School of Public Health

Fetal Alcohol Spectrum Disorder and Fine Motor Skills: A Population-based Study of Children in the Fitzroy Valley

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Declaration

To the best of my knowledge and belief this thesis contains no material previously published by any other person except where due acknowledgment has been made.

This thesis contains no material that has been accepted for the award of any other degree or diploma in any university.

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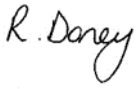
Signature: 

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Abstract

Background

Alcohol consumption during pregnancy is teratogenic to fetal development, and can cause delayed growth, dysmorphic facial features and irreversible neurodevelopmental impairment. Individuals with prenatal alcohol exposure (PAE) and significant impairment across a range of areas can be diagnosed with a Fetal Alcohol Spectrum Disorder (FASD). Impairments can include cognitive, language, behavioural, emotional and sensory-motor impairments, including damage to fine motor skills. Fine motor skills include precision, visual-motor integration, dexterity and upper-limb coordination, and facilitate performance of many everyday tasks. They are particularly important for school-aged children because many academic tasks, such as handwriting and drawing (graphomotor skills), are essential to effectively communicate ideas and demonstrate learning, and rely on sound fine motor skills. However, few studies have comprehensively reported fine motor skills in children with PAE or FASD.

Aboriginal leaders in the Fitzroy Valley region of Western Australia were concerned about the high levels of alcohol misuse by people in their communities, especially by pregnant women. They were worried that many children in their communities had undiagnosed FASD and would be unable to pass on their Aboriginal language and culture to successive generations. Consequently, they conceived and developed the Lililwan Project, which was Australia's first population-based prevalence study of FASD. 'Lililwan' means 'all the little ones' in Kimberley Kriol. This thesis reports the fine motor skills of children involved in the Lililwan Project, including those with and without PAE and those who were diagnosed with a type of FASD. This is the first such study among Australian Aboriginal children.

The thesis aims to fill two gaps in the scientific literature: 1) the paucity of information regarding the fine motor abilities of Aboriginal children in remote regions of Australia (this is important considering the children's consistent underperformance on national academic testing) and 2) the lack of a comprehensive profile of fine motor impairment in children with PAE and/or FASD, and the implications this has for FASD diagnosis and treatment.

Methods

In 2011, children in the Fitzroy Valley who were born in 2002 or 2003 (*n* = 108, 7.5 to 9.6 years) underwent comprehensive health and neurodevelopmental assessments by a multidisciplinary team including an audiologist, occupational therapist (the doctoral candidate), ophthalmologist, paediatrician, physiotherapist, psychologist, and speech pathologist. FASD diagnoses were made based on modified Canadian FASD Diagnostic Guidelines. Fine motor assessments were conducted by the occupational therapist using observational assessment (graphomotor skills: human-figure drawing; name and sentence writing; pencil grasp; writing pressure) and standardised tools (Beery Buktenica Developmental Test of Visual-Motor Integration (Beery VMI); and Bruininks-Oseretsky Test of Motor Proficiency (BOT-2)). Graphomotor samples were analysed retrospective to the Lililwan Project by two independent occupational therapists and scored according to the Evaluation Tool of Children's Handwriting and Miller Function and Participation Scales criteria.

Descriptive statistics, including means and standard deviations, were reported for the overall cohort. Parametric and non-parametric tests were used to compare children i) without PAE; ii) with PAE, but who were not diagnosed with FASD because they did not meet full diagnostic criteria; and iii) who were diagnosed with a type of FASD. Rates of 'moderate' (≤ 16th percentile) and 'severe' (≤ 2nd percentile) fine motor impairment were determined.

Results

The overall cohort had a distinct fine motor profile, with specific areas of strengths and difficulties. Although the mean Fine Motor Composite scores from the BOT-2 were in the average range, this composite score masked strengths and weaknesses: the BOT-2 Fine Motor Integration (copying geometric shapes) scores approached 'below average', which was consistent with the 'below average' Beery VMI scores; on the other hand BOT-2 Upper-Limb Coordination (ball skills) scores approached 'above average'. Many children used a pencil grasp, which was delayed for their age, and handwriting legibility was poor.

Children with PAE who were not diagnosed with FASD did not score significantly different from children without PAE or those with FASD, but children with FASD had specific deficits. Children with FASD scored significantly lower on BOT-2 Fine Motor Composite and Manual Coordination tests than children without PAE. Children with FASD also had lower Beery VMI Fine Motor Coordination scores, and were more

likely to use a cross-thumb pencil grasp (which was delayed for their age), apply heavy writing pressure, be unable to write a short sentence, and show poorer word legibility.

Few children in the cohort, including those with PAE and/or FASD, had a 'severe' fine motor impairment. However, rates of 'moderate' impairment were very high for many skills, even in children without PAE. Moderate impairment can indicate considerable dysfunction and indicate a need for intervention. Children with FASD had the highest rates of impairment across most types of fine motor skills.

Conclusion

Children in the Fitzroy Valley had a distinct profile of fine motor abilities, as did the sub-group of children with FASD. A range of fine motor skills should be assessed in children with PAE and suspected or confirmed FASD, and results should be reported as discrete skills rather than as a combined motor score. The BOT-2 and Beery VMI tests appear to be well-suited for FASD diagnostic assessment, as they assess a range of skills and report them as distinct skill areas.

Rates of 'severe' impairment were very low, which was an unexpected finding given the high rates of PAE and FASD. The Canadian FASD Diagnostic Guidelines recommend that only fine motor scores below the 2nd percentile should be deemed significant for FASD diagnosis, but this cut-off may have been too restrictive. Scores below the 16th percentile were common, and this suggests significant clinical impairment. It may be clinically useful to include observations of functional fine motor performance, such as graphomotor skills, as part of the FASD diagnostic process, rather than relying solely on scores from standardised assessment tools.

As in many remote regions of Australia, allied health services, including occupational therapy, are limited in the Fitzroy Valley region. The high rates of PAE and FASD, along with identified fine motor impairment, warrant an increase in coordinated services by multi-disciplinary teams with a focus on developing sustainable service delivery.

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Abbreviations

AEDI	Australian Early Developmental Index
ANOVA	Analysis of Variance
AUDIT-C	Alcohol Use Disorders Identification Test - Consumption
ARND	Alcohol Related Neurodevelopmental Disorder
Beery VMI	Beery-Buktenica Developmental Test of Visual-Motor Integration
BOT-2	Bruininks-Oseretsky Test of Motor Proficiency (2nd edition)
CAT	Critical Appraisal Tool
CDC	Centers for Disease Control
CI	Confidence Interval
CNS	Central nervous system
ETCH	Evaluation Tool of Children's' Handwriting
FAS	Fetal Alcohol Syndrome
FASD	Fetal Alcohol Spectrum Disorder/s
FM	Fine motor
GM	Gross motor
HSD	Tukey's Honestly Significant Difference test
IOM	Institute of Medicine
IQ	Intelligence Quotient
M	Mean
M-FUN	Miller's Function and Participation Scales
NAPLAN	National Assessment of Performance – Literacy and Numeracy
ND-AE	Neurodevelopmental Disorder – Alcohol Exposed
ND-PAE	Neurodevelopmental Disorder – Prenatal Alcohol Exposed
PAE	Prenatal alcohol exposure
pFAS	Partial Fetal Alcohol Syndrome
SD	Standard deviation
UNIT	Universal Non-verbal Intelligence Test
US	United States
UW	University of Washington
VMI	Visual-motor integration
WA	Western Australia/n

Terminology

- The term ‘Fetal Alcohol Spectrum Disorder’ (FASD) is used interchangeably with ‘Fetal Alcohol Spectrum Disorders’ in the literature, with Canadian sources tending to favour the former and the United States the latter. Australian sources use both terms, and occasionally use the English spelling for ‘Foetal’. This thesis uses the term ‘Fetal Alcohol Spectrum Disorder’ except in some instances in publications because the editor or reviewers requested alternative terminology be used.
- Fetal Alcohol Spectrum Disorder is not a diagnosis, but a collective term to describe the spectrum of disorders that can result following prenatal alcohol exposure (PAE). Thus, it is technically correct to write ‘a type of FASD’ or ‘a diagnosis on the FASD spectrum’ to avoid insinuating that FASD is a diagnostic term. However, for the sake of brevity and readability, this thesis does not conform to this convention (except when requested by editors or reviewers of some of the publications), in keeping with much of the published literature in the area.
- The term ‘Aboriginal’ rather than ‘Indigenous’ is used when referring to people in the Fitzroy Valley as it is the preferred terminology of the local people. ‘Indigenous’ is used when discussing results from the NAPLAN and other studies because this is more commonly accepted terminology Australia-wide, and includes people of the Torres Strait Islands.
- The Beery-Buktenica Developmental Test of Visual-Motor Integration Motor Coordination (Beery VMI) subtest is referred to as the ‘Fine Motor Coordination’ subtest in this thesis to avoid confusion with tests of gross motor coordination skills.
- Traditionally, birth and child-rearing are considered ‘women’s business’ in Aboriginal culture, and men were unable to comment or be involved in some aspects of the Lililwan Project. As such, this thesis often refers to ‘the women of the Fitzroy Valley’. This does not imply that men weren’t involved or consulted when appropriate, or that they are not involved in issues related to FASD.
- Visual-motor integration is sometimes considered a type of fine motor skill, but it differs somewhat from other fine motor skills as it incorporates different neural pathways and is closely tied with visual perception, as well as some aspects of executive function. Sometimes visual-motor integration and fine motor skills are differentiated in this thesis, but when only the term ‘fine motor skills’ is used, this should be considered to include visual-motor integration.

Publications

The following publications are included as part of this thesis:

1. Doney, R., Lucas, B. R., Jones, T., Howat, P., Sauer, K., & Elliott, E. J. (2014). Fine motor skills in children with prenatal alcohol exposure or Fetal Alcohol Spectrum Disorder. *Journal of Developmental and Behavioral Pediatrics*, *35*(9), 598-609. doi:10.1097/dbp.0000000000000107
2. Doney, R., Lucas, B. R., Watkins, R. E., Tsang, T. W., Sauer, K., Howat, P., Latimer, J., Fitzpatrick, J. P., Oscar, J., Carter, M., & Elliott, E. J. (2017). Fine motor skills in a population of children in remote Australia with high levels of prenatal alcohol exposure and Fetal Alcohol Spectrum Disorder. *BMC Pediatrics, 17*(193), 1-10. doi: 10.1186/s12887-017-0945-2
3. Doney, R., Lucas, B. R., Watkins, R. E., Tsang, T. W., Sauer, K., Howat, P., Latimer, J., Fitzpatrick, J. P., Oscar, J., Carter, M., & Elliott, E. J. (2016). Visual-motor integration, visual perception, and fine motor coordination in a population of children with high levels of Fetal Alcohol Spectrum Disorder. *Research in Developmental Disabilities, 55*, 346-357. doi:10.1016/j.ridd.2016.05.009
4. Doney, R., Lucas, B. R., Jirikowic, T., Tsang, T. W., Watkins, R. E., Sauer, K., Howat, P., Latimer, J., Fitzpatrick, J. P., Oscar, J., Carter, M., & Elliott, E. J. (2016). Graphomotor skills in children with prenatal alcohol exposure and Fetal Alcohol Spectrum Disorder: A population-based study in remote Australia. *Australian Occupational Therapy Journal*, *64*(1), 68-78. doi: 10.1111/1440-1630.12326

Statement of Contribution of Others

The doctoral candidate was the sole occupational therapist with the Lililwan Project's multi-disciplinary neurodevelopmental assessment team. The author designed the fine motor assessment protocol for the Lililwan Project and selected the fine motor assessment tools; applied for ethics approval relevant to the fine motor aspects of the Lililwan Project; carried out and scored the fine motor assessments then analysed fine motor data and interpreted results under the supervision of qualified statisticians; and drafted and finalised the published papers and the thesis.

The publications included in this thesis were written by the thesis author with contributions and input from the following supervisors and/or co-authors:

- **Professor Peter Howat**: supervised the thesis, assisted with drafting and critical review of the thesis and published papers.
- **Dr Kay Sauer**: supervised the thesis; assisted with drafting and critical review of the thesis and published papers.
- **Dr Jonine Jancey**: supervised the thesis; assisted with drafting and critical review of the thesis.
- **Professor Elizabeth Elliott**: supervised the thesis as an associate supervisor; assisted with drafting and critical review of the thesis and published papers; conceptualised, designed, and applied for ethics approval for the Lililwan Project.
- **Ms Barbara Lucas**: assisted with analysing fine motor data from the Bruininks-Oseretsky Test of Motor Proficiency; assisted with drafting and critical review of the published papers.
- **Dr Rochelle Watkins**: assisted with statistical analysis and interpretation of results; assisted with drafting and critical review of the published papers.
- **Dr Tracey Tsang**: assisted with statistical analysis and interpretation of results; assisted with drafting and critical review of the published papers.
- **Professor Jane Latimer**: conceptualised, designed, and applied for ethics approval for the Lililwan Project; assisted with drafting and critical review of the published papers.
- **Dr James Fitzpatrick**: conceptualised, designed, and applied for ethics approval for the Lililwan Project; assisted with drafting and critical review of the published papers.

- **Ms Maureen Carter**: conceptualised, designed, and applied for ethics approval for the Lililwan Project; assisted with drafting and critical review of the published papers.
- **Ms June Oscar**: conceptualised, designed, and applied for ethics approval for the Lililwan Project; assisted with drafting and critical review of the published papers.
- **Ms Taryn Jones**: assisted with data extraction, drafting, and critical review of the systematic review published paper.
- **Dr Tracy Jirikowic**: assisted with drafting and critical review of the graphomotor published paper.
- **Dr Jennifer Nash**: assisted with scoring the graphomotor assessments.
- **Ms Dianne Rios**: assisted with scoring the graphomotor assessments.

Signed statements for each of the above co-authors are included in Appendix F.

Additional Publications

The following publications were co-authored by the doctoral candidate as part of the Lililwan Project, but do not form part of this thesis:

1. Fitzpatrick, J., Elliott, E. J., Latimer, J., Carter, M., Oscar, J., Ferreira, M. L, Carmichael-Olson, H., Lucas, B. R., **Doney, R.**, Salter, C., Peadon, E., Hawkes, G., Hand, M. (2012). The Lililwan Project: Study protocol for a population-based active case ascertainment study of the prevalence of Fetal Alcohol Spectrum Disorders (FASD) in remote Australian Aboriginal communities. *BMJ Open, 2*, 1-11. doi:10.1136/bmjopen-2012-000968
2. Fitzpatrick, J. P., Latimer, J., Carter, M., Oscar, J., Ferreira, M. L., Carmichael-Olson, H., Lucas, B. R., **Doney, R.**, Salter, C., Try, J. (2015). Prevalence of Fetal Alcohol Syndrome in a population-based sample of children living in remote Australia: The Lililwan Project. *Journal of Paediatrics and Child Health, 51*(4), 450-457. doi:10.1111/jpc.12814
3. Fitzpatrick, J. P., Latimer, J., Olson, H. C., Carter, M., Oscar, J., Lucas, B. R., **Doney, R**., Salter, C., Try, J., Hawkes, G., Fitzpatrick, E., Hand, M., Watkins, R. E., Tsang, T. W., Bower, C., Ferreira, M. L., Boulton, J., Elliott, E. J. (2017). Prevalence and profile of neurodevelopment and Fetal Alcohol Spectrum Disorder (FASD) amongst Australian Aboriginal children living in remote communities. *Research in Developmental Disabilities, 65*, 114-126. doi:10.1016/j.ridd.2017.04.001
4. Lucas, B. R., Latimer, J., **Doney, R**., Ferreira, M. L., Adams, R., Hawkes, G., Fitzpatrick, J. P., Hand, M., Oscar, J., Carter, M. (2013). The Bruininks-Oseretsky Test of Motor Proficiency-Short Form is reliable in children living in remote Australian Aboriginal communities. *BMC Pediatrics, 13*(1), 135. doi:10.1186/1471-2431-13-135
5. Lucas, B. R., Latimer, J., Pinto, R. Z., Ferreira, M. L., **Doney, R.**, Lau, M., Jones, T., Dries, D., Elliott, E. J. (2014). Gross motor deficits in children prenatally exposed to alcohol: A meta-analysis. *Pediatrics*, *134*(1), e192-e209. doi:10.1542/peds.2013-3733
6. Lucas, B. R., Latimer, J., Fitzpatrick, J. P., **Doney, R**., Watkins, R. E., Tsang, T. W., Jirikowic, T., Olson, H. C., Oscar, J., Carter, M., Elliott, E. J. (2016). Soft neurological signs and prenatal alcohol exposure: a population-based

study in remote Australia. *Developmental Medicine and Child Neurology*, *58*(8), 861-867. doi: 10.1111/dmcn.13071

7. Lucas, B. R., **Doney, R.**, Latimer, J., Watkins, R. E., Tsang, T. W., Hawkes, G., Fitzpatrick, J. P., Oscar, J., Carter, M., Elliott, E. J. (2016). Impairment of motor skills in children with Fetal Alcohol Spectrum Disorders in remote Australia: The Lililwan Project. *Drug and Alcohol Review, 35*(6), 719-727. doi:10.1111/dar.12375
8. Lucas, B. R., Latimer, J., **Doney, R.**, Watkins, R. E., Tsang, T. W., Hawkes, G. B., Fitzpatrick, J. P., Oscar, J., Carter, M., Elliott, E. J. (2016). Gross motor performance in children prenatally exposed to alcohol and living in remote Australia. *Journal of Paediatrics and Child Health*, *52*(8) 814-824. doi:10.1111/jpc.13240

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This thesis is dedicated to the children of the Fitzroy Valley, who often made me laugh, and at times made me cry, but always filled my days with smiles. To the resilient and courageous families who welcomed our team into their lives and communities, and shared their world and stories with us; and to the teachers and schools who magically created time in their hectic schedules to support the project because they believed in better outcomes for the children — I thank you with all my heart.

