Sensory Integration Difficulties and Dysfunctions in Children with Fetal Alcohol Spectrum Disorders

by

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Sensory Integration Difficulties and Dysfunctions in Children with Fetal Alcohol Spectrum Disorders (FASD).

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In the words of Eloise McGraw….

“To all children

who have ever felt *different”*

DECLARATION

I, …………………………………., declare that the

Master’s Degree research dissertation entitled

Sensory Integration Difficulties and Dysfunctions in Children

with

Fetal Alcohol Spectrum Disorders

that I herewith submit for the Master’s Degree qualification

Magister in Occupational Therapy at the University of the Free State,

is my independent work,

and that I have not previously submitted it for a qualification

at another institution of higher education

…………………………………..

Maritha du Plooy

"I continue to believe that if children are given the necessary tools to succeed, they will succeed beyond their wildest dreams!"

David Vitter, U.S. Senator

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ACRONYMS AND ABBREVIATIONS

AOTA	American Occupational Therapy Association
ARBD	Alcohol-related birth defects
ARND	Alcohol-related neurodevelopmental disorders
ASI	Ayres Sensory Integration
BRAM	Bonnievale, Robertson, Ashton, Montagu
CNS	Central nervous system
CWED	Cape Winelands Education District
FARR	Foundation for Alcohol Related Research
FAS	Fetal Alcohol Syndrome
FASD	Fetal Alcohol Spectrum Disorders
FASER-SA	Fetal Alcohol Syndrome Epidemiological Research - South Africa
HSREC	Health Sciences Research Ethics Committee
IOM	Institute of Medicine
ND-PAE	Neurobehavioural disorder associated with prenatal alcohol exposure
NIAAA	National Institute on Alcohol Abuse and Alcoholism
PFAS	Partial Fetal Alcohol Syndrome
PRN	Postrotary Nystagmus
SI	Sensory integration
SIPT	Sensory Integration and Praxis Tests
SP	Sensory Profile
SPSC	Sensory Profile School Companion
WCED	Western Cape Education Department
WISC-IV	Wechler Intelligence Scale for Children

CONCEPT CLARIFICATION

In the following section, terms will be defined as they will be used in this project.

Adaptive response	An appropriate action in which the individual responds successfully to environmental demands. Good sensory integration is a prerequisite for adaptive responses (Ayres, 2005, p. 199).
Alcohol-related birth defects (ARBD)	Specific physical anomalies resulting from confirmed prenatal alcohol exposure. These may include heart, skeletal, vision, hearing and fine/gross motor problems (Blackburn, Carpenter, & Egerton, 2012, p. 102).
Alcohol-related neurodevelopmental disorders (ARND)	Children with these disorders exhibit central nervous system (CNS) damage resulting from a confirmed history of prenatal alcohol exposure. This may be demonstrated in learning difficulties, poor impulse control, poor social skills and problems with memory, attention and judgement (Blackburn, Carpenter, & Egerton, 2012, p. 102).
Children	Children are vulnerable human beings under the age of 18; only as adults will they be able to protect themselves (Humanium, 2008).
Foundation for Alcohol Related Research (FARR)	The Foundation for Alcohol Related Research (FARR) is a non-governmental organization doing extensive research on Fetal Alcohol Spectrum Disorders (FASD) and Fetal Alcohol Syndrome (FAS) in South Africa. Dedicated also to FASD prevention, they conduct numerous community programmes (FARR , n.d.).
Fetal Alcohol Syndrome Epidemiological Research - South Africa (FASER-SA)	FASER-SA was established as a collaborative endeavour of the University of North Carolina, the University of New Mexico, the Faculty of Medicine and Health Sciences of Stellenbosch University, and the Medical Research Council of South Africa. The mission of the group is to reduce the prevalence of FASD through comprehensive prevention activities developed in collaborative research with local communities. (Medicine and Health Sciences - Research Department of Psychiatry, 2013).

Fetal Alcohol Spectrum Disorders (FASD)	An umbrella term that encompasses the range of possible effects of prenatal exposure to alcohol. These effects may include physical, cognitive, behavioural, and/or learning disabilities with lifelong implications (Blackburn, Carpenter, & Egerton, 2012, p. 104). (The spelling “fetal” is used in most South African and international literature on FASD; it will therefore be used in this dissertation.)
Fetal Alcohol Syndrome (FAS)	A term used to describe a specific identifiable group of children with FASD who all share certain characteristics: a specific set of facial features, central nervous system dysfunction and often growth deficiency and a scattering of other birth defects resulting from confirmed prenatal maternal alcohol exposure (Blackburn, Carpenter, & Egerton, 2012, p. 104).
Occupations	Daily life activities in which people engage (American Occupational Therapy Association (AOTA), 2014, p. S6).
Partial Fetal Alcohol Syndrome (PFAS)	A child with PFAS exhibits some, but not all of the physical signs of FAS and also has learning and behavioural difficulties due to CNS damage (Blackburn, Carpenter, & Egerton, 2012, p. 105).
Sensory input	Streams of electrical impulses flowing from the sensory receptors in the sense organs of the body to the spinal cord and/or brain (Ayres, 2005, p. 201).
Sensory integration	“The organization of sensory input for ‘use’. ‘Use’ may be a perception of the body or the world, an adaptive response, a learning process or the development of some neural function. Through sensory integration many parts of the nervous system work together to ensure that a person can interact with the environment effectively with appropriate success and satisfaction” (Ayres, 2005, p. 201).

Teratogen	Any substance, such as alcohol or drugs, or condition, such as measles, capable of causing damage to the development of a fetus, resulting in deformed fetal structures. Alcohol causes birth defects and brain damage, resulting in neurobehavioural problems in exposed offspring and is thus classified as teratogenic (Blackburn, Carpenter, & Egerton, 2012, p. 106).

SUMMARY

Sensory Integration Difficulties and Dysfunctions in Children with Fetal Alcohol Spectrum Disorders

Introduction:

The effects of Fetal Alcohol Spectrum Disorders (FASD) are devastating and enduring, impacting on performance skills and limiting successful participation in activities of daily life. The high prevalence of FASD in the Western Cape, together with poor results in school performance, are reasons for concern.

Symptoms of sensory integration dysfunctions are reported as challenges faced by children with FASD. The aim of this study was to describe sensory integration difficulties and dysfunctions experienced in a group of children aged five to eight years old from the Cape Winelands Education District (CWED), both with and without a diagnosis of FASD.

Objectives:

The main objectives were to investigate sensory modulation, as measured by the Sensory Profiles, and sensory processing and practic abilities, as measured by the SIPT, of an identified group of children with FASD and a matched controlled group without a diagnosis of FASD. A final objective was to identify patterns of sensory integration difficulties and dysfunctions, to describe these findings, and to draw conclusions about the distinctive patterns of sensory integration difficulties and dysfunctions among children with FASD.

Methodology:

Thirty children with FASD (cases) were compared with a matched sample (controls) without FASD. Standardized measuring instruments, the Sensory Profile (SP), Sensory Profile School Companion (SPSC) as well as the Sensory Integration and Praxis Tests (SIPT), were used in this quantitative, observational, analytical study.

Results:

Higher percentages of cases than controls experienced challenges in 22 of the 23 categories of the Sensory Profile, with significant differences in nine of the categories. According to the results of the Sensory Profile School Companion, cases experienced more challenges in all 13 of the categories, with significant differences in three. Comparison of the mean scores of the matched sample showed significant differences in eight categories of the SP and three of the SPSC, with the cases performing poorer. The highest percentages of cases in the Definite Difference and Probable Difference ranges were recorded for Sensory Seeking (90% (n=18)), Inattention/Distractibility (85% (n=17)), Auditory Processing (70% (n=14)), Multisensory Processing (85% (n=17)), Registration (62% (n=19)) and Avoiding (62% (n=19)). The identified sensory processing difficulties seemed to occur in multiple sensory systems, ranging from under- to over-responsiveness. It is important to note that sensory processing difficulties were also reported for the controls, although at lower percentage levels.

The paired t-test results indicated significant differences between the paired cases and controls in eleven of the seventeen test items of the SIPT, with the cases performing poorer. According to the results, 14 (46.7%) of the cases adhere to the criteria recognised as a Visuo- and Somatodyspraxia pattern of dysfunction.

The difficulties and dysfunctions identified by the Sensory Profiles and SIPT could contribute to the challenges experienced in occupations of the children with FASD.

Conclusion:

Although further research is needed, the results from this study confirm sensory integration difficulties and dysfunctions of the FASD population impacting on their daily functioning and performance. Consistencies with previous research results were found.

The outcome of this study has clinical importance for occupational therapists working with children with FASD, and for educators and caregivers in terms of intervention, education and caring.

Keywords:

Fetal Alcohol Spectrum Disorders, occupational therapy, sensory integration, occupations

CHAPTER 1

INTRODUCTION AND ORIENTATION TO RESEARCH

1.1 INTRODUCTION

The prevalence rate of Fetal Alcohol Spectrum Disorders (FASD) in the Western Cape, South Africa, is alarmingly high. Numerous studies on FASD have been conducted and reported on in the Western Cape from as early as 2000 (May, et al., 2000, p. 1905). Currently, the main researchers on FASD in the Western Cape are FASER-SA (Fetal Alcohol Syndrome Epidemiological Research - South Africa) and FARR (Foundation for Alcohol Related Research). Research done by FASER-SA is focused on two geographical areas, the Wellington and Bonnievale, Robertson, Ashton, Montagu (BRAM) areas. Their current research focus includes the following:

- Community surveys
- Prevention activities
- Brief interventions and assessment of the risk of having a child with FAS, conducted at antenatal clinics
- Assessment and screening of babies, with follow-up assessments
- In-school examination of Grade 1 children by dysmorphologists.

A Grade 1 schools screening study was carried out in the BRAM area during 2009 by FASER-SA as part of the *Fetal Alcohol Syndrome Prevention in South Africa: A Trial of the Institute of Medicine Model*. The study reported a prevalence rate of 18 to 26% for FASD, which is the highest rate ever documented in a general population study (May, et al., 2016, p. 207 & 216; Marais, 2017). A further Grade 1 school study, in a similar kind of community in the Western Cape (Witzenberg area), carried out by FARR, reported a FASD prevalence rate of 9.6%, with numbers as high as 18.2% in one of the towns (Olivier, 2013, p. 7). These two studies alone offer confirmation of the alarmingly high prevalence rate of FASD among children in these communities.

Although the diagnosis of Fetal Alcohol Syndrome (FAS) was only formalised in 1973 (Jones & Smith, 1973, p. 999), the effect of alcohol on the developing brain was described as early as 1968 (Lemoine, Harousseau, Borteyru, & Menuet, 2003, p. 132). Among other features, the authors described characteristics such as growth retardation and psychosomatic alterations, with defining facial features such as a low arched forehead, flattened nasal base, short upturned nose, retracted upper lip, and poorly implanted deformed ears with horizontal upper edges (Lemoine, Harousseau, Borteyru, & Menuet, 2003, p. 132). Alcohol is a neurobehavioral teratogen with a disruptive effect on fetal development, especially in early brain development (Stratton, Howe & Battaglia, in Jirikowic, Olson, & Kartin, 2008a, p. 118). Exposure to alcohol before birth may result in fetal alcohol syndrome (FAS), with very specific characteristics or a continuum of effects, including physical, mental, behavioural and learning disabilities, with possible lifelong implications (Jirikowic, Olson, & Kartin, 2008a, p. 118; Bertrand, Floyd, & Weber, 2005, p. 2). This continuum falls under the umbrella of FASD and includes fetal alcohol syndrome (FAS), partial fetal alcohol syndrome (PFAS), alcohol-related neurodevelopmental disorders (ARND), and alcohol-related birth defects (ARBD) (May, Blankenship, & Marais, 2013, p. 820).

The brain damage associated with prenatal exposure to alcohol leads to a range of deficits, including cognitive, motor, executive function, language, visuo-spatial, learning, auditory processing, sensory processing and attention deficits (Mattson & Riley, 1998, p. 291; Bertrand, Floyd, & Weber, 2005, p. viii). Secondary disabilities, such as deficits in adaptive behaviour, social competence, communication and daily living skills, may also have an effect on work, school and social functioning (Franklin, Deitz, Jirikovic, & Astley, 2008, p. 265 & 270).

It is clear that FASD impacts on purposeful and meaningful participation in the activities of daily life. The scope of practice of the occupational therapist includes the promotion of health, well-being and involvement in life through engagement in activities of daily life (American Occupational Therapy Association (AOTA), 2014, p. S2). Occupational therapists therefore have an important role to play in the management of the realities of FASD. In the domain of occupational therapy, two of the key client factors which are affected by FASD are body functions and body structures. These body functions and body structures support the performance skills of individuals and are important components for successful participation in occupations (American Occupational Therapy Association (AOTA), 2014, p. S7). They are of primary concern in paediatric occupational therapy and assessing them provides decisive information for the occupational therapist when planning interventions (Luebben, Hinojosa, & Kramer, 2010, p. 48). Different frames of reference are used in paediatric occupational therapy to guide the planning and implementation of intervention. Examples are a sensory integration frame of reference, a frame of reference to enhance childhood occupations, a frame of reference for visual perception and a frame of reference to enhance teaching-learning (Kramer & Hinojosa, 2010, p. 99; 266; 349; 234). The question arises which frame of reference or intervention modality could contribute to enhanced performance and engagement in the occupations of children with FASD.

Cognitive and behavioural deficits associated with FASD have been thoroughly covered in the literature (Glass, Ware, & Mattson, 2014). Sensory integration difficulties and dysfunctions, on the other hand, is one area of neurobehavioral functioning which has not been well documented in the research of children with FASD. Jirikowic, Olson and Kartin conducted a study in 2008 which had an important clinical outcome for occupational therapists. They described the sensory processing behaviours and sensory-motor abilities of children with FASD and explored the relationship of these to home and school functions (Jirikowic, Olson, & Kartin, 2008a, p. 120). Marked sensory processing impairments and subtle sensory-motor performance deficits were identified among a large proportion of the children with FASD. Significant correlations were found, supporting the hypothesised relationships between sensory processing and decreased adaptive and academic function among such children (Jirikowic, Olson, & Kartin, 2008a, p. 131). This study was important in that its results supported earlier evidence of sensory processing disorders in children with FASD (Morse et al, 1995, in Jirikowic, Olson, & Kartin, 2008a, p. 31) and were consistent with the theory described in the sensory integration theoretical framework.

Carr, Agnihotri and Keightley (2010, p. 1023) confirmed these findings and found a significant positive correlation between sensory processing and adaptive behaviour of children with partial Fetal Alcohol Syndrome (PFAS), those with Alcohol-related Neurodevelopmental Disorder (ARND), and those who were prenatally exposed to alcohol, but did not meet the criteria for a FASD diagnosis. A third study, by Franklin, Deitz, Jirikowic and Astley (2008), found that children with FASD who demonstrated difficulties with processing and integrating sensory information, also had significantly more behavioural problems, specifically in the domains of socialisation, attention, rule breaking and thought problems.

Comparative studies from a South African context could not be found in an EBSCO Host online search, including the databases Academic Search Complete, Africa-Wide Inform, CINAHL, Health Source and Medline.

As far back as 1972, Ayres (Bundy, Lane, & Murray, 2002, p. 4) described sensory integration as “the neurological process that organizes sensation from one’s own body and from the environment and makes it possible to use the body effectively within the environment” (Ayres, 2005, p. 5). Dunn (1999, p. 11) stated that "learning occurs when a person receives accurate sensory information, processes it, and uses it to organise behaviours." She hypothesised that a continuum of interaction exists between neurological processing of sensory input and behavioural responses (adaptive responses) (Franklin, Deitz, Jirikowic, & Astley, 2008, p. 265). An adaptive response is a "purposeful, goal-directed response to a sensory experience," involving mastering a challenge or learning something new (Ayres, 2005, p. 7). Within a sensory integration framework, sensory integration is considered an important foundation for learning, adaptive behaviour, social-cognitive functioning, skill development and participation in activities (Ayres, 1972; Jirikowic, Olson, & Kartin, 2008a, p. 119; Schaaf & Mailloux, 2015, p. 5), and thus has relevance for children with FASD.

Many of the symptoms of sensory integration difficulties and dysfunctions, such as clumsiness, inattention and distractibility, emotional reactivity, learning problems and sensory sensitivity, have also been consistently reported in children with FASD (Franklin, Deitz, Jirikowic, & Astley, 2008, p. 266; Mattson & Riley, 1998, p. 287). Sensory integration thus offers a framework within which to evaluate, intervene and deepen our understanding of these children, with their challenging behaviours and many functional problems.

Since children with FASD may be affected in almost every area of functioning, educators working with them are faced with unique challenges. Even after the children are diagnosed, classroom educators often do not have the information, tools or skills necessary to assess and support them on an intervention level (Florida State University: Centre for Prevention & Early Intervention Policy, 2005, p. 8). In 2015, the researcher launched an occupational therapy developmental programme, emphasising multisensory stimulation, throughout Grade R in the Cape Winelands Education District (CWED) (one of the eight districts in the WCED). This was designed to support the Western Cape Education Department's (WCED) turnaround plan to improve the Grade 1 passing rate, as set out in departmental documentation from the head of the department (Vinjevold, 2015). The main aim of the programme was to improve school readiness. Two Inclusive Education team occupational therapists were part of the development and played a vital role in ensuring the sustainability of the programme. In order to ascertain the effectiveness of the programme, pre-testing and post-testing were done in selected schools. The pre-testing of one of the Grade R classes during the first term of 2015 (unpublished findings) indicated that 73.0% of the children scored one standard deviation or more below the norm for their age group in the Beery-Buktenica Test for Visual-Motor Integration (Beery & Beery , 2010). In the same class, between 53.0% and 68% had difficulty assuming and maintaining antigravity postures, while 59.0% had problems with smooth eye pursuits according to the Ayres Clinical Observations (South African Institute of Sensory Integration Research Committee, 2005, pp. 13-16).

Given the high prevalence of FASD, the poor academic performance of the children and the significant difficulties they face with processing and integrating sensory information into age-appropriate postural responses and visuo-motor integration, this study set out to investigate the neurobehavioral functions dependent on sensory integration of a group of South African learners with FASD.

The results of this research will be used to plan and design intervention strategies for educators and parents to help these children to optimally participate in daily life activities.

1.2 PROBLEM STATEMENT

Taking into account the effects of the brain damage associated with prenatal alcohol exposure, as discussed in 1.1, the high prevalence of FASD in the Western Cape's school-going population appears to be one of the reasons for the developmental delays and failure to progress academically (1.1). The effects of FASD are devastating and enduring, including cognitive, motor, executive function, language, visuo-spatial, learning, social skills, sensory processing and attention deficits (Mattson & Riley, 1998, p. 291; Bertrand, Floyd, & Weber, 2005, p. viii). All of these impact on what is described in occupational therapy literature as performance skills (American

Occupational Therapy Association (AOTA), 2014, p. S7). These are fundamental to the degree to which an individual can participate meaningfully and successfully in activities of daily life and thus occupations. The high incidence of FASD in the Western Cape, together with the poor results in school performance, were reasons for concern for the researcher and motivated her to investigate the sensory integration difficulties and dysfunctions experienced by children with FASD.

To the researcher's knowledge, no previous study describing the sensory integration difficulties and dysfunctions of South African children with FASD had been conducted. This study therefore investigated and then described the sensory integration difficulties and dysfunctions of an identified group of children with FASD in the Western Cape. Results obtained from this research will be used in future to plan and design occupational therapy intervention strategies to be implemented by educators and parents/caregivers in the WCED. The goal will be to enhance the physical, mental and social well-being and therefore the optimal participation in daily life activities of the child with FASD. Before that can be accomplished, however, scientific knowledge will be needed on the sensory integration difficulties and dysfunctions experienced by children with FASD growing up in the South African context.

1.3 PURPOSE OF THE STUDY

The purpose of the study will be discussed in terms of its aim and objectives.

1.3.1 Aim

The aim of this study was to describe sensory integration difficulties and dysfunctions experienced by an identified group of children aged five to eight years old from the CWED in the Western Cape, South Africa, both with and without a diagnosis of FASD.

1.3.2 Objectives

In order to achieve the main aim, the following objectives were set:

- To investigate sensory modulation of children with FASD aged five to eight years old and a control group of non-exposed, typically functioning children, as measured by the Sensory Profiles.

- To investigate the sensory processing and practic abilities of children aged five to eight years old with FASD and those of a control group of non-exposed, typically functioning children, as measured by the SIPT.
- To identify the patterns of sensory integration difficulties and dysfunctions in both the FASD group and the control group, to describe these findings and to draw conclusions from them about the unique and distinctive patterns of sensory integration difficulties and dysfunctions among children with FASD.

1.4 DELIMITATIONS

The research population was limited to the boundaries of the Cape Winelands Education District where the researcher was employed. The children, both with and without a diagnosis of FASD, were selected for the study from two sub-studies; these were part of a 5-year longitudinal study by FASER-SA with multiple aims which commenced on 1 June 2013 and was carried out in the towns of Wellington, Robertson and Ashton. Both sub-studies from which the study sample was selected were undertaken in this area.

Although more children both with and without FASD were available from the FASER-SA studies, the researcher had to limit the numbers of this study due to time constraints as well as a lack of funding and the availability of occupational therapists qualified in the administration of the SIPT used in the study. Test materials for both the sensory profiles and SIPT are costly and the occupational therapists who assisted had to be remunerated (6.2).

1.5 METHODOLOGY

A quantitative observational analytical design was used in this study. Two groups of children with and without FASD (30 each) were selected through non-random sampling from two sub-studies which were part of an umbrella study on the "Trajectory of Fetal Alcohol Spectrum disorders across the Lifespan: New Understandings in Interventions," conducted in the CWED by FASER-SA at the same time as this study. Two standardised questionnaires, the Sensory Profile (SP) and Sensory Profile School Companion (SPSC), as well as a standardised test, the Sensory Integration and Praxis Tests (SIPT), were used to collect the data. Caregivers were individually interviewed by the researcher to complete the SP, and the educators completed the SPSC. The researcher and one assistant occupational therapist, both qualified in its use, administered the SIPT.

Data were analysed by the Department of Biostatistics, University of the Free State, using SAS/STAT© version 13.2 of the SAS System for Windows. Demographic data were reported using frequencies and percentages, or, where appropriate, means and medians. The various test scores (both raw and standardised) were reported, their means indicated with standard deviations, medians and ranges, as well as 95% confidence intervals.

Because the children of the two groups were matched in age and gender, differences between the typical and FASD children were computed using one-sided, paired t-tests, computed on the raw scores for each subscale. Categorical variables were analysed by means of cross-tabulations with chi-square analysis and Fisher's exact p-values. The results were interpreted, and recommendations and conclusions were made in accordance with the findings.

1.6 THE IMPORTANCE AND VALUE OF THE STUDY

This study was aimed at a better understanding of sensory integration difficulties and dysfunctions of the young South African child with FASD. The results of the study will add to the body of knowledge about children with FASD. It will be of value not only for the researcher but also of clinical importance for occupational therapists. Information acquired from the outcome of this study could contribute to the development of best practices and of new intervention strategies for children with FASD. Furthermore, caretakers and educators of children with FASD will also benefit from the information gathered, once intervention strategies and programmes have been compiled to support them both at home and in the classroom.

A better understanding of the impact of sensory integration difficulties and dysfunctions on the occupations of these children will help occupational therapists to develop intervention programmes and strategies which will optimise functions supported by sensory integration.

1.7 ETHICAL CONSIDERATIONS

In keeping with the obligation to professional ethics and the regulations prescribed by the Health Professions Council of South Africa, the Occupational Therapy Association of South Africa, as well as the University of the Free State, the following relevant ethical obligations were adhered to: basic ethical principles, duties to research participants (e.g. informed consent, respect, confidentiality), as well as duties to the healthcare professions (HPCSA, 2008, pp. 1-11).

Approval for this research was obtained from the Health Sciences Research Ethics Committee (HSREC), University of the Free State (ECUFS no 137/2015) (Appendix A). The researcher obtained written permission from Prof Soraya Seedat to include children identified by FASER-SA as being with or without FASD in this study (Appendix B), as well as permission from the Western Cape Education Department (Appendix C) and the director of the CWED (Appendix D). Permission was obtained from the participating school principals to use their facilities and perform the tests during school hours. Written consent was obtained from the caregivers (Appendix E), as well as written assent from the children (Appendix F). The principals gave permission allowing the researcher to ask the educators involved to complete the SPSC's.

1.8 OUTLINE OF CHAPTERS

The dissertation consists of six chapters arranged as follows:

Chapter 1 - Introduction and Orientation: This chapter provides background and a short introduction to the study. The problem as stated and the purpose of the study discussed in terms of its specific aims and objectives. The researcher describes the scope, value, methodology and ethical considerations of the study, as well as giving a short summary of what is to be expected in the relevant chapters.

Chapter 2 - Literature overview: The literature review gives an essential background to the investigation into the sensory integration difficulties and dysfunctions of children with FASD in the CWED as carried out in this study. The review includes overviews of FASD, of the child with FASD in South Africa – specifically in the Cape Winelands Education District in the Western Cape – of FASD and occupational therapy, FASD and education, as well as of FASD interventions.

Chapter 3 - Research methodology: Chapter 3 focuses on the details of research methodology. The study followed a quantitative method to collect data with an observational, analytical design. The discussion on methodology includes the study design, population, sampling, pilot study, data collection, data analysis, methodological and measurement errors, and ethical considerations.

Chapter 4 - Presentation of results: This chapter gives the results obtained from the demographic information, the Sensory Profile, Sensory Profile School Companion, and the Sensory Integration and Praxis Tests that were used as measuring instruments to identify sensory integration difficulties and dysfunctions in children with FASD. The results are given in the form of tables with brief summaries of the findings.

Chapter 5 - Discussion of results: Chapter 5 discusses, interprets and compares the results presented in Chapter 4. For the purpose of the discussion, existing research and literature are used, together with the researcher's own experience and insights.

Chapter 6 - Conclusions and recommendations: Chapter 6 presents the conclusions and recommendations of the study. The researcher offers recommendations for further research and examines the implications of this research for the design of future intervention programmes for educators and caretakers.

1.9 SUMMARY

This chapter gave an outline of the study, including a background summary orientating the reader to FASD and its debilitating effects on the brain and behaviour, the problem statement and purpose of the study, delimitation factors that needed consideration, the methodology used, the importance and value of the study, ethical considerations that had to be adhered to, as well as an outline of the chapters of the dissertation to follow.

The next chapter will discuss the literature that supported the research throughout.

CHAPTER 2

LITERATURE REVIEW

2.1 INTRODUCTION

Chapter one gave an overview of the purpose, importance and value of this particular study. The methodology used, ethical considerations and delimitations were also discussed briefly in order to introduce the reader to what is to be expected in the different chapters.

The literature review in chapter two will be presented in the following six sections. An overview of fetal alcohol spectrum disorders (FASD), covering its history, the diagnostic process, FASD in South Africa, as well as the effects of alcohol on the brain and behaviour (2.2). An overview of the child with FASD in the Western Cape, South Africa (2.3), followed by FASD and occupational therapy, with a discussion of the occupational therapy scope of practice, sensory integration, sensory-motor development and sensory integration in children with FASD, as well as the value of using the sensory integration framework to assess children with FASD (2.4). This is followed by FASD and education (2.5), and lastly (2.6) a discussion of different intervention methods to try and ameliorate the effects of prenatal alcohol exposure.

2.2 OVERVIEW OF FETAL ALCOHOL SPECTRUM DISORDERS

2.2.1 HISTORY

Fetal alcohol spectrum disorders (FASD) are currently the leading cause of preventable developmental disabilities in the world (Hoyme, et al., 2016, p. 2). The term describes the spectrum of disorders seen when a pregnant woman consumes sufficient amounts of alcohol to cause harm to the developing fetus (Mukherjee, 2015, p. 580).

The negative effects of maternal drinking on the fetus have been suspected throughout history. Evidence exists that pregnant mothers were warned against alcohol consumption in England in the first half of the 18th century, and the belief that alcohol consumption during pregnancy caused “weak, feeble and distempered children” continued until the early 20th century (Mattson & Riley, 1998, p. 279). The idea that alcohol could be harmful to the developing foetus was, however, dismissed during the post-prohibition era as a morally inspired one. It was only in 1968, when Lemoine and three other French researchers (Lemoine, Harousseau, Borteyru, & Menuet, 1968) published their findings after investigating the offspring of alcoholic parents, that interest in the harmful effect of alcohol on the fetus was rekindled (Mattson & Riley, 1998, p. 279). Lemoine et al. (1968, p. 132) wrote:

> *After investigating 127 offspring of alcoholic parents (mostly mothers), we consider the role of chronic alcoholism on the offspring to be very harmful, causing the following: miscarriages, stillbirths, prematurity, growth retardation, psychosomatic alterations with very specific facial features and malformations*.

According to Mattson and Riley (1998, p. 279), these findings went virtually unnoticed until 1973, when Kenneth Jones and his colleagues, morphologists at the University of Washington School of Medicine (Jones, Smith, Ulleland, & Streissguth, 1973, p. 1267), observed and documented similar findings. Their findings were based on observing eight unrelated children whose mothers were chronic alcoholics during pregnancy. They were convinced that their findings were sufficient to conclude that maternal alcoholism can cause abnormal fetal development (Jones, Smith, Ulleland, & Streissguth, 1973, p. 1271). In a second article by Jones and Smith in 1973 (p. 999), the term “fetal alcohol syndrome” was introduced to the medical world for the first time (Riley, Infante, & Warren, 2011, p. 73).

2.2.2 DIAGNOSIS OF FETAL ALCOHOL SYNDROME

Although the diagnosis of Fetal Alcohol Syndrome (FAS) has been expanded and refined over the years, it still includes many of the abnormalities originally described by Kenneth Jones and his colleagues in 1973 (Mattson & Riley, 1998, p. 74). Soon after the initial description of the diagnosis of FAS, it became clear that not all the individuals demonstrated all the features required for a diagnosis, and that the features required for a diagnosis covered a spectrum from mild to severe. In 1996, the Institute of Medicine (IOM) suggested new terminology to include the whole range of consequences following prenatal alcohol exposure (Mattson & Riley, 1998, p. 74; Institute of Medicine, 1996). The IOM suggested the following diagnostic criteria:

- FAS with confirmed maternal alcohol exposure
- FAS without confirmed maternal alcohol exposure
- Partial FAS with confirmed maternal alcohol exposure (PFAS)
- Alcohol-related birth defects (ARBD)
- Alcohol-related neurodevelopmental disorder (ARND) (Institute of Medicine, 1996, pp. 4-8)

The wide spectrum of effects caused by prenatal alcohol exposure led to the term "fetal alcohol spectrum disorders" (FASD) (Sokol, Delaney-Black, & Nordstrim, 2003, p. 2996; Bertrand, Floyd, & Weber, 2005, p. 2; Riley, Infante, & Warren, 2011, p. 74). FASD is therefore "a non-diagnostic umbrella term identifying the range of outcomes from gestational alcohol exposure" (Riley, Infante, & Warren, 2011, p. 74). FAS, PFAS, ARND and ARBD would thus all be included under the FASD umbrella term (Sokol, Delaney-Black, & Nordstrim, 2003, p. 2996; Bertrand, Floyd, & Weber, 2005, p. 2).

In 2005, Hoyme et al. published a report, firstly to clarify specific 1996 IOM criteria and to enhance its practical application in paediatric practice (p. 39) and secondly to attempt more meaningful service to children with FASD (p. 43). For the "Diagnostic guidelines for specific fetal alcohol spectrum disorders (FASD) according to the IOM as clarified by Hoyme et al. (2005)," see Appendix G.

Hoyme et al. (2005, p. 46) stated the strengths of the proposed revised criteria as the following:

- A correction of the vagueness of the original IOM criteria by better definition of certain physical deficiencies and anomalies,
- specifically defined ARND and ARBD,
- the multidisciplinary diagnostic approach,
- using an evidence-based approach and data from previous studies,
- evidence of a rigorous and accurate method, and
- the fact that diagnosis is not only based on prenatal alcohol exposure.

The diagnostic process, using the Diagnostic guidelines for specific fetal alcohol spectrum disorders (FASD) according to the IOM, as clarified by Hoyme et al. (2005), follows the sequence illustrated in figure 2.1 (May, et al., 2016, p. 210):

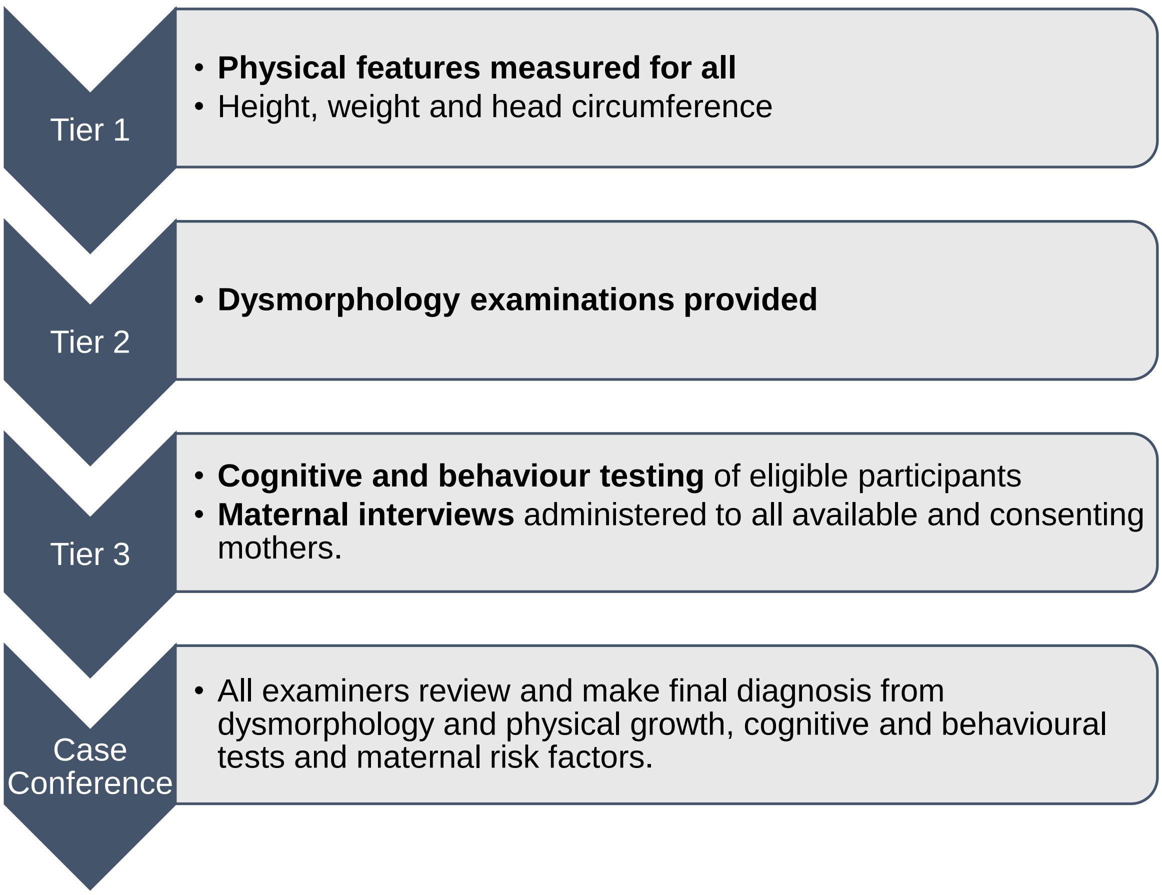

Figure 2.1 The diagnostic process, using the diagnostic guidelines for specific fetal alcohol spectrum disorders (FASD) according to the IOM, as clarified by Hoyme et al. (2005)

A weakness at that stage was the fact that the normative data applicable to growth and facial morphology were largely based on white populations (Hoyme, et al., 2005, p. 46).

A second updated report with diagnostic guidelines was therefore carried out in 2016, following a thorough review of the literature. Drawing on the combined professional expertise of the authors, it was based on the evaluation of more than 10 000 children with potential FASD (Hoyme, et al., 2016, pp. 1-2). Specific areas addressed were the following:

- *Precise documentation of prenatal alcohol exposure,*

- *neurobehavioral criteria for diagnosis of foetal alcohol syndrome, partial fetal alcohol syndrome and alcohol-related neurodevelopmental disorder,*
- *revised diagnostic criteria for alcohol-related birth defects,*
- *an updated comprehensive research dysmorphology scoring system*
- *and a new lip/philtrum guide for the white population, incorporating a 45-degree view* (p. 1)*.*

Assessment of the maternal prenatal alcohol drinking pattern is an essential step and part of the diagnostic process. Quantity of alcohol consumed per occasion, frequency of consumption and timing of consumption during gestation, are all crucial information for diagnostic purposes. The literature indicates that the latter can cause different physical and neurobehavioral phenotypes, with binge drinking (3-5 or more drinks per occasion) being the most detrimental to the developing fetal brain (Hoyme, et al., 2016, p. 5). Consensus among the researchers led to the specific guidelines (Hoyme, et al., 2016, p. 5). See Appendix H.

During the dysmorphology examination, measurement of the height, weight and head circumference is done first, followed by the three cardinal facial features or any other physical malformations of FASD. Appendix I gives the updated criteria for FASD diagnosis.

The next important step in the diagnostic process is the neurodevelopmental assessment. This is essential, since the primary manifestations of the teratogenic effects of alcohol on the brain are cognitive and behavioural deficits (Hoyme, et al., 2016, p. 6; Doyle & Mattson, 2015, p. 175). The use of standardised tests is recommended in this stage of the process (Hoyme, et al., 2016, p. 6).

Once all the information from the interviews, examinations and tests has been obtained, it is strongly recommended that a final diagnosis, whether of FASD or otherwise, is made in a multidisciplinary case conference (Hoyme, et al., 2016, p. 7).

Four other FASD diagnostic guidelines have been published since the IOM criteria in 1996. This reflects the ongoing debate on how most accurately to diagnose FASD (Astley, 2011, p. 6&8; Rendall-Mkosi, et al., 2008, p. 9) They are the following:

- FASD 4-Digit Code, first created in 1997
- CDC FAS Guidelines, published in 2004
- Canadian FASD Guidelines, published in March 2005
- FASD guide for healthcare professionals, published by the British Medical Association in 2007.

The inclusion of the proposed diagnostic criteria for "neurobehavioral disorder associated with prenatal alcohol exposure" (ND-PAE) in the 2013 DSM-5 adds a new dimension to the diagnostic field of FASD (Olson, 2015, p. 187). Three domains of impairment are included in diagnosing ND-PAE: neurocognitive functioning, self-regulation, and adaptive functioning. In contrast to the diagnostic criteria for FAS and PFAS, where the emphasis is primarily on the physical features, the focus here shifts to the cognitive and behavioural deficits caused by prenatal alcohol exposure (Doyle & Mattson, 2015, p. 175).

Olson (2015, p. 187) believes that using the neurobehavioural criteria will not only allow for a wider identification of individuals on the fetal alcohol spectrum but will also facilitate the referral of those identified by health and mental health workers as needing further, more specialised, assessment and tests. Once it has its own ICD-10 code, it will also improve payment for much needed services, interventions and treatments for individuals on the fetal alcohol spectrum (Hoyme, et al., 2016, p. 12).

ND-PAE is currently not an official psychiatric disorder with its own ICD-10 code. It is still listed in Section III of the *DSM-5,* which lists conditions in need of further research before being officially included (Elleseff, 2014).

Although there are still differences of opinion and ongoing debate as to how to make the most accurate FASD diagnosis, consensus has been reached on the following:

- A FASD diagnosis is best accomplished by a multi-disciplinary team.
- Very precise case-defined, validated FASD diagnostic criteria should be used (Astley, 2011, p. 3; Hoyme et al., 2016, p. 2).

Numerous studies conducted in South-Africa (for example, May et al., 2000; Viljoen et al., 2005; May et al., 2007; Urban et al., 2008; May, Blankenship, & Marais, 2013; May et al., 2016) have contributed to the design of a methodology for multidisciplinary studies of fetal alcohol syndrome in developing countries. These studies have also contributed to the refinement of the initial IOM (1996) and the 2005/2016 revised criteria (May et al., 2000, p. 1905; Rendall-Mkosi et al., 2008, p. 9).

A project which began as part of an initiative between South Africa and the United States (US), allowing professionals from the US to lecture, share information and survey for research opportunities in this country, has bequeathed to South African researchers a vast amount of technical expertise and knowledge about the use of these specific diagnostic guidelines – with the revised (2005) IOM diagnostic guidelines for FASD. It also provided South African professionals with locally applicable criteria for future diagnostic research projects (May et al., 2007, p. 1905; Rendall-Mkosi et al., 2008, p. 9). This is demonstrated for instance in the development of a more specific and sensitive lip/philtrum evaluation guide for the Cape Coloured population (Hoyme, et al., 2015, p. 752). It has been recommended that the revised (2005) IOM diagnostic guidelines for FASD be used for research purposes in South Africa (Rendall-Mkosi, et al., 2008, p. 9). To the researcher's knowledge, the 2005 criteria are currently being used for all research in this regard in South Africa.

2.2.3 DIAGNOSTIC PROCESS RELEVANT TO THIS STUDY

The children, both with and without FASD, included in this specific research project were diagnosed by multidisciplinary teams of Fetal Alcohol Syndrome Epidemiological Research - South Africa (FASER-SA). FASER-SA was established as a collaborative endeavour of the University of North Carolina, the University of New Mexico, the Faculty of Medicine and Health Sciences of Stellenbosch University, and the Medical Research Council of South Africa. The research was, at the time of this study, funded by a grant from the USA National Institute on Alcohol Abuse and Alcoholism (NIAAA) (Medicine and Health Sciences - Research Department of Psychiatry, 2013).

FASER-SA was at the time of this research conducting different parts of a study, "Trajectory of Fetal Alcohol Spectrum Disorders across the Lifespan: New Understandings in Interventions," in the Western Cape under the guidance of Prof Philip May of the University of North Carolina.

Permission was granted by Prof May and Prof Soraya Seedat of the University of Stellenbosch for the researcher to access the children with Fetal Alcohol Spectrum Disorders (FASD) and their control group, who had been recruited by them for the above-mentioned studies (Appendix B).

The diagnostic guidelines for specific fetal alcohol spectrum disorders (FASD) according the IOM, as clarified by Hoyme et al. (2005), were used.

The intention here is not to provide results but to inform the reader of the importance and relevance of the literature that will now be reviewed and discussed. The children who were approached/considered for inclusion in the present study were drawn from the larger study described below:

Trajectory of FASD across the life span: New understandings and interventions

This 5-year longitudinal study with multiple aims commenced on 1 June 2013 and is being carried out in the towns of Wellington, Robertson and Ashton. The aims include the following:

a) initiation of early intervention/remediation research on development through nutritional and cognitive/behavioural enhancement techniques for children with FASD from 24 months of age onwards, including physical and behavioural markers at ages 6 weeks, 9, 18, 42 and 60 months,
b) the continuation and initiation with a new cohort, of detailed longitudinal study of the physical and cognitive/behavioural developmental trajectory of children from the newborn period to seven years of age,
c) the initiation of an efficacy study of biomarkers for alcohol consumption,
d) the initiation of new methodology to study the nutrition of women while pregnant via a survey of multiple nutrients through 24-hour dietary recall with the Nutrition Data System for Research (NDRS) and blood sample analysis and blood analysis, and
e) completion of all IOM prevention activity and an intensive evaluation of the impact of nine years of IOM-recommended prevention efforts in the broader community, with a repetition of in-school studies of the prevalence and characteristics of FASD, to be compared with the baseline and a third survey of the community on Knowledge, Attitudes, Beliefs and Behaviours (KABB).

The children were selected from two studies supporting aims (a) and (b). Children from the Newborn Screening study (a) were either recruited at the clinics before they were born or as newborns at the Robertson and Montagu hospitals and were from the towns and farms of Robertson, Ashton, Montagu and Bonnievale. The number of children taking part in this particular study is 98.

The 2014 Wellington schools study (e), done in 13 primary schools in the Wellington area, involved a total number of 390 Grade 1 children. This study commenced in May 2014. Informed consent letters were sent to the parents of 1127 Grade 1 learners in these schools, regardless of age, race or gender. First tier screening was done on the 728 learners with parental consent. Screening consisted of measurement of the learner's length, weight and head circumference. Those with a head circumference less than the 25th centile or length and weight less than the 25th centile, together with randomly selected controls, were invited to take part in the 2nd tier screening, consisting of a brief physical examination for characteristic physical features of FASD. A total of 553 learners participated in the 2nd tier screening. 182 children have been diagnosed thus far (Marais, 2017).

For cognitive and behavioural testing, the following measuring tools were used (Marais, 2016):

a) The 2014 Wellington schools study

- Test of Reception of Grammar (TROG), a measure of verbal intelligence,
- Raven Coloured Progressive Matrices, a measure of nonverbal intelligence
- WISC-IV Digit Span Scaled Score,
- Achenbach Teacher Report Form (behaviour checklist).

b) Newborn Screening Study

- Bayley Scales of Infant and Toddler Development at six, nine, eighteen and 42 months,
- Kaufmann Assessment Battery for Children, second edition (KABC-II), at 60 months.

Having discussed the development of the diagnostic process for FASD according to Hoyme, as well the process used to identity the children with FASD for this study, the next section will provide an overview of FASD in South Africa.

2.2.4 FETAL ALCOHOL SPECTRUM DISORDERS IN SOUTH AFRICA

According to Parry (2005, p. 426), “alcohol has played a central and often controversial role in the life of South Africa since the arrival of European settlers.” From as early as the colonial settlement in the Cape Colony, alcohol was used to pay the indigenous population for cattle and work done on the farms (Parry, 2005, p. 426). This was the beginning of the so called “DOP” system, where farm workers would partly be paid in alcohol. This system, which seems to be unique to South Africa, became an institutionalised part of the farming industry, especially in the Western and Northern Cape, over the next 300 years. Te Water Naude, London, Pitt and Mahomed (1998, pp. 103-104), in their research on farms in the Stellenbosch area, found that the weekly amounts of alcohol given to the farm workers ranged from 750ml to more than 6750ml.

Although the DOP system is no longer legal, it is believed that one of the unfortunate consequences of this practice is the high rate of alcohol abuse and excessive alcohol consumption among farm workers (London, Sanders, & Te Water Naude, 1998, p. 1093; London, 2000, pp. 199-200).

Between the 17th and 20th centuries, South Africa saw the growth of large wine and brewing industries which are still important businesses in the global alcohol market today. The second half of the 20th century was marked by the growth of “shebeens” (illegal alcohol outlets where beer is brewed and sold) as a form of resistance against the “apartheid” laws prohibiting black South Africans from using alcohol (Parry, 2005, p. 426; Olivier, Curfs, & Viljoen, 2016, p. S103).

According to the World Health Organization (Global Status Report on Alcohol and Health 2014), South Africa has a reported absolute alcohol (AA) per capita consumption rate of 11 litres per year, compared to 6.2 litres worldwide (p. 128). With a reported abstinence rate of 43.7% for South African males and 73.7% for females above 15 years of age, the amount consumed per drinker per year is about 20 litres of absolute alcohol. This rate is among the highest in the world (Schneider, et al., 2007,

p. 665). The South African prevalence of heavy episodic drinking (at least 60 grams or more of pure alcohol on at least one occasion consumed in the past 30 days) is 31.7% for males and 13.7% for females (25,6% in total for drinkers only), with a worldwide average of 7.5% (World Health Organization, 2014, p. 35 & 128). This ties in with the finding of the South African Demographic and Health survey (SADHS) of 1998, which reported risky drinking over weekends (five or more drinks per day for men and three or more per day for women) (Schneider, et al., 2007, p. 665), as well as with the statement of De Vries et al. (2016, p. 2) that:

> *weekend drinking is institutionalised and is seen as a normal way of life and a valued form of recreation. Drinking is not only a social affair and a regular source of relaxation, but also a way of coping with unpleasant realities.*

Research done in the Western Cape found that it is quite common for up to 40% of women of childbearing age to drink between two to nine alcoholic beverages over weekend nights (De Vries, et al., 2016, p. 2).

The effect of alcohol consumption on health is detrimental. In 2000 in South Africa, alcohol harm accounted for an estimated 7.1% (95% Confidence Interval 6.6 – 7.5%) of all deaths and 7.0% (95% Confidence Interval 6.6 – 7.4%) of total DALY's (disability-adjusted-life-years) (Schneider, et al., 2007, p. 664; Peltzer, Davids, & Njuho, 2011, p. 30). In terms of alcohol-attributable disability, alcohol use disorders ranked first (44.6%), interpersonal violence second (23.2%), and foetal alcohol syndrome third (18.1%) (Peltzer, Davids, & Njuho, 2011, p. 30; Schneider et al., 2007, p. 664). The negative effects of excessive alcohol use are thus far-reaching in terms of trauma, violence, crime, unsafe sexual practices, brain injuries to the developing fetus, and labour costs in terms of lost productivity at work and absenteism. The combined total tangible and intangible costs of alcohol harm to the economy were estimated at 10 - 12% of the 2009 gross domestic product (GDP). The tangible financial cost of harmful alcohol use alone was estimated at R37.9 billion (Matzopoulos, Truen, Bowman, & Corrigall, 2014, p. 127).

Fetal alcohol spectrum disorders are among the burdens of alcohol consumption. The condition remained under-reported in South Africa until the end of the last century (Olivier, Curfs, & Viljoen, 2016, p. S103). The first study to determine the characteristics of FAS in a South African community was done after collaboration between the vice presidents of South Africa and the United States of America (May, et al., 2000, p. 1905). An epidemiological study of FAS, done in 1997 and reported on in 2000, was carried out in a small town with its surrounding rural settlements just outside Cape Town in the Western Cape (Wellington) (May, et al., 2000). There was a clear need for further epidemiological studies in this province, where the economy is to a large extent based on grape farming and wine production, which has a serious influence on the regional drinking patterns. The residual patterns of regular and heavy alcohol consumption of the DOP system, where wine was used as partial payment for farm workers, remained till today. The problem of heavy drinking has been exacerbated by the availability of inexpensive liquor in shebeens and "papsakke" (A plasticized foil bag in which cheap wine is sold). Heavy binge drinking as a form of recreation has become institutionalised over weekends (May et al., 2000, pp. 1905-1906; Parry et al., 2012, p. 66). There are an estimated 121 000 farm workers in the fruit and wine industry of the Western Cape. This is more than in any other province in South Africa (History of Labour Movements in South Africa, 2015, p. 1).

According to research, it seems that the Western Cape is particularly problematic with regard to harmful alcohol use. In 2005 and 2009, National HIV/AIDS surveys showed that the Western Province had the highest proportion of the general population aged 15 and older scoring eight or above on the Alcohol Use Disorders Identification Test (AUDIT) questionnaire (scores of 8 or more are considered an indicator of hazardous and harmful alcohol abuse) (Babor, Higgins-BiddleJohn, Saunders, & Monteiro, 2001, p. 19). Higher levels of binge drinking of young persons in Grades 8 to 11 were also reported by the National Youth Risk Behaviour Surveys of 2003 and 2008 (Parry C. D., et al., 2012, p. 66).

