumference were below the 10th percentile were referred for Tier II assessments. Three types of Tier II assessment were conducted for each child: (1) dysmorphology assessments; (2) developmental testing; and (3) a maternal interview.

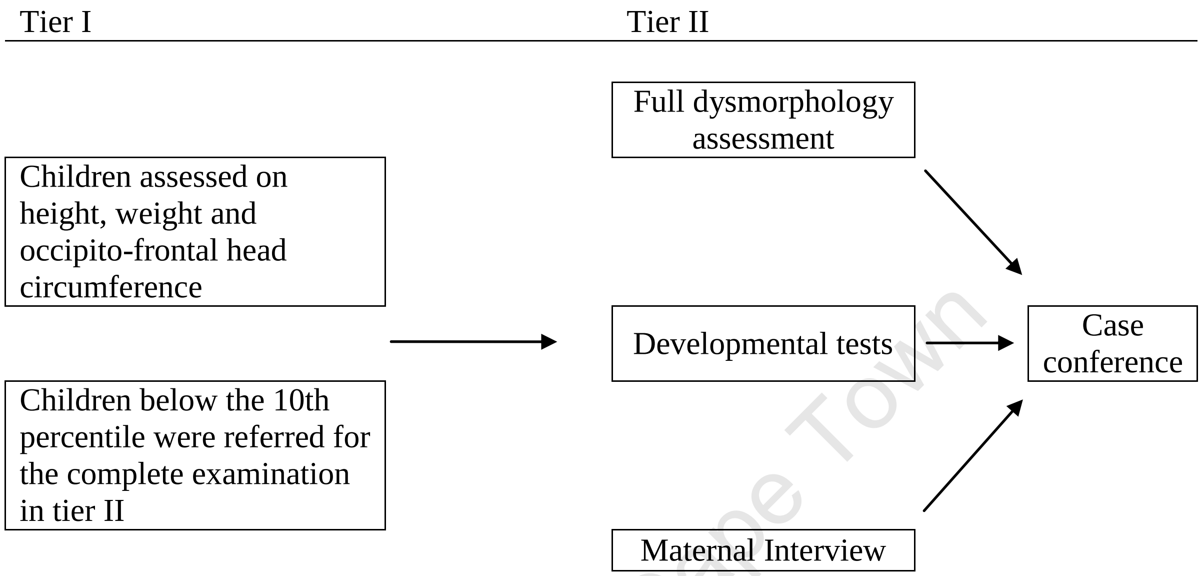

Figure 4.1. The two-tier diagnostic approach.

The Tier II dysmophology assessments were conducted by two independent teams in the children's classrooms. The revised Institute of Medicine's (IOM) criteria were used for diagnosis (Hoyme et al., 2005). The IOM criteria can be found in Appendix A. At the assessment, each participating child was examined by two physician teams. They measured the child's occipito-frontal circumference; palpebral fissure length; philtrum length; inner and outer canthal distance; and other indicators such as abnormalities in joints, heart function, and palmar creases. The findings were recorded on data forms and the physicians in each team verified each others' finding. All physicians were 'blinded' from any prior knowledge of the child or mother. Once seen by one team, the child was directed to another blinded team that repeated the examination and measurements as a reliability check.

Neurodevelopmental tests and behavioural questionnaires were used to assess the neuropsychological functioning of the children. The Ravens Coloured Progressive Matrices (Raven, Raven & Court, 1998) was used as a test of non-verbal intellectual functioning and Test of Reception of Grammar (Bishop, 1983) was used as a verbal proxy for intelligence.

A maternal interview was conducted to confirm prenatal exposure to alcohol and the dosage of alcohol consumed. A primary diagnosis along the FASD spectrum without confirmation of drinking is not typically due to the mother's denial of alcohol consumption but is often as a result of the mother not being available for the interview (Viljoen et al., 2005). In these cases the maternal interview was conducted with the nearest relative to gather information on alcohol consumption, difficulties during the pregnancy, developmental milestones and demographic data such as socio-economic status.

The information from the neurodevelopmental tests, maternal interview and dysmorphology examination was examined together for each child at the case conference. Here the child was assigned a diagnosis of 'not FAS', 'deferred', or a primary diagnosis within the FASD spectrum based on this information.

4.1.2 Recruitment of Participants

All participants were initially recruited for the CIFASD-II study in 2009 and 2010. For this original study the children and parents were given consent and assent forms (Appendix B) detailing the tests, procedures, confidentiality and the ability to withdraw from the testing at any time.

The participants recruited for this language study were the first 25 control children and 25 FASD children, from the cohort of CIFASD-II participants who met the age range criteria for this study (14 years to 16 years 11 months) and who consented to participate. All children in the FASD group had a diagnosis of FAS or partial FAS. These children and parents were given new consent and assent forms (Appendix C) detailing the language tests and procedure for the language battery of tests.

4.2 Neuropsychological tests and outcome measures

The CIFASD-II neuropsychological assessment study has employed a wide battery of standardised neuropsychological tests and questionnaires across all sites (Table 4.1 in Appendix D). The intelligence tests from this battery were included in the language study as it is expected that intelligence, which typically differs between control and FASD groups, may have an impact on the language ability of the participants and this measure needed to be included in the analysis.

The neurobehavioural study battery was translated into Afrikaans for the assessment of participants in South Africa. The translation of these tests was performed by a first language Afrikaans speaking clinical psychologist. The tests were then back-translated. Some of the items were modified to be more culturally appropriate for the study population.

The language measures used in the present study are additional to the CIFASD-II battery. The tests and the domains of language they assess are summarised in Table 4.2 in Appendix E. The language battery chosen was based on tests that are typically used in language research for this age group and availability in South Africa. The battery of language tests used in this study could not be based on tests used in the FASD literature as the language ability for this age range has not previously been investigated. The translation of the tests into Afrikaans is detailed below.

Two tests for language comprehension and two for language production were used. Multiple tests for the same domain were used to allow for correlations to be made between the tests. It was hypothesised that correlations between the tests of the same measure would help justify the validity of the tests, as the validity of the tests was called into question as a result of translation into Afrikaans. If the language tests used in this study measured the same domain, then these results would be expected to positively correlate. In the translation of the tests distortions may have occurred. It was expected that if a relationship between the tests could not be inferred then the construct validity of the test was more doubtful.

4.2.1 General Intelligence

4.2.1.1 Weschler Intelligence Scale for Children- Fourth Edition (WISC-IV)

The WISC-IV is the most widely used assessment of intellectual ability for children (Weschler, 2004). It includes norms for children between the ages of 6 and 11 years of age, based on a USA sample. The 15 sub-tests of the WISC-IV are categorised into 4 composite scores, which sum to give a Full Scale IQ (FSIQ) estimate. The composite scores are Verbal Comprehension, Perceptual Reasoning, Working Memory and Processing Speed.

Verbal Comprehension requires language based reasoning to solve problems whereas *Perceptual Reasoning* is the ability to solve problems using visual perception, organisation and reasoning with visually presented, nonverbal material.

4.2.2 Language tests

4.2.2.1 Clinical Evaluation of Language Fundamentals-Revised (CELF-R)

The Clinical Evaluation of Language Fundamentals - Revised subtests measure expressive and receptive language (Semel, Wiig & Secord, 2004). The subtests can be grouped into expressive and receptive language scores that are added together to form a total language score. The receptive language subtests are oral directions, word classes and semantic relationships. The expressive language subtests are formulated sentences, recalling sentences and sentence assembly. Figure 4.2 is a flow diagram of the CELF-R subtests and scores that form the total language score. This battery has an age range of 3 years to 16 years, 11 months. Although this is not the most current version of the CELF series, this is the only version available for use in South Africa.

The CELF-R subtests are:

Oral directions; requires the child to point to a number of coloured shapes in an instructed sequence, the task gradually increasing in complexity.

Word classes; requires the child to categorise words by semantic class, opposites, spatial and temporal features.

Semantic relationships; assesses semantic relationships in spoken sentences. The test is divided into comparative, spatial, passive and temporal relationships. The child is presented with four words, two of which are related to a spoken sentence.

Formulated sentences; requires the subject to produce a one sentence response to a picture using a specific word in the sentence.

Recalling sentences; requires repetition of increasingly complex sentences.

Sentence assembly; requires the participant to form two sentences from a number of words presented to the participant.

An expressive and receptive standard score as well as total language score can be computed for the CELF-R. These three scores are the outcome measures of these tests.

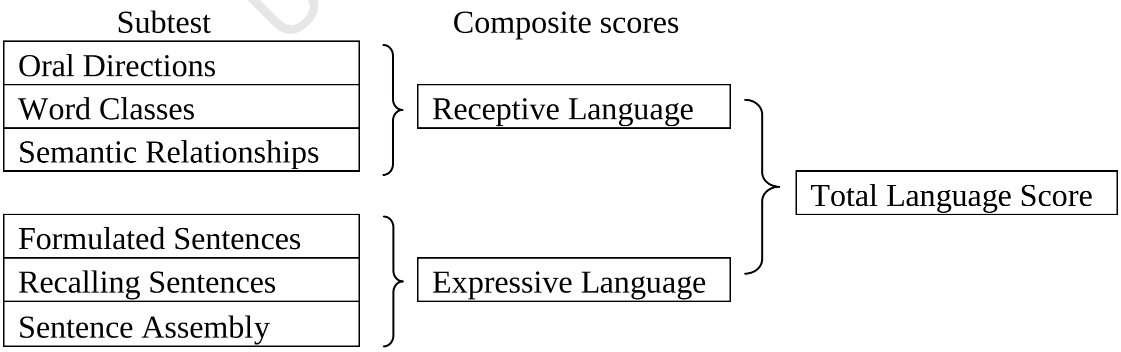

Figure 4.2.3: The subtests that summate to form the CELF-R total language score.

4.2.2.2 The Cookie Jar Theft Picture

The Cookie Jar Theft Picture is a test of spontaneous language production. The participant is required to produce a comprehensive explanation of a scene drawing presented to them. The Cookie Jar Theft Picture has been found to yield relatively short language descriptions and have predictable content (Hux, Wallace, Evans & Snell, 2008). It was unknown how the Afrikaans population's responses would differ from that of the English language population's responses. Standard administration of the test applied and the total number of utterances and complexity index (number of clauses per utterance) score is the outcome measure for this test.

4.2.2.3 Test for Reception of Grammar, Second Edition (TROG-2)

The Test for Reception Of Grammar (Bishop, 1983) has been used in FASD research both in South Africa and internationally (Adnams et al., 2007; Aragon et al., 2008). The number of blocks passed as a standard score is the typically used outcome measures for this test. The TROG-2 (Bishop, 2003) is a revised version of the TROG with an increased difficulty level, age range and ceiling to allow for effective testing of the receptive language skills of secondary school aged children and young adults. The TROG-2 has been standardised for children of 8 to 16 years of age and for adults. Test stimuli are presented in a multiple choice format with lexical and grammatical foils, more specifically, 20 constructs are tested four times each, using different stimuli.

4.2.2.4 The Boston Naming Test

The Boston Naming Test (BNT) is a measure of word naming (Goodglass & Kaplan, 1972). The 60-item standard form has been shortened and translated into a 16-item Afrikaans Naming test. This shorter version of the test was used. The age range is 5 years to adulthood. The outcome measure for this test is the total number of correct items.

4.3 Translation of the language tests

The translation of the tests into Afrikaans, the home language of the participants, was imperative to obtain an understanding of each child's language functioning. There are very few normalised and validated neuropsychological tests in South Africa due to the difficulty in translating and validating test measures for the South African population. The difficulty is, in part, due to the wide cultural and language differences across South Africa, the effect of socio-economic and educational status upon the tests as well as socio-political disadvantage (Paterson & Uys, 2005). The scarcity of Afrikaans language measures is currently being addressed, however there are currently no validated and normalised Afrikaans language measures in South Africa for children over 14 years of age (personal communication, B. Gerber, April 22 2010).

To assess language production, comprehension and word naming in Afrikaans children between 14 and 16 years of age, commonly used and age-appropriate tests in the language literature that assessed the desired domains were, of necessity, translated into Afrikaans. As a result of translating the tests into Afrikaans, the validity of these tests has been called into question. The measures are also not comparable to international norms. The test measures are thus only comparable between the FASD and non-exposed control group. Although the question of validity of the translated tests is a limitation in this study, there were no alternative options. Thus all results are carefully interpreted in light of this limitation. It is hoped that this research will promote the creation of valid language measures for the South African population.