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Thank you to my colleagues from the Lililwan Project who shared many funny and the occasional not-so-funny moments (involving large snakes and flat tyres) in our year of camping in the Fitzroy Valley. A special thanks to Ms Barbara Lucas for her support and for caring about motor skills as much as I do, and to Ms Sharon Eadie for her tireless help. Special thanks to the local people of the Fitzroy Valley who conceptualised and assisted at all stages of the Lililwan Project and shared your beautiful country and culture: Ms June Oscar, Ms Maureen Carter, Ms Marmingee Hand, Ms Annette Kogolo, Mr Stanley Marr, Ms Marilyn Oscar and Ms Natalie Davies.

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Preface

This thesis reports fine motor skills of children who participated in the Lililwan Project. The Lililwan Project was the first comprehensive health and neurodevelopmental evaluation of a cohort of almost two entire age groups of Australian Aboriginal children, and the first population-based prevalence study of Fetal Alcohol Spectrum Disorder in a remote setting in Australia.

This thesis constitutes the research component of the Doctor of Public Health degree. In addition to this thesis, six public health-related units have been completed by the doctoral candidate to meet the requirements for the doctoral degree.

The thesis is presented to meet the requirements for a thesis by publication:

The first chapter provides background to the Lililwan Project, including the history of alcohol consumption in the Fitzroy Valley, existing knowledge about fine motor skills of Aboriginal children and the impact of prenatal alcohol exposure on neurodevelopment. Chapter 1 also outlines the significance of the research and the study aim and objectives.

The second chapter comprises a published systematic review of the literature related to FASD and fine motor skills in primary school aged children.

Chapter three is an expanded methods section, providing extra detail to that which is available in each of the publications.

Chapter four comprises three publications, each of which include a literature review relevant to the specific type of fine motor skill outcome reported, detailed methods pertaining to administration of each assessment tool, a discussion of the implications of the fine motor outcomes reported in each paper and a reference list. Each section is formatted as per the publication requirements of the respective publishing journals. Facsimile PDF versions of each of the published papers are included in Appendix C. Chapter four reports additional data to that included in the publications, specifically the number of children with a significant fine motor impairment according to the diagnostic criteria used in the Lililwan Project.

The fifth chapter provides an overview of key findings, discusses strengths and limitations of the research, and makes recommendations incorporating research findings into clinical practice and service delivery.

Chapter 1 Introduction

1.1 The Fitzroy Valley

The Fitzroy Valley is in the central Kimberley region, in northern Western Australia (Appendix A - Map of the Fitzroy Valley). It spans an area approximately 200km in diameter, and is bounded by the Great Sandy Desert to the south, Napier Ranges and Leopold Ranges to the west and north, and extends towards Halls Creek to the east. It is bisected by the Fitzroy River, one of the largest rivers in Australia.

The Fitzroy Valley has a population of approximately 3,500 people. About 1,000 people live in the main service town of Fitzroy Crossing and six adjoining communities (Morphy, 2010). The remainder of the population lives in approximately 45 outlying communities, which can range from few or no people at certain times of the year, to permanently established communities with populations of up to 300 people.

Approximately 80% of the population identify as being Australian Aboriginal, with most non-Indigenous people living in Fitzroy Crossing. The Aboriginal people of the Fitzroy Valley include five distinct language and cultural groups: the Bunaba, Gooniyandi, Nykina, Walmajarri and Wangkatjungka peoples.

The climate ranges from sunny, warm days of 32 to 36 degrees Celsius and cool nights in the 'Dry Season' (May to September), to hot, humid days and nights in the 'Build-up Season' (October to November), and tropical downpours in the 'Wet Season' (October to March) (Australian Government Bureau of Meteorology, 2016). Many communities are isolated from access and services during the 'Wet Season', when extensive flooding can occur.

Fitzroy Crossing has had a troubled, and at times violent, social history (Hawke, 2013; Morphy, 2010). The Aboriginal people of the Fitzroy Valley are thought to have inhabited the area for up to 40,000 years prior to non-Indigenous settlement (Isaacs, 2006). The Fitzroy Valley was established as a pastoral region in the 1890s, which resulted in the loss of traditional Aboriginal lands. However, many Aboriginal families worked for rations for local pastoralists and, by continuing to live close to their traditional lands, retained a connection to their culture, language and traditions (Hawke, 2013). The situation changed in the late 1960s with the introduction of equal wages for Aboriginal people, because many pastoralists could not afford, or refused to pay, equal wages to Aboriginal workers. People were removed from their traditional lands and a shanty town of displaced Aboriginal people from different cultural and language groups grew to become the current Fitzroy Crossing town. In 1967,

Aboriginal people were acknowledged as Australian citizens, which allowed them to purchase alcohol. The dire consequences of displacement, coupled with access to alcohol, are discussed by Hawke (2013, p. 189):

> The other element that could not be missed was the degradation and misery that revolved around the Crossing Inn. The binge-drinking culture of the white stockmen had been passed on to the men who got their citizenship rights, and from them to the mob at large. Moderate social drinking was never a concept that gained a foothold in Fitzroy and the towns of the north.

Olive Knight (Hawke, 2013, p. 190), a local Aboriginal elder and leader, recalls:

> The women didn't drink at all. Only the men at first…But they were into it with a force you know. They bin (sic) beating up their people and all that sort of thing and in the process draining themselves too. Then it started going out to the older men and then to the women.

The social situation in Fitzroy Crossing continued to worsen. Similar to other displaced Indigenous cultures around the world, excessive alcohol consumption was both a result, and also a perpetuation, of the trauma experienced by loss of land, language, and culture (Bartlett, 2003; Beauvais, 1998; Waldram, Herring, & Young, 1995). An enquiry into 22 deaths in the Fitzroy Valley, led by the Coroner's Court of Western Australia, noted that excessive alcohol consumption was involved in most of the 22 deaths. The report described the social situation in Fitzroy Crossing (State Coroner of Western Australia, 2008, p. 23):

> It was clear that the living conditions for many Aboriginal people in the Kimberley were appallingly bad. The plight of the little children was especially pathetic and for many of these the future appears bleak. Many already suffer from foetal alcohol syndrome and unless major changes occur most will fail to obtain a basic education, most will never be employed and, from a medical perspective, they are likely to suffer poorer health and die younger than other Western Australians. In this context the very high suicide rates for young Kimberley Aboriginal persons were readily explicable.

Local Aboriginal leaders campaigned for a trial of alcohol restrictions for the town, and in 2007 the state government agreed to restrictions that are still in place. Take-away sales of alcohol in Fitzroy Crossing are restricted to low-strength beer. However, it is legal to consume any type of alcohol at either of the town's two licensed hotels, although in accordance with state laws it is illegal to provide alcohol to drunk or

intoxicated people (*Liquor Control Act 1988*), and to bring small amounts of alcohol into the Fitzroy Valley for personal consumption (the nearest location to purchase full-strength take-away beer, wine, or spirits is Derby, 260 kilometres west of Fitzroy Crossing), but it is illegal to on-sell it to others.

The impact of the alcohol restrictions has been noticeable. In the 12 months after restrictions were imposed on the sales of take-away alcohol, an independent review found significant improvements were reported by police, health, and education staff. These included a 50% reduction in Emergency Department presentations, 27% reduction in alcohol-related domestic violence, and a 14% increase in school attendance (Kinnane, Farringdon, Henderson-Yates, & Parker, 2009). The benefits were sustained two years after the imposition of alcohol restrictions (Kinnane, Farringdon, Henderson-Yates, & Parker, 2010).

1.2 Fine Motor Skills of Aboriginal Children in Australia

There is a paucity of data related to fine motor skills of Australian Indigenous children. However, the National Assessment Program – Literacy and Numeracy (NAPLAN), which is an annual Australian-wide test of reading, writing, language, and numeracy conducted with all students in Years 3, 5, 7, and 9, shows that students in the Fitzroy Valley have much lower academic performance than the national average (Australian Curriculum Assessment and Reporting Authority, 2015). Although the NAPLAN does not directly assess fine motor skills, these skills are essential for successful academic performance (Kulp, 1999; McHale & Cermak, 1992; Tomchek & Schneck, 2006), and students with poor handwriting skills receive lower grades despite the content of their work (Chase, 1986). In Western Australia (WA), 50.2% of Year 3 Indigenous children in very remote areas (including Aboriginal children in the Fitzroy Valley) had reading skills below the national average, compared to just 3.7% of non-Indigenous children in WA. Spelling abilities were also low, with 56.1% of remote Indigenous children in WA being below the national average. Poor results were similar across different year levels, including for persuasive writing and numeracy skills (Australian Curriculum Assessment and Reporting Authority, 2015).

There are few data specific to fine motor skills of Australian Indigenous children, and the limited existing data are drawn from small sample sizes, and do not provide a comprehensive profile of abilities. One small study (n = 13) of Indigenous children in an urban school setting assessed visual-motor integration skills using the Beery-Buktenica Developmental Test of Visual-Motor Integration (Beery VMI) (Beery &

Beery, 2010) as part of a pre- and post-assessment for a school skills program (McGarrigle & Nelson, 2006). However, Beery VMI scores were reported as raw scores rather than standardised scores, so comparisons to normative data were not possible. An earlier study of 9 to 18-year-old students (*n* = 81) in Fitzroy Crossing found that both visual-motor and visual-perception skills were below average in relation to norms based on children from Westernised cultures, but the focus of the study was related to visual perception rather than fine motor skills (Maples, Leslie, & Atchley, 1993). The Australian Early Developmental Index (AEDI) is based on teacher reports of student competence. Teachers in Fitzroy Crossing reported that 55.9% of students in their first year of full-time school (*n* = 34) were below the 10th percentile in the Physical Health and Wellbeing domain, including 20.6% below the 10th percentile for fine and gross motor skills (The Royal Children's Hospital Melbourne, 2012). However, the AEDI results are not based on comprehensive evaluation of fine motor skills using standardised assessments, nor are results available for older children.

1.3 Impact of Prenatal Alcohol Exposure on Neurodevelopment

Alcohol is teratogenic to the fetus when consumed in pregnancy. Alcohol freely crosses the placenta and the fetal blood-brain barrier, where it damages developing neural cells (Goodlett, Horn, & Zhou, 2005). PAE can cause permanent, irreversible damage to brain structures, especially the cerebellum, basal ganglia, and corpus callosum, as well as disrupting the interconnectivity between brain regions (Autti-Rämö et al., 2002; Coles & Li, 2011; Mattson et al., 1996; Wozniak et al., 2009). It can also damage nerve conductivity (de los Angeles Avaria et al., 2004) and impair muscle development (David & Subramaniam, 2005).

Fetal Alcohol Spectrum Disorder (FASD) is a term used to describe a set of diagnoses, which result from prenatal alcohol exposure (PAE). Diagnoses on the spectrum include Fetal Alcohol Syndrome (FAS), partial FAS (pFAS), and Alcohol-Related Neurodevelopmental Disorder (ARND) or Neurodevelopmental Disorder – Alcohol Exposed (ND-AE) (Astley & Clarren, 2000; Bertrand et al., 2004; Chudley et al., 2005; Hoyme et al., 2005) (see 3.3 FASD diagnostic criteria).

FAS, the most severe form of FASD, was first identified separately and almost simultaneously by French and US doctors in the late 1970s (Jones & Smith, 1973; Lemoine, Harousseau, Borteyru, & Menuet, 1968). It was noted that babies whose mothers had chronic alcohol misuse problems displayed typical patterns of 'craniofacial, limb, and cardiovascular defects' (Jones & Smith, 1973, p.7815).

Subsequently, it was reported that the children with FAS had persistent poor growth and a range of learning and behavioural delays, and that many of the physical and developmental delays persisted into adulthood. Longitudinal studies of infants with FAS (*n* = 415) found that as adolescents and adults, 70% had had disrupted schooling; 94% had mental health problems, including 23% attempting suicide; 60% had been in trouble with the law; 53% of males and 70% of females had substance misuse disorders; 80% were unemployed; and 80% were unable to live independently (Streissguth, Barr, Kogan, & Bookstein, 1997; Streissguth et al., 2004).

1.4 Significance of the Research

This thesis addresses two notable gaps in the scientific literature. First, little is known about the fine motor skills of Aboriginal children living in remote regions of Australia. Occupational therapists working in the Fitzroy Valley have anecdotally reported that many children in the region have difficulties with fine motor skills, and teachers have reported that many children have difficulties with handwriting. National literacy and numeracy testing shows that many students in the Fitzroy Valley perform below the national average, and it is known that proficiency in fine motor skills is a key component of academic success. However, the only existing evidence of fine motor abilities of children in the Fitzroy Valley is based on teachers' observational reports of general motor abilities of children in their first year of school. No systematic, in-depth study of fine motor skills using standardised assessment tools has been conducted with children in the Fitzroy Valley, who are predominantly of Australian Aboriginal descent, nor with children living in other remote regions of Australia, who plausibly experience similar neurodevelopmental risk factors, including PAE and FASD.

Second, few studies have comprehensively reported a range of fine motor skills in children with FASD, and no studies have reported the prevalence of fine motor impairment in a population-based cohort with high levels of PAE and FASD. Further, despite anecdotal reports of graphomotor impairments in children with FASD, there has been limited detailed exploration of the possible functional effects of PAE on functional fine motor skills.

The findings presented in this thesis provide the first detailed profile of fine motor skills of remote-dwelling Aboriginal children, which will be of benefit to therapists and educators. The findings also add evidence to the growing body of knowledge regarding possible neurodevelopmental effects of PAE.

1.5 Hypothesis

It was predicted that primary school children in the Fitzroy Valley would have higher than expected rates of fine motor impairment, and that they would be higher in children with PAE and/or FASD than those without PAE.

1.6 Aim and Objectives

The aim of the study was to develop a comprehensive profile of the fine motor abilities of 7 to 9-year-old children living in the Fitzroy Valley, including children with PAE and/or FASD. This aim was achieved through the following objectives:

1.6.1 General objectives

1. Develop a comprehensive profile of fine motor skills for children living in the Fitzroy Valley.
2. Compare the fine motor skills of children with and without PAE and/or FASD.
3. Determine if commonly used fine motor assessment tools are appropriate for use as part of a FASD diagnostic assessment.
4. Report the prevalence of moderate and severe fine motor impairment of children in the Fitzroy Valley.
5. Report, and discuss possible implications of, the proportion of children with PAE and/or FASD who have a significant fine motor impairment according to FASD diagnostic criteria used during the Lililwan Project.
6. Consider the implications of rates of fine motor impairment for existing therapy services for children in the Fitzroy Valley.

1.6.2 Specific objectives

Specific objectives were:

Fine motor skills (Chapter 4.1)

1. Assess and evaluate fine manual control (fine motor precision and fine motor integration) and manual coordination (manual dexterity and upper-limb coordination) using the BOT-2.
2. Compare fine motor skills of children i) without PAE; ii) with PAE but not FASD; and iii) with FASD.
3. Determine the prevalence of moderate (≤ 16th percentile) and significant (≤ 2nd percentile) fine motor impairments in the cohort.

Visual-motor integration (Chapter 4.2)

1. Assess and evaluate visual-motor integration, visual perception, and fine motor coordination skills using the Beery VMI.
2. Compare visual-motor integration, visual perception, and fine motor coordination, including types of errors and time to complete subtests, in children i) without PAE; ii) with PAE but not FASD; and iii) with FASD.
3. Determine the prevalence of moderate (≤ 16th percentile) and severe (≤ 2nd percentile) visual-motor integration, visual perception, and fine motor coordination impairments in children i) without PAE; ii) with PAE but not FASD; and iii) with FASD.

Graphomotor skills (Chapter 4.3)

1. Assess and evaluate pencil grasp; writing pressure; and ability to write their name and a short sentence, using clinical observation.
2. Assess and evaluate drawing abilities in terms of motor accuracy and body awareness, using the Miller Function and Participation Scales (Miller, 2006).
3. Assess and evaluate handwriting legibility in terms of percentage of letters and words formed correctly when writing their name and a short sentence, using the Evaluation Tool of Children's Handwriting (Amundson, 1995).
4. Compare graphomotor skills of children i) without PAE; ii) with PAE but not FASD; and iii) with FASD.

Prevalence of significant domains of fine motor or visual-motor integration (Chapter 4.4)

1. Report the proportion of children i) without PAE; ii) with PAE but not FASD; and iii) with FASD who had an impaired fine motor or visual-motor integration domain of impairment according to FASD diagnostic criteria used during the Lililwan Project.