The research team of the first epidemiological study done in the Western Cape in 1997 was able to access all children in 11 predominantly Coloured and Black schools and one predominantly White school. A high FAS rate of 40.5 to 46.4 per 1000 children aged 5 to 9 years was found. These rates were 18 to 141 times higher than in the United States (May, et al., 2000, p. 1905). Results indicated that all the children with

FAS were within the Coloured group, who also had the lowest socio-economic indicators. May et al. concluded that maternal data in this population were generally consistent with the spectrum of effects exhibited by the children (May et al., 2008, p. 738). The mothers of children with FAS were mostly from rural areas, were living unmarried with their partners and had the lowest education and income levels (some as little as R94 per week). Mothers of both FAS and PFAS children were on average significantly smaller on weight, height and BMI measures, indicating poor nutrition or even malnutrition. Smaller mothers who drink thus have children with more dysmorphology (May et al., 2008, p. 750). May et al. (2008, p. 751) summarise this succintly when they state:

> *So in addition to the fact that the mothers of children with FASD drink in binges during the index pregnancies, their slightness, inadequate diet, and length of drinking career may put the fetus at even greater risk via high blood alcohol concentrations and therefore partially explain the extraordinary high FASD rates in this population.*

This confirmed that low socio-economic communities are at high risk for FASD, as also suggested in earlier US and European studies (Abel, 1995, p. 437; May et al., 2000, p. 1910).

Since 1997, four studies have described the prevalence and characteristics of FASD as recorded in a single municipality and surrounding rural areas in the western part of the Western Cape:

- FAS rate of 40.5 – 46.4 per 1000 children aged five to nine years (May, et al., 2000, p. 1905)
- FAS rate of 65.2 – 74.2 per 1000 children in Grade 1 (Viljoen, et al., 2005, p. 593)
- FAS and PFAS rate of 68.0 – 89.2 per 1000 Grade 1 children (May, et al., 2007, p. 259)
- FASD rate of 135.1 – 207.5 per 1000 (13.6 to 20.9%) Grade 1 children (May, Blankenship, & Marais, 2013, p. 818)

Various other FAS(D) prevalence studies have also been conducted between 2000 and 2016 by other researchers in other parts of the country, as cited by Olivier L. (2013, p. 6). FASD reported were 27 per 1000 in Johannesburg (2000), 122 per 1000 in (2002) and 86 per 1000 in 2010 in De Aar, 69 per 1000 in Upington (2003), 80 – 130 per 1000 in Aurora (2011) and 96 per 1000 in Witzenberg (2013) (Olivier L. , 2013, p. 6). Urban et al. (2015, p. 1016) found a high prevalence rate of FASD (64 per 1000) in a predominantly Black African population in Kimberley. A prevalence rate of 18 to 26%, the highest ever reported in the world, was found by May et al. (2016, p. 207) in four rural communities (Bonnieval, Robertson, Ashton and Montagu) in the Western Cape.

Similar studies done in other parts of the world reported FAS and PFAS rates of 10.9 to 25.2 per 1000 in a Rocky Mountain community and a FASD rate of 24 to 48 per 1000 in the Northern Plains (Hoyme, et al., 2016, p. 2), FASD rates of 10 per 1000 in Canada, 35 per 1000 in Italy, 18 per 1000 in France, 20 per 1000 in Poland and 12 per 1000 in Croatia. In 2015 a study from a community in rural Australia reported a FASD rate of 120 per 1000 (Fitzpatrick, et al., 2015, p. 454). This was the first study outside South Africa to report rates close to those of South Africa. In comparison to these studies from around the world, the extent of the FASD challenge facing South Africa is clear and concerning (Olivier, Curfs, & Viljoen, 2016, p. S104).

2.2.5 THE EFFECTS OF ALCOHOL ON BRAIN AND BEHAVIOUR

In this section, the teratogenic impact of alcohol on the developing fetus will be discussed. This will be followed by a review of current findings related to central nervous system dysfunction, primary neurobehavioural disabilities, as well as associated behavioural, social and secondary problems.

Sensory integration and processing dysfunctions have been linked to a wide range of neurobehavioural difficulties, such as problems with motor coordination, language, visual-perceptual skills, behaviour, attention, learning problems and emotional regulation (Franklin, Deitz, Jirikovic, & Astley, 2008, p. 266). Since many of the symptoms of sensory integration difficulties and dysfunctions, such as clumsiness, inattention and distractibility, emotional reactivity, learning problems and sensory sensitivity, have also been reported in children with FASD (Franklin, Deitz, Jirikovic, &

Astley, 2008, p. 266), this section is of particular importance for this study. Sensory integration difficulties and dysfunctions is an area of neurobehavioural functioning which has not been extensively documented in the research on children with FASD (Jirikowic, Olson, & Kartin, 2008a, p. 118). How sensory integration difficulties and dysfunctions factor into the wide range of complex neurobehavioural problems in children with FASD will therefore be looked at in this study.

2.2.5.1 RISK FACTORS FOR TERATOGENIC EFFECTS OF ALCOHOL

Alcohol use during pregnancy can have detrimental effects on the developing fetal brain and central nervous system, causing permanent changes in brain growth, structure and neurophysiology, as well as both cognitive and behavioural deficits (Glass, Ware, & Mattson, 2014, p. 435; Hoyme, et al., 2016, p. 9). The extent and severity of damage to the central nervous system are largely dependent on how much alcohol the pregnant mother consumes per occasion, how often she consumes alcohol and at what time during gestation this happens (Mattson, Schoenfeld, & Riley, 2001, p. 185; Hoyme, et al., 2016, p. 5). According to Hoyme et al. (2016, p. 5), research has shown that binge drinking of 3-5 drinks or more per occasion has the most detrimental effects on the developing fetus.

The following conclusions on the specific quantity, frequency and timing of prenatal exposure to alcohol that will produce a child with a diagnosis within the FASD spectrum were made by May et al. (2013, p. 510):

- It varies greatly from mother to mother.
- Binge drinking of at least two days a week during all three trimesters may produce a child with FAS or PFAS.
- Reduced average and peak consumption in the later trimesters may produce a child with ARND and exposed children without FASD.
- Regular binge drinking is the most common pattern for all mothers of children with a FASD diagnosis of any kind.

Despite the fact that mothers of FASD children regularly binge drink during their pregnancies, other factors such as inadequate diet, slightness (smaller on weight and

BMI) and length of drinking career, may put the foetus at an even greater risk because of high blood alcohol concentrations (May et al., 2008, pp. 750-751).

2.2.5.2 NEUROANATOMICAL AND NEUROPHYSIOLOGICAL CHARACTERISTICS

The neurological and neurobehavioural difficulties of persons on the fetal alcohol spectrum are probably related to underlying central nervous system abnormalities, especially structural and functional brain abnormalities. Early autopsy studies revealed diffuse brain abnormalities, including microcephaly and microencephaly, callosal dysgenesis and malformations, reduced cerebellar volume and neuronal abnormalities (Glass, Ware, & Mattson, 2014, p. 444; Jones, Smith, Ulleland, & Streissguth, 1973, p. 1268). More modern, advanced neuro-imaging techniques, particularly magnetic resonance imaging (MRI), now allow for in-depth evaluation of neural structural deficits and advance our understanding of brain-behaviour relationships in FASD (Glass, Ware, & Mattson, 2014, pp. 444-445; Spadoni, McGee, Fryer, & Riley, 2007, p. 243).

These neuroimaging studies have revealed several brain differences between alcohol-exposed and non-exposed individuals and also suggest that certain areas of the brain seem to be especially vulnerable to prenatal alcohol exposure (Mattson, Schoenfeld, & Riley, 2001, p. 187; Spadoni, McGee, Fryer, & Riley, 2007, p. 239). These areas include the corpus callosum, cerebellar vermis, basal ganglia, persylvian, orbito-frontal and parietal brain regions (Spadoni, McGee, Fryer, & Riley, 2007, p. 243).

The neuroimaging characteristics of FASD are summarized in Table 2.1 below.

Table 2.1: Neuroimaging characteristics of FASD

NEUROIMAGING CHARACTERISTICS OF FASD				
Brain Area	**Volume/Size**	**Structural Abnormalities**	**Brain-Behaviour Relation**	**Normal Function**
Total brain volume	Reduced overall	Schizencephaly Polymicrogyria	Volume negativity associated with IQ in children and young adults.	
Frontal lobe	Reduced overall Regional volume deficits	Anterior and orbital surface area reductions Cortical atrophy Increased cortical thickness	Surface area, volume and cortical thickness associated with verbal learning and memory.	Involved with movement, decision making, problem solving, and planning.
Temporal lobe	Reduced Regional volume deficits	Increased grey-matter density Increased cortical thickness		Involved with memory, emotion, hearing, and language.
Parietal lobe	Reduced (most affected) Regional volume deficits	Increased grey-matter density Increased cortical thickness		Involved in the reception and processing of sensory information from the body.
Occipital lobe	No reductions	Increased cortical thickness in lateral regions		Main centre for visual processing.
Corpus Callosum	Reduced (Partial and full agenesis)	Decreased length and area, increased displacement and shape variability. Decreased area in midsagital and splenium.	Posterior/anterior displacement associated with verbal learning and recall. Thickness associated with executive function.	Cerebral interhemispheric transfer. Role in: Bimanual motor tasks

			Surface area thinning correlates with motor abilities. IQ associated with surface area. Splenium mean diffusivity values associated with working memory and perceptual organization. Splenium fractional anisotropy values positively associated with visuo-motor performance.	Sustained attention Vision Spatial and visual working memory
Cerebellum	Reduced	Atrophy hypoplasia and sulci distinction. Displaced anterior vermis.	Volume associated with verbal learning and memory. Abnormalities correlates with motor impairments.	Cognitive and motor skills: Balance Coordination Learning
Hippocampus	Disproportionately reduced and may be individually lateralized		Right volume associated with verbal and non-verbal recall. Left volume associated with verbal recall.	Essential component in memory. Synthesis of learned information. Information recovery during retrieval. May be involved in spatial learning abilities.
Basal Ganglia	Reduces caudate, putamen, globus, pallidus and lenticular	Moderate exposure associated with caudate asymmetry	Putamen volume positively correlates with IQ. Caudate volume associated with cognitive control, verbal learning and recall.	Related to movement and procedural learning. Involved in cognitive, affective and motivational functions.

(Compiled by the researcher from Glass, Ware, & Mattson, 2014, pp. 446-448; Bailey, 2016; Spadoni, McGee, Fryer, & Riley, 2007, pp. 241-243)

The development of the central nervous system is characterised by a series of functional, structural, molecular, neurochemical and cellular processes. Experimental evidence shows that prenatal alcohol exposure disrupts many of these processes of normal developmental patterns (Guerri, 1998, p. 304; Guerri, 2002, p. 327). Some brain areas, even within the same region, are more affected than others due to a difference in cell vulnerability (Guerri, 2002, p. 327).

Alcohol disrupts the normal cellular processes and causes alteration in cell migration and neuronal survival and differentiation (Guerri, 2002, p. 327). Changes in neurotransmitter developmental patterns have been reported, decreasing the content of several neurotransmitters as well as the number of uptake sites and certain classes of receptors (Guerri, 1998, pp. 308-309). It seems that, in the case of prenatal alcohol exposure, alcohol also changes the endocrine environment by inducing hormonal changes, causing changes in stress responses and sexual behaviour (Guerri, 1998, p. 309).

It appears that many children prenatally exposed to alcohol show no signs of structural brain changes, yet display a wide range of neurobehavioral problems suggestive of organic brain damage (Jirikowic, 2003, p. 11). Norman, Crocker, Mattson and Riley (2009, p. 212) report on this as follows:

> *While clear structural changes exist in the brains of alcohol-exposed individuals, studies examining brain differences in individuals with FASD also document changes in metabolic levels and ratios, cerebral blood flow and neurotransmitters, even in the absence of structural deficits*.

Functional imaging findings suggest lower levels of serotonin transporter in the medial frontal cortex and higher levels of dopamine transporter binding in the basal ganglia. This may bring about behavioural problems as seen in children with FASD, since serotonin has been linked with inhibition and impulsive aggression and dopamine with attention deficit hyperactivity disorder. The latter is frequently diagnosed in individuals with FASD (Norman, Crocker, Mattson, & Riley, 2009, p. 212).

In a study by Clark, Li, Conry, Conry and Loock (2000), in which they analysed the neuroimages of nineteen individuals who met the criteria for FAS but did not demonstrate severe cognitive dysfunction (mean full scale IQ = 80.2 with range: 66-92), the results indicated that only one individual demonstrated structural brain abnormalities. The abnormality found was for the individual with the lowest IQ.

However, when regional cerebral metabolic rates were analysed in comparison with those of a typically developing control group, the findings showed significant differences in the subcortical areas of the thalamus and areas of the basal ganglia. With the thalamus being a sensory relay area to the cortex for all sensory systems, except for olfaction and the basal ganglia associated with control and coordination of movement, this may suggest evidence of subtle brain differences that potentially could impact on sensory and motor functions (relevant to this study) in the absence of severe cognitive or structural impairment (Clark, Li, Conry, Conry, & Loock, 2000, p. 1099; Jirikowic, 2003, p. 12).

2.2.5.3 NEUROBEHAVIOURAL CHARACTERISTICS

Although external physical features are an indication of prenatal alcohol exposure and are the basis for a FAS diagnosis, the majority of children affected by prenatal alcohol exposure do not show these physical markers. The effects of alcohol on the central nervous system are clinically very significant, since neurocognitive impairment and abnormal behaviour are the most important sources of disability in FASD (Glass, Ware, & Mattson, 2014, p. 435; Hoyme, et al., 2016, p. 9). Researchers have been working for years on a neurodevelopmental profile that is both sensitive (it must correctly identify alcohol-affected individuals) and specific (it must correctly exclude individuals who are not alcohol affected) (Mattson & Riley, 2011, p. 53). A study by Mattson et al. (2013) concluded that the neuropsychological effects of prenatal alcohol exposure are clinically meaningful and can be used to accurately distinguish alcohol-affected children from both typically developing children and those with ADHD. The variables with the largest effect size were those measuring executive function, spatial working memory and delayed matching to sample (Mattson, et al., 2013, p. 525).
The following section will report on the neurobehavioural characteristics of children with FASD.

a) Global Intellectual Ability

FAS is related to decreased intellectual functioning, with average IQs ranging from the borderline to low average ranges (Mattson & Riley, 1998, p. 287; Riley & McGee, 2005, p. 361). This decrease in performance is seen not only in those children with alcohol-related dysmorphia but in all children prenatally exposed to alcohol, and the levels of performance are stable across time (Mattson & Riley, 1998, p. 287; Glass, Ware, & Mattson, 2014, p. 438). Verbal and non-verbal scores are both affected in the FASD population (Glass, Ware, & Mattson, 2014, p. 435).

b) Cognition

- **Executive functioning**

Executive dysfunction is one of the most prominent features of FASD, occurring in alcohol-exposed children both with and without a FAS diagnosis. Research indicates that deficits exist in executive function domains such as planning, set shifting, cognitive flexibility, concept formation, working memory and response inhibition (Glass, Ware, & Mattson, 2014, p. 438).

- **Learning and memory**

Empirical evidence exists of deficits in these areas, particularly those relating to the verbal domain (Glass, Ware, & Mattson, 2014, p. 439; Riley & McGee, 2005, p. 361). Research has found both encoding and retrieval deficits in children with FASD, with retention (the amount of material recalled after adjusting for initial learning) spared. Memory deficits, therefore, do not seem as global as was once thought. It seems that although the FAS group demonstrated deficits in learning verbal information, these resulted from difficulties with acquisition of the information rather than with the ability to remember it (Glass, Ware, & Mattson, 2014, p. 439; Riley & McGee, 2005, p. 361).

Non-verbal memory deficits are less clear, reflecting an insufficient and sometimes inconsistent literature (Glass, Ware, & Mattson, 2014, p. 439).

- **Language**

The effects of prenatal alcohol exposure on language and communication seems to be an area that is still developing. Glass, Ware and Mattson (2014, p. 438) are of the opinion that, although children with FASD have demonstrated deficits in expressive language, in the ability to produce verbal or written expression and receptive language, the underlying mechanisms of these deficits still need clarification, although they may relate to physical changes or impaired auditory and verbal processing.

Mattson and Riley (1998, p. 289) also maintain that research outcomes have been mixed and unclear. They do however conclude that it appears as if children with FAS have deficits in speech and language and noted that similar effects were seen in groups of alcohol-exposed children. Church and Kaltenbach (1997, p. 507) reported delays in language acquisition as well as deficits in both expressive and receptive language of FAS children, and Riley and McGee (2005, p. 361) reported a study where children prenatally exposed to alcohol performed significantly less well when evaluated on word comprehension and naming ability.

More recent research by Wyper and Rasmussen (2011) found that children with FASD scored significantly lower than control children on both receptive and expressive language. Younger children scored significantly lower on relational vocabulary and sentence imitation, while the older children were delayed on word ordering, grammatical comprehension and malapropisms (Wyper & Rasmussen, 2011, p. e364).

- **Cognitive processing speed**

Cognitive processing speed deficits are considered one of the major impairments of infants, children and adolescents on the fetal alcohol spectrum. These deficits are more apparent in the context of complex cognition than in automatic processing. Children on the fetal alcohol spectrum also exhibit deficits in vigilance, reaction time, sustained attention and information processing (Glass, Ware, & Mattson, 2014, pp. 439-440).

Research shows that alcohol-exposed children have deficits in both premotor and motor reaction times, indicating that central as well as peripheral mechanisms are affected (Riley & McGee, 2005, p. 439).

- **Attention**

Hyperactivity and attention deficits are very prominent features of prenatal alcohol exposure. These children have been described as tremulous, hyperactive and irritable, as well as “on the go” and “never sit still” (Mattson & Riley, 1998, p. 287). The problems seem to persist from infancy throughout childhood (Mattson & Riley, 1998, p. 287). A large number of children with FASD would in fact qualify for an ADHD diagnosis. Although some researchers have highlighted similarities between children with attention deficit and FAS or alcohol-related diagnosis, others have found clear distinctions between the neurocognitive and behavioral profiles of the two groups (Jirikowic, 2003, p. 14; Coles et al., 1997, p. 150). Recent research also suggests that attention deficits of children prenatally exposed to alcohol are more domain-specific, with greater impairments in sustained visual attention compared to auditory attention. Coles, Platzman, Lynch and Freides (2002) as well as Mattson, Calarco and Lang (2006) confirm that sustained attention, processed in the visual modality, is more affected than that in the auditory modality. According to Mattson, Calarco and Lang (2006, p. 366), their data suggest that children with heavy prenatal alcohol exposure have deficits in attention which are not global in nature, with consistent and significant deficits in visually focused attention exhibiting lower accuracy levels and slower reaction time. Coles, Platzman, Lynch and Freides (2002) confirmed this in their research and reported that performance deficits result from failure to respond as well as from impulsivity. These outcomes suggested that some aspect of visual processing might be affected by alcohol exposure, and that deficits in visual processing could result from insensitivity to target stimuli.

Further studies to specify these visual attention impairments found impaired alerting (ability to achieve and maintain attention, i.e. getting ready to respond) and executive control (the ability to control behavior, eg. the ability to monitor, detect and resolve conflicts to achieve goals) (Glass, Ware, & Mattson, 2014, p. 440).

- **Academic achievement**

Learning and academic problems have been widely described in children with FASD. These problems exist in all three domains, mathematics, reading and spelling, and extend beyond the effect of low IQ (Glass, Ware, & Mattson, 2014, p. 438; Jirikovic, Olson, & Kartin, 2008, p. 128).

Children prenatally exposed to alcohol consistently demonstrate impairments in mathematics, including basic numerical processing and cognitive estimation (Glass, Ware, & Mattson, 2014, p. 438). Data of a study done by Crocker, Riley and Mattson (2015, p. 114) suggest that heavy prenatal alcohol exposure is associated with deficits in mathematical achievement above and beyond the contribution on general cognitive abilities. They also present with dysfunction in reading and spelling. Deficits occur in pre-reading and spelling abilities such as phonologic processing, as well as comprehension and reading speed (Glass, Ware, & Mattson, 2014, p. 438).

- **Visio-spatial abilities**

Numerous studies report deficits in the Beery Developmental Test of Visual Motor Integration, as well as in impaired spatial location recall and spatial reasoning (Mattson & Riley, 1998, p. 289). In 2008, Jirikowic, Olson and Kartin (p. 128) tested both children with FASD and typically developing children and found that the FASD group scored significantly lower on Design Copying (A Developmental Neuropsychological Assessment - NEPSY). A recent study by Crocker, Riley and Mattson (2015, p. 114) demonstrated that deficits in spatial aspects of attention, working memory and visual memory are related to global mathematics scores in FASD children. Doney et al. (2016, p. 355) reported on below average visual-motor integration problems in a group of FASD and prenatally alcohol-exposed Aboriginal children in the Fitzroy Valley, Australia.

Mattson et al. (2013, p. 1649), following more thorough reseach in their quest for an accurate neurobehavioural profile of FASD, reported that spatial processing is especially sensitive to prenatal alcohol exposure. Several of the measures they used tapped into spatial processing, including spatial recognition memory, spatial span, spatial working memory, spatial learning and visual-motor integration.

- **Motor skills**

Motor impairment is another field of central nervous system function associated with prenatal alcohol exposure. Although a few studies have found no effect of prenatal alcohol exposure on motor development, most studies of motor development and skill suggest an effect (Mattson & Riley, 1998, p. 289). The first known meta-analysis to define and measure the effect of prenatal alcohol exposure on gross motor proficiency was done by Lucas et al. (2014, p. 204), and confirmed that gross motor impairment was consistently found, regardless of the assessment tool used. Gross motor impairments demonstrated in this meta-analysis were in the domains of balance, coordination and ball skills (Lucas, et al., 2014, p. 207). Kooistra et al. (2009, p. 538) reported postural stability problems on a static balance task, whereas a previous study found that, when sensorimotor input (visual and/or somatosensory) was reliable, there was no difference between children with FASD and the controls (Roebuck, Simmons, Mattson, & Riley, 1998, p. 255). The latter also found that the alcohol-exposed children were unable to make effective use of either visual or vestibular information when somatosensory information was inaccurate.

Gross motor deficits were also found in force regulation, atypical trajectories in goal-directed arm movements, and atypical gait (Glass, Ware, & Mattson, 2014, p. 443).

Early descriptions of fine motor dysfunction in children with FAS included tremors, weak grasp, poor eye-hand coordination and fine motor speed (Riley & McGee, 2005, p. 362). Numerous studies have been done on fine motor dysfunction in children with FASD. Adnams et al. (2001, p. 560) reported in a South African study on poor eye-hand coordination in a group of FAS children, as did Kalberg et al. (2006, p. 2037) on significant fine motor delays in a group of Native American children. Both these studies found the fine motor skills significantly more delayed than the gross motor skills (Adnams et al., 2001, p. 557; Kalberg et al., 2006, p. 2042). Jirikovic, Olson and Kartin (2008, p. 132) found that tasks requiring visual-motor speed and precision and design copying speed were particularly problematic for children with FASD.

A review of 24 studies which assessed fine motor skills in primary-school-aged children with prenatal alcohol exposure done by Doney et al. (2014) concluded that complex fine motor skills such as visual-motor integration were consistently impaired in children with moderate to high prenatal alcohol exposure and with PFAS or FAS diagnoses (p. 606). They found that only a few studies investigated foundational fine motor skills, such as grip strength, finger tapping, praxis and kinesthesia. Comparison between the existing studies in this area was difficult since different assessment tools had been used.

In the first study of functional handwriting of children with FASD, as reported by the authors, Duval-White, Jirikovic, Rios, Deitz and Olson (2013, p. 538), challenges were identified with handwriting legibility across letter, sentence and paragraph writing tasks and during performance of visual motor tasks requiring precision and accuracy. Handwriting speed was also below average. Speed and accuracy both decreased with level of difficulty of the task from simple alphabet writing to copying paragraphs.

As had other authors, they also found a decline in the performance of the FASD children as the complexity of the task increased, from speed of simple finger movements to coordinated hand function, tool use and visual-motor abilities (Kodituwakku, 2007, p. 199; Doney et al., 2014, p. 608).

- **Processing of sensations**

Children with FASD have sensory processing irregularities across multiple modalities (Glass, Ware, & Mattson, 2014, p. 441). This will be discussed in depth later in Chapter 2.4.2.3.

c) Soft neurological signs

Children with prenatal alcohol exposure also show evidence of soft neurological signs which may suggest central nervous system immaturity or minor neurological dysfunction (Larroque & Kaminski, 1998, p. 302; Lucas et al., 2016, p. 866). A significant relationship was found between prenatal alcohol exposure and the presence of minor neurological soft signs in preschool children with moderate levels of alcohol exposure (Larroque & Kaminski, 1998, p. 302).

These results were confirmed by a more recent population-based study on children living in remote Aboriginal communities in the Fitzroy Valley, Western Australia. Although the median scores for all the participants were within the normal category, there was significant underperformance in some subgroups, including children with prenatal alcohol exposure and FASD (Lucas, et al., 2016, p. 866). The areas showing higher rates of “severe discrepancies” were Sound Patterns, Figure Recognition and Production and/or Hand Skills and Stand on One Leg. In her commentary on this study, Bertrand (2016, pp. 794-795) holds that, although a consistent pattern of deficits was not identified, the brain-based nature of prenatal alcohol exposure was highlighted. It also played an important role in demonstrating the increase of adverse neurological signs in children who were prenatally exposed to alcohol, but did not meet the FASD criteria. This reinforces the understanding that neurological harm can occur at any level and any time of alcohol exposure, even if this cannot be confirmed with a FASD diagnosis.

The use of soft neurological signs as a measure of central nervous system integrity should be done with caution though, since typically developing children also exhibits moderate rates of soft neurological signs or immaturities which may last until eight or nine years of age (Jirikowic, 2003, p. 17). Larroque and Kaminski (1998, p. 301) are however of the opinion that the presence of soft neurological signs are an indication of the integrity of the central nervous system, similar to psycohomotor development, and that assessing them may be of importance since they may be less affected by the social environment than mental development.

d) Behaviour

Difficult behaviours have been cited as a universal problem among children with prenatal alcohol exposure throughout their lifespan, and seem to be one of the primary reasons causing families to seek help (Jirikowic, 2003, p. 18).

High rates of externalizing problem behaviours such as defiance, aggression and delinquency, have been reported. Children with FASD demonstrate delinquent behaviour, impaired moral decision-making, and lying about their behaviour at a young age. Internalizing behaviours which are not always observable, such as anxiety, depression and withdrawal, are also recorded for this population (Glass, Ware, & Mattson, 2014, p. 441). Challenging behaviour stemming from attention deficits and hyperactivity is also frequently reported (Jirikowic, 2003, p. 19).

It is important to bear in mind that, although many of the behavioural problems may be manifestations of underlying central nervous system dysfunction, factors such as poor environment, family dysfunction, poor socio-economic circumstances, genetic or postnatal influences may also complicate behavioural profiles (Institute of Medicine, 1996, p. 18; Jirikowic, 2003, p. 18).

e) Adaptive and social behaviour

Interpersonal and social skills of children with FASD seem to be affected too. Problems such as clinginess, reduced social competence, not getting along with others, poor social judgment and problems with responding to social cues have been reported in the literature (Glass, Ware, & Mattson, 2014, p. 440). At an early age, children with FASD appear to lack social boundaries, while the older children seem to be socially immature, with poor peer relationships (Jirikowic, 2003, p. 19). A review of social skills deficits in individuals with FASD and prenatal alcohol exposure done by Kully-Martens, Denys, Treit, Tamara and Rasmussen (2012, p. 568 & 570) confirmed social deficits persisting from infancy and childhood into adolescence and adulthood.

Adaptive behaviour in this population also seems to be significantly compromised, especially in relation to communication and social interaction (Whaley, O'Connor, & Gunderson, 2001, p. 1022; Jirikowic, Kartin, & Olson, 2008b, p. 238). A longitudinal study investigating FAS in adolescents and adults reported their adaptive functioning levels to be equivalent to those of a 7-year old, with the most prominent deficits in socialisation skills (perceiving and responding to social cues, cooperating with peers and friendships) (Streissguth et al., 1991 in Kully-Martens, Denys, Treit, Tamara, & Rasmussen, 2012, p. 570). It seems that although young children who are exposed to alcohol prenatally show deficits in all domains of adaptive functioning, the deficits do not differ from those exhibited by non-exposed children with psychiatric problems. Exposed children do however show a more rapid decline in socialization standard scores with age (Whaley, O'Connor, & Gunderson, 2001, p. 1018).

Kully-Martens, Denys, Treit, Tamara, & Rasmussen (2012, p. 568) conclude that "abnormalities in neurobiology, executive function, sensory processing and communication likely interact with contextual influences to produce the range of social deficits observed in FASD." The role of sensory processing will be discussed in Chapter 2.4.2.3.

2.2.5.4 SECONDARY DISABILITIES AND LONG-TERM OUTCOME

In many children affected by prenatal alcohol exposure, functional and behavioural problems underlie poor outcomes and secondary disabilities such as conduct problems, school failure, trouble with the law and poor vocational abilities. Secondary disabilities are defined as deficits not directly attributable to central nervous system dysfunction, but rather as subsequent manifestations of underlying problems which persist over time (Jirikowic, 2003, p. 19). Prenatal alcohol exposure, for instance, does not directly cause school failure. Learning difficulties which are not properly understood can result in poor academic achievement, which in turn can lead to frustration, low self-esteem and anger. The latter can further impact on school performance or even result in drop-out (Jirikowic, 2003, p. 20).

Long-term outcomes for many adolescents and adults with prenatal alcohol exposure are poor, since they demonstrate persisting problems with academic, social, vocational and adaptive skills. These adolescents are reported as having a high rate of secondary disabilities, such as academic failure and school drop-out, difficulties with independent living, staying employed, managing money, trouble with the law and engaging in satisfying relationships (Jirikowic, 2003, p. 20). In a study done by Streissguth et al. (2004) with 415 individuals with either FAS or fetal alcohol effects, the life span prevalence was 61% for disrupted school experience, 60% for trouble with the law, 50% for confinement (in detention, jail, prison or psychiatric or alcohol/drug inpatient facility), 49% for inappropriate sexual behaviors on repeated occasions, and 35% for alcohol/drug problems.

Despite the reports of very challenging futures for children prenatally exposed to alcohol, the chances of escaping these adverse life outcomes increase two- to fourfold with receiving a FASD diagnosis at an early age and by being raised in supportive stable environments (Streissguth, et al., 2004, p. 228). Both Olson, Jirikowic, Kartin and Astley (2007, p. 175) and Streissguth et al. (2004, p. 235) emphasise the importance of caring families, early diagnosis and early intervention. They refer to research indicating that early identification and intervention for children with FASD may be especially important, since central nervous system function might still improve in enriched motor and learning environments (Olson, Jirikowic, Kartin, & Astley, 2007, p. 175). Targeted remedial interventions from caregivers and educators have also been found to facilitate the remediation of academic achievement and behaviour. Coles, Kable and Taddeo (2009, p. 7) report on a study with three- to ten-year-old children with FASD whose maths skills and behavior improved significantly through interventions designed to meet their specific learning and behavior needs. Caregivers as well as educators were involved in these interventions. In a South African study by Adnams et al. (2007, p. 404) a group of nine- to ten-year-old children with FASD showed significant improvements in language and literacy after specific areas were targeted by classroom interventions. The mean test scores of these children did however remain lower than those of the non-exposed controls.

2.3 AN OVERVIEW OF THE CHILD WITH FASD IN SA – SPECIFICALLY IN THE CAPE WINELANDS EDUCATION DISTRICT IN THE WESTERN CAPE

In this section, the realities of children with FASD in the Cape Winelands Education District will be addressed, specifically as they pertain to (a) early diagnosis, (b) early intervention, (c) other factors that impact on the services these children receive, as well as the long-term outcome for them.

a) Early diagnosis

In South Africa, developmental delays in babies and young children are detected at the Primary Health Care clinics or crèches. Developmental screenings should be done when children attend the clinics to be immunized. With a shortage of specialized staff such as therapists to perform thorough and more detailed screenings, nurses are expected to identify problems and refer those children suspected of underlying problems. Therefore babies and young children are often diagnosed too late or not diagnosed at all.

Even in a well-resourced area where specialist screening is available, few doctors or therapists are sufficiently aware of FASD diagnostic criteria or trained to make a diagnosis (Rendall-Mkosi, London, Adnams, Morojele, McLoughlin & Goldstone, 2008, p. 47).

No specific training in diagnosing children with FASD is routinely provided, either to young doctors doing their community service in rural hospitals or to registrars specialising in pediatrics in South Africa. According to a registrar in the Western Cape (Du Plooy, 2016), they often draw conclusions from the mother's drinking history or only see the children once they have been referred for serious academic or behavioural problems. This lack of training for doctors and others in the health services in the identification of FASD is not limited to South Africa. In a study done in Connecticut, Rojmahamongkol, Cheema-Hasan and Weitzman (2015, p. 197) reported that pediatricians "under-recognize FASDs, lack confidence in making this diagnosis and are unfamiliar with the diagnostic criteria." They recommended more training to increase the chances of a diagnosis FASD when children present with developmental delays or behavioral problems. In South Africa, the Foundation for Alcohol Related Research (FARR) established an accredited Training Academy in

2008 which among other functions builds the capacity of health care workers, teachers, social workers, undergraduate students and other relevant stakeholders to identify pregnant women at risk, offer suitable interventions to stop alcohol use during pregnancy and make appropriate referrals for diagnosis (FARR, 2014). Their training is done through workshops on demand.

b) Early intervention

It does, however, seem that the cultural context and physical environment from which the vast majority of the children with FASD come, are not conducive to early intervention and support during the preschool and school years. Most of the children with FASD in the Western Cape come from low socio-economic, rural areas with mothers whose educational level and income, and therefore resources, are low and limited (May et al., 2016, p. 207; May et al., 2008, p. 738). From personal observations during a four-year period as district therapist in the Cape Winelands Education District (CWED), the researcher found houses to be small and often limited to one or two rooms. Mothers were often contract workers on farms, working from dusk to dawn during the season. Off-season months, however, left them with no income and insufficient resources (De Vries, et al., 2016, p. 2). Children witnessed heavy binge drinking, often accompanied with violence over weekends. Given these factors, it is not surprising that support from the home is often poor and erractic.

In the area of community support for early identification and intervention, there seem to be many gaps. According to Rendall-Mkosi et al. (2008, p. 45), very little is published on the interventions needed to assist the young child with FASD in South Africa. When compiling their situational and gap analysis in 2008 they had to rely heavily on information from informant interviews.

Early childhood development programmes for children with FASD, with developmental delays or other disabilities in South Africa seem to be limited. However, non-governmental organizations do play a prominent role in the Western Cape. Pebbles and Early Years Services educate and train parents and practitioners to guide the children with developmental delays (FASD South Africa). Early Years Services, for example, state that “our vision is to ensure that children are nurtured and educated in a stable, caring and stimulating environment” (FASD South Africa). FARR is also involved in a preventative initiative, the Healthy Mother Healthy Baby Programme

(FARR, 2014 b). This programme runs in De Aar, Prince Alfred's Hamlet and Wolseley, with some of the core principles incorporated in the Department of Health's Western Cape Antenatal Personal Support Programme in Delft and Mitchell's Plain. The ultimate goal is for women to have healthy FASD-free babies (FARR, 2014 b). To address one of the aims of their study, Trajectory of FASD across the Life Span: New understandings and Interventions, FASER is currently conducting an early intervention study in the Wellington and Robertson areas. The specific aim is to initiate early intervention/remediation research on development through nutritional and cognitive/behavioural enhancement techniques for children from 12 months of age and upwards, including physical and behavioural markers at various ages (six weeks, nine, 18, 36 and 60 months). Babies measuring up to specific criteria, as well as a control group, were identified to take part. This information was provided by the project manager of FASER in South Africa via e-mail (Marais, 2016).

c) Other factors affecting the services of children with FASD

Once in Grade R and in public schools where classes are large, with numbers of between 30 and 40 children per class, the needs of the learner with FASD are very difficult to accommodate. Where a school has the luxury of a learning support educator assigned to the school, support is done in groups once or twice a week. While the intention of White Paper 6 (Department of Education, 2001) to include children with special needs is noteworthy, educators are faced with overcrowded classes, and do not have sufficient knowledge of or insight into the underlying neurological deficits and behavioural problems of the child with FASD (Rendall-Mkosi, et al., 2008, p. 50). Workshops or training for educators on the educational needs and specific remedial programmes for learners with FASD exhibiting learning and behavioral difficulties are not well aligned in the WCED. During the researcher's time of employment at the CWED office, the only information sessions for educators were the initiatives organised by the component for specialized learner and educational support. No initiative being implemented at the time reached all the educators. Keeping in mind that these learners have very special educational needs and make up a considerable percentage of the children (up to 26% in certain areas of the WCED, as indicated by May et al., 2016), introducing sufficient training for the educators seems to be an urgent need.

2.4 FASD AND OCCUPATIONAL THERAPY

2.4.1 OCCUPATIONAL THERAPY SCOPE OF PRACTICE

The effects of brain damage associated with prenatal alcohol exposure include a range of neuro-behavioural deficits, as described in 2.2.5.3 (Glass, Ware, & Mattson, 2014, pp. 435-443, Riley & McGee, 2005, pp. 361-362). In combination with adaptive behavioral deficits relating to social interaction and communication, personal and community living, all these factors may affect work, school and social functioning (Jirikowic, Kartin, & Olson, 2008b , p. 242; Franklin, Deitz, Jirikowic, & Astley, 2008, p. 265). These effects have all been discussed in depth in Chapter 2.2.5.3.

The scope of practice of the occupational therapist includes the promotion of health, well-being and involvement in life through engagement in activities of daily life (American Occupational Therapy Association (AOTA), 2014, p. S4). Occupational therapy services include assessment and intervention of individuals or groups whose capacity to engage in purposeful and meaningful activities, are threatened. From the extant literature, it is evident that FASD impacts on purposeful and meaningful participation in the activities of life of children with this diagnosis. Within the domain of occupational therapy, the key **client factors** ("capacities, characteristics, or beliefs that reside within the person and that influence performance" (AOTA, 2014, p. 41) which are affected by FASD are **body functions** and **body structures**.

Before discussing the sensory integration difficulties and dysfunctions specific to those children with FASD who took part in this research study and who are attended to in Chapter 5, it is important to have a clear picture of the extent to which the discussed neurobehavioural deficits impact on these children's participation in occupations. Since the ultimate aim of this study is to use the information gained to plan and design intervention strategies for educators and parents to help children with FASD to optimally participate in their occupations, it is important to know how compromised their client factors, performance skills and ultimately their participation in their occupations are. The effect of FASD on **client factors**, **performance skills** and **occupations** within their specific environments will now be discussed.

CLIENT FACTORS:

a) In the domain of **body functions**, the following functions are or may be affected, as noted above:

1) Mental functions such as:

- executive functioning (Glass, Ware, & Mattson, 2014, p. 438),
- attention (Mattson & Riley, 1998, p. 287),
- memory (Kodituwakku, 2007, p. 197; Glass, Ware, & Mattson, 2014, p. 439),
- visual and auditory perception (Mattson et al., 2013, p. 1649; Glass, Ware, & Mattson, 2014, p. 441),
- and emotions (Glass, Ware, & Mattson, 2014, p. 441).

2) Sensory functions such as:

- **Visual functions** which may include one or more of the following:
  - high frequency of ocular pathology and visual impairments such as refractive errors, strabismus, and optic nerve hypoplasia (Glass, Ware, & Mattson, 2014, p. 441),
  - slow processing and slower primary visual cortex activity (Glass, Ware, & Mattson, 2014, p. 441), and
  - difficulty with initial visual encoding (Glass, Ware, & Mattson, 2014, p. 441).

- **Hearing and auditory functions** may include one of more of the following:
  - delayed auditory processing and filtering: central auditory delay (Hoyme, et al., 2016, p. 10),
  - sensorineural hearing loss, central hearing loss or conductive hearing loss due to frequent episodes of otitis media (Glass, Ware, & Mattson, 2014, p. 441; Church & Kaltenbach, 1997, p. 495), and
  - developmental delays in auditory maturation (Glass, Ware, & Mattson, 2014, p. 441; Church & Kaltenbach, 1997, p. 495).

- **Vestibular functions:**
    - although observations made during several animal studies suggest that prenatal alcohol exposure can cause abnormal vestibular function, evidence in human studies has been ambiguous (Church & Kaltenbach, 1997, p. 495 & 506),
    - preliminary results from a very small study (Jirikowic, et al., 2013, p. e212) showed small group differences in sensorimotor and sensory weighting behaviours, specifically those which rely on the integration of vestibular sensation. The results from this particular study contribute to the existing, but limited, information on vestibular functions in this population.

- **Touch functions:**
    - tactile sensitivity has been reported on sensory processing measures (Jirikowic, Olson, & Kartin, 2008a, p. 126),
    - tactile sensitivity, as well as the fact that prenatal stress significantly increases withdrawal responses, was also reported by Schneider et al. (2008, p. 100),
    - lower performance on fine motor tactile performance tests (Osborn, Harris, & Weinberg, 1993, p. 603).

- **Sensory modulation/regulation:**
    - regulatory disorders in infants, such as difficulty with self-soothing and being soothed, decreased affective functioning, increased jitteriness, greater stress vulnerability (Hoyme et al., 2016, p. 10; Olson, Jirikowic, Kartin, & Astley, 2007, p. 181; Jirikowic, Chen, Nash, Gendler, & Olson, 2016, p. 171 & 184),
    - sensory modulation/regulation difficulties have been reported – caregivers revealed that children with FASD demonstrated patterns of overresponsivity to tactile, auditory and visual stimuli, as well as patterns of sensory underresponsivity and poor auditory filtering (Hoyme et al., 2016, p. 10; Jirikowic, Olson, & Kartin, 2008a, p. 132; Franklin, Deitz, Jirikowic, & Astley, 2008, p. 269),

- indication of the inability to effectively use available and accurate sensory systems – multisensory abnormality (Roebuck, Simmons, Mattson, & Riley, 1998, p. 256).

3. Neuro-musculoskeletal and movement-related functions such as:

- **Muscle functions:**
  - weak grasp in infants, poor grip strength in older children (Glass, Ware, & Mattson, 2014, p. 443; Osborn, Harris, & Weinberg, 1993, p. 603,)
  - hypotonia in infants (Harris, Osborn, Weinberg, Loock, & Junald, 1993, p. 610),
  - poor postural stability, deficits in static postural stability (Glass, Ware, & Mattson, 2014, p. 443; Jirikowic et al., 2013, p. 22)

- **Movement functions:**
  - persistent primitive reflexes or significant alterations in reflex behaviour (Harris, Osborn, Weinberg, Loock, & Junald, 1993, pp. 610-612; Osborn, Harris, & Weinberg, 1993, p. 604),
  - poor balance (Glass, Ware, & Mattson, 2014, p. 443),
  - poor eye-hand coordination, poor hand or fine motor skills, deficits in fine motor speed and coordination (Lucas et al., 2016, p. 864; Hoyme et al., 2016, p. 10; Glass, Ware, & Mattson, 2014, p. 443; Osborn, Harris, & Weinberg, 1993, p. 603),
  - impaired oculomotor control (Green, et al., 2009, p. 1302),
  - increased saccadic reaction time (Glass, Ware, & Mattson, 2014, p. 441),
  - variability in saccade endpoint accuracy (Paolozza, Titman, Brien, Munoz, & Reynolds, 2013, p. 1491),
  - wide-based gait and “clumsy” gait have been reported (Osborn, Harris, & Weinberg, 1993, p. 604).

b) **Body structures** which may be affected are first and foremost the structure of the nervous system. Neuroanatomical changes to the brain have been discussed in depth in Chapter 1.2.5.2. Other malformations found in children with ARBD include the cardiovascular system, for example atrial and ventricular septal defects, the structures related to movement, for example large joint contractures and scoliosis, as well as eyes, ears and related structures, for example strabismus and conductive hearing loss (Hoyme, et al., 2016, p. 4).

PERFORMANCE SKILLS:

For a person to actively participate in the activities of daily life, performance skills relating to motor actions, process skills such as attending to, initiating, sequencing and terminating tasks appropriately, as well as social interaction skills, need to be developed over time. Performance skills are defined as “goal-directed actions that are observable as small units of engagement in daily life occupations” (AOTA, 2014, p. S7). Effective performance skills are reliant on body functions and structures, as well as personal and environmental contexts.

Considering all the deficits and delays relating to body functions and structures mentioned above, it seems inevitable that performance skills will be affected.

a) Motor skills

The researcher concluded that deficits in visual, hearing and auditory, vestibular, touch, sensory modulation, muscle and movement functions (see above), could affect the fine and gross motor skills needed for certain activities of daily life as the child engages in occupations. Activities such as getting dressed, moving around freely during play in a well-balanced, coordinated manner, as well as motor skills needed in school, such as sitting upright in a chair, writing and manipulating small objects, are all compromised to different degrees, as discussed earlier. Deficits in mental and sensory functions such as visual perception and touch also limit effective motor skills.

b) Process skills

Process skills, such as initiating, sequencing, pacing and terminating tasks in the classroom, are affected by deficits in mental functions, especially executive functions (Glass, Ware, & Mattson, 2014, p. 438), and slow the cognitive processing speed. Poor concentration (Glass, Ware, & Mattson, 2014, p. 440) will affect the child’s ability to attend to a task until completed.

c) Social interaction

This is a skill with which most children with FASD experience problems (Jirikowic, Kartin, & Olson, 2008b, p. 238; Glass, Ware, & Mattson, 2014, p. 440). Mental body functions such as executive, perceptual and emotional functions play a vital role in regulating the range and appropriateness of interaction with other people. It is clear from earlier discussions that problems are experienced in all three of these areas of functioning (Glass, Ware, & Mattson, 2014, p. 441; Glass, Ware, & Mattson, 2014, p. 441).

Problems with social interaction make it exceptionally challenging for the child with FASD to comply with the role of learner in a school setting. Problems with sensory modulation, social interaction, concentration, understanding of verbal instructions, planning and executing tasks, all contribute to the poor academic outcomes of many children in the Cape Winelands Education District (CWED), as reflected by the annual Western Cape Education Department's (WCED) systemic tests (Cornelissen, 2017).

OCCUPATIONS:

The occupations or daily life activities in which people engage are influenced by the interplay between client factors, performance skills and performance patterns within a particular environment (AOTA, 2014, p. S6). Due to the described deficits in client factors and performance skills, as well as environmental shortcomings as discussed in 2.4.1, children with FASD face numerous challenges in fulfilling their specific occupations.

The main occupations of these children are activities of daily living (ADL), rest and sleep, education, play and social participation. From the reviewed literature it seems clear that children with FASD experience problems in fully participating in such daily activities. Jirikowic, Kartin and Olson (2008b, p. 245) found that such children are slower to aquire age-appropriate ADL skills relating to dressing, mealtime and toileting, compared to typically developing children. Disrupted sleep patterns and short duration of sleep associated with sensory processing problems, with consequent adverse effects on both the child and parent, were reported by Wengel, Hanlon-Derman and Fjeldsted (2011, p. 384). Problems with the activities needed for learning and participating in the educational environment have been widely described in children with FASD. These problems exist in the domains of mathematics, reading and spelling, and extend beyond the effect of low IQ (Glass, Ware, & Mattson, 2014, p. 438; Jirikovic, Olson, & Kartin, 2008, p. 128). Social participation in all spheres (school, family and friends) is problematic. They struggle to meet the expectations of the social conventions and rules needed to engage with other people (Jirikowic, Kartin, & Olson, 2008b, p. 245). It is thus clear that children with FASD demonstrate significant occupational performance deficits.

Given the potential for occupational dysfunction posed by a diagnosis of FASD, occupational therapists clearly have a role to play in the management of children with FASD, particularly in the school system. The occupational therapist's clinical reasoning skills, based on professional training, evidence and expertise, guides her to select one or more appropriate frames of reference to guide intervention, such as the sensory integration frame of reference (Smith Roley, Bissell, & Clark, 2009, p. 824). The role of the occupational therapist in the school system includes the application of specialized knowledge and skills to facilitate adaptive interaction in the school environment and to support both behaviour and learning (Smith Roley, Bissell, & Clark, 2009, p. 824 & 826). The occupational therapist can support children with FASD through direct intervention, consultation with the educators and parents, as well as through early identification and monitoring of children with developmental delays or disability (Jirikowic 2003, p. 45). Occupational therapists often use a sensory integration frame of reference to gain a better understanding of the relationship between central nervous system dysfunction, behaviour and learning difficulties, since sensory processing and integration difficulties and dysfunctions can have a significant impact on school performance (Jirikowic, 2003, p. 46). According to Smith Roley, Bissell and Clark (2009, p. 824), occupational therapists are able to provide evaluation and interventions designed to identify, prevent and remediate deficits related to a child's sensory sensitivities, sensory-perceptual skills, motor and praxis skills, as well as related patterns of performance. All these are potential risks for the child with FASD, as discussed in 2.2.5.3 and earlier in this section. The outcome of occupational therapy using sensory integration theory is therefore to improve function in relevant daily occupations (Smith Roley, Bissell, & Clark, 2009, p. 824). A survey done in the United States as part of a master's thesis, found that out of 131 occupational therapists 81% used a sensory integration frame of reference when working with children with

FASD. Sensory integration dysfunction was also a primary area of focus (Jirikowic, et al., 2013, p. 46).

Using a sensory integration frame of reference with FASD learners in schools, the therapist can include recommendations for sensory-motor activities throughout the day, environmental modifications to assist with regulation and modulation, and consultation with parents and educators to enhance understanding of FASD, offering insight into specific problems. Designing programmes with a sensory integration frame of reference for Grade R and 1 learners could also play an important role in facilitating development and preventing behavioral and academic problems later.

2.4.2 SENSORY INTEGRATION

Cognitive and behavioural deficits associated with FASD have been extensively covered in the literature and are described in section 2.2.5.3. Sensory integration difficulties and dysfunctions, on the other hand, comprise an area of neurobehavioral functioning which has not been thoroughly documented in the research on children with FASD (Jirikowic, Olson, & Kartin, 2008a, p. 118).

Ayres describes sensory integration as “the neurological process that organizes sensation from one’s own body and from the environment and makes it possible to use the body effectively within the environment” (Ayres, 2004, p. 9), or simply put, “the organization of sensations for use” (Ayres, 2005, p. 5). The theory of sensory integration combines concepts from human development, neuroscience, psychology as well as occupational therapy into a framework in which one can view behaviour and learning (Schaaf & Mailloux, 2015, p. 5; Schaaf et al., 2010, p. 100). It assumes that “learning is a function of the brain” (Ayres, 1972, p. 1) and that adequate integration and processing of sensory information are an important foundation for adaptive behaviour (Schaaf & Mailloux, 2015, p. 5).

The sensory integrative approach focuses on sensory-motor factors, such as visio- and somatodyspraxia and vestibular and proprioceptive bilateral integration and sequencing which affects behavior, skill development, learning and participation in activities (Schaaf & Mailloux, 2015, p. 5). Despite the fact that this frame of reference has been updated and expanded, the basic concept of sensory integration theory remains the assumption that “the sensory systems and the integration of their inputs are important contributors to behavior and learning” (Schaaf & Mailloux, 2015, p. 5).

Sensory integration is an evolving theory and practice framework, with a search for growth and a broadening of the knowledge base. To preserve the carefully chosen terms, theory and research-based Ayres Sensory Integration (ASI) body of knowledge, as well as to ensure future development and growth in the way Ayres intended it, the term has been trademarked (Smith Roley, Mailloux, Miller-Kuhaneck, & Glennon, 2007, pp. CE-6).