The WISC-IV, DKEFS and Boston Naming tests were translated into Afrikaans for the CIFASD-II and other neuropsychological studies in South Africa. These tests were translated and back-translated for the studies but are not, as yet, normalised or validated. For this study the TROG-2, CELF-R and Cookie Jar Theft Picture were translated into Afrikaans. The tests were then independently back-translated by an independent first-language Afrikaans Masters graduate in psychology at UCT. Any discrepancies were noted and discussed and the appropriate changes made. Many of the test items could be directly translated from English into Afrikaans without changing the intention of the test.

The tests that were directly translated are the Cookie Jar Theft Picture, TROG-2, CELF-R Oral Directions and Recalling Sentences subtests. For instance, the instruction ‘Point to the small square and white circle, begin.’ could be directly translated into Afrikaans. Similarly the recalling sentences subtest could be directly translated. Some of the sentences from this test became shorter or longer due to grammatical differences between English and Afrikaans. It was felt that the length changes of the items did not significantly alter the difficulty of the item.

Items in the CELF-R, formulated sentences, word classes, semantic relationships and sentence assembly tests were changed to accommodate for cultural changes and grammatical differences between English and Afrikaans. For instance in the CELF-R semantic relationships test, the item ‘Thanksgiving comes between…’ had to be changed to accommodate cultural differences. These changes are illustrated in Appendix F with the English and Afrikaans version of an item from each test. Extra care was taken in the modification of these items to ensure that a true reflection of the original content was maintained.

4.4 Procedure

Participants and their caregivers were contacted to explain the study, obtain consent and organise a date for testing. The schools were then informed of the child’s participation as they were required to miss a day of school for administration of the CIFASD-II battery. The language tests were conducted at the participant’s school in a quiet room.

4.4.1 Neuropsychological testing

The neuropsychological tests were administered in two stages. The first stage was the neuropsychological testing for the CIFASD-II study. Parts of the CIFASD-II battery of tests have been used in this language study. The second stage of the neuropsychological testing was the administration of the specific language tests translated for this study.

For the CIFASD-II neuropsychology tests the participants were tested in a private, quiet, two-roomed assessment office in Wellington. A blinded CIFASD-II researcher administered the neuropsychological battery. The battery was administered such that the majority of the WISC-IV subtests were administered to the child over approximately 2 hours. This was followed by a break. The CANTAB (Cambridge Neuropsychological Test Automated Battery) was then administered to the child for one hour. Following a second break the D-KEFS (Delis Kaplan Executive Function System) and the remainder of the WISC-IV subtests were administered for approximately 2 hours. In total the neuropsychological tests for the CIFASD-II study took approximately 5 hours to administer. The neuropsychological battery administration sequence can be found in Table 4.3 in Appendix G.

The language measures were administered to the children in a quiet room at their respective schools. The language tests took approximately 80 minutes to complete. No breaks were given during the testing unless required by the participant. The language battery administration sequence can be found in Table 4.4 in Appendix H.

4.4.2 Data checking and capturing

The data from the neuropsychological tests for the CIFASD-II study underwent a rigorous capturing and checking process to minimise inter-rater differences and data capturing errors. The data was initially scored by the researcher who administered the neuropsychological tests. The data was then independently checked and any discrepancies were noted and discussed by the researchers before being corrected. The data was then uploaded onto the respective test scoring tools. These tools produce a printout of the raw scores, scaled-scores and summary scales. The data from the electronic scoring tool was then uploaded onto a CIFASD-II computer based tool that contains all the demographic, neuropsychological and questionnaire data for each participant under the participant's study number, thus ensuring confidentiality. The data on this CIFASD-II tool was then checked against the data generated by the scoring tool.

The language data was stored separately from the CIFASD-II neuropsychological data. To ensure confidentiality yet ensure comparability across studies the same study number as that of the CIFASD-II study was used for the language data.

4.5 Data Analysis

4.5.1 Variables

The independent variable in this study is the attribute variable of FASD vs. non-exposed control. The small sample size of this study did not allow for comparisons between the primary diagnoses of FAS and partial FAS. However, the aim of this study was to compare the language ability of FASD to normal controls and not the comparison of language ability within the FASD spectrum. Both the FAS and partial FAS primary diagnoses represent the severe end of the spectrum of disorders. The dependent variables are the IQ and language scores from the tests referred to earlier. These dependent variables are listed in Table 4.5 in Appendix I.

4.5.2 Statistical Analyses

All statistical analyses were conducted using STATA version 10.0 (StataCorp, 2007). The significance level applied was 95% i.e. $\alpha = .05$. The statistical analyses were designed to address the three hypotheses as outlined in the specific aims of this study, repeated as follows:

1. The FASD group would have a significantly lower intellectual functioning than the control group;

2. Language comprehension, production and word naming would be significantly worse in the FASD group; and

3. Language comprehension, production and word naming deficits in the FASD group would be accounted for by deficits in intellectual functioning.

4.5.2.1 Preliminary data analysis

The demographic data shows the composition of the sample in terms of age, gender, race, home language, school grade and the distribution of the children in primary, secondary, and special schools. These variables, as well as the primary diagnosis along the FASD spectrum, were recorded and descriptive statistics were calculated.

Once the demographic data had been recorded, the IQ and language data for the groups were inspected. It was anticipated that the results for the IQ and language tests when compared to the standard scores would show a basement effect because of the known low performance of South African children on neuropsychological measures (Skuy, Schutte, Fridjhon & O'Carroll, 2000).

To explore this, the percentage of FASD children that scored the lowest possible standard score was calculated. To ameliorate a basement effect, the raw scores, instead of the standard scores, of the FASD group were compared to the scores of the control group. To examine if the assumption of normality was upheld histograms and box-plots of the raw scores for the language and IQ data of each group were produced. To investigate if the assumption of equality of variance was upheld, variance-comparison tests were calculated. t-tests, using Welch's approximation of the degrees of freedom, were used if the variables did not have equal variances.

4.5.2.2 Correlations between language tests

As described earlier, multiple tests of language production and comprehension were used to minimise the risk of the tests being invalid. To explore the relationship between language tests of the same domain, correlation matrices between the tests of language comprehension and between the tests of language production were calculated.

4.5.2.3 Hypotheses 1 and 2: Differences in IQ and language between the groups

Intellectual functioning of children with FASD is known to be lower than that of control children (Mattson & Riley, 1998). As this difference in intellectual functioning may influence the language abilities of the children, the possibility of a difference in intellectual functioning between the groups was examined. The intellectual functioning of the two groups was compared by independent sample Welch t-tests. T-tests of the Full Scale IQ, Verbal Comprehension and Perceptual Reasoning composite scores between the two groups were calculated. The mean Full Scale IQ of the control group was compared to the average WISC-IV Full Scale IQ.

The CELF-R composite scores, namely the expressive, receptive and total language scores, were used in the analyses. The CELF-R subtest scores each measure a separate element of language and combined give an understanding of language comprehension and language production. These composite scores were used due to their stronger psychometric properties in comparison to the individual subtest scores (McGee et al., 2009). To investigate the language functioning between the two groups, independent sample Welch t-tests were performed between the word naming, language comprehension and language production measures.

T-tests between the CELF-R receptive and expressive language standard scores were calculated to investigate differences between language comprehension and language production within the FASD and control groups.

4.5.2.4 Hypothesis 3: IQ-independent language functioning

Intelligence (for which IQ is a common proxy) is an important variable to be covaried when estimating the impact of FAS on language, as intelligence may play a role in language ability. To partial out the effects of IQ, hierarchical regression analyses were calculated.

A child's language ability is reflected in the Verbal Comprehension and Full Scale IQ indexes. Perceptual Reasoning IQ is an index of IQ that does not rely on a child's language ability. The Perceptual Reasoning IQ was used as a language-independent proxy of intelligence in the regression analyses.

The nested models of the hierarchical regression for the total language score, language comprehension, language production and naming, with the Independent Variable (IV) and the Dependent Variables (DV) are shown in Tables 4.6 – 4.9 below. In each case model “a” was nested in model “b” to determine the unique contribution of diagnosis to the variance of the language score when Perceptual Reasoning was partialled out. The change in R^2 between model *a* and *b* was calculated to find the amount of variance explained uniquely by diagnosis.

Differences in school grade and gender between the FASD and control groups were controlled for in a hierarchical regression, of the total language raw score, to determine the unique contribution of diagnosis. The nested regression models are shown in Table 4.10 below. In this nested regression, model “a” was nested in model “b” to determine the contribution of the school grade over and above that of IQ. This was then nested in model “c” to determine the contribution of gender over and above that of IQ and school grade. This was then nested in model “d” to determine the contribution of diagnosis over and above that of IQ, school grade and gender. Correlations between grade and Perceptual Reasoning IQ and the Total Language score were calculated. A separate nested regression was computed to allow for distinction between the regressions that controlled for IQ only and the regression that controlled for school grade and gender.

Table 4.6

Regression models of the total language score that controlled for Perceptual Reasoning IQ.

Model	Dependent Variable	Independent Variables
1a	CELF-R Total language score	Perceptual Reasoning composite score
1b	CELF-R Total language score	Perceptual Reasoning composite score Diagnosis

Table 4.7

Regression models of language comprehension that controlled for Perceptual Reasoning IQ.

Model	Dependent Variable	Independent Variable
2a	CELF-R receptive language score	Perceptual Reasoning composite score
2b	CELF-R receptive language score	Perceptual Reasoning composite score Diagnosis
3a	TROG-2	Perceptual Reasoning composite score
3b	TROG-2	Perceptual Reasoning composite score Diagnosis

Table 4.8

Regression models of language production that controlled for Perceptual Reasoning IQ.

Model	Dependent Variable	Independent Variable
4a	CELF-R expressive language score	Perceptual Reasoning composite score
4b	CELF-R expressive language score	Perceptual Reasoning composite score Diagnosis
5a	Cookie Jar Test Complexity Index	Perceptual Reasoning composite score
5b	Cookie Jar Test Complexity Index	Perceptual Reasoning composite score Diagnosis

Table 4.9

Regression models of naming that controlled for Perceptual Reasoning IQ.

Model	Dependent Variable	Independent Variable
6a	Boston Naming Test total score	Perceptual Reasoning composite score
6b	Boston Naming Test total score	Perceptual Reasoning composite score Diagnosis

Table 4.10

Regression models that controlled for Perceptual Reasoning IQ, school grade and gender.

Model	Dependent Variable	Independent Variable
1a	CELF-R Total language score	Perceptual Reasoning composite score
1b	CELF-R Total language score	Perceptual Reasoning composite score School grade
1c	CELF-R Total language score	Perceptual Reasoning composite score School grade Gender
1b	CELF-R Total language score	Perceptual Reasoning composite score School grade Gender Diagnosis

I considered using discriminant function analysis to find the combination of independent variables that best discriminated the FASD and control groups. This analysis was rejected due to the small sample size and the limited variables in this study. The predictive value of language variables in the diagnosis of FASD should be investigated in a much larger sample that also considers other factors that are associated with FASD, such as behaviour problems (Rasmussen, Horne & Witol, 2006) and executive dysfunction (Kodituwakku, 2007). The primary aims of this study were not compromised by the rejection of this analysis.

CHAPTER 5
RESULTS

5.1 Synopsis of main findings

The FASD and Control groups were compared on measures of IQ and language. There were significant differences in Full Scale IQ, Perceptual Reasoning and Verbal Comprehension composite scores. There were also highly significant differences between the FASD and control groups on all language tests, except the Cookie Jar Picture test. When controlling for Perceptual Reasoning IQ it was identified that the diagnosis of the child uniquely accounts for a significant amount of total language and expressive language variance. Diagnosis did not significantly predict for the variance in language comprehension. This chapter will outline the demographic results, the correlations between the tests, and then the group comparisons of the intelligence and language tests, and the language test results controlling for IQ.