Chapter 2 Systematic Literature Review

This chapter contains the peer-reviewed, published systematic literature review:

Doney, R., Lucas, B. R., Jones, T., Howat, P., Sauer, K., & Elliott, E. J. (2014). Fine motor skills in children with prenatal alcohol exposure or Fetal Alcohol Spectrum Disorder. *Journal of Developmental and Behavioral Pediatrics*, *35*(9), 598-609.

Further, more detailed literature reviews relevant to fine motor, visual-motor integration, and graphomotor skills are included in each of the published papers in the relevant results sections.

2.1 Abstract

Objective: Prenatal alcohol exposure (PAE) can cause Fetal Alcohol Spectrum Disorders (FASD) and associated neurodevelopmental impairments. It is uncertain which types of fine motor skills are most likely to be affected following PAE, or which assessment tools are most appropriate to use in FASD diagnostic assessments. This systematic review examined which types of fine motor skills are impaired in children with PAE or FASD; which fine motor assessments are appropriate for FASD diagnosis; and whether fine motor impairments are evident at both 'low' and 'high' PAE levels.

Method: A systematic review of relevant databases was undertaken using key terms. Relevant studies were extracted using a standardized form and methodological quality was rated using a critical appraisal tool.

Results: Twenty-four studies met inclusion criteria. Complex fine motor skills, such as visual-motor integration, were more frequently impaired than basic fine motor skills, such as grip strength. Assessment tools that specifically assessed fine motor skills more consistently identified impairments than those that assessed fine motor skills as part of a generalized neurodevelopmental assessment. Fine motor impairments were associated with 'moderate' to 'high' PAE levels. Few studies reported fine motor skills of children with 'low' PAE levels so the effect of lower PAE levels on fine motor skills remains uncertain.

Conclusion: Comprehensive assessment of a range of fine motor skills in children with PAE is important to ensure an accurate FASD diagnosis and develop appropriate therapeutic interventions for children with PAE-related fine motor impairments.

Prenatal alcohol exposure (PAE) can result in a range of lifelong neurological impairments in offspring termed Fetal Alcohol Spectrum Disorders (FASD).[1-3] Diagnoses on the spectrum include Fetal Alcohol Syndrome (FAS), which includes characteristic dysmorphic facial features and growth impairments; partial FAS, with some facial and growth impairments; and alcohol related neurodevelopmental disorder (ARND), in which individuals do not have facial or growth changes. All diagnoses have significant neurological impairments, which can include impaired cognition, executive function, memory, language, attention, social and adaptive skills, and motor skills.[2]

2.2 Introduction

PAE and motor skills

PAE can affect both fine and gross motor skills.[1] Early reports noted that infants with FAS had motor impairments[4], and subsequently orthopaedic and structural defects were also recorded, including clinodactyly (fixed laterally curved fifth finger), camptodactyly (fixed finger flexion), impaired upper limb pronation/supination, tapering of the distal phalanges, and resting and kinetic hand tremors.[5-7] In animal studies, PAE has been associated with impaired myelination of spinal and peripheral nerves,[8,9] and impaired motor coordination, response, speed, activity, reflexes, and tone.[10,11] Neuroimaging studies of individuals with PAE or FASD have identified damage to specific brain regions. Damaged regions which may affect motor skills include the cerebellum,[12] basal ganglia,[13] corpus callosum,[14] and hippocampus.[12] PAE can also damage neural circuits, including projections which extend into motor and premotor cortices.[15] There are limited studies of the motor cortex in relation to PAE. However, one study of adolescents with PAE concluded that observed motor impairments likely were due to damage to the motor cortex.[16]

Fine motor skills in FASD diagnosis

Fine motor (FM) skills require the use of small hand muscles and include speed, accuracy, control, coordination, dexterity, visual-motor skills, and eye-hand coordination.[17] FM skills are important in children because they facilitate increasing independence in self-care tasks such as dressing, eating, brushing hair, and cleaning teeth; academic skills including handwriting, drawing, and using scissors; and participation in play and social activities.[18] Parents and teachers of primary school aged children with FASD often report they have difficulty with many of these functional tasks.[19]

FASD diagnostic guidelines universally advise that motor skills should be assessed in children with PAE, but variation exists regarding whether FM skills are differentiated from gross motor (GM) skills, and the recommended assessment tools. The Canadian Guidelines recommend specific tools, such as the Movement Assessment Battery for Children (M-ABC), the Bruininks-Oseretsky Test of Motor Proficiency, and the Beery-Buktenica Developmental Test of Visual-Motor Integration.[2] The UW 4-Digit Diagnostic Code guidelines do not recommend specific assessment tools, but advise that they should be standardised and validated.[20] The Institute of Medicine (IOM) and Centers for Disease Control (CDC) guidelines offer no advice about which assessment tools to use.[1,3]

It has been suggested that generalised neurodevelopmental assessments may not detect subtle and specific impairments resulting from PAE.[21,22] Many neurodevelopmental assessment tools used to assess children with PAE are problematic in terms of FM assessment: some, such as the WISC-IV,[23] do not include a motor skills component; others, such as the McCarthy Scales of Children's Abilities,[24] provide an overall motor score which is a composite of FM and GM skills, which may mask specific areas of FM or GM impairment;[25] while others, such as the Griffiths Mental Development Scales,[26] assess only limited types of FM skills.

FM skills in children with PAE or FASD

Three systematic reviews have examined the relationship between PAE and motor skills. The first review concluded that only high levels of PAE (10-30 drinks/week) were associated with impaired motor function, although findings were not consistent between studies.[27] However, this study did not explore whether different types of motor skills were more likely to be impaired, or if FM skills were affected independently of GM skills. A second systematic review and meta-analysis found that GM skills were 2.9 times more likely to be impaired in children aged 0-18 years with 'moderate' to 'high' PAE compared to children without PAE, but this review did not include FM skills or lower levels of PAE within the scope of the review.[28] A third study of neurological impairments included 13 studies of 'visual and motor' skills and concluded that impairments were not associated with 'mild', 'moderate', or 'binge' PAE, but none of the included assessed children older than 5 years.[29]

Aims and hypotheses

Whilst FASD diagnostic guidelines recommend assessing motor skills in children with PAE, it is unclear which types of FM skills are most likely to be impaired or which assessment tools are best to use. It is also uncertain whether impaired FM

skills account for the functional difficulties reported by caregivers and teachers of children with FASD. This knowledge is essential to ensure accurate and timely FASD diagnosis, contribute to the development of a FASD neurological profile, employ objective measurement of FM skills over time, and develop therapeutic programs which promote independence by enhancing FM strengths and supporting areas of difficulty.

This systematic review examined FM skills in primary school aged children (4-12 years) with PAE or FASD, and aimed to:

1. Establish which types of FM skills are impaired following PAE;
2. Identify which FM assessment tools are commonly used with children with PAE or FASD; and
3. Investigate whether different levels of PAE are associated with FM impairments

Given the diversity of neural regions affected by PAE, we hypothesised that a range of FM skills would be impaired in children with PAE or FASD, especially those which are more complex and involve multiple neural regions and connectivity. We further hypothesised that FM impairments would be more evident amongst children with high levels of PAE, and that neurodevelopmental assessments which assessed FM skills as part of a generalised assessment battery would not identify FM impairments.

2.3 Methods

Literature Search

A search strategy was used to systematically search peer-reviewed journals in the Web of Knowledge, Ovid (Medline, PsychInfo, Maternity and Infant Care, Embase, and Amed), EBSCOhost (CINAHL), ProQuest, SciVerse, The Cochrane Library, Emerald, Informit, OT Seeker, and PEDRO databases. The following key words were combined to identify relevant articles: alcohol*, fetal, maternal, prenatal*, *in-utero* and fine motor. No articles were identified which specifically reported FM skills in relation to PAE, so search terms were broadened to include motor, neuro*, development*, sensor*, and visu*. Experts in the field were contacted to identify unpublished studies and reference lists of relevant articles were manually searched.

Inclusion Criteria and Selection Process

Full texts were obtained for articles which met the study inclusion criteria (Table 2.1). Included FM skills were: control, precision, speed, or accuracy; visual-motor

integration (VMI) or visuomotor precision; in-hand manipulation; hand grasp and release; manual dexterity or coordination; and foundational skills including praxis, grip strength, and finger tapping. Functional FM skills included pencil grasp and writing pressure; handwriting or drawing skills; and adaptive skills with a FM component such as tying shoelaces or doing up buttons. Studies were excluded if they only reported neuromuscular performance such as range of motion, reflexes, or skeletal deformities; GM skills (walking, running, strength, or balance); visual perception (acuity, convergence, tracking, and eye-blink conditioning); visual cognitive skills (visual discrimination, visual memory, form constancy, spatial skills, figure ground, or visual closure); or sensory processing skills.

Table 2.1

Inclusion and exclusion criteria

Attribute	Inclusion criteria	Exclusion criteria
Time	Any time until December 2012	January 2013 onwards
Language	English	All other languages
Design	RCTs Cohort studies Case-control studies	Descriptive studies Case studies Reviews
Publications	Original research Peer-reviewed journals	Non peer-reviewed articles Conference abstracts/ posters Post-graduate theses
Participants	Humanistic studies 4-12 years (all or some participants within this range)	Animal studies Babies, infants, adolescents, adults
PAE	FASD diagnoses PAE (including low-high, binge, any trimester)	Nil
FM Assessment	Direct assessment of skills by clinician/ therapist Published, commonly available, standardized assessments Assessments using standardized equipment e.g. dynamometer; mechanical finger tapper	Parental or teacher report Observational data, e.g. handedness; tremor Assessments not commonly available to clinicians (e.g. robotic or computerized tasks)
FM results	Reported separately to GM and other neurological results Statistical significance of results reported	Composite GM/FM outcomes (e.g. McCarthy Scales of Children's Abilities)

Note: RCT: randomized control trial; PAE: Prenatal Alcohol Exposure; FASD: Fetal Alcohol Spectrum Disorder; FM: fine motor; GM: gross motor

Data Extraction and Quality Assessment

There were 24 studies which met all the inclusion criteria (Table 2.1). These were summarized by two authors (RD, TJ) using a data extraction form (Appendix B). A 10-point critical appraisal tool (CAT) was developed based on STROBE and Health Evidence guidelines to assess the methodological quality and relevance (Table 2.2). The studies were independently rated by two authors (RD, TJ) and consensus agreement reached. Studies were classified as having either 'low' (0-4), 'moderate' (5-7), or 'strong' (8-10) methodological quality. Studies which quantified PAE levels as ounces of absolute alcohol/ day or week were converted to drinks/ day or drinks/ week (1oz absolute/ alcohol = 28.35g; 14g approximates 1 standard drink). A meta-analysis was not conducted due to the heterogeneity of FM skills and assessment tools, and the variability of PAE levels and FASD diagnoses between studies.

Table 2.2

Study quality and relevance

	Sample				Assessment				Bias		
Study	Age [a]	Defined sample [b]	PAE/ FASD [c]	Matching [d]	Size [e]	FM depth [f]	Psycho-metrics [g]	FM specificity [h]	Blinding [i]	Confounders [j]	Total
Adnams et al., 2001	✓	✓	✓	✓	×	×	×	×	✓	✓	6
Aragón et al., 2008	×	✓	✓	✓	×	×	✓	✓	×	✓	6
Aronson et al., 1985	×	×	×	✓	×	×	×	×	×	✓	2
Astley et al., 2009	×	✓	✓	✓	✓	×	×	✓	✓	✓	7
Barr et al., 1990	✓	✓	✓	×	×	✓	×	✓	✓	✓	7
Bay et al., 2012	✓	✓	✓	✓	×	×	✓	✓	✓	✓	8
Chiodo et al., 2009	✓	✓	✓	✓	×	✓	✓	✓	✓	✓	9
Coles et al., 1997	✓	✓	✓	✓	×	×	×	✓	✓	✓	7
Conry, 1990	×	✓	✓	✓	×	✓	✓	✓	✓	✓	8
Fried & Watkinson, 1990	✓	✓	✓	×	×	×	×	✓	✓	✓	6
Henry et al., 2007	×	×	✓	×	×	✓	✓	×	×	✓	4
Irner et al., 2012	×	×	✓	✓	×	×	×	×	×	✓	3
Janzen et al., 1995	×	✓	✓	✓	×	✓	✓	✓	×	✓	7
Jirikowic et al., 2008	✓	✓	✓	×	×	×	✓	×	✓	✓	6
Kooistra et al., 2009	✓	✓	✓	×	×	×	✓	✓	✓	✓	7
Korkman et al., 1998	✓	✓	✓	×	×	✓	✓	✓	×	✓	7
Laforce et al., 2001	×	✓	×	✓	×	×	×	✓	×	✓	4
Mattson et al., 1998	×	✓	✓	✓	×	✓	×	✓	×	✓	6
Mattson et al., 2010	×	✓	✓	×	×	✓	✓	✓	×	✓	6
Russell et al., 1991	✓	✓	✓	✓	×	✓	×	✓	✓	✓	8
Sowell et al., 2008	×	✓	✓	✓	×	×	✓	✓	×	✓	6
Uecker & Nadel, 1996	×	×	✓	✓	×	×	×	✓	×	✓	4
Vaurio et al., 2011	×	✓	✓	✓	×	✓	×	✓	✓	✓	7
Zhou et al., 2011	×	✓	✓	✓	×	×	×	×	×	✓	4

Note: ✓ criterion met; × criterion not met/ uncertain. [a] Entire sample aged 4-12 years. [b] Cohort studies: selection process provided; case control studies: inclusion/ exclusion criteria provided. [c] Method of PAE ascertainment provided (e.g. interview; review of medical records); or FASD diagnostic criteria provided. [d] Cohort studies: sample comparable to FM assessment normative data; or case control studies: matched cases and control groups. [e] Sample size/ power calculation; or justification provided for sample size (e.g. population study). [f] More than one FM skill reported, either using two different assessments; or one assessment which assesses multiple types of FM skills. [g] Psychometrics for assessment provided; or justification for assessment choice. [h] FM specific assessment (i.e. not part of general neuropsychological assessment). [i] Assessors blinded to PAE or FASD status at time of assessment. [j] Consideration given to potential confounding factors

Table 2.3

Fine motor assessments and outcomes

	Visual motor skills			Dexterity		Foundational FM skills				Functional FM skills	
Study	VMI	Eye-hand	VM precision	VMI	Eye-hand	Grip strength	Finger tapping	Praxis	Sensori-motor	Grapho-motor	Motor inhibition
Undefined; or low, moderate and high PAE											
Barr et al., 1990				+/-[1] GPT	+/-[2] WFMSB	- Dyna	+ HRNB				
Bay et al., 2012					- M-ABC						
Coles et al., 1997	+ BVMI										
Chiodo et al., 2009	+/-[3] PENTB			+/-[4] PPT							
Fried and Watkinson, 1990				- GPT							
Irner et al., 2012		+/-[5] GMDS									
Russell et al., 1991	- BVMI										- CATB
Moderate to high PAE; or pFAS/FAS											
Adnams et al., 2001		+ GMDS									
Aragon et al., 2008				- GPT							
Aronson et al., 1985		+ GMDS									
Janzen et al., 1995	+ BVMI			-[6] GPT							

	Visual motor skills			Dexterity		Foundational FM skills				Functional FM skills	
Study	VMI	Eye-hand	VM precision	VMI	Eye-hand	Grip strength	Finger tapping	Praxis	Sensori-motor	Grapho-motor	Motor inhibition
Jirikowic et al., 2008									+ NEPSY[b]		
Korkman et al., 1998	+ BVMI		- NEPSY[a]					+/-[7] NEPSY[c]			
Laforce et al., 2001				- PPT							
Mattson et al., 1998	+/-[8] BVMI			+/-[9] GPT							
Mattson et al., 2010	+ BVMI			+ GPT							
Sowell et al., 2008	+ BVMI										
Uecker and Nadel, 1996	+ BVMI										
Vaurio et al., 2011	+ BVMI			- GPT							
FASD (PAE levels unspecified)											
Astley et al., 2009	+/-[10] BVMI										
Conry, 1990	+/-[11] BVMI			+/-[12] DTLA		+/-[13] Sphyg	+/-[4]				
Henry et al., 2007								- PEEX		- PEEX	
Kooistra et al., 2009					+ M-ABC						
Zhou et al., 2011			+ NEPSY[a]								