For the purpose of this study, the terms sensory integration difficulties and dysfunctions will be used. Both difficulties and dysfunctions will be looked at, since there are children who experience sensory integration difficulties, meaning that, while their performance on the SI tests was not so low as to be identified as a dysfunction, they nevertheless faced challenges with sensory integration.

2.4.2.1 CORE CONCEPTS OF AYRES SENSORY INTEGRATION

Ayres regarded sensory integration as a brain behaviour process. Six core concepts provide the basis for the theory and its foundation (Schaaf & Mailloux, 2015, p. 7):

a) Sensory information provides an important foundation for learning and behaviour

Sensory integration theory contributes to child development through its focus on the sensory systems as information sources. Although all the sensory systems are seen as valuable contributors to behaviour and learning, the three “body-related” senses (tactile, vestibular and proprioceptive) are emphasized. These are seen as providing reference points about the body relative to the environment, critical to all learning and behaviour (Schaaf & Mailloux, 2015, p. 7).

b) Sensory integration is a developmental process

Sensory integration takes place in a natural, developmental order and each child follows the same sequence, which in turn is influenced by the child’s experiences (Ayres, 2005, p. 13; Schaaf & Mailloux, 2015, p. 7). Therapy using an Ayes Sensory Integration framework therefore also takes a developmental approach, starting at the child's current level and progressing to higher-level interaction and actions (Schaaf & Mailloux, 2015, p. 7).

c) Successful integration of sensory information results in and is further developed by adaptive responses

An adaptive response is "an appropriate action in which the individual responds successfully to some environmental demand" (Ayres, 2005, p. 199). In an adaptive response, we master a challenge and learn something new. Simultaneously, the formation of an adaptive response enhances brain development and organization (Ayres, 2005, p. 7), and therefore provides motivation and skill to engage in more complex, challenging activities (Schaaf & Mailloux, 2015, p. 8). Ayres stated that "children are designed to enjoy activities that challenge them to experience new sensations and develop new motor functions" (2005, p. 14). The ability to make adaptive responses to constantly changing sensory environments is a crucial factor in the sensory integration frame of reference (Schaaf & Mailloux, 2015, p. 8).

d) The "just-right challenge" provides the milieu for sensory integration to occur

The term "just right challenge" refers to "the activity that has the capacity to build new skills and abilities while adjusting for the current level of function of the child" (Schaaf, et al., 2010, p. 106). Learning can only occur when a child meets and successfully accomplishes a challenge, or as Ayres (2005, p. 8) put it in simple terms, "when the child experiences challenges to which he can respond effectively, he 'has fun'". Sensory integration-guided therapy requires the facilitation of the "just right challenge," thus creating an environment in which learning occurs. With just right sensory and motor challenges, the child is engaged, leading to participation and subsequent success (Schaaf, et al., 2010, p. 106).

e) Children have an innate drive to seek meaningful experiences from their environment

Children have an inner drive toward sensory integration through interaction with and mastery of their environments. They are driven to explore and find meaning in their environments (Schaaf & Mailloux, 2015, p. 8; Ayres, 2005, p. 15).

f) Sensory integration promotes neuroplasticity

"Neuroplasticity is the nervous system's ability to change in response to environmental input and demands and is one of the key theoretical concepts of the sensory integration frame of reference" (Schaaf & Mailloux, 2015, p. 8). Ayres built her theory on research showing that the nervous system, and consequently behaviour is shaped by environmental and sensory inputs or experiences (Schaaf & Mailloux, 2015, p. 8; Schaaf et al., 2010, p. 107).

More contemporary literature has shown that the nervous system is even more plastic, complex and integrated than Ayres and others believed it to be. Kraemer (2001, p. 53) wrote that plasticity is constantly taking place in the nervous system and that it is affected predominantly by the organism's experiences with the environment. Jacobs and Schneider (2001, p. 37) confirm the brain's vulnerability to prenatal stress and alcohol exposure and how it can modify the developing neural system. Of particular importance for this study is the fact that they also demonstrated that the nervous system is changeable and can be altered by environmental influences. In their review of literature between 1964 and 2005, Lane and Schaaf (2010, p. 386) confirmed support for the role of neuroplasticity in many brain regions in response to enriched environments or to direct sensory input, which can be enhanced by motor activity. Many parallels can therefore be drawn between basic science studies and the sensory integration theory of Ayres (1972), meaning that the original principles on which Ayres built her theory are still held in high regard.

Two more recent research studies also validate the above findings. Of specific relevance for this study is the study of Helfer, Goodlett, Greenough and Klintsova (2009), in which mice were induced to binge on alcohol post-natally. Both studies indicated that active sensory-motor activities (wheel-running) induced increases in cell proliferation and neurogenesis in the hippocampal dendate gyrus of mice (Helfer, Goodlett, Greenough, & Klintsova, 2009, p. 1; Brown et al., 2003, p. 2042). This means that exercise improves performance on hippocampal-dependent learning and memory tasks. Similarly, Ayres hypothesized that, with the provision of enriched sensory opportunities and just-right sensory and motor challenges, change will occur both neurologically and behaviorally, as the child makes adaptive responses and is more willing to take on challenges in everyday life (Schaaf & Mailloux, 2015, p. 9).

These studies provide evidence that neuro-plasticity, as stimulated through sensory integration interventions, may also potentially be possible in the child with FASD.

Schaaf et al. (2010, p. 108) added a further core concept to the list:

g) Sensory integration is a foundation for participation

"Sensory integration supports development and occupational engagement through organizing the individual's nervous system" (Spitzer & Smith Roley, 2001, p. 22). The basic processes of sensory modulation and sensory discrimination interact to support praxis and postural/occular/oral control, as well social and emotional development, which eventually form the basis for organized behavior, motivating the individual to act and interact in the environment and adapt to environmental demands. All of these support engagement in occupations or the child's ability to "participate meaningfully in the contexts within which they live and thrive" (Schaaf et al., 2010, pp. 108-109; Spitzer & Smith Roley, 2001, p. 22).

2.4.2.2 KEY SENSORY INTEGRATIVE ABILITIES, DIFFICULTIES AND DYSFUNCTIONS

In 2.3.2.2 the dysfunctions as identified by research were discussed. In this section the key sensory integration dysfunctions, as identified and confirmed by research, together with the difficulties described in the literature, will be discussed in more detail. The aim is to highlight the challenges and functional difficulties experienced by children with FASD.

The abilities supported by sensory integration are consistent with the patterns of function and dysfunction as described. They include sensory modulation, sensory discrimination, postural-occular control, praxis and bilateral integration, and sequencing and visual perception (Schaaf, et al., 2010, p. 112).

a) Sensory modulation

“Sensory modulation refers to the ability of the nervous system to regulate, organize and prioritize incoming sensory information, inhibiting or suppressing irrelevant information and prioritizing and helping the child focus on relevant information” (Murray-Slutsky & Paris, 2000, p. 108). A well-modulated nervous system contributes to the capacity to sustain engagement, despite changes in the intensity of sensations from the body and environment, and thus ensures an appropriate response in direct proportion to the input. In doing so, it contributes to emotional stability, behaviour, arousal, activity level and attention (Schaaf et al., 2010, p. 112; Murray-Slutsky & Paris, 2000, p. 108).

Disorders of modulation can involve over- or under-responsiveness to sensation from either the internal or external environment. As part of the nervous system’s maturation, a balance between habituation (“the nervous system’s recognition that something familiar has occurred”) and sensitization (“the nervous system mechanism that enhances potentially important stimuli”) must develop to support adaptive behaviour (Dunn, 1999, pp. 7-8). The specific point along a child’s neurological threshold that is most likely to produce a response is referred to as that child’s threshold for that stimulus (Dunn, 1999, p. 9). There are two ranges of thresholds – those which support adaptive behaviour and those behaviours which lie outside acceptable ranges for functional performance. Children with thresholds that are too high, tend to be under-responsive to sensory stimuli (“it takes a lot of stimuli to reach the threshold, as when the children don’t respond to cues around them”) (Dunn, 1999, p. 9). Those with thresholds that are too low tend to be over-responsive (“very little stimuli causes a reaction, as when children are distracted by every stimuli”) (Dunn, 1999, pp. 8-9). Miller, Anzalone, Lane, Cermak and Osten (2007, p. 137) also add sensory seeking to describe people who crave an unusual amount of or type of sensory input. They seem to have an insatiable desire for sensation and their neurological thresholds are high.

Problems with modulation can occur in any sensory system, sometimes in a single system, but also in multiple systems simultaneously. Clinical observations suggest that over- and under-sensitivity can be experienced simultaneously in different systems. It is also possible that responsiveness within one system can fluctuate between under- and over-responsiveness (Schaaf, et al., 2010, p. 118).

Other researchers hold that there are three subtypes of sensory modulation disorders, over-responsivity, under-responsivity and sensory seeking (Schaaf, et al., 2010, p. 134).

Children with overresponsivity respond to sensations faster, more intensely and for longer than those with normal responsivity. The atypical responses are automatic, unconscious physiological reactions to sensations. The behaviours elicited can range from active, negative and impulsive, to withdrawal or avoidance of sensations. Sympathetic nervous system activation eliciting exaggerated fight, flight or freeze responses is a marker of sensory overresponsivity (Miller, Anzalone, Lane, Cermak, & Osten, 2007, p. 137).

Functionally overresponsive children can be bothered by specific textured materials or tags in clothing, or lumps in food when the tactile system is involved. They may also shy away from hugs and dislike standing in line close to other children. In the case of auditory over-responsivity, they can try to withdraw from noisy gatherings or become distressed by environmental noises, such as a vacuum cleaner or blender. Children with over-responsive proprioceptive systems tend to avoid and dislike climbing or hanging activities, while vestibular system overresponsivity leads to the avoidance of swings, slides and climbing apparatus. Visual oversensitivity causes sensitivity to bright lights or brightly coloured patterns, smell oversensitivity can cause an aversion to the smell of food and household products, and lastly taste oversensitivity may lead to an avoidance of spicy or salty food or an aversion to mixed consistencies (Schaaf, et al., 2010, p. 135).

Children with underresponsivity disregard or do not respond to sensory stimuli in their environments (Miller, Anzalone, Lane, Cermak, & Osten, 2007, p. 137). They need longer reaction time or a higher intensity or longer duration of sensory stimuli before they react. This makes them appear apathetic and passive (Schaaf, et al., 2010, p. 136).

Functionally they may not respond to being touched or to pain, such as from bumping or falling. These children do not respond when called or to verbal instructions only given once. With smell and taste, they do not notice strong odours and are able to tolerate very spicy food. Movements are performed in a slow, plodding fashion and their grasp is often weak. They do not express a like or dislike of movement experiences (Schaaf, et al., 2010, p. 136).

Sensory seeking children crave an unusual amount or a certain type of sensory input and can be present in any of the sensory sytems (Miller, Anzalone, Lane, Cermak, & Osten, 2007, p. 137). These children are often in constant motion, spinning, bumping and crashing on purpose. They may also engage in unsafe behaviours. Sensory seeking children are over-active, touching people and objects to the point of irritation, watching moving objects and often seeking loud sounds, e.g. the TV or radio, or intense smells and strong taste experiences (Schaaf, et al., 2010, p. 136). They are themselves loud, noisy children, who like making noise for its own sake. With regard to taste, they prefer chewy, crunchy foods and also chew on non-food objects or mouth them (Schaaf, et al., 2010, p. 136).

All three of these subtypes of modulation dysfunctions can interfere with the child's ability to engage in social interactions or to participate in home and school occupations (Schaaf, et al., 2010, p. 136).

b) Sensory discrimination

The discrimination of sensory information involves "the ability to identify and interpret the qualities of the sensory experience" (Schaaf, et al., 2010, p. 121). Instead of only registering and tolerating the sensory information, as in the case of modulation, the person now has to interpret the qualities of the experience. For example, somatosensory discrimination informs the individual about size, weight, texture, location and colour, integrating or associating these with past memories or experiences and formulating perceptions and motor memories to guide planned movements (Schaaf, et al., 2010, p. 121&139).

The sensory integration frame of reference focuses on sensory discrimination in the somatosensory, vestibular, proprioceptive and visual systems.

Indicators of functional and dysfunctional sensory discrimination are the following:

- **Touch/proprioception (somatosensory):** ability to identify an object in the hand without looking, to identify touch on the body, and to be aware of the body parts and their positions. Combined tactile and proprioceptive information facilitates the development of a child's "body awareness" or "body scheme," which in turn provides the foundation for praxis (Schaaf, et al., 2010, p. 113). Dysfunction in this regard leads to challenges with performing fine motor tasks without looking, an inability to localize touch without vision, and difficulty moving through space without bumping into objects (Schaaf, et al., 2010, p. 139).

- **Visual:** the ability to track objects or localize and track a moving target. Visual discrimination is the foundation of form, space and contrast discrimination and therefore also of visual perception, as well as visual and motor skills (Van Jaarsveld, 2011, p. 6). Poor visual discrimination leads to difficulty keeping track of place on a page, following a moving object with the eyes, or copying from the blackboard.

- **Proprioception:** the ability to detect body position in relation to gravity, awareness of body positions and their movement, the ability to use graded force, timing and distance appropriately in play and daily activities, and to maintain posture at the table. Poor proprioceptive discrimination can cause difficulties with balancing without the help of vision, judging timing and distance in ball play, using appropriate pressure with writing implements, or with extension against gravity and balance. It can also cause decreased core stability, lack of postural background movements and low endurance. These children often feel floppy and cannot maintain upright postures during table-top activities (Schaaf, et al., 2010).

- **Vestibular:** the ability to sit still, move through space without tripping and falling, or to establish coordinated use of the two sides of the body and both hands. Children with deficits in vestibular discrimination may have difficulty maintaining good posture and fall over easily. Their body spatial awareness is poor, and they coordinate the two sides of the body poorly, avoid crossing the midline and have difficulty sequencing tasks.

The integration of the sensory information from the different senses allows for the interpretation and understanding of environmental cues, this information then being used to act and interact with the environment. Instead of being able to obtain the information from the multisensory environment, the child with sensory discrimination deficits cannot extract appropriate meaning from the stimuli received (Schaaf, et al., 2010, p. 122). Children with sensory discrimination problems often demonstrate poor body awareness, poor visual perception, as well as difficulty with fine coordinated movements relying on sensory feedback from the muscles and joints for effective performance (Schaaf, et al., 2010, p. 122).

c) Postural-ocular and postural control

"Postural-ocular control involves activating and coordinating muscles in response to the position of the body relative to gravity and sustaining functional positions during transitions and while moving" (Schaaf, et al., 2010, p. 114). Postural responses are required for all the actions needed while physically engaging. It provides a stable yet mobile base for refined movement of the head, eyes and limbs. Postural control is dependent on adequate muscle tone, co-contraction of muscles and the ability to activate muscle synergies. Adequate sensory integration of information from the vestibular, proprioceptive, visual and tactile systems is also essential. These systems play a role in the individual's ability to maintain upright antigravity postures, as well as to move efficiently through the environment. Balance and equilibrium are also components of postural control which are modulated by the vestibular, proprioceptive and visual systems (Schaaf et al., 2010, pp. 114-115; Miller, Anzalone, Lane, Cermak, & Osten, 2007, p. 138).

Postural-ocular dysfunction can lead to difficulty in staying upright for long periods of time or to slouching, difficulty maintaining or adjusting a position to improve task performance, poor trunk rotation, or poor oculomotor control, causing trouble in keeping the eyes on a moving target or tracking words while reading. It can also manifest in poor equilibrium reactions (Schaaf & Mailloux, 2015, p. 22; Schaaf et al., 2010, p. 124; Miller, Anzalone, Lane, Cermak, & Osten, 2007, p. 138).

d) Visual perception and visual-motor integration

Visual perception is crucial to many of the abilities already described and is a common deficit addressed by the sensory integration frame of reference. Visual perceptual abilities are an important part of many cognitive skills that are needed for success in the activities of daily life, from finding one's way in the environment, to school performance and simple self-care activities such as finding clothes in the drawer to get dressed. Visual motor skills include the ability to use vision to guide hand and body movements. These skills need adequate visual tracking, coordination of eye-head movements, as well as coordination of eye-hand movements. Visual motor skills are key components for successful participation in academic and sporting pursuits, as well as play activities (Schaaf, et al., 2010, p. 145&146).

Poor visual perceptual and visual motor skills can lead, among other drawbacks, to copying and handwriting difficulties, the inability to find objects in distracting backgrounds, problems negotiating movements in the environment, and the inability to judge the speed of moving objects (Schaaf, et al., 2010, p. 145).

e) Bilateral integration and sequencing

Bilateral integration and sequencing is the ability to coordinate the two sides of the body effectively in motor activities (Schaaf, et al., 2010, p. 142). There has been an ongoing debate about the inclusion of bilateral integration and sequencing dysfunction as a praxis dysfunction together with somatodyspraxia (Van Jaarsveld, 2011, p. 7). More recent research by Mailloux et al. (2011, p. 148) indicates that vestibular and proprioceptive bilateral integration and sequencing is a separate pattern of dysfunction. This is also confirmed in recent literature by Schaaf and Mailloux (Schaaf & Mailloux, 2015, p. 20).

Effective, well developed bilateral integration is observed when activities that require the use of the two sides of the body together are executed smoothly and skilfully. Examples of these are jumping, skipping, riding a bicycle, or bilateral tool use such as cutting with scissors or eating with a knife and fork (Schaaf, et al., 2010, p. 142).

Bilateral integration and sequencing deficits or dysfunction have their origin in poor vestibular and proprioceptive discrimination, which interferes with the ability to coordinate, sequence and execute motor actions. These problems are found especially in actions that require coordination of the two sides of the body, as well as those needing sequencing and timing through space (Schaaf, et al., 2010, p. 124). Postural-ocular problems, which also have their base in vestibular and proprioceptive processing, are often seen in conjunction with bilateral integration and sequencing problems (Schaaf, et al., 2010, p. 142).

Difficulties with lateralization skills, establishment of hand dominance and poor crossing of the midline are characteristic of difficulties in bilateral integration and sequencing. The influence of postural problems can often be seen in an inability to stay upright for long and with slouching (Schaaf & Mailloux, 2015, p. 22). With sporting activities, problems are often seen with skilled coordination of actions that require efficient timing and spatial accuracy. Sequencing of motor actions seems to be particularly challenging, even when the child can complete the separate parts successfully. These children have difficulty coordinating two parts of the body in bi-manual tasks, such as holding paper down to write or using a knife and fork. They struggle with bilateral tasks, such as hopscotch, riding a bicycle or pumping a swing. Coordinating eye-hand activities such as catching and throwing a ball is often problematic (Schaaf, et al., 2010, p. 142).

f) Praxis

"Praxis is the ability to conceptualize, plan and execute intentional and meaningful motor actions" in response to environmental demands (Schaaf, et al., 2010, p. 122). It is therefore a neurocognitive process of identifying and planning how to make effective goal-directed motor responses to specific situations. Praxis consists of several sub-processes which include ideation, motor planning, sequencing, execution and feedback. It requires the retrieving of sensory and motor memories of previous actions when a new plan is needed. Successful motor experiences allow for the availability of more motor engrams or "neural maps" in our nervous system on which we can draw when building new motor plans. Accurate sensory perceptions enhance the creation of an accurate plan of action (Schaaf, et al., 2010, pp. 122-123; 116).

A deficit in planning new motor actions, learning new motor actions and generalizing motor plans is called a somatodyspraxia. A somatodyspraxia has its origin in somatosensory discrimination deficits, interfering with the development of body scheme and body awareness. These children struggle to complete new or complex motor tasks (Schaaf, et al., 2010, p. 124 & 140). They have difficulty with body boundaries, are clumsy, disorganized and struggle with activities of daily living such as brushing teeth, getting dressed and using eating utensils. They also often develop low self-esteem and have a strong need to be in control. They often find transitions or changes of schedule throughout the day difficult (Schaaf, et al., 2010, p. 141).

Execution, or the production of the motor movement, is the last step in the process. Motor control is essential for accurate execution of an action that has been planned. It is therefore more critical than praxis in the execution of motor acts (Schaaf, et al., 2010, p. 142).

The most recent development in the understanding of praxis is reflected in work done by Theresa May-Benson on the ideational part of the process (Van Jaarsveld, 2011, p. 8; Schaaf et al., 2010, p. 125). May-Benson and Cermak stated that ideation underlies planning, sequencing and organization of actions and may therefore influence the way children engage in activities and occupations in unstructured or novel situations. They developed the Test for Ideational Praxis, described as an "objective assessment of a child's ability to demonstrate awareness of object affordances" (May-Benson & Cermak, 2007, p. 152). Since the evaluation of ideational praxis is not part of the SIPT, and the test is not readily available without training, it has not been covered in this specific research.

Successful participation in self-care, play, social and academic activities is the end-product outcome of the sensory integration process for children. Participation in the relevant daily activities is dependent on the production of a suitable adaptive response to all the demands of the environment. Mastery of the environment depends on adequate sensory modulation, discrimination and integration of the sensory systems, praxis and the organisation of behaviour. Together all these processes form the basis or foundation for successful and meaningful participation in the occupations of daily life (Schaaf, et al., 2010, p. 130).

2.4.2.3 PATTERNS OF SENSORY INTEGRATION DYSFUNCTION

Studies over more than sixty years have shown that there are common patterns of sensory integration dysfunction (Schaaf & Mailloux, 2015, p. 17). Ayres carried out numerous factor analytic studies, first using the Southern California Sensory

Integration Test (SCSIT) and later the Sensory Integration and Praxis Tests (SIPT) that validated sensory integration theory (Schaaf, et al., 2010, p. 111). These factor and cluster analyses included children with a variety of developmental disorders as well as typically developing children. Patterns emerged from these test scores and observations which reflected shared underlying commonalities loading together in clusters (Schaaf & Mailloux, 2015, p. 17). Ayres conducted various cluster analyses through which four dysfunctional groups were identified:

- Low-average bilateral and sequencing deficits
- Visual and somatodyspraxia
- Dyspraxia on verbal command
- Generalized sensory integrative dysfunction

Mulligan (1998, p. 827) confirmed these findings of Ayres when her cluster analytic studies revealed many similarities to the previous model. Mulligan suggested a four-factor model of sensory integration dysfunctions:

- Visual perceptual deficit
- Bilateral integration and sequencing deficit
- Dyspraxia
- Somatosensory deficit (Mulligan, 1998, p. 826; Schaaf et al., 2010, p. 111)

Several of Ayres' factor analyses also indicated high factor loading between tactile defensiveness and hyperactiviry and distractibility (Schaaf, et al., 2010, p. 111). Tactile defensiveness is defined as "the tendency to react negatively and emotionally to touch sensations" (Ayres, 2005, p. 106). Unusual responsiveness in other sensory systems was also identified by Ayres. This included gravitational insecurity, auditory hyperresponsivity and visual distractibility (Schaaf, et al., 2010, p. 111). Ayres noted that children on the autistic spectrum often exhibited behaviours suggesting poor modulation of sensory information. These observations started the formulation of a new subtype of sensory integrative dysfunction in sensory modulation. According to Schaaf et al. (2010, p. 111), people like Dunn and Miller expanded on and refined this factor by using data from instruments such as the Sensory Profile and the Sensory Processing Measure.

Mailloux et al. (2011, p. 143) identified similar patterns to those found by Ayres in earlier studies in a retrospective study. The patterns identified in this study were the following:

a) Visuo- and Somatodyspraxia

This factor is characterised by high loadings on the tests of visual perception and visuopraxis functions and includes Space Visualization, Design Copying, Constructional Praxis, Postural Praxis, Praxis on Verbal Command and Manual Form Perception. The fact that Postural Praxis, Praxis on Verbal Command and Manual Form Perception are also present in this factor suggests a mild association with a pattern of Somatodyspraxia. The factor therefore represents Visuodyspraxia with secondary aspects of Somatodyspraxia (Mailloux, et al., 2011, p. 148).

Visuodyspraxia occurs when a person has difficulty with visual perception as well as with visual-motor planning. The main feature of this pattern therefore is the association between visual perception and visual-motor skills. Although this pattern has been identified individually, a combined pattern of visuo- and somatodyspraxia was found in several studies, for example those of Ayres (2004, p. 142) and Mailloux et al. (2011, p. 147). The combined pattern will be used in this study.

b) Vestibular and Proprioceptive Bilateral Integration and Sequencing

This pattern involves inefficient vestibular processing associated with poor postural, ocular and bilateral function. It is characterised by high loadings on Oral Praxis, Standing and Walking Balance, Postrotary Nystagmus (PRN), Bilateral Motor Coordination, Graphestesia, Motor Accuracy and Sequencing Praxis. Although the Motor Accuracy test item does not involve bilateral or sequencing actions of note, this test item also loaded statistically within the group in this study. It is hypothesized to reflect the vestibular and bilateral functions needed for coordination of the eye, head and hand movements, postural adjustments and crossing of the midline needed for this test (Mailloux, et al., 2011, p. 148). Kinesthesia loaded modestly on this factor and together with Standing and Walking Balance led to adding the term “proprioception” to the name of the factor. The presence of PRN on this factor has been inconsistent, and Mulligan (1998, p. 842) noted in her study that this test had no association with any of the patterns of dysfunction. In a retrospective study to verify the patterns of sensory integration dysfunction, Mailloux et al. (2011) did however find a link between the PRN test and low scores on tests of vestibular and bilateral functions (p. 149). The relationship of hyporesponsiveness to vestibular input, shown by low PRN scores, had been hypothesized, but this was the first time it had been empirically supported. This finding therefore supported Ayres' theory that bilateral integration problems and other signs of verstibular dysfunction are associated with a shortened duration of postrotary nystagmus (Mailloux, et al., 2011, p. 149).

The PRN test is unique in the sense that it is the only SIPT subtest in which both high (Z scores >1) and low scores (Z score <1) are indicative of dysfunction, whereas for all the other tests only low scores indicate dysfunction (Mailloux et al., 2011, p. 145; Mulligan, 1998, p. 822). A low PRN score is further believed to be associated with a problem different from a high PRN score (Mailloux, et al., 2011, p. 145). This had caused problems in previous studies, since combining low and high PRN scores results in mean scores for the group which appear average. To avoid this, Mailloux et al. (2011, p. 146) entered this variable in several different ways, including entering high and low scores separately. Data of this study did however indicate that a very small percentage (about 8%) of the sample demonstrated prolonged PRN ($\geq$ 1.0 standard deviation). Since the cancelling effect was not present in this sample, PRN scores were entered in the same way as the other SIPT scores (Mailloux, et al., 2011, p. 146).

c) Tactile and Visual Discrimination

This factor is characterised by two tests of tactile discrimination, Localization of Tactile Stimuli and Finger Identification, and one test of visual discrimination, Figure-Ground Perception. This dysfunction indicates discriminatory difficulties in the tactile- and visual systems in the absence of praxis difficulties (Mailloux, et al., 2011, p. 149).

d) Tactile Defensiveness and Attention

This factor is consistent with the hypothesised patterns between inattention and hyperactivity and tactile defensiveness as earlier decribed by Ayres (Mulligan, 1998, p. 149).

This concludes the discussion on the patterns of sensory integration dysfunction identified and confirmed by research. Essential for this specific study, however, is the fact that Van Jaarsveld, Mailloux, Smith Roley and Raubenheimer (2014, p. 2) confirmed similarities in these patterns of dysfunction in a study on South African children experiencing sensory integration difficulties. This supports the value of the SIPT in identifying sensory integration dysfunction across countries. It also confirms that South African children experience sensory integration dysfunctions, similar to those already identified in research, which interfere with their occupations (Van Jaarsveld, Mailloux, Smith Roley, & Raubenheimer, 2014, p. 5). However, in another study on typically developing South Afrcian children from middle- to higher socio-economic settings, the authors did find that typically developing South African children performed moderately to significantly better in five of the SIPT tests in the older age band (6y – 8y 11m). In order to prevent South African children with sensory integration dysfunction going unidentified by the SIPT, they recommend that the scores of the five identified SIPT test items of the children in the older age band be adapted by ½ a standard deviation unit toward the negative side, before clinically interpreting the scores (Van Jaarsveld, Mailloux, & Herzberg, 2012, p. 12). The five tests concerned are Design Copying, Oral Praxis, Bilateral Motor Coordination, Standing and Walking Balance, and Motor Accuracy.

The confirmation of the patterns of sensory integration dysfunction, identified in numerous studies from 1966 to the present, guided the development and refinement of Ayres Sensory Integration theory, assessment and intervention (Schaaf & Mailloux, 2015, p. 18). Schaaf and Mailloux (2015, p. 17) emphasised that these patterns should assist the evaluator in recognizing relationships among scores and thus to interpret the set of scores and observations seen. They did not intend to suggest that the assessment data of a child would fit clearly into a specific pattern.

The patterns verified and clarified by Mailloux et al. (2011) and confirmed as being valid for South African children by Van Jaarsveld, Mailloux, Smith Roley, and Raubenheimer (2014, p. 2) will be used in the analysis and clinical reasoning for this study as they are the most recent and have proved relevant for children from South Africa. As sensory modulation difficulties and dysfunctions are not identified by the SIPT, the Sensory Profile Caregiver Questionnaire as well as Sensory Profile School Companion will be utilised in this study.

2.4.3 SENSORY-MOTOR DEVELOPMENT AND SENSORY INTEGRATION IN CHILDREN WITH FASD

This section will review the effects of prenatal alcohol exposure on sensory integration, sensory processing and sensory-motor development in children with FASD.

2.4.3.1 Studies examining sensory-motor development of the child with FASD from a sensory integrative perspective

Although many researchers have investigated aspects of motor, neuromotor and neuropsychological development and performance in children with FASD (as previously discussed and referenced) only a few, however, have examined sensory-motor development from a sensory integration perspective (Jirikowic, 2003, p. 37). According to Jirikowic, (2003, p. 37), Morse et al. published one of the first and few studies in 1995 which examined sensory processing and integration in children with FAS and alcohol-related diagnoses. Their results suggested that children with prenatal alcohol exposure experienced problems particularly in the domains of visual-spatial processing, as well as auditory and tactile processing. Although these findings provided preliminary evidence of sensory processing problems in this population, they were limited by reliance on questionnaires still under development (not standardized), a sample with a very large age range (2 to 19 years), and subjects diagnosed by methods not as systematic and clearly defined as are currently available (Jirikowic, Olson, & Kartin, 2008a, p. 119; Jirikowic, 2003, p. 37).

Jirikowic, Olson and Kartin conducted a study with an important clinical outcome for occupational therapists (Jirikowic, Olson, & Kartin, 2008a). They described the sensory processing behaviours and sensory-motor abilities of 25 children with FASD and a control group of 26 typically developing children and explored the relationship between them in home and school functions (Jirikowic, Olson, & Kartin, 2008a, p. 117). Marked sensory processing impairments and more subtle sensory-motor performance deficits were identified among a large proportion of the children with FASD. Significant correlations were found, providing support for the hypothesized relationships between sensory processing and decreased adaptive and academic function among such children (Jirikowic, Olson, & Kartin, 2008a, p. 131). This study was important in that the results supported earlier evidence of sensory processing disorders in children with FAS, as described by Morse et al. in 1995 and by Jirikowic (2003, p. 37). Their findings are consistent with the theory described in the sensory integration theoretical framework.

Franklin, Deitz, Jirikowic, and Astley (p. 265) also published a retrospective data anaysis in 2008, describing the sensory processing and behavior profiles of 44 children aged five to ten years with FASD. They found that children who demonstrated difficulties in processing and integrating sensory information also had significantly more behavioral problems, specifically in the domains of socialization, attention, rule breaking and throught problems (Franklin, Deitz, Jirikowic, & Astley, 2008, p. 270). Again, however, they felt that their sample was too small and only drawn from a limited population. They also had to rely on caregiver reports which were dependent on the caregiver's own observations (Franklin, Deitz, Jirikowic, & Astley, 2008, p. 271).

Carr, Agnihotri and Keightley (2010) conducted a secondary data analysis on 46 children between the ages of three and 14 years with FASD. The participants were diagnosed according to the Canadian guidelines, including an assessment for ADHD. Only those whose caregivers had completed a Short Sensory Profile (SSP) and Adaptive Behavior Assessment System – Second Edition (ABAS-II) were included. The children were divided into three distinct groups: those with prenatal alcohol exposure, those diagnosed with ARND, and those diagnosed with PFAS (Carr, Agnihotri, & Keightley, 2010, p. 1024). Only one child in the FASD groups had a concomitant diagnosis of ADHD (Carr, Agnihotri, & Keightley, 2010, p. 1030). A significant correlation was found between scores on the SSP and ABAS-II (a General Adaptive Composite score that reflects a child's overall adaptive behavior and Practical domain, including skills of community use, home living, health and safety and self-care) (Carr, Agnihotri, & Keightley, 2010, p. 1030). This suggests that "deficits in a child's ability to process sensory information from their environment may consequently be related to the adaptive behaviors they use in response" (Carr, Agnihotri, & Keightley, 2010, p. 1030). A similar link was found by Jirikowic, Olson and Kartin (2008a), as well as by Franklin, Deitz, Jirikowic and Astley (2008).

The authors suggested future studies with a larger sample size and more direct methods of measurement of sensory processing and adaptive behavior, rather than relying on caregiver reports (Carr, Agnihotri, & Keightley, 2010, p. 1031). They warned that the relationship between sensory processing and adaptive behaviors should be interpreted with caution, since concomitant diagnoses, for example of ADHD and mental health and psychiatric diagnosis, might influence the results (Carr, Agnihotri, & Keightley, 2010, p. 1030).

Comparative studies from a South African context could not be found in an EBSCO Host online search, including the databases Academic Search Complete, Africa-Wide Inform, CINAHL, Health Source and Medline.

2.4.3.2 Studies examining components of sensory processing and sensory integrative functions of the child with FASD

Numerous studies, as will be discussed, have examined tasks related to sensory-motor performance in children with prenatal alcohol exposure, with inconsistent results. Although the findings from these studies are informative, they only address limited components of sensory processing and integrative functions (Jirikowic, 2003, p. 38).

a) Fine and gross motor skills, eye-hand coordination and visual perception

- Mattson, Riley, Gramling and Delis (1998, p. 146) used a set of neuropsychological tests and compared the results of three groups: children with FAS, those prenatally exposed to alcohol, and typically developing children. The results showed that both the FAS group and the group of children prenatally exposed to alcohol experienced difficulties on tests of fine motor speed and visual-motor integration when compared to the typically developing group. They also performed poorly on the other cognitive and neuropsychological tests.

  In contrast, Janzen, Nanson and Block (1995, p. 276) found no significant differences between the FAS and control groups for fine motor ability. There were however significant differences in visual-motor, motor and perceptual performances. When visual perceptual abilities were measured without a motor component, no significant difference was found.

More recent studies indicated the following:

- Doney et al. (2014, p. 353-355) reported on a study done in Western Australia in a population-based cohort of predominantly Australian Aboriginal children consisting of children with and without prenatal alcohol exposure and children with FAS. Visual-motor integration was moderately impaired across the cohort, especially in children with FASD. Visual perceptual scores were in the average range for the cohort, with no significant differences between the groups. Fine motor coordination was in the average range, but below average and significantly lower in the children with FASD.
- In a South African study by Adnams et al. (2001, p. 560), the FAS group performed significantly worse than the control group on a combination of four Griffiths subscales which included eye-hand coordination.
- Doney et al. (2014a, pp. 606-608) reviewed 24 studies which assessed fine motor skills in primary-school-aged children with prenatal alcohol exposure or FASD. Complex fine motor skills such as visual-motor integration were consistently poor in all groups. This implies that complex skills are more likely to be impaired after prenatal alcohol exposure. The studies concluded that complex skills were more likely to be affected than basic fine motor skills such as grip strength.
- Kalberg et al. (2006) assessed the gross as well as fine motor development of three groups of children (14 children with FAS, 11 children not FAS but prenatally exposed to alcohol, and 11 not prenatally exposed to alcohol). Results indicated clinically important delays in the motor development of children with FAS, with fine motor skills being more delayed. They suggest that fine motor delays in children with FAS may be related to specific neurobehavioral deficits affecting fine motor skills.
- Since handwriting is a critical skill for school success, Duval-White, Jirikovic, Rios, Deitz and Olson (2013) conducted a study to investigate handwriting skills in children with FASD. Participants scored below average on measures of handwriting legibility, speed and visual-motor precision. The study suggests deteriorating visual-motor skills and increased difficulty

as the task complexity increases (Duval-White, Jirikovic, Rios, Deitz, & Olson, 2013, p. 534).

It is important to note that visual-motor deficits and greater difficulty performing complex tasks are challenges which have also been found in other studies of children with FASD (Duval-White, Jirikovic, Rios, Deitz, & Olson, 2013, p. 540).

- With regard to gross motor deficits, Kalberg et al.(2006, p. 2037) found fine motor development to be significantly more delayed than gross motor development in children with FAS. No significant group differences were found between the gross motor scores of children with FAS, those with prenatal alcohol exposure who did not have FAS, and those with no reported prenatal alcohol exposure.
- This finding was confirmed by Adnams et al. (2001, p. 557), who found no significant group differences between a group of children with FAS and a typically developing group on the gross motor subscale of the Griffiths Mental Development Scales.
- A systematic review with meta-analysis conducted by Lucas et al. (2014), however, found gross motor deficits in the domains of balance, coordination and ball skills (Lucas, et al., 2014, p. 207).
- An earlier study by Roebuck, Simmons, Mattson and Riley (1998, pp. 252; 256-257) suggests that prenatally alcohol-exposed children exhibit balance deficits under specific environmental conditions which may indicate a multisensory abnormality. The children seemed to be overreliant on somatosensory input, and when this was inaccurate they were unable to compensate using available visual or vestibular information. Unavailability of somatosensory input caused significantly greater anterior-posterior body sway. The researchers considered the possibility that the children were unable to integrate visual and vestibular information and were unable to use vestibular information alone to maintain balance.
- Church and Kaltenbach (1997, p. 506), however, were of the opinion that "the abnormal balance and gait exhibited by FAS children and animal

models are more likely due to a cerebellar dysfunction than vestibular dysfunction." Nevertheless, they did not rule out central vestibular (e.g. vestibulo-cerebellar) dysfunction (Roebuck, Simmons, Mattson, & Riley, 1998, p. 256).

- In a small, more recent study by Jirikowic et al. (2013) examining sensorimotor performance and the sensory control of balance, evidence was found of "diminished functional sensorimotor performance on clinical measures for children with FASD compared to matched controls with typical development" (Jirikowic, et al., 2013, p. e224). The clinical severity of postural dysfunction in the children with FASD was minimal to moderate. The lower total mean scores and vestibular scores were considered to be clinically significant and suggestive of decreased postural stability under inaccurate or conflicting sensory conditions. The findings were therefore interpreted as evidence of possible inefficient adaptation to and use of sensory subsystems, namely vestibular input, for balance control (Jirikowic, et al., 2013, p. e225).
- The corpus callosum is one of the brain structures affected in children with FASD. This part of the brain plays a very important role in coordinating motor activity from opposite sides of the body, and bilateral manual coordination deficits have been reported in children with FASD. A study by Roebuck-Spencer, Mattson, Deboard Marion, Brown, and Riley (2004) was conducted to assess speed and accuracy of bimanual tasks (Roebuck-Spencer, Mattson, Deboard Marion, Brown, & Riley, 2004, p. 536). The Bimanual Coordination Test was used, where the two hands must coordinate to guide a cursor through angled pathways, providing measures of the ability of the two hemispheres to coordinate activity via the corpus callosum. The results indicated that FASD children were equally accurate but slower than the non-exposed control children on basic visio-motor tasks. Variable and inaccurate performance of the FASD group did however increase as task complexity, and reliance on interhemispheric interaction increased (Roebuck-Spencer, Mattson, Deboard Marion, Brown, & Riley, 2004, p. 536).

- Wass, Simmons, Thomas and Riley (2002) conducted a study "to determine the extent to which prenatal alcohol exposure affects perception, movement planning and movement execution during tasks that require temporal processing" (Wass, Simmons, Thomas, & Riley, 2002, p. 1887). The children had to perform two timing tasks, a co-incident-anticipation timing task primarily assessing central processing, and a movement-speed timing task focusing on the motor component of temporal processing. The children with prenatal alcohol exposure were significantly less accurate and more variable than the control group on both tasks. This meant that both the sensory-perceptual and motor components of temporal processing were disrupted in the children prenatally exposed to alcohol. This may be due to damage to the basal ganglia or cerebellum (Wass, Simmons, Thomas, & Riley, 2002, p. 1887).

b) Infant studies

The existing literature reports several neurobehavioral concerns in infants prenatally exposed to alcohol which could be regarded as indications of sensory processing and integration difficulties and dysfunctions.

- Regulatory behaviours in infants prenatally exposed to alcohol have been reported in a few studies. Harris, Osborn, Weinberg, Loock and Junald (1993, pp. 610-612) discuss five infants who were followed up from approximately 5 months of age to a final visit between 18 and 20 months. The problems reported included generalised low tone, delayed head-righting reactions, delays in fine and gross motor milestones, gagging on any other than pureed or junior foods at 18 months of age, failure to show any response to the Bailey bell, refusal to bear weight on feet when placed in supported standing, flat affect, sneezing, tactile hypersensitivity to the scalp, irritability, lack of fear and a lack of realisation of dangers.
- Davis Eyler and Behnke (1999, p. 107) summarised peer-reviewed literature published during the two decades preceding 1999, examining representative studies of the developmental outcomes of drug-exposed infants during the first two years of life. They reported on a study where the

researchers found that higher alcohol use in midpregnancy was significantly related to poorer habituation and lower arousal scores. The investigators in an earlier newborn study (part of this same project) found five main effects of alcohol. Alcohol exposure was significantly related to:

- more eye opening,
- more head turning to the left, which is atypical for neonates,
- more body tremors,
- lower levels of body activity, and
- less hand-to-face activity.

Babies of the heaviest drinkers and smokers also demonstrated more dazed eyes, yawns and sneezes.

- Davis, Eyler and Behnke (1999) also reviewed an observational study of newborns by Rosett et al. (1979) to measure the amplitude and duration of activity in wake-sleep states. This study was done because sleep disturbances are common among alcohol-exposed infants and state of regulation is important for infant-caregiver interaction. They found that infants born to heavy alcohol users sleep less than those born to moderate users. They had more interruptions of quiet sleep, were more restless, and had more body movements when compared to the controls. There was a significant negative correlation between the amount of alcohol exposure in the third trimester and total sleep (Davis Eyler & Behnke, 1999, p. 112).

- In a South African study on 85 Cape Coloured infants who were assessed on the Alarm Distress Baby Scale at 6.5 months, Molteno, Jacobson, Carter, Dodge and Jacobson (2014) found a direct effect of fetal alcohol exposure on emotional withdrawal in infancy (p. 479). They speculate that infant withdrawal may reflect a neurological impairment attributable to alcohol exposure, independent of the postpartum social environment. It is important to note that children who were later diagnosed with FAS and PFAS at five years old exhibited more emotional withdrawal and less responsivity and activity as infants. It seems that infant withdrawal is a significant predictor of nine-year IQ (Molteno, Jacobson, Carter, Dodge, & Jacobson, 2014, p. 479). In another study to examine regulatory behaviors and stress reactivity, conducted on 18 infants (6-15 month old) with moderate to high prenatal alcohol exposure in comparison with infants with low or no prenatal exposure to alcohol, Jirikowic, Chen, Nash, Gendler and Olson (2016) found that the group with moderate to high prenatal alcohol exposure demonstrated significantly fewer social monitoring behaviors compared to the controls (Jirikowic, Chen, Nash, Gendler, & Olson, 2016, p. 171). They exhibited significantly less social monitoring, such as orientation or attunement to their caregiver's facial behaviors, than controls (both during play and reunion episodes). Physiologically the infants with high-risk prenatal alcohol exposure had significantly high baseline levels of salivary cortisol. This is an indicator of heightened reactivity after a stressor. The physiological dysregulation and early affective differences noted in these infants may place them at an increased risk for early regulatory problems (Jirikowic, Chen, Nash, Gendler, & Olson, 2016, p. 181). The authors of this study maintain that early identification and intervention can take advantage of neuroplasticity and build essential protective factors, such as responsive caregiving, to soften early adversity and help improve early regulatory challenges (Jirikowic, Chen, Nash, Gendler, & Olson, 2016, p. 185). Continued research on early regulatory processes among children with prenatal alcohol exposure is essential also in South Africa.

c) Sleep studies

Two other studies link the sleep challenges to sensory processing deficits:

- Fjeldsted and Hanlon-Dearman (2009) conducted a pilot study which was the first work done to positively correlate sensory processing differences in the sleep patterns of children affected by prenatal alcohol exposure. They collected data from 20 children aged 0-36 months with confirmed alcohol exposure. The results of two questionnaires completed by the caregivers were analysed for correlation. They found that children who slept less during the day were more sensation seeking and active, most probably working actively to meet their high neurological thresholds. Those who remained awake more at night were avoiding sensory stimuli, probably due to having low sensory thresholds. They work actively to avoid sensory overload (Fjeldsted & Hanlon-Dearman, 2009, p. 27). These findings are of great importance both for this study and for occupational therapists in general, since sleep is impacted by sensory modulation problems.

- Wengel, Hanlon-Derman and Fjeldsted (2011, p. 384) assessed sleep using actigraphy, a sleep log and the Children's Sleep Habits Questionnaire. The Sensory Profile was used to evaluate the child's sensory processing difficulties. The children with FASD presented with significantly more sensory processing deficits compared to the controls, with the sensation seeking pattern in the quadrant scores indicating the highest departure from the normal. Data showed that children with FASD had significantly more sleep disturbances than typically developing children, including increased bedtime resistance, shortened sleep duration, increased sleep anxiety and more frequent night awakenings (Wengel, Hanlon-Derman, & Fjeldsted, 2011, p. 384).

When looking at the interactions between sleep and sensory physiology it is important to note that the processing of sensory information is present during sleep, although it exhibits different characteristics during slow-wave sleep (deep sleep) and paradoxical sleep (REM sleep) (Velluti, 1997, p. 61). Sensory processing is vital in the initiation and maintenance of sleep. The reticular formation is both responsible for sensory processing and the induction of wakefulness, and must be down-regulated during sleep (Wengel, Hanlon-Derman, & Fjeldsted, 2011, p. 389). Since sensory stimuli of any of the senses have the potential to disrupt sleep if the brain is unable to regulate the incoming signals, effective sensory modulation is crucial (Wengel, Hanlon-Derman, & Fjeldsted, 2011, p. 389). "Missing sensory input as well as the enhancement of several sensory inputs can produce sleep and waking imbalances, augmenting or diminishing the proportion of sleep or waking" (Velluti, 1997). Wengel, Hanlon-Derman and Fjeldsted (2011, p. 390) confirm this statement when they suggest that children with FASD lack the organised filter system to accomplish self-regulation during sleep. This is a very important finding, since it indicates that sensory integration treatment addressing sensory modulation difficulties may be helpful in the FASD population.

2.4.4 The value of using the Sensory Integration Framework to evaluate children with FASD

It seems clear from all the literature discussed that sensory integration and sensory processing problems are associated with a wide range of behavioural and developmental difficulties, many of which have been reported in children with FASD. Jirikowic (2003 p. 41) believes that, considering the strengths and weaknesses of the framework, sensory integration and processing should be further studied in young children with prenatal alcohol exposure. She offers the following reasons for her opinion (Jirikowic, 2003, pp. 41-42):

- Sensory integration provides a theoretical framework for examining neurobehavioural disorders related to more subtle central nervous system organization.
- Studies to examine sensory integration and processing in children with FASD remain limited (and have not been done in South Africa according to the search mentioned in Chapter 2.4.3), despite preliminary research evidence of difficulties in this area.
- Sensory integration and processing problems can be identified in young children, creating an opportunity for early identification and intervention.

The framework of sensory integration can be used to examine behaviours related to the central nervous system processing, organization and integration of sensory input. Looked at from a theoretical standpoint, the diffuse problems in central nervous system organization and subtle information processing in children with prenatal alcohol exposure correlate in many aspects with those of children with sensory integration difficulties and dysfunctions (see section 2.4.3). Dysfunctional behaviour that results from an underlying organic basis is theorised within the sensory integration framework, as opposed to dysfunctional behaviour, which is seen only as being a psychological or movitational problem. (Jirikowic, 2003, p. 42).

The systematic assessment of sensory integration and related behaviors in young children with prenatal alcohol exposure is still limited. The three studies discussed all recommended the use of more expanded and direct measures of sensory integration, rather than making use only of caregiver and teacher questionnaires and tests for sensory-motor development (Carr, Agnihotri, & Keightley, 2010, p. 1031; Franklin, Deitz, Jirikowic, & Astley, 2008, p. 271; Jirikowic, Olson, & Kartin, 2008a, p. 133). Jirikowic, Olson and Kartin (2008a, p. 133) specifically recommend the use of the SIPT. With the highest rates of FASD in the world, as discussed in Chapter 2.2.4, a study examining the sensory integration difficulties and dysfunctions of the children with FASD in South Africa is long overdue.

Finally, sensory integration and processing dysfunctions can be identified in early childhood. Early identification and intervention can facilitate the prevention of secondary disabilities later in life and can play an important role in supporting children with FASD and their families ((Jirikowic, 2003, p. 43).

Streissguth stated in 1997 that "alcohol-related diagnosis by age six was related to reduced odds of secondary disabilities later in life and considered a protective factor in later development." While early identification of sensory integration problems in children with FASD may not directly impact diagnosis, it could have important implications for the planning of intervention services to prevent secondary disabilities.

2.5 FASD AND EDUCATION

Many of the symptoms of sensory integration difficulties and dysfunctions such as clumsiness, inattention, and distractibility, emotional reactivity, learning problems and sensory sensitivity have consistently been reported in children with FASD (Franklin, Deitz, Jirikowic, & Astley, 2008, p. 266; Mattson & Riley, 1998, p. 287).

Keeping in mind that children with FASD may be affected in almost every area of functioning, it is clear that their educators are faced with numerous unique challenges. Even after the children are diagnosed, their educators may not have the information, tools or skills necessary to assess and support them on an intervention level (Florida State University: Centre for Prevention & Early Intervention Policy, 2005, p. 8).

School-aged children will usually be referred for an evaluation because of learning problems, especially with reading and mathematics, or because of behavioural abnormalities (Canadian Paediatric Society, 2002, p. 7; Rendall-Mkosi et al., 2008, p. 49). In South Africa, especially in under-resourced communities, the most common time for a child with FASD to be identified as having serious disability is in the foundation phase in primary school. The child's educator may observe learning difficulties and/or behavioural problems and refer the child to the District Based Support Team (DBST) (Rendall-Mkosi, et al., 2008, p. 50). Rendall-Mkosi et al. (2008) summarise the South African situation accurately when they state: "Again, due to poor resources, even if the psychologist, therapist or remedial teacher identifies the particular difficulties of the child, the ongoing remedial support required is not available unless the child is placed in a special school" (p. 50). Since these children often do not qualify for placement in a special school, it remains the task of the class educator to accommodate the child with his or her specific needs. Even if a child qualifies, the distances from the rural areas to the special schools, together with the finances involved, often make placement impossible. In some cases, where a learning support teacher is assigned to the school, the child will get more specialized help in a small group. From experience in the Department of Education, the researcher is aware that in most instances neither the teacher nor the learning support teacher have suffiicient specialized knowledge on the educational needs of the child with FASD. Even if the class educator has the knowledge, with classes of up to 40 children in one class (as was the case with the Grade 3 classes who participated in this study), the educational needs of the child with FASD are very difficult to accommodate (Rendall-Mkosi, et al., 2008, p. 50).