5.2 Demographic data

The two groups were matched on sample size. The groups were also similar in average age. All the children were coloured and first language Afrikaans speakers. The number of females in the control group was higher than that of the FASD group. The demographic data for the two groups can be seen in Table 5.1 below.

Of note are the differences in school grades for the two groups, the FASD group being on average, two grades lower than the control group. Of the children in the FASD group, three were in a special school, one was enrolled in a special class (within a mainstream school), and one was no longer enrolled in school. The remaining twenty children attended standard school classes. All children in the control group were enrolled in main stream schools. The socio-economic status of the two groups was deemed the same as all participants came from a low socioeconomic background. Similarly the maternal education of the two groups was considered matched as all mothers obtained fewer than 10 years of schooling.

Table 5.1

A summary of the demographic data for FASD and Control groups

	Control	FASD
Sample size	25	25
Mean Age (*SD*)	15.3 (0.476)	15.6 (0.641)
Age range	14.10 -15.8	14.3 - 16.2
% Ratio Females: Males	68:32	32:68
Modal Grade	9	7
Grade range	7 - 9	3-11

On the spectrum of fetal alcohol disorders, the FASD children in this study were diagnosed as FAS or partial FAS with or without confirmation of drinking by their mothers. A summary of the primary diagnoses is shown in Table 5.2. A diagnosis of FAS or partial FAS without confirmation was often due to a maternal interview being unavailable, rather than the denial of drinking by the mother.

Table 5.2

Summary of the primary diagnoses of children along the FASD spectrum

Primary diagnosis	*N*
FAS with confirmation	15
FAS without confirmation	4
Partial FAS with confirmation	5
Partial FAS without confirmation	1
Total FASD sample	25

5.3 Neuropsychological data

5.3.1 Summary of the data pre-analysis

A summary of the overall raw scores for the Language and IQ data are summarised in Table 5.3 below to give an overview. The standard scores for the TROG-2 and CELF-R are summarised in table 5.4 in Appendix J. The standard scores have been tabulated for comparison of the CELF-R scores within the same group.

Table 5.3

A summary of the overall language and IQ results for the FASD and Control groups

Variable	Group	*N*	Score		
			Mean	SD	Range
BNT total raw	Control	25	12.96	1.337	10-15
	FASD	25	11.12	2.127	6-14
TROG2 Raw	Control	25	12.68	2.657	4-17
	FASD	25	9.84	3.923	2-15
Cookie Jar complexity	Control	25	0.749	0.137	0.44-1
	FASD	25	0.807	0.168	0.5-1
Oral Directions	Control	25	17.32	3.375	9-22
	FASD	25	13.08	5.859	2-22
Recalling sentences	Control	25	61.96	61.96	53-75
	FASD	25	49.84	49.84	5-66
Formulated sentences	Control	25	45.76	10.059	18-60
	FASD	25	34.16	12.72	12-60
Word Classes	Control	25	19.16	4.2	11-25
	FASD	25	12.68	7.9	0-23
Semantic Relationships	Control	25	14.84	5.048	4-24
	FASD	25	9.76	4.78	3-19
Sentence Assembly	Control	25	13.72	5.3	5-21
	FASD	25	7.8	5.2	1-20
CELF receptive	Control	25	51.32	10.84	31-67
	FASD	25	35.52	16.65	7-60
CELF Expressive	Control	25	121.44	15.75	89-153
	FASD	25	91.8	27.1	31-146
CELF total	Control	25	74.8	14.27	53-100
	FASD	25	59.96	11.7	50-93
Full Scale IQ	Control	25	72.84	10.02	56-97
	FASD	25	55.16	11.12	40-74

	Control	25	73.88	13.39	57-112
Verbal Comprehension Composite Index	FASD	25	56.64	10.99	45-81
	Control	25	81	10.03	61-106
Perceptual Reasoning Composite Index	FASD	25	66.4	12.42	45-94

It is inappropriate to compare the results from translated tests on Afrikaans children to the normative data from the UK and USA. In order to avoid normative comparisons the analyses in this paper are based on the raw scores of the tests. It was also found that the standard scores result in a *basement effect*, i.e. 40% of the FASD group attained at most the lowest standard scores in the TROG2 and all the CELF-R tests. The raw scores of these tests show a greater spread of results and ameliorate this basement effect. Figure 5.1 below shows that the standard scores of the FASD group are negatively skewed as a high percentage of the participants obtained the lowest standard score. Figure 5.2 of the raw scores for the same test can be seen to have a much greater spread.

The distribution of the TROG-2 and all the CELF-R test raw scores as shown by histogram and box-plot distributions did not show clear evidence of violation of the assumption of normality (whereas the distribution of standard scores did). The TROG-2 and CELF-R scores were analysed as normal distributions.

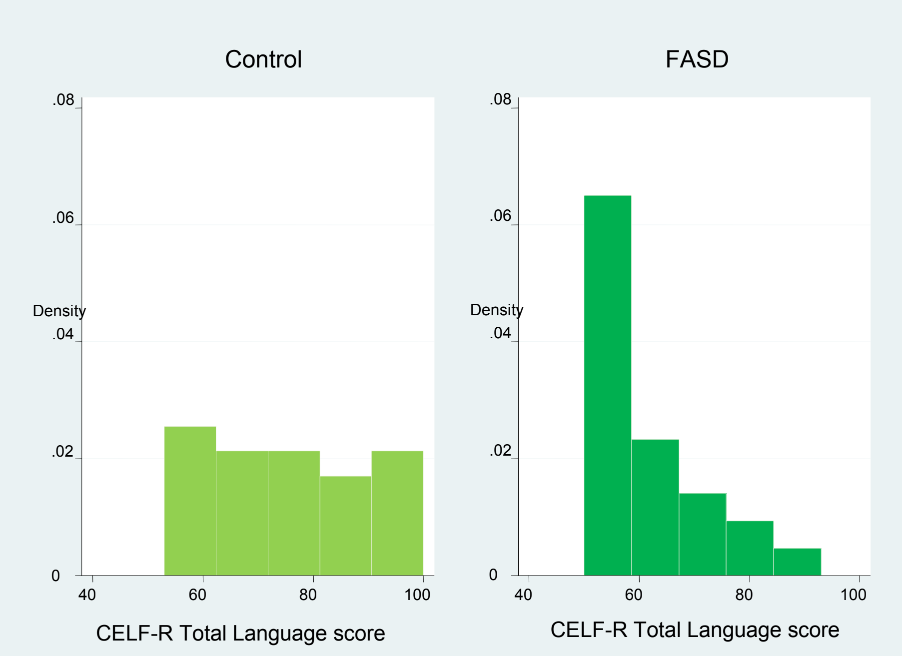

Figure 5.1. Distribution of the CELF-R total language *standard* score for the Control and FASD groups

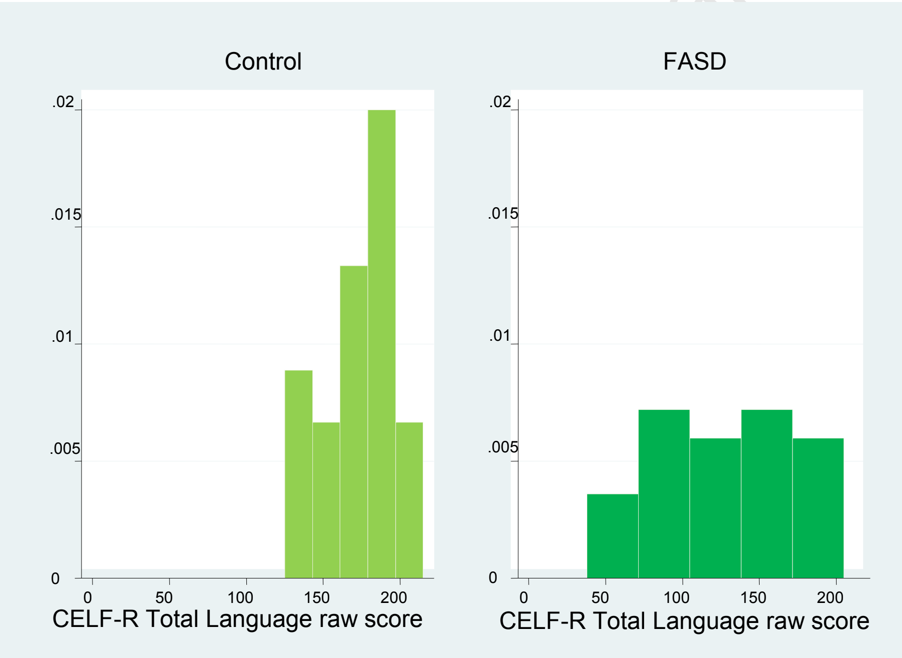

Figure 5.2. Distribution of the CELF-R total language *raw* score for the Control and FASD groups

5.3.2 Correlations between language tests

The correlations between language comprehension tests and the language production tests are shown in Table 5.5 below. For measures of language comprehension the TROG-2 and the CELF-R receptive language subtests were used. Within the CELF-R set of receptive language tests the scores show a significant positive correlation between all the tests.

Table 5.5

Correlations between language comprehension tests

Test	TROG2 Raw	CELF-R Oral Directions	CELF-R Word Classes	CELF-R Semantic Relationships	CELF-R Receptive Language
TROG2 Raw	1				
CELF-R Oral Directions	0.794 *	1			
CELF-R Word Classes	0.69 *	0.788 *	1		
CELF-R Semantic Relationships	0.666 *	0.65 *	0.704 *	1	
CELF-R Receptive Language	0.79 *	0.895 *	0.938 *	0.865 *	1

* *p* <0.001; *N pairs* = 10

Two measures of language production, the Cookie Jar Theft Picture and the CELF-R expressive language subtests were used in the study. Table 5.6 shows the correlations between the language production tests. Relationships between the Cookie Jar Theft Picture and the CELF-R subtests could not be inferred given that the correlations between the tests are not significant. There are significant positive correlations between the CELF-R subtests.

Table 5.6

Correlations between language productions test

Test	Cookie Jar Complexity Index	CELF-R Recalling Sentences	CELF-R Formulated Sentences	CELF-R Sentence Assembly	CELF-R Expressive Language
Cookie Jar Complexity Index	1				
CELF-R Recalling Sentences	-0.221	1			
CELF-R Formulated Sentences	-0.074	0.558 *	1		
CELF-R Sentence Assembly	-0.051	0.572 *	0.698 *	1	
CELF-R Expressive Language	-0.146	0.855 *	0.898 *	0.818 *	1

* *p* <0.001; *N pairs* = 10

Strong and significant positive correlations between the TROG-2 and CELF-R expressive language tests were found. Highly significant positive correlations between the CELF-R expressive language scores were also found. No significant correlations between the CELF-R expressive language tests and the Cookie Jar Theft Picture were found.

5.3.3 Group Comparisons

5.3.3.1 Intelligence tests

Strong effect sizes and significant differences between the FASD and control groups for all the composite scores of the WISC-IV intelligence tests were found. Welch t-tests were used to test the differences in IQ scores between the groups. Table 5.7 shows the Welch t-tests for the Full Scale IQ, Processing Speed and Verbal Comprehension composite scores.

The mean Full Scale IQ of the control group (M= 72.84, SD= 10.02) is significantly lower than the average WISC-IV IQ of 100 ($t(24)$= -13.55, p<0.001) and ranged from 56 to 97. Verbal Comprehension IQ is significantly lower than Perceptual Reasoning IQ, in children with FASD ($t(24)$= -5.396, p<0.001) and in the control group ($t(24)$= -2.85, p<0.004).

Table 5.7

Welch t-tests for comparison of IQ measures between the FASD and control groups

IQ measure	Group	Mean	Std Error	Standard Deviation	*t*	Welch's degrees of freedom	*p*	Cohen's d
Full Scale IQ	Control	72.84	2.004	10.019	5.907	49.449	<0.001	3.85
	FASD	55.16	2.223	11.116				
Verbal Comprehension	Control	73.88	2.678	13.389	4.976	48.093	<0.001	3.49
	FASD	56.64	2.198	10.988				
Perceptual Reasoning	Control	81	2.006	10.029	4.573	47.791	<0.001	3.08
	FASD	66.4	2.484	12.419				

5.3.3.2 Language tests

There are strong effect sizes and highly significant differences between the control and FASD groups in word naming, language comprehension and language production tests. Welch t-tests were used in the analyses of the language tests that follow.