Note. + = Significant difference between children with PAE/FASD and a control group; +/- = Both significant and non-significant outcomes, e.g. between FAS and controls, but not FAE and controls; or for the dominant hand but not the non-dominant hand; - = Non-significant difference.
Assessments: BVMI: Beery Buktenica Test of Visual Motor Integration; CATB: The Cincinnati Autonomy Test Battery (Draw a line slowly test); Dyna: Dynamometer; DTLA: Detroit Tests of Learning Aptitude (The motor speed and precision test); GMDS: Griffiths Mental Development Scales (Eye and hand coordination subscale); GPT: Grooved pegboard test; HRNB: Halstead Reitan Neuropsychological Battery (Finger tapping test); M-ABC: Movement Assessment Battery for Children (Fine motor subarea); NEPSY: Developmental Neuropsychological Examination (a Visuomotor precision subtest; b Sensorimotor domain (includes visuomotor precision; finger tapping; imitating hand positions; and manual motor series (praxis) subtests); c Manual motor series (dynamic praxis) and Imitating hand positions (kinesthetic praxis) subtests); PENTB: Pediatric Environmental Neurobehavioral Test Battery (Visual-motor integration test); PEEX: Pediatric Early Entry Examination or Pediatric Examination of Educational Readiness at Middle Childhood (Fine motor/graphomotor test); PPT: Purdue pegboard test; Sphyg: Sphygmomanometer; WFMSB: Wisconsin Fine Motor Steadiness Battery (Vertical/ Horizontal Groove Board; Grooved Maze Board; Resting Steadiness Hole Board)

[1] Significant (p = .018) impairments in time to complete for children with early pregnancy PAE (time to complete); peg drop count outcomes and mid-pregnancy PAE outcomes not reported

[2] Significant (p = .011; p = .000) number of errors and latency to correct for children with early pregnancy PAE; mid-pregnancy PAE outcomes not reported

[3] Significant (p < .025) for 'at-risk alcohol measure' only

[4] Significant (p < .025) for 'at-risk alcohol measure' only

[5] Significant (p = .03) for children with PAE 'at time of birth', but non-significant (p = .30) for children with PAE 'during pregnancy' (NB comparisons for 'during pregnancy' made between children with PAE/ no polydrug; polydrug/ no PAE; and polydrug/ PAE groups only)

[6] Non-significant number of pegs placed in 1 minute, left (p = .011) or right (p = .047) hands; authors have used conservative p values (p <.01)

[7] Significant (p < .0001) for dynamic praxis; non-significant for kinesthetic praxis

[8] Significant (p < .001) between FAS and controls; non-significant between PAE and controls, or FAS and PAE

[9] Significant (p < .01) non-dominant hand; non-significant dominant hand

[10] Significant (p < .05) between controls, pFAS, FAS, neurodevelopmental disorder/alcohol exposed, and static encephalopathy/alcohol exposed (SE/AE) ; non-significant between pFAS/FAS and SE/AE

[11] Significant (p < .001) between controls and FAS, and FAS and FAE; non-significant between controls and FAE

[12] Significant (p = .002) between controls and FAS; non-significant between controls and FAE; and FAS and FAE

[13] Significant (p < .001) dominant and non-dominant hands between controls and FAS, and controls and FAE; non-significant between FAS and FAE

[14] Significant (p = .005) dominant hand between controls and FAS, and controls and FAE; non-significant between FAS and FAE. Significant (p = .006) non-dominant hand between controls and FAS; non-significant between controls and FAE, and FAS and FAE

2.4 Results

Literature Search

We identified 6,259 studies after removal of duplicates (*Figure 2.1*). An additional 10 papers were identified by manually searching reference lists. No unpublished papers were identified from experts in the field. A total of 6,173 papers were excluded because they did not meet inclusion criteria, and a further 72 were excluded for not meeting FM-related criteria. This resulted in 24 studies eligible for inclusion.

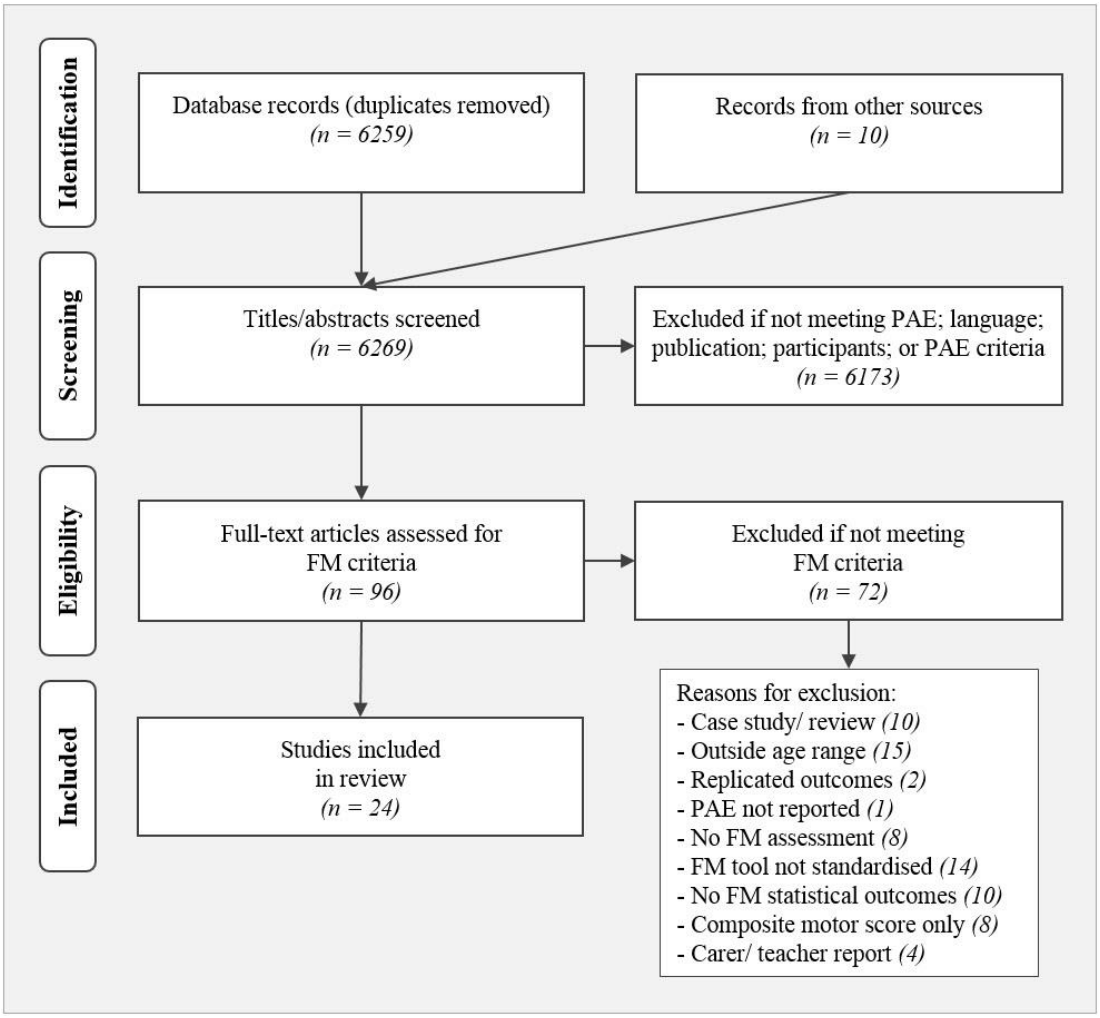

Figure 2.1 Flowchart for article inclusion

Study Characteristics

Characteristics of the 24 included studies, including PAE levels and FASD diagnoses when provided, are outlined in Appendix B. Included studies were published between 1985 and 2012. Sample sizes ranged from 10 to 685, and included eight small (n < 50), seven medium (n = 50-100), and nine large (n > 100) studies.

Risk of Bias and Quality Assessment

Studies were rated using the CAT (Table 2.3). Most of the studies had moderate (58.3%) or strong (16.7%) methodological quality. Half of the studies (50%) blinded assessors to PAE levels or FASD diagnoses as a strategy to reduce bias. All studies discussed potential confounding factors, but variation existed regarding which confounding factors were considered relevant, and whether these were controlled for statistically. The majority of studies used at least one assessment tool which specifically assessed motor skills, as opposed to generalized neurodevelopmental assessments with a FM component (75.0%), but less than half of the studies assessed more than one type of FM skill (41.7%).

FASD and PAE

Most studies included children with FASD diagnoses (70.8%), but seven of these included only children with the more severe diagnoses of pFAS or FAS (29.2%). Only three studies reported FM outcomes for children with 'no/low' PAE <0.14 oz/ day (<2 drinks/ week);[30] 'low' PAE (1- 4 drinks/ week);[31] or 'light/ moderate' PAE (0-1 oz/ day (0-14 drinks/ week).[32] A further three studies included children with 'low' PAE but did not stratify FM outcomes by PAE levels.[33-35] Thirteen studies only included children with 'moderate' or 'high' PAE, or children with pFAS or FAS (who likely had 'moderate to high' PAE as these diagnoses include the most severe effects of PAE). Not all studies quantified PAE levels, but those that did variously defined 'moderate' and 'high' PAE as: 'moderate'(5-8 drinks/ week);[31] 'moderate to high/ heavy' (>10drinks/ week);[36] >0.14 oz/day (or >2 drinks/ week);[30] 'high/ heavy' (9-14 drinks/ week);[31] 0.5-1.5 oz/day (or 7-21 drinks/ week);[33] >13 or >14 drinks/ week *or* >4 drinks/ occasion;[37,38] (1-3.5 oz/day (or 14-49 drinks/ week);[32] and 'very heavy' (>3.5 oz/ day (or >49 drinks/ week).[32] A further four studies included children with a range of FASD diagnoses but did not specify PAE levels.[39-42]

Fine Motor Assessments and Outcomes

The most common types of FM skills assessed were visual-motor skills and manual dexterity. Researchers used a range of different FM assessment tools (Table 2.3).

1. Visual-motor skills (n = 17). Studies of visual-motor skills included visual-motor integration (VMI), eye-hand coordination, and visuomotor precision. Assessments included a combination of drawing and functional tasks such as threading beads or posting coins.

(i) Beery-Buktenica Developmental Test of Visual-Motor Integration (Beery VMI)

(*n* = 11). The Beery VMI[43] was the most commonly used assessment in this review. The Beery VMI requires children to copy increasingly complex geometric shapes. All but one study[32] which used this assessment tool reported significantly impaired VMI skills, including in children with a range of FASD diagnoses.[38,39,44,45,46] One study identified VMI impairments in children with 'moderate to high' PAE (>10 drinks/ week) in the first trimester; first and second trimesters; and all three trimesters.[36] In contrast, another study with 5-16 year old children with FAS had impaired VMI skills, but these findings did not extend to children with 'heavy' PAE (unquantified levels) but no FASD diagnosis.[47] However, a subsequent larger, multi-site study by the same researchers did identify significant VMI impairment in children with 'heavy' PAE (>13 drinks/ week; or >4 drinks/ occasion) but no FASD diagnosis.[37] Only one study did not report VMI impairments when these skills were assessed using the Beery VMI.[32] This study included 6 year old children with 'none'; 'light to moderate' (0-2 drinks/ day); 'heavy' (2-7 drinks/ day); or 'very heavy' (>7 drinks/day) PAE early in pregnancy. In contrast to other studies, PAE in this study included any alcohol consumption in the year leading up to pregnancy, as the authors considered that this was the most accurate proxy for PAE early in the first trimester.

(ii) Pediatric Environmental Neurobehavioral Test Battery (PENTB): Visual-motor integration test (*n* = 1). The PENTB includes a VMI component, which consists of copying line drawings.[48] Only one study assessed VMI skills using the PENTB.[34] When VMI skills were analysed using the newly developed 'at-risk alcohol exposure' measure, impairments remained significant after adjustment for potential confounding factors.

(iii) Griffiths Mental Development Scales (GMDS): Eye and hand coordination subscale (*n* = 3). Three studies used the eye and hand coordination subscale of the GMDS, which assesses eye-hand coordination by having the child copy geometric figures, as well as functional tasks such as threading beads.[26] Two studies found significant eye and hand coordination impairments, including in children with FAS,[21] and children of alcohol-dependent mothers.[49] A third study of children aged 3-7 years, at least half of whom had 'high' PAE (>15 drinks/ week), identified significant eye-hand coordination impairments.[35]

(iv) A Developmental Neuropsychological Assessment (NEPSY): Visuomotor precision subtest (*n* = 3). The NEPSY visuomotor precision subtest assesses FM speed and accuracy through a timed pencil and paper maze.[50] One study found no

significant visuomotor precision impairments in 5-9 year old children with 'moderate to high' PAE (>10 drinks/ week) in either the first; first and second; or all trimesters.[36] In contrast, a second study found significant visuomotor precision impairments in 6-12 year old children with FASD. PAE levels were unspecified in this study.[42] A third study used the NEPSY's sensorimotor domain, which includes visuomotor precision, finger tapping, imitating hand positions, and manual motor series subtests, to assess 5–8.5 year old children with FASD, of whom 60% had 'high' PAE.[51] This study did not report visuomotor precision findings independently of the other subtests, but found that the overall sensorimotor domain scores were significantly impaired.

2. FM dexterity (speed and coordination) (*n* = 13). FM dexterity was reported in 13 studies using four different assessment tools.

(i) Pegboard tests (*n* = 9). Pegboard tests assess a range of FM skills, including manual dexterity, precision, speed, and accuracy. Nine studies used pegboard tests and reported varying outcomes. Assessment tools included the Purdue Pegboard Test, the Grooved Pegboard Test, and pegboard tests that comprise part of generalized neurodevelopmental assessment tools, such as the Wisconsin FM Steadiness Battery (WFMSB). Three studies reported non-significant results in children with pFAS or FAS (unspecified PAE levels);[52]; FAS or Fetal Alcohol Effects (a term previously used to describe FASD diagnoses other than FAS) with 'heavy' PAE (unquantified levels);[53] and children with 'heavy' PAE (>14 drinks/ occasion, or >14 drinks/ week).[38] One study reported non-significant results amongst children with 'none/light' PAE (<0.14 oz of absolute alcohol per day, or <2 drinks/ week) compared to children with 'moderate/heavy' PAE (>0.14 oz of absolute alcohol per day, or >2 drinks/ week).[30] Another study reported non-significant results in left or right hands in children with FAS born to mothers with alcohol dependency, although conservative *p* values were used which may have been considered significant in other studies.[54] At 4 years, children with PAE (7-21 drinks/ week) early in pregnancy, and children who were exposed to an average of more than 1 drink per day throughout pregnancy, took longer to complete the pegboard but did not make more errors than children without PAE.[33] A different study examined whether different ways of quantifying PAE affected the findings.[34] This study found that a dichotomous measure of 'at-risk alcohol exposure' was associated with fewer pegs being placed accurately, but these findings were not significant when other measures of PAE were used. A further study reported some significant results when comparing different FASD diagnoses, but findings were not consistent between dominant and non-dominant hands.[47] In contrast, a larger, multi-site study by the same authors found participants with FAS and 'heavy' PAE

(>13 drinks/ week; or >4 drinks/ occasion) had impairments in both their dominant and non-dominant hands.[37]

(ii) Movement Assessment Battery for Children (M-ABC): Manual dexterity subarea

(*n* = 2). The M-ABC manual dexterity component includes items such as posting coins, threading beads, and completing a paper and pencil drawing trail.[55] The M-ABC was used in two studies to assess manual dexterity skills and reported contrasting results. The first study found no significant manual dexterity impairments in 5 year olds stratified by different PAE levels.[31] This study mostly included 'low to moderate' PAE (≤8 drinks/ week), with only 1.6% (*n* = 11) of the sample being exposed to more than 9 drinks per week, and no children being exposed to more than 14 drinks per week. The second study found manual dexterity skills were significantly impaired amongst 7-10 year olds with various FASD diagnoses, as well as children with ADHD.[41] This study did not report levels of PAE amongst the children with FASD.

(iii) Detroit Tests of Learning Aptitude (DTLA): The timed motor and precision test

(*n* = 1). One study[39] used the DTLA Timed Motor and Precision Test which assesses FM speed and precision by rapidly drawing crosses in circles of decreasing size.[56] This study found significant differences in FM speed and precision skills amongst children aged 5-18 years with FAS, but not children with FAE. PAE levels were not defined in this study.

(iv) Wisconsin FM Steadiness Battery (WFMSB) (*n* = 1). One study used the FM tests from the WFMSB[57] to assess 4 year old children with no PAE; children with PAE only in early pregnancy; and children with PAE throughout pregnancy.[33] This study reported that the children with 'moderate to heavy' PAE (7-21 drinks/ week) early in pregnancy made significantly more errors and were slower to self-correct their errors compared to children with no PAE. These impairments were significant for children with PAE early in pregnancy as well as children with PAE (average ≥1 drink/ day) throughout pregnancy. These difficulties remained significant after adjustment for IQ and other confounding factors.

3. Foundational FM skills: grip strength, finger tapping, and praxis.

(i) Grip strength (*n* = 2). Two studies reported grip strength findings with mixed results. The first study identified no significant differences in grip strength amongst 4 year olds with PAE early or throughout pregnancy.[33] PAE levels in this study ranged from 0.5 to 1.5 oz of absolute alcohol/ week (7-21 drinks/ week). The second study identified that 5-18 year olds with FAS or FAE had significant impairments when

using both their dominant and non-dominant hands.[39] PAE levels were not defined for the children in this study.