According to Education White Paper 6 (Department of Education, 2001) on Inclusive Education for children with special needs in South Africa, the idea is to increase support so that children with disabilities can attend mainstream schools in their own communities (Rendall-Mkosi, et al., 2008, p. 51). Class educators should be well equipped to adapt the curriculum according to the child's abilities, use teaching strategies suited for the child with special education needs, focusing on strengths and providing an emotionally nurturing and accepting environment. Unfortunately, the emphasis is still mostly on scholastic achievement (pass rates) with the consequent effect of poor achievement on self-esteem, causing most of these children to drop out of school (Rendall-Mkosi, et al., 2008, p. 50). The drop-out rate before Grade 12 is unacceptably high. Spaull (2015) reported that "in 2014, only 532 860 learners wrote matric (and 403 874 passed), even though there were 1 085 570 learners in the cohort that started Grade 1 twelve years earlier" (p. 36). The reality therefore is that only about 36% of children who started school in 2003, passed matric in 2014. One of the reasons given for the high drop-out rate is the failing of grades (Spaull, 2015, p. 36). With a FASD rate of 18 - 26% (May, et al., 2016, p. 207), children on the fetal alcohol spectrum, with all their neurobehavioural dysfunctions and accompanying learning difficulties, certainly contribute to the number of children failing grades. Specific statistics in this area are not yet available, although Dr Luther Robinson, a member of the FASER-SA team, is currently busy conducting a follow-up study in Wellington, according to FASER's project manager in South Africa (Marais, E-mail correspondence, 2016) .

The Western Cape Education Department's annual systemic tests confirm that problem areas already exist in the foundation phase. Table 2.2 shows the 2016 performance in mathematics and language of the Grade 3 learners in the Cape Winelands Education District, compared to those of the whole WCED (Cornelissen, 2017).

Table 2.2: WCED Systemic Test Results 2016

Grade 3 Mathematics Item Analysis 2016		
	Pass%	
	CWED	Province
Arrangement of numbers	60.3	64.7
Counting	60.3	62.1
Graphs	77.5	80.0
Shape	68.0	70.8
Time	10.4	12.6
Word Sum (Fractions)	44.5	47.8
Addition	54.6	60.3
Division	38.3	40.9
Multiplication	48.5	53.8
Subtraction	38.3	41.9

Word sum (Addition)	50.9	55.4
Word sum (Division)	44.9	48.1
Word sum (Subtraction)	40.8	46.8
Word sum (Multiplication)	49.8	55.7

Grade 3 Language Item Analysis 2016		
	Pass %	
	CWED	Province
Multiple Choice Questions - Recall	58.5	62.8
Ordering Sentences	29.5	34.1
Reasoning – Sentence writing	32.4	36.6
Recall – Sentence writing	49.6	53.8
Reviewing – Sentence writing	35.3	41.7
Writing (Five sentences)	42.5	48.2
Matching Words (DNA)	18.0	18.3
Matching Words (Correct)	67.5	67.1
Recall Words	29.8	36.8
Synonyms (DNA)	24.2	22.1
Synonymns (Correct)	28.4	36.2
Word Meaning	22.8	27.0

It is interesting to note that the CWED is one of the three districts with the lowest scores of the eight districts in the WCED. The three districts with the lowest scores are also the three rural districts. This may be because demographics and socio-economic factors can impact on early childhood (Davies, et al., 2011, p. 298). Davies et al. (2011,

p. 298) state that: “With age, the developmental delay between groups becomes more evident and the apparent influence of socio-economic factors more marked.” According to Davies et al. (2011, p. 298), their findings concur with previous research showing that children from poor socio-economic environments tend to score within the normal range on cognitive assessments during early infancy, but if they remain in disadvantaged environments for the first two years of life, all developmental domains become significantly more deprived. Since the control group of this study was selected from the same low socio-economic areas as the cases, it will be interesting to see whether this study can confirm this statement as it manifests in the development of sensory integration and sensory processing.

The main areas of challenges identified by the WCED for the Grade 3 learners in mathematics and language seem to be the following (Cornelissen, 2017):

- **Mathematics**
  - Arranging numbers either from smallest to biggest or from biggest to smallest
  - Understanding the concept of time moving from morning to afternoon
  - Word problems – failure to grasp which one of the four basic operations to use
- **Language**
  - Following instructions on paper
  - Weak basic sentence construction. Inability to construct five basic sentences
  - Weakness in grammar, spelling and punctuation
  - Incorrect transcribing

As discussed in 1.1, an occupational therapy developmental programme, emphasizing multisensory stimulation, was launched during 2015 in all the Grade R classes of the CWED. As part of the WCED’s turnaround plan (Vinjevold, 2015), it was designed to improve the Grade 1 passing rate. With the main aim of the programme being the improvement of school readiness, pre- as well as post-testing was done in two identifiied schools. These pre-tests highlighted significant difficulties with processing and integrating sensory information into age-appropriate postural responses and visuo-motor integration.

The high prevalence of FASD in the CWED, the poor academic performance of the children, as well as the difficulties highlighted by the pre-tests as described in 1.1, together warranted this investigation into the neurobehavioural functions dependent on sensory integration of a group of children with FASD in South Africa.

2.6 FASD INTERVENTIONS

Despite the devastating effects of prenatal alcohol exposure, women continue to drink during pregnancy. The ideal would be to intervene at the time of alcohol exposure, directly preventing or reducing maternal consumption. Given that prevention is not always possible, effective treatments that will reduce developmental alcohol-related deficits are urgently needed (Idrus & Thomas, 2011, p. 76&83).

Different strategies, both in use and as described in the literature, can be followed to try and control or improve outcomes:

2.6.1 Interventions during alcohol exposure

“Alcohol disrupts development through numerous mechanisms, both direct and indirect” (Idrus & Thomas, 2011, p. 76). Attempts have been made by researchers to prevent alcohol-induced damage by blocking these mechanisms.

Findings from animal studies suggest that blocking N-methyl-D-aspartatereceptors during withdrawal in the fetus or newborn may offer protection against some of the neuropathology associated with prenatal alcohol exposure. It has also been demonstrated that the administration of serotonin agonists, either in vivo or in vitro, can prevent alcohol-induced cell death. Other findings suggest the development of pharmacological agents to prevent alcohol-induced inhibition of L1 cell adhesion in an effort to reduce the severity of FASD. Since neurotropic factors influence cell metabolism and growth, the proliferation and differentiation, as well as the migration and maturation of cells, administering these agents may reduce alcohol’s teratogenic effects by promoting cell growth and survival.

As discussed in 2.2.5.5, alcohol may also lead to cell death due to an “imbalance between the production of damaging reactive oxygen species and the ability of cells to protect themselves with endogenous antioxidants” (Idrus & Thomas, 2011, p. 79). Successful protection against alcohol-related growth retardation, physical abnormalities and neuropathologies has been demonstrated in a number of studies. Although negative side-effects have been reported in some of these, identification of antioxidants that could be effective clinically remains an area of great interest.

Nutrients such as vitamins C and E possess antioxidant properties and may modify alcohol’s teratogenic effects. Research has indicated that a wide variation in outcomes of children with FASD is still a reality and that this variation may also be attributed to nutritional factors (May P. A., et al., Maternal Risk Factors for Fetal Alcohol Syndrome and Partial Fetal Alcohol Syndrome in South Africa: A Third Study, 2008, p. 750). Malnutrition can worsen fetal alcohol effects. Nutritional supplements may therefore serve as a relatively easy way to improve outcomes among children prenatally exposed to alcohol. There are, however, synergistic effects among nutrients which call for caution in the prescription. A balanced multisupplement diet throughout pregnancy may be the most effective, whether or not it is compensating for a deficiency. Although research in this area has been done, further studies are, according to literature, warranted (Idrus & Thomas, 2011, pp. 80-81).

Researchers state that performing the experimental treatments discussed in 2.6.1 in the clinical field will have challenges (Idrus & Thomas, 2011, p. 83). These include the difficulty of administering treatments during prenatal periods and safety concerns. According to Idrus and Thomas (2011, p. 83), the ideal would be to intervene during the time of alcohol exposure in order to directly prevent and/or reduce maternal alcohol comsumption. Prevention, however, is not always possible, hence the need to seek for effective treatments to reduce developmental alcohol-related deficits. Continued research and refinement of interventions may become viable in the future.

2.6.2 Intervention for individuals with FASD

Other types of interventions are easier to implement. They include the following:

- **Nutritional and pharmacological interventions**

Nutritional supplementation such as choline administration and intervention targeting the enhancement of the plasticity of the brain are currently being investigated in animal studies. Choline has the ability to reduce the severity of adverse physical and behavioral oucomes (Idrus & Thomas, 2011, pp. 81-82).

- **Environmental interventions**

Neuronal plasticity can be enhanced with environmental enrichment such as social, motor and sensory stimulation (Idrus & Thomas, 2011, pp. 82-83). This kind of stimulation is known to enhance a number of central nervous system responses, such as increasing neurotropic factor levels, structural changes including dendritic arborization, as well as neurogenesis and improved learning. Animal experimental research has found that running on exercise wheels can soften the adverse effects of alcohol exposure on hippocampal plasticity and learning, and acrobatic motor learning can enhance the functioning of the cerebellum and weaken alcohol-induced motor skill impairments.

It is clear that the postnatal environment can play a key role in influencing the adverse effects of prenatal alcohol exposure (Idrus & Thomas, 2011, p. 83). This was also an important factor for this particular study.

- **Educational and cognitive interventions**

These include interventions such as teaching strategies, classroom modifications, support and resources for teachers and cognitive and academic skills training (Paley & O'Connor, 2009, p. 260). Two South African studies fall into this category. In Paley and O'Connor (2009, p. 260), a pilot study by Adnams et al is discussed. Cognitive control therapy was given to a small group of school-aged children with FAS, aimed at teaching them strategies that would facilitate their ability to aquire and organize information more effectively. The results showed improvements in classroom behavior, academic achievement, writing and communication skills as reported by the educators, as well as improvements in self-efficacy, motivation, self-confidence and emotionality. The cognitive control therapy group and the control group, however, did not differ significantly (Paley & O'Connor, 2009, p. 260). Adnams et al. (2007) also conducted a study to demontrate the efficacy of a school-based language and literacy training intevention in a group of nine-year-old children with FASD. No significant gains by the study group over the control groups were found on general scholastic measures. Significant improvements in specific catergories of language and early literacy of the study group did however occur. These catergories included syllable manipulation, letter-sound knowledge, written letters word-reading and non-word reading and spelling (Adnams, et al., 2007, p. 404).

Studies like the above suggest that children with FASD can benefit from interventions aimed at remediating some of the impairments associated with prenatal alcohol exposure. There seem to be limits to the improvements, as mentioned earlier in this section, and additional studies to identify treatment responses such as IQ and level of alcohol exposure are suggested in the literature. Ongoing research in other areas of neurocognitive functioning, such as speech and language delays, executive dysfunction and specific learning deficits, is also encouraged (Paley & O'Connor, 2009, p. 261). Since sensory integration is also an area of neurocognitive functioning, the present study may prove to contribute to the current body of knowledge on intervention for children with FASD.

- **Parenting interventions**

Parenting a child with FASD is usually challenging for parents, particularly in the domains of behavioural, emotional and cognitive functioning. Programs to enhance the quality of the relationship between the parent and child, reducing parental stress and improving the parents' skills, are critical in an approach to individuals with FASD (Paley & O'Connor, 2009, p. 261).

- **Adaptive skills training**

Children with FASD show deficits across multiple domains of adaptive skills, as discussed in 2.2.5.3(e). There is a definite need for interventions promoting the development of age-appropriate adaptive skills to enhance independent living. Studies to promote social and safety skills have been carried out. O'Connor, et al. (2006) conducted a study to assess the efficacy of child friendship training (CFT) versus a delayed treatment control (DTC) for children with FASD (p. 639). The children in the

CFT showed evidence of improved knowledge of appropriate social behaviour, and parents reported important improved social skills with less problem behaviours in comparison to the DTC intervention. Gains were maintained over a three-month period. Teachers, however, did not report improved social skills in the classroom (O'Connor, et al., 2006, p. 639).

According to Coles, Strickland, Padgett and Bellmoff (2007, p. 518), teaching safety skills to children with FASD is challenging, given their cognitive limitation and characteristic behavioural problems. They conducted a successful study teaching fire and street safety through computer games which employed “virtual worlds”. The children showed knowledge of the game they were exposed to immediately after intervention, and on follow-up 72% were able to generalise the steps in a behavioural setting (Coles, Strickland, Padgett, & Bellmoff, 2007, p. 518).

- **Pharmacological interventions**

“Findings suggest that children with prenatal alcohol exposure exhibit significantly more psychopathology, including symptoms of anxiety, disruptive behaviours, and mood disorders when compared to children without alcohol exposure” (Walthall, O'Connor, & Paley, 2008, p. 69). They also seem to be at risk for substance use or abuse and psychiatric hospitalisation (Paley & O'Connor, 2009, p. 263). With these findings in mind, it is no surprise that children with FASD often receive pharmacological interventions. Research support for the efficacy of the medications presribed is however limited, with mixed patterns of findings (Paley & O'Connor, 2009, p. 263). Although some preliminary evidence indicates the efficacy of at least stimulant medications in some individuals with FASD, more research in this area is urgently needed according to the literature (Paley & O'Connor, 2009, p. 263).

- **Case management**

The mental health issues often seen in individuals with FASD make up a major focus of the case management needed for this population (Paley & O'Connor, 2009, p. 263). In children with FASD, case management is often necessary where they are at risk because of medical problems, including cardiac problems, skeletal defects, sensory deficits and dental problems (Paley & O'Connor, 2009, p. 264).

Case management seems to be more frequently used in the case of adults with FASD, to manage comorbid psychiatric issues, substance and alcohol abuse problems, sexuality and legal problems (Paley & O'Connor, 2009, pp. 263-264).

- **Treatments specifically related to sensory integration theory**

Two important studies for occupational therapists have been conducted in recent years, those by Nash et al. (2015) and Wells, Chasnoff, Schmidt, Telford and Schwartz (2012), on the use of the Alert Programme® as published or adapted, to improve the executive functioning and emotional problem-solving skills in children with FASD.

The Alert Programme® (Williams & Shellenberger, 1996, p. iv) "promotes awareness of how individuals regulate their arousal states and encourages the use of sensorimotor strategies to manage our levels of alertness." This program endeavours to improve knowledge of self-regulation and gives strategies to improve learning abilities, encouraging socialising within the work or play environments. It also aims at improving self-esteem, self-confidence and self-monitoring skills.

As discussed earlier, children with FASD face difficulties in almost all aspects of executive functioning. Cognitive and socio-affective executive functions also seem to be dependent on self-regulation abilities (Nash, et al., 2015, p. 192). Studies of children with brain injury (other than FASD) have shown that damage occuring between 0 and 3 years of age primarily affects the self-regulation component of executive functioning. Nash et al. (2015, p. 192) state that "given the timing of the early brain insult in children with FASD, it is not surprising that several researchers have proposed that self-regulation may represent a core deficit in this population."

Deficits in self-regulation in children with FASD are evident throughout their development, as discussed in 2.4.3.2, ranging from problematic sleep-wake cycles and fluctuating arousal states as infants to elevated reactivity, high levels of distractability and sensory processing difficulties as toddlers. Eventually higher order executive function disorders present at school age (Nash, et al., 2015, p. 192).

A study by Wells, Chasnoff, Schmidt, Telford and Schwartz (2012) had the overall therapeutic goal of improving executive functioning skills and emotional regulation related to the children's home and school environments. They incorporated components of the Alert Programme® strategies, used with pediatric traumatic brain injury patients, which included occupational therapy, family psycho-education as well as interventions designed to improve memory, emotional awareness and cause-and-effect reasoning. Parents were informed about FAS and ARND during the psycho-educational groups (Wells, Chasnoff, Schmidt, Telford, & Schwartz, 2012, p. 26). Results indicated significant improvements in executive and emotional functioning of the intervention group in comparison to the control group (Wells, Chasnoff, Schmidt, Telford, & Schwartz, 2012, p. 31). Wells, Chasnoff, Schmidt, Telford and Schwartz (2012, p. 32) make a very valid deduction from this study when they state that, unlike clinical testing settings, the children's everyday situations, such as home and school, are environments with multiple stimuli and constantly changing expectations. Children with prenatal alcohol exposure may be unable to use their available executive functioning skills in these complex environments and consequently become progressively more dysregulated as they are distracted or overwhelmed by stimulation in their environments. The "speed" analogy of the Alert Programme® was used to teach the children how to identify internal indicators of dysregulation and to use new modulating strategies to improve their self-regulation and emotional control within a group setting (Wells, Chasnoff, Schmidt, Telford, & Schwartz, 2012, p. 32).

The study by Nash et al. (2015, p. 202) aimed to determine whether a treatment program to improve self-regulation could also be generalised to other areas of executive functioning, behavior and social skills in children with FASD. In this study only the Alert Programme® was used, as treatment modality and feedback on progress were given to the parents afterwards to encourage translation of the therapeutic tools into the home and classroom practice.

Results similar to those of the study by Wells, Chasnoff, Schmidt, Telford and Schwartz (2012) have indicated improvements in parent-rated executive function abilities, accompanied by significant improvements in behavioural regulation, especially emotional control. The children showed the greatest improvements in simple functions, e.g. Nepsy inhibition-naming test item, rather than complex executive functions (Nash, et al., 2015, p. 202).

In an article related to and of importance for this study, Kodituwakku (2010) presents a neurodevelopmental framework for designing new therapies for children with FASD, after summarising published intervention studies. The framework assumes a neuroconstructionist view, which postulates “that reciprocal interactions between neural activity and the brain’s hardware lead to the progressive formation of intra- and interregional neural connections,” or simply put, experience alters the “brain hardware,” which leads to new experiences and further alterations of the neural systems (p. 717 & 722). Kodituwakku therefore proposes that the idea of interaction between experiences and the neural system offers a practical framework for understanding the neurocognitive profile of the child with FASD and the development of subsequent appropriate interventions (2010, p. 722).

He states that the effects of attention and self-regulation training are more extensive or far-reaching than those resulting in domain-specific training (p. 723). There is growing evidence that early intervention focusing on self-regulation and executive attention is very crucial, since:

- the effects self-regulation and attention training are more far-reaching than those of domain-specific training,
- the development of specific self-regulatory skills during preschool years predicts long-term developmental outcomes, including academic outcomes independent of IQ (Kodituwakku, 2010, pp. 723-724).

Kodituwakku (2010) suggests other principles for the treatment of children with FASD which could be of value for occupational therapy. He encourages practitioners to design a program that pays careful attention to the child’s overall cognitive-behavioural profile. He maintains that the processing and integration of information is a general deficit characterising the core of the cognitive-behavioural phenotype of the child with FASD (Kodituwakku, 2010, p. 723). He further suggests the use of strategies appropriate for the child’s developmental age and the use of enriched environments in a guided manner (Kodituwakku, 2010, p. 724).

These findings are important in the development of effective treatment interventions for the the FASD population. They offer the promise of an effective use of a sensory modulation program as an intervention to improve executive functioning deficits and emotional problem-solving skills. Other researchers also recommend that focus should be maintained on the creation and refinement of intervention programs (Wells, Chasnoff, Schmidt, Telford, & Schwartz, 2012, p. 33).

2.7 CONCLUSION

The first phase of the literature review focused on an overview of FAS/FASD, with emphasis on the history, diagnosis and burden of FASD in South Africa. The teratogenic effects of alcohol on the brain and behaviour were discussed extensively, with a short of overview of the realities of the child with FASD in the Cape Winelands, in the Western Cape Province of South Africa. This was followed by a discussion on FASD and occupational therapy. This section covered the scope of occupational therapy practice, taking in the influence of FASD on body functions and structures, as well as the eventual limiting effect on the occupations of the child with FASD. A short summary on the general role of the occupational therapist with regard to this population was also given. This was followed by a background of sensory integration, including the core concepts of Ayres Sensory Integration, patterns of SI dysfunction and the key SI abilities and difficulties/dysfunctions.

A discussion was then presented of the known sensory-motor and sensory integration difficulties of children with FASD, as well as the value of using the SI framework to evaluate these children. The possible contribution of FASD to the poor academic results of a large percentage of learners in the CWED was highlighted, and parallels were drawn between the neurobehavioral problems children with FASD face and the current problems experienced by the district's children.

Current interventions were finally discussed, with an emphasis on those related to sensory integration and the possibility of including similar principles in a local treatment program.

In conclusion, this chapter gave an extensive background of the child with FASD, faced with all the challenges and difficulties of coping with their occupations at home and in the current South African school system. The background provided by the literature was essential to this investigation into the sensory integration difficulties and dysfunctions of such children, allowing the researcher to explore and understand how these difficulties and dysfunctions might factor into the broader array of neurobehavioural problems experienced and the extent to which they impact on specific adaptive abilities.

In Chapter 3 the research methodology used in the investigation into the sensory integration difficulties and dysfunctions experienced by a group of children living in the Western Cape Province, South Africa, will be presented and discussed.

CHAPTER 3

RESEARCH APPROACH AND METHODOLOGY

3.1 INTRODUCTION

Chapter two discussed the literature in five phases, giving an overview of FASD in general and of the child with FASD in South Africa in particular (with attention focused on the child with FASD growing up in the CWED), of FASD and occupational therapy and education, as well as FASD and intervention.

An extensive overview of children with FASD highlighted the challenges and difficulties they are facing, specifically in the school system. Since many of the symptoms of sensory integration dysfunction have been reported in children with FASD, an in-depth discussion was included on sensory integration theory and studies examining components of sensory processing and sensory integrative functions of these children. The literature overview in Chapter 2 provided an essential background for an investigation into the sensory integration difficulties and dysfunctions of children with FASD in the CWED.

This chapter focuses on the research approach and methodology used in the study. The discussion on methodology includes the research approach, study design, population, sampling, pilot study, data collection, data analysis, error of measurement and ethical considerations.

3.2 RESEARCH APPROACH AND STUDY DESIGN

A quantitative observational analytical design was used to investigate and describe sensory integration difficulties and dysfunctions in children both with and without FASD.

3.2.1 Quantitative approach

A formal, objective and systematic approach of quantitative research (Leedy & Ormrod, 2010, p. 96) suited this research best. It aimed to confirm and develop existing knowledge about the sensory integration difficulties and dysfunctions of children with foetal alcohol spectrum disorders. An objective and systematic approach allows for the use of standardised measuring instruments to objectively measure the variables of interest (Leedy & Ormrod, 2010, p. 96), and was chosen to support this research in the process of data collection. Numerical data obtained were statistically analysed and subjected to deductive reasoning before conclusions were drawn about the sensory integration difficulties and dysfunctions of a group of children with FASD from the Cape Winelands in South Africa.

3.2.2 Observational analytical design

Quantitative observational studies focus on a certain aspect of behaviour which is to be quantified in some way (Leedy & Ormrod, 2010, pp. 182-183). Analytical studies set out to quantify the relationship between two factors, that is, the effect of an exposure on an outcome. To quantify the effect, one has to know the rate of outcomes in a control group, as well as in an exposed group (Centre for Evidence-Based Medicine, 2017). In an observational analytical study, the researcher does not actively change a factor or use an intervention (Centre for Evidence-Based Medicine, 2017). This study design was chosen since the effect of FASD on the cases was investigated without changing any factors or using interventions. The control group was used to quantify the effect of FASD on the sensory integration difficulties and dysfunctions of the cases.

3.3 RESEARCH POPULATION

A research population is the entire group of people or items which meet the criteria or characteristics set by the researcher (Bailey, 1997, p. 45). The research population in this study consisted of children both with and without FASD, as diagnosed during two parts of a longitudinal study conducted in the Cape Winelands, Western Cape, by Fetal Alcohol Syndrome Epidemiological Research – South Africa (FASER-SA), as described in 2.2.3. Children diagnosed with FASD will be refered to as the “cases” and children that did not receive a diagnosis of FASD will be refered to as “controls”.

3.4 SAMPLING

Sampling is the statistical process of selecting a subset (called a “sample”) of a population of interest for the purpose of making observations and statistical deductions about that population (Bhattacherjee, 2012, p. 65). Because of feasibility and cost limitations, one cannot study entire populations. Instead, a representative sample is selected from the population of interest for observation and analysis. It is extremely important to choose a sample of the population which is truly representative, so that the conclusions drawn from it can be generalized to the larger community (Bhattacherjee, 2012, p. 65). Criteria to ensure that the study would be reflective and representative of the characteristics of children with FASD were thus established. Sections 3.4.1 (inclusion criteria) and 3.4.2 (exclusion criteria) discuss the inclusion and exclusion criteria.

3.4.1 Inclusion criteria

"Inclusion criteria are a set of predefined characteristics used to identify subjects who will be included in a research study" (Salkind, 2010). Such criteria should be sensitive to the objective of the study and are crucial to its accomplishment. A proper selection of inclusion criteria has many advantages, among them optimising the external and internal validity, improving the attainability of the study, ensuring the homogeneity of the sample population, and minimising ethical concerns (Salkind, 2010). Criteria for diagnosing children both with and without FASD, for demographical requirements and for the requirements of the SIPT, were determined for this study.

The inclusion criteria selected for this study are set out in Table 3.1 below.

Table 3.1: Inclusion criteria

<table>
<tr><th colspan="2">INCLUSION CRITERIA</th></tr>
<tr><th>FASD Group (Cases)</th><th>Control Group</th></tr>
<tr><td>Diagnosed with FASD by an accredited diagnostic team using the IOM criteria reflecting the key diagnostic features of FASD.</td><td>Found NOT to be FASD after being subjected to the examinations of the same accredited diagnostic team using the IOM criteria reflecting the key diagnostic features of FASD.</td></tr>
<tr><th colspan="2">MATCHING CRITERIA FOR BOTH GROUPS</th></tr>
<tr><td colspan="2">Sex, race, age (±1 month), geographical area and family structure.</td></tr>
<tr><td colspan="2">Between the ages of 5 years and 8 years 11 months old, due to the fact that the SIPT was designed for use with children from 4 years to 8 years 11 months old. The inclusion age for this study started at 5-years of age, since the children enter Grade R at that age.</td></tr>
</table>

All children had to be from schools in the CWED, Western Cape, South Africa.
Availability of a suitable caregiver to complete the sensory profile in the company of the assigned therapist. This had to be the person who physically took care and spent time with the child in the same household. This included spending time together during activities of daily living such as feeding, dressing, grooming and socialising. The person had to be able to comprehend and answer questions about the child in a reliable manner.

3.4.2 Exclusion criteria

"Exclusion criteria are a set of predefined definitions that are used to identify subjects who will not be included or who have to withdraw from a research study after being included" (Salkind, 2010). Together, inclusion and exclusion criteria make up the desirable criteria that rule participants in or out of a research study. Exclusion criteria are also guided by the study objective and have important implications for the scientific accuracy of a study, as well as for assuring ethical principles (Salkind, 2010). As with the inclusion criteria, the researcher established criteria that would exclude those children who, if included, might negatively influence the validity of the study.

The following were the exclusion criteria for this study:

- Children with serious physical or neurological disabilities, such as spina bifida or cerebral palsy, blindness or deafness, were excluded, as these could have had an effect on the manner in which sensory information was processed. Since one of the measuring instruments was performance dependent, such conditions could have interfered with test performance. Most of the tests required sight, which would have made them impossible for a blind child.
- Children with severe intellectual disability were excluded, as this would have influenced the child's comprehension of test instructions and therefore limited participation.
- Children who were subjected to the Occupational Therapy Programme introduced in the Grade R classes during 2015 were also excluded, as the programme contained activities stimulating the vestibular, proprioceptive and tactile systems. The tests would not have given a true reflection of the children's initial ability to process and integrate sensory information since they had already been exposed to intervention in the past (2.5).

- Children not attending school as the SP School Companion, completed by the child's educator, formed part of the assessment instruments.

3.4.3 Sample size

Sample size was calculated on the basis of the practical considerations relating to the diagnosis of FASD in the FASER study, as well as age and geographical location of the children. The researcher's available time and funding also had to be taken into consideration. It was estimated that at most 30 matched pairs of children with and without FASD should be tested.

The initial sample size was computed as follows: The means and standard deviations published in Van Jaarsveld, Mailloux, Smith Roley and Raubenheimer (2014) were used as normative values for a South African sample. Positing that paired t-tests would be performed using a mean difference of one standard deviation, it was computed that between 4 and 13 pairs (N=8 to N=26) per SIPT group would be sufficient. However, for Fisher's exact tests these numbers might have been too small. Power calculations indicated that, with 15 pairs, most of the paired t-tests would have power approximating 95%, and, depending on the initial proportion and the size of the difference, power with 30 pairs for the Fisher's tests would range between 33% (for a proportion of .7 in the control group and .5 in the case group) to 97% (for a proportion of .85 in the control group and .35 in the case group). It was therefore decided to select 30 children from the FASD group and pair them according to the inclusion criteria with children not diagnosed with FASD.

3.4.4 Sampling process

Once the sample size of 60 had been established and the pilot study completed, two groups of children, one group with and one group without FASD (27 each – this excluded the 6 children from the pilot study), were selected through non-random sampling from the larger ongoing studies. FASER-SA made lists with the research number, sex, date of birth, race, final diagnosis and school of the children, with informed consent by the parents to participate in further research projects, available to the researcher. Twenty-seven children were selected from each of the two research studies (2.2.3) to ensure a wider age group and bigger demographic area. Since the two studies are focused on two different demographic areas in the Cape Winelands

Education District, this study was also divided in two phases in order to simplify the logistics. Since the FASER-SA studies also included typically developing controls (the children not diagnosed with FASD), the control group for the present study was selected from the same control group, ensuring that the children in this study were not on the fetal alcohol spectrum. Once the children were selected, the research numbers were sent to FASER-SA who then released the names of the children to the researcher. In most instances, the caregiver's information was also available. Demographic information was obtained either from the caregivers during the interview arranged for completion of the Sensory Profiles or requested from the FASER-SA Maternal Questionnaire information.

The inclusion of a matched control group was essential, since the study population was mainly from low socio-economic communities which in itself may have had a negative effect on the children's sensory integration. Such integration is developed optimally in a stimulating environment with ample opportunities for interacting and adapting the body and brain to physical challenges from the environment (Ayres, 2005, p. 7). Including a matched control group in the study and describing both gave valid information on the unique and distinctive patterns of sensory integration difficulties and dysfunctions among children with FASD (Howell, Lynch, Platzman, Smith, & Coles, 2006, p. 124).

3.5 RESEARCH PROCEDURE

The research procedure is shown in Figure 3.1 below.

PERMISSION FOR RESEARCH

Researcher	- Obtained research approval from the Health Sciences Research Ethics Committee, UFS
Researcher	- Obtained permission from Department of Education for research (Appendix C)
Researcher	- Obtained permission from Director of Cape Winelands Education District (Appendix D)
Researcher	- Obtained permission from school principals; confirmed date, times and logistics for pilot study

PREPARATION FOR PILOT STUDY

Biostatistician	- Selected 3 FASD and 3 NOT FASD children from the study population
Researcher	- Obtained children's names and relevant caregiver information from FASER-SA
Parents	- Obtained Parental Consent (Appendix E)
School principals	- Confirmed dates, times and logistics
Educators	- Informed individually how to complete SPSC

PERFORM PILOT STUDY

Children	- Obtained Children's Assent (Appendix F)
Researcher	- Assessment: SIPT
Educators	- Completed: SPSC
Researcher	- Assessment: SP with parents

PREPARATION AND EXECUTION - PHASE 1

Biostatistician	- Selection of sample
Researcher	- Pre-test arrangements
Educators	- Completed SPSC
Researcher and additional therapist	- Assessments: SIPT

PREPARATION AND EXECUTION - PHASE 2

Biostatistician	- Selection of sample
Researcher	- Pre-test arrangements
Educators	- Completed SPSC
Researcher and additional therapist	- Assessments: SIPT

Researcher and additional therapist	- Scored SIPTS
Researcher	- Captured all data
Researcher	- Reviewed and interpreted results
Researcher	- Wrote dissertation
Language editing	
Final technical editing	

Figure 3.1: Research procedure

3.6 PILOT STUDY

A pilot study was done to determine the feasibility of the study with regard to the test procedures and infrastructure needed. It also served to detect flaws in the protocol, so these could be rectified before the real study started (Joubert, Bam, & Cronjé, 2008, p. 52). The pilot study was only performed once the HSREC of the Faculty of Health Sciences, University of the Free State, had given their approval for the study to commence.

The researcher therefore used the pilot study to determine the viability of the study, to ensure that all processes ran smoothly, and that the study would as far as possible secure the achievement of the objectives.

Six participants in total (three with FASD and three NOT FASD) were randomly selected from the list provided by FASER-SA for the pilot study. The selection was done by the biostatistician from the Department of Biostatistics, Free State University, who was involved in this study. All the participants of the pilot study were assessed with the measuring instruments selected for the project. The researcher tested all six children, since no additional therapists were available to help with the assessments at that stage. The primary caregivers were interviewed and they, as well as the educators, completed the standardised questionnaires. The caregivers were individually guided through their questionnaires, and the educators each received verbal instructions on completing the SPSC. The researcher was available to answer questions about any of the items they did not understand. The six children were part of the group officially diagnosed by FASER-SA, from which the study sample was selected, and the selection criteria were the same as for the official study participants.

The data obtained from the children in the pilot study were included as part of the data from the 30 children from the Wellington area in the main study, since no adjustments were made to the measuring techniques or process.

The pilot study indicated the following:

a) Caregiver appointments: It became apparent that parents needed a few dates and time options from which to choose when they were unable to come in the afternoons as initially planned. This caused major difficulties for the researcher, since time was very limited. Reminders of their appointments were sent out during the main study, since a few of the pilot study parents/caregivers had not shown up for the original appointments.

b) Questionnaires: Due to the low educational level of most of the caregivers, they were unable to read and complete the SP independently. We therefore decided to go through the questionnaires with the parents/caregivers on an individual basis, making sure that they understood each question. Educators were able to complete the SPSC independently, but it became evident that the larger the classes, the harder it was for them to pinpoint specific behaviours as described in the questionnaire. This led to more general answers, such as "Occasionally" or "Seldom", as included on the questionnaire. This however was a reality that the researcher could not change or adapt.

c) Data collection process: No changes were made.

d) Additional therapists: The researcher was able to recruit one occupational therapist, trained in the use of the measuring instruments, to assist with the data collection. The fact that only one person was available and willing to assist markedly extended the time frame within which the researcher initially planned to complete the data collection process.

3.7 MEASUREMENT

Measurement is a process whereby the data relating to any phenomenon are limited in order to be interpreted and compared to a particular standard (Leedy & Ormrod, 2010, p. 21). In this study, the phenomenon in question was the sensory integration difficulties and dysfunctions of children with FASD.

3.7.1 The measurement Instruments

The measurement instruments chosen for a specific research project provide the basis upon which the entire research endeavour rests (Leedy & Ormrod, 2010, p. 91). Faulty measurement tools will be of little value in solving the problem under investigation, and it is thus important to use instruments with a reasonable degree of validity and reliability (Leedy & Ormrod, 2010, p. 91).

Three standardised assessment instruments, the Sensory Profile (SP), Sensory Profile School Companion (SPSC) and the Sensory Integration and Praxis Tests (SIPT), were used to collect data in this study. Two of these were standardised questionnaires which were completed by the primary caregiver and educator of each child. All three instruments were developed in the United States of America and were used in the absence of similar valid and reliable instruments developed for South African children. While studies have been conducter to validate the use of the SIPT on the South African population (cf. p. 109), the Sensory Profiles have not been standardised on the South African population, but are currently the preferred instrument for use in South Africa.

3.7.1.1 Standardised questionnaires:

The Sensory Profile (SP) and Sensory Profile School Companion (SPSC) provide information on children’s responses to commonly occurring sensory events (Dunn, 1994, p. 967). With the assistance of the researcher and the additional occupational therapist, they were completed by the primary caregivers and educators. Both the SP and SPSC were used to collect comprehensive data on the children’s responses to sensory experiences both at home and at school. The child has to meet different demands relating to focus and behaviour at home and school, since the sensory environments can differ significantly, so it was important to obtain information from both. In the case of the SP, both the Sensory Profile Summary Score Sheet and Worksheet for Calculating Quadrant Scores on the Sensory Profile were used to calculate scores.

a) Sensory Profile (Winnie Dunn, 1999)

The **Sensory Profile** was originally developed as part of research to test the application of sensory integration theory in consultative services in classrooms (Dunn, 1999, p. 13; Kemmis & Dunn, 1996; Dunn, 1994). Researchers used these studies to determine the children's sensory needs and sensory processing difficulties in order to enable consultative occupational therapy school services to address these needs in the classroom. These studies by Dunn (1999, p. 13) showed that it was possible to address sensory processing successfully in the context of the daily routines and expectations of the classroom. Therapists showed an interest in discovering more about the SP as an assessment tool to aid in designing effective interventions. With the support of organisations such as The American Occupational Therapy Foundation, graduate students worked on the development of the SP from 1993 to 1999 (Dunn, 1999, p. 13).

The SP offers professionals a method to measure a child's sensory processing abilities and to profile the effect of sensory processing on functional performance in the daily life of the child (Dunn, 1999, p. 1). The profile is most appropriate for children 5-10 years of age.

The **Sensory Profile** is a judgment-based caregiver **questionnaire**. Each item is written as a behavioural statement, e.g. "Holds hands over ears to protect ears from sound," and describes the child's responses to different sensory experiences. The caregiver with daily contact with the child completes the questionnaire by reporting the frequency of different behaviours. This is done by using a 5-point Likert scale (Always, Frequently, Occasionally, Seldom or Never) (Dunn, 1999, p. 1). The therapist then scores the caregiver's responses on the questionnaire. Certain patterns of performance are indicative of difficulties with sensory processing and performance. The SP consists of 125 items grouped into three main sections: Sensory Processing, Modulation, and Behavioural and Emotional Responses (Dunn, 1999, p. 1). The summary score sheet includes a **Factor** Summary which "reveals patterns related to the child's responsivity to stimuli in the environments" (Dunn, 1999, p. 29) as well as a **Section** Summary with provides information on the child's sensory processing, modulation and behavioural/emotional response abilities (Dunn, 1999, p. 29).

Problems with sensory processing may raise barriers to everyday functional performance and behaviour. The SP was included as an assessment instrument of choice in this research because it links sensory processing to daily functional performance as well as to behaviour (Dunn, 1999, p. 3).

The Worksheet for calculating quadrant scores on the Sensory Profile (Dunn, 1999) has also been used in this study. Dunn proposed a model for sensory processing, based on a child's neurological thresholds and behavioural response patterns (Dunn, 1999, p. 32). This model illustrates the neurological thresholds as the vertical axis, with low thresholds at the bottom and high thresholds at the top, anchoring the behavioural response continuum on the horizontal axis, indicating behaviour in accordance with or counteracting the neurological threshold. Children acting more in accordance with thresholds tend to respond more passively to the the environment, while children who counteract their thresholds, tend to actively work to oppose the thresholds (Dunn, 1999, p. 32). The continuum interacts with each other to create four basic quadrants of responsivity namely poor registration, sensitivity to stimuli, sensation seeking and sensation avoiding (Dunn, 1999, p. 32).

b) Sensory Profile School Companion (Winnie Dunn, 2006)

The **Sensory Profile School Companion (SPSC)** provides a standardised assessment of a student's sensory processing abilities and gives an indication of the educator's association with the student's functional performance in the classroom and school environments (Brown, Morrison, & Stagnitti, 2010, p. 59). It is completed by the educator who has routine contact with the child by reporting the frequency of different behaviours. This is done using a 5-point Likert scale (Always, Frequently, Occasionally, Seldom or Never). Certain patterns of performance are indicative of difficulties with sensory processing and performance. The SPSC consists of 62 items, covering five domains: auditory, visual, movement, touch and classroom behaviours. The school companion is most appropriate for children from 3 years to 11 years 11 months.

Validity and reliability of the Sensory Profile and Sensory Profile School Companion

The validity of a measuring instrument is the "extent to which the instrument measures what it is intended to measure," and the reliability is the extent to which it "yields consistent results when the characteristic being measured hasn't changed" (Leedy & Ormrod, 2010, p. 28 & 93).

Content validity was established while developing the SP by ensuring that the test sampled the full range of children's sensory processing behaviours and that the items were appropriately assigned to the different sections (Dunn, 1999, p. 52). Sensory processing literature, sensory histories from the children, as well as expert reviews from therapists experienced in applying sensory integration theory to practice, were all used in this process (Dunn, 1999, p. 52).

The SP has a moderate to large and meaningful correlation with the SFA (School Function Assessment) (Dunn, 1999, pp. 53-54) and a moderate level of convergent validity with the Sensory Processing Measure (Brown, Morrison, & Stagnitti, 2010, p. 56).

The reliability of the SP was estimated using internal consistency, which indicates the extent to which the items in the different sections measure a single construct. Cronbach's Alpha was calculated for each section of the SP. The values for the various sections ranged from .47 to .91, meaning that reliability ranged from average to good (Dunn, 1999, p. 47; Polit & Beck, 2010, p. 375).

Two statistical methods were used to estimate the reliability of the SPSC – internal consistency and test-retest reliability. Cronbach's Alpha was used to calculate the internal consistency of the SPSC and alpha coefficients ranging from .83 to .95, indicating a high degree of internal consistency (Dunn, 2006, p. 83). Test-retest reliability seemed to be good to excellent, with reliability coefficients ranging from .80 to .95 (Dunn, 2006, p. 84).

Validity related to the content of the SPSC was done extensively through exploratory studies, interviews and collecting data from pilot studies to ensure that the test adhered to Dunn's Model of Sensory Processing, that the test sampled the full range of learners' sensory processing behaviours at school, and that the items were assigned correctly (Dunn, 2006, p. 88). Although the validation process of the test was not completed by the publication of the test, there is enough evidence to claim that the SPCS can provide reliable and valid information about a learner's sensory processing abilities (Dunn, 2006, p. 99).

The two sensory profiles have not formally been translated to Afrikaans for the purposes of this research. Although all the educators involved in completing the Sensory Profile School Companion are Afrikaans speaking, they have all matriculated with English as their second home language. Apart from that, they have also obtained tertiary teaching qualifications. The researcher was therefore of the opinion, that it would be within their capabilities to understand and complete the profile. They were also informed of the availability of the researcher and the assistant occupational therapist to answer any questions that might arise. The researcher performed all the individual interviews with parents/caregivers, completing the Sensory Profile Caregiver Questionnaires herself. This was done in Afrikaans since not all the caregivers were literate enough to understand the English questions. Since the Sensory Profile is a questionnaire and not a standardised test, the reasoning was that it was more beneficial to obtain accurate answers than to use the original language of the profile, with an inaccurate outcome. The fact that the researcher was familiar with this instrument and have used similar explanations with all the caregivers, secured more test reliability. Further research into the translation of the sensory profiles is recommended.

3.7.1.2 Standardised instrument

The Sensory Integration and Praxis Tests (SIPT) were administered by the researcher and an occupational therapist who was recruited to assist with data collection. Both occupational therapists involved in the testing procedure had completed the South African Institute for Sensory Integration (SAISI) training courses in sensory integration testing and interpretation inclusive of the SIPT, as this is a measurement tool that calls for postgraduate training.

a) **Sensory Integration and Praxis Tests - SIPT** (Ayres, 2004) (4 years – 8 years 11 months)

Evolved over three decades of research, the **SIPT** contribute to the clinical understanding of children with difficulties in learning and/or behaviour (Ayres, 2004, p. 1). The SIPT "were designed to assess several different practic abilities, various aspects of the sensory processing status of the vestibular, proprioceptive, kinesthetic, tactile, and visual systems, and the major behavioural manifestations of deficits in integration of sensory inputs from these systems" (Ayres, 2004, p. 1). Originating in the USA, the SIPT were designed for use with children from 4 years to 8 years 11 months of age.

The validity of the tests to discriminate between typically developing children and those with learning, behavioural, attention or suspected sensory integration deficits, has been researched over a long period of time. While continued research, as recommended by Mailloux et al. (2011, p. 150), is still necessary, the SIPT stems from one of occupational therapy's most developed frames of reference – the Ayres Sensory Integration® (Mailloux, et al., 2011, pp. 144, 150). The subtests of the SIPT are reliant on performance, and severe neuro-motor problems such as spasticity will therefore interfere with performance on at least half of the tests (Ayres, 2004, p. 1). All the tests, except for those evaluating tactile and kinesthetic function, as well as standing balance, require sight. No verbal response is required for the SIPT, and tasks can usually be communicated non-verbally. Background information, including a brief history, the presenting problems, as well as relevant medical information, will assist in bringing the SIPT data into proper perspective. All SIPT subtests are individually administered (Ayres, 2004, pp. 1-2).

The SIPT consists of 17 tests, divided as follows:

Table 3.2: SIPT Subtests (Ayres, 2004, pp. 2-8); (Bundy, Lane, & Murray, 2002, p. 172)

Primary domain	Test	Description
Form and Space Tests • Measures visual perception	Space Visualization (SV)	Ability to mentally manipulate objects in space.
	Figure-Ground (FG)	Visually separate foreground from a rival background.
	Motor Accuracy (MAC)	Eye-hand coordination and control of movement.
Somatic Processing Tests • Sensory input from the brain to the body.	Kinesthesia (KIN)	Muscle and joint sense.
	Graphesthesia (GRA)	Perception of the spatial and temporal qualities of a series of tactile stimuli.
	Finger Identification (FI)	Differentiate fingers from tactile stimuli only.
	Localization of Tactile Stimuli (LTS)	Localize single tactile stimuli.
	Manual Form Perception (MFP)	Matching of block held in hand with visual counterpart or block held in other hand.
	Standing and Walking Balance (SWB)	Static and dynamic balance on one or both feet with eyes open or closed.
Vestibular Processing Tests	Postrotary Nystagmus (PRN)	Duration of vestibulo-ocular reflex.
	Standing and Walking Balance (SWB)	Static and dynamic balance on one or both feet with eyes open or closed.
Praxis Tests	Praxis on Verbal Command (PrVC)	Translate verbal commands into acts of praxis.
	Constructional Praxis (Pr)	Three-dimensional construction.

	Sequencing Praxis (SPr) Oral Praxis (OPr) Postural Praxis (PPr) Design Copying (DC)	Remembering and executing a series of hand and finger movements. Imitation of movements and positions of tongue, lips and jaw. Assuming different and unfamiliar body positions. Copying designs.
Bilateral Integration and Sequencing Tests	Bilateral Motor Coordination (BMC)	Move both hands and both feet in smooth integrated patterns.

Ayres conducted various clusters analysis through which four dysfunctional groups were identified (Ayres, 2004, pp. 140-145):

- Low-average bilateral and sequencing deficits
- Visual and somatodyspraxia
- Dyspraxia on verbal command
- Generalized sensory integrative dysfunction

As discussed in 2.4.2.3, the patterns of dysfunction were updated by Mulligan (1998) and expanded on by people like Dunn and Miller (Schaaf, et al., 2010, p. 111). The patterns of sensory integration dysfunction used in this study, as described in 2.4.2.3, were identified by Mailloux et al. (2011, p. 143). They are Visuo- and Somatodysparaxia, Vestibular and Proprioceptive Bilateral Integration and Sequencing, Tactile and Visual Discrimination. The patterns of Visuo- and Somatodysparaxia, Vestibular and Proprioceptive Bilateral Integration and Sequencing have remained consistent over the years of research. In the pattern of Tactile and Visual Discrimination, described in the research of Mailloux et al., a child scores poorly on the tactile discrimination tests, as well as in one or more of the other sensory systems. Loadings of praxis, bilateral and postural measures are not present in this pattern (Mailloux, et al., 2011, p. 149). Indications of dysfunction in this pattern include difficulty moving through space without bumping into objects and trouble with performing fine motor tasks or identifying something in the hand without looking. Such children also find difficulty in copying from the blackboard, keeping track of a place on a page, or following a moving object with the eyes (Schaaf, et al., 2010, p. 139).

The SIPT assess the sensory processing status of five sensory systems, as well as manifestations of sensory integrative deficits, are cited in the sensory integration literature as the ”most comprehensive and statistically sound measure for assessing some important aspects of sensory integration” (Bundy, Lane, & Murray, 2002, pp. 170-171), and are regarded as the “gold standard” for evaluating sensory integration and praxis (Windsor, Smith Roley, & Szklut, 2001, p. 218). All these factors made them a suitable instrument for obtaining data relevant to achieving the aim of this research. It could be argued that the SIPT are not relevant for children with cognitive impairments, but Windsor, Smith Roley and Szklut (2001, p. 218) claim that, although the SIPT are not appropriate for children with significant cognitive impairments, the information they provide may still be valuable in assessing children with diagnoses of mild cognitive delays. All the children who took part in this study were in mainstream schools at the time of the research. Some of the older children, however, did receive extra academic support from the learning support educator assigned to the school.

In their comprehensive study investigating sensory processing, school performance and adaptive behaviour in young school-age children with FASD, Jirikowic, Olson and Kartin (2008a) recommended the future use of a more comprehensive standardized measure for sensory integration abilities. Although the SIPT are a time-consuming measure for which specialised training is needed, they have been described as the “most comprehensive and statistically sound means for assessing some important aspects of sensory integration” (Bundy, Lane, & Murray, 2002, p. 170). For these reasons, the researcher decided to include them among the measuring instruments used to address the research aim.

Reliability and validity of the SIPT

The two types of test reliability reported for the SIPT are test-retest and inter-rater reliability. Several researchers conducted test-retest reliability studies on the SIPT and results indicated that the group of praxis tests, including those such as Postural Praxis and Sequencing Praxis, had the highest test-retest reliability compared with the reliability of other tests within the acceptable range (Mailloux, 1990, p. 591). Four of the tests, namely Postrotary Nystagmus, Kinesthesia, Localization of Tactile Stimuli and Figure-Grond Perception, had a low test-retest reliability (Ayres, 2004, p. 209). Inter-rater reliability coefficients for most of the SIPT scores were greater than .90 when administered by trained therapists (Ayres, 2004, p. 212).

To be a valid test, the SIPT needs to achieve its primary aims, which are to evaluate the sensory integrative and praxis abilities of children and guide the treatment thereof (Ayres, 2004, p. 171). The construct validity of the SIPT encompassed a large body of work developed over several decades of clinical work and research (Mailloux, 1990, p. 593). Mailloux (1990, p. 593) summarises the solid base on which the SIPT's content validity is built when she states: "The content validity for SIPT was established through the work that led to the development of the SCSIT, use and refinement of the SCSIT, use of current research in related areas." All of the prior research led to the development of the SIPT, representing a large body of work.

Research done by using the SIPT on a South African population of 223 children from throughout the country, confirms the existence of patterns of sensory integration dysfunction consistent with patterns identified through research worldwide over the past 50 years. The patterns identified were similar to those found in the US, where the SIPT originated (Van Jaarsveld, Mailloux, Smith Roley, & Raubenheimer, 2014, p. 5). The fact that it was found to identify similar patterns of dysfunctions in children identified as experiencing sensory integration challenges and growing up in South Africa, confirms it as a useful instrument to use on South African children, although research is still needed on children growing up in low socio-economic settings in South Africa.