5.3.4 IQ-dependent language differences

The results for the three domains of language, comprehension, production and naming, are presented separately below.

5.3.4.1 IQ-dependent comprehension differences

The children in the control group scored significantly higher and had very high effect sizes on *all* tests of language comprehension as well as in the CELF-R receptive language score, which is the sum of all the CELF-R language comprehension tests. The Welch t-tests of language comprehension are shown in Table 5.8 below.

Table 5.8

Welch t-test comparisons of language comprehension between Control and FASD groups

Test measure	Group	Mean	Std Error	Std Dev	*t*	Welch's degrees of freedom	*P*	Cohen's d
TROG-2	Control FASD	12.68 9.84	0.532 0.785	2.657 3.923	2.997	43.707	<0.002	1.1
CELF-R Oral Directions	Control FASD	17.32 13.08	0.675 1.171	3.375 5.859	3.135	39.547	<0.002	1.4
CELF-R Word Classes	Control FASD	19.16 12.68	0.84 1.582	4.2 7.909	3.618	37.58	<0.001	1.86
CELF-R Semantic Relationships	Control FASD	14.84 9.76	1.009 0.956	5.047 4.781	3.654	49.848	<0.001	1.62
CELF-R Receptive Language	Control FASD	51.32 35.52	2.168 3.331	10.839 16.654	3.976	42.676	<0.001	3

5.3.4.2 IQ-dependent production differences

Similarly the control group had significantly higher language production scores than the FASD group for all the CELF-R language tests. There was no significant difference between the FASD and control group on the complexity index of the Cookie Jar test. The Welch t-tests of language production are shown in Table 5.9 below.

Table 5.9

Welch t-test comparisons of language production between the Control and FASD groups

Test measure	Group	Mean	Std Error	Std Dev	*t*	Welch's degrees of freedom	*P*	Cohen's d
CELF-R Recalling Sentences	Control	61.96	1.181	5.905	4.153	33.813	<0.001	2.76
	FASD	49.84	2.668	13.341				
CELF-R Formulated Sentences	Control	45.76	2.012	10.059	3.577	47.377	<0.001	2.73
	FASD	34.16	2.544	12.72				
CELF-R Sentence Assembly	Control	13.72	1.061	5.303	3.981	49.984	<0.001	1.83
	FASD	7.8	1.042	5.212				
CELF-R Expressive Language	Control	121.44	3.151	15.754	4.728	39.777	<0.001	4.53
	FASD	91.8	5.419	27.094				

5.3.4.3. IQ-dependent naming differences

There is also a strong effect size, of almost one standard deviation, and a significant difference in word naming ($t(41.78)=3.66$, $p<0.001$, *Cohen's d*=0.989), with the control group having a better naming ability on the Boston Naming test than the FASD group. The control group had significantly higher scores on almost all tests of language comprehension, production and word naming than the FASD counterparts. The only exception was the Cookie Jar Complexity Index, with the FASD group scoring better results than the controls.

5.3.4.4 Comparison of language production and comprehension on the CELF-R

The CELF-R allows for comparisons between the expressive and receptive language scores by using the standard scores for the tests. As discussed previously, the standard scores resulted in a large basement effect. Due to differences in score allocation between the CELF-R receptive and expressive language measures the raw scores could not be compared. The distribution of the FASD group's standard scores on the receptive and expressive language measure of the CELF-R can be seen in figure 5.3.

No statistically significant differences were found between the CELF-R expressive and receptive language scores for either of the groups (Table 5.10). The CELF-R is designed such that differences between expressive and receptive language would indicate a strength or weakness in that domain.

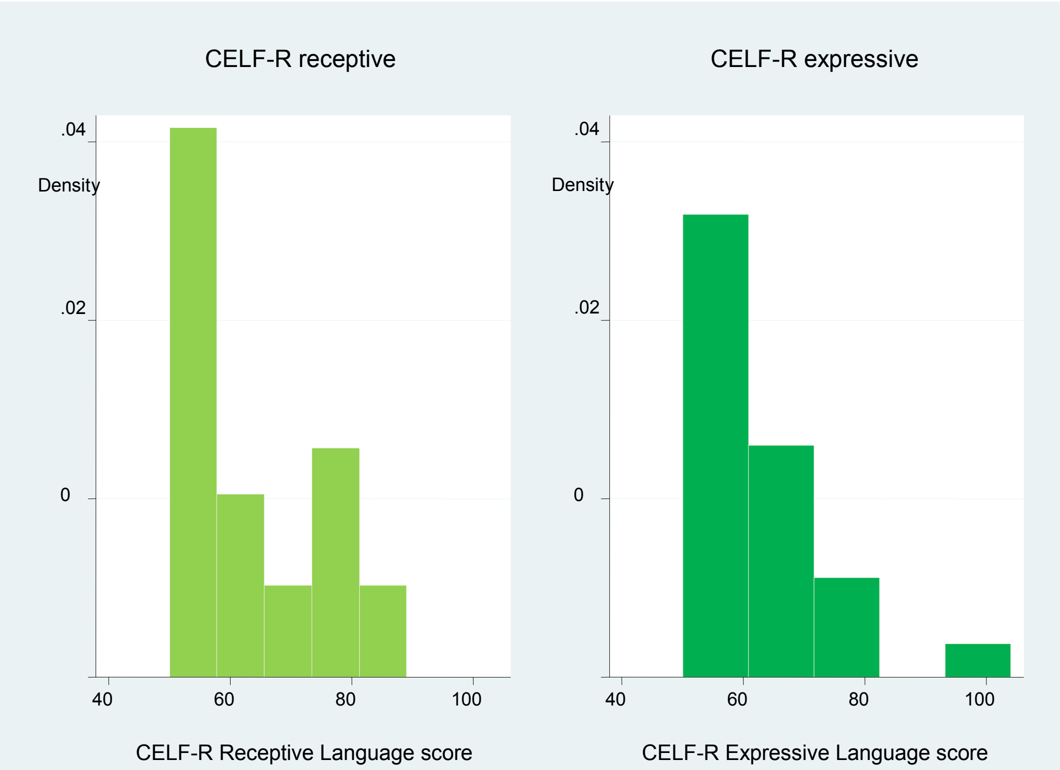

Figure 5.3. Distribution of standard scores for the FASD group on the CELF-R receptive and expressive language measures.

Table 5.10

T-tests between the CELF-R expressive and receptive score for the two groups

Group	Measure	Mean	Std Error	Std Dev	*t*	Degrees of freedom	*p*	Cohen's d
Control	Expressive	76.28	3.133	15.665	-0.209	24	<0.837	-0.113
	Receptive	76.76	2.715	13.575				
FASD	Expressive	62.04	2.592	12.96	0.859	24	<0.399	0.399
	Receptive	60.56	2.55	12.748				

5.3.5 IQ-independent language differences

As shown above there is a significant difference between the control and FASD groups in tests of intelligence. The Perceptual Reasoning IQ score was partialled out to ascertain the unique contribution of diagnosis to the language outcome measures. It can be seen in each model that Perceptual Reasoning is significantly associated with language so Perceptual Reasoning is an important variable to partial out.

5.3.5.1 Total language

Language differences between the two groups after the effect of Perceptual Reasoning IQ has been partialled out, show that Perceptual Reasoning IQ significantly predicts the CELF-R Total language score (F(1,48)=31,35, p<0.001, R^2=0.395). Thirty-nine percent of the CELF-R Total language score variance is accounted for by Perceptual Reasoning. When a regression is calculated with both Perceptual Reasoning and diagnosis as the independent variables, both variables significantly predict the CELF-R Total language score (F(2,47)=19,97, p<0.001, R^2=0.459). Thus 45.9% of the Total language score variance is accounted for by Perceptual Reasoning IQ and the diagnosis of the child. The significant change in R^2 of 0.064 (p<0.022) shows that 6.4% of the CELF-R Total language score variance is uniquely accounted for by the diagnosis of the child. The results from this hierarchical regression can be seen in Table 5.11 below.

Table 5.11

The results of the hierarchical regressions and the change statistics for the CELF-R total language score

Model	Dependent Variable	Parameters		Model Statistics		Change Statistics		
		β	*p*	R^2	*df*	ΔR^2	Δ F	*p*
1a	IQ	0.628	<0.001	0.395	1, 48			
1b	IQ, diagnosis	0.461; 0.304	<0.001; 0.022	0.459	2, 47	0.064	5.6	<0.022

5.3.5.2 Language comprehension

When using only the CELF-R receptive language score it can be seen that Perceptual Reasoning IQ significantly predicts the CELF-R Receptive Language score (F(1, 48)=37.99, p<0.001, R^2=0.442). Thus 44% of the variance of the CELF-R Receptive Language score is accounted for by Perceptual Reasoning IQ.

When a regression is calculated for both Perceptual Reasoning and diagnosis as the independent variables, it can be seen that the total model significantly predicts CELF-R Receptive Language (F(2,47)=20.56, p<0.001, R^2=0.467). Thus 46.7% of the variance of CELF-R Receptive Language is accounted for by Perceptual Reasoning IQ and diagnosis. The change in R^2 of 0.026 is non-significant and shows that only 2% of the variance of CELF-R Receptive language can be uniquely accounted for by diagnosis. The results from the hierarchical regressions for language comprehension can be seen in Table 5.12 below.

The TROG-2, a second measure of language comprehension, shows similar results of Perceptual Reasoning significantly predicting for the TROG-2 score (F(1, 48)=21.87, p<0.001, R^2=0.313). Thus 31% of the variance of the TROG-2 is explained by Perceptual Reasoning IQ.

When a regression is calculated for both independent variables, Perceptual Reasoning and diagnosis, it can be seen that the total model significantly predicts the TROG-2 score (F(2, 47)=11.28, p<0.001, R^2=0.324). Thus 32% of the TROG-2 variance is explained by both variables. The non-significant change in R^2 of 0.011, shows that diagnosis is uniquely accountable for only 1% of the variance of the TROG-2 score. As expected, a non-significant amount of the variance of the TROG-2 score is accounted for by diagnosis, in keeping with the non-significant amount of variance of CELF-R receptive language score explained by diagnosis.

Table 5.12

The results of the hierarchical regressions and the change statistics for the language comprehension measures

Model	IV	DV	Parameters		Model Statistics		Change Statistics		
			β	p	R^2	df	ΔR^2	ΔF	p
2a	CELF-R Receptive Language Score	IQ	0.665	<0.001	0.442	1, 48	0.025	2.19	<0.146
2b		IQ, diagnosis	0.561; 0.189	<0.001; 0.146	0.467	2, 47			
3a	TROG-2	IQ	0.56	<0.001	0.313	1, 48	0.012	0.79	<0.38
3b		IQ, diagnosis	0.49; 0.128	<0.001; 0.379	0.324	2, 47			

5.3.5.3 Language production

When a regression for the CELF-R expressive language score is calculated with Perceptual Reasoning IQ remaining constant, Perceptual Reasoning IQ significantly predicts expressive language (F(1, 48)= 23.37, p<0.001, R^2=0.327). This means that 32.7% of the variance of expressive language is accounted for by Perceptual Reasoning. The results from the hierarchical regression for language production measures can be seen in Table 5.13 below.

When a regression is calculated for both independent variables, Perceptual Reasoning and diagnosis, it can be seen that both variables significantly predict for expressive language (F(2, 47)=16.75, p<0.001, R^2= 0.416). Thus 41.6% of the variance is explained by both variables. Given that almost 9% of the variance is uniquely accounted for by diagnosis (ΔR^2= 0.089, p<0.01), diagnosis is significantly associated with language production.

The Cookie Jar Theft Picture Complexity Index variance is significantly predicted by Perceptual Reasoning IQ (F(1,48)= 4.88, p<0.032, R^2=0.092).When both Perceptual Reasoning IQ and diagnosis are controlled for the variance of the Cookie Jar Theft Picture is not significantly accounted for by both variables (F(2, 47)= 2.41, p<0.85, R^2=0.093). The non-significant change in R^2 means that diagnosis does not uniquely account for any of the variance in the Cookie Jar Theft Picture, IQ on its own however does significantly account for 9.2% of the variance.