(ii) Finger tapping (*n* = 3)*.* Finger tapping was assessed in three studies with mixed results. Two studies used a mechanical finger tapper to record how many times the child could tap their finger while holding the other fingers stationary, and a third study measured finger tapping as part of the NEPSY sensorimotor domain. One study reported a significant finger tapping impairment in 5-18 year old Native Americans with FAS or FAE in both their dominant and non-dominant hands.[39] In contrast, the second study found 4 year old children with 'moderate to heavy' PAE in early pregnancy, but not mid-pregnancy, had impaired finger tapping skills.[33] A third study assessed finger tapping skills in 5-8.5 year old children with FASD, of whom 60% had 'high' PAE (unquantified levels), but finger tapping results were not reported independently of other tasks in the sensorimotor domain.[50]

(iii) Praxis (*n* = 2)*.* One study assessed kinesthetic and dynamic praxis using the 'Imitating Hand Positions' and 'Manual Motor Series' subtests from the Sensorimotor Domain of the NEPSY.[50] These tests require imitation of static hand positions and rhythmic hand movements. This study found that dynamic praxis, but not kinesthetic praxis, was affected in 5-9 year old Finnish children with 'moderate to high' PAE (≥10 drinks per week).[36] Dynamic praxis was impaired in children who had been exposed to 'moderate to high' PAE in the first; first and second; and all three trimesters of pregnancy.

(iv) Combined foundational FM skills (*n* = 2)*.* Two studies reported combined outcomes of different types of foundational hand skills. The first study reported outcomes from the NEPSY sensorimotor domain, which includes dynamic and kinesthetic praxis, finger tapping, and visuomotor precision.[51] These authors found that children with FASD (60% with 'high' levels of PAE) had impaired sensorimotor skills. A second study assessed FM skills in 6-16 year old children with FASD, who had also experienced significant trauma and trauma, using the Pediatric Early Elementary Examination (PEEX-2) or the Pediatric Examination of Educational Readiness at Middle Childhood (PEERAMID-2).[40] Tasks included lateral preference, imitative finger movements, finger tapping, and sequential finger opposition. These authors did not identify significant differences between children with or without FASD. PAE levels were not defined in this study.

4. Functional FM skills. Two studies included assessment of functional FM skills as part of a generalized neurodevelopmental assessment and neither found significant

impairments. The first study assessed graphomotor skills using the PEEX-2 or the PEERAMID-2 with 6-16 year old children who had experienced trauma, and compared outcomes between children with and without FASD. Tasks included pencil control, pencil speed, and writing the alphabet.[40] A second study used the 'Draw a Line Slowly' test from The Cincinnati Autonomy Test Battery to assess motor inhibition skills. No impairments were identified in 6 year old children with 'none'; 'light to moderate' (0-2 drinks/ day); 'heavy' (2-7 drinks/ day); or 'very heavy' (>7 drinks/day) levels of PAE early in pregnancy.[32]

2.5 Discussion

This review identified 24 studies which assessed FM skills in primary-school aged children with PAE or FASD. A range of FM skills, including visual-motor skills (VMI, visuomotor precision, and eye-hand coordination), manual dexterity (speed, coordination, and precision), foundational FM skills (grip strength, finger tapping, and praxis), and graphomotor (handwriting, drawing, and motor inhibition) skills were assessed using a variety of FM-specific and generalized neurodevelopmental assessments.

Complex FM skills, such as VMI, were consistently impaired in children with 'moderate to high' PAE, and with pFAS or FAS diagnoses. These findings support the rationale that complex skills, which are controlled by several neural regions and involve multiple neural pathways, are more likely to be impaired following PAE.[58] VMI skills were impaired in most studies which used the Beery VMI, but findings were less consistent when visual-motor skills were assessed using generalized neurodevelopmental assessments such as the PENTB or GMDS. It may be that generalized neurodevelopmental assessments do not assess visual-motor skills in sufficient detail, or alternatively that the constructional visual-motor skills are less sensitive to PAE than graphomotor visual-motor skills. VMI deficits were evident in both young children and adolescents, and also across different trimesters of PAE exposure, making this an important FM skill to assess in children with PAE for both diagnostic and therapeutic purposes.

Pegboard tests were used to assess manual dexterity, precision, speed and coordination, which are also relatively complex FM skills. The Purdue Pegboard Test requires placement of pegs into round holes, while the Grooved Pegboard Test requires pegs to be rotated for accurate placement, and thus may be considered a more complex task. However, despite the differences in complexity, studies which used pegboard tests as a FM outcome measure did not consistently identify

impairments, despite most studies only including children with ‘moderate to ‘high’ PAE, who would be most likely to show impairments. It is possible that the pegboard tasks may be too simple to adequately detect subtle and complex FM impairments resulting from PAE.

Few studies investigated foundational FM skills, such as grip strength, finger tapping, praxis, and kinaesthesia, and each used different assessment tools, so outcomes were difficult to compare between studies. Only two studies reported grip strength and finger tapping skills, which are relatively basic FM skills, and reported varying results.[33,39] The contrasting outcomes may be due to age differences of the children in each study, which is consistent with the theory that PAE-related impairments become more pronounced with maturity.[59] While it is important to assess foundational FM skills in individual children with PAE as they may be affected and therefore warrant therapeutic intervention and support, evidence suggests that they do not provide a reliable marker for FASD presentation and diagnosis.

Only two studies reported functional outcomes of FM skills, and both used assessments of handwriting or drawing as a small part of a generalized neurodevelopmental assessment.[32,40] One study included children with exposure to trauma, and compared outcomes between those with and without FASD with undefined levels of PAE.[40] The second study included children with various levels of PAE, which included alcohol consumption in the year prior to pregnancy as a proxy measure for PAE early in pregnancy.[32] The demographic and methodological differences between these two studies make it difficult to compare outcomes, but neither study identified impaired graphomotor (handwriting or drawing) skills.

Only three studies stratified FM outcomes specific to ‘low’ PAE, and these studies did not identify FM impairments. ‘Low’ PAE was variously defined as <2 drinks/ week;[30] 1- 4 drinks/ week;[31] and 0-14 drinks/ week in the year leading up to pregnancy (as a proxy measure for early first trimester PAE).[32] Each of these studies reported different types of FM skills (VMI; motor inhibition; and manual dexterity), and each used different assessment tools, so outcomes were difficult to compare. The effect of ‘low’ PAE on specific type of FM skills remains uncertain.

Studies of functional FM skills which did not meet inclusion criteria

Several other studies of functional FM skills were identified that did not meet inclusion criteria for this review. However, impaired functional FM skills are frequently reported by parents and teachers of children with FASD, and these studies warrant mentioning. Caregivers of 5-8 year old children reported difficulties with adaptive function, many of

which require FM skills, including name writing, fastening clothing, and brushing teeth.[51] In another study, only 7% of the children with FAS had adaptive fine motor skills in the 'adequate' range, with the remainder scoring in the 'low' or 'moderately low' range.[60] One study assessed handwriting abilities in 20 primary school-aged children with FASD and found that they scored well-below average in handwriting legibility, speed, and visuomotor precision. This was an exploratory descriptive study and no control group was used for comparison.[61]

Neuroanatomical explanations for FM impairments

Damage to the cerebellum,[12] basal ganglia,[13] corpus callosum,[14] and parietal lobes[12] may account for some FM impairments. The cerebellum is commonly associated with balance, but also incorporates information from the visual system to control oculomotor function[62] which may contribute towards VMI impairments. The basal ganglia modulates emotional responses, but also has connections to motor areas, including in the frontal cortex and thalamus. Neuroimaging studies have shown that the corpus callosum may be reduced, misplaced, or missing in individuals with FASD, [14] and the role of the corpus callosum in relaying communications between hemispheres may explain bimanual coordination and manual dexterity deficits. One study identified that children with FASD had corpus callosum damage which may have resulted in difficulties with complex VMI tasks which required inter-hemispheric interaction.[63] The parietal lobe is involved with sensorimotor integration and visual perception, and damage to these regions may also contribute to VMI deficits observed in children with FASD.[64] Further, neural myelination can also be damaged by PAE,[46] which may explain why more complex FM skills – which require input from multiple areas as well as effective connectivity between regions – are more likely to be affected following PAE.

Limitations

This review had several limitations. Studies had significant methodological differences regarding PAE levels and FASD diagnoses, which hindered the comparability of FM outcomes. Studies were included only if they used standardized assessments commonly available to clinicians. Consequently, novel assessments of FM skills, such as computer and robotic-based studies, were excluded, as were observational assessments, such as finger-nose touching, tremors, and finger localization. Some studies assessed clock and person drawing, and although these tasks require FM skills, they were excluded because outcomes were reported as visual-perceptual and visual-spatial skills.[49,64] The comparability of outcomes

between studies was problematic because of the varying requirements of FM skills and assessment tools used as outcome measures, and also the use of different sets of normative data, comparison groups, and outcome criteria. For example, some studies used unmatched or matched control groups as a comparison, while others compared outcomes to varying sets of normative data. Studies which used pegboard tests variously reported outcomes based on dominance, laterality, or a combined score. More than half of the studies included children older or younger than the target age range of 4-12 years. This reduced certainty that the conclusions are specific to primary school aged children, although FM impairments were still evident in studies that included younger children and adolescents. As with all systematic reviews, there is a risk of positive publication bias. This risk was minimized by including studies which reported a range of neurodevelopmental outcomes, not just studies which reported FM skills, and by contacting experts in the field to identify unpublished studies.

Future Directions

FASD diagnostic guidelines should be updated to advise clinicians that a range of FM skills, particularly those which are more complex, may affected by PAE, and that these should be assessed separately to other motor skills using FM-specific assessment tools. Future studies should clarify what degree of FM impairment should contribute towards a FASD diagnosis. Establishing whether there is a relationship between specific FM skills and functional performance at school and home in children with PAE would be beneficial to those involved in FASD diagnosis, as well as clinicians, caregivers, and teachers of children with PAE.

Conclusion

This review identified that FM skills are impaired in children with ‘moderate to high’ PAE, and children with a pFAS or FAS diagnosis. It remains uncertain if ‘low’ PAE affects FM skills, because few studies reported FM skills in children with PAE, and those that did assessed different types of FM skills and each used different assessment tools. Consistent with our hypothesis, complex FM skills, such as VMI, were more likely to be impaired following PAE than basic FM skills such as grip strength. Further, specific FM assessment tools, such as the Beery VMI, were more likely to detect impairments than generalized neurodevelopmental assessments. However, pegboard tests, although they are a specific FM assessment tool, and manual dexterity is a relatively complex FM skill, did not consistently detect manual dexterity impairments. It is uncertain whether pegboard tests are too basic to detect

manual dexterity impairments resulting from PAE, or if this type of skill is not affected by PAE. In all likelihood, it is probably a combination of both of these factors. However, the heterogeneity of the FM skills assessed and variability amongst assessment tools make it difficult to draw definite conclusions. Comprehensive assessment of FM skills in children with PAE or FASD is essential to contribute towards an accurate FASD diagnosis, and inform therapeutic interventions which support specific areas of FM difficulties and enhance individual strengths.

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Chapter 3 Research Methods

This chapter provides an overview of the development and implementation of the Lililwan Project, including for FASD diagnostic and fine motor impairment criteria. Additional information related to each of the fine motor assessment tools, and relevant statistical analyses, is provided in the results chapter.

The Lililwan Project provided both a neurodevelopmental and FASD diagnostic clinic and a research study (Fitzpatrick et al., 2012). It was the first comprehensive health and neurodevelopmental evaluation of a population of Australian Aboriginal children, and the first population-based study of FASD prevalence in Australia.

3.1 Development of the Lililwan Project

Around the time that alcohol restrictions were introduced to Fitzroy Crossing (in 2007), local leaders began to learn about the harmful effects of PAE (Australian Government, 2013). They had noticed that many of the children in their communities had learning and behavioural problems, and were concerned that these deficits may have been the result of women consuming alcohol during pregnancy. At the time, the level of community awareness regarding the harmful effects of PAE and potential FASD were low. Subsequently, they were concerned about the impact of PAE and FASD on children, families, and on the continuation of their culture. June Oscar, a local Bunaba elder and CEO of Marninwarntikura Fitzroy Women's Resource Centre, stated (World Health Organization, 2014, np):

> FASD is a tragedy that somehow transcends other aspects of grief and trauma. Here is innocent young life, the future of our people and all that goes with it – our culture, our language, deep knowledge of our creation and the laws of our country – being born into this world with brains and nervous systems that are so impaired that life for that person, from birth to death, is cruelly diminished.

However, at the time there was limited evidence about the prevalence of FASD in Australia. National Australian paediatric reports showed FAS (the most severe diagnosis on the FASD spectrum) had a prevalence of 0.06 per 1,000 births (Elliott, Payne, Morris, Haan, & Bower, 2008), with higher rates in Indigenous populations (Elliott, Payne, Morris, Haan, & Bower, 2008; Harris & Bucens, 2003). These rates were thought to be an underestimate due to a lack of systematic screening and reporting (Bower, Silva, Henderson, Ryan, & Ryan, 2000). Fitzroy Valley leaders

were certain that rates were high in their communities, and that support was urgently needed for affected children and families.

A ‘circle of friends’ was established in the Fitzroy Valley, which included community members and representatives from local health and education departments. In 2009, following extensive community consultation, Nindilingarri Cultural Health Services and Marninwarntikura Women’s Resource Centre approached The George Institute for Global Health and the Discipline of Paediatrics and Child Health at The University of Sydney Medical School to form a partnership. The Marulu Strategy was developed, with the overarching aim of overcoming FASD and early life trauma in the Fitzroy Valley (Australian Government, 2013; Latimer et al., 2010). ‘Marulu’ is a local Bunaba word, meaning ‘precious, worth nurturing’. Early life trauma was included as part of the strategy in recognition of the multiple and sometimes extreme experiences of historical and current trauma experienced by many families in the Fitzroy Valley, including loss of land and culture, the experiences of the stolen generation, and ongoing effects of poverty. Early life trauma is often intertwined with issues related to alcohol misuse, and can exacerbate the detrimental effects of excessive alcohol consumption, including in children with FASD (Henry, Sloane, & Black-Pond, 2007; Hyter, 2007).

The Lililwan Project formed part of the Marulu Strategy, and aimed to determine the prevalence of FASD in a cohort of children in the Fitzroy Valley as well as to identify children and families needing support services. ‘Lililwan’ is a Kimberly Kriol word, meaning ‘all the little ones’.

3.2 The Lililwan Project procedures

Communities, families, and women of the Fitzroy Valley were consulted extensively before, during, and after the Lililwan Project, and were almost universally in support of the Lililwan Project. Mick Gooda, the Australian Aboriginal and Torres Strait Islander Commissioner, commended the Lililwan Project for being ‘a genuine partnership, one where research is done with the community, and not just about the community’ (Latimer et al., 2010). The Lililwan Project chief investigators were aware of the potentially stigmatising effects of the results on the people of the Fitzroy Valley. Hence, they were cognisant of ensuring any media coverage emphasised that FASD is not an ‘Aboriginal problem’. They also acknowledged that the women in the Fitzroy Valley showed courage by addressing the issue of FASD in their communities for the benefit of affected children. Accordingly, they were keen not to apportion blame or shame to the women.

The Lililwan Project aimed to assess all children born in 2002 or 2003 who were living in the Fitzroy Valley (Fitzpatrick et al., 2012). The children were aged between 7 and 9 years of age at the time of assessment. These age cohorts were chosen because: i) the children were old enough to tolerate the rigorous neurodevelopmental testing required for accurate FASD assessment; ii) some FASD-related deficits, such as executive function, become more pronounced with developmental maturity; iii) the children were young enough to benefit from therapeutic interventions; and iv) these age cohorts were born prior to the community alcohol restrictions, and thus likely represented some of the most alcohol affected age groups.

In 2010, parents and caregivers of eligible children were approached by local leaders to discuss the goals and requirements of the project. Families who consented for participation (*n* = 137, 95% participation) completed an in-depth verbal questionnaire with local 'Community Navigators', local Aboriginal people who ensured cultural safety of assessments and procedures. Aside from prenatal alcohol exposure (PAE), families also provided information related to a range of pre- and post-natal health, developmental, and socioeconomic circumstances which may have affected each child's neurodevelopment (Fitzpatrick et al., 2013).

In 2011, the children completed multi-disciplinary health and neurodevelopmental assessments with a multidisciplinary team consisting of an audiologist, occupational therapist (the doctoral candidate), ophthalmologist, paediatrician, physiotherapist, psychologist, and speech pathologist to develop a neurodevelopmental profile for each child and determine if they met FASD diagnostic criteria (Fitzpatrick et al., 2012). Children were assessed in their local communities or schools. Children completed approximately six hours of testing conducted over two days to minimise fatigue. Clinicians were cognisant of the potential for exhaustion in children due to rigorous testing. Assessments were timetabled in a way to be least burdensome. For example, the psychological testing was often completed in the morning, followed by the paediatrician evaluation in the afternoon which did not require cognitive responses from the child. The assessments completed by the occupational therapist, speech pathologist, and physiotherapist were designed to be perceived by children as fun. As such, they were arguably less arduous than the demands of school classes. The assessments were conducted during school hours. The six hours of assessment included the assessments completed by the doctoral candidate which form the basis of this thesis.