3.7.2 Data collection process

PREPARATION FOR PHASE 1 OF STUDY

Biostatistician	- Selected 12 FASD and 12 NOT FASD children from the Wellington study population
Researcher	- Obtained children's names and relevant caregiver information from FASER-SA
Additional Therapist	- Prepared therapist for data collection - consolidated and adjusted work plan after Pilot Study
Educators	- Revised and answered questions about completion of SPSC
Parents	- Obtained Parental Consent (Addendum E)
School principals	- Obtained permission, confirmed dates, times and logistics

PHASE 1

Children	- Obtained Children's Assent (Addendum F)
Occupational Therapist	- Assessment: SIPT (Mornings)
Researcher	- Assessment: SP with parents (Mornings) Assessment: SIPT (Mornings)

PREPARATION FOR PHASE 2 OF THE STUDY

Biostatistician	- Selected 15 FASD and 15 NOT FASD children from the BRAM study population
Researcher	- Obtained children's names and relevant caregiver information from FASER-SA
Educators	- Informed individually how to complete SPSC
Parents	- Obtained Parental Consent (Addendum E)
School principals	- Got permission, confirm dates, times and logistics

PHASE 2

Children	- Obtained Children's Assent (Addendum F)
Occupational Therapist	- Assessment: SIPT (Mornings)
Researcher	- Assessment: SP with parents (Mornings)
	- Assessment SIPT (Mornings)

DATA MANAGEMENT PHASES 1 AND 2

Researcher and therapist	- Scored SIPTS
Researcher	- Captured all data (SIPTS and Sensory Profiles)

Figure 3.2: Data collection process

The data collection process as shown in Figure 3.2 took place as follows:

The original intention was to randomly select participants from the larger FASER-SA group. However, once the criteria for both inclusion and exclusion and the matching requirements had been applied, and consent had been obtained, the study population of FASER-SA participants had been exhausted. This necessitated the use of non-random sampling of eligible participants in order to obtain a statistically valid study population. More diagnosed children from the Wellington School Study finally became available in May 2016, but time and money constraints did not allow for taking on more children. For the purpose of this study, therefore, the sampling is considered as non-random.

A Biostatistician from the University of the Free State was responsible for the selection of children from the two FASER-SA studies, as described in Chapter 2.2.3 and 3.3. The study was executed in two parts. The first part was conducted in Wellington schools for which 24 children (12 children with FASD and 12 matched children not diagnosed with FASD) were selected from the 2014 Wellington Schools study. The other three children in each group were part of the pilot study as described in 3.6. The second part was conducted in the BRAM area and for this leg of the research 30 children (15 children with FASD and 15 matched children not diagnosed with FASD) were selected from the population of the Newborn Screening Study.

Once the children had been identified, the relevant school principals were contacted to inform them which children had been chosen for the research. Pre-research arrangements with the principals included allotting space in which to do the assessments, deciding dates and times, as well as access to research participants for testing purposes.

Parental consent was obtained by sending the information and consent letters (Addendum E) with possible dates for interviews to the parents via the class educator. In the case of a parent refusing consent or not returning the letter, the biostatistician was informed and another child fitting the criteria was selected.

The assisting occupational therapist was prepared for data collection by the researcher during a training session in which all relevant matters were discussed to ensure valid and reliable data collection.

Educators were shown how to complete the SPSC in individual sessions, either by the researcher or the assisting therapist. Questions about the content of the questionnaire were answered to ensure valid and reliable data collection.

The SIPT assessments took place in the mornings at the schools. All assessments were done according to the guidelines in the administration manual of the SIPT. This included specifications on the test setting, test materials, administration time and placement and handling of the materials. All the assessments were performed in Afrikaans which is currently one of the two languages the SIPT is available in in South Africa (Van Jaarsveld, Mailloux, & Herzberg, 2012, p. 13).

With regard to the setting, the SIPT was administered individually in quiet (as quiet as the schools would allow) well-lit rooms, as free from distractions as possible. A table was provided with two matching children's chairs, all of appropriate height (Ayres, 2004, p. 11). Where possible, two children were tested per day. Since the researcher did all the interviews with the caregivers to complete the SPs, this was often not possible. The assisting occupational therapist, however, was usually able to assess two children per day. The children were allowed a break halfway through the test, during which they were allowed to play outside and eat their food.

Selecting children from two different sub-studies of FASER-SA made it possible to cover a wider age group, as well as a bigger geographical area in the CWED.

Data management

The researcher and assisting occupational therapist were responsible for scoring the SIPTS of the children they tested. It was the responsibility of the researcher to capture all data from the Sensory Profiles and SIPTs into the relevant computer programmes. The data from the Sensory Profiles were captured in an Excel programme, with the help of a co-data capturer to ensure accuracy. Since the SIPT data are captured in a clinician-administered computerised scoring programme, (which forms an intergral part of the SIPT), this was done by the researcher herself.

3.8 DATA ANALYSIS

The Sensory Profiles and Sensory Profile School Companions raw and standardised scores, as well as the demographic data, were captured by the researcher in an Excel spreadsheet. The SIPT raw scores were captured in the clinician-administered computerised scoring programme. Data were analysed by the Department of Biostatistics, University of the Free State, using SAS/STAT© version 13.2 of the SAS System for Windows. Demographic data were reported using frequencies and percentages, or, where appropriate, means and medians.

The various test scores (both raw and standardised) were reported, their means indicated with standard deviations, medians and ranges, as well as 95% confidence intervals.

Because children of the two groups were matched in age and gender, differences between the typical and FASD children were computed using one-sided, paired t-tests, computed on the raw scores for each subscale. Categorical variables were analysed by means of cross-tabulations with chi-square analysis and Fisher's exact p-values.

It has been decided not to adjust the test results for the five SIPT tests (DC, OPr, BMC, SWB and MAc) with a half a standard deviation to the negative side as suggested by Van Jaarsveld (2011, p. 17), for the age group 6 years to 8 years 11months since the children from this study were mostly from low socio-economic backgrounds. The majority children from both South African studies (Van Jaarsveld, Mailloux, &

Herzberg, 2012; Van Jaarsveld, Mailloux, Smith Roley, & Raubenheimer, 2014) were primarily from middle to high socio-economic settings and therfore it was decided not to adapt the scores but to use the data from the standardised sample as is. Research has indicated that children from low socio-economic settings do experience sensory integration difficulties, although the specific patterns of dysfunction still need to be confirmed (Van Jaarsveld, Mailloux, Smith Roley, & Raubenheimer, 2014, p. 5).

3.9 METHODOLOGICAL AND MEASUREMENT ERRORS

Methodological and measurement errors can occur during the research process and can lead to incorrect conclusions (Joubert, Bam, & Cronjé, 2008, p. 44). Errors of this nature can be due to variation, bias, or confounding variables.

3.9.1 Measurement errors

Measurement errors were possible with any of the components that were involved in this research. The components involved in the measuring process in this study were:

- The researcher and the assisting occupational therapist,
- the parents and educators,
- the children with FASD and those identified as not having FASD as well as the
- measuring instruments (Questionnaires and standardized test).

Types of measurement error:

a) Random error (variation)

Random errors usually result from the test administrator's inability to take the same measurement in exactly the same manner so as to get the exact same number/score. This could influence the reliability of the results. Random error can be caused by the observer, the participant or the instrument (Joubert, Bam, & Cronjé, 2008, p. 45).

Variation associated with the observer that needed consideration:

- **Inter-observer variation** can occur in a study when different observers measure the same characteristic in the same participant. In this study, it could have occurred when caregivers and educators had to complete the

Sensory Profiles (SP and SPSC) on the same child. To minimize this, the researcher and therapist took time to explain the questions and gave similar instructions to the educators. The researcher did the same with all the caregivers. Consensus was reached with the therapist during the training period about the amount and type of specific assistance allowed both during the completion of the questionnaires and in answering additional questions. The pilot study also eliminated some of the problems before the actual study commenced.

b) Systematic error (bias)

This occurs when the measurement of a characteristic gives values which differ systematically from the true value (Joubert, Bam, & Cronjé, 2008, p. 46). The following bias was a possibility in this study:

- **Non-responder bias**

  The possibility existed that some of the parents approached to take part in the study would refuse due to the sensitivity around the FASD diagnosis. This happened in the case of a few caregivers of children not diagnosed with FASD. It had to be explained to them why it was important to have a control group and that their child would benefit from a free, specialised occupational therapy evaluation. The caregivers understood that children with severe problems would be referred to a relevant district-based team member, such as a psychologist, social worker or inclusive education team occupational therapist, as defined by referral policies. They were also motivated by the idea that all the children would potentially benefit from an intervention programme following on the study, since it would be designed for use in the classroom and not for individual use. It also became evident that the rural caregivers were often negligent in sending the letters back to the class educator as requested. In some cases, the researcher only received a response after the educator had sent follow-up messages to remind the caregiver.

- **Membership bias**

  A sample of children with FASD in the Cape Winelands Education District does not represent all children with FASD. Samples are often selected from such subgroups to simplify sampling (Joubert, Bam, & Cronjé, 2008, p. 47). In this case, however, the CWED was the area of employment of the researcher and was thus the obvious choice of a research setting.

- **Drop-out**

  Had more than 20%, either of the children with FASD or the typically developing children, been unable to complete the study, the results may have been influenced to such an extent that they did not reflect the truth (Joubert, Bam, & Cronjé, 2008, p. 48). Drop-out could occur during the sampling or measurement phases of the research process. It could also occur due to parents withdrawing their children from the study, parents not showing up for interviews, or children absent from school. In order to prevent drop-out, follow-up appointments had to be made for those caregivers and participating children who did not show up for their first appointment. Follow-up appointments had to be made for several caregivers who had not shown up for their first appointments. Only one child was replaced, after being absent from school on a number of occasions during the researcher's visits.

- **Recall bias**

  Failure of memory by the parents could result in inaccurate information given in questionnaires. This could negatively influence the reliability of the information from the questionnaires. This possibility was taken into account when deciding who was the child's primary caregiver. In rural areas, this role is often taken over by grandparents. It was thus important to involve a primary caregiver who had the most recent knowledge of the child taking part in the research (See Inclusion Criteria in 3.4.1).

- **Observational bias**

  Observations are always vulnerable to bias. Factors which may influence objective observation are:

  - emotions, prejudices and values of observer
  - personal views

- anticipation of what is to be observed
- hasty decisions (Polit & Beck, 2010, p. 358).

Observational bias can be reduced to a minimum by careful training of the observers (Polit & Beck, 2010, p. 358). The occupational therapist who assisted in this research had been trained in the use of the SIPT. Administering and interpreting the SIPT is a postgraduate training course presented for occupational therapists by SAISI and ensures uniformity to a large extent with regard to instructions, administration of the test and observations made during the test. The educators were guided in the process of completing the Sensory Profiles, and the caregivers were also individually guided through the questionnaire by the researcher to ensure that they all understood the questions in exactly the same way. However, as described in 3.6(b), it was observed that the educators of the classes with an average of 40 children tended to allocate higher, more general scores on the SPSC, unless the child's behaviour or academic performance were extremely good or bad.

METHODS TO LIMIT VARIATION AND BIAS IN THIS STUDY:

- Only one additional occupational therapist assisted with the data collection.
- The assisting therapist was fully certified in the administration of the SIPT and was familiar with the use of the SP questionnaires.
- Only standardized tests and observations were used.
- Inclusion and exclusion criteria as well as measurement procedures were clearly defined in advance and used consistently and accurately during the study. Information was collected according to standardized questionnaires (See Appendices J & K).
- Quality checks were made by the researcher throughout the evaluation period to ensure that she and the assisting therapist maintained a high level of accuracy in the test administration and scoring. Use of a co-data capturer for the SP and SPSC data also ensured as much accuracy as possible.
- Parents were fully informed about the value of the study to reinforce their continuous participation (Appendix E).

- One extra assisting occupational therapist took part in the study to reduce the first therapist's exhaustion. It was still a daunting task and certainly not ideal to assess 60 children with only one assistant, but no other willing therapists could be found. To prevent exhaustion of the participants, the children's assessments were scheduled with a break in the middle. The duration of the sessions was planned according to their attention span and endurance. Some of the children needed more than one break to regain focus.

3.9.2 Confounding variables

A confounding variable can influence a study, leading to results that are not true (Joubert, Bam, & Cronjé, 2008, p. 50). One of the ways to limit confounders in this study was careful selection of the matched sample. The group of children with FASD and the typically developing group were matched in terms of sex and age (±1 month) and geographical area. This allowed the researcher to make comparisons between the two groups, and to ascribe any differences in sensory integration found between the two groups primarily to the FASD diagnoses of children in the FASD group and not to other confounding variables.

Differences in the levels of education of the caregivers and educators could also influence the outcome of information recorded on the questionnaires. Guidance was given before and during the data collection period in an effort to minimize this effect. The primary caregivers completed the sensory profile in the company of the researcher who provided individual guidance in an effort to ensure correct interpretation of questions and to help with their understanding. The researcher and assisting occupational therapists used similar explanations when preparing the educators for completion of the SPSC.

3.10 ETHICAL ASPECTS

This study was guided by ethical rules of conduct prescribed by the **Health Professions Council of South Africa** (HPCSA). The final responsibility for ethical conduct lay with the researcher who at all times strived for the highest possible ethical conduct.

The researcher of this study:

- only commenced with the research once the approval of the **Health Sciences Research Ethics Committee (HSREC)** of the University of the Free State (Tel 051-4017794) had been obtained (Appendix A),
- obtained permission for the study from relevant authorities – WCED (Appendix C), Director of CWED (Appendix D) and the principals of schools involved.

The following core ethical values and standards applied:

- **Informed consent:** The researcher provided the caregivers with sufficient information about the nature and purpose of the study in a language that they could understand. This included the goal of the study, information about sensory integration, a brief description of the tests that would be done, benefits of participating and the credibility of researcher. Parents had the opportunity to ask questions. They were told that participation was voluntary at all times. The parents had to give consent for their children's participation in the study, but the children also had the right to refuse or withdraw at any stage (Appendix E). The children themselves gave assent (agreed to take part) and a child's refusal to participate would have been respected (Council for International Organizations of Medical Sciences, 2012, p. 43) (See Appendix F).
- **The principle of best interest or well-being:** The potential benefits of this study for the participants outweighed any potential risks. The chance of hurting or traumatising the children during the evaluation process was minimal and was a small risk to take in comparison to the benefits of the results and the expected future programme planning. It is foreseen that the results of this study will contribute to effective intervention planning by those occupational therapists who assist children with FASD in the classroom and at home to improve their adaptive behaviour and school performance within their cognitive capabilities.
- **The principle of respect for persons:** The primary caregivers had the right to decide whether or not they wanted their child to participate in the study. They would have been respected had they decided to withdraw their child from the study. The child also had the right to withdraw and would not have been discriminated against

afterwards (principle of autonomy). In fact, no child chose to withdraw. The perception was that the children enjoyed participating in the assessment process.

The parents' right to keep their children's FASD status confidential was respected. One principal wanted the researcher to disclose the children's FASD status, but he was referred to FASER, who had made the diagnoses. Only children whose parents/caregivers had given permission during the initial research studies in which they took part were considered for this study. The researcher ensured that all data collected were kept in a locked filing cabinet with no access to unauthorized persons (principle of confidentiality). In this study, the FASD diagnoses were known only to the researcher, who kept the information confidential.

- **The principle of justice:** It is believed that this study will eventually leave the participants and community better off. It was not done for any purpose other than to fulfil the aim of the research and to obtain a postgraduate qualification. Nor was it done either to exploit the children or their parents. Once the planned programme to help educators and caregivers manage and stimulate the children with FASD is put into practice, it is hoped that the children from this area will benefit from the outcomes of the study in the long run. No one was unjustly excluded from it because of race, gender or religion. Since the children with FASD were from a vulnerable community, special care was taken to consider them, to respect their decisions, and to avoid taking advantage of them or exploiting them. Their right to withdraw from the research at any time, for example, was always respected.
- **The right to privacy:** The researcher made sure that the participants' right to privacy was maintained during the study and that the procedure was no more intrusive than it needed to be (Polit & Beck, 2010, p. 125). To ensure this, a procedure of confidentiality was followed. Numbers instead of names were used to identify the participants. Teachers and assisting occupational therapist did not know the difference between the control group and FASD subjects, and parents were informed of what would be done with the test results after completion of the research project.
- **Minimal risk:** Evaluating the children did not pose greater risks to them than those they encountered in daily life (Polit & Beck, 2010, p. 125). They were evaluated at

school, and the test items of the SIPT resembled ordinary gross or fine motor activities.

- **Harm to participants:** The researcher protected the subjects against any form of physical discomfort, within reasonable limits. The only anticipated physical discomfort was the effect of the PRN test. Depending on the child's vestibular processing, he or she may have experienced discomfort in the form of dizziness and nausea caused by the fast rotatory movements. Both therapists administering the tests were trained in the use of inhibitory techniques to counteract discomfort. This test item was also terminated in cases where a child showed discomfort or when the therapist observed signs of disorganisation. Where this happened, the test was immediately terminated and inhibitory techniques were applied. Nor were the children forced to get onto the post-rotary board.
- **Deception of participants:** No information about the study was withheld from the participants, nor were they given any false information (Polit & Beck, 2010, p. 123).
- **Competence of researcher:** The researcher was appropriately qualified to carry out the research. She had sufficient experience and training in sensory integration theory and testing, FASD and in dealing with parents. She is a qualified occupational therapist, registered with the Health Professionals Council of South Africa, and successfully completed the South African Institute for Sensory Integration's training in the administration and interpretation of the Sensory Integration and Praxis Tests. She also has vast experience in the use of sensory profiles.
- **Co-worker:** The assisting occupational therapist was well briefed beforehand. This ensured that she knew what was expected of her. She was kept well informed of the progress throughout the study. She was a qualified occupational therapist, registered with the Health Professionals Council of South Africa and had completed the South African Institute for Sensory Integration's training in the administration and interpretation of the Sensory Integration and Praxis Tests.
- **Conflict of interest** was avoided, for example in the case of sponsors who may want commercial gain from taking part in the study or who may try to dictate to the researcher what to do. At no time did the researcher engage in any activity which

might have caused conflict of interest. Although she received financial support of R5000 from SAISI, this did not cause any conflict of interest.

- **Debriefing sessions:** The caregivers were given the researcher's contact number in order to report any stress caused by the interviews or assessments. Time was allowed during completion of the SP to give the parents the chance to discuss their worries or fears about their children's behaviour or progress at school.
- Children with suspected health problems were referred to the nearest clinic for further investigation.
- The researcher strove to write an accurate, objective, clear and unambiguous **research report** at the conclusion of the study. Plagiarism was avoided by recognising all sources consulted and the people who collaborated and contributed to the research. Shortcomings and errors are discussed in the final research report. Involved parents as well as the Department of Education will be given an information brochure, recording the findings of this research in an objective way, while protecting the confidentiality of the participants.

3.11 SUMMARY

Chapter 3 provided a detailed discussion of the research methodology used during the study to determine the sensory integration difficulties and dysfunctions of children with FASD. A quantitative research approach and observational analytical design were used. The population from which the sample was selected consisted of children, both with and without FASD, who had taken part in two studies conducted by FASER-SA in the CWED (2.2.3). The matched study sample of 30 children with FASD and 30 not on the fetal alcohol spectrum was selected from these children according to inclusion (3.4.1) and exclusion (3.4.2) criteria specifically designed for this study. Measurement was done using two standardised questionnaires (SP and SPSC), as well as one standardised test (SIPT). Ethical considerations relating to informed consent, principles of best interest and respect, the right to privacy, and minimal risk were discussed.

Chapter 4 presents demographic and diagnostic information about the study participants and their parents/caregivers, together with the results of the Sensory Profiles and the SIPT.

CHAPTER 4

RESULTS

4.1 INTRODUCTION

The previous chapter provided a discussion of the research methodology used in this study. Chapter 4 presents the results of the Sensory Profile (SP and SPSC) questionnaires and the Sensory Integration and Praxis Tests (SIPT). Given the descriptive nature of the study, the results will be presented in tables, followed by discussions of focus areas in the results. The aim of this chapter is to present the results obtained as they relate to the sensory integration difficulties and dysfunctions of children on the fetal alcohol spectrum in the Cape Winelands Education District (CWED).

The results were obtained from two questionnaires, the Sensory Profile Caregiver Questionnaire and the Sensory Profile School Companion, and from the SIPT, a standardized test. A clinical diagnosis of sensory integration dysfunctions was made on the basis of categories (Sensory Profiles) or patterns of dysfunction (SIPT), identified and confirmed through research. For this reason, the researcher considered it to be of practical importance to review how the two groups performed when compared to these categories/dysfunctions. Hence the reflection of these results in the tables presented, giving a clear picture of the outcomes of testing for all the children included in the sample. While the study investigated the sensory integration difficulties and dysfunctions of children with FASD, the results of the matched sample could not be ignored. It was therefore deemed necessary to present and discuss any significant problems which presented in the control group.

The study design used children matched on age and gender for the exposed and control groups. It is important to remember that the group comparisons by cross-tabulation would be statistically inappropriate unless the pairing were taken into consideration. Paired t-tests were therefore used to investigate the effect of fetal alcohol spectrum symptoms on sensory integration, as they were statistically more appropriate for the study design.

The research results are presented in the following sequence:

(4.2) demographic information, (4.3) diagnostic information, (4.4) sensory profiles (caregiver questionnaire and sensory profile school companion), (4.5) SIPT, (4.6) patterns of sensory integration dysfunction, and a summary (4.7).

The following section gives the demographic information of the study participants.

4.2 DEMOGRAPHIC INFORMATION

Table 4.1 presents demographic information on the 60 study participants as provided by the primary caregiver, as well as FASER-SA's Maternal Questionnaire (with parental consent) in some instances. The information includes gender, age, ethnic grouping and grade. Where the primary caregiver was available, information was obtained about the family structure.

Table 4.1 Study Participant Demographic Information

Variable		FASD group (n=30)	Control group (n=30)
Gender	Male	14 (46.7%)	14 (46.7%)
	Female	16 (53.3%)	16 (53.3%)
Age	Range	5 yrs 6 mths – 8 yrs 11 mths	5 yrs 6 mths – 8 yrs 10 mths
	Mean	7 years 5 months	7 years 4 months
Ethnic grouping	Coloured	30 (100.0%)	30 (100.0%)
Grade	Grade R	8 (26.7%)	8 (26.7%)
	Grade 1	7 (23.3%)	7 (23.3%)
	Grade 2	6 (20.0%)	1 (3.3%)
	Grade 3	9 (30.0%)	14 (46.7%)

Variable		FASD group (n=23)	Control group (n=20)
Family structure	Family with two parents (both biological or one biological with new partner/boyfriend)	14 (60.9%)	9 (45.0%)
	Single parent	7 (30.4%) (4 live with parents)	7 (35.0%) (3 live with parents)
	Foster	2 (8.7%)	4 (20.0%)

The representation of gender in Table 4.1 indicates that there were 14 males (46.7%) and 16 females (53.3%) in both the FASD and control groups.

Ages ranged from 5 years, 6 months to 8 years, 11 months for the FASD group, and 8 years, 10 months for the control group. The mean age was 7 years, 5 months for the FASD group, and 7 years, 4 months for the control group.

The children in both groups were represented from Grades R to 3. Eight children (26.7%) of each group were in Grade R, while seven of each (23.3%) were in Grade 1. Six children (20.0%) of the FASD group were in Grade 2, while only one (3.3%) of the control group was in Grade 2, and 9 (30.0%) and 14 (46.7%) of children of the FASD and control groups respectively were represented in Grade 3. It should be remembered that children from the FASD and control groups were paired in terms of gender and age, not gender and grade. Thus, although the age and gender distributions were always similar, this would not necessarily be case with the grade distribution.

In terms of ethnicity, all the children from both the FASD and control groups were from the Coloured population. Both groups came from a variety of family structures. Seven of the cases and 10 of the controls were not reported on. The largest percentage of each group came from families with two parents (14/60.8% of the cases and 9/45.0% of the controls). Some of these families consisted of two biological parents, while others consisted of one biological parent and a new partner or boyfriend/girlfriend. Seven of the single mothers from the combined groups lived with their parents (4 cases and 3 controls), and the other seven lived on their own with their children. As reported, six children in total were in foster care, of whom 2 (8.7%) were from the cases and 4 (20.0%) from the controls.

Tables 4.2 and 4.3 report on maternal educational level and employment status.

Table 4.2 Maternal Educational Level

Variable		FASD group (n=20)	Control group (n=20)
Maternal education – highest grade	Gr 4	2 (10.0%)	0 (0.0%)
	Gr 5	1 (5.0%)	0 (0.0%)
	Gr 6	0 (0.0%)	1 (5.0%)
	Gr 7	6 (30.0%)	4 (20.0%)
	Gr 8	4 (20.0%)	4 (20.0%)
	Gr 9	3 (15.0%)	2 (10.0%)
	Gr 10	2 (10.0%)	5 (25.0%)
	Gr 11	1 (5.0%)	2 (10.0%)
	Gr 12	1 (5.0%)	2 (10.0%)

Table 4.2 records that the highest grades achieved in maternal education varied from Grade 4 to Grade 12. Nine of the cases and five of the controls had not progressed beyond primary school, while only three (one case and two controls) finished secondary school Grade 12.

Table 4.3 Maternal Employment Status

Variable		FASD group (n=24)	Control group (n=22)
Employment status	Work full-time	7 (29.2%)	12 (54.6%)
	Work part-time	2 (8.3%)	2 (9.1%)
	Seasonal work	1 (4.2%)	6 (27.3%)
	Unemployed	14 (58.3%)	2 (9.1%)

Of the 46 parents who reported their employment status, only seven of the case group and 12 of the control group worked full-time. Three of the parents from the case group and eight from the control group were employed part-time or were doing seasonal work. Two (9.1%) of the control group and 14 (58.3%) of the case group parents were unemployed.

4.3 DIAGNOSTIC INFORMATION

Table 4.4 shows a summary of the diagnoses of the children studied in this research. Diagnoses were made by the interdisciplinary team of FASER using the Diagnostic guidelines for FASD, according to the IOM as clarified by Hoyme et al. (2005). A three-tier method, as described in 2.2.2, was used. Thirty-three of the children were from one town, with its rural surroundings, and 27 were from four other small towns, also with their rural surroundings. The towns are all in the CWED, Western Cape, South Africa. The latter area is more rural, remote and of lower socioeconomic status than the first town (May, et al., 2016, p. 208).

Table 4.4 Diagnoses of Study Sample

Diagnosis		
Diagnosis	**Frequency**	**Percent**
FAS	17	28.3
PFAS	7	11.7
ARND	6	10.0
Not FASD	30	50.0

From Table 4.4, it is clear that the largest percentage of the FASD children (56.7%, representing 28.3% of the total sample) who took part in this project had been diagnosed with FAS. Twelve (70.6%) of these seventeen children were from the most remote rural area. Seven (11.7% of the total sample) were diagnosed with PFAS, and six (10.0% of the total sample) with ARND. None of the children in the more remote rural area were diagnosed with PFAS. They were also the younger children in the study sample, as indicated on Table 4.1. The 30 children in the control group were selected on the basis of their having no diagnosis of FASD.

4.4 SENSORY PROFILES

4.4.1 Sensory Profile Caregiver Questionnaire

Scores were not available for 20 (33.3%) of the children in the group (10 FASD and 10 controls). Their parents or caregivers were not available for the interviews, mainly because of transport and work responsibilities. None of the rural parents had transport to get to the schools, and especially those parents doing seasonal work were unable to attend the interviews.

4.4.1.1 Sensory Profile Caregiver Questionnaire Classification Categories

The results of each classification category (Typical Performance, Probable Difference, Definite Difference) of the Sensory Profile Caregiver Questionnaire and each of the fourteen sections of the two groups (cases and controls) are shown in Table 4. Typical performance recorded scores at or above the point of one standard deviation below the mean for children without disabilities, probable difference at or above the point of two standard deviations below the mean, but lower than one standard deviation below the mean and a definite difference from scores below the point of two standard deviations below the mean (Dunn, 1999, p. 19). Fisher's exact p-values are also presented in Table 4.5. This test is used to examine the significance of association between two categorical variables (Maree, 2007, p. 247) in the case of smaller samples.

Table 4.5 Sensory Profile Caregiver Questionnaire Classification Categories

	Typical performance		Probable Difference		Definite Difference		
Scale	**Cases (n=20)**	**Controls (n=20)**	**Cases (n=20)**	**Controls (n=20)**	**Cases (n=20)**	**Controls (n=20)**	**Fisher's exact P**
FACTOR							
Sensory Seeking	2 (10.0%)	10 (50.0%)	7 (35.0%)	4 (20.0%)	11 (55.0%)	6 (30.0%)	0.024*
Emotionally Reactive	13 (65.0%)	14 (70.0%)	3 (15.0%)	3 (15.0%)	4 (20.0%)	3 (15.0%)	1.000
Low Endurance / Tone	12 (60.0%)	15 (75.0%)	1 (5.0%)	2 (10.0%)	7 (35.0%)	3.(15.0%)	0.450
Oral Sensory Sensitivity	8 (42.1%)	16 (80.0%)	8 (42.1%)	3 (15.0%)	3 (15.8%)	1 (5.0%)	0.060
Inattention/Distractibility	3 (15.0%)	13 (65.0%)	3 (15.0%)	1 (5.0%)	14 (70.0%)	6 (30.0%)	0.004*
Poor Registration	17 (85.0%)	18 (90.0%)	1 (5.0%)	2 (10.0%)	2 (10.0%)	0 (0.0%)	0.600
Sensory Sensitivity	16 (80.0%)	17 (85.0%)	1 (5.0%)	2 (10.0%)	3 (15.0%)	1 (5.0%)	0.690
Sedentary	17 (85.0%)	14 (70.0%)	2 (10.0%)	1 (5.0%)	1 (5.0%)	5 (25.0%)	0.290
Fine Motor/Perceptual	9 (45.0%)	16(84.2%)	1 (5.0%)	3 (15.8%)	10 (50.0%)	0 (0.0%)	0.0005*

*Statistically significant difference: $P<0.05$

	Typical performance		Probable Difference		Definite Difference		
Scale	**Cases (n=20)**	**Controls (n=20)**	**Cases (n=20)**	**Controls (n=20)**	**Cases (n=20)**	**Controls (n=20)**	**Fisher's exact P**
SECTION							
SENSORY PROCESSING							
Auditory Processing	6 (30.0%)	14 (70.0%)	4 (20.0%)	3 (15.0%)	10 (50.0%)	3 (15.0%)	0.030*
Visual Processing	8 (40.0%)	17 (85.0%)	5 (25.0%)	2 (10.0%)	7 (35.0%)	1 (5.0%)	0.010*
Vestibular Processing	3 (15.0%)	7 (35.0%)	3 (15.0%)	7 (35.0%)	14 (70.0%)	6 (30.0%)	0.040*
Touch Processing	11 (55.0%)	18 (90.0%)	2 (10.0%)	1 (5.0%)	7 (35.0%)	1 (5.0%)	0.020*
Multisensory Processing	3 (15.0%)	15 (75.0%)	12 (60.0%)	14 (20.0%)	5 (25.0%)	1 (5.0%)	0.0005*
Oral Sensory Processing	7 (35.0%)	14 (70.0%)	6 (30.0%)	4 (20.0%)	6 (30.0%)	2 (10.0%)	0.110

Statistically significant difference: P<0.05

	Typical performance		Probable Difference		Definite Difference		
Scale	**Cases (n=20)**	**Controls (n=20)**	**Cases (n=20)**	**Controls (n=20)**	**Cases (n=20)**	**Controls (n=20)**	**Fisher's exact P**
MODULATION							
Sensory Processing Related to Endurance/Tone	12 (60.0%)	15 (75.0%)	1 (5.0%)	2 (10.0%)	7 (35.0%)	3 (15.0%)	0.440
Modulation Related to Body Position and Movement	5 (25.0%)	8 (40.0%)	4 (20.0%)	5 (25.0%)	11 (55.0%)	7 (35.0%)	0.440
Modulation of Movement Affecting Activity Level	11 (55.0%)	13 (65.0%)	7 (35.0%)	6 (30.0%)	2 (10.0%)	1 (5.0%)	0.790
Modulation of Sensory Input Affecting Emotional Responses	9 (45.0%)	16 (80.0%)	7 (35.0%)	1 (5.0%)	4 (20.0%)	3 (15.0%)	0.030*
Modul of Visual Input Affecting Emotional Responses/Activity Level	9 (45.0%)	10 (50.0%)	9 (45.0%)	9 (45.0%)	2 (10.0%)	1 (5.0%)	1
BEHAVIORAL & EMOTIONAL RESPONSES							
Emotional/Social Responses	12 (60.0%)	15 (75.0%)	4 (20.0%)	3 (15.0%)	4 (20.0%)	2 (10.0%)	0.640
Behaviour Outcomes of Sensory Processing	9 (45.0%)	14 (70.0%)	4 (20.0%)	3 (15.0%)	7 (35.0%)	2 (10.0%)	0.190
Items Indicating Thresholds for Response	11 (55.0%)	17 (85.0%)	5 (25.0%)	2 (10.0%)	4 (20.0%)	1 (5.0%)	0.170

Statistically significant difference: $P<0.05$

The results summarized in Table 4.5 indicate that the percentages of the cases falling in the Typical Performance range of the SP Caregiver Questionnaire were less than the percentages for the controls in 22 of the 23 categories, e.g. only 10.0% (2) of cases fell in the Typical Performance range, compared to 50.0% (10) of the controls in the category Sensory Seeking. The only category with a higher percentage of cases (85.0%) than controls (70.0%) was Sedentary. This meant that in the Definite Difference range, the cases had a higher representation in all but one category; for example, for the category Visual Processing, 35.0% (7) of the cases were represented, as against only 5.0% (1) of the controls (p=0.010). Representation of cases and controls in the Probable Difference range varied, with higher percentages of cases than controls in 13 categories, similar representation in two categories, and lower percentages of cases than controls in eight of the 23 categories.

Significant differences on Fisher's exact test were found between the two groups on the Sensory Profile in nine of the 23 categories (Table 4.5).

The summary of the results from the Sensory Profile Caregiver Questionnaires therefore indicate that the cases scored worse than the controls in 22 of the 23 categories. In nine of these categories the scores were significantly poorer, while in the only category where the cases performed better than the controls (Sedentary) the difference was not statistically significant.

4.4.1.2 Sensory Profile Caregiver Questionnaire Mean Scores

Descriptive data for the Sensory Profile factor and section raw scores are shown in Table 4.6. A paired sample t-test was used to calculate whether the means of the pairs in the two groups differed significantly from each other (Trochim, 2006; Lani, 2016).

A high drop-out rate of caregivers completing the Sensory Profile (n = 15 (pairs)) as reflected in Table 4.6 was experienced (cf. 3.9.1. for discussion thererof). Even though 20 Sensory Profiles each for the cases and controls were available, only 15 of the cases could be paired with their matching controls, considering the inclusion criteria. Since it was computed by the biostatistician that between 4 and 13 pairs would be sufficient for data analysis, as discussed in 3.4.3, the conclusions made from this smaller sample are thus considered valid.

Table 4.6 Sensory Profile Caregivers Questionnaire Mean Scores

		MEANS							
Scale	**N (pairs)**	**Controls**	**SD**	**Cases**	**SD**	**Difference: Controls - cases**	**95% Confidence level**	**t-value**	**P-value**
FACTOR									
Sensory Seeking	14	60.8	11.9	49.9	12.0	8.8	[-0.9; 18.4]	1.97	0.07
Emotional Reactive	14	61.2	9.8	58.1	10.5	2.4	[-4.9; 9.6]	0.70	0.50
Low Endurance/Tone	14	40.3	5.1	38.0	6.9	4.4	[-0.6; 9.3]	1.89	0.08
Oral Sensitivity	14	36.8	5.9	31.7	6.1	5.1	[0.01; 10.3]	2.17	0.05*
Inattention/Distractibility	14	24.1	5.7	18.4	6.2	6.9	[1.9; 11.8]	3.00	0.01*
Poor Registration	14	36.9	3.1	36.3	3.9	0.9	[-1.4; 3.3]	0.85	0.41
Sensory Sensitivity	14	18.2	2.9	17.8	4.1	0.1	[-1.9; 2.2]	0.15	0.88
Sedentary	14	13.6	4.6	14.7	2.9	-1.3	[-4.7; 2.1]	-0.82	0.42
Fine Motor/Perceptual	13	12.7	2.2	8.9	3.9	3.5	[0.4; 6.7]	2.47	0.03*

*Statistically significant difference: $P<0.05$

		MEANS							
Scale	**N (pairs)**	**Controls**	**SD**	**Cases**	**SD**	**Difference: Controls - cases**	**95% Confidence level**	**t-value**	**P-value**
SECTION									
SENSORY PROCESSING									
Auditory Processing	14	30.8	4.8	26.1	6.3	5.9	[1.4; 10.3]	2.82	0.01*
Visual Processing	14	35.0	4.4	28.9	6. 5	4.9	[0.2; 9.5]	2.25	0.04*
Vestibular Processing	14	45.1	5.4	41.5	6.5	4.6	[1.2; 7.9]	2.97	0.01*
Touch Processing	14	77.7	7.3	70.9	10.6	8.7	[2.2; 15.2]	2.89	0.01*
Multisensory Processing	14	28.0	4.4	24.7	3. 8	3.1	[-0.8; 6.9]	1.72	0.11
Oral Sensory Processing	14	49.3	7.2	41.1	11.2	6.8	[0.8; 12.7]	2.46	0.03*

*Statistically significant difference: P<0.05

		MEANS							
Scale	**N (pairs)**	**Controls**	**SD**	**Cases**	**SD**	**Difference: Controls - cases**	**95% Confidence level**	**t-value**	**P-value**
MODULATION									
Sensory Processing Related to Endurance/Tone	14	40.3	5.1	38	6.9	4.4	[-0.62; 9.3]	1.89	0.08
Modulation Related to Body Position & Movement	14	39.1	6.0	36.1	5.4	1.8	[-2.0; 5.5]	1.03	0.32
Modulation of Movement Affecting Activity Level	14	23.9	4.2	22.4	2.4	1.3	[-1.0; 3.6]	1.22	0.24
Modulation of Sensory Input Affecting Emotional Responses	14	16.6	3.2	15.6	3.8	0.8	[-2.2; 3.8]	0.56	0.58
Modulation of Visual Input Affect. Emotional Resp. & Activity Level	14	14.0	2.1	14.3	1.8	0.4	[-1.2; 2.0]	0.48	0.64
BEHAVIORAL & EMOTIONAL RESPONSES									
Emotional/Social Responses	14	67.0	8.3	65.5	9.8	0.8	[-5.6; 7.2]	0.27	0.79
Behavioural Outcomes of Sensory Processing	14	22.9	5.7	20.2	5.2	3.0	[-1.3; 7.3]	1.50	0.16
Items Indicating Thresholds for Response	14	13.3	1.9	11.4	1.9	1.5	[-0.3; 3.3]	1.84	0.09

*Statistically significant difference: P<0.05

The mean raw scores for each of the Sensory Profile factor and section scores were lower for the cases than for the controls, with the exception of the Sedentary scores. The p-values indicated significant differences between the cases and controls for Oral Sensitivity (p=0.05), Inattention/Distractibility (p=0.01), Fine Motor/Perceptual (p=0.03), Auditory (p=0.01), Visual (p=0.04), Vestibular (p=0.01) and Touch Processing (p=0.01), as well as Oral Sensory Processing (p=0.03).

4.4.1.3 Sensory Processing – Four Quadrants of Responsivity

Table 4.7. depicts the four basic quadrants of responsivity in Dunn's (1999, p. 32) theoretical model of sensory processing. These are Low Registration, Sensation Seeking, Sensory Sensitive and Sensation Avoiding. The performance of the cases and controls are indicated in the ranges Typical Performance, Probable Difference More than Others, Probable Difference Less than Others, and Definite Difference. Fisher's exact test was used to examine the significance of association between the cases and controls.

Table 4.7 Sensory Processing – Four Quadrants of Responsivity Classification Categories

	Typical Performance		Probable Difference, More than others		Probable Difference, Less than Others		Definite Difference, More than Others		
	Cases (n=20)	**Controls (n=20)**	**Cases (n=20)**	**Controls (n=20)**	**Cases (n=20)**	**Controls (n=20)**	**Cases (n=20)**	**Controls (n=20)**	**Fisher's exact P**
Low Registration	6 (30.0%)	12 (60.0%)	5 (25.0%)	3 (15.0%)	1 (5.0%)	1 (5.0%)	8 (40.0%)	4 (20.0%)	0.25
Sensation Seeking	2 (10.0%)	7 (35.0%)	4 (20.0%)	6 (30.0%)			14 (70.0%)	7 (35.0%)	0.06
Sensory Sensitive	2 (10.0%)	13 (65.0%)	2 (10.0%)	5 (25.0%)			16 (80.0%)	2 (10.0%)	<.0001*
Sensation Avoiding	11 (55.0%)	14 (70.0%)	5 (25.0%)	5 (25.0%)	0 (0.0%)	1 (5.0%)	4 (20.0%)	0 (0.0%)	0.17

*Statistically significant difference: P<0.05

The results in Table 4.7 show that higher percentages of cases fell in the Definite Difference range in all four quadrants of responsivity, although only one was statistically significant. In the quadrant for Sensory Sensitivity, 80.0% (16) of the cases fell in the Definite Difference range, compared to 10.0% (2) of the controls (Fisher's exact P = <.0001). Percentages of cases and controls in the Probable Difference More than Others range varied, with more cases in Low Registration, less in Sensation Seeking and Sensory Sensitive, and an equal number in Sensation Avoiding. Probable Difference Less than Other had a small representation of one (5.0%) each for Low Registration, and less cases (0.0%) than controls (5.0%) for Sensation Avoiding.

4.4.2 Sensory Profile School Companion

Sensory Profile School Companion Teacher Questionnaires were completed by the educators for 59 of the children (29 cases and 30 controls). One of the score sheets was not included in the statistical analysis because the educator had allocated the same score (5) for all 62 questions on the Teacher Questionnaire.

4.4.2.1 Sensory Profile School Companion Classification Categories

The results of each classification category (Typical Performance, Probable Difference, Probable Difference More than Others, Probable Difference Less that Others, Definite Difference) of the Sensory Profile School Companion for the two groups (cases and controls), are shown in Table 4.8. All 13 categories are illustrated. Fisher's exact p-values are also presented for each category.

Table 4.8 Sensory Profile School Companion Classification Categories

	Typical Performance		Probable Difference, More than Others		Probable Difference, Less than Others		Definite Difference		
	Cases (n=29)	Controls (n=30)	Cases (n=29)	Controls (n=30)	Cases (n=29)	Controls (n=30)	Cases (n=29)	Controls (n=30)	Fisher's exact P
Registration	10 (34.5%)	21 (70.0%)	9 (31.0%)	2 (6.7%)	1 (3.5%)	2 (6.7%)	9 (31.0%)	5 (16.7%)	0.01*
Seeking	17 (58.6%)	23 (76.7%)	5 (17.2%)	2 (6.7%)			7 (24.1%)	5 (16.7%)	0.29
Sensitivity	14 (48.3%)	20 (66.7%)	6 (20.7%)	5 (16.7%)			9 (31.0%)	5 (16.7%)	0.32
Avoiding	11 (37.9%)	20 (66.7%)	7 (24.1%)	6 (20.0%)			11 (37.9%)	4 (13.3%)	0.04*
School Factor 1	12 (41.4%)	22 (73.3%)	8 (27.6%)	3 (10.0%)			9 (31.0%)	5 (16.7%)	0.05*
School Factor 2	19 (65.5%)	21 (70.0%)	4 (13.8%)	7 (23.3%)			6 (20.7%)	2 (6.7%)	0.26
School Factor 3	11 (37.9%)	19 (63.3%)	6 (20.7%)	5 (16.7%)			12 (41.4%)	6 (20.0%)	0.12
School Factor 4	13 (44.8%)	21 (70.0%)	7 (24.1%)	5 (16.7%)			9 (31.0%)	4 (13.3%)	0.14
Auditory	11 (37.9%)	20 (66.7%)	7 (24.1%)	4 (13.3%)	1 (3.5%)	2(6.7%)	10 (34.8%)	4 (13.3%)	0.08
Visual	13 (44.8%)	19 (63.3%)	7 (24.1%)	8 (26.7%)			9 (31.0%)	3 (10.0%)	0.13
Movement	15 (51.7%)	22 (73.3%)	6 (20.7%)	3 (10.0%)			8 (27.6%)	5 (16.7%)	0.25
Touch	11 (37.9%)	18 (60.0%)	6 (20.7%)	5 (16.7%)			12 (41.4%)	7 (23.3%)	0.23
Behaviour	13 (44.8%)	19 (63.3%)	7 (24.1%)	3 (10.0%)	0 (0.0%)	2 (6.7%)	9 (31.0%)	6 (20.0%)	0.15

*Statistically significant difference: $P<0.05$

A higher percentage of cases fell within the Definite Difference range of all 13 categories of the Sensory Profile School Companion, although only three were statistically significant, with 37.9% (11) cases against 13.3% (4) controls in the category Avoiding. Fewer cases than controls fell in the Typical Performance range. An example of this tendency is observed in the category Registration, where 34.5% (10) cases fell in the Typical Performance range, compared to a much higher 70.0% (21) of the controls. This tendency was also true for all the other categories on the profile.

Higher percentages of cases fell in the Probable Difference range, more than in the Others range in all the categories, except for School Factor 2 (representing the learner's awareness and attention in the learning environment), where 13.8% of the cases and 23.3% of the controls were recorded. Only three categories were represented in the Probable Difference Less than Others range, namely Registration, Auditory and Behaviour. In all three of these, fewer cases than controls were recorded, with 3.5% (1) case and 6.7% (2) controls in the category Registration. Significant differences between the cases and controls were found for Registration (p=0.01), Avoiding (p=0.04) and School Factor 1 (p=0.05), indicating poorer performance of the case group children in the Typical Performance range of these subsections.

4.4.2.2 Sensory Profile School Companion mean scores

Means and standard deviations for the Sensory Profile School Companion section raw scores are presented in Table 4.9. Paired sample t-tests were used to calculate whether the means of the pairs in the two groups differed significantly (Trochim, 2006; Lani, 2016).

Table 4.9 Sensory Profile School Companion mean scores

		MEANS							
Scale	N (pairs)	Controls	SD	Cases	SD	Difference: Controls - Cases	95% Confidence Level	t-value	P-value
Registration	29	67.9	15.5	58.6	16.2	8.7	[-0.2; 17.6]	2.00	0.06
Seeking	29	47.1	10.2	42.0	13.3	4.9	[-1.5; 11.2]	1.58	0.13
Sensitivity	29	62.3	10.5	58.0	13.8	4.2	[-2.0; 10.3]	1.39	0.17
Avoiding	29	72.3	10.1	65.7	12.1	6.3	[0.4; 12.2]	2.20	0.04*
School Factor 1	29	85.0	18.9	73.8	21.5	10.5	[-0.6; 21.6]	1.94	0.06
School Factor 2	29	48.9	8.9	46. 5	11.9	2.5	[-2.9; 7.9]	0.34	0.35
School Factor 3	29	71.3	11.7	64.0	13.9	6.9	[0.5; 13.3]	2.19	0.04*
School Factor 4	29	44.3	7.9	40.0	8.8	4.1	[-0.5; 8.8]	1.84	0.07
Auditory	29	39.5	8.8	34.5	9.4	4.8	[0.002; 9.6]	2.05	0.05*
Visual	29	40.8	8.5	35.9	11.4	4.76	[-0.9; 10.4]	1.72	0.09
Movement	29	59.0	10.5	53.1	14.3	5.7	[-0.7; 12.0]	1.83	0.08
Touch	29	50.0	8.6	45.9	10.6	3.9	[-1.2; 9.0]	1.56	0.13
Behaviour	29	60.1	10.4	54.9	12.3	4.9	[-1.2; 11.0]	1.64	0.11

*Statistically significant difference: $P<0.05$

The mean raw scores for each of the Sensory Profile School Companion section scores were lower for the cases than for the controls. The p-values indicated significant differences between the cases and controls for Avoiding, School Factor 3, representing the learner's range of tolerance for sensory input (Dunn, 2006, p. 43), and Auditory.

4.5 SENSORY INTEGRATION AND PRAXIS TESTS

All 60 children were tested on the SIPT. This test, designed to assess different practic abilities and various aspects of sensory processing, was discussed in 3.7.1.

4.5.1 SIPT Clinical observations results by group

Table 4.10 summarises the performance of the children on the three clinical observations which forms part of the pre-observations of the test. Scores are reported in the ranges Adequate, Slightly Deficient and Poor (South African Institute of Sensory Integration Research Committee, 2005).

Table 4.10 SIPT Clinical observations results by group

	Adequate		Poor		Slightly deficient		
	Cases (n=30)	Controls (n=30)	Cases (n=30)	Controls (n=30)	Cases (n=30)	Controls (n=30)	Fisher's Exact P
Prone extention	5 (16.7%)	8 (26.7%)	8 (26.7%)	13 (43.3%)	17 (56.7%)	9 (30.0%)	0.14
Supine flexion	12 (40.0%)	18 (60.0%)	5 (16.7%)	4 (13.3%)	13 (43.3%)	8 (26.7%)	0.31
Occular pursuits	8 (26.7%)	6 (20.0%)	8 (26.7%)	6 (20.0%)	14 (46.7%)	17 (56.7%)	0.65

* Statistically significant difference: P<0.05

The results on the clinical observations were mixed, with no significant differences between the cases and controls in any of the three items. With prone extension, five cases (16.7%), as compared with eight (26.7%) controls, tested in the Adequate range. More children of the control group (13 or 43.3%) tested in the Poor range, in comparison with eight (26.7%) of the cases. Lastly, 17 (56.7%) of the cases tested Slightly Deficient, as against nine (30.0%) of the control group, resulting in the majority of the children displaying slight deficient prone extension.

The tendency with supine flexion seemed to be different, with 12 (40.0%) of the cases and 18 (60.0%) of the controls scoring in the Adequate range. Five of the cases scored in the Poor and 13 Slightly Deficient, against the 13 (43.3%) and eight (26,7%) respectively of the controls. Poor crossing of the midline and insufficient postural adjustments in 33 (55.0%) of the children (cases and controls) were additional observations made during the execution of the Motor Accuracy test.

Eight (26.7%) of the cases had Adequate ocular pursuits, as against six (20.0%) of the controls. The exact same number of the cases and controls scored in the Poor range, while 14 (46.7%) of the cases and 17 (56.7%) of the controls presented with Slightly Deficient ocular pursuits.

4.5.2 SIPT Subtests mean scores

In Table 4.11 the paired t-test procedure was used to compare the differences between the two groups. The sample size, means, standard deviations and difference of the means between the controls and cases are presented. The 95% confidence intervals, as well as the p- and t-values for each subtest of the SIPT, are also illustrated in the table.