Table 5.13

The results of the hierarchical regressions and the change statistics for the language production measures

Model	IV	DV	Parameters		Model Statistics		Change Statistics		
			β	*p*	R ^{2}	*df*	Δ R^2	Δ F	*p*
4a	Expressive Language	IQ	0.572	<0.001	0.327	1, 48	0.089	7.14	<0.01
4b		IQ, diagnosis	0.376; 0.357	<0.007; 0.01	0.416	2, 47			
5a	Cookie Jar Theft Picture Complexity Index	IQ	-0.303	<0.032	0.092	1,48	0.001	4.86	<0.854
5b		IQ, diagnosis	-0.027; -0.031	<0.091; 0.854	0.093	2,47			

5.3.5.4 Naming

When a regression for the Boston Naming Test is calculated with Perceptual Reasoning IQ remaining constant, Perceptual Reasoning IQ significantly predicts for naming ($F(1, 48)= 15.18$, $p<0.001$, $R^2=0.24$). This means that 24% of the variance of expressive language is predicted by Perceptual Reasoning.

When a regression is calculated for both independent variables, Perceptual Reasoning and diagnosis, it can be seen that both variables significantly predict for expressive language ($F(2, 47)=9.88$, $p<0.001$, $R^2= 0.296$). Thus 30% of the variance is explained by both variables. A non-significant change in R^2 of 0.056, shows that 5.6% of the variance in naming is accounted for by diagnosis. The results from the hierarchical regression can be seen in table 5.14 below.

Table 5.14

The results of the hierarchical regressions and the change statistics for the Boston Naming test

Model	DV	Parameters		Model Statistics		Change Statistics		
		β	*p*	R^2	*df*	ΔR^2	Δ F	*p*
6a	IQ	0.49	<0.001	0.24	1, 48			
						0.056	3.73	<0.059
6b	IQ, diagnosis	0.334; 0.283	<0.027; 0.06	0.296	2, 47			

5.3.5.5. Regression controlling for school grade, sex, Perceptual Reasoning IQ and diagnosis

The school grade for the FASD group was, on average, two grades below the control group. The FASD group also had a far higher percentage of males than females. Thus these demographic differences in school grade and gender were controlled for. The model and change statistics for the nested regression of the total language score are shown in Tables 5.15 and 5.16 respectively.

When school grade and gender were controlled for it was found that gender did not significantly account for any of the total language variance. School grade however, uniquely accounted for almost 50% of the total language score variance ($F(2, 42)=20.81$, $p<0.001$, $R^2=0.498$). The explanation of the variance by the school grade resulted in the diagnosis variable having no significant explanation of the language variance.

There are significant positive correlations between grade and Perceptual Reasoning IQ and grade and the total language score as can be seen in table 5.17.

Table 5.15

The model statistics of the CELF-R total language score

Model	Dependent Variable	Parameters	
		β	*p*
1a	IQ	0.629	<0.001
1b	IQ, grade	0.442; 0.382	<0.001; 0.004
1c	IQ, grade, gender	0.462; 0.323; -0.014	<0.001; 0.004; 0.416
1d	IQ, grade, gender, diagnosis	0.439; 0.303;-0.095; 0.053	<0.005; 0.059; 0.476; 0.75

Table 5.16

The change statistics of the total language score

Model	Change Statistics		
	ΔR^2	ΔF	*p*
1b	0.114	17.15	<0.001; 0.004
1c	0.114; 0.008	16.47	<0.001; 0.004; 0.416
1d	0.114; 0.008; 0.001	16.37	<0.001; 0.004; 0.416; 0.75

Table 5.17

Correlations between school grade, Perceptual Reasoning IQ and Total language score

	School grade	Perceptual Reasoning IQ	Total Language Raw Score
School grade	1		
Perceptual Reasoning IQ	0.464	1	
Total Language Raw Score	0.587	0.619	1

* *p* <0.001; *N pairs* = 3

CHAPTER 6
DISCUSSION AND CONCLUSION

Studies on younger children with prenatal alcohol exposure have consistently found language deficits. Two studies have reported that these language difficulties were accounted for by the intellectual abilities of the children (Becker at al., 1990; McGee et al., 2009). Others have reported language difficulties in children with prenatal alcohol exposure, beyond that which may be accounted for by intellectual dysfunction (Hamilton, 1981; Shaywitz et al., 1981)

Language ability has been found to change as children mature. It has been found that the language abilities of children with prenatal alcohol exposure develop at a normal but slower rate to normally developing children. It was expected the older children in this study would have similar language deficits to the younger children. However, as the abilities of older children have been shown to change over time it would also not be unexpected for some differences to be found at an older age. As there are no available studies on the language abilities of adolescents with FASD, predictions were based on available data from assessments of younger children.

6.1 Summary of findings

The similar demographic data between the FASD and Control groups allowed for ease of comparisons between the groups. The FASD group had a greater number of males and was on average two school grades below the control group.

The first hypothesis that the FASD group would have significantly lower intellectual functioning than the control group was confirmed. All the IQ indexes were significantly lower in the FASD group than the control group. The FASD and control group had a significantly lower Verbal Comprehension IQ than Perceptual Reasoning IQ. The average full scale IQ of the Control group in this study was significantly lower than that of a normally developing child from the USA.

Correlations between the language tests show that the TROG-2 and CELF-R receptive language tests have strong positive correlations, whilst language production tests, namely the Cookie Jar Theft Picture and CELF-R expressive language tests, do not demonstrate strong correlations.

In agreement with the second hypothesis that language comprehension, production and word naming would be significantly worse in the FASD group, there was a markedly poorer performance of the FASD group on measures of language ability. However, the Cookie Jar theft picture test showed no significant differences between the groups.

Contrary to the third hypothesis that these language difficulties would be accounted for by intellectual functioning, it was found that when Perceptual Reasoning IQ was controlled for, diagnosis contributed significantly to the variance of language production and the CELF-R total language score. There was also a gender and school grade difference between the two groups. These demographic differences were of concern regarding the comparability of the sample. When controlling for these variables it was found that gender did not significantly contribute to the variance in language scores. School grade accounted for a substantial amount of language variance which completely masked the contribution of diagnosis to language variance. The relevance of this variable in the explanation of language ability is argued in section 6.4.2.1 below.

6.2 Application of international, translated tests to a South African sample

One of the challenges to psychological assessment in South Africa is the lack of standardised tests that ensure an assessment yields valid and reliable results (Foxcroft, Paterson, leRoux & Herbst, 2004). There are a number of hurdles that have prevented the development of local tests, specifically the very diverse cultural and language differences throughout the country (Paterson & Uys, 2005). Variation in neuropsychological test performance is documented as a factor of ethnic/cultural group membership, socio-economic and educational status, socio-political disadvantage and cognitive/educational limitations (Ardila, 1995; Skuy et al., 2000). This variation creates difficulties comparing scores across groups and creating norms applicable to all South Africans.

This study has faced similar challenges of a lack of culturally and language appropriate tests needed for this research. The first challenge of using international batteries is the inability to compare the South African scores to standardised norms. The IQ and language results from these translated tests are interpreted with caution in light of the differences between the normative groups and the study population. Despite these precautions it is difficult to draw firm conclusions from these translated tests. There is a great need for psychological tests to be developed for the South African context so that research results can be interpreted with greater confidence.

6.3 Intelligence tests

The WISC-IV intelligence test has been developed for USA populations and normed on a sample of USA children. The children in this study come from very different cultural, language and socio-economic backgrounds. The IQ results are interpreted in a context of cultural, language and educational differences between the USA norms and this sample.

The South African control group had a mean full scale IQ score over one standard deviation below the USA normed mean. This translated to the control group falling, on average, in the borderline range as classified by the WISC-IV norms. This large difference in full scale IQ is tangible considering that the children in the South African control group obtain similar IQ scores to children with FASD in the USA (Benton Gibbard et al., 2003; Becker et al., 1990; Carney & Chermak, 1991; Mattson & Riley, 1998; Shaywitz et al., 1981).

The poor performance by the control group on tests of intelligence was not unexpected as similar findings of typically developing South African children having below average to borderline ranges of intellectual functioning, as measured by the WISC-R, have been reported by Skuy et al. (2000).

The poor average performance of the control group may also be accounted for by a significant percentage of the control children having very low IQ scores. The full scale IQ for the control group ranged from 56 to 97. Ten of the children in the control group had full scale IQ scores of or below 70, so falling into a low IQ category. This high proportion of the control group falling within the low IQ range follows a similar pattern to the larger South African CIFASD-II sample.

The state of education of the South Africa population is poor in comparison to other developing countries. Education is understood to impact on IQ outcomes (Husen & Tuijnman, 1991; Reynolds, Chastain, Kaufman & McLean, 1987). The poor educational state of South Africa may be a possible contributing factor to the low IQ outcomes in South Africa and of this study. Significant numeracy and literacy shortcomings have been reported in South Africa (Chinapah et al., 2000; Department of Education, 2009). Numeracy and literacy are correlated to IQ (Kroesbergen, Van Luit, Van Lieshout, Van Loosbroek, & Van der Rijt, 2009; Bishop, 2001) and so the literacy and numeracy shortcoming of South Africa in general may offer a possible explanation for the low IQ scores of the participants in this study. The competencies of South African grade 4 learners in numeracy, literacy and life skills, measured by a monitoring and learning project in 12 African countries, was found to be exceptionally poor (Chinapah et al., 2000). In comparison to the other African countries South Africa scored the lowest average in numeracy (30.2%), the fifth lowest average in literacy (48.1%) and the third lowest average in life skills (47.1%). Similarly the reading and mathematics competencies of grade 6 learners, as assessed by the Southern African Consortium for Monitoring Educational Quality (SACMEQ), were found to be poor (Department of Education, 2009). South Africa achieved just under the mean SACMEQ score, ranking 8^{th} out of 14 developing countries for reading and 9^{th} for mathematics. The low measured IQ scores of the children in this study are thus less surprising in light of the poor overall education status of children in South Africa.

A combination of language, cultural differences and deficits in education, may offer a possible explanation for the poor performance of the control group on tests of intelligence. However, the effect of these factors may not comprehensively account for the very low IQ scores of the control group. The low socio-economic and historically disadvantaged background of the children may also have further contributed to the poor IQ scores. Children from impoverished backgrounds are understood to face multiple challenges from conception (Scott, Nonkin Avchen & Holloman, 1991). These challenges include poor prenatal care, inadequate nutrition, deficient medical care and insufficient education. Nutrition, poor health and non-optimal care-giving are well recognised to have a detrimental effect on cognitive development (Grantham-McGregor, Pollitt, Wachs, Meisels & Scott, 1999). Low maternal education (under 12 years) combined with low birth-weight, common in developing countries, is reported to result in children being 3.1 times more likely to be identified as requiring special education (Hollomon, Dobbins & Scott, 1998).

In summary, the low IQ scores for the control group may thus be attributed to a multitude of factors. Possible differences in the nutritional, care-giving and home environments between the groups have not been specifically accounted for in this study. All the children in this study received one meal per day at their school as provided for by the national school nutrition programme. None of the children in this study were in foster care, and all reside with family members - their biological parents, grandparents or other relatives. As such the children in this study come from similar backgrounds, and for the purpose of this study we have assumed negligible differences in these aspects. The cognitive levels of the FASD group can be understood to be impacted by the same factors as the control group and additionally by prenatal alcohol exposure.

Children with FASD are known to have deficits in intellectual functioning (Mattson & Riley, 1998). In light of the poor performance of the control group on tests of intelligence it was not surprising that the FASD group obtained significantly lower IQ scores. The mean full scale IQ of the FASD group was 55.16. This meant that the FASD group fell, on average, in the mild mental retardation range. The FASD group performed significantly worse than the control group on all the IQ composite scores. The poor outcomes on all the composite scores of IQ for the FASD group were expected as the literature reports similar deficits for children with FASD relative to control groups (Benton Gibbard et al., 2003; Becker et al., 1990; Carney & Chermak, 1991; Mattson & Riley, 1998; Shaywitz et al., 1981).