Following assessment, clinicians conducted case conferences to compile data from neurodevelopmental assessments and determine if the child met FASD diagnostic criteria. Clinicians were blinded to each child's social and developmental history, including PAE, during assessment and case conferencing phases, to minimise bias, and were made aware of these details only once consensus had been reached regarding neurodevelopmental function and resultant FASD diagnosis (if any). Case conferences lasted for approximately one to three hours per child. Feedback was provided to families, including information about FASD if the child was diagnosed. Families whose child was diagnosed with FASD were provided the option of receiving support from an Aboriginal woman and FASD educator, along with referral to the local social worker. Following case conferences, a comprehensive report and individual management plan was compiled and, with the family's permission, provided to the child's school and local health service.

The Lililwan Project posed several logistical and clinical challenges. Many communities in the Fitzroy Valley are inaccessible during the Wet Season (October to April), so assessments had to be completed between May and September. Night temperatures during the dry season in desert communities approached zero degrees Celsius, but by September the day temperatures were 40 degrees and higher. As many communities are located hundreds of kilometres from the town of Fitzroy Crossing, some along poorly maintained gravel roads, the Lililwan Project team camped in communities for several weeks at a time to complete assessments. Many families in the Fitzroy Valley move between communities, making it difficult sometimes to locate children for assessment and families for feedback. Cultural issues also had to be negotiated. For example, if a community member had died and the community was in 'sorry business', the team could not enter the community. Death of a community member meant their name could not be spoken or used by anyone of the same name, creating some confusion in accurately identifying children. Two Lililwan multidisciplinary team members were known by pseudonyms for part of the project because their names were the same as people who had passed away.

In addition to logistical challenges, there were clinical challenges. No neurodevelopmental assessment tools existed for use with Australian Aboriginal children, especially those living in a remote context with diverse language and cultural backgrounds. This can potentially invalidate the assessment procedures, content or the normative data used for comparison. It was a critical issue, because FASD diagnosis relies on determining the performance of each child in comparison to cut-off points (see 3.4 Definition of a significant neurodevelopmental impairment).

Clinicians chose assessment tools that were least likely to be affected by language or cultural factors and interpreted results with caution. For example, a non-verbal assessment of cognition (Universal Non-verbal Intelligence Test: UNIT) (Bracken & McCallum, 1998) was used by the psychologist due to the high levels of hearing impairment in the cohort, and because many children spoke English as a second language. Information was sought from families and teachers about each child's performance in home and school settings to compliment and validate outcomes from assessment tools, and to determine if they had concerns about any aspects of the child's development. Observations were made of the child's functional performance in their everyday environments; these observations were facilitated by the Lililwan Project team staying and participating in communities for extended periods of time. A detailed study protocol outlining assessment tools and procedures has been published (Fitzpatrick et al., 2012).

3.3 FASD diagnostic criteria

Australian guidelines for diagnosing FASD did not exist at the time of the Lililwan Project. Existing international guidelines were considered for their suitability for use in the Lililwan Project. They included those published by the American IOM (Hoyme et al., 2005), Canadian Guidelines for FASD Diagnosis (Chudley et al., 2005), the US CDC (Bertrand et al., 2004), and the University of Washington 4-digit FASD Diagnostic Code (Astley & Clarren, 2000). The Canadian FASD Diagnostic Guidelines (here-after called 'The Canadian Guidelines') were deemed most appropriate for use in the Lililwan Project. However, minor modifications were made to suit the resources available in the Lililwan Project, including the availability of clinicians, timeframe available to assess each child, and validity of suitable assessments.

Diagnoses on the FASD spectrum include Fetal Alcohol Syndrome (FAS), in which individuals display a set of characteristic facial dysmorphology, growth impairment, and neurodevelopmental delays; partial Fetal Alcohol Syndrome (pFAS), in which individuals display only some of the facial dysmorphology and/or growth impairment, and neurodevelopmental delays; and Neurodevelopmental Disorder – Alcohol Exposed (ND-AE), in which individuals display only some or none of the facial dysmorphology and/ or growth impairment, and neurodevelopmental delays. In evaluating whether an individual meets diagnostic criteria for FASD, clinicians evaluate four key areas:

1. Evidence of PAE: preferably self-report by the biological mother, or sometimes corroboration by family members or documentation in medical or legal records; and
2. Growth impairment: pre- or postnatal growth deficiency, defined as height or weight below the 10th percentile; and
3. Facial dysmorphology: shortened palpebral fissures (eye openings); smooth philtrum (upper-lip groove); and/or thin vermillion border (upper lip); and
4. Neurodevelopmental impairment: evidence of significant impairment in at least 3 out of 10 developmental, behavioural, and emotional domains.

The Canadian Guidelines used at the time of the Lililwan Project included fine motor skills and visual-motor integration under the 'hard and soft neurologic signs' domain. This domain includes a broad range of skills, including 'hard' signs, such as seizure activity, and 'soft' neurological signs, including sensory processing; fine and gross motor skills; visual perception; and articulation, phonology and motor speech (Chudley et al., 2005). The Canadian Guidelines were updated following the Lililwan Project, and this domain was renamed 'motor skills' (Cook et al., 2015). The newly defined 'motor skills' domain includes formal assessment of fine and gross motor skills; visual-motor integration, and graphomotor skills, with consideration given to tone, reflexes, balance, coordination, strength, and other abnormal findings on the neurological examination. It no longer includes hard neurological signs, sensory processing, visual perception, articulation, phonology, or motor speech (Cook et al., 2015).

During the Lililwan Project, the paediatrician assessed children for evidence of hard neurological signs; the paediatrician, physiotherapist, and occupational therapist (the doctoral candidate) assessed different aspects of general soft neurological signs. The occupational therapist assessed sensory processing, fine motor skills and visual-motor integration using a range of different assessment tools (see 3.5.1 Fine motor assessment tools); the physiotherapist assessed gross motor skills; and the speech pathologist assessed articulation, phonology, and motor speech. The Canadian Guidelines do not advise how many of these individual skill sets need to be significantly impaired for the whole domain to be impaired. At the end of the assessment phase of the Lililwan Project, the multidisciplinary team reviewed the results from each of the assessment tools to ensure that the diagnostic guidelines had been consistently applied when determining whether a domain was significantly impaired.

3.4 Definition of a significant neurodevelopmental impairment

The Canadian Guidelines stipulate that nine neurodevelopment ('neurobehavioral') domains should be evaluated during FASD diagnosis (Chudley et al., 2005). These domains were adapted slightly in the Lililwan Project, which evaluated 10 domains:

- CNS structure (including head circumference < 3rd percentile or other structural CNS abnormality
- Hard and soft neurological signs; seizure disorder; gross and fine motor functioning; articulation, phonology and motor speech
- Visual-motor integration
- Cognition (IQ or uneven cognitive profile)
- Communication: receptive and expressive
- Academic achievement
- Memory
- Executive functioning and abstract reasoning
- Attention deficit/hyperactivity +/- other behavioural problems; abnormal sensory processing
- Adaptive behaviour, social skills, social communication.

The Canadian Guidelines suggest that a neurodevelopmental impairment exists if an individual has 'significant' impairment in a minimum of three different domains. A 'significant' impairment is defined as being a score that is: i) 2 standard deviations (*SD*) or more below the mean on a standardised assessment tool; or ii) a discrepancy of at least 1 *SD* between subdomains; or iii) a discrepancy of at least 1.5 to 2 *SD* between subtests. The Canadian Guidelines also advise that 'in areas where standardized measurements are not available, a clinical judgement of 'significant dysfunction' is made' (Chudley et al., 2005, p.18).

3.5 Assessment of fine motor skills

Sensory-motor skills, including sensory processing, fine motor skills, and visual-motor integration, were evaluated by the occupational therapist (the doctoral candidate). This thesis reports fine motor and visual-motor integration skills (from here-on collectively termed 'fine motor skills' for brevity). Fine motor skills were assessed in a single-session of approximately 1 hour, in a one-to-one session. A Community Navigator was present to explain task requirements to the child if required. Fine motor skills use the smaller muscles of the hand and wrist to perform tasks with speed, accuracy, control, coordination and dexterity (Exner, 2005). Fine

motor skills facilitate independence and successful performance of many self-care, academic, and recreational or play activities. Visual-motor integration is sometimes considered a fine motor skill, but as outlined in section 4.2 Visual-motor , the neural processes underpinning visual-motor integration are complex and differ from many other types of fine motor skills. It became apparent in the early stages of the Lililwan Project that visual-motor integration was often impaired despite other fine motor skills being intact, so the clinical assessment team decided to classify visual-motor integration as a separate domain of impairment in terms of FASD diagnosis. However, in accordance with the Canadian Guidelines, care was taken to ensure that the two domains did not overlap. Also in accordance with the Canadian Guidelines, clinical judgement was applied in determining whether a domain was impaired, as opposed to relying solely on cut-offs based on standard deviations, because assessment tools had not been validated with the local cohort. To this end, children completed graphomotor (drawing and handwriting) tasks to supplement outcomes from standardised assessment tools.

3.5.1 Fine motor assessment tools

When choosing assessment tools, the occupational therapist considered which assessment tools were commonly used in international FASD diagnostic clinics, as well as those commonly used in Australia and by local occupational therapists in the Kimberley. Assessment tools were chosen to minimise potential cultural and language bias where possible. For example, the Bruininks-Oseretsky Test of Motor Proficiency (BOT-2) includes tasks which are designed to be novel for all children regardless of background or experience (Bruininks & Bruininks, 2005), and the developers of the Beery-Buktenica Developmental Test of Visual-Motor Integration (Beery VMI) claim it is ‘virtually culture-free’ as it does not rely on alphabetical or numerical knowledge (Beery & Beery, 2010, p. 15). Further, the children in our cohort had been attending mainstream schools for at least four years, and were familiar with fine motor tasks such as pencil grasp, and writing their names and short sentences.

Each child was asked to write their name, a short sentence, and draw a picture of themselves to determine hand dominance, pencil grasp, and pressure exerted through the pencil. These functional graphomotor tasks were initially included to supplement and corroborate outcomes from the standardised assessment tools, and provide knowledge about the functional impact of deficits to ensure therapeutic recommendations for teachers and families addressed specific difficulties. However, as the Lililwan Project progressed, it became evident that these observations of

functional fine motor performance provided valuable information about the functional impact of fine motor and visual-motor integration impairments. The drawing and handwriting samples were not evaluated using standardised assessment tools during the Lililwan Project because it was not feasible to introduce new assessment tools part-way through the project. However, the graphomotor samples seemed to provide valuable information about the functional impact of fine motor deficits, and thus could be of value to future FASD diagnostic processes and guide the choice of therapeutic interventions. The samples were evaluated retrospective to the Lililwan Project by two independent occupational therapists who were blinded to each child's PAE and FASD status (see 4.3 Graphomotor). The handwriting samples were evaluated using the Evaluation Tool of Children's Handwriting (ETCH) (Amundson, 1995), and the drawing samples evaluated with the Miller Function and Participation Scales (M-FUN) (Miller, 2006).

3.5.2 Definition of a significant fine motor or visual-motor integration impairment in the Lililwan Project

Using the Canadian Guidelines as a framework, fine motor skills were considered significantly impaired if the BOT-2 Fine Motor Composite, Fine Manual Control or Manual Coordination composite standard scores were: i) ≥ 2 *SD* below the mean, or ii) ≥ 1.5 *SD* below the mean *and* supporting evidence from functional graphomotor assessments (e.g. immature pencil grasp; exerting excessive pencil pressure in handwriting) *and* supporting evidence of fine motor impairment from fine motor related subtests in other standardised assessment tools (e.g., the Beery VMI Fine Motor Coordination subtest; or a 'moderate discrepancy' score on the Quick Neurological Screening Test (Mutti, Martin, Sterling, & Spalding, 1998) Hand Skill; Figure Recognition and Production, or Thumb and Finger Circles subtests). The occupational therapist (the doctoral candidate) and physiotherapist retrospectively reviewed BOT-2 data from all participants to determine if it was feasible to apply the Canadian Guidelines regarding discrepancy between domains or subtests. However, this diagnostic criteria did not appear to be valid for the BOT-2, as many children had a 'significant' discrepancy between different types of fine and gross motor skills, and it was deemed clinically and diagnostically irrelevant.

However, in contrast to the BOT-2 subtests, the Beery VMI subtests were designed to determine if discrepancies existed between the core Beery VMI test and the Visual Perception and Fine Manual Coordination subtests, so more detailed diagnostic criteria were applied. The visual-motor integration domain was considered significantly

impaired if the standard score on the Beery VMI was: i) ≥ 2 *SD* below the mean; or ii) ≥ 1.5 *SD* below the mean with additional functional impairments such as handwriting difficulties; or iii) 'Below average' performance in BOT-2 Fine Motor Integration subtest; or iv) ≥ 1.5 *SD* between either the core Beery VMI test and the Visual Perception subtest *or* the core Beery VMI test and Fine Motor Coordination subtest (but only when the main Beery VMI standard score was 'below average', 'low', or 'very low'). Clinical judgement was applied to ensure fine motor and visual-motor integration impairments that contributed towards a FASD diagnosis did not overlap.

3.5.3 Prevalence of fine motor impairment

The prevalence rates of 'significant' impairments were determined retrospectively to the Lililwan Project once data from the whole cohort could be analysed. Data did not exist on the prevalence of fine motor impairment in Australian Aboriginal children, nor in the fine motor or visual-motor integration skills of children with PAE or FASD. Further, while the Lililwan Project generally used the somewhat conservative cut-offs of ≥ 2 *SD* below the mean to indicate a significant impairment, other diagnostic guidelines, including those published by the U.S. Centers for Disease Control, propose a less conservative cut-off of ≥ 1 *SD* below the mean (Bertrand et al., 2004). Additionally, impairments ≥ 1 *SD* below the mean (below the 16th percentile) often indicate clinical impairment, and are used by clinicians as a basis for initiating therapeutic intervention (Williams, Lee, & Anderson, 2010). For these reasons, each of the published papers report the prevalence of both 'severe' and 'significant' (≥ 2 *SD* below the mean) and 'moderate' (≥ 1 *SD* below the mean) rates of impairment.

3.6 Cohort groupings

Little was known about the fine motor skills of Aboriginal children in remote Australia prior to this study, nor of children with PAE or FASD, so it was considered important to report results on two levels. First, results were reported for the total cohort to provide important information about the general level of fine motor performance across the cohort. Second, results were compared between children: i) without PAE ('no PAE' group); ii) with PAE but who did not meet FASD diagnostic criteria ('PAE, no FASD' group); and iii) who met FASD diagnostic criteria (PAE; with significant neurodevelopmental impairments in a minimum of three domains). With regard to the 'PAE, no FASD' group, the group is diverse, and includes children who may have had no neurodevelopmental impairment; or who had some degree of impairment but not at a 'significant' level in a minimum of three domains; the small

numbers of children in some of the FASD diagnostic groupings did not allow comparison between the different FASD diagnoses.

3.7 Summary

This chapter has provided an overview of the Lililwan Project and the role of fine motor assessment within the FASD framework. The following chapter includes further detail about the fine motor assessments, provides results and discusses the results in relation to the literature.

Chapter 4 Results

This chapter is based on three papers which have been published in peer-reviewed academic journals. Each of the papers report the outcomes from different fine motor assessment tools used in the Lililwan Project. They are:

4.1 Fine Motor : The BOT-2

4.2 Visual-motor Integration: The Beery VMI

4.3 Graphomotor : graphomotor assessments (clinical observations, the ETCH and the M-FUN)

4.4 Prevalence of Significant Domains of Fine Motor or Visual-motor Integration : an additional section to report how many children were defined as having a 'significant' fine motor or visual-motor integration impairment according to criteria used in the Lililwan Project (see 3.5.2 Definition of a significant fine motor or visual-motor integration impairment in the Lililwan Project).

4.1 Fine Motor Skills

This section is the peer-reviewed, published paper:

> Doney, R., Lucas, B. R., Watkins, R. E., Tsang, T. W., Sauer, K., Howat, P., Latimer, J., Fitzpatrick, J. P., Oscar, J., Carter, M., & Elliott, E. J. (2017). Fine motor skills in a population of children in remote Australia with high levels of prenatal alcohol exposure and Fetal Alcohol Spectrum Disorder. *BMC Pediatrics, 17*(193), 1-10. doi: 10.1186/s12887-017-0945-2

This chapter reports results from the Bruininks-Oseretsky Test of Motor Proficiency (BOT-2) (Bruininks & Bruininks, 2005).

4.1.1 Abstract

Background: Many children in the remote Fitzroy Valley region of Western Australia have prenatal alcohol exposure (PAE). Individuals with PAE can have neurodevelopmental impairments and be diagnosed with one of several types of Fetal Alcohol Spectrum Disorder (FASD). Fine motor skills can be impaired by PAE, but no studies have developed a comprehensive profile of fine motor skills in a population-based cohort of children with FASD. We aimed to develop a comprehensive profile of fine motor skills in a cohort of Western Australian children;

determine whether these differed in children with PAE or FASD; and establish the prevalence of impairment.

Methods: Children (*n* = 108, 7 to 9 years) were participants in a population-prevalence study of FASD in Western Australia. Fine motor skills were assessed using the Bruininks-Oseretsky Test of Motor Proficiency, which provided a Fine Motor Composite score, and evaluated Fine Manual Control (Fine Motor Precision; Fine Motor Integration) and Manual Coordination (Manual Dexterity; Upper-Limb Coordination). Descriptive statistics were reported for the overall cohort and comparisons made between children with and without PAE and/or FASD. The prevalence of severe (≤ 2nd percentile) and moderate (≤ 16th percentile) impairments was determined.