Since space in Table 4.11 is limited, the following abbreviations will be used for the subtests:

Space Visualization	SV	Constructional Praxis	CPr
Figure-Ground Perception	FG	Postural Praxis	PPr
Manual Form Perception	MFP	Oral Praxis	OPr
Kinesthesia	KIN	Sequencing Praxis	SPr
Finger Identification	FI	Bilateral Motor Coordination	BMC
Graphesthesia	GRA	Standing and Walking Balance	SWB
Localization of Tactile Stimuli	LTS	Motor Accuracy	MAc
Praxis on Verbal Command	PrVC	Postrotary Nystagmus	PRN
Design Copying	DC		

Table 4.11 SIPT Subtests mean major scores

		MEANS							
Scale	N (pairs)	Controls	SD	Cases	SD	Difference: Controls – Cases	95% Confidence Level	t-value	P-value
SV	30	-0.6	0.6	-1.0	0.7	0.4	[0.1; 0.8]	2.41	0.02*
FG	30	-0.9	0. 9	-1.4	0.9	0.5	[0.1; 0.9]	2.51	0.02*
MFP	30	-0.8	1. 3	-1.5	1.5	0.8	[0.1; 1.5]	2.30	0.03*
KIN	30	-0.5	1.0	-1.8	1.2	0.7	[0.2; 1.1]	2.76	0.009*
FI	30	-0.1	1.0	-0.9	0.9	0.8	[0.3; 1.3]	3.21	0.003*
GRA	30	0.0	0. 9	-0.5	0.9	0.5	[-0.01; 1.0]	2.00	0.05*
LTS	30	0.8	1.0	0.6	1.1	0.1	[-0.3; 0.6]	0.54	0.6
PrVC	30	-1.3	1.3	-2.0	1. 2	0.8	[0.2; 1.4]	2.73	0.01*
DC	30	-0.3	1.2	-1.7	1.1	1. 4	[0.8; 2.0]	4.86	<0.0001*
CPr	30	-0.1	0. 6	-0.7	0.8	0.6	[0.2; 0.9]	3.51	0.0015*
PPr	30	-0.0	0.9	-0.3	1. 1	0.3	[-0.2; 0.7]	1.23	0.23
OPr	30	-0.3	0.8	-0.4	0.9	0.1	[-0.3; 0.5]	0.67	0.51
SPr	30	-0.1	0.9	-1	0.9	0.9	[0.3; 1.4]	3.28	0.003*
BMC	30	0.5	1. 2	-0.4	1.0	0. 9	[0.2; 1.5]	2.77	0.01*
SWB	30	0.1	0.9	-0.2	1.2	0.3	[-0.2; 0.8]	1.30	0.2
MAc	30	0.2	0.9	-0.0	1.2	0.2	[-0.3; 0.8]	0.79	0.43
PRN	27	-0.2	0.8	-0.3	1.2	0.06	[-0.6; 0.7]	0.21	0.38

*Statistically significant difference: P<0.05

The data in Table 4.11 indicate a significant difference between the cases and the control group for 11 of the 17 SIPT subtests. Significant differences were found for Space Visualization (p=0.02), Figure-Ground Perception (p=0.02), Manual Form Perception (p=0.03), Kinesthesia (p=0.009), Finger Identification(p=o.003), Graphesthesia (p=0.05), Praxis on Verbal Command (p=0.01), Design Copying (p=<0.0001), Constructional Praxis (p=0.0015), Sequencing Praxis (p=0.003) and Bilateral Motor Coordination (p=0.01). In all 11 of these subtests the case group performed poorer than the control group.

4.5.3 SIPT Subtests by group

The performance of the two groups (cases and controls) in the 17 subtests of the SIPT is illustrated in Table 4.12. The SIPT Manual (Ayres, 2004, p. 112) states that SD scores <-1.0 may be indicative of clinical problems or dysfunction, while SD scores between -1.0 and +1.0 represent typical performance. Performance is thus shown in Table 4.9 in terms of typical range (-1.0 and +1.0 SD) and dysfunctional range (<-1.0 SD), and is recorded as percentages with the corresponding number of children for each group. Fisher's exact test was used to examine the significance of the association between the cases and controls. It is presented as Fisher's exact p-value.

Table 4.12 SIPT Subtest Classification Categories

	Typical range scored between -1.0 & +1.0 SD		Dysfunctional range scored < 1.0 SD		
	Cases (n=30)	Controls (n=30)	Cases (n=30)	Controls (n=30)	Fisher's exact P
Space Visualization	14 (46.7%)	19 (63.3%)	16 (53.3%)	11 (36.7%)	0.29
Figure-Ground	9 (30.0%)	17 (56.7%)	21 (70.0%)	13 (43.3%)	0.067
Manual Form Perception	13 (43.3%)	20 (66.7%)	17 (56.7%)	10 (33.3%)	0.12
Kinesthesia	17 (56.7%)	20 (66.7%)	13 (43.3%)	10 (33.3%)	0.59
Finger Identification	20 (66.7%)	27 (90.0%)	10 (33.3%)	3 (10.0%)	0.05*
Graphesthesia	24 (80.0%)	26 (86.7%)	6 (20.0%)	4 (13.3%)	0.73
Localization of Tactile Stimuli	29 (96.7%)	29 (96.7%)	1 (3.3%)	1 (3.3%)	1
Praxis on Verbal Command	8 (26.7%)	15 (50.0%)	22 (73.3%)	15 (50.0%)	0.11
Design Copying	8 (26.7%)	20 (66.7%)	22 (73.3%)	10 (33.3%)	0.004*
Constructional Praxis	19 (63.3%)	27 (90.0%)	11 (36.7%)	3 (10.0%)	0.03*
Postural Praxis	22 (73.3%)	25 (83.3%)	8 (26.7%)	5 (16.7%)	0.53
Oral Praxis	25 (83. 3%)	25 (83. 3%)	5 (16.7%)	5 (16.7%)	1
Sequencing Praxis	19 (63.3%	26 (86.7%)	11 (36.7%)	4 (13.3%)	0.07
Bilateral Motor Coordination	23 (76.7%)	25 (83. 3%)	7 (23.3%)	5 (16.7%)	0.74
Standing and Walking Balance	24 (80.0%)	26 (86.7%)	6 (20.0%)	4 (13.3%)	0.73
Motor Accuracy	24 (80.0%)	26 (86.7%)	6 (20.0%)	4 (13.3%)	0.73
Post Rotary Nystagmus	19 (63.3%)	26 (86.7%)	11 (36.7%)	4 (13. 3%)	0.07

*Statistically significant difference: $P<0.05$

Table 4.12 indicates that a higher percentage of the cases scored in the dysfunctional range (<-1.0) on 15 of the 17 subtests of the SIPT, and that two subtests had the same percentages of children from each group in the dysfunctional range: Localization of Tactile Stimuli (3.3%) and Oral Praxis (16.7%).

Significant differences were found between the cases and controls in three of the BPraxis (p=0.03), with the cases performing relatively poorly. More than 50% of the cases scored in the dysfunctional range (≤ -1.0) on five of the subtests (Space Visualization, Figure Ground, Manual Form Perception, Praxis on Verbal Command, Design Copying). Both groups performed best on Localization of Tactile Stimuli, where 96.7% (n=29) of both cases and controls fell within the normal range. Scores for the cases and controls were exactly the same for Oral Praxis, with17.7% in the dysfunctional range.

4.5.4 SIPT patterns of dysfunction

As described in 2.4.2.3, patterns of sensory integration dysfunction are groupings of poor test scores and observations clustering together. These patterns of dysfunctions were demonstrated in numerous factor and cluster analytic studies over the years, reflecting shared underlying commonalities of the test scores and behaviours observed, that load together on a cluster or factor (Schaaf & Mailloux, 2015, p. 17). The patterns of sensory integration dysfunction, as verified and clarified by Mailloux, et al. (2011) and confirmed as valid for South African children by Van Jaarsveld, Mailloux, Smith Roley and Raubenheimer (2014, p. 2), are given in Table 4.13. The identified three patterns of dysfunction, Visuo- and Somatodyspraxia, Bilateral Integration and Sequencing and Tactile Discrimination, are illustrated. The paired t-test procedure was used to assess whether the means of the paired cases and controls differed significantly (Trochim, 2006). P-values indicating the significance of these differences are presented.

Table 4.13 SIPT Patterns of Dysfunction Mean Scores

		MEANS							
Scale	**N (pairs)**	**Controls**	**SD**	**Cases**	**SD**	**Difference**	**95% Confidence level**	**t-value**	**P-value**
Visuo- & Somatodyspraxia[1]	30	-3.1	3.7	-7.3	4.2	4.16	[2.5; 5.8]	5.09	<.0001*
Bilateral Integration & Sequencing[2]	27	-0.4	4.2	-4.3	4.2	4.19	[2.1; 6.3]	4.13	0.0003*
Tactile Discrimination[3]	30	-0.2	1. 4	-1.6	1.9	1.43	[0.7; 2.2]	3.92	0.0005*

*Statistically significant difference: P<0.05

Poor performance on test items listed, is reflective of a specific pattern of SI dysfunction:

[1] Space Visualization
Manual Form Perception
Praxis on Verbal Command
Design Copying
Constructional Praxis
Postural Praxis

[2]Kinesthesia
Graphesthesia
Oral Praxis
Sequencing Praxis
Bilateral Motor Coordination
Standing and Walking Balance
Motor Accuracy
Postrotary Nystagmus

[3]Figure-Ground Perception
Finger Identification
Localization of Tactile Stimuli

The means of all three patterns of dysfunctions as illustrated in Table 4.13 were lower for the cases than for the controls; for example, the mean for the cases for Visuo- and Somatodyspraxia was -7.3, as compared to -3.1 for the controls. For all three dysfunctions, the p-values indicated a significant difference between the cases and the controls.

The results thus showed that the control children scored significantly better on the test items, revealing a specific pattern of dysfunction as compared to the paired children in the case group, meaning that significantly more of the case children could be identified with one of the three identified patterns of dysfunction.

4.6 CONCLUSIONS

Chapter 4 presented the results in terms of the three research objectives. Given the quantitative approach and the descriptive study design, the results were shown in tabular format together with brief discussions.

A higher percentage of cases than controls was recorded in 22 of the 23 categories of the Sensory Profile, with significant differences in nine of these. In the Sensory Profile School Companion, a higher percentage of cases was recorded in all 13 categories, with significant differences in three. Comparing the mean scores of the paired cases and controls, significant differences were found in eight categories of the SP and three of the SPSC.

The paired t-test procedure indicated significant differences between the pairs of cases and controls for 11 of the SIPT tests, a finding which will be discussed in more detail in the next chapter. Significantly more cases were also identified with one of the three identified SIPT patterns of dysfunction.

Chapter 5 will cover interpretation of the results, with a discussion of the conclusions as they compare with the relevant literature.

CHAPTER 5

DISCUSSION OF RESULTS

5.1 INTRODUCTION

Chapter 4 presented the results of the Sensory Profile Caregiver Questionnaire, the Sensory Profile School Companion, and the Sensory Integration and Praxis Tests. Demographic information of the case and control groups, followed by the composition of the diagnosis, were also given. In this chapter, the results will be discussed and compared with the relevant literature in order to make appropriate conclusions about the sensory integration difficulties and dysfunctions of children with FASD from the CWED. The clinical importance of these findings will also be covered in this chapter. In keeping with the structure of Chapter 4, the research objectives were used as a guide for the discussion of the results.

The results will be reviewed in the following sequence: (5.2) demographic information, (5.3) diagnostic information, (5.4) sensory profiles (Caregiver Questionnaire and School Companion Teacher Questionnaire), (5.5) SIPT, (5.6) patterns of sensory integration dysfunction and, in conclusion, a summary (5.7).

The following section gives a description of the study population.

5.2 DEMOGRAPHIC INFORMATION

The information in this section will be discussed under three headings: (5.2.1) study participants' demographic information, (5.2.2) family structure in which the participants grew up, and (5.2.3) maternal information, including educational level and employment status. Howell, Lynch, Platzman, Smith and Coles (2006, p. 124) emphasise the importance of using a socio-economic matched control group when carrying out research on alcohol-affected children. Using this approach, the chance of attributing deficits caused by the poor circumstances to prenatal alcohol use is eliminated. By using socio-economic matched groups in this study, the deficits caused by prenatal alcohol use could thus be isolated.

5.2.1 Study participants' demographic information

There were 14 males (46.7%) and 16 sixteen females (53.3%) in each of the case and control groups. Although this was not planned, the gender distribution of this study correlated with those of previous prevalence studies done in the same two areas of the Western Cape (May, et al., 2016, p. 211; May, Blankenship, & Marais, 2013, p. 823).

Mean ages for the cases (7 years, 5 months) and controls (7 years, 4 months) were one month apart, which correlates with the inclusion criteria of the study as discussed in Chapter 3 (3.4.1).

All the children from the study group were from the Coloured ethnic group. This is in line with the composition of another FASD prevalence study performed in the BRAM area with an ethnic composition of 92,9% coloured, 5,6% black and 1,5% white (May, et al., 2016, p. 208). The diagnostic process of the two studies, from which the study population of this particular study was non-randomly selected, was described in Chapter 2 (2.2.3).

and controls were enrolled in Grades R and 1. Nine of the cases and 14 of the controls were in Grade 3, which meant that the six cases and one control in Grade 2 did not meet the minimum requirements to be promoted to Grade 3. There were therefore five more cases than controls who did not meet the minimum requirements for Grade 3, which might indicate that more children with FASD already faced difficulties with meeting academic requirements in the foundation phase.

5.2.2 Family structure

The highest percentage of cases (60.9%) as well as controls (45.0%) were from families with two parents. The biological mother lived either with the biological father or with a new partner or boyfriend. Fourteen children (seven cases and seven controls) from the study sample were raised by single mothers. From the case group, four of the mothers together with their children lived with their parents, while the same was true for only three of the control group. These mothers had extra support with raising their children, as well as financial support. Lastly, two of the cases and four controls were in foster care. The remainder of the children were not reported on and thus no information was available. During the FASER maternal interviews, 30 of the 53

(56.6%) mothers who responded (groups combined) described their pregnancies as being stressful, ranging from medium to extremely stressful, with no significant difference between the two groups. Fifteen of the 26 mothers (57.7%) of both groups in the in-school study reported being physically abused at home. There were no statistical differences between the groups regarding abuse against the mothers (8 FASD group/7 controls). In a group of rhesus monkeys, Schneider et al. (2008, p. 100) found that prenatal stress significantly increased withdrawal responses to repetitive tactile stimulation, compared to no prenatal stress. This is an important consideration for this study as well, since a large percentage of both the cases and controls had been subjected to prenatal stress.

5.2.3 Maternal information

- **Educational level**

Information from the FASER-SA maternal interviews indicated that the baseline of highest grade achieved at school was lower for the mothers of the FASD group. Two of these mothers only achieved Grade 4 and only one reached Grade 5, while the baseline for the control group was Grade 6. In total, nine of the mothers of the FASD group did not progress past primary school, compared to five of the mothers of the control group. Only one mother from the FASD group and two from the controls finished secondary school. The baseline of scholastic achievement was thus lower for the mothers of the children with FASD than for the mothers of the children not affected, while the highest level of achievement was the same. Both May et al., (2008, p. 742) and May and Gossage (2011, p. 19) reported that women with FASD children had lower levels of education than mothers of controls. May et al. (2008, p. 742) found the lowest mean education among mothers of FAS children and the highest in the controls. According to Sutherland (2015, p. 1), better educated parents usually also meant a higher household income, which translates into better opportunities and stimulation for the children. The importance of early intervention and enriched environments in the early years for children with FASD has been discussed in 2.4.2.1 and 2.6.2 (Jacobs & Schneider, 2001, p. 37; Kraemer, 2001, p. 53; Lane & Schaaf, 2010, p. 386; Idrus & Thomas, 2011, p. 83).

- **Maternal employment status**

As noted in Chapter 4 (Table 4.3), information was available on the employment status of 24 mothers of the FASD group and 22 mothers of the controls. Twice as many mothers of the control group were employed, either part-time or full-time, than mothers of the cases. Of the mothers of the controls, 54.6% were employed full-time, while 58.3% of the mothers of the FASD group were unemployed. The difference in employment status between the mothers of the two groups was significant, with the mothers of the control group in a much better position to contribute to the financial status of their families. One must assume that unemployed mothers, living in poverty, would have monetary resources too limited to supply their children with enriched environments or to foster their development, as discussed under Educational Level. In the researcher's experience, the knowledge of these mothers about early childhood stimulation was also limited. Unless it was pointed out to them, or the child suffered from severe developmental delays, they seldom sought help during the child's preschool years. May and Gossage (2011, p. 19) found that in most population-based studies, women with FASD children were more frequently unemployed or underemployed, which was also true of the mothers of the children taking part in this study.

In an important study, Hart and Risley (2003) observed 42 families with one- and two-year-old children who were learning to talk, over a period of two and a half years. These parents were from three different socio-economic groups, higher socio-economic (professional), middle to lower socio-economic (working class), and families on welfare. The unfortunate conclusion of this study was that the average child on welfare had less than one third of the experience of words heard by the average child in a professional home and half of that of the average working-class child. This means that a child in a welfare family may hear up to 13 million words less than the average child in a working-class family. Children with FASD from welfare families (58.3%) would also have more limited vocabularies than their peers in the working-class. This phenomenon may have been one of the factors influencing the outcome of Praxis on Verbal Command, as will be discussed in 5.5.2 (g).

5.3 DIAGNOSTIC INFORMATION

It is important to note that the prevalence of FASD proved to be highest in rural areas surrounding the towns studied in the Western Cape Province (May, et al., 2016, p. 208). Numerous other studies confirm that factors such as regular binge drinking, low socio-economic status, insufficient nutrition, high fertility and challenging conditions for pre- and postnatal development were prevalent in these rural areas and in combination elevated the prevalence and severity of FASD (May et al., 2016, p. 208; May, P. A. et al., 2008, p. 738; May P. A. et al., 2013, p. 510).

The children in this study were selected from those in the longitudinal study, and were from the area described by May et al. (2016, p. 208) as more rural and of lower socio-economic status than the community in previous Western Cape studies. Of the children with FAS (17) in this study, 64.7% (11) were from this area. One would thus expect these children to present with more severe sensory integration difficulties and dysfunctions.

The results of the Sensory Profiles and the SIPT will be discussed in 5.4 and 5.5. As explained in 4.1, clinical diagnosis of sensory integration dysfunctions is based on categories or patterns of dysfunction identified and confirmed through research. For this reason, the researcher considered it to be of practical importance to review how the two groups performed when compared to these categories/dysfunctions. This would give a clearer picture of the practical outcomes of testing for all the children included in the sample. As discussed, it is statistically inappropriate to cross-tabulate the group comparisons without taking into account the pairings. To investigate the effect of fetal alcohol spectrum symptoms on sensory integration, paired t-tests were therefore used, as they were statistically more appropriate for the study design. The results should always be interpreted bearing this distinction in mind.

5.4 SENSORY PROFILES

5.4.1 Sensory Profile Caregiver Questionnaire

Until recently, sensory integration difficulties and dysfunctions were areas of neurobehavioral function in children with FASD which were not well documented (Jirikowic, Olson, & Kartin, 2008a, p. 118), although it was recognised that sensory processing information could enhance our understanding of disorders (Dunn, 2001, p. 614). From the research of people such as Abele-Webster, Magill-Evans and Pei (2012), Jirikowic, Olson and Kartin (2008a), Franklin, Deitz, Jirikovic and Astley (2008), Carr, Agnihotri and Keightley (2010), it became evident that sensory integrative problems are prevalent in children with FASD.

As noted in Chapter 4 (4.4.1.1) (Table 4.5), the results of the SP revealed that the caregivers of the group with FASD in this study reported more indicators of sensory processing difficulties than did the caregivers of the control group without FASD. More children with FASD showed probable or definite differences in sensory processing on 22 of the 23 subsections of the SP, when compared to the controls. Significant differences (indicated by Fisher's exact test) between the two groups were present in nine of the subsections, namely Sensory Seeking, Inattention/Distractibility, Fine Motor/Perceptual, Auditory Processing, Visual Processing, Vestibular Processing, Touch Processing, Multisensory Processing, and Modulation of Sensory Input Affecting Emotional Responses.

The three factors (Table 4.5) of Sensory Seeking, Inattention/Distractibility and Fine Motor/Perceptual, mentioned above, where the cases scored significantly lower than the controls are also three of the four factors found to be the best discriminators between children with and without disabilities (Ermer & Dunn, 1998, p. 286). The fourth factor, Oral Sensory Sensitivity, also indicated a notable difference between the cases and controls of this study according to the paired sample t-test results, although the difference was not statistically significant. It could thus be concluded that the children with FASD participating in this research obtained scores on the SP which correspond to those of children with disabilities.

The prominence of **sensory seeking behaviour** of the cases (90% (n=18) scored in the Definite and Probable Difference ranges) correlates with similar findings of previous studies, as discussed in 2.4.3.1 (Jirikowic, Olson, & Kartin, 2008a, p. 126; Carr, Agnihotri, & Keightley, 2010, p. 1026; Franklin, Deitz, Jirikovic, & Astley, 2008, p. 270). The Short Sensory Profile (SSP) has a combined section for Underresponsive/Seeks Sensation, and although the cases in this study correlated with sensory seeking, this was not the case with Poor Registration. The researcher felt that the caregivers either did not properly understand some of the more abstract questions in the Poor Registration factor relating to the children's emotional responses or were unsure about the answers due to a lack of knowledge of their child. This included questions such as: "Doesn't perceive body language or facial expressions (for example, unable to interpret)" or "Doesn't have a sense of humour." The caregivers tended rather to say that something seldom or never happened; as a result, the children scored higher when the caregivers did not fully understand the meaning of the question or when their knowledge of the children was lacking.

The highest percentage (70.0% (n=14)) of cases in any of the factors was reported in the Definite Difference range for **Inattention/Distractibility** (Table 4.5). In combination with the children reported in the Probable Difference range, this amounted to 85% (n=17), as opposed to the 35.0% (n=7) of controls (p=0.0042). The majority of the cases therefore experienced difficulties with attention, which correlates with the literature as discussed in 2.2.5.3 (b). In four of the seven questions in this section the cases obtained the lowest scores, suggesting the presence of low neurological thresholds causing sensory sensitivity. This correlates with finding of Kooistra et al. (2009, p. 205), that although both children with ADHD and FASD experience problems with sustained attention, the ADHD group have problems handling understimulation, while the children with FASD may experience problems with overstimulation. Coles, Platzman, Lynch and Freides (2002) and Mattson, Calarco and Lang (2006) both confirmed that sustained attention, processed in the visual modality, was more affected than that in the auditory modality. This however could not be confirmed from the information obtained with the SP, since none of the seven questions in this section was specifically related to the visual system. Nevertheless, it is worth noting that five of the questions in this section were related to auditory function and the results support the literature discussed in Chapter 2.3.1, where the numerous auditory processing problems of children with FASD were documented.

The difference between the cases and controls with **Fine Motor Perceptual** was also significant (p=0.0005). A discrepancy between the two groups was recorded, with 50.0% (n=10) of the cases in the Definite Difference range and none of the controls (Table 4.5). The lowest scores in this section suggested possible visual perceptual and fine motor difficulties such as poor handwriting in a large percentage of the cases.

Fine motor and handwriting difficulties in children with FASD were recorded in the literature as discussed in 2.4.3.2(b).

When comparing the groups, significant differences were found in five of the six items in the grouping of **Sensory Processing** (Table 4.5). The Sensory Processing grouping measures the child's responses to sensory stimuli in relation to five modalities as well as multisensory processing (Dunn, 1999, p. 14). Sensory processing refers to the way in which the central and peripheral nervous system receives incoming sensory information from tactile, vestibular, proprioceptive, visual, auditory, olfactory and gustatory sensory systems and turns them into responses (Bundy, Lane, & Murray, 2002, p. 401; Miller, 2006, p. 4). Of the six items, the highest percentage of cases in the Definite Difference range were with Auditory Processing (50% (n=10)). This correlates with the findings on the SSP (Auditory Filtering) of all three previous studies investigating sensory processing in children with FASD, as discussed in 2.4.3.1. Similarly to this current study, the previous studies also reported significant differences with visual and tactile sensory processing, keeping in mind that the SSP only reports on sensory sensitivity. Although only 35.0% of the cases scored in the Definite Difference range and 10.0% in Probable Difference with Touch Processing, this was still significantly more than the controls with 5.0% and 5.0% respectively (p=0.02).

Seventy percent of the cases in this study scored in the Definite Difference range for Vestibular Processing. The lowest means in this section were scored in statements such as "Seeks all kinds of movement and this interferes with daily routines (for example, can't sit still, fidgets)", "Seeks all kinds of movement activities (for example, being whirled by an adult, merry-go-rounds, playground equipment, moving toys)", and "Twirls/spins self frequently throughout the day." This is an indication of high neurological thresholds with sensory seeking behaviour. From analysing the separate items in the sections for Auditory, Visual and Multisensory Processing, it appears that the sensory responsivity of the children with FASD was not just either high or low with regard to these sections. In the auditory section, low scores were for example noted in "Has trouble completing tasks when the radio is on," which indicates a low neurological threshold, in contrast with "Doesn't respond when name is called, but you know the child is hearing OK," which indicates a high neurological threshold.

This may mean that some of the children have fluctuating responsivity to incoming stimuli. Because of the underlying neurological problem, the brain fails to organize, regulate and assimilate incoming sensory information. Children with fluctuating responsivity may become easily overstimulated due to a lack of appropriate inhibition of sensory stimuli, or they may appear unaware of sensory input, remaining oblivious of their surroundings (Murray-Slutsky & Paris, 2000, p. 112). These children are characteristically unpredictable and inconsistent in their behaviour.

Although responsivity may fluctuate in some of the cases, it seems that the neurological thresholds may also differ between children with FASD. Schneider et al. (2009, p. 5) found in their studies that "the timing of prenatal alcohol exposure affected the behavioural responsivity of monkeys to sensory stimuli," with the response depending on both genotype and timing of exposure.

Eighty-five percent of the cases scored in the Definite Difference (25.0%) and Probable Difference (60.0%) ranges of Multisensory Processing (Table 4.5). Two of the items with the lowest scores in this area suggest poor attention/distractibility, and one suggests the sensory seeking behaviour inherent in high neurological thresholds. Both these items suggest that the children may have trouble focusing on tasks and in the case of those with sensory seeking behaviour, may need sensory strategies which will increase input to meet their high thresholds (Dunn, 1999, p. 36)

With regard to the **Modulation** section, a significant difference between the cases and controls was only recorded for Modulation of Sensory Input Affecting Emotional Responses. Four of the cases (20.0%) and three (15.0%) of the controls fell in the Definite Difference range, with seven cases (35.0%) and only one control (5.0%) in the Probable Difference range of this section.

It is also important to note that 55.0% (n=11) of the cases scored in the Definite Difference range and 20.0% (n=4) in the Probable Difference range for Modulation Related to Body Position and Movement. An analysis of the separate questions on the SP indicated, as in the case of Vestibular Processing, a tendency toward high neurological thresholds. The low scores of the cases in this area indicate that these children enjoy excessive movement experiences and like movement which involves risks such as climbing. According to Dunn (1999, p. 36), they are probably creating opportunities to increase sensory input to meet their high thresholds. It is especially important that educators are aware of this, since they will need to include sufficient movement experiences in their daily routine to help these children meet their thresholds and be able to focus.

While the number of cases in comparison to controls was also higher in all three sections of **Behavioural and Emotional Responses**, the differences between them were not significant. Despite this, 55.0% (n=11) of the cases still fell in the Definite and Probable Difference ranges for Behavioural Outcomes of Sensory Processing, reflecting the child's ability to meet performance demands (Dunn, 1999, p. 15). Items with the lowest scores which might indicate possible problem behaviours include stubborn and uncooperative behaviour, poor frustration tolerance, sensitivity to criticisms, and immature reaction to situations. Although the difference was not statistically significant (p=0.19), only 25.0% (n=5) of the controls fell in this category. These problem behaviours may be challenging for caregivers as well as for educators, and will necessitate a firm knowledge of the child's strengths and weaknesses in order to adapt expectations to a level the child can master and thus elicit an adaptive response. Problem behaviours have been frequently discussed in the literature, as presented in 2.2.5.3 (d).

The paired sample t-test, comparing the means of the pairs of controls and cases (Table 4.6), showed that, as in the case of comparing the two groups, the means of the cases were lower than those of the controls in all the Factor and Section scores, with the exception of Sedentary. T-test results showed significant differences between the cases and controls in eight subsections (Table 4.6).

A comparison of the matched pairs indicated a noteworthy difference between the cases and controls in the sensory processing category. The only difference was that Oral Sensory Processing was also implicated when comparing the pairs, while Multisensory Processing was implicated when comparing the two groups. From this one can deduce that the children in the group with FASD experienced problems processing sensory information. According to Dunn (1999, p. 11), learning occurs when a child receives accurate sensory information, processes it and uses it in subsequent organised behaviour. When the brain receives inaccurate or unreliable sensory information, the child's ability to process the information and produce responses is disrupted, negatively influencing participation in daily occupations.

When comparing the pairs, noticeable differences were also observed in three factors (Table 4.6), Oral Sensitivity, Inattention/Distractibility and Fine/Motor Perceptual. The last two factors were also significantly different when comparing the two groups. Although there was a noticeable difference with the factor Sensory Seeking when comparing the two groups (p=0.024), the difference was not significant in the comparison of the pairs (p=0.07). The discrepancy between the groups was significant (90.0% of the cases in comparison to 50.0% of the controls scored in the Definite and Probable Difference ranges), but when the groups were paired according to gender and age, the p-value was 0.07, which did not reflect a significant difference.

A summary of the results as shown in the **four basic quadrants of responsivity** in Table 4.7 indicates a significant difference between the cases and controls with the **Sensory Sensitive** quadrant. A total of 80.0% (n=16) of the cases scored within the Definite Difference range, which is more than in any of the other quadrants. The items with the lowest scores were related to low auditory and oral thresholds (related to taste) and multisensory processing causing distractibility. High percentages were also recorded in **Sensation Seeking** (90.0%, n=18), scored in Definite and Probable Difference (more than others) ranges, as well as **Low Registration** (65.0%, n=13), scored in Definite and Probable Difference (more than others) ranges. The fact that 65.0% of the cases reflected dysfunction in this quadrant when using the Worksheet for Calculation of Quadrant Scores, as against the 15.0% when using the SP Summary Score Sheet, confirmed the researcher's hypothesis that the specific questions asked may have influenced the outcome of the latter summary. The two scoring methods used different questions from the questionnaire to determine dysfunction.

The least number of cases obtained scores in the Definite and Probable Difference (more than others) ranges for **Sensation Avoiding** (45.0%, n=9). Most prominent in this quadrant seemed to be emotional avoiding behaviour, such as stubbornness and an unwillingness to cooperate, as well as poor frustration tolerance.

The results from the Sensory Profile School Companion will be discussed next, followed by a summary of both profiles and a conclusion.

5.4.2 Sensory Profile School Companion

A general observation from the completed School Companion Teacher Questionnaires was that the bigger the classes, the more the educators were inclined to describe the child's behaviour as occurring "occasionally" or "seldom", unless the child was especially unruly or had exceptional learning difficulties (3.9.1). The 30 children in Grades 2 and 3 who took part in the study were all in classes with between 30 and 40 learners, making it very hard for the educator to pay detailed attention to individuals. The other half of the study population were in Grades R and 1, in classes with between 20 and 30 learners.

It is clear from the information on Table 4.8 on the Sensory Profile School Companion Teacher Questionnaire, that the children with FASD in this study group obtained lower scores than the children not diagnosed with FASD in all 13 sections on the Summary Score Sheet.

Significant differences between the cases and controls were reported for three of the sections, Registration (p=0.01), Avoiding (p=0.04) and School Factor 1 (0=0.05), as seen in Table 4.8.

Sixty-two percent of the cases scored in the Definite and Probable Difference ranges for **Registration**. The cases obtained the lowest scores in this section for questions relating to the missing of auditory instructions, inefficiency in doing things, for example by moving slowly, wasting time and making tasks more complicated than they were, as well as a lack of showing emotion. The same percentage (62.0%) was obtained by the cases in the Definite and Probable Difference ranges for Avoiding. The lowest mean scores in this section were for statements such as "Doesn't have a sense of humour" and "Avoids eye contact", meaning that these behaviours were more frequent in the classroom. It is interesting to note that both these questions were scored one point higher by the caregivers in the SP. The opposite happened with statements such as "Is stubborn or uncooperative" and "Is frustrated easily", which the caregivers scored as happening more frequently than did the educators. This may mean that the children were less inclined to negative behaviour in the classroom and possibly tried to conform to the group as far as possible.

A third section where a significant difference between the cases and controls was observed was **School Factor 1**, which represents the student's need for external support (Dunn, 2006, p. 41). The 58.0% of cases with Probable/Definite Difference scores in School Factor 1 will need a lot of extra sensory input to activate their high thresholds and prepare them for learning (Dunn, 2006, p. 41). School Factor 1 incorporates both Seeking and Registration sensory patterns, since both are high threshold conditions (Dunn, 2006, p. 41). The implication here is that the sensory needs of these learners must be met to optimise their learning experiences (Dunn, 2006, p. 41).

According to the educators' perceptions, 62.0% of the cases scored in the Definite and Probable Difference ranges for **Touch**. The lowest score in this section was for the statement "Plays or 'fiddles' with objects or school supplies (e.g. pencils, notebooks, folders)". Playing or fiddling with objects may be a modulating strategy for children with either high or low sensory thresholds. In the case of high thresholds, the educator may have to supply the child with extra sensory input to keep their high thresholds activated (Dunn, 2006, p. 41). This may call for strategies such as movement breaks or objects to fiddle with or chew on. Children with low thresholds may become overloaded quickly in a normal classroom setting, which may interfere with their ability to get instructions and complete tasks independently. In order for them to participate successfully, they will need reductions in stimuli which can be ensured for example by correct placement or less visual stimuli on the walls. The lowest scores, contributing to a percentage of 55.0% cases in the Definite and Probable Difference ranges for Visual, were for visual distractibility, which is usually related to low sensory thresholds and sensitivity. These children will become quickly overloaded in a classroom with bright lights, with the walls covered in colourful posters, and where learners constantly move around. The educator will have to adjust to their needs so as to enhance learning in these children.

With **Movement**, the educators reported 48.0% of the cases and 26.7% of the controls in the Definite and Probable Difference ranges. This was much less than the caregivers' score of 85% for Vestibular as discussed earlier. This may have been because of the more restricted, disciplined classroom environment compared to the home environment. The lowest scores in this section reflect mainly high sensory thresholds, but with active seeking behaviour, as well as passive responses. If children with high neurological thresholds are not given the opportunity to meet these thresholds they will struggle to focus on instructions or complete tasks. The educator's goal should therefore be to incorporate additional sensory input, such as movement breaks, into the learner's daily classroom routine, so that the high thresholds can be met (Dunn, 1999, p. 36).

With **Behaviour**, 55.0% of the cases scored in the Definite and Probable Difference ranges according to the observations of the educators. These results agree with those reported by the caregivers in the section Behaviour Outcomes of Sensory Processing on the SP.

When comparing the matched pairs, a statistically significant difference of p=0.05 between the means of the cases and controls was reported for **Auditory** (Table 4.9). Sixty-two percent of the cases scored within the Definite and Probable Difference ranges. The educators reported some cases to be easily distracted by auditory stimuli from the environment and others as oblivious of instructions and their immediate environment. This correlates with the reports of the caregivers as discussed in 5.4.1.

School Factor 3 also indicated a significant difference (p=0.04) between the scores of the pairs (Table 4.9). This factor represents the learner's range of tolerance for sensory input and is more related to Sensitivity and Avoiding patterns (Dunn, 2006, p. 43).

Results from the SPSC confirm the presence of sensory processing and integration difficulties in the FASD population in the classroom. In general, the percentages recorded in the Definite and Probable Difference ranges were lower than those recorded by the caregivers on the SP in the home settings.

It should also be noted that, similar to the study of Jirikowic (2003, p. 104), an average of 28.7% of those children not diagnosed with FASD were classified in the Definite and Probable Difference ranges on the SP. This suggests that, although fewer, the controls also demonstrated behavioural symptoms associated with sensory processing difficulties.

The percentages of controls scoring within the Definite and Probable Difference ranges on the SPSC were lower than on the SP. Despite the possibility of other diagnoses such as ADHD, the outcome on the SP suggests that sensory processing difficulties also occurred in that part of the sample not diagnosed with FASD, although to a lesser extent than those with FASD.

5.4.3 Results summary of sensory profiles

Before drawing a final conclusion on the results from the two Sensory Profiles, one should keep in mind that there is a significant variation in the diagnostic groupings of FASD due to the quantity, frequency and timing of prenatal maternal alcohol consumption, as discussed in Chapter 2 (May P. A., et al., 2013, p. 502). Evidence from numerous animal and human studies suggests a considerable variability in the frequency and magnitude of the neurobehavioral, neurological and neuroimaging manifestations of prenatal alcohol exposure (Paley & O'Connor, 2011, p. 64; Florida State University: Centre for Prevention & Early Intervention Policy, 2005, p. 9). One should also keep in mind that timing of prenatal alcohol exposure has an effect on the behavioural responsivity of monkeys to sensory stimuli, as noted earlier in this chapter (Schneider, et al., 2009, p. 1).

Considering both the literature and the results of this study, it is no surprise that the sensory processing and integration difficulties and dysfunctions of children with FASD as observed with the Sensory Profiles seem to occur in multiple sensory systems, ranging from under- to over-responsiveness and, as suggested earlier in this section, fluctuating responsivity. The same tendency was reported by Jirikowic (2003, p. 119). From the data in this study it seems that the children with FASD experience significant difficulties with low registration, causing sensation seeking behaviour frequently presented as excessive movement. Sensory sensitivity, especially with regards to the auditory and visual systems can also lead to overstimulation, excessive movement and to a lesser degree, avoiding behaviour. Avoiding behaviour was observed more frequently by the educators in the classroom than by the caregivers at home, perhaps because of the more confined space in the classroom, with more movement and sounds.

Auditory processing difficulties seem to be a prominent difficulty in the cases, but were also recorded in 30.0% of the controls. Difficulties with processing the incoming sensations were prevalent in the visual and vestibular systems, and were also recorded frequently by the educators. The behavioural outcomes of sensory processing were reported as problematic by more than half of the caregivers, as well as by the educators of the cases. Difficulties with Modulation of Sensory Input Affecting

Emotional Responses were also reported by more than half of the caregivers of the cases.

Lastly, according to the SP, Inattention and Distractibility appeared to be prominent problems. This correlates with School Factors 1 and 3, which represent the student's need for support and range of tolerance for sensory input in the classroom (Dunn, 2006, p. 41 & 43). As reported by the educators, the results confirm the observation that sensory responsivity varies and fluctuates between high and low. On the SP, Fine Motor/Perceptual was a noticeable difficulty, with 50% of the cases falling in the Definite Difference range.

Modulation and integration of sensory input is a crucial function of the central nervous system and a prerequisite for daily occupational performance (Dunn, 1999, pp. 10-11). Performance problems and outcomes relating to the identified sensory integration difficulties and modulation problems could therefore by expected. A discussion will follow of the possible correlations between sensory processing and modulation problems identified and the functional problems experienced by the children as consequence of this.

In Chapter 2.4, we discussed the disconcertingly low pass rate of Grade 3 learners in the CWED in mathematics and language. Jirikowic, Olson and Kartin (2008a, p. 129) found that sensory processing behaviours (SSP) correlated significantly with mathematics and spelling. Similarly, a pattern of auditory filtering problems and sensory under-responsiveness with sensory seeking behaviour was associated with academic underachievement in a study by Ashburner, Ziviani and Rodger (2008, p. 564). Similarly to the finding in this study population, they found that children with ASD who had trouble attending to verbal instructions in the presence of background noise, and who often focused on sensory seeking behaviour, appeared more likely to underachieve academically (Ashburner, Ziviani, & Rodger, 2008, p. 570). The auditory filtering difficulties were also associated with attention problems. Although deficits in cognitive processing speed are a core impairment of children with FASD (Glass, Ware, & Mattson, 2014, p. 439), sensory processing also affects cognitive performance (Dunn, 2001, p. 611). Research found that auditory processing seems to be particularly relevant, and that poor auditory processing slows down the speed of cognitive processing even more (Dunn, 2001, p. 611). As discussed in Chapter 2.5.2., the importance of a well-regulated sensory system for long-term development, including academic outcomes, was emphasized by Kodituwakku (2010, pp. 723-724) and confirmed in a study by Nash et al. (2015), in which improved sensory regulation facilitated improvements in behaviour and simple executive function skills.

Both the caregivers and educators who took part in this study reported sensory-based behavioural problems for 55% of the cases. Franklin, Deitz, Jirikowic, & Astley (2008, p. 270) found that children with FASD who demonstrated sensory processing deficits demonstrated significantly more externalising behavioural problems, specifically in the domains of attention, rule breaking, socialisation and thought problems. Neither the SP nor SPSC in their questions address the issue of serious behavioural problems directed toward the external environment. During the inverviews with the caregivers to complete the Sensory Profiles, only two of the caregivers of children with FASD reported aggressive or destructive behaviour. Serious socialisation problems have been reported neither by the caregivers nor by educators on the SPs. This may be explained by research done by Whaley, O'Connor and Gunderson (2001), who suggested that deficits in socialisation become more prominent with age. Jirikowic, Olson, & Kartin (2008a, p. 133) found no significant relationships between sensory processing behaviours and teacher-rater performance as was expected. They also came to the conclusion that, in their sample, the children with FASD demonstrated a variable behavioural profile, rather than one consistent behavioural pattern (Jirikowic, Olson, & Kartin, 2008a, p. 113). They suggested the use of a context-specific measure, such as a teacher-rated sensory processing measure. This observation and recommenation of Jirikowic, Olson and Kartin (2008a) prompted the researcher to use both Sensory Profiles in their specific contexts.

Although the SPs do not specifically target sleeping patterns, a study by Wengel, Hanlon-Derman and Fjeldsted (2011, p. 388), as discussed in Chapter 2.4.3, found significant correlations between items on the SP and the Children's Sleep Habits Questionnaire. Significant correlations were found with the following items, on which the cases of this study also performed poorly:

- Fine Motor/Perceptual correlated with bedtime resistance.
- Behavioural Outcome of Sensory Processing correlated with sleep onset delay.

- Sensory Sensitivity and Behavioural Outcomes of Sensory Processing correlated with sleep duration.
- Registration and Modulation of Visual input, Affecting Emotional responses/Activity Level correlated with night wakening.
- Behavioural outcomes of Sensory Processing correlated with parasomnias.

At least 55% of the cases scored in the Definite and Probable Difference ranges on the SP in the specifically mentioned items. One could therefore deduce that these cases may also be at risk of sleep disturbances. This is quite alarming, since Touchette et al. (2007, pp. 1217-1218) reported that specific cognitive deficits and high hyperactivity scores at age six were most strongly associated with a pattern of short sleep duration at the age of 2,5 years, despite the increase to normative sleep patterns from age 3,5 through six years. This suggests that insufficient sleep during the first few years of life may have long-standing consequences. Adding to the sensory processing difficulties, the low socio-economic environment of the children was not conducive to a good sleeping pattern. The children hardly ever had their own bedrooms and in many cases not even their own beds. Informal settlement housing mostly consists of one room where everything happens. A foster mother, for example, reported to the researcher that only a cupboard separated her and the child, sleeping in the same bed, from the father and his girlfriend. She complained of the father being regularly drunk and very noisy.

The results of a study by White, Mulligan, Merrill and Wright (2007, p. 158) suggest that children identified with sensory processing difficulties on the SP are likely to experience certain challenges in performing everyday occupations. These children seem likely to face difficulties with at least Personal and Instrumental Activities of Daily Living.

Although correlations between sensory processing and adaptive behaviour in the existing literature were discussed in Chapter 2.4.3, the questions in the two sensory profiles do not provide sufficient evidence from which to draw conclusions. A few of the mothers of the cases mentioned that their children still needed help with selfcare activities, but questions in that area were not asked formally.

Lastly, a moderate to low correlation was found by Jirikowic, Olson and Kartin (2008a, p. 129) between sensory processing behaviours (SSP) and sensori-motor performance, including visio-spatial abilities. The sensory processing difficulties identified in this study may therefore have influenced the poor results of the Grade R class in the CWED on Ayres's Clinical Observations and the test for visio-motor integration that were previously done with them, as described in Chapter 2.5.

This study needs to take into account the possibility that the identified sensory processing problems of the cases, and to a lesser degree some of the controls, may have had a debilitating effect on their occupational performance with sleep, school (academic work), behaviour at home and school, motor performance and adaptive skills.

To be conclusive, more research and a bigger study sample are still needed, but it is clear that the results from the Sensory Profiles (Caregiver Questionnaire as well as School Companion) of this study, together with consistencies with findings from other studies, acknowledge the reality of sensory processing and integration difficulties in the FASD population.

5.5 SIPT

The data obtained from the SIPT will be presented in three sections: Clinical Observations, SIPT Subtests, and Patterns of Sensory Integration Dysfunction.

5.5.1 Clinical observations

Before the administration of the SIPT, clinical observations regarding prone extention, supine flexion and occular pursuits needed to be executed and recorded. No significant differences were found between the cases and controls in the comparison of the two groups, as indicated in Table 4.10.

The results showed that both the cases and controls encountered problems in assuming and maintaining the antigravity postures. With prone extention, 83% of the cases and 73% of the controls scored within the Slightly Deficient and Poor ranges. Assuming and maintaining the supine flexion position scored slightly better, but was still a cause for concern. In this observation, 60% of the cases and 40% of the controls scored in the Slightly Deficient and Poor ranges. With occular pursuits, 73% of the cases and 76% of the controls obtained scores in the Slightly Deficient and Poor ranges. Seven percent more of the cases fell in the Poor range on occular pursuits. Poor scores on occular pursuits were also reflected in a previous setting as part of a Grade R pre-test by the researcher, as desribed in Chapter 2.5. The results of this study indicate that the irregularities with eye movements cannot only be attributed as part of the FASD picture, as described in Chapter 2 by Green et al. (2009) and Paolozza, Titman, Brien, Munoz and Reynolds (2013). The latter state that the altered accuracy of eye movements can impact the everyday functions of children, for example in sports and reading (p. 1497), and the fact that a noteworthy percentage of both the cases and the controls had difficulties with eye movements warrants futher attention.

Postural control for stability and orientation of the body in space requires an intricate interaction of muscoloskeletal and neural systems, of which sensory/perceptual processes, involving the organisation and integration of visual, vestibular and somatosensory systems, form part (Shumway-Cook & Woollacott, 2012, p. 165). These systems contribute to our ability to maintain upright and antigravity postures and to move efficiently (Schaaf, et al., 2010, p. 115). An example of this is the development of antigravity postures through which the infant can raise his or her head against gravity and subsequently initiate movement (Schaaf, et al., 2010, p. 115). Well developed antigravity postures seem to form the basis for more complex postural mechanisms which allow the child to move in and out of midline positions (Schaaf, et al., 2010, p. 115). It was clear that the research sample (cases and controls) experienced problems with the latter when performing the Motor Accuracy test (Table 4.11). Poor crossing of the midline and insufficient postural adjustments were, in addtition to the formal test scores, observed by the researcher and her occupational therapy assistant in 33 (55%) of the children.

Evidence from the clinical observations thus indicates the likelihood of poor postural-ocular control in both the cases and controls.

The reason for this phenomenon warrants further reasearch. The researcher is of the opinion that cultural differences and the low socio-economic conditions of the children may have affected early development. Space in both the homes and crèches are limited, and babies are left to lie their backs for long periods with very little visual stimulation. As they get older, they are often propped into a sitting position, which means that they miss the important developmental sequence of getting up against gravity by themselves. Although not a South African study, Saccani and Valentini (2013, p. 350) compared the motor development of three population samples and found a non-linear motor developmental trend. They concluded that the differences in ways of motor development may be due to cultural differences in infant care.

5.5.2 SIPT subtests

Table 4.12 indicates that the cases in this research project scored lower on 15 of the 17 subtests of the SIPT. On the tests Localization of Tactile Stimuli and Oral Praxis the numbers of children in the dysfunctional range were equal.

Before discussing the patterns of dysfunction as described in Chapter 2.4.2.2, the individual SIPT tests will be discussed in terms of the performance of the cases, the skills for which each was assessed, and their significance for the children's occupations. The percentage of dysfunction regarding the controls will also be mentioned in each instance.

Statistically significant differences between the pairs of cases and controls were identified through the paired t-test procedure. In this case, the difference was statistically significant in 11 of the subtests (Table 4.11), including Space Visualization (p=0.02), Figure Ground Perception (p=0.02), Manual Form Perception (p=0.03), Kinesthesia (p=0.009), Finger Identification p(=0.003), Graphesthesia (p=0.05), Praxis on Verbal Command (p=0.01), Design Copying (p=<0.0001), Constructional Praxis (p=0.0015), Sequencing Praxis (p=0.003), and Bilateral Motor Coordination (p=0.01).

a) Space Visualization

The results indicate that 53.3% (n=16) of the cases scored within the dysfunctional range for Space Visualization. It should be noted that 36.7% (n=11) of the controls also experienced problems in this test (Table 4.12). The results of the paired groups (Table 4.11) showed that a significant number of cases scored below the norm compared to the controls (p=0.02).

Since motor performance is not required and does not enter the score for Space Visualization, it is regarded as a visual rather than a visual-motor test. It requires visual space perception and, in the more advanced items, mental manipulation of objects in space as well (Ayres, 2004, p. 2).

Significance for childhood occupations

"The ability to perceive shapes and designs is a foundation skill for reading and writing" (Bodison & Mailloux, 2006, pp. CE-4). Children who struggle on this measure are likely to show confusion about the spatial aspects of many classroom, playground and ADL tasks. The ability to envisage what an object would look like in another orientation is needed for tasks such as putting on a shirt, moving a chair through a door, or organizing letters within a word (Bodison & Mailloux, 2006, pp. CE-4).

Problems with visual spatial perception were reported by Jirikowic (2003, p. 87) for the FASD population. She also found a moderate correlation between visio-spatial perception and mathematical performance. Her findings offered important support for this study's results, since mathematics is one of the problem areas in the performance of the foundation phase learners of the CWED, as described in Chapter 2.5.

In their study to examine the relationship between mathematics and attention, working memory and visual memory in children with prenatal alcohol exposure and controls, Crocker, Riley and Mattson (2015, p. 8) suggested that "it appears that spatial processing may be driving these relationships, rather that actual attention and working memory abilities, per se." They reasoned that more basic visual-spatial processing may be of value for capabilities such as the alignment of digits in mathematical calculation, as well as for the concepts of borrowing and carrying (Crocker, Riley, & Mattson, 2015, p. 8). Their evidence also suggests that children in general represent numerical ranges on a left-to-right mental number line. Thus impairments in the ability to visually manipulate information, as was expected in the Space Visualization test as well, may result in inefficient use of this mental representation of the number line and consequently impaired mathematics performance (Crocker, Riley, & Mattson, 2015, p. 8). According to the chief marker of the WCED on the 2016 Grade 3 provincial systemic tests, one of the areas of weakness was the inability of the children to arrange numbers either from smallest to largest or from largest to smallest (Cornelissen, 2017).

Poor performance in Space Visualization may thus play a role in poor mathematical performance.

b) Figure-Ground Perception

Seventy percent of the cases (n=21) and 43.3% (n=13) of the controls fell within the dysfunctional range for figure-ground perception. A significant difference was found when comparing the pairs (p=0.02) (Table 4.11).

This test assesses the motor-free ability to perceive and distinguish a foreground figure from a rival background. Since the test is not directly related to somatosensory processing or praxis, it may help to identify a primary visual perceptual deficit (Ayres, 2004, p. 134). The test is more sensitive than most of the other SIPT tests for high-level central nervous system integrity (Ayres, 2004, p. 135).

Significance for childhood occupations

Seventy percent of the cases may experience difficulties with tasks/activities such as finding objects in a desk, locating and isolating specific information or attending to items on a blackboard, building puzzles or finding a friend among others in the playground (Bodison & Mailloux, 2006, pp. CE-4).