Verbal Comprehension IQ was significantly worse in the FASD group compared to the control group and so it was expected that the language ability of the children would similarly be poorer in the FASD group. The differences in general intelligence may, however, inform a difference in language performance. Thus all language differences between the groups have been analysed and understood in light of the differences in IQ.

There are conflicting accounts in the literature as to whether Verbal Comprehension IQ is poorer than Perceptual Reasoning IQ. In accordance with Conry (1990), Verbal Comprehension IQ was significantly lower than Perceptual Reasoning IQ in the FASD group. An unexpected outcome was that the *control* group also performed significantly worse on the Verbal Comprehension IQ index than the Perceptual Reasoning IQ index. The Perceptual Reasoning IQ and Verbal Comprehension IQ indexes are typically similar and a significant difference in performance between them often indicates a specific problem rather than general intellectual difficulties.

Again, the translation of the WISC-IV into Afrikaans may be cited as the problem resulting in this discrepancy, specifically as the translation of the tests into Afrikaans would result in greater problems in the Verbal Comprehension items than the Perceptual Reasoning items. However the very poor performance of South African children in literacy measures, compared to other developing countries (Chinapah et al., 2000; Department of Education, 2009), gives additional cause for concern that the verbal abilities of South African children are indeed significantly below their non-verbal abilities.

6.4 Language tests

6.4.1 Correlations

Multiple tests for the same language domain were used to allow for correlations between the tests to be computed. It was anticipated that the inter-correlations of translated tests would be strong as the tests were designed as a measure for the same domain.

The positive correlations between the language comprehension tests were strong and significant. These high correlations between the tests allow us to infer that the domain that the tests assess is similar. The TROG-2 and CELF-R receptive language tests are also designed similarly. Each test gives practice examples and has many items per test which allow the child a number of items to become familiar with the test format before the test is discontinued. Thus the correlation between these tests may be due to the tests measuring the same language domain and the similar format of the tests.

Contrary to the strong correlations between the language comprehension tests there were no significant correlations between language production tests. The Cookie Jar Theft Picture is a test of spontaneous language production. The CELF-R expressive language tests include a test of sentence repetition, sentence assembly and formulated sentences. These tests were expected to positively correlate as they are both a measure of language production.

The administration of CELF-R expressive language tests are more structured than the Cookie Jar Theft Picture. Thus a possible explanation for the lack of correlations between these tests is the format of the test administration. An alternate explanation of the lack of strong correlations may also be attributed to the different types of language production measured by these tests. As no strong correlations between these language production tests were found they cannot be interpreted as tests of a similar language function and are discussed individually in section 6.4.4 below.

6.4.2 Language abilities: controlling for school grade and gender

6.4.2.1 School grade as an unsuitable regression variable

The control and FASD groups differed on average by two school grades. When controlled for in the regression analysis, school grade accounted for almost 50% of the variance and completely masked the impact of diagnosis on the language test variance. A likely explanation is that grade is directly determined by both intellectual functioning and diagnosis. A child who was prenatally exposed to alcohol is likely to have lower intellectual functioning and due to this is more likely to fail and repeat grades at school. These variables account, somewhat, for the large grade difference. The strong positive correlations between these variables also support this explanation. In this study we aimed to tease apart the correlation of diagnosis and intellectual functioning on language ability. As grade appears to amalgamate these variables it is not a suitable variable to use to meet the aims of this study.

6.4.2.2 Impact of sex differences on language abilities

Typically developing females have higher verbal ability than males, whereas normally developing males have greater visual-spatial mathematical abilities (Vogel, 1990). General intellectual functioning is not significantly different between genders and the WISC has been designed to reduce major sex differences in the Full Scale IQ score (Weschler, 2004).

In children with learning disabilities however, it is reported that males with learning disabilities have higher verbal IQ scores than females (Vogel, 1990). Gender differences in general intellectual functioning and language abilities in children prenatally exposed to alcohol are unknown.

In this study, a greater percentage of males were in the FASD group than in the control group. This gender difference did not significantly account for any of the variance of general intellectual functioning or language abilities. The lack of gender differences in language ability may be due to no difference in the cognitive abilities between the genders being present. However, the sample studied is small and inferences to the larger population are tenuous. Alternatively the very low scores of all the children on the tests may mask any gender differences in language.

6.4.3 Language Comprehension

The language comprehension abilities in young children exposed to heavy doses of alcohol prenatally have been shown to be deficient in numerous studies (Becker et al., 1990; Carney & Chermak, 1991; Coggins et al., 2007; Hamilton, 1981; Iosub et al., 1981; McGee et al., 2009; Shaywitz et al., 1981). Consistent with these studies, the FASD group performed significantly worse than the control group on both tests of language comprehension: the CELF-R receptive language test and the TROG-2.

These results differ from the finding of Greene et al. (1990) of no significant differences between normally developing children and children prenatally exposed to alcohol. The low level of prenatal alcohol exposure almost certainly accounts for these differences.

These difficulties in comprehension were found to be accounted for by intellectual functioning. This is consistent with the finding of two studies that have controlled for both non-verbal intellectual functioning (Becker at al., 1981) and full scale IQ (McGee et al.,

2009) between children prenatally exposed to alcohol and normally developing children. This suggests that language comprehension in children with prenatal alcohol exposure develops normally, but at a slower rate than expected. Language comprehension, relative to intellectual functioning in children prenatally exposed to alcohol, does not represent a specific strength or weakness for these children.

The language comprehension of adolescents with prenatal alcohol exposure is similar to the pattern of comprehension abilities of younger children.

6.4.4 Language Production

6.4.4.1 CELF-R Expressive Language Measure

The greatest variation in language performance has been found on production measures, both within (Hamilton, 1981) and between studies. Our results showed significant differences between the FASD and control group on the CELF-R expressive language score. This is consistent with numerous findings that language production in children prenatally exposed to alcohol is impaired (Becker et al., 1990; Carney & Chermak, 1991; Coggins et al., 2007; Hamilton, 1981; McGee et al., 2009; & Shaywitz et al., 1981).

Again, these results differ from the study on children exposed to lower levels of alcohol prenatally (Greene et al., 1990) that found no significant differences between normally developing children and children prenatally exposed to alcohol.

In contrast to the finding on language comprehension, language production, as measured by the CELF-R, the FASD group showed impairment in language production over and above that which could be explained by non-verbal IQ. These results differ from those found by Becker et al. (1990) and McGee et al. (2009) that language production was accounted for by intellectual functioning. One possible explanation for the difference to McGee et al.'s (2009) results is that this study controlled for intelligence using the Perceptual Reasoning IQ whereas McGee et al. (2009) controlled for intelligence using the full scale IQ measure. The Perceptual Reasoning IQ measure was specifically selected in this study so that difference in language functioning between the groups would not be partialled out in the regression analyses, which we anticipated would occur using the full scale IQ. When the full scale IQ measure replaces the Performance IQ measure in this study, the amount of variance uniquely explained by diagnosis is no longer significant.

The study by McGee et al. (2009) also sampled a relatively high functioning sample of children exposed to high levels of alcohol prenatally. The mean full scale IQ for the FASD group in the McGee et al. study was 95 (*SD*= 9.45). As the sample of children from this study is functioning at a lower level intellectually, the difference in language performance may also be as a function of these general intellectual differences in the studies. Thus the differences in language production may only be apparent in children with FASD with severe intellectual deficits.

Our results suggest that language production in adolescents prenatally exposed to alcohol does not follow a normal developmental trajectory and is a significant weakness for children with FASD. The language production ability of the adolescents appears more impaired than that of younger children as reported by Becker et al. (1990) and McGee et al. (2009). These results give further evidence supporting the understanding that the language abilities of children change as they mature (Benton Gibbard et al., 2003; Carney and Chermak, 1991; & Coggins et al., 2007) and suggest that a deficit in language production may represent a specific characteristic of a neurobehavioural phenotype in adolescents of this FASD population.

Regression analysis of the subtests within the CELF-R expressive language score revealed that the greatest variation in language was accounted for by the recalling sentences subtest. This sample is too small to make conclusions based on these findings. However it does raise important avenues to explore in further research. The repetition abilities of younger children are known to be preserved (Benton Gibbard et al., 2003) but this finding suggests it may be impaired in older children. A study of a larger cohort would allow for the subtests within the CELF-R to be explored.

No differences between expressive and receptive language scores on the CELF-R were found when Perceptual Reasoning IQ was not controlled for. A possible explanation may arise from the large basement effect in the FASD group. The low scores of the children may have masked possible differences between the expressive and receptive scores before intellectual functioning was accounted for.

Language production is also understood to be more complex than language comprehension (Verhoeven & van Balkom, 2004). As children with FASD are understood to have difficulties with the complexity of tasks it is understandable that production may be more impaired than comprehension (Kodituwakku, 2007).

6.4.4.2 The Cookie Jar Theft Picture test

The second measure of language production, the Cookie Jar Theft Picture test yielded unexpected results. It was expected that the children in the FASD group would perform poorly as it has been found by Coggins et al. (2007) that on a test of narrative production, *Frog, Where are you?*, older children (aged 12) performed significantly worse than younger children (aged 8). We hypothesised that this finding of poorer narrative production in older children would follow into adolescence. It was also expected that as a test of language production, the results from the Cookie Jar Theft Picture would follow the trend of the other language production tests in this study, with the FASD group performing significantly worse than the control group. However no significant differences were found between the control and FASD group on this test. The Cookie Jar Theft Picture has not been used in other studies of language in FASD.

It could be argued, in light of these results, that adolescents exposed to alcohol prenatally may have similar narrative production to normally developing control children. However a qualitative appraisal of the test performance shows that the standardised scoring of the Cookie Jar Theft Picture may have favoured the FASD group. In general, the control group formulated a small number of short whole sentences with minimal content, resulting in a higher proportion of, grammatically correct, single clause sentences. In general the FASD group did not formulate grammatically correct sentences and joined most clauses with an "and". The FASD group also evidenced a lot of repetition throughout the test. This resulted in the FASD group having a higher number of multiple clause sentences that resulted in a higher complexity index. The transcriptions for a typical Cookie Jar Theft Picture performance for the FASD and control groups can be found in Appendix K.

The nested regression analyses showed that a higher Cookie Jar Theft Picture Complexity Index score was significantly predicted for by Perceptual Reasoning IQ. Diagnosis however, did not contribute uniquely to any variance in the Cookie Jar Theft Picture test.

These results suggest that the two groups' performance on this test was similar and that spontaneous language production in adolescents with FASD is equivalent to that of adolescents in the control group. However the different qualitative performance of the groups indicates that their performance is not similar and these results may be due to the inability of the test to measure the difference.

Unfortunately no other published literature on spontaneous language production in FASD adolescents could be found, so these results cannot be compared. Further research into the spontaneous language production of children of all ages with FASD is warranted. However the methodology should be chosen with caution as higher-level language skills are not well assessed by standard language tests as evidenced in this study and concluded by Coggins et al. (2007).

The development of one area of the brain is not isolated from the development of other areas, and the entire brain is interconnected (Anderson et al., 2001). Language development occurs simultaneously to executive functioning development. Children with prenatal alcohol exposure are known to have executive functioning difficulties (Kodituwakku, 2001). The ability to spontaneously generate words and ideas, one of the functions under the umbrella term of executive functioning, is deficient in children with prenatal alcohol exposure (Mattson & Riley, 1998) and may affect the narrative abilities of the children. This association should be considered in further research.

6.4.5 Production more impaired than comprehension

The evidence from the regression analyses suggests that language production is more impaired than language comprehension. This has not been found in the language studies of younger children prenatally exposed to alcohol, although production was found to be more variable than that of comprehension (Hamilton, 1981 as cited in Carney & Chermak, 1991). No differences between expressive and receptive language scores on the CELF-R were found when Perceptual Reasoning IQ was not controlled for. A possible explanation may arise from the large flooring effect in the FASD group. The low scores of the children may have masked possible differences between the expressive and receptive scores before intellectual functioning was accounted for. Language production is also understood to be more complex than language comprehension (Verhoeven & van Balkom, 2004). As children with FASD are understood to have difficulties with the complexity of tasks (Kodituwakku, 2007) it is understandable that production may be more impaired than comprehension; however the mechanisms accounting for production being more impaired than comprehension is unknown.