Results: Overall, Fine Motor Composite scores were 'average' (M = 48.6 ± 7.4), as were Manual Coordination (M = 55.7±7.9) and Fine Manual Control scores (M = 42.5±6.2). Children with FASD had significantly lower Fine Motor Composite (M = 45.2±7.7 *p* = 0.046) and Manual Coordination scores (M = 51.8±7.3, *p* = 0.027) than children without PAE (Fine Motor Composite M = 49.8±7.2; Manual Coordination M = 57.0±7.7). Few children had severe impairment, but rates of moderate impairment were very high.

Conclusions: Different types of fine motor skills should be evaluated in children with PAE or FASD. The high prevalence of fine motor impairment in our cohort, even in children without PAE, highlights the need for therapeutic intervention for many children in remote communities.

4.1.2 Introduction

Local Aboriginal leaders in the remote Fitzroy Valley region of Western Australia introduced alcohol restrictions in 2007 because they were concerned about the social and health effects of chronic alcohol misuse. These concerns included the potential harm caused by alcohol consumption during pregnancy, which can cause Fetal Alcohol Spectrum Disorder (FASD). In 2009 local leaders initiated 'The Lililwan Project' ('Lililwan' is Kimberley Kriol for 'all the little ones') to determine the prevalence of FASD [1]. Diagnoses on the FASD spectrum include Fetal Alcohol Syndrome (FAS) and partial Fetal Alcohol Syndrome (pFAS), both with characteristic facial anomalies and impaired growth; and Alcohol-Related Neurodevelopmental Disorder (ARND) or Neurodevelopmental Disorder – Prenatal/ Alcohol Exposed (ND-PAE/ND-AE) with neurodevelopmental impairment in the absence of physical features [2, 3].

PAE can affect the development and function of the corpus callosum [4], cerebellum [5], basal ganglia [6], and motor cortex [7], and children with FASD may have skeletal malformations [8], abnormal muscle development [9], tremor [10], and impaired nerve conductivity [11]. All these factors may impair fine motor performance. Fine motor skills include basic skills such as grip strength, and more complex skills including visual (or fine) motor integration, manual dexterity, and upper-limb coordination. These skills underpin many self-care, academic, and recreational activities, including handwriting, dressing, and ball sports. Fine motor skills are particularly important in primary school aged children, who can spend more than half of their day completing tasks which require fine motor skills [12]. Handwriting quality can be affected by poor fine motor skills, and students with poor handwriting often receive poorer grades [13]. Teacher reports indicate that 20.6% of first year students at Fitzroy Fitzroy Crossing are below the Australian population 10th percentile for fine and gross motor skills [14]. Many Australian Aboriginal students perform below-average on the National Assessment Program – Literacy and Numeracy (NAPLAN), which is conducted annually with students in Years 3, 5, 7, and 9 [15].

Few studies of children with PAE or FASD have reported whether they have a motor impairment, and of those that do, many report a motor score that is a combination of fine motor and gross motor skills [16-18], or a score based on subtests of generalised developmental assessment tools [19], such as the Eye and Hand Coordination subscale from the Griffith's Mental Development Scales [20]. Individuals with FASD can have subtle neurological impairment, and researchers have highlighted the importance of assessing a range of specific areas of function rather than reporting amalgamated scores [18, 19]. Motor scores that are an average of fine and gross motor skills provide little insight into deficits, which is essential for understanding the child's neurological profile and developing appropriate therapy goals.

Several studies have assessed a range of fine motor skills in children with PAE or FASD [21-24], but each has used varying assessment tools and none report data from an entire population age-cohort. Motor skills in children with PAE or FASD are summarised in three systematic reviews. In one review, 'visual and motor' skills were not associated with mild, moderate, or binge PAE, however, none of the included studies assessed children older than 5 years [25]. Another review found an association between motor impairment and levels of PAE, but did not differentiate between fine and gross motor skills [26]. We reviewed fine motor skills in primary school aged children with PAE or FASD [27], and found that complex fine motor

skills, such as visual-motor integration, were more likely to be impaired than basic skills, such as grip strength. We identified a range of assessment tools used to assess fine motor skills in children with PAE or FASD, but few that comprehensively assessed a range of different skills.

Study hypotheses

In this study, we report the fine motor proficiency and prevalence of impairment amongst children in the remote Fitzroy Valley, Western Australia. We hypothesised that rates of fine motor impairment would be high due suspected high rates of neurodevelopmental and socioeconomic risk factors, including PAE. We also hypothesised that children with PAE, particularly those with FASD, would have the most impairment due to the teratogenic effect of alcohol on the central and peripheral nervous systems involved in performance of fine motor skills.

Study aims

1. Assess and evaluate fine manual control (fine motor precision and fine motor integration) and manual coordination (manual dexterity and upper-limb coordination) in a cohort of children in the Fitzroy Valley.
2. Compare fine motor skills of children (i) without PAE; (ii) with PAE but not FASD; and (iii) with FASD.
3. Determine the prevalence of moderate (≤ 16th percentile) and significant (≤ 2nd percentile) fine motor impairments in the cohort.

4.1.3 Methods

Setting

We evaluated fine motor data from the Lililwan Project, a population-based study of FASD prevalence in the Fitzroy Valley in the West Kimberley region of northern Western Australia. The Fitzroy Valley has a population of 4,500 people living in communities across a 200km radius, 80% of whom identify as being Australian Aboriginal [28].

Procedures

All children born in 2002 or 2003 and living in the Fitzroy Valley during 2010 and 2011 were eligible for inclusion. In Stage 1 of the study parents and carers of 127 children (95% participation) provided information about prenatal and childhood exposures, including PAE, antenatal drug exposures, nutrition, living conditions, and

exposure to early life trauma [29]. The Alcohol Use Disorders Identification Test – Consumption (AUDIT-C) was used to classify PAE as 'low', 'risky', or 'high risk' [30].

In Stage 2, 108 of the children completed comprehensive neurodevelopmental assessments by qualified paediatricians and allied health practitioners. Attrition occurred because families moved out of the Fitzroy Valley (*n* = 15), we were unable to locate families or children (*n* = 3), or clinical assessment was declined (*n* = 1).

Assessors were blinded to alcohol and other pre-and post-natal exposures. Adapted Canadian FASD Diagnostic Guidelines were used to assign FASD diagnoses, including FAS, pFAS, and ND-AE. To be diagnosed with one of the FASD diagnoses, a child was required to have 'significant' impairment (defined as ≥ 2 *SD* below the mean, or clinically significant variability between subtests on standardised assessments) in a minimum of 3 of 10 neurodevelopmental domains. The diagnoses of pFAS or FAS additionally required evidence of characteristic facial features or growth impairment. A study protocol detailing assessment tools and diagnostic criteria has been published [1]. Children were referred to local health services for medical or therapeutic treatment if required. Families whose child had a FASD diagnosis were referred to a social worker and an Indigenous support worker with extensive experience working with families affected by FASD. Fine motor skills were assessed in a one hour session by the primary author (RD), an occupational therapist with experience working with children in the Fitzroy Valley. Overall motor proficiency and gross motor skills were assessed by a paediatric physiotherapist (BRL), and have been reported [31, 32].

Instrumentation

The Bruininks-Oseretsky Test of Motor Proficiency (Second edition)

The Bruininks-Oseretsky Test of Motor Proficiency (BOT-2) is a standardised, norm-referenced tool suitable for motor assessment in children and young adults aged 4-21 years [33]. Complete (53 tasks) and short versions (14 tasks) are available. The complete version of the BOT-2 was chosen for use in our study because it evaluates a diverse range of fine motor skills, is frequently used in Australia [34] and international FASD diagnostic clinics [35], and is recommended in the Canadian FASD Diagnostic Guidelines [3]. The BOT-2 provides a Fine Motor Composite score, which is an overall measure of fine motor proficiency. The Fine Motor Composite score is derived from the Fine Manual Control and Manual Coordination composite scores, which in turn are derived from Fine Motor Precision (which assesses precise hand and finger control through paper and pencil tasks, folding paper, and scissor

skills), Fine Motor Integration (which assesses ability to reproduce a series of eight geometric shapes), Manual Dexterity (which assesses reaching, grasping, and bimanual control through timed tasks such as stringing blocks and placing pegs in a pegboard), and Upper-Limb Coordination (which assesses coordinated arm and hand movement in terms of catching, throwing, and dribbling a tennis ball) subtest scores (*Figure 4.1*). Composites are reported as standardised scores (mean (M) = 50.0, standard deviation (*SD*) = 10.0), and subtest scores are reported as scale scores (M = 15.0, *SD* = 5.0). Descriptive categories are defined as 'well-above average'(standard score ≥ 70; scale score ≥ 25; ≥ 98th percentile); 'above average' (standard score 60 to 69; scale score 20 to 24; 84th to 97th percentile); 'average'(standard score 41 to 59; scale score 11 to 19; 18th to 83rd percentile); 'below average'(standard score 31 to 40; scale score 6 to 10; 3rd to 17th percentile); and 'well-below average' (standard score ≤ 30; scale score ≤ 5; ≤ 2nd percentile) [33].

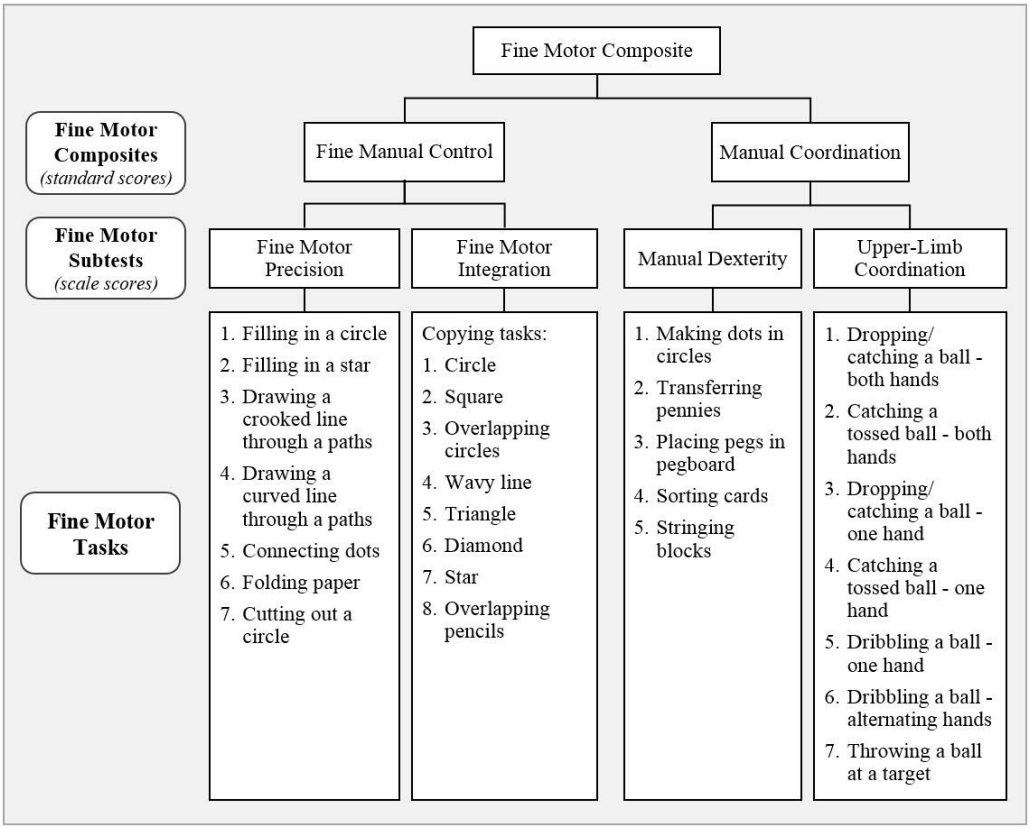

Figure 4.1 BOT-2 Fine motor composites, subtests, and tasks

BOT-2 tasks are designed to be novel for all children, including those from diverse cultural backgrounds, regardless of familiarity with the tasks, and the composites and subtests have well-established internal consistency and test-retest reliability [33]. The BOT-2 Short Form was trialled in a subset of children from the Lililwan project and we found it to have excellent inter-rater reliability (0.88 to 0.92) and fair

to good test-retest reliability (0.62 to 0.73) in this population [35]. The BOT-2 is endorsed as a suitable measure of motor skills in FASD diagnostic assessment [3].

Statistical analysis

Data were scored using the sex-specific norms of the BOT-2 ASSIST scoring software. The Fine Motor Composite score was calculated using the online Q-global^{TM} scoring system. Means and standard deviations were obtained for all BOT-2 fine motor composite standardised scores and subtest scale scores. Fine motor scores were assessed for normality and analysed using a one-way between groups analysis of variance (ANOVA). Children with unconfirmed or unknown PAE (n = 5) were excluded from the between-groups analysis. Group differences were analysed using ANOVA between children without PAE ('No PAE' group); children with PAE who did not have multiple, significant neurodevelopmental impairments and were therefore not diagnosed with a type of FASD ('PAE (no FASD)' group); and children with confirmed PAE plus FASD ('FASD' group). Significance was set at $p < 0.05$. Effect sizes (eta^2) were calculated, with 0.01 being deemed a small effect size; 0.06 a medium effect size; and 0.14 a large effect size [36]. Tukey's Honestly Significant Difference (HSD) test was utilised as a post-hoc test to determine which groups differed. Prevalence of severe (≥ 2 *SD* below the mean; ≤ 2nd percentile) and moderate (≥ 1 *SD* below the mean; ≤ 16th percentile) impairment was reported for each fine motor composite and subtest for the cohort, and also by exposure group. Statistical analysis was completed using IBM SPSS Statistics for Windows, version 21.0 (Armonk, NY: IBM Corp.).

4.1.4 Results

Participants

Participants were aged between 7.5 to 9.6 years (M = 8.7 years) at assessment. The majority were of Australian Aboriginal descent (Table 4.1). Of the children with PAE (n = 60, 55.6%), most (95%) were exposed to 'risky' or 'high risk' levels according to AUDIT-C criteria [37]. Children who participated in Stage 1 only (n = 15) were slightly less likely to have PAE (36.8%) than children who participated in both Stage 1 and 2 (55.6%) but were otherwise similar. Children with and without PAE were born at similar weeks of gestation, and the incidence of pre-term births were also similar [37]. The Universal Non-Verbal Intelligence Test [38] formed part of the assessment battery during the Lililwan Project and was used to evaluate cognitive abilities. Full-scale standard scores were similar between groups with and

without PAE or FASD (No PAE M = 89.9, *SD* = 8.5; PAE, no FASD M = 89.4, *SD* = 9.1; FASD M = 85.0, *SD* = 12.3; *p* = 0.329).

Many children lived in overcrowded households (M = 6.1, range 2 -16), and many had lived in more than four homes since birth (*n* = 17, 15.8%). Most children (*n* = 89, 82.4%) attended school 4 to 5 days a week, with only one child (who did not have FASD) not attending school at all. Half of the children's biological mothers had studies beyond Year 10. These socioeconomic factors were similar between children with and without FASD [39].

Table 4.1

Cohort characteristics

	Total Cohort[a] *N* = 108		No PAE *n* = 43		PAE (no FASD) *n* = 39		FASD *n* = 21	
	n	(%)	n	(%)	n	(%)	n	(%)
Australian Aboriginal	106	(98.1)						
Gender								
Male	57	(52.8)	24	(55.8)	18	(46.2)	13	(61.9)
Handedness								
Right	101	(93.5)	41	(95.3)	38	(97.4)	19	(90.5)
Hearing[d]								
Normal	42	(38.9)	16	(37.2)	14	(35.9)	10	(47.6)
Mild loss	38	(35.2)	15	(34.9)	13	(33.3)	7	(33.3)
Moderate loss	13	(12.0)	7	(16.3)	3	(7.7)	3	(14.3)
Missing	15	(13.9)	5	(11.6)	9	(23.1)	1	(4.8)
Prenatal nicotine exposure[e]								
Yes	67	(62.0)	18	(41.9)	32	(82.1)	15	(71.4)
Unknown	7	(6.5)	0	(0.0)	1	(2.6)	3	(14.3)
Prenatal marijuana exposure[e]								
Yes	13	(12.0)	2	(4.7)	10	(25.6)	1	(4.8)
Unknown	7	(6.5)	0	(0.0)	1	(2.6)	2	(9.5)
PAE risk levels[f]								
No exposure	43	(100.0)	0	(0.0)	0	(0.0)	0	(0.0)
Low (1-3)	4	(3.7)	0	(0.0)	4	(10.3)	0	(0.0)
Risky (4-5)	4	(3.7)	0	(0.0)	3	(7.7)	1	(4.8)
High risk (≥ 6)	46	(42.6)	0	(0.0)	29	(74.4)	17	(81.0)
PAE, uncertain risk	6	(5.6)	0	(0.0)	3	(7.7)	3	(14.3)
Unknown PAE	5	(4.6)	0	(0.0)	0	(0)	0	(0.0)

[a] 'Total cohort' includes *n* = 5 children with unknown PAE who are not included in the No PAE, PAE (no FASD), or FASD groups

[b] Reduced visual acuity defined as ≤6/9 in one or both eyes

[c] Not all children completed audiology and ophthalmology testing

[d] Mild hearing loss 26 – 40dB; moderate hearing loss 41 – 55dB

[e] Some prenatal exposure information not available, either due to the primary carer not knowing, or the birth mother choosing not to disclose this information

[f] Risk level according to AUDIT-C scoring criteria

Fine motor composites and subtests

For the total cohort, all fine motor composite and subtest scores were in the 'average' range (Table 4.2). Children with FASD had significantly lower Fine Motor Composite scores and Manual Coordination scores than children without PAE (Fine Motor Composite eta^2 = 0.06, Tukey's HSD p = 0.038; Manual Coordination eta^2 = 0.07, Tukey's HSD p = 0.024) (Table 4.2). There were no other significant differences between groups, but the mean scores of the PAE (no FASD) and FASD groups were consistently lower than in children without PAE in almost all composites and subtests (aside from the Upper-Limb Coordination subtest), and the scores of children with FASD were lower again (*Figure 4.2*).