Children who experience difficulty with visually differentiating objects, shapes, people or structures, may be slow workers, making more incorrect choices than their peers (Bodison & Mailloux, 2006, pp. CE-4). From her experience in the education system, the researcher was aware of educators' complaints about inaccurate and incomplete copying from the blackboard and work cards.

c) Manual Form Perception

A majority of cases (56.7%, n=17) and 33.3% (n=10) of the controls were recorded in the dysfunctional range for Manual Form Perception (Table 4.12).

When comparing the means of the pairs, a significant difference was found (p=0.03) (Table 4.11).

This test helps in identifying problems with form and space perception and has a strong inherent visualization component (Ayres, 2004, p. 135&194). Being successful in it requires the ability to combine tactile and kinesthetic information contributing to the perception of stereognosis. The child also has to sequentially analyse what he/she feels, forming a higher level of meaning and understanding in order to perform the next steps of the task (Bodison & Mailloux, 2006, pp. CE-6).

Significance for childhood occupations

The ability to identify shape, texture and size by touch enables a person to select and use objects without overreliance on vision (Bodison & Mailloux, 2006, pp. CE-6). Children who struggle with this are more likely to experience difficulty retrieving and finding objects in a pocket, backpack or drawer, and will be dependent on their visual system for support.

The test results indicated that more than half of the cases were probably more reliant on vision than the average child when matching shapes by touch to a visual form. With this in mind, the educator can allow the child extra time and supplementation with visual and verbal cues where appropriate (Bodison & Mailloux, 2006, pp. CE-6).

d) Kinesthesia

In the test for Kinesthesia, 43.3% (n=13) of the cases and 33.3% (n=10) of the controls scored within the dysfunctional range (Figure 4.12).

A comparison between the pairs indicated a significant difference (p=0.009) (Figure 4.11).

Kinesthesia is one of the somatosensory tests, assessing proprioception (Ayres, 2004, p. 135). The test is dependent on the ability to focus attention and the ability to rely on proprioceptive awareness to accurately move the arms and hands in space. The successful execution of the test also depends on appropriate postural adjustments, allowing the subject to freely move the arm in isolation from the trunk (Ayres, 2004, p. 135; Bodison & Mailloux, 2006, pp. CE-7).

Significance for childhood occupations

Awareness of position helps the child to maintain a position at a desk, in line, or on the playground during games. Accurate feedback of position is also essential for maintaining an optimal position for holding a crayon or pencil, ruler or scissors. Clinical observations made by the researcher and her assistant during the Motor Accuracy test found that 16 (53%) of the cases had poor pencil grips and 7 (23.3%) exhibited inadequate postural adjustments. Pressing very hard and working very slowly, having difficulty moving forward with the pen, were also noted. It therefore seems that the poor Kinesthesia recorded in 43.3% of the cases may have influenced the children's handwriting and work speed. Poor postural adjustments may, however, also correlate with poor antigravity postures as reported in 5.5.1.

e) Finger Identification

Thirty-three percent of the cases (n=10) and only 10% (n=3) of the controls scored in the dysfunctional range for Finger Identification (Figure 4.12).

A significant difference of p=0.003 was found in comparing the means of the pairs.

Finger Identification is regarded as one of the most important measures for tactile perception in the SIPT (Ayres, 2004, p. 135). The test assesses the ability to rely on tactile proprioceptive awareness alone to discriminate and identify fingers touched (Bodison & Mailloux, 2006, pp. CE-7).

Significance for childhood occupations

Since touch perception guides actions by helping us make refined adjustments, it enhances coordinated function, for example, grasp and release and selfcare activities such as getting dressed (Bodison & Mailloux, 2006, pp. CE-7; SAISI, 2011, p. 9). In combination with Kinesthesia, as mentioned in (d), dysfunctional Finger Identification may therefore contribute to the poor pencil grips of the cases.

f) Graphesthesia

Twenty percent of the cases (n=6) and 13.3% (n=4) of the controls scored within the dysfunctional range for Graphesthesia (Table 4.12).

Although there was a significant difference between the performance of the pairs (p=0.05) on Graphesthesia (Table4.11), the percentages recorded in the dysfunctional range of the two groups were fairly low.

The test for Graphesthesia evaluates the ability to translate tactile input into motor responses (South African Institute for Sensory Integration, 2011, p. 10). Spatial and temporal analysis of passively received tactile stimuli is needed to form a visual image of the stimuli. This image is used to produce a motor response to replicate what was actually perceived (Bodison & Mailloux, 2006, pp. CE-7).

Significance for childhood occupations

A low score on Graphesthesia, in combination with low scores on visual perception, may contribute to difficulty with tasks such as writing. Since high percentages of dysfunction were recorded on the visual perceptual tests of the SIPT for the cases, the low scores of 20% for Graphesthesia may add to the handwriting problems these children experience.

g) Praxis on Verbal Command

The highest percentages of dysfunction in any of the SIPT subtests were recorded for both the cases and controls in Praxis on Verbal Command. In this test, 73.3% of the cases and 50% of the controls fell in the dysfunctional range (Table 4.12).

A significant statistical difference was found when comparing the pairs using the t-test (p=0.01), meaning that when the pairs of cases and controls were compared, the cases scored significantly less well on Praxis on Verbal Command.

Skills needed for this test include the following of verbal instructions, the planning of movements based on these instructions, and the ability to rely on tactile and proprioceptive awareness to motor plan the movements of the body and limbs without a visual model (Bodison & Mailloux, 2006, pp. CE-5). The sensory systems involved include the auditory, proprioceptive and tactile systems (South African Institute for Sensory Integration, 2011, p. 16).

The researcher believes that the poor performance of the cases on this test may be due to several reasons.

- Results from the Sensory Profiles indicated auditory sensory processing problems. High as well as low sensory thresholds were identified, causing either a slow or no reaction to instructions or, conversely, auditory distractibility. This agrees with the literature discussed in Chapter 2.4.1 (d), indicating delayed auditory processing and filtering in the population with FASD.

- Limited vocabularies and/or poor body concept could have played a role in the poor performance of both cases and controls. There were, in fact, children who confused their “elbows” with their “shoulders” and who did not understand concepts such as “back of your foot”. The latter may have been aggravated by their underlying spatial perceptual problems. This limitation, in the opinion of the researcher and as supported in the literature already mentioned, seems to be inherent in the low socio-economic culture from which these children come. Since both the cases and controls were from the same socio-economic backgrounds, difficulties with Praxis on Verbal Command due to prenatal alcohol use, could thus be isolated.
- With 43.3% (n=13) of the scores for the cases in the dysfunctional range for Kinesthesia, the ability to rely on tactile proprioceptive awareness to motor plan how to move the body without a visual model, as decribed by Bodison and Mailloux (2006, pp. CE-5), also played a role. Twelve of the cases who tested within the dysfunctional range for Praxis on Verbal Command were also in the dysfuntional range for Kinesthesia. There was a distinct difference when the cases were given a visual model (without verbal instructions) to copy, as was seen in the test for Postural Praxis, where only 26.7% were in the dysfunctional range.

Significance for childhood occupations

This test may help clarify difficulties with language comprehension or auditory processing in the light of the “motor-planning ability needed to translate a verbal direction into an action” (Bodison & Mailloux, 2006, pp. CE-5). An analysis of the SIPT tests will help clarify the question of whether the child has difficulties with both language and praxis problems or with just one aspect.

Difficulty on this test may relate to functional problems such as following instructions, especially sequential ones, as well as difficulty completing tasks (South African Institute for Sensory Integration, 2011, p. 16). It seems that the poor performance of the cases on Praxis on Verbal Command does not have a single cause, but rather a combination of factors influencing their performance. Having children with dysfunctional Praxis on Verbal Command in the classroom seems to be a serious problem for educators, as it implies that a large percentage of their learners have difficulty understanding instructions and/or executing them, or understand them, but at a much slower pace than their peers because of slow auditory processing.

h) Design Copying

In the test for Design Copying, 73.3% of the cases and 33.3% of the controls scored within the dysfunctional range. Comparison of the means of the pairs of cases and controls using the t-test indicated a significant difference (p=<0.0001).

Ayres (2004, p. 137 & 203) described Design Copying as the best single indicator of visiopractic ability in the SIPT, especially drawing on practic management of two-dimensional visual space.

Significance for childhood occupations

Scores of dysfunction on this test may relate to difficulty with drawing and writing, as well as difficulty copying from the blackboard (South African Institute for Sensory Integration, 2011, p. 5).

Numerous studies have reported on the presence of visio-motor integration difficulties in the FASD population (Chapter 2.2.5.3). The data collected in this study therefore support previous findings as well as the regular complaints the researcher received from the foundation phase educators in the CWED about handwriting difficulties in their classes. The study by Duval-White, Jirikovic, Rios, Deitz and Olson (2013) that was discussed in Chapter 2.2.5.3, described the challenges the children with FASD face with handwriting legibility across alphabet, sentence and paragraph writing (p. 538). A recent study by Carlson, Rowe, and Curby (2013) confirmed that visio-spatial integration ("creating a mental representation of an image and replicating it with controlled small muscle movements" - p. 515) was an important predictor of written expression and that advances in these skills resulted in significant increases in written expression scores. Skills in alphabet writing, sentence and essay composition and spelling all rely on handwriting ability, which in its turm has been linked to visual-spatial copying skills. The reasoning is that when writing is easier, more cognitive resources are available to devote to the production of grammatically correct sentences (Carlson, Rowe, & Curby, 2013, p. 529). Although it will not alleviate underlying language and punctuation problems, research indicates that improved visio-spatial skills result in better performance on writing assignments. Improved Design Copying should therefore lead to improvement in the areas of weakness of the foundation phase learners of the CWED, namely weak basic sentence construction and spelling (Cornelissen, 2017).

Performance of numerical operations has been linked to the same specific areas of the brain which are tied to visual-spatial processing abilities. This also suggests a link between the two abilities (Carlson, Rowe, & Curby, 2013, p. 528). The understanding of numbers and quantities is linked to an understanding of spatial organisation and related movements within the parietal cortex, partly because number knowledge needs to exist in the same area as the understanding of general magnitude (Carlson, Rowe, & Curby, 2013, p. 528). Children with advanced visual-spatial integration skills should therefore be more likely to have better developed numerical abilities (Carlson, Rowe, & Curby, 2013, p. 528).

The knowledge that visio-motor integration skills are associated with achievements in maths and written expression may offer an avenue of targeted intervention for children of all ages, including those with FASD in the CWED (Carlson, Rowe, & Curby, 2013, p. 514).

i) Constructional Praxis

Eleven (36.7%) of the cases and three (10%) of the controls tested within the dysfunctional range. These results indicated that problems with Constructional Praxis do not affect the majority of the cases.

A comparison of the means of the pairs using the t-test indicated a noteworthy difference (p=0.0015).

Constructional praxis taps a practic skill unique to the SIPT. The child's ability to relate objects to one another and motor-plan a construction within three-dimensional space is evaluated with this test (Bodison & Mailloux, 2006, pp. CE-5).

Significance for childhood occupations

Dysfunction on this test may relate to practical problems such as the inability to organise desk space or construct with blocks (Bodison & Mailloux, 2006, pp. CE-5; SAISI, 2011, p. 7). In correlation with this, Howell, Lynch, Platzman, Smith and Coles (2006, p. 121) found significantly more problems with block design on the Wechler

Intelligence Scale for Children (3rd edition) in children with FASD than those without. This correlates with complaints the researcher often received from the psychologists testing the children in the CWED. According to them, the children had difficulty constructing with blocks in the Block Designs subtest of the Senior South African Individual Scale – Revised (SSAIS-R). Constructing in a three-dimensional space is a foundational skill for two-dimensional drawing copying (thus contributing to poor scores on DC) and for writing between lines as well as understanding mathematical concepts (Cornelissen, 2017), with all of which the FASD children were experiencing problems.

j) Sequencing Praxis

In this test, eleven (36.7%) of the cases and four (13.3%) of the controls fell in the dysfunctional range.

Though the difference between the groups was not significant, the t-test proved the difference between the means of the pairs to be significant (p=0.003).

This test primarily measures bilateral integration and sequencing (Ayres, 2004, p. 206). Skills required for successful execution of this test include the ability to coordinate the two sides of the body, visual, auditory as well as kinesthetic memory, and sufficient tactile proprioceptive awareness to plan how to move the hands and arms (Bodison & Mailloux, 2006, pp. CE-6).

Significance for childhood occupations

Dysfunctional Sequencing Praxis may influence rhythmical playground games and ADL activities, such as tying shoelaces. Children experiencing difficulties find it challenging to follow through on multi-step instructions (South African Institute for Sensory Integration, 2011, p. 22). Sequencing problems were evident in the 2016 Grade 3 mathematics and home language WCED systemic test results. With mathematics, the children had problems arranging numbers from small to large and from large to small, and with understanding the concept of time moving from morning to afternoon (Cornelissen, 2017). With regard to home language they were unable to adequately construct five basic sentences consisting of a sequencing of words (Cornelissen, 2017).

k) Bilateral Motor Coordination

In this test, 23.3% of the cases and 16.67% of the controls fell within the dysfunctional range.

Although there was a significant difference between the performance of the pairs (p=0.01) on Bilateral Motor Coordination, the percentages recorded in the dysfunctional range of the two groups were fairly low.

This test measures the ability of the child to move both arms and feet together in a smooth, integrated pattern (South African Institute for Sensory Integration, 2011, p. 24). Ayres warns that it should not be interpreted in isolation from other SIPT scores (Ayres, 2004, p. 139).

Significance for childhood occupations

The ability to use both sides of the body in smooth, rhythmic and sequenced actions is integral to many school and ADL activities, such as cutting, buttoning, pedaling and skipping. From experience, the researcher found that the Grade R children had difficulty performing rhythmic **contralateral** bilateral activities. The majority were able to perform the unilateral activities with ease, as was also the case in this test, indicating that the difficulty with the bilateral contralateral activities may instead be aggravated by poor crossing of the midline or insufficient trunk rotation.

No noteworthy differences were found between the two groups or between the matched pairs of the remaining six subtests. These will now be discussed, since their results are still of value.

l) Localization of Tactile Stimuli

Only one child (3.3%) in each of the cases and controls fell within the dyfunctional range for Localization of Tactile Stimuli.

Both the cases and controls did unexpectedly well in this test, considering the high level of concentration it demands. It is however seen as a fairly simple test which is easy to understand and requires little or no practic ability (Ayres, 2004, p. 136). It is also considered one of the less reliable measures in the SIPT and shows its closest positive praxis correlation with Postural and Oral Praxis (Ayres, 2004, p. 136).

Although slightly poorer, the cases also did well in these two tests, as will be discussed under (m) and (n) in this section.

The researcher observed that the mean for the cases in this research was 0.6, while the mean for the South African children with possible sensory integration dysfunction in the study by Van Jaarsveld, Mailloux, Smith Roley and Raubenheimer (2014, p. 3) (sample consisting of 78.03% white children) was -0.47, while the discrepancy between the means of the other tactile tests was substantially smaller. Comparing the results of the researcher's study to that done by Lombard (1995, p. 91), it seems there is a possibility that children from the Coloured populaton may score high in tests for Localization of Tactile Stimuli when compared to white children. Although the test used by Lombard (1995, pp. 46-47) was not standardized and is still a pilot study, the test mechanism was similar to that of the SIPT. This study found that from the four tactile tests administed (KIN, LTS, MFP and FI), the white children obtained higher scores in all the tests, except for Localization of Tactile Stimuli.

Significance for childhood occupations

Children who have difficulty with this test may experience practical problems with activities requiring general tactile awareness, such as grasp and release (South African Institute for Sensory Integration, 2011, p. 11). With only one of the cases in the dysfunctional range, this was however not the case in this study.

m) Postural Praxis

Only eight (26,7%) of the cases and five (16.7%) controls scored in the dysfunctional range for Postural Praxis.

Postural Praxis measures general practic abilities. It is important to note that it was significantly harder, especially for the cases, to assume postures on verbal command. Cermak, Morris and Koomar (1990, p. 643) found that performance on verbal command was significantly less accurate in 4-year olds compared to 6-year olds. One of their possible explanations was that the language skills of the 4-year olds had not developed sufficiently for them to accurately follow the verbal commands in comparison with the 6-year olds. Although the study referring to was already published in 1990, it seems that the limited vocabularies of the cases in this study could therefore have contributed to their poor performance, with a better performance on imitation only.

n) Oral Praxis

Five (16.7%) of both the cases and controls scored in the dysfunctional range for Oral Praxis.

Oral Praxis is closely related to perception and interpretation of sensation from the body, such as tactile, proprioceptive and visual sensations, but especially those which are tactile (Ayres, 2004, p. 138; SAISI, 2011, p. 20).

Significance for childhood occupations

Difficulty on this test may relate to problems with feeding, articulation or blowing (South African Institute for Sensory Integration, 2011, p. 20). Although a few children with articulation problems were identified during the testing, Oral Praxis does not seem to be an area of concern.

o) Motor Accuracy

Although the dysfunctional scores for Motor Accuracy, which is primarily an index of eye-hand coordination or visual space management, were only 20.0% (6) for the cases and 13.3% (4) for the controls, the clinical observations made by the researcher and her assistant during the test showed that 30.0% of the cases worked with slow, jerky movements, perseverating on one spot, and kept lifting their pens from the paper. Difficulties with pencil grasp, postural adjustments and crossing of the midline were discussed under (d). Since complaints about handwriting and fine motor difficulties have been raised regularly by foundation phase educators in the CWED, the outcome of this test was a surprise for both the researcher and her assistant. According to Ayres, this test “is not a good screening instrument for sensory integrative dysfunction in 7- and 8-year old children” (Ayres, 2004, p. 135). One should bear this in mind, since 65% of the research sample fell in this age category.

The majority (67%) of the cases in the dysfunctional range had a FAS diagnosis and the remaining two (33%) were diagnosed with ARND. This correlates to a large extent with the study by Doney et al. (2014) who found significant differences in fine motor speed and precision skills among children with FAS but not in children on the other diagnostic criteria of FASD.

It is also noteworthy that among all the studies the above-mentioned authors reviewed, more complex fine motor skills, such as visuo-motor integration, were consistently impaired in children with FASD. Studies assessing skills such as manual dexterity, grip strength and manipulation of pegboards did not consistently identify deficiencies (Doney, et al., 2014, p. 606).

p) Standing and Walking Balance

Twenty percent (6) of the cases and 13.3% (4) of the controls scored within the dysfunctional range on the test for Standing and Walking Balance.

Given the poor performance of the children with assuming and maintaining the antigravity postures, the researcher expected similar difficulties with Standing and Walking Balance. This however was not the case.

According to the systems model that evolved from the work of Bernstein, “the nervous system is seen as part of a flexible complex of systems and subsystems sharing in the control process” (Woollacott & Shumway-Cook, 1990, p. 800). Movement is therefore always an emergent property, coming from the interaction of these systems. Woollacott and Shumway-Cook (1990, p. 800) based their work on the hypothesis that the development of independent stance and movement emerges from the interaction among multiple neural and mechanical components contributing to balance control. Two factors, body alignment and muscle tone, contribute to quiet stance or balance. Three main factors contribute to muscle tone during static balance or quiet stance: a) the inherent stiffness of the muscles themselves, b) the background muscle tone, due to neural contributions, which normally exists in all muscles, and c) postural tone, the activation of antigravity muscles during quiet stance (Shumway-Cook & Woollacott, 2012, p. 167).

In an upright standing position, activity increases in antigravity postural muscles to counteract the force of gravity. This increased activity is called postural tone. Visual, vestibular and somatosensory inputs are critical to postural tone (Shumway-Cook & Woollacott, 2012, p. 168). In the past, many clinicians suggested that postural tone in the trunk was the key component for normal postural control in the upright position.

EMG studies of the muscles' activity in quiet stance, however, found that many muscles in the body are tonically active, including the soleus and gastrocnemius, tibialis anterior, gluteur medius and tensor faciae latae, iliopsoas and the thoracic erector spinae (Shumway-Cook & Woollacott, 2012, p. 170). This suggests that muscles throughout the body are actually tonically active in order to maintain balance. Postural control is therefore a dynamic process, involving active sensory processing with a "constant mapping of perception to action so that the postural system is able to calculate where the body is in space and can predict where it is going and what actions will be necessary to control this movement" (Shumway-Cook & Woollacott, 2012, p. 170). Many mechanisms are active during quiet stance and, because it is characterised by body sway, passive skeletal alignment in cooperation with muscle and postural tone is not enough. Movement strategies are also essential to maintaining stability, even when standing still. One can thus assume that these children, despite poor prone extension and supine flexion, would be able to compensate with the other muscles involved to maintain a quiet stance. The children from the test sample often go barefoot and play outside. This could support the more recent train of thought of seeing control of the body in quiet stance as a multilink pendulum with two coexisting modes of control. These modes can be described as the ankle strategy, where both the leg and trunk segments move in phase when sway frequencies are low, and the hip strategy, where the leg and trunk segments move out of phase when sway frequencies are higher (Shumway-Cook & Woollacott, 2012, p. 170).

q) Postrotary Nystagmus

Lastly, 36.67% (11) of the cases and 13.33% (4) of the controls were recorded in the dysfunctional range for **Postrotary Nystagmus**. Of the cases, eight of the children recorded a short duration of postrotary nystagmus with very low SIPT scores (<-1.0), two recorded prolonged duration with subsequent high scores (>+1.0), and two exhibited abnormal reactions (one fell off while spinning and one refused to get back on after spinning in one direction), which are both interpreted as reactions of a vestibular system that overinhibits incoming information. The four controls in the dysfunctional range all recorded a short duration of nystagmus.

Short duration of postrotary nystagmus has been interpreted as a central nervous system underreactivity to vestibular sensory input and not as a peripheral disorder (inner ear or vestibular nerve) (Ayres, 2004, p. 199). According to Ayres, a short duration of nystagums may be the result of overinhibition of the vestibular nuclei – possibly through connections with the cerebellum (South African Institute for Sensory Integration, 2011, p. 3). Short duration of nystagmus has been found in a number of studies of learning-disabled children (Ayres, 2004, p. 199). In the process of validating the SIPT, a significant positive correlation was found between shortened nystagmus and Design Copying. This may suggest a relationship between short nystagmus and poor visual spatial management. Five of the eight cases with dysfunctional low PRN scores showed this relationship.

Ayres hypothesised that prolonged PRN results from an insufficient amount of inhibition from higher level cortical centres acting on the vestibular nuclei and pontine reticular formation (South African Institute for Sensory Integration, 2011, p. 3). The fact that all the cases with prolonged PRN had a diagnosis of FAS may correlate with research suggesting that prolonged PRN may be associated with more extensive damage, which affects both cortical and sub-cortical functions. According to the literature as described in Chapter 2.2.2, children with FAS are the most severely affected by prenatal alcohol use. High PRN scores are often associated with low Praxis on Verbal Command. Both cases with dysfunctionally high PRN scores also scored dysfunctionally low on Praxis on Verbal Command. Four more cases with borderline PRN scores between +0.8 and +1.0 also had dysfunctionally low scores for Praxis on Verbal Command. According to Ayres (2004, p. 145), this could mean either a primary linguistic or postural problem, but a linguistic interpretation is preferred in the company of an average or high score on Postrotary Nystamus. This reasoning is strengthened by the fact that five of the six cases mentioned received diagnoses of FAS, meaning that they exhibit deficient brain growth/structure of neurophysiology (Hoyme, et al., 2016, p. 9).

Having reviewed all 17 SIPT subtests, it is clear that the performance of the cases was poorer than that of the controls in 15 of the SIPT items. An equal number of children scored within the dysfunctional range on two of the subtests (Localization of Tactile Stimuli and Oral Praxis). There was a significant difference between the cases and controls when comparing the pairs (cases and controls) in 11 of the subtests. From the discussion of the results it seems evident that there are definitely areas of concern regarding the FASD children, but equally concerning is the reality that the controls also experienced problems on some of the test items .

In the next section the possible patterns of dysfunction in the research data will be identified and discussed.

5.6 PATTERNS OF SENSORY INTEGRATION DYSFUNCTION

Numerous research studies over more than six decades have revealed common patterns of sensory integration dysfunctions, as discussed in Chapter 2.4.2.2. A study by Van Jaarsveld, Mailloux, Smith Roley and Raubenheimer (2014, p. 2) confirmed the similarities between patterns of dysfunction in children growing up in South Africa and those already identified in wider research, thus confirming the value of the SIPT in identifying sensory integration dysfunction across cultures. One of the limitations of this study was that it was based on a convenience sample which was not representative of all the children in South Africa, especially those from low to very low socio-economic backgrouds (Van Jaarsveld, Mailloux, Smith Roley, & Raubenheimer, 2014, p. 5). It is noteworthy, however, that the three patterns identified with the SIPT in the research done in 2014 were similar to those identified in a study done by Mailloux et al. in 2011 in the United States. The patterns identified in the South African study will therefore be used in identifying the patterns of dysfunction in the current study.

Knowledge of the patterns of dysfunction helps us to understand the nature of the difficulties interfering with a child's performance, contributes to more precise intervention planning, and offers a means of measuring outcomes of intervention (Mailloux, et al., 2011, p. 150). Schaaf and Mailloux (2015, p. 17) remind us that, although the patterns offer guidance for interpreting data in order to plan treatment, they are not intended to suggest that "assessment data on an individual child will fall clearly into these patterns." The statistical analysis of the matched pairs showed that the control children scored significantly better on the test items, indicating a specific pattern of dysfunction compared to their paired children in the case group. This meant that significantly more of the case children could be identified with one of the three identified patterns of dysfunction.

The researcher, in collaboration with her study leader, therefore decided to consider a possible pattern of dysfunction when 60% or more of the identifying SIPT measures were met. Scores for the individual subtests included in a specific pattern of dysfunction had to be <-0.8, as this was what was used in the process of clinical reasoning on patterns of dysfunction. It should also be mentioned that the identification was done solely on the available SIPT scores and not through a process of clinical reasoning, as is usually done when a child is suspected of having a sensory integration dysfunction. In a clinical setting, much more data from tests and background information are available to assist in making a decision on a relevant pattern of dysfunction. The decisions made in this research on patterns of dysfunctions were made only on the available SIPT scores.

5.6.1 General observations

The analysis of the performance of the children on the SIPT indicated that 16 (53.33%) of the cases and 8 (26.67%) of the controls could fall into one of the patterns of dysfunction. Four of the cases could possibly fall in two, and two of the cases in three different patterns of dysfunction.

Ten of the cases possibly falling in a pattern of dysfunction were diagnosed with FAS, five with ARND, and only one with PFAS. It is interesting to note that 83% of the children in this study with ARND could fall in a pattern of dysfunction, followed by 58.8% of the children with FAS and only 14% of those with PFAS. Cognitive and behavioural checklist results of an in-school study reported by May et al. (2016, p. 211) also found that the children with ARND performed most poorly, followed by those with FAS and then PFAS. The findings of this research thus confirm the findings of the work done by May et al. (2016), although different constructs were measured.

5.6.2 Identified patterns in this study

According to Van Jaarsveld, Mailloux, Smith Roley and Raubenheimer (2014), the best fit for the South African sample was found in a three-factor solution – Visuo- and Somatodyspraxia, Bilateral Integration and Sequencing, and Tactile and Visual Discrimination.

- **Visuo- and Somatodyspraxia**

The analysis showed that fourteen (46.7%) of the cases could fall in the pattern Visuo- and Somatodyspraxia. The matched paired results also indicated a significant difference between the cases and controls (Table 4.13). Similarly to the South African sample of Van Jaarsveld, Mailloux, Smith Roley and Raubenheimer (2014, p. 4), Postural Praxis was the least represented in the pattern. Representations of Space Visualization, Design Copying, Constructional Praxis, Manual Form Perception and Praxis on Verbal Command were consistent with the pattern.

With the main feature of this pattern being the association between visual perception and visual-motor planning, these children may have difficulty with writing, colouring, drawing and following visual models to construct something. Correlations described under Space Visualization and Design Copying in 5.5.2 are also relevant here. Praxis on Verbal Command and Manual Form Perception suggest a mild association with a pattern of Somatodyspraxia. Secondary difficulties of somatodyspraxia can cause these children to struggle with the completion of new or complex motor tasks (Schaaf et al., 2010, p. 124 & 140). The literature indicates that children with FASD have more difficulty as task complexity increases (Roebuck-Spencer, Mattson, Deboard Marion, Brown, & Riley, 2004, p. 536; Doney et al., 2016, p. 606). They may have problems with body boundaries, are often disorganized and struggle with the activities of daily living such as brushing teeth, getting dressed and using eating utensils. Although not specifically tested in this research, the literature indicates that the FASD population often struggle with activities of daily living (White, Mulligan, Merrill, & Wright, 2007, p. 158). Schaaf et al. (2010, p. 141) state that they may have problems with transitions or schedule changes. Both the caregivers and educators agreed in the Sensory Profiles that this was occasionally the case with the FASD children in this study.

- **Bilateral Integration and Sequencing**

Four of the cases could fit into this pattern of dysfunction, as described in Chapter 2.4.2.2. Only two of the six children displaying this pattern of dysfunction had dysfunctionally low PRN scores, as could be expected. Two of the PRN scores were dysfunctionally high and two were within normal limits. PRN did not load within the South African research of Van Jaarsveld, Mailloux, Smith Roley and Raubenheimer (2014, p. 4), probably due to the equalising effect of low and high PRN scores when both are dysfunctional. PRN is the only SIPT item that could have this equalising effect during statistical analysis. All four of the children experienced problems assuming and maintaining the antigravity postures during pre-SIPT clinical observations, which ties in with this pattern of dysfunction.

- **Tactile and Visual Discrimination**

The Tactile and Visual Discrimination pattern of dysfunction as described in Chapter 2.4.2.2 could fit five of the cases. Similarly to the South African research of Van Jaarsveld, Mailloux, Smith Roley and Raubenheimer (2014), dysfuntional scores for Figure-Ground Perception and Finger Identification were present in all the six times this pattern of dysfunction was recorded in this research. Dysfunctional Localization of Tactile Stimuli was only recorded twice.

5.7 SUMMARY

The results of the Sensory Profile Caregiver Questionnaire, the Sensory Profile School Companion, and the SIPT, as presented in Chapter 4, were discussed and compared with relevant literature in this chapter, in order to draw appropriate conclusions about the sensory integration difficulties and dysfunctions of children with FASD from the CWED.

Although further research will be needed before a final conclusion can be reached, the results from the Sensory Profile Caregiver Questionnaire, the Sensory Profile School Compation and the SIPT of this study provide support for reports of sensory processing and integration difficulties and dysfunctions in the FASD population. These difficulties and dysfunctions seem negatively to impact on purposeful and meaningful participation in daily occupations such as activities of daily living, from tying shoelaces and getting dressed, to sleeping and eating, to classroom behaviour, including the ability to focus and follow instructions, and academic performance relating to constructing, writing, reading and mathematics. In all these, the findings from this research are consistent with previous studies.

CHAPTER 6

CONCLUSIONS AND RECOMMENDATIONS

6.1 INTRODUCTION

In the previous chapter, the findings from this study were discussed. Where possible, interpretations were made and the relevance of the existing literature was taken into account. The findings described and compared the sensory integration difficulties and dysfunctions both of children with Fetal Alcohol Spectrum Disorders (FASD) and of matched controls without a diagnosis of FASD, as identified using the Sensory Profile, Sensory Profile School Companion and the Sensory Integration and Praxis Tests. The possible influence of the findings on the occupational performance of the children with FASD involving sleep, school (academic work), behaviour at home and school, motor performance and adaptive skills were also discussed.

Chapter 6 concludes the study. The limitations of the study will be discussed, followed by conclusions and recommendations for future research, with a final closure.

6.2 LIMITATIONS OF THE STUDY

The findings from this study need to be interpreted in the context of the limitations within which it was carried out. In terms of sample size, Gay, Mills, and Airasian (2009, p. 133) suggest a basic rule, that in the case of smaller populations, the entire population should be included in the research sample. The bigger the sample, the more valid will be the findings. This was not possible in this study, since the inclusion- and exclusion criteria and the matching requirements were needed to ensure a homogenous sample. More children could not be selected due to time and financial constraints, as well as the unavailability of SIPT-trained occupational therapists to help with data collection. These factors limited the sample size, despite the fact that more children formally diagnosed both with and without FASD became available on the Wellington side during May 2016. During data collection, distances in the rural areas as well as work responsibilities prevented 20 of the caretakers from attending interviews to complete the SP at the schools (3.9.1). Had more therapists been available, a better transport system, either to fetch the parents at home or to interview them at work, could have been implemented. With only two therapists involved, as well as the financial constraints, this was not possible.

The lack of funding proved to be a further limitation, since the cost of the test material and of remunerating the occupational therapist who assisted with data collection was very high. The NGOs who were approached were very reluctant to sponsor individual research. One NGO, actively involved in early childhood development in the CWED, showed an initial interest and requested the protocol to present to their board, but never contacted the researcher again. An overseas-funded organization supporting and developing schools in the area was approached, but said it was not their policy to fund individual research. Two wine farms, well-known for their development work among the farm workers in the Robertson area, were also approached. One of the wine farms asked for a protocol, but did not take it further. The other wine farm did not respond at all. Although the researcher was employed by the WCED, no scholarship was available from them. While some funding was secured from SAISI, it was not enough to cover the expenses of the study.

The use of the long version of the Sensory Profile as well as the SIPT also limited the sample size. Both are extremely time-consuming, and the SIPT in addition is an expensive test in the SA context. The low educational level of the majority of caregivers (Table 4.2) further added to the time spent in completing the Sensory Profile with each of the caregivers individually. Both the SIPT and SP are well researched, valid occupational therapy tools, giving in-depth information on the sensory processing and practic abilities of children (3.7.1). This reinforced the researcher’s decision to use them instead of shorter screening tools in favour of a bigger sample size. Jirikowic, Olson and Kartin (2008a, p. 133) also recommend the use of comprehensive standardised tests, such as the SIPT, to measure sensory processing abilities.

All the FASD children in this study came from low socio-economic backgrounds. This may also have contributed to their sensory integration difficulties and dysfunctions, since such environments are usually marked by a dearth of stimulation for young children. Howell, Lynch, Platzman, Smith and Coles (2006, p. 124) emphasise the importance of using a socio-economically matched control group when carrying out research on alcohol-affected children. Using a matched sample minimises the possibility that a low socio-economic environment alone is the cause of the difficulties and/or dysfunctions. The fact that socio-economic status was factored in was therefore a strength of this study.

The researcher believes that the average class size of 40 learners in Grade 3 may have influenced the reliability of the information gathered on the Sensory Profile School Companion (3.9.1). It was noticeable that the teachers were unable to answer in-depth questions about each learner, unless a particular child exhibited behaviour demanding individual attention. In general, the educators of classes with less learners were able to give more detailed information about each learner. The SP of one child where the educator scored all the questions a four, was discarded and not analysed as it was deemed invalid.

Performance-based tests such as the SIPT reflect only a limited area of a child's level of overall function. The SIPT assess different practic abilities and aspects of the sensory processing status of different sensory systems. These tests, however, do not assess sensory modulation. The caregiver and educator questionnaires were therefore needed for a more comprehensive measure of behaviour and daily functioning over time, as well as to assess sensory modulation. Questionnaires however are subject to reporting bias, which should be taken into account in the interpretation of the results (3.9.1 and 5.4.1). The use of both performance-type measures as well as caregiver and teacher reports accounted both for these factors and for the belief that children perform differently in different physical and social environments (Jirikowic, 2003, p. 129).

The main limitations of this study were therefore the small sample size due to selection, time and financial constraints, as well as the shortage of SIPT-trained occupational therapists to assist with data collection. The high drop-out rate of the caregivers, as discussed in 3.9.1, was a further factor, and the use of the long version of the SP as well as the SIPT, both time-consuming measuring instruments, limited the sample size even more. The possibility of reporting bias with the SP and SPSC questionnaires, as described in 3.9.1 and 5.4.1, also had to be considered in the interpretation of the results.

6.3 CONCLUSIONS

The aim of this study, as laid out in Chapter 1, was to describe sensory integration difficulties and dysfunctions experienced by a group of children aged five to eight years old with a diagnosis of FASD from the CWED in the Western Cape, using a quantitative observational analytical study design.

The objective was to investigate sensory modulation as measured by the Sensory Profiles and sensory processing and practic abilities as measured by the SIPT of a group of children with FASD and a matched control group without a diagnosis of FASD. The inclusion of the control group, subjected to the same measuring instruments as the group with FASD, provided a baseline against which the effects of FASD on sensory integration difficulties and dysfunctions could be measured (Polit & Beck, 2010, p. 550). Lastly the objective was to identify the patterns of sensory integration difficulties and dysfunctions, to describe these findings and to draw conclusions from them about the unique and distinctive patterns of sensory integration difficulties and dysfunctions among children with FASD. (1.3.2).

The aim of the study having been achieved, the problem of the high prevalence of FASD in the Western Cape, together with the poor results in school performance (as stated in 1.2), can now be addressed. Primarily, this will mean planning and designing occupational therapy-centred intervention strategies to be implemented by both educators and parents/caregivers.

A summary of the conclusions of the study as they relate to the aim and objectives will now be presented.

The results of this research confirm extant reports of sensory processing and integration difficulties and dysfunctions in the FASD population.

In the data from both the Sensory Profile and the Sensory Profile School Companion the caregivers and educators of the children with FASD reported more behavioural indicators of sensory processing differences than did the caregivers and educators of those children not diagnosed with FASD. These sensory processing difficulties seemed to occur in multiple sensory systems ranging from under- to over-responsiveness. This may be due either to a difference in neurological thresholds of children with FASD, as described in the literature of Schneider et al. (2009, p. 5) and discussed in 5.4.1., or to inconsistent responses to sensory input, thereby displaying a tendency of fluctuating responsivity. Higher percentages of cases than controls were recorded in 22 of the 23 categories of the SP, with significant differences in nine. For the SPSC, higher percentages of cases were found in all 13 categories, with significant differences in three. Comparing the mean scores of the pairs of cases and controls revealed significant differences in eight categories of the SP and three categories of the SPSC.

The children with FASD had significantly more difficulties with low registration than their typically developing peers, causing sensation seeking behaviour in the form of excessive movement, especially at home. In the classroom, it more frequently showed as inattention or distractibility. A similar effect was seen with sensory sensitivity, which caused avoiding behaviour, mainly reported in the classroom. These children tend to become quickly overloaded in a noisy, crowded classroom, leading them either to withdraw or to become overstimulated and even more active.

Children with FASD presented with a higher prevalence of fluctuating responsivity relating to the auditory system than their peers not diagnosed with FASD. Auditory processing difficulties and sensory under-responsiveness, as were reported for the FASD population of this study, had previously been associated with academic underachievement (Ashburner, Ziviani, & Rodger, 2008, p. 564).

Noticeable differences between the children with FASD and those not diagnosed on the spectrum were also recorded for Inattention and Distractibility. Fluctuating responsivity in the auditory and visual systems was a major factor in this difficulty.

Fine Motor/Perceptual was a noticeable problem for the FASD population, correlating with complaints about poor handwriting and difficulty with writing sequential sentences.

From a review of the results on the SIPT subtests, it was clear that the performance of the cases was poorer than that of the controls in 15 of the SIPT items. Significant differences were found when comparing the pairs (cases and controls) in 11 of the subtests, with poorer performance from the cases. From the results, it seems evident that there are definite areas of concern (Space Visualization, Figure-Ground, Design Copying, Manual Form Perception, Praxis on Verbal Command, Kinesthesia, Constructional Praxis) for the children with FASD. Five of these tests (Space Visualization, Design Copying, Manual Form Perception, Praxis on Verbal Command, Constructional Praxis) were, as expected, strongly related to the pattern of visuo- and somatodyspraxia. However, a particular concern was the poor performance of the controls on some of the test items (specifically Space Visualization, Figure-Ground, Praxis on Verbal Command). Poor performance on Space Visualisation and Figure Ground has, among other factors, implications for a child's visual perceptual abilities and the development of academic skills such as reading and writing.

The most prominent pattern of sensory integration dysfunction experienced by the children diagnosed with FASD seemed to be visuo- and somatodyspraxia. The main feature of this is the association between visual perception and visual-motor planning. This was clearly observed in the difficulties the cases had with Design Copying and Space Visualization. This was demonstrated in their problems with drawing, writing, following visual models to construct objects, as well as with related academic difficulties, as described in 5.5.2. Secondary difficulties of somatodyspraxia, also described in the literature, were observed in the cases as they struggled with the completion of complex motor tasks, difficulties with body boundaries, as well as with activities of daily living such as tying shoelaces. The mild association of Praxis on Verbal Command with this pattern of dysfunction may partly explain the high incidence of dysfunction with Praxis on Verbal Command. Skills needed for Praxis on Verbal Command, apart from following verbal instructions, include planning movements based on these instructions and using tactile and proprioceptive awareness to motor plan the movements of the body and limbs without a visual model (Bodison & Mailloux, 2006, pp. CE-5). The proprioceptive and tactile systems were therefore also involved (South African Institute for Sensory Integration, 2011, p. 16).

The difficulties and dysfunctions identified by the Sensory Profiles and SIPT could have contributed to challenges experienced in occupations of the children with FASD. This includes difficulties with:

- ADL skills such as tying shoelaces impaired by poor kinesthetic awareness and somatopraxis skills;
- Sleep: disrupted sleep patterns and short duration of sleep associated with sensory processing difficulties such as sensory sensitivity (Table 4.7), behavioural outcomes of sensory processing, modulation related to body position and movement and fine motor perceptual difficulties;

- Education: difficulties with regards to performance components needed for learning and participation in the educational environment exist. Examples are the inability to focus and follow instructions due to fluctuating responsivity in the auditory and vestibular systems and poor academic performance pertaining to constructing, writing, reading and mathematics. All the mentioned academic skills are affected by sensory processing difficulties such as poor visuo- and somatodyspraxia, space visualization challenges and poor figure-ground perception;
- Social participation – poor modulation of sensory input affecting emotional responses can contribute to problems reported by caregivers for example stubbornness, uncooperative behaviour and sensitivity to criticism.

6.4 RECOMMENDATIONS

Given the potential for occupational dysfunction caused by sensory integration difficulties and dysfunctions posed by a diagnosis of FASD, occupational therapists clearly have a role to play in the management of children with FASD, particularly in the school system. It is thus recommended that a follow-up study, designing suitable programmes for educators to address the sensory integration difficulties and dysfunctions in the classroom, be undertaken.

The results of this study will be presented to the WCED for consideration in terms of possible intervention strategies. Occupational therapists working with children with FASD should also take note of the results in terms of their implication for best practices and treatment strategies.

More research is also warranted into the following aspects:

- The results of children in the CWED from low socio-economic environments and not diagnosed with FASD (controls) revealed that a substantial percentage of them also presented with sensory integration difficulties and dysfunctions and thus warrants further investigation. This correlates with findings of Davies, et al. (2011, p. 298) showing that when children remain in poor, disadvantaged socio-economic environments for the first two years of their lives, all developmental domains are compromised as discussed in 2.5.

- More in-depth research on the poor performance of the FASD children on Praxis on Verbal Command.
- An in-depth look into the poor antigravity postures and eye movements of the total population. Antigravity postures forms part of the basis for more complex postural mechanisms (Schaaf, et al., 2010, p. 115). Daily actions such as sitting upright at a table in class, applying postural adjustments and crossing the midline when writing, reading, playing ball games as well as performing sequential activities all require postural-ocular control.
- More research on early regulatory processes among children with prenatal alcohol exposure as discussed in 2.4.3.2 (b). This recommendation is made on the assumption that early identification and intervention can take advantage of neuroplasticity to soften early adversity and help improve early regulatory challenges (Jirikowic, Chen, Nash, Gendler, & Olson, 2016, p. 185).
- Research in the use of the Sensory Profiles on the South African population.

6.5 CLOSURE

The significant differences in sensory processing and integration of the FASD children found in this study and supported by previous findings need consideration as factors contributing to the overall behavioural, cognitive and psychosocial manifestations of FASD (Dunn, 2001, p. 614).

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APPENDIX A: Letter of Approval from Ethics Committee

UNIVERSITY OF THE FREE STATE
UNIVERSITEIT VAN DIE VRYSTAAT
YUNIVESITHI YA FREISTATA

UFS·UV
HEALTH SCIENCES
GESONDHEIDSWETENSKAPPE

IRB nr 00006240
REC Reference nr 230408-011
IORG0005187
FWA00012784

28 August 2015

MS M DU PLOOY
DEPARTMENT OF OCCUPATIONAL THERAPY
FACULTY OF HEALTH SCIENCES
UFS

Dear Ms M Du Plooy

ECUFS NR 137/2015 **DEPARTMENT OF OCCUPATIONAL THERAPY**
PROJECT TITLE: SENSORY INTEGRATION DIFFICULTIES AND DYSFUNCTIONS IN CHILDREN WITH FETAL ALCOHOL SPECTRUM DISORDERS

1. You are hereby kindly informed that the Ethics Committee approved the above project and it will be ratified at the meeting scheduled for 15 September 2015.

2. Any amendment, extension or other modifications to the protocol must be submitted to the Ethics Committee for approval.

3. A progress report should be submitted within one year of approval of long term studies and a final report at completion of both short term and long term studies.

4. Kindly use the ECUFS NR as reference in correspondence to the Ethics Committee Secretariat.

5. The Ethics Committee functions in compliance with, but not limited to, the following documents and guidelines: The SA National Health Act. No. 61 of 2003; Ethics in Health Research: Principles, Structures and Processes (2015); SA GCP(2006); Declaration of Helsinki; The Belmont Report; The US Office of Human Research Protections 45 CFR 461 (for non-exempt research with human participants conducted or supported by the US Department of Health and Human Services- (HHS), 21 CFR 50, 21 CFR 56; CIOMS; ICH-GCP-E6 Sections 1-4; The International Conference on Harmonization and Technical Requirements for Registration of Pharmaceuticals for Human Use (ICH Tripartite), Guidelines of the SA Medicines Control Council as well as Laws and Regulations with regard to the Control of Medicines, Constitution of the Ethics Committee of the Faculty of Health Sciences.

Yours faithfully

....
DR SM LE GRANGE
CHAIR: ETHICS COMMITTEE
Cc: Ms A van Jaarsveld

Ethics Committee
Office of the Dean: Health Sciences
T: +27 (0)51 401 7795/7794 | F: +27 (0)51 444 4359 | E: ethicsfhs@ufs.ac.za
Block D, Dean's Division, Room D104 | P.O. Box/Posbus 339 (Internal Post Box G40) | Bloemfontein 9300 | South Africa
www.ufs.ac.za

APPENDIX B: Study Permission from Prof. Seedat

UNIVERSITEIT•STELLENBOSCH•UNIVERSITY
jou kennisvennoot • your knowledge partner

2 July 2015

Ms. Maritha du Plooy
Cape Winelands Education District
Private Bag X3102
WORCESTER
6849

Dear Ms. Du Plooy

Your Project: "Sensory Integration difficulties and dysfunctions in children with FASD"

I hereby confirm that permission has been granted for you to access children with Fetal Alcohol Spectrum Disorders (FASD) who are recruited through the "Trajectory of FASD across the Lifespan: New Understandings in Interventions" for your project as named above. This will be done in consultation with the FASER-SA study team.

Furthermore, I confirm that your control group will also be selected from the FASER-SA study participants and that you will select the older participants from the "Wellington in School" study.

Please feel free to contact me should you require any further information.

With best wishes

Soraya Seedat
Professor and Executive Head of Department

Fakulteit Geneeskunde en Gesondheidswetenskappe
•
Faculty of Medicine and Health Sciences

Departement Psigiatrie • Department of Psychiatry
Posbus/PO Box 241 • Cape Town/ Kaapstad • 8000 • Suid-Afrika/South Africa
Tel +27 21 938 9227/9116 • Faks/Fax: +27 21 938 9738

APPENDIX C: Permission to conduct research in Public Schools within the Western Cape

Education

Audrey.wyngaard@westerncape.gov.za
tel: +27 021 467 9272
Fax: 0865902282
Private Bag x9114, Cape Town, 8000
wced.wcape.gov.za

REFERENCE: 20150826-2754
ENQUIRIES: Dr A T Wyngaard

Mrs Maritha Du Plooy
PO Box 254
Worcester
6849

Dear Mrs Maritha Du Plooy

RESEARCH PROPOSAL: SENSORY INTEGRATION DIFFICULTIES AND DYSFUNCTIONS IN CHILDREN WITH FETAL ALCHOHOL SPECTRUM

Your application to conduct the above-mentioned research in schools in the Western Cape has been approved subject to the following conditions:

1. Principals, educators and learners are under no obligation to assist you in your investigation.
2. Principals, educators, learners and schools should not be identifiable in any way from the results of the investigation.
3. You make all the arrangements concerning your investigation.
4. Educators' programmes are not to be interrupted.
5. The Study is to be conducted from **28 August 2015 till 24 June 2016**
6. No research can be conducted during the fourth term as schools are preparing and finalizing syllabi for examinations (October to December).
7. Should you wish to extend the period of your survey, please contact Dr A.T Wyngaard at the contact numbers above quoting the reference number?
8. A photocopy of this letter is submitted to the principal where the intended research is to be conducted.
9. Your research will be limited to the list of schools as forwarded to the Western Cape Education Department.
10. A brief summary of the content, findings and recommendations is provided to the Director: Research Services.
11. The Department receives a copy of the completed report/dissertation/thesis addressed to:

    The Director: Research Services
    Western Cape Education Department
    Private Bag X9114
    CAPE TOWN
    8000

We wish you success in your research.

Kind regards.
Signed: Dr Audrey T Wyngaard
Directorate: Research
DATE: 27 August 2015

APPENDIX D: Explanatory Document and Letter of Consent to Director of the Cape Winelands Education District

EXPLANATORY DOCUMENT AND LETTER OF CONSENT TO DIRECTOR OF THE CAPE WINELANDS EDUCATION DISTRICT

TITLE OF RESEARCH STUDY:

Sensory integration difficulties and dysfunctions in children with Fetal Alcohol Spectrum Disorder (FASD).

RESEARCHER:

Mrs. Maritha du Plooy, B. Occupational Therapy, (US)

Dear Mr Benjamin

I, Maritha du Plooy, am an occupational therapist in the Western Cape Education Department (Cape Winelands Education District). The aim of this this study, as part of Master's Degree in Occupational Therapy at the University of the Free State, is to investigate the sensory integration difficulties and dysfunctions of children with FASD in our district. The learners taking part in the study will be selected from two bigger research studies conducted by Prof. Philip A. May from the University of North Carolina and Proff. Charles D.H. Parry and Soraya Seedat both from the University of Stellenbosch, in the Wellington, Robertson, Ashton, Montagu and Bonnievale areas.

This study has been approved by the Ethics Committee of the Faculty of Health Sciences, University of the Free State as well as the Western Cape Education Department.

FASD, sensory integration and children:

The effects of the brain damage associated with prenatal alcohol exposure are devastating and enduring and include cognitive-, motor-, executive function-, language-, visuo-spatial-, learning-, social skills-, sensory processing- and attention deficits. Cognitive and behavioural deficits associated with FASD have been extensively covered in the literature. Difficulty processing and integrating sensory information, on the other hand, is one area of neurobehavioral functioning that has not been adequately documented in the research of children with FASD although these difficulties may result in challenges meeting appropriate developmental expectations and achieving success at home, in the classroom and in the community.