6.4.6 Naming

The majority of studies have focused on language comprehension and production. Few studies have investigated the naming abilities of children prenatally exposed to alcohol. Iosub et al. (1981) found significant naming impairments on the Peabody Picture Vocabulary test. In contrast, Becker et al. (1990), found no significant differences on the naming on confrontation subtest of the Clinical Evaluation of Language Functions. What Becker et al. (1990) did find was that the children prenatally exposed to alcohol took markedly longer to complete the test than the intellectually matched controls, suggesting a difficulty in this subtest.

Our results are consistent with those of Iosub et al. (1981), in that the FASD group had significant difficulties on the Boston Naming Test in comparison to the control group. These difficulties were accounted for by intellectual functioning. This suggests that naming is not a specific strength or weakness for children prenatally exposed to alcohol.

6.5 Implications of language dysfunction

Impairments in language functioning are understood to affect cognitive and behavioural functioning. Language is a necessary tool for academic and social achievement (Tomblin, Zhang, Buckwalter & Catts, 2000). Language delays have been correlated with academic underachievement (Hinshaw, 1992) and children with language impairment are more likely to be considered socially inept due to their interpersonal communication difficulties (Redmond & Rice, 1998). In addition, the processing limitations of children with language impairments may account for difficulties in non-linguistic domains, such as response inhibition (Im-Bolter, Johnson & Pascual-Leone, 2006).

Externalising behaviour is a common behavioural problem associated with receptive language impairments (Tomblin et al., 2000). A deficit in the verbal mediation component of self-regulation has been proposed to result in externalising behaviour problems, such as acting-out (Hinshaw, 1992). Ford, Farah, Shera & Hurt, 2007, reported that in 55 children prenatally exposed to cocaine both executive functioning and receptive language were associated with problem behaviour It was reasoned that the language impairment reduced the child’s ability to process and understand rules as well as resulting in a reduced ability to use internal language skills to guide and control behaviour.

Villemarette-Pittman, Stanford & Greve (2002) found language to be the most handicapping cognitive deficit in a group of 20 early adult impulsive aggressive individuals. However it was not language in isolation that resulted in these difficulties. An executive impairment in the language domain was concluded to result in a difficulty in verbal mediation that accounted for the resultant behavioural problems.

Early language deficits have also been shown to result in later behaviour problems. Hinshaw (1992) found a language delay at 3 years of age correlated with later behaviour problems at age 8. A relationship between language delay and problem behaviours at home and in the classroom was found at 3, 7, 9 and 11 years of age throughout a longitudinal study (Silva, Williams & McGee, 1987). Language impairments at age 5 were associated with anxiety disorders, internalising and externalising behaviour problems in early adulthood (Beitchmanet al., 2001). Stattin and Klackenberg-Larsson (1993) found that language difficulties at 5 years of age had a stronger relationship with future registered criminality than intelligence.

Children prenatally exposed to alcohol have been shown to have language difficulties which are likely to lead to other academic, cognitive and behavioural problems. These learning and behavioural difficulties associated with FASD may result in children with FASD becoming more susceptible to criminal behaviour.

In a sample of 415 adolescents and adults with FASD the lifespan prevalence of trouble with the law was 60% and of confinement in prison, psychiatric or detention centres 50% and 61% of the participants had disrupted school experiences (Streissguth et al., 2004). Similarly, a disproportionately large number of youth in the U.S.A juvenile justice system have been diagnosed with FASD (Fast, Conry, Loock, 1999). The behavioural difficulties and poor social outcomes associated with language difficulties may also be associated with the high prevalence of youth with FASD in conflict with the legal system.

This research suggests that language impairment is a facet of behaviour problems that may result in high risk outcomes such as social delinquency. The language deficits identified in adolescents with FASD in this study may predispose this FASD and possibly other populations with FASD in South Africa to high risk outcomes. Interventions focussed on language remediation may domino into changes in behaviour regulation and this would have a far greater impact than an effect confined to an improved academic performance.

6.6 Limitations

The major limitation of this study was the lack of standardised Afrikaans language tests for this age group in South Africa. The lack of normative data made age appropriate comparisons difficult. To overcome this limitation, multiple tests assessing language comprehension and language production were used. A control group was used to allow for comparisons in place of normative data.

This study is novel in its assessment of language in adolescents with FASD. There is no available published research literature on language in children with FASD in this age group and the expectations and outcomes of this study could only be based on, and compared to studies on younger children.

The sample size of this study, although large compared to other studies assessing the language of children with FASD did not allow for comparisons within the diagnostic spectrum. It was expected that children exposed to heavier doses of alcohol prenatally would experience greater difficulties in language functioning than children exposed to lower levels of alcohol but this sample was too small to differentiate between the language ability of children with FAS and partial FAS. Similarly the small sample size did not allow for analysis of the subtests within the CELF-R.

6.7 Conclusions and Recommendations

This study established that language is impaired in adolescents with FASD. As the first known study of this age group many more questions are raised than answered. There is significant scope and need for further research. This study has found that changes in the language ability of children with FASD may emerge as they get older; language production in adolescents with FASD is more impaired than what can be accounted for by intellectual functioning. The factors that underpin this finding are unknown and should be investigated. The language production impairment may be as a result of epigenetics, or due to environmental influences on the children. Longitudinal studies assessing language development, following prenatal alcohol exposure, from childhood into adolescence may provide insight into the developmental trajectory of comprehension and production and how and why they may differ. Environmental factors, such as maternal interaction, may influence language development and should be investigated.

Language has been consistently reported as impaired in the literature on younger children. This study determined that these impairments continue into adolescence. The mechanism behind this impairment is unknown. Results from imaging studies on children with FASD have shown brain anomalies in the language processing areas of the brain. These anomalies and their potential correlation to language impairments should be investigated. Longitudinal studies that examine the development of language processing areas of the brain may provide an explanation for the worse performance in language production than comprehension.

Few studies have examined narrative production in FASD. This is an inherently difficult undertaking as standard neuropsychological tests are not adequate measures of this domain. The Cookie Jar Theft Picture test, using standard administration and scoring, yielded unexpected results with no impairment found in the FASD group, whereas other studies have found impairments to emerge as children develop. Further research into this domain is warranted; however care as to the test choice should be taken as the findings from this study were attributed to the standard scoring of the Cookie Jar Theft Picture being an inappropriate measure.

The impact of the low resource setting on these findings also raises the question as to whether the findings from this study are generalisable to other multicultural settings. Language production could represent a core deficit and a characteristic of a neurobehavioural phenotype in adolescents with FASD in South Africa. Further research in less impoverished sites with higher functioning adolescents would elucidate whether language production is a core deficit in FASD in general.

Future studies should aim to use a larger sample to allow for the exploration of language differences between children within the FASD spectrum. A larger sample would also allow for analysis of the subtests within the CELF-R test.

There is a great need for standardised neuropsychological tests appropriate for the South African population. For this research a number of language and intelligence tests were translated and adjusted to be relevant for the local population. Further research to validate and normalise the translated tests would be an asset to South African clinical and research work.

The finding of language impairment in adolescents with FASD has numerous implications. An improved understanding of the language impairments in people with FASD as well as research into the mechanisms underlying the deficits can inform intervention strategies. Teaching and testing of children with FASD through a verbal based medium would not elicit the best outcomes. Support for teachers of learners with FASD should be developed as many classrooms in the Wellington site of this study have a substantial proportion of children with FASD but use a verbal based medium of teaching. Language impairments are also associated with other cognitive and behavioural deficits, the significant language impairments found in these adolescents with FASD may negatively affect their social functioning and behavioural regulation, which in turn, may place them at risk for further long-term problems.

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Appendix A

Revised IOM criteria for diagnosis within the FASD continuum (Hoyme et al., 2005)

Diagnostic criteria for FAS or PFAS (with or without confirmed maternal alcohol exposure):

FAS requires all features A–C

PFAS requires A and: B or C or evidence of a complex pattern of behavior or cognitive abnormalities inconsistent with developmental level and that cannot be explained by genetic predisposition, family background, or environment alone [see ARND]).

A. Evidence of a characteristic pattern of minor facial anomalies, including at least two of the following:

1. Short palpebral fissures (less than or equal to the 10th centile)
2. Thin vermilion border of the upper lip
3. Smooth philtrum (score 4 or 5 on the lip/philtrum guide)

B. Evidence of prenatal and/or postnatal growth retardation: height or weight less than or equal to the 10th centile

C. Evidence of deficient brain growth or abnormal morphogenesis, including one or more of the following:

1. Structural brain abnormalities
2. Head circumference less than or equal to the 10th centile

Diagnostic criteria for alcohol-related effects (ARBD and ARND):

A diagnosis in these categories requires a confirmed history of prenatal alcohol exposure. ARBD requires the characteristic facies as above plus specific congenital structural defects (including malformations and dysplasias) in at least one organ system (if the patient displays minor anomalies only, at least two must be present). This category assumes the subject to have normal growth and intellectual/behavioral characteristics. ARND assumes the subject to have normal growth and structure and at least one of the following (A or B):

A. Evidence of deficient brain growth or abnormal morphogenesis, including one or more of the following:

1. Structural brain abnormalities
2. Head circumference less than or equal to the 10th centile

B. Evidence of a complex pattern of behavior or cognitive abnormalities inconsistent with developmental level and that cannot be explained by genetic predisposition, family background, or environment alone. This pattern includes: marked impairment in the performance of complex tasks (complex problem solving, planning, judgment, abstraction, metacognition, and arithmetic tasks); higher-level receptive and expressive language deficits; and disordered behavior(difficulties in personal manner, emotional lability, motor dysfunction, poor academic performance, and deficient social interaction)

Appendix B

Consent and assent forms for the parent and participants in the CIFASD-II study

CONSENT TO PARTICIPATE IN A RESEARCH PROJECT

An analysis of the learning process and behaviour in South African and American children with Fetal Alcohol Spectrum Disorders (FASD)

Dear Parent

Dr Colleen Adnams and her team from the University of Cape Town are busy conducting a research project. The goal of the project is to understand the development and learning process of school-children who have been exposed to alcohol before birth.

This research will be conducted in Wellington as well as in certain states in the United States of America (USA). This study is funded by the government of the USA's National Institute on Alcohol Abuse and Alcoholism (NIAAA). One Thousand children between 8 and 16 years of age and their families will be involved in this study. All these families will come from Wellington as well as the certain states in the USA. For the research outcomes to be comparable there are children in this study that have not been exposed to Alcohol before birth.

PROSEDURE

If you decide to participate in this study, and give your consent for your Child to participate in this study, the following shall happen:

Your Child will be evaluated on a range of tests that examine his/her aptitude in a range of areas. You, your Child and your Child's teacher will be asked to fill out a number of questionnaires about your Childs' behaviour and emotions. You may also be asked to take part in face-to-face interview. You and your Child can decline to answer a question or withdraw from the study at anytime.

The research will begin in June 2009 and run until 2012. The testing of your Child will take place during school hours in our research office in Wellington. Your Child will be transported by a reliable member of the research team.

Information about the tests and the progress your child has made in this research will shared with you, your Child and your Childs' school.

VIDEO FILMING, AUDIO-RECORDING AND CONFIDENTIALITY

We also ask your permission to film your Child during the testing. The identity of your Child will be maintained. Only a study number assigned to your Child will be used in the research. You can decline the filming and recording of your Child.

RISKS AND ACCIDENTS

In the case of an accident, wherein you or your Child was involved during the time you were participating in the study, the study will cover the costs through insurance.

BENEFITS

There may or may not be direct benefits to you or your Child by participating in this study. We hope that what is learned through this study will make people more aware of the interventions needed for children that have been exposed to alcohol before birth.

CONFIDENTIALITY

Any information that you give during this study will be reliably handled. Your name will not be used in any publications about this study.

COST OF THE STUDY

There will be no cost incurred to you connected to this study. You will not be accountable for any of the costs of the tests or procedures connected with this research.

PARTICIPATION

Your participation in this study completely voluntary. You have the right to not take part in the study or to withdraw from the study at any time.

QUESTIONS

If you have any questions in connection with this study you can contact the principal investigator or project coordinator at the University of Cape Town.