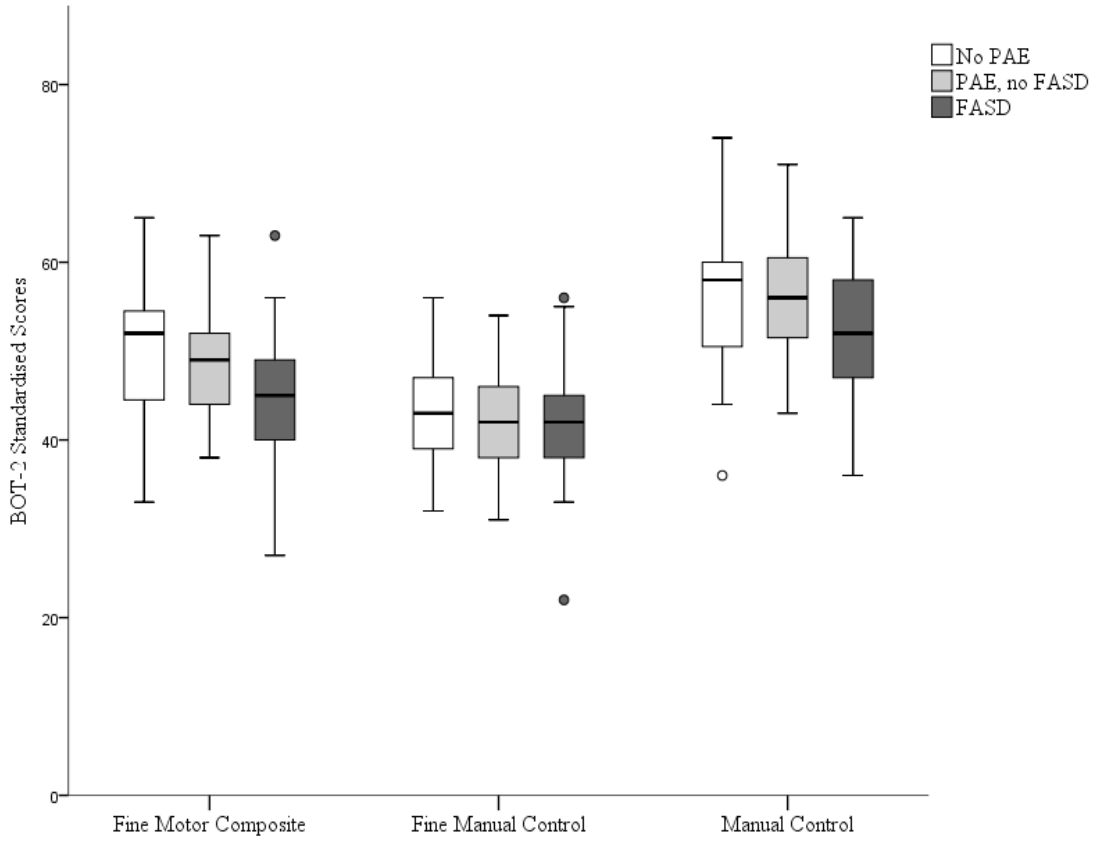

Figure 4.2 BOT-2 Fine Motor Composite, Fine Manual Control, and Manual Coordination composite scores for children with no PAE; PAE but not FASD; and FASD

Table 4.2

BOT-2 Fine motor composite standardised scores and subtest scale scores in children with no PAE; PAE (no FASD); and FASD

	Total Cohort[1] *N* = 108			No PAE *n* = 43			PAE (no FASD) *n* = 39			FASD *n* = 21					
	M	(*SD*)	95% CI	M	(*SD*)	95% CI	M	(*SD*)	95% CI	M	(*SD*)	95% CI	df	F	ANOVA[3] (*p*)
FINE MOTOR COMPOSITE	48.6	(7.4)	47.2 - 50.0	49.8	(7.2)	47.6 - 52.0	48.8	(6.2)	46.8 - 50.8	45.2	(7.7)	41.7 - 48.7	2,100	3.17	0.046*[4]
Fine Manual Control[2]	42.5	(6.2)	41.3 - 43.6	43.4	(6.2)	41.4 - 45.3	41.9	(5.3)	40.2 - 43.6	41.1	(7.3)	37.8 - 44.5	2,100	1.10	0.336
Fine Motor Precision[3]	12.3	(3.3)	11.7 - 12.9	12.7	(3.4)	11.7 - 13.8	11.9	(2.6)	11.0 - 12.7	11.8	(4.0)	10.0 - 13.6	2,100	0.94	0.393
Fine Motor Integration[3]	11.0	(2.9)	10.5 - 11.6	11.3	(2.7)	10.4 - 12.1	11.2	(2.9)	10.3 - 12.2	10.1	(3.0)	8.8 - 11.5	2,100	1.29	0.279
Manual Coordination[2]	55.7	(7.9)	54.2 - 57.2	57.0	(7.7)	54.6 - 59.4	56.2	(7.0)	53.9 - 58.5	51.8	(7.3)	48.4 - 55.1	2,100	3.74	0.027*[4]
Manual Dexterity[3]	14.9	(3.7)	14.2 - 15.6	15.4	(3.5)	14.3 - 16.4	15.1	(3.1)	14.1 - 16.1	13.2	(4.0)	11.4 - 15.0	2,100	2.97	0.056
Upper-Limb Coordination[3]	19.6	(4.4)	18.7 - 20.4	19.8	(4.4)	18.5 - 21.2	20.0	(4.5)	18.5 - 21.5	18.0	(3.8)	16.3 - 19.7	2,100	1.64	0.200

1 'Total Cohort' includes n = 5 children with unknown PAE who are not included in the No PAE, PAE (no FASD), or FASD groups
2 BOT-2 norms M = 50, SD = 10
3 BOT-2 norms M = 15, SD = 5. Lower scores represent poorer performance in composites and subtests
4 Tukey's HSD: No PAE > FASD
* $p < 0.05$

Prevalence of fine motor impairment

Prevalence of severe impairment (range 0 to 0.9%) was low in all composites and subtests (Table 4.3). Prevalence of moderate impairment for the Fine Motor Composite (14.8%) was derived from a high prevalence of moderate impairment in the Fine Manual Control composite (38.9%), and low prevalence in the Manual Coordination composite (1.9%) (Table 4.3). Only one child with PAE (who had FASD) had severe impairment in any fine motor composite or subtest (Table 4.3). Prevalence of moderate impairment in the Fine Motor Composite was slightly lower than BOT-2 norms for children without PAE (11.6%) and PAE (no FASD) (7.7%), but much higher in children with FASD (28.6%). Moderate impairment was very high in the Fine Manual Control composite (and its associated subtests) for all exposure groups, but highest in children with FASD (47.6%). Moderate impairment was less than expected in the Manual Coordination composite for all exposure groups (range 0 – 4.8%), but this composite was an amalgamation of the Manual Dexterity subtest, which had high rates of moderate impairment, particularly for children with FASD (23.8%), and the Upper-Limb Coordination subtest, in which few children had moderate impairment (range 4.7 to 5.1%).

Table 4.3

Prevalence of severe (≥ -2SD) and moderate (≥ -1SD) fine motor impairment in children with no PAE; PAE (no FASD); and FASD

	Total Cohort[1] *N* = 108		No PAE *n* = 43		PAE (no FASD) *n* = 39		FASD *n* = 21	
	n	(%)	n	(%)	n	(%)	n	(%)
FINE MOTOR COMPOSITE								
- ≥ 2*SD*	1	(0.9)	0	(0.0)	0	(0.0)	1	(4.8)
- ≥ 1*SD*	16	(14.8)	5	(11.6)	3	(7.7)	6	(28.6)
Fine Manual Control								
- ≥ 2*SD*	1	(0.9)	0	(0.0)	0	(0.0)	1	(4.8)
- ≥ 1*SD*	42	(38.9)*	16	(37.2)*	14	(35.9)*	10	(47.6)**
Fine Motor Precision								
- ≥ 2*SD*	1	(0.9)	0	(0.0)	0	(0.0)	1	(4.8)
- ≥ 1*SD*	33	(30.6)	12	(27.9)	11	(28.2)	9	(42.9)*
Fine Motor Integration								
- ≥ 2*SD*	1	(0.9)	0	(0.0)	0	(0.0)	1	(4.8)
- ≥ 1*SD*	48	(44.4)*	17	(39.5)*	15	(38.5)*	13	(61.9)**

	Total Cohort[1] *N* = 108		No PAE *n* = 43		PAE (no FASD) *n* = 39		FASD *n* = 21	
	n	(%)	n	(%)	n	(%)	n	(%)
Manual Coordination								
- ≥ 2*SD*	0	(0)	0	(0.0)	0	(0.0)	0	(0.0)
- ≥ 1*SD*	2	(1.9)	1	(2.3)	0	(0.0)	1	(4.8)
Manual Dexterity								
- ≥ 2*SD*	1	(0.9)	0	(0.0)	0	(0.0)	1	(4.8)
- ≥ 1*SD*	11	(10.2)	5	(11.6)	0	(0.0)	5	(23.8)
Upper-Limb Coordination								
- ≥ 2*SD*	0	(0)	0	(0.0)	0	(0.0)	0	(0.0)
- ≥ 1*SD*	5	(4.6)	2	(4.7)	2	(5.1)	1	(4.8)

- ≥ 2*SD* = ≤ 2nd percentile; - ≥ 1*SD* = ≤ 16th percentile
* = at least twice, and ** = at least three times, the rate of BOT-2 norms
[1] 'Total Cohort' includes *n* = 5 children with unknown PAE who are not included in the No PAE, PAE (no FASD), or FASD group

4.1.5 Discussion

This is the first study to comprehensively assess fine motor skills in a population-based cohort of predominantly Aboriginal children in Australia. Many children in our study had high levels of PAE and were diagnosed with FASD. The cohort's mean BOT-2 Fine Motor Composite scores were in the 'average' range, an unexpected finding given the high levels of PAE and other neurodevelopmental risk factors in our cohort. However, in keeping with our hypothesis, children with FASD had poorer fine motor skills than children without PAE. Manual coordination skills, including fine motor speed, manual precision, and coordinated arm and hand movement were specific areas of difficulty for children with FASD. Few children had severe impairment (below the 2nd percentile), but rates of moderate impairment (below the 16th percentile) were very high.

Similar to our findings, other studies of fine motor impairment in children with PAE or FASD have also reported a mixed profile of strengths and difficulties. A range of assessment tools have been used to evaluate fine motor skills in children with PAE or FASD, including the Visuomotor Precision subtest from the Developmental Neuropsychological Evaluation (NEPSY) [40], the Movement Assessment Battery for Children (M-ABC) [41], and The Beery Buktenica Developmental Test of Visual-Motor Integration (Beery VMI) [42]. Other studies have reported mixed findings for fine motor precision [24, 43] and manual dexterity [44, 45] skills, which weren't impaired in children with PAE or FASD in our study. Ball skills were also not impaired, which is consistent with other reported findings [44-46]. We found that visual-motor integration (termed 'fine motor integration' in the BOT-2) wasn't impaired, but this contradicts other studies which commonly report visual-motor

integration impairment in children with FASD [47-49]. This may be due to the limited number of tasks used to evaluate this skill in the BOT-2 (*n* = 8), compared to the more commonly used Beery VMI (*n* = 30). The Beery VMI formed part of the neurodevelopmental assessment battery in the Lililwan Project, and we reported that the Fine Motor Coordination subtest of the Beery VMI was significantly lower in children with FASD [50].

Only one other study group [17] has published motor outcomes in children with FASD using the BOT. These authors used an earlier version of the BOT (1st edition), which does not include a Fine Motor Composite score. The authors reported that the motor score (an amalgamation of fine and gross motor skills) was not significantly different in children with FASD (M = 49.1) compared to 'typically developing' (M = 57.7, *p* = 0.36) children. These non-significant findings may result from areas of stronger skills masking fine motor impairments, in much the same way that children in our cohort with FASD had an 'average' Fine Motor Composite score (M = 45.2), which was derived from relatively stronger Manual Coordination (M = 51.8) and weaker Fine Manual Control scores (M = 41.1).

Implications of prevalence rates

The very low prevalence of severe fine motor impairment in our cohort has implications for FASD diagnosis. The University of Washington 4-digit Diagnostic Code [51] and the Canadian FASD Diagnostic Guidelines [3] each advise that scores 2 *SD* below the mean (≤ 2nd percentile) indicate impairment when diagnosing FASD. In contrast, 1 *SD* below the mean (≤ 16th percentile) indicates impairment according to the Centers for Disease Control (CDC) [2]. Other authors have also proposed a 1 *SD* cut-off for identifying impairment for ND-PAE [52]. Only one child in our cohort (who had FASD) had fine motor scores below the 2nd percentile, which seems conservative given the high levels of PAE and other neurodevelopmental risk factors in our cohort. This issue warrants further consideration and investigation.

Strengths

This study is the first comprehensive, population-based study of fine motor skills in Aboriginal children in Australia. It is also the first to use a standardised fine motor assessment to develop a comprehensive profile of fine motor skills in children with PAE and/or FASD.

Limitations

Most children in our study identified as Australian Aboriginal and all were living in remote communities, and so the results should not be generalised. Nevertheless,

outcomes may be relevant to other populations with similar demographics. Although the study involved almost two entire age cohorts and had a high participation rate (%), the sample size was too small to statistically control for potentially confounding factors. However, many risk factors, such as early life trauma and low socioeconomic status, were common to almost all children in our study. Many children without PAE also had a moderate level of fine motor impairment, and thus impairments cannot be solely attributed to PAE. However, the high proportion of children in our cohort with “risky” or “high risk” levels of PAE make it likely that PAE contributed, at least in part, to the identified fine motor impairment.

Recommendations and future directions

This study highlights the importance of comprehensively assessing a range of fine motor skills in children with PAE or suspected FASD. Other researchers have expressed concerns that composite scores may not be sensitive enough to detect subtle neurological impairment in children with FASD [18, 19]. Our findings support these concerns. We recommend that a range of fine motor skills be assessed in children with PAE, and outcomes not be amalgamated with other fine or gross motor scores, because an averaged ‘motor’ score could mask specific difficulties, resulting in inaccurate diagnoses and missed opportunities for therapeutic support.

Conclusions

Children in our cohort had Fine Motor Composite scores in the ‘average’ range. Upper-limb coordination (ball skills) was a strength, while fine motor integration skills (copying complex shapes) were an area of weakness. Children with FASD had significantly lower Fine Motor Composite and Manual Coordination scores than children without PAE. These outcomes highlight the importance of reporting specific types of fine motor skills, rather than an amalgamated ‘motor’ or even ‘fine motor’ score. The very high levels of impaired fine motor precision and fine motor integration skills highlight the need for therapeutic intervention for many children in the Fitzroy Valley, regardless of PAE, to encourage successful participation in self-care, academic, and recreational activities.

4.1.6 References

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4.2 Visual-motor Integration

This section is the peer-reviewed, published paper that reports results from the Beery VMI (Beery & Beery, 2010).

> Doney, R., Lucas, B. R., Watkins, R. E., Tsang, T. W., Sauer, K., Howat, P., Latimer, J., Fitzpatrick, J. P., Oscar, J., Carter, M., & Elliott, E. J. (2016). Visual-motor integration, visual perception, and fine motor coordination in a population of children with high levels of Fetal Alcohol Spectrum Disorder. *Research in Developmental Disabilities, 55*, 346-357.

4.2.1 Abstract

Background: Visual-motor integration (VMI) skills are essential for successful academic performance, but to date no studies have assessed these skills in a population-based cohort of Australian Aboriginal children who, like many children in other remote, disadvantaged communities, consistently underperform academically. Furthermore, many children in remote areas of Australia have prenatal alcohol exposure (PAE) and Fetal Alcohol Spectrum Disorder (FASD), which are often associated with VMI deficits.

Methods: VMI, visual perception, and fine motor coordination were assessed using The Beery-Buktenica