Many of the symptoms of sensory integration difficulties such as clumsiness, inattention and distractibility, emotional reactivity, learning problems and sensory sensitivity have also been consistently reported in children with FASD. The fact that research has indicated a FASD prevalence of up to 20%, in certain communities in our district and which is among the highest in the world, poses a real challenge not only to our children, but also to our educators who are working with them on a daily basis.

These children often exhibit behavioural problems, battle to focus and concentrate and generally progress slower than their peers. Keeping in mind that children with FASD may be affected in almost every area of functioning, it is clear that the educators working with these children are faced with unique challenges. Even after the children are diagnosed, the educators may not have the tools or information to correctly identify the cause of the cognitive or behaviour problems or know about intervention strategies to help them.

With the already mentioned high prevalence of FASD, poor academic performance of the children in our district as reflected in die Western Cape Education Department's Systemic Test Results and the significant difficulties with processing and integrating sensory information into age-appropriate postural responses and visuo-motor integration in the grade R pre-screenings by the occupational therapists, this study will be an effort to start investigating learners with FASD's neurobehavioral functions dependant on sensory integration. The results of this research will be utilised to plan and design intervention strategies for educators to help these children reach their full potential.

Course of the study:

A total of thirty children with FASD and 30 typically developing children, aged five to eight years, will be randomly selected from the studies already mentioned. They will be tested by occupational therapists qualified to administer and interpret the Sensory Integration and Praxis Tests. The children will perceive the tests as games and the risk involved of experiencing discomfort, is far outweighed by the eventual benefits of the study. The parents/caregivers of all the children, as well as their respective teachers will also be invited to complete a four page standardised questionnaire on the child's sensory preferences and behaviour. This will be done with the purpose of collecting information on how the children's sensory environments (home and school) influence their behaviour. A time that will be convenient for both the teacher and the parent to complete the questionnaire will be individually arranged with them.

The tests will be administered in the morning at the child's school and participants will miss three hours of one school day. This, as well as the availability of a quiet venue where parent questionnaire can be conducted and the tests can be administered without interruption, will be arranged with the school principal and educators beforehand.

Ethical considerations:

Informed consent and assent will be obtained from the participating parents and children respectively before the commencement of the testing. All information will be handled confidentially and neither the school, nor the child will be identified in any report or presentation. Participants and schools may withdraw from the study at any time without the fear of being discriminated against.

A copy of the research report will be made available to the Department of Education on completion of the study.

If you have any further questions regarding the research study, you are welcome to contact any of the following persons:

Contact details:

Study leader: Mrs. Annamarie van Jaarsveld 9h00 – 16h00 Tel: 051-4012829
Researcher: Mrs. Maritha du Plooy 9h00 – 16h00 Tel: 023-3484670
Ethics Committee, Faculty of Health Sciences, University of the Free State
9h00– 16h00 Tel: 051-4017794

Thank you for your time and co-operation.

Yours sincerely

Maritha du Plooy
Occupational Therapist

CONSENT FORM

I hereby give my permission that the study, *Sensory Integration Difficulties and Dysfunctions in Children with Fetal Alcohol Spectrum Disorder,* may be conducted in selected schools in the CWED.

I have read the information document and understand that.

- participation of the schools are voluntary and that they may withdraw from the study at any time if they wish to,
- specific children with FASD and typically developing children will be invited to take part in the study,
- only children where both the parent/caregiver and children have given consent and assent will take part in the study,
- all information gathered will be regarded as highly confidential,
- participants may withdraw from the study at any time without being discriminated against and
- a copy of the research findings will be made available to the Cape Winelands Education District Office.

T. Bro[illegible] ______ Director

______ Signature

31/8/15 ______ Date

APPENDIX E (i): Explanatory Document and Letter of Consent to Parents/Caregivers of Children with FASD

INLIGTINGSDOKUMENT EN INGELIGTE TOESTEMMINGSVORM AAN OUERS

Navorsingsdeelnemer Identifikasienommer: ________________________________

TITEL VAN NAVORSINGSTUDIE:

Sensoriese Integrasie Disfunksies en Hindernisse in kinders met Fetale Alkohol Spektrum Afwykings.

NAVORSER:

Maritha du Plooy, B. Arbeidsterapie, Universiteit van Stellenbosch

Geagte Ouer/Versorger,

Ek, Maritha du Plooy, is 'n arbeidsterapeut by die Wes-Kaapse Onderwysdepartement se Wynland Onderwysdistrik. Die doel van hierdie studie, as deel van 'n magister graad in Arbeidsterapie aan die Universiteit van die Vrystaat, is om die sensoriese integrasie disfunksies en hindernisse van jong kinders met Fetale Alkohol Spektrum Afwykings (FASA) in ons omgewing te ondersoek. Die kinders wat aan die studie sal deelneem was en is almal deel van groter navorsingstudies deur Proff. May, Parry en Seedat waarvan u kind ook deel was/is. Ons neem die vrymoedigheid om u vir hierdie voorgenome studie te kontak, aangesien u tydens die aanvanklike navorsing aangedui het dat u met die oog op verdere navorsing gekontak mag word.

Hierdie brief en toestemmingsvorm is om u in te lig oor die studie waarby ons graag u kind wil betrek. Hierdie studie is goedgekeur deur die Etiekkomitee van die Fakulteit Gesondheidswetenskappe van die Universiteit van die Vrystaat.

Wanneer ‘n swanger moeder alkohol drink, plaas sy haar ongebore baba in gevaar. Die groep afwykings wat veroorsaak word deur die moeder se alkoholgebruik tydens swangerskap, staan bekend as FASA. Mense met FASA ondervind probleme met sekere funksies en sensoriese integrasie disfunksie is 'n voorbeeld hiervan.

Sensoriese integrasie is die proses waardeur die brein inligting vanaf die sintuie ontvang en organiseer vir gebruik. Wanneer hierdie proses nie glad verloop nie, kan dit lei tot verskeie leer-, gedrags- en funksionele probleme. Voorbeelde hiervan is:

- swak beheer oor gedrag (hiperaktiwiteit, onttrekking van ander mense, emosionele onstabiliteit),
- probleme met aandag en konsentrasie,
- probleme met die beplanning en uitvoering van growwe en fyn motoriese aksies bv. lompheid en handskrifprobleme
- leerprobleme bv. leesprobleme en
- sensitiwiteit vir sekere sensasies bv. druk ore toe vir harde geluide.

Hierdie probleme verhinder die kind om optimaal te ontwikkel en om sukses by die huis, in die skool en in die gemeenskap te ervaar.

Baie van die probleme soos hierbo beskryf word deur kinders met FASA ervaar en indien ons deur middel van hierdie studie op 'n Suid-Afrikaanse populasie die voorkoms van sensoriese integrasie disfunksies en hindernisse kan identifiseer en ontleed, kan daar 'n doelgerigte intervensieprogram ontwikkel word waaraan die kind in die klas blootgestel kan word. Die potensiële doelwitte van so ‘n program is die volgende:

- Om die kinders te help om hulle gedrag en emosies beter te reguleer om sodoende makliker aan te pas in multisensoriese omgewings soos die skool.
- Om die kinders te help om beter met ander op skool en by die huis oor die weg kom.
- Om beter in die klas kan konsentreer en op take te fokus.

So ‘n doelgerigte program het ook die potensiaal om 'n positiewe effek op probleme soos lompheid, handskrif-, en leesprobleme te hê.

Die studie behels dat u onder andere’n bietjie meer oor u kind moet vertel deur 'n vraelys oor u kind se sensoriese voor- en afkeure en gedrag in te vul. U sal hulp ontvang om die vraelys in te vul. As bewys van waardering vir u samewerking sal u gesin 'n geskenkpakkie met skoolbenodigdhede vir u kind ontvang.

U kind gaan ook deelneem aan gestandaardiseerde ontwikkelingstoetse wat in dieselfde volgorde met al die kinders wat aan die studie deelneem, gedoen sal word. Dit gaan afgeneem word in een sessie van ongeveer drie ure by u kind se skool. Die sessie sal ‘n 15 minute pouse na elke 45 minute hê, waartydens u kind kan rus en iets kan eet en drink wat verskaf sal word. Die toetse gaan afgeneem word deur arbeidsterapeute wat spesifiek opgelei is in die korrekte gebruik daarvan. Dit behels dat u kind 'n verskeidenheid motoriese en perseptuele take moet doen wat vir hom/haar soos speletjies sal voel en geen gevaar vir beserings vir u kind inhou nie. Indien u kind ongemak tydens enige van die toetse ervaar, mag hy/sy dit dadelik aan die terapeut sê. Sommige kinders kan byvoorbeeld effens naar en dronk voel tydens die toets waar hulle in die rondte gedraai word. Indien dit gebeur, sal die toets dadelik gestop word en die terapeut sal spesifieke tegnieke toepas wat die reaksie sal omkeer.

Elke kind se opvoeder gaan ook gevra word om 'n vraelys oor u kind se sensoriese voor- en afkeure by die skool asook sy gedrag en hoe hy/sy met die maats oor die weg kom, in te vul. Dit gaan gedoen word omdat kinders se sensoriese omgewings by die huis en by die skool verskil en hulle ook dikwels verskillend optree in verskillende sensoriese omgewings.

Alle inligting wat gedurende die navorsing gebruik word, sal vertroulik hanteer word en sal in ‘n toegesluite kabinet gehou word deur die navorser. Daar sal slegs ‘n kontrolenommer gebruik word om inligting vir die navorsingsdoeleindes te identifiseer. U of u kind se naam sal nêrens met die navorsingsresultate verbind word of in publikasies wat oor die navorsing sal verskyn, gebruik word nie. As gevolg van hierdie stappe wat ons sal neem om u privaatheid te beskerm, is die risiko vir die verlies aan vertroulikheid, minimaal. Net soos in die geval van die vorige studie waaraan u kind deelgeneem het, gaan hier ook ‘n kontrolegroep kinders sonder FASA wees. U kind met FASA sal dus nie deur deelname aan die studie identifiseer kan word nie.

U en u kind se deelname aan die navorsingstudie is heeltemal vrywillig en die kinders hoef nie deel te neem as hulle nie wil nie. Hulle kan ook op enige tyd gedurende die studie onttrek sonder enige negatiewe gevolge.

Indien u en u kind instem om aan die studie deel te neem, sal dit u steeds vrystaan om op enige stadium daarvan te onttrek sonder enige gevolge. U moet slegs die navorser betrokke, sowel as die kind se opvoeder of skoolhoof daarvan in kennis stel. U inligting sal dan van die navorsingsprojek onttrek word. U kind het ook die reg om 'n antwoord op, of deelname aan 'n vraag of aspek van die navorsingsproses wat u of u kind as ontoepaslik, beledigend of as skending van sy/haar regte beskou, te weier. Dit staan ook die navorser vry om u kind van die studie te onttrek indien sy voel dat dit in die beste belang van die kind is. U het die reg om te eniger tyd vrae te vra oor die studie.

Deur deel te neem aan die studie onvang u kind dus ‘n gratis gespesialiseerde ondersoek deur 'n arbeidsterapeut. Die terapeut sal na afloop van die toetse met u ‘n afspraak maak om aan u teruvoer gee oor hoe u kind gevaar. Dit kan baie leersaam vir u wees. Indien u kind wel sensoriese disfunksies/uitdagings ervaar, sal hy/sy kan baat by die intervensieprogram wat saamgestel gaan word na aanleiding van die resultate van die navorsing. Hierdie program sal in die klasse geïmplementeer word sodat u kind, sowel as die res van kinders, daarby kan baat.

Indien u enige navrae of bekommernisse het aangaande hierdie versoek om aan die navorsingstudie deel te neem, mag u die navorser, die studieleier of die Etiekkomitee van die Fakulteit Gesondheidswetenskappe, Universiteit van die Vrystaat, enige tyd gedurende die studie kontak:

Kontakbesonderhede:

Studieleier: Mev. Annamarie van Jaarsveld 9h00 – 16h00 Tel: 051-4012829
Navorser: Mev. Maritha du Plooy 9h00 – 16h00 Tel: 023-3484670
Etiekkomitee, Fakulteit Gesondheidswetenskappe, Universiteit van die Vrystaat
9h00 – 16h00 Tel: 051-4017794

U kan ook skryf aan die navorser by:
Posbus 254
Worcester
6849

U handtekening onderaan beteken:

- dat u besluit het om u kind te laat deelneem,
- dat u die inligting hierby gelees het (of dat dit aan u voorgelees is) en
- dat u 'n afskrif van hierdie toestemmingsvorm ontvang het om te hou.

Indien u enige vrae oor u regte as ouer/versorger of oor die navorsingsonderwerp het, mag u ook die Sekretariaat van die Etiese Komitee van die Fakulteit van Gesondheidswetenskappe, UVS skakel by (051) 405 2812.

Baie dankie vir u tyd.

Vriendelik die uwe,

Maritha du Plooy
Arbeidsterapeut

TOESTEMMINGSVORM

Deur hierdie dokument te onderteken, gee ek, ..., my toestemming dat my kind ...., aan die studie mag deelneem.

Geboortedatum van die kind: ____

Adres van die kind: ____

Naam en van van Ouer/versorger:

____

Ek verklaar dat:

- Ek die inligting en toestemmingsvorm gelees het of dat dit aan my voorgelees is in 'n taal wat ek vlot kan praat en goed verstaan.
- Ek verstaan dat die resultate gebruik gaan word vir navorsingsdoeleindes en dat alle inligting vertroulik gehou sal word.

- Ek verstaan dat die kennis wat deur die navorsing verkry gaan word, tot voordeel van my kind en ander kinders met FASA kan lei,
- Ek die geleentheid gehad het om vrae te vra en dat al my vrae bevredigend beantwoord is.
- Ek verstaan dat my kind se deelname aan die navorsingstudie vrywillig is en dat nie onder enige druk geplaas is om toestemming te gee nie.
- Ek my kind te eniger tyd uit die studie kan onttrek sonder om gepenaliseer te word of teen gediskrimineer te word op enige manier.
- Ek gevra mag word om my kind uit die studie te onttrek voordat dit voltooi is indien die navorser voel dat dit in die beste belang van my kind is of indien ek of my kind nie die navorsingsprosedure soos ooreengekom, nakom nie.

Geteken te .... op .... 2015.

....	....
Handtekening van ouer/voog/versorger	Handtekening van getuie

Kontak vir Toekomstige Studies

Ons vra graag u toestemming om u moontlik in die toekoms te kontak omtrent ander studies op die gebied van kinderontwikkeling en –gestremdhede, wat moontlik uitgevoer mag word. Deur vandag aan die huidige studie deel te neem, word u op geen manier verplig om deel te neem aan toekomstige studies nie.

Kies asseblief SLEGS EEN van die volgende opsies:

_______ JA, ek sal in die toekoms gekontak wil word.

Handtekening van ouer/voog: ________________________________

_______NEE, ek sal nie in die toekoms gekontak wil word nie.

Handtekening van ouer/voog/versorger: ________________________________

Gebruik van Data vir Toekomstige Studies

Ons vra graag u toestemming om u kind se data vir toekomstige studies te gebruik. Enige toekomstige studies sal gedoen word in die veld van kinderontwikkeling en -gestremdhede en moet goedgekeur word deur 'n Etiese Komitee. Die Etiese Komitee is 'n groep mense wat navorsingstudies beoordeel om die regte en welstand van navorsingsdeelnemers te beskerm. Die Etiese Komitee sal bepaal of die navorsing volgens erkende standaarde uitgevoer word. Hulle sal ook bepaal of dit vir ons nodig sal wees om u te kontak om vir toestemming om gebruik te maak van u gestoorde data vir spesifieke toekomstige navorsingstudies. Om u te beskerm, sal die data slegs gemerk word met 'n navorsingsdeelnemer-identifikasienommer (navorsingsnommer). Ons sal u navorsingsnommer, wat verbind kan word aan u naam, in ons plaaslike rekords bewaar (toegesluit en veilig). Indien u sou besluit om die gestoorde data te laat vernietig terwyl ons hierdie verbintenis tussen u navorsingsnommer en naam het, kan u ons laat weet en dan sal ons die inligting, wat gemerk is met u navorsingsnommer, laat vernietig. Die verbintenis sal teen die einde van die studie vernietig word. Sodra die verbintenis vernietig is, sal die data, wat nie identifiseerbaar is nie, vir 'n onbepaalde tyd gestoor word. U kind se naam of inligting sal nie ingesluit word in enige publikasies nie.

Kies asseblief SLEGS EEN van die volgende opsies:

_______ JA, ek gee toestemming dat my kind se data vir toekomstige navorsing gebruik mag word.

Handtekening van ouer/voog/versorger: ______________________________

_______ NEE, ek gee nie toestemming dat my kind se data vir toekomstige navorsing gebruik mag word nie.

Handtekening van ouer/voog/versorger: ______________________________

APPENDIX E (ii): Explanatory document and letter of consent to Parents/Caregivers of Children in Control Group

INLIGTINGSDOKUMENT EN INGELIGTE TOESTEMMINGSVORM AAN OUERS

Navorsingsdeelnemer Identifikasienommer: ________________________________

TITEL VAN NAVORSINGSTUDIE:

Sensoriese Integrasie Disfunksies en Hindernisse in kinders met Fetale Alkohol Spektrum Afwykings.

NAVORSER:

Maritha du Plooy, B. Arbeidsterapie, Universiteit van Stellenbosch

Geagte Ouer/Versorger,

Ek, Maritha du Plooy, is 'n arbeidsterapeut by die Wes-Kaapse Onderwysdepartement se Wynland Onderwysdistrik. Die doel van hierdie studie, as deel van 'n magister graad in Arbeidsterapie aan die Universiteit van die Vrystaat, is om die sensoriese integrasie disfunksies en hindernisse van jong kinders met Fetale Alkohol Spektrum Afwykings (FASA) in ons omgewing te ondersoek. Die kinders wat aan die studie sal deelneem was en is almal deel van groter navorsingstudies deur Proff. May, Parry en Seedat waarvan u kind ook deel was/is. Ons neem die vrymoedigheid om u vir hierdie voorgenome studie te kontak, aangesien u tydens die aanvanklike navorsing aangedui het dat u met die oog op verdere navorsing gekontak mag word.

Hierdie brief en toestemmingsvorm is om u in te lig oor die studie waarby ons graag u kind wil betrek. Hierdie studie is goedgekeur deur die Etiekkomitee van die Fakulteit Gesondheidswetenskappe van die Universiteit van die Vrystaat.

Wanneer ‘n swanger moeder alkohol drink, plaas sy haar ongebore baba in gevaar. Die groep afwykings wat veroorsaak word deur die moeder se alkoholgebruik tydens swangerskap, staan bekend as FASA. Mense met FASA ondervind probleme met sekere funksies en sensoriese integrasie disfunksie is 'n voorbeeld hiervan.

Sensoriese integrasie is die proses waardeur die brein inligting vanaf die sintuie ontvang en organiseer vir gebruik. Wanneer hierdie proses nie glad verloop nie, kan dit lei tot verskeie leer-, gedrags- en funksionele probleme. Voorbeelde hiervan is:

- swak beheer oor gedrag (hiperaktiwiteit, onttrekking van ander mense, emosionele onstabiliteit),
- probleme met aandag en konsentrasie,
- probleme met die beplanning en uitvoering van growwe en fyn motoriese aksies bv. lompheid en handskrifprobleme
- leerprobleme bv. leesprobleme en
- sensitiwiteit vir sekere sensasies bv. druk ore toe vir harde geluide.

Hierdie probleme verhinder die kind om optimaal te ontwikkel en om sukses by die huis, in die skool en in die gemeenskap te ervaar.

Baie van die probleme soos hierbo beskryf word deur kinders met FASA ervaar en indien ons deur middel van hierdie studie op 'n Suid-Afrikaanse populasie die voorkoms van sensoriese integrasie disfunksies en hindernisse kan identifiseer en ontleed, kan daar 'n doelgerigte intervensieprogram ontwikkel word waaraan die kind in die klas blootgestel kan word. Die potensiële doelwitte van so ‘n program is die volgende:

- Om die kinders te help om hulle gedrag en emosies beter te reguleer om sodoende makliker aan te pas in multisensoriese omgewings soos die skool.
- Om die kinders te help om beter met ander op skool en by die huis oor die weg kom.
- Om beter in die klas kan konsentreer en op take te fokus.

So ‘n doelgerigte program het ook die potensiaal om 'n positiewe effek op probleme soos lompheid, handskrif-, en leesprobleme te hê.

Dit het uit die studie waaraan u kind voorheen deelgeneem het geblyk dat hy/sy nie voor geboorte deur alkohol geaffekteer is nie. Ons het vir hierdie studie ook kinders sonder FASA nodig sodat ons ‘n duidelike prentjie kan kry van die kinders wat wel voor geboorte deur alkohol geaffekteer is.

Die studie behels dat u onder andere’n bietjie meer oor u kind moet vertel deur 'n vraelys oor u kind se sensoriese voor- en afkeure en gedrag in te vul. U sal hulp ontvang om die vraelys in te vul. As bewys van waardering vir u samewerking sal u gesin 'n geskenkpakkie met skoolbenodigdhede vir u kind ontvang.

U kind gaan ook deelneem aan gestandaardiseerde ontwikkelingstoetse wat in dieselfde volgorde met al die kinders wat aan die studie deelneem, gedoen sal word. Dit gaan afgeneem word in een sessie van ongeveer drie ure by u kind se skool. Die sessie sal ‘n 15 minute pouse na elke 45 minute hê, waartydens u kind kan rus en iets kan eet en drink wat verskaf sal word. Die toetse gaan afgeneem word deur arbeidsterapeute wat spesifiek opgelei is in die korrekte gebruik daarvan. Dit behels dat u kind 'n verskeidenheid motoriese en perseptuele take moet doen wat vir hom/haar soos speletjies sal voel en geen gevaar vir beserings vir u kind inhou nie.

Indien u kind ongemak tydens enige van die toetse ervaar, mag hy/sy dit dadelik aan die terapeut sê. Sommige kinders kan byvoorbeeld effens naar en dronk voel tydens die toets waar hulle in die rondte gedraai word. Indien dit gebeur, sal die toets dadelik gestop word en die terapeut sal spesifieke tegnieke toepas wat die reaksie sal omkeer.

Elke kind se opvoeder gaan ook gevra word om 'n vraelys oor u kind se sensoriese voor- en afkeure by die skool asook sy gedrag en hoe hy/sy met die maats oor die weg kom, in te vul. Dit gaan gedoen word omdat kinders se sensoriese omgewings by die huis en by die skool verskil en hulle ook dikwels verskillend optree in verskillende sensoriese omgewings.

Alle inligting wat gedurende die navorsing gebruik word, sal vertroulik hanteer word en sal in 'n toegesluite kabinet gehou word deur die navorser. Daar sal slegs 'n kontrolenommer gebruik word om inligting vir die navorsingsdoeleindes te identifiseer. U of u kind se naam sal nêrens met die navorsingsresultate verbind word of in publikasies wat oor die navorsing sal verskyn, gebruik word nie. As gevolg van hierdie stappe wat ons sal neem om u privaatheid te beskerm, is die risiko vir die verlies aan vertroulikheid, minimaal.

U en u kind se deelname aan die navorsingstudie is heeltemal vrywillig en die kinders hoef nie deel te neem as hulle nie wil nie. Hulle kan ook op enige tyd gedurende die studie onttrek sonder enige negatiewe gevolge.

Indien u en u kind instem om aan die studie deel te neem, sal dit u steeds vrystaan om op enige stadium daarvan te onttrek sonder enige gevolge. U moet slegs die navorser betrokke, sowel as die kind se opvoeder of skoolhoof daarvan in kennis stel. U inligting sal dan van die navorsingsprojek onttrek word. U kind het ook die reg om 'n antwoord op, of deelname aan 'n vraag of aspek van die navorsingsproses wat u of u kind as ontoepaslik, beledigend of as skending van sy/haar regte beskou, te weier. Dit staan ook die navorser vry om u kind van die studie te onttrek indien sy voel dat dit in die beste belang van die kind is. U het die reg om te eniger tyd vrae te vra oor die studie.

Deur deel te neem aan die studie onvang u kind dus 'n gratis gespesialiseerde ondersoek deur 'n arbeidsterapeut. Die terapeut sal na afloop van die toetse met u 'n afspraak maak om aan u teruvoer gee oor hoe u kind gevaar. Dit kan baie leersaam vir u wees. Indien u kind wel sensoriese disfunksies/uitdagings ervaar, sal hy/sy kan baat by die intervensieprogram wat saamgestel gaan word na aanleiding van die resultate van die navorsing. Hierdie program sal in die klasse geïmplementeer word sodat u kind, sowel as die res van kinders, daarby kan baat.

Indien u enige navrae of bekommernisse het aangaande hierdie versoek om aan die navorsingstudie deel te neem, mag u die navorser, die studieleier of die Etiekkomitee van die Fakulteit Gesondheidswetenskappe, Universiteit van die Vrystaat, enige tyd gedurende die studie kontak:

Kontakbesonderhede:

Studieleier: Mev. Annamarie van Jaarsveld 9h00 – 16h00 Tel: 051-4012829
Navorser: Mev. Maritha du Plooy 9h00 – 16h00 Tel: 023-3484670
Etiekkomitee, Fakulteit Gesondheidswetenskappe, Universiteit van die Vrystaat
9h00 – 16h00 Tel: 051-4017794

U kan ook skryf aan die navorser by:
Posbus 254
Worcester
6849

U handtekening onderaan beteken:

- dat u besluit het om u kind te laat deelneem,
- dat u die inligting hierby gelees het (of dat dit aan u voorgelees is) en
- dat u ‘n afskrif van hierdie toestemmingsvorm ontvang het om te hou.

Indien u enige vrae oor u regte as ouer/versorger of oor die navorsingsonderwerp het, mag u ook die Sekretariaat van die Etiese Komitee van die Fakulteit van Gesondheidswetenskappe, UVS skakel by (051) 405 2812.

Baie dankie vir u tyd.

Vriendelik die uwe,

Maritha du Plooy
Arbeidsterapeut

TOESTEMMINGSVORM

Deur hierdie dokument te onderteken, gee ek, ..., my toestemming dat my kind ...., aan die studie mag deelneem.

Geboortedatum van die kind: ____

Adres van die kind: ____

Naam en van van Ouer/versorger:

____

Ek verklaar dat:

- Ek die inligting en toestemmingsvorm gelees het of dat dit aan my voorgelees is in 'n taal wat ek vlot kan praat en goed verstaan.
- Ek verstaan dat die resultate gebruik gaan word vir navorsingsdoeleindes en dat alle inligting vertroulik gehou sal word.
- Ek verstaan dat die kennis wat deur die navorsing verkry gaan word, tot voordeel van my kind en ander kinders met FASA kan lei,
- Ek die geleentheid gehad het om vrae te vra en dat al my vrae bevredigend beantwoord is.
- Ek verstaan dat my kind se deelname aan die navorsingstudie vrywillig is en dat nie onder enige druk geplaas is om toestemming te gee nie.
- Ek my kind te eniger tyd uit die studie kan onttrek sonder om gepenaliseer te word of teen gediskrimineer te word op enige manier.
- Ek gevra mag word om my kind uit die studie te onttrek voordat dit voltooi is indien die navorser voel dat dit in die beste belang van my kind is of indien ek of my kind nie die navorsingsprosedure soos ooreengekom, nakom nie.

Geteken te .... op .... 2015.

....	....
Handtekening van ouer/voog/versorger	Handtekening van getuie

Kontak vir Toekomstige Studies

Ons vra graag u toestemming om u moontlik in die toekoms te kontak omtrent ander studies op die gebied van kinderontwikkeling en –gestremdhede, wat moontlik uitgevoer mag word. Deur vandag aan die huidige studie deel te neem, word u op geen manier verplig om deel te neem aan toekomstige studies nie.

Kies asseblief SLEGS EEN van die volgende opsies:

_______ JA, ek sal in die toekoms gekontak wil word.

Handtekening van ouer/voog: ______________________________

_______NEE, ek sal nie in die toekoms gekontak wil word nie.

Handtekening van ouer/voog/versorger: ____________________________

Gebruik van Data vir Toekomstige Studies

Ons vra graag u toestemming om u kind se data vir toekomstige studies te gebruik. Enige toekomstige studies sal gedoen word in die veld van kinderontwikkeling en -gestremdhede en moet goedgekeur word deur ‘n Etiese Komitee. Die Etiese Komitee is ‘n groep mense wat navorsingstudies beoordeel om die regte en welstand van navorsingsdeelnemers te beskerm. Die Etiese Komitee sal bepaal of die navorsing volgens erkende standaarde uitgevoer word. Hulle sal ook bepaal of dit vir ons nodig sal wees om u te kontak om vir toestemming om gebruik te maak van u gestoorde data vir spesifieke toekomstige navorsingstudies. Om u te beskerm, sal die data slegs gemerk word met ‘n navorsingsdeelnemer-identifikasienommer (navorsingsnommer). Ons sal u navorsingsnommer, wat verbind kan word aan u naam, in ons plaaslike rekords bewaar (toegesluit en veilig). Indien u sou besluit om die gestoorde data te laat vernietig terwyl ons hierdie verbintenis tussen u navorsingsnommer en naam het, kan u ons laat weet en dan sal ons die inligting, wat gemerk is met u navorsingsnommer, laat vernietig. Die verbintenis sal teen die einde van die studie vernietig word. Sodra die verbintenis vernietig is, sal die data, wat nie identifiseerbaar is nie, vir ‘n onbepaalde tyd gestoor word. U kind se naam of inligting sal nie ingesluit word in enige publikasies nie.

Kies asseblief SLEGS EEN van die volgende opsies:

_______ JA, ek gee toestemming dat my kind se data vir toekomstige navorsing gebruik mag word.

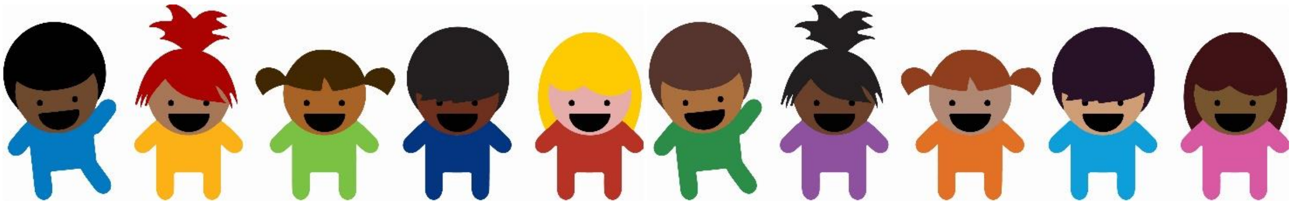
Handtekening van ouer/voog/versorger: ______________________________

_______ NEE, ek gee nie toestemming dat my kind se data vir toekomstige navorsing gebruik mag word nie.

Handtekening van ouer/voog/versorger: ______________________________

APPENDIX F (i): Explanatory Document and Letter of Assent of Participant

INLIGTINGSPAMFLET EN INSTEMMINGSVORM AAN DEELNEMERS



Navorsingsdeelnemer Identifikasienommer: ________________________________

TITEL VAN DIE NAVORSINGSTUDIE:

Sensoriese Integrasie Disfunksies en Hindernisse in kinders met Fetale Alkohol Spektrum Afwykings (FASA).

NAAM VAN NAVORSER:

Mev. Maritha du Plooy, B. Arbeidsterapie, (Universiteit van Stellenbosch)

Wat is NAVORSING?

Navorsing is iets wat ons doen om meer oor mense en hoe dinge werk uit te vind. Ons gebruik navorsingsprojekte of studies om ons te help om meer oor mense wat anders is of probleme en siektes het uit te vind. Navorsing help ons ook om beter maniere te vind om kinders met probleme of wat siek is, te help en te behandel sodat hulle ook na die beste van hulle vermoë kan presteer.

Waaroor gaan hierdie navorsingstudie?

Wanneer ‘n swanger moeder alkohol drink, plaas sy haar ongebore baba in gevaar. FASA is ‘n groep groei-, brein- en fisiese probleme wat mag voorkom in ‘n baba wanneer die moeder **alkohol drink tydens haar swangerskap.** Hierdie navorsingstudie word gedoen om te kyk hoe kinders wat deur alkohol geaffekteer is voordat hulle gebore is, se breine inligting vanaf die sintuie soos die ore en oë ontvang en organiseer vir gebruik. Dit is belangrik om te weet omdat dit hulle manier van dink en leer asook hulle gedrag beïnvloed.

Hoekom is ek uitgenooi om aan hierdie navorsingsprojek deel te neem?

Jy word gevra om aan hierdie studie deel te neem omdat jy in Graad R of 2 in jou skool is en voorheen aan die studie deelgeneem het waar jy gemeet is en die dokter na jou gesig, ore en hande gekyk het. Jy het dalk ook 'n paar kort toetsies saam met die sielkundige gedoen. Jou ouers het ingestem dat jy weer gevra mag word om aan ‘n studie deel te neem.

Wie doen hierdie navorsing?
Mev. Maritha du Plooy, 'n nagraadse student van die Fakulteit Gesondheidswetenskappe, Universiteit van die Vrystaat, doen hierdie studie.

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Jy kan ook aan die navorser skryf by die volgende adres:

Posbus 254
Worcester
6849

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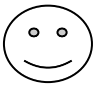
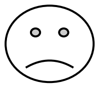
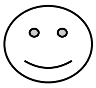
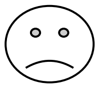
Jy hoef nie aan die studie deel te neem nie. As jy eers besluit het om deel te neem kan jy jou besluit enige tyd verander. Die arbeidsterapeute wat met die studie help, sal nie ontsteld of kwaad wees as jy van besluit verander nie.

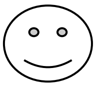
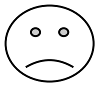
Verstaan jy hierdie navorsingstudie en sal jy daaraan deelneem?

Het die tannie al jou vrae beantwoord?

Verstaan jy dat jy enige tyd mag besluit om nie meer aan die studie deel te neem nie?

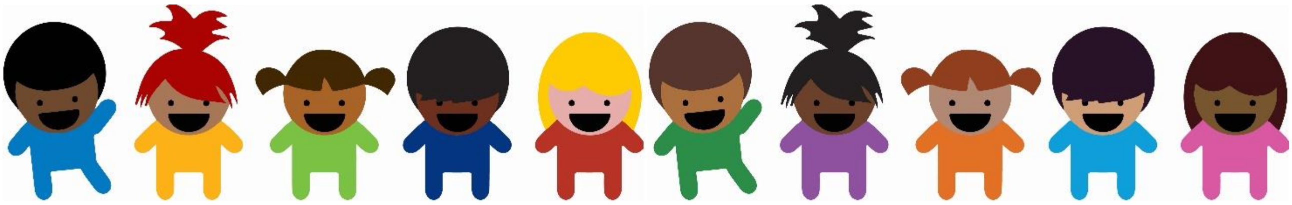
 

____________________________ ______________________

Handtekening van die kind (Naam of X) Datum

APPENDIX F (ii): Explanatory Document and Letter of Assent of Participant (Control Group)

INLIGTINGSPAMFLET EN INSTEMMINGSVORM AAN DEELNEMERS



Navorsingsdeelnemer Identifikasienommer: ________________________________

TITEL VAN DIE NAVORSINGSTUDIE:

Sensoriese Integrasie Disfunksies en Hindernisse in kinders met Fetale Alkohol Spektrum Afwykings (FASA).

NAAM VAN NAVORSER:

Mev. Maritha du Plooy, B. Arbeidsterapie, (Universiteit van Stellenbosch)

Wat is NAVORSING?
Navorsing is iets wat ons doen om meer oor mense en hoe dinge werk, uit te vind. Ons gebruik navorsingsprojekte of studies om ons te help om meer oor mense wat anders is of probleme en siektes het, uit te vind. Navorsing help ons ook om beter maniere te vind om kinders met probleme of wat siek is, te help en te behandel sodat hulle ook na die beste van hulle vermoë kan presteer.

Waaroor gaan hierdie navorsingstudie?
Wanneer ‘n swanger moeder alkohol drink, plaas sy haar ongebore baba in gevaar. FASA is ‘n groep groei-, brein- en fisiese probleme wat mag voorkom in ‘n baba wanneer die moeder **alkohol drink tydens haar swangerskap.** Hierdie navorsingstudie word gedoen om te kyk hoe kinders wat deur alkohol geaffekteer is voordat hulle gebore is, se breine inligting vanaf die sintuie soos die ore en oë ontvang en organiseer vir gebruik. Dit is belangrik om te weet omdat dit hulle manier van dink en leer asook hulle gedrag beïnvloed.

Hoekom is ek uitgenooi om aan hierdie navorsingsprojek deel te neem?
Jy word gevra om aan hierdie studie deel te neem omdat jy in Graad R of 2 in jou skool is en voorheen aan die studie deelgeneem het. Jy is tydens daardie studie gemeet en die dokter het na jou gesig, ore en hande gekyk. Jy het dalk ook 'n paar kort toetsies saam met die sielkundige gedoen. Hulle het gevind het dat jy nie voor jou geboorte deur alkohol geaffekteer is nie en daarom sal jy ons, deur deel te neem aan

hierdie studie, kan help om te verstaan wat die verskille is tussen kinders wat wel geaffekteer is en ‘n kind soos jy, wat nie is nie. Jou ouers het ingestem dat jy weer gevra mag word om aan ‘n studie deel te neem.

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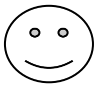
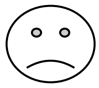
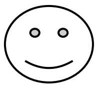
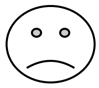
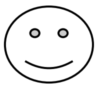
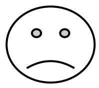



Het die tannie al jou vrae beantwoord?

Verstaan jy dat jy enige tyd mag besluit om nie meer aan die studie deel te neem nie?

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Handtekening van die kind (Naam of X) Datum

APPENDIX G: Diagnostic guidelines for specific fetal alcohol spectrum disorders (FASD) according to the IOM as clarified by Hoyme et al. (2005)

Reprinted with written permission from Dr Hoyme (Hoyme, et al., 2005, p. 44)

I. FAS With Confirmed Maternal Alcohol Exposure (requires all features A- D)

A. Confirmed maternal alcohol exposure

B. Evidence of a characteristic pattern of minor facial anomalies, including ≥2 of the following
   1. Short palpebral fissures (≤10th percentile)
   2. Thin vermilion border of the upper lip (score 4 or 5 with the lip/philtrum guide)
   3. Smooth philtrum (score 4 or 5 with the lip/philtrum guide)

C. Evidence of prenatal and/or postnatal growth retardation
   1. Height or weight ≤10th percentile, corrected for racial norms, if possible

D. Evidence of deficient brain growth or abnormal morphogenesis, including ≥1 of the following
   1. Structural brain abnormalities
   2. Head circumference ≤10th percentile

II. FAS Without Confirmed Maternal Alcohol Exposure

IB, IC, and ID, as above

III. Partial FAS With Confirmed Maternal Alcohol Exposure (requires all features, A-C)

A. Confirmed maternal alcohol exposure

B. Evidence of a characteristic pattern of minor facial anomalies, including ≥2 of the following
   1. Short palpebral fissures (≤10th percentile)
   2. Thin vermilion border of the upper lip (score 4 or 5 with the lip/philtrum guide)
   3. Smooth philtrum (score 4 or 5 with the lip/philtrum guide)

C. One of the following other characteristics
   1. Evidence of prenatal and/or postnatal growth retardation
      a. Height or weight ≤10th percentile corrected for racial norms, if possible

2. Evidence of deficient brain growth or abnormal morphogenesis, including ≥1 of the following
    a. Structural brain abnormalities
    b. Head circumference ≤10th percentile
3. Evidence of a complex pattern of behavioural or cognitive abnormalities inconsistent with developmental level that cannot be explained by genetic predisposition, family background, or environment alone
    a. This pattern includes marked impairment in the performance of complex tasks (complex problem solving, planning, judgement, abstraction, metacognition, and arithmetic tasks); higher-level receptive and expressive language deficits; and disordered behaviour (difficulties in personal manner, emotional ability, motor dysfunction, poor academic performance, and deficient social interaction)

IV. Partial FAS Without Confirmed Maternal Alcohol Exposure

IIIB and IIIC, as above

V. ARBD (requires all features, A-C)

A. Confirmed maternal alcohol exposure

B. Evidence of a characteristic pattern of minor facial anomalies, including ≥2 of the following
    1. Short palpebral fissures (≤10th percentile)
    2. Thin vermilion border of the upper lip (score 4 or 5 with the lip/philtrum 1 guide)
    3. Smooth philtrum (score 4 or 5 with the lip/philtrum guide)

C. Congenital structural defects in ≥1 of the following categories, including malformations and dysplasias (if the patient displays minor anomalies only, ≥2 must be present): *cardiac:* atrial septal defects, aberrant great vessels, ventricular septal defects, conotruncal heart defects; *skeletal*: radioulnar synostosis, vertebral segmentation defects, large joint contractures, scoliosis; *renal*: aplastic/hypoplastic/dysplastic kidneys, "horseshoe" kidneys/ureteral duplications; *eyes*: strabismus, ptosis, retinal vascular anomalies, optic nerve hypoplasia; *ears*: conductive hearing loss, neurosensory hearing loss; *minor anomalies*: /hypoplastic nails, short fifth digits, clinodactyly of fifth fingers, pectus carinatum/excavatum, camptodactyly, " hockey stick" palmar creases, refractive errors, "railroad track" ears

VI. ARND (requires both A and B)

A. Confirmed maternal alcohol exposure

B. At least 1 of the following
   1. Evidence of deficient brain growth or abnormal morphogenesis, including ≥1 of the following
      a. Structural brain abnormalities
      b. Head circumference ≤10th percentile
   2. Evidence of a complex pattern of behavioural or cognitive abnormalities inconsistent with developmental level that cannot be explained by genetic predisposition, family background, or environment alone.
      a. 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 behaviour (difficulties in personal manner, emotional lability, motor dysfunction, poor academic performance, and deficient social interaction)

In the proposed diagnostic criteria, the following considerations apply. Each of the categories assumes that genetic and medical assessment has ruled out a phenocopy, including other genetic and malformation syndromes. Confirmed maternal alcohol exposure is defined as a pattern of excessive intake characterized by substantial regular intake or heavy episodic drinking. Evidence of this pattern may include frequent episodes of intoxication, development of tolerance or withdrawal, social problems related to drinking, legal problems related to drinking, engaging in physically hazardous behaviour while drinking, or alcohol-related medical problems such as hepatic disease. Confirmation may be from maternal interview or reliable collateral sources.

ARBD and ARND refer to clinical conditions in which there must be a history of maternal alcohol exposure, and in which clinical and/or animal research must link maternal alcohol ingestion to the observed outcome. ARBD encompasses children with major and /or minor structural anomalies who display normal growth and intellectual development. ARND comprises a specific pattern of disordered behaviour and development among children with normal growth and structural development.

APPENDIX H: Definition of Documented Prenatal Alcohol Exposure (as Applied to the Diagnostic Categories Set Forth in APPENDIX I)

Reprinted with written permission from dr E Hoyme from (Hoyme, et al., 2016, p. 5)

One or more of the following conditions must be met to constitute documented prenatal alcohol exposure during pregnancy (including drinking levels reported by the mother 3 months before her report of pregnancy recognition or a positive pregnancy test documented in the medical record). The information must be obtained from the biological mother or a reliable collateral source (e.g., family member, social service agency, or medical record):

- ≥6 drinks/week for ≥2 weeks during pregnancy[a]
- ≥3 drinks per occasion on ≥2 occasions during pregnancy[a]
- Documentation of alcohol-related social or legal problems in proximity to (before or during) the index pregnancy (e.g., history of citation[s] for driving while intoxicated or history of treatment of an alcohol-related condition)
- Documentation of intoxication during pregnancy by blood, breath, or urine alcohol content testing
- Positive testing with established alcohol-exposure biomarker(s) during pregnancy or at birth (e.g., analysis of fatty acid ethyl esters, phosphatidylethanol, and/ or ethyl glucuronide in maternal hair, fingernails, urine, or blood, or placenta, or meconium) 50 – 55
- Increased prenatal risk associated with drinking during pregnancy as assessed by a validated screening tool of, for example, T-ACE (tolerance, annoyance, cut down, eye-opener) or AUDIT (alcohol use disorders identification test) [56]

Assignment of documented prenatal alcohol exposure to any individual case requires the sound judgment of an experienced clinician.

[a] These criteria for maternal drinking are based on large epidemiologic studies that demonstrate adverse fetal effects from ≥3 drinks per occasion 26, 57 and others that indicate 1 drink/day as a threshold measure for FASD. [58 –60]

APPENDIX I: Updated Criteria for the Diagnosis of FASD

Reprinted with written permission from dr Hoyme from (Hoyme, et al., 2016, pp. 3-4)

Diagnostic Categories
(See Table 2 for definition of documented prenatal alcohol exposure)
I. FAS (With or without documented prenatal alcohol exposure) A diagnosis of FAS requires all features, A–D: A. A characteristic pattern of minor facial anomalies, including ≥2 of the following: 1. Short palpebral fissures (≤10th centile) 2. Thin vermilion border of the upper lip (rank 4 or 5 on a racially normed lip/philtrum guide, if available) 3. Smooth philtrum (rank 4 or 5 on a racially normed lip/philtrum guide, if available) B. Prenatal and/or postnatal growth deficiency 1. Height and/or weight ≤10th centile (plotted on a racially or ethnically appropriate growth curve, if available) C. Deficient brain growth, abnormal morphogenesis, or abnormal neurophysiology, including ≥1 of the following: 1. Head circumference ≤10th percentile 2. Structural brain anomalies 3. Recurrent nonfebrile seizures (other causes of seizures having been ruled out) D. Neurobehavioral impairment 1. For children ≥3 y of age (a or b): a. WITH COGNITIVE IMPAIRMENT: ▪ Evidence of global impairment (general conceptual ability ≥1.5 SD below the mean, or performance IQ or verbal IQ or spatial IQ ≥1.5 SD below the mean) OR ▪ Cognitive deficit in at least one neurobehavioral domain ≥1.5 SD below the mean (executive functioning, specific learning impairment, memory impairment or visual-spatial impairment)

b. WITH BEHAVIORAL IMPAIRMENT WITHOUT COGNITIVE IMPAIRMENT:

- Evidence of behavioural deficit in at least one domain ≥1.5 SD below the mean in impairments of self-regulation (mood or behavioural regulation impairment, attention deficit, or impulse control)

2. For children <3 y of age:
   - Evidence of developmental delay ≥1.5 SD below the mean

II. PFAS

- **For children with documented prenatal alcohol exposure, a diagnosis of PFAS requires features A and B:**

A. A characteristic pattern of minor facial anomalies, including ≥2 of the following:
   1. Short palpebral fissures (≤10th centile)
   2. Thin vermilion border of the upper lip (rank 4 or 5 on a racially normed lip/philtrum guide, if available)
   3. Smooth philtrum (rank 4 or 5 on a racially normed lip/philtrum guide, if available)

B. Neurobehavioral impairment
   1. For children ≥3 y of age (a or b):

a. WITH COGNITIVE IMPAIRMENT:

- Evidence of global impairment (general conceptual ability ≥1.5 SD below the mean, or performance IQ or verbal IQ or spatial IQ ≥1.5 SD below the mean)

  OR

- Cognitive deficit in at least 1 neurobehavioral domain ≥1.5 SD below the mean (executive functioning, specific learning impairment, memory impairment or visual-spatial impairment)

b. WITH BEHAVIORAL IMPAIRMENT WITHOUT COGNITIVE IMPAIRMENT:

- Evidence of behavioural deficit in at least one domain ≥1.5 SD below the mean in impairments of self-regulation (mood or behavioural regulation impairment, attention deficit, or impulse control)

2. For children <3 y of age:
   - Evidence of developmental delay ≥1.5 SD below the mean

- **For children without documented prenatal alcohol exposure, a diagnosis of PFAS requires all features, A–C:**

A. A characteristic pattern of minor facial anomalies, including ≥2 of the following:
   1. Short palpebral fissures (≤10th centile)
   2. Thin vermilion border of the upper lip (rank 4 or 5 on a racially normed lip/philtrum guide, if available)
   3. Smooth philtrum (rank 4 or 5 on a racially normed lip/philtrum guide, if available)

B. Growth deficiency or deficient brain growth, abnormal morphogenesis, or abnormal neurophysiology
   1. Height and/or weight ≤10th centile (plotted on a racially or ethnically appropriate growth curve, if available), or:
   2. Deficient brain growth, abnormal morphogenesis or neurophysiology, including ≥1 of the following:
      a. Head circumference ≤10th percentile
      b. Structural brain anomalies
      c. Recurrent nonfebrile seizures (other causes of seizures having been ruled out)

C. Neurobehavioral impairment
   1. For children ≥3 y of age (a or b):
      a. WITH COGNITIVE IMPAIRMENT:
         - Evidence of global impairment (general conceptual ability ≥1.5 SD below the mean, or performance IQ or verbal IQ or spatial IQ ≥1.5 SD below the mean)

           OR

         - Cognitive deficit in at least 1 neurobehavioral domain ≥1.5 SD below the mean (executive functioning, specific learning impairment, memory impairment, or visual-spatial impairment)

      b. WITH BEHAVIORAL IMPAIRMENT WITHOUT COGNITIVE IMPAIRMENT:
         - Evidence of behavioural deficit in at least 1 domain ≥1.5 SD below the mean in impairments of self-regulation (mood or behavioural regulation impairment, attention deficit, or impulse control)

   2. For children <3 y of age:
         - Evidence of developmental delay ≥1.5 SD below the mean

III. ARND

Requires features A and B (this diagnosis cannot be made definitively in children <3 y of age):

A. Documented prenatal alcohol exposure

B. Neurobehavioral impairment [a]

1. For children ≥3 y of age (a or b):
    a. WITH COGNITIVE IMPAIRMENT:
    - Evidence of global impairment (general conceptual ability ≥1.5 SD below the mean, or performance IQ or verbal IQ or spatial IQ ≥1.5 SD)

      OR

    - Cognitive deficit in at least two neurobehavioral domains ≥1.5 SD below the mean (executive functioning, specific learning impairment, memory impairment or visual-spatial impairment)

    b. WITH BEHAVIORAL IMPAIRMENT WITHOUT COGNITIVE IMPAIRMENT:
    - Evidence of behavioural deficit in at least two domains ≥1.5 SD below the mean in impairments of self-regulation (mood or behavioural regulation impairment, attention deficit, or impulse control)

IV. ARBD

Requires features A and B:

A. Documented prenatal alcohol exposure

B. One or more specific major malformations demonstrated in animal models and human studies to be the result of prenatal alcohol exposure: cardiac: atrial septal defects, aberrant great vessels, ventricular septal defects, conotruncal heart defects; skeletal: radioulnar synostosis, vertebral segmentation defects, large joint contractures, scoliosis; renal: aplastic/hypoplastic/dysplastic kidneys, “horseshoe” kidneys/ureteral duplications; eyes: strabismus, ptosis, retinal vascular anomalies, optic nerve hypoplasia; ears: conductive hearing loss, neurosensory hearing loss

Diagnostic Caveats: The assignment of an FASD is a complex medical diagnostic process best accomplished through a multidisciplinary approach. As is the case with many medical conditions, sound clinical judgment must be used. Differential diagnoses should always include genetic disorders or conditions arising from other teratogens. Additionally, because head circumference, growth, and many cognitive and behavioural characteristics have moderate to high degrees of heritability, when information is available about the biological parents, these data should be considered in the final diagnostic decision.

[a] Adaptive skills should be assessed, but such deficits cannot stand alone for diagnosis.