Dr Colleen Adnams - UCT Tel. 021 404-2173

Project Coordinator - UCT Tel. 021 404 5385

If you have any ethical questions in connection with the project you can contact the University of Cape Town Research Ethics Committee:
Dr. Mark Blockman, Head : UCT Ethics Committee Tel 021 406-6942.

CONSENT

If your child is older than seven years he/she must also give his/her assent to partake in this study and the rest of this form must be completed. If your child is younger than seven years this part of the form does not need to be completed.

____

Childs' name (Print)

______________________________ ____________________

Childs' signature Date

You, as parent, give permission for your child to participate in this study. Your signature hereunder shows that you and your child have decided to participate in this study and that you have read the above information of that is was read to you.

I have read the above information (or have had it read to me). I have had the opportunity to ask questions and all my questions have been answered to my satisfaction. By signing this consent form, I give consent for my and my child's participation in this study.

____

Childs' name (type or print)

____

Parent/Guardians' Name

________________________________ _________________

Parent/Guardians' Signature Date

Address ____________________________Telephone number__________________

____

Witness **if one is present** (Type or print)

_____________________________ ________________

Signature of Witness Date

If a researcher has explained the information:

I have explained all information and answered all questions related to this research project to the child and his/her guardian. I believe that he/she has understood the information in this consent form and has voluntarily decided to participate in the study.

____ ________________

Name and signature of research team member Date

Appendix C

Consent and assent forms for the additional language tests

CONSENT TO ADMINISTER ADDITIONAL TESTS

Language production, comprehension and word naming in adolescents with fetal alcohol spectrum disorder.

Dear Parent

A study on the language functioning of children that have been exposed to alcohol before birth is being conducted at the University of Cape Town. This study forms a part of the study being conducted by Dr Colleen Adnams and her team. This is the study to which you have already consented. The goal of the project is to understand the language deficits that may follow exposure to alcohol before birth. Information from the tests your child has already done will be used in this study. For this study additional tests on language also need to be conducted on some of the children that are participating in Dr Adnams' study.

This test will be conducted in Wellington. Fifty children between 14 and 16 years of age that are involved in Dr Adnams' study will be involved in this study. For the research outcomes to be comparable there are children in this study that have not been exposed to Alcohol before birth.

PROCEDURE

If you decide to give your consent for your Child to participate in this study, the following shall happen:

Your Child will be evaluated on some tests of language. These tests take approximately 80 minutes to administer. This test will be conducted at your Childs' school, during school hours, in July, August or Sepetember 2010.

CONFIDENTIALITY

The study number assigned to your Child in the study by Dr Adnams will be used in this research. Any information given during this test will be reliably handled. Your Childs' name will not be used in any publications.

BENEFITS

There may or may not be direct benefits to you or your Child by participating in this test. We hope that this test will allow us to better understand the language so that interventions for children that have been exposed to alcohol before birth can be created.

COST OF THE STUDY

There will be no cost incurred to you connected with this. You will not be accountable for any of the costs of the tests or procedures connected with this test.

PARTICIPATION

Your Child's participation in this test is completely voluntary. You have the right to not take part in the test or to withdraw from the test at any time.

QUESTIONS

If you have any questions in connection with this test you can contact the researcher responsible for conducting this test at the University of Cape Town.

Claire Corbett - UCT Tel. 021 404 5385

If you have any ethical questions in connection with the project you can contact the University of Cape Town Research Ethics Committee:

Dr. Mark Blockman, Head : UCT Ethics Committee Tel 021 406-6942.

CONSENT

Your child must also give his/her assent to participate in this study.

____

Childs' name (Print)

________________________________ ____________________

Childs' signature Date

You, as parent, give permission for your child to participate in this test. Your signature hereunder shows that you and your child have decided to participate in this test and that you have read the above information of that is was read to you.

I have read the above information (or have had it read to me). I have had the opportunity to ask questions and all my questions have been answered to my satisfaction. By signing this consent form, I give consent for my and my child's participation in this test.

____

Childs' name (type or print)

____

Parent/Guardians' Name

_________________________________ ________________

Parent/Guardians' Signature Date

Address____

Telephone number_________________________

____

Witness **if one is present** (Type or print)

______________________________ ________________

Signature of Witness Date

If a researcher has explained the information:

I have explained all information and answered all questions related to this test to the child and his/her guardian. I believe that he/she has understood the information in this consent form and has voluntarily decided to participate in the test.

____ ________________

Name and signature of research team member Date

Appendix D

Table 4.1

CIFASD-II Neuropsychological battery

Test	Sub-test
Weschler Intelligence Scale for Children-Fourth Edition (WISC-IV)	
	Block Design
	Similarities
	Digit Span
	Picture Concepts
	Coding (and Coding Recall)
	Vocabulary
	Letter-Number Sequencing
	Matrix Reasoning
	Comprehension
	Symbol Search
	Arithmetic Process Approach Parts A&B
	Written Arithmetic
	Letter Span
	Visual Digit Span
	Letter-Number Sequencing PA
Delis-Kaplan Executive Function System (D-KEFS)	
	Trail Making
	Verbal Fluency
	Tower
	Design Fluency
Cambridge Neuropsychological Test Automated Battery (CANTAB)	
	Motor Screening
	Big/Little Circle
	Simple Reaction Time
	Delayed Matching to Sample
	Spatial Working Memory
	Choice Reaction Time

Appendix E

Table 4.2

The language test battery and the respective domains the tests measure

Test	Domain
CELF-R (Expressive Language Scale) Formulated sentences Recalling sentences Sentence assembly	Expressive language
Cookie Jar Theft Picture	Expressive language
TROG-2	Receptive language
CELF-R (Receptive Language Scale) Oral directions Word classes Semantic relationships	Receptive language
Boston Naming Test	Word Naming

Appendix F

Examples of translation differences

CELF-R Sentence Assembly

Item 16 was not directly translated from English to Afrikaans. Changes were made to accommodate grammatical differences (the double negative in Afrikaans) whilst maintaining the number of blocks in the stimulus and number of potential sentences that could be assembled from the blocks of words from the stimulus.

English item

Stimulus:

the lamp	the woman	the table	put	didn't	on

Potential sentences:

_____ a) The woman didn't put the lamp on the table

_____ b) Didn't the woman put the lamp on the table?

1	0	NR

Afrikaans item

Stimulus

die lamp	die vrou	op die tafel	gesit nie	het	nie

Potential sentences

_____ a) Die vrou het nie die lamp op die tafel gesit nie

_____ b) Het die vrou nie die lamp op die tafel gesit nie?

1	0	NR

CELF-R Semantic relationships

Item 28 was not directly translated into Afrikaans to allow for cultural differences. The number of holidays used in Afrikaans item is reduced as there are few holidays commonly known by the population being tested.

English item

28. Thanksgiving comes between:

_____ a) Labour day and Valentines day

_____ b) Easter and the 4th of July

_____ c) Halloween and Christmas

_____ d) New Year's and memorial day

Afrikaans item

28. Nuwe Jaar kom tussen

_____ a)	Kersfees en Valentynsdag	_____ c)	Paasfees en Kersfees
_____ b)	Valentynsdag en Paasfees	_____ d)	Kersfees en Paasfees

CELF-R Word classes

Item 22 was altered such that the word pair "*ahead; front"* became opposites ahead and behind (as in other items) as the translation into Afrikaans would result in the same word, *voor*, for both ahead and front.

English item

22. amoung ahead until front

Afrikaans item

22. tussen voor tot agter

Appendix G

Table 4.3

Neuropsychological test battery administration sequence

Standardised Test		Sub-test
WISC-IV		
		Block Design
		Similarities
		Digit Span
		Picture Concepts
		Coding (and Coding Recall)
		Vocabulary
		Letter-Number Sequencing
		Matrix Reasoning
		Comprehension
		Symbol Search
		Arithmetic Process Approach Parts A&B
		Written Arithmetic
	1 Hour rest break	
CANTAB		Motor Screening
		Big/Little Circle
		Simple Reaction Time
		Delayed Matching to Sample
		Spatial Working Memory
		Choice Reaction Time
	1 Hour rest break	
WISC-IV		Letter Span
D-KEFS		Trail Making
WISC-IV		Visual Digit Span
D-KEFS		Verbal Fluency
D-KEFS		Tower
WISC-IV		Letter-Number Sequencing PA
D-KEFS		Design Fluency

Appendix H

Table 4.4

Language test battery administration sequence

Test	Subtest
Boston Naming Test	
TROG-2	
Cookie Jar Theft Picture	
CELF-R	Oral directions
	Recalling sentences
	Formulated sentences
	Word classes
	Semantic Relationships
	Sentence Assembly

Appendix I

Table 4.5

Dependent variables: List of outcome measures

Test	Subtest	Outcome measure
WISC-IV		Full Scale IQ
		Verbal Comprehension composite score
		Perceptual Reasoning composite score
Boston Naming test		Total Correct
TROG-2		Total Correct
Cookie Jar Theft Picture		Complexity Index
CELF-R		Total Language Score
		Expressive Language Score
		Receptive language Score
	Oral directions	Raw Score
	Recalling sentences	Raw Score
	Formulated sentences	Raw Score
	Word classes	Raw Score
	Semantic relationships	Raw Score
	Sentence assembly	Raw Score

Appendix J

Table 5.4

A summary of the TROG-2 and CELF-R standard scores.

Variable	Group	Mean	SD	Range
	Control	75.6	10.21	55-95
TROG2 Raw	FASD	67.04	11.01	55-85
	Control	7.44	3.12	3-14
Oral Directions	FASD	5.12	2.666	3-13
	Control	6.26	2.11	3-12
Recalling sentences	FASD	4.08	1.441	3-7
	Control	5.84	3.67	3-14
Formulated sentences	FASD	4.08	2.532	3-14
	Control	6.4	2.5	3-11
Word Classes	FASD	4.4	1.7	3-9
	Control	5.76	2.006	3-11
Semantic Relationships	FASD	4.16	1.313	3-7
	Control	7.48	2.4	5-13
Sentence Assembly	FASD	5.24	1.69	4-11
	Control	76.28	15.665	50-101
CELF Receptive Language Score	FASD	62.04	12.96	50-89
	Control	76.76	13.575	59-110
CELF Expressive Language Score	FASD	60.56	12.748	50-104
	Control	74.8	14.265	53-100
CELF Total Language Score	FASD	59.96	11.706	50-93

Appendix K

Cookie Jar Theft Picture transcription for a 16 year 1 month old male prenatally exposed to alcohol, in the equivalent of grade 9 in a special school.

Die seun prober koekies vat maar die stoel is besig om oor te val. / Die Ma was skottlegoed maar die water loop oor. / Die meisie wil ook a koekie hê. / Die vrou kyk uit die venster / sy het klaar twee koppies en ‘n bord afgedroog. / Daar is baie koekies in die bakkie. / Die huis is groot. / Die vrou het ‘n rok aan / die seun dra ‘n boek en skoene / die meisie het ‘n rok aan. / Die vrou werk in die huis. / Die gordyn en die vensters is oop. / Daar is kasse in die huis / daar is ‘n kraan in die huis.

Translation:

The boy is trying to steal cookies but the chair is busy falling over. The mother is washing the dishes but the water is over-flowing. The girl also wants a cookie. The lady is looking out the window; she has already dried two cups and a plate. There are many cookies in the jar. The house is big. The lady is wearing a dress, the boy is wearing a pair of shorts and shoes, the girl is wearing a dress. The lady is working in the house. The gate and windows are open. There are cupboards in the house, there is a tap in the house.

Cookie Jar Theft Picture transcription for a 15 year 7 month old female, in grade 9, from the non-exposed control group.

Die vrou droog water af./ Die seun haal koekies uit die kas. / Die water loop uit die wasbak. / Die stoel is besig om the val. / Die dogtertjie kyk na hom. / Die vrou staan in die water. / Die gordyn is oop. / Die venster is ook oop, / en die kraan van die wasbak is ook oop. / Die seun en die dogterjie is besig om die koekies te vat sonder dat die vrou weet.

Translation:

The lady is drying off the water. The boy is taking cookies out the cupboard. The water is running over the sink. The chair is falling over. The girl watches him. The lady stands in the water. The gate is open. The window is also open and the tap from the sink is open. The boy and girl are taking cookies without the lady knowing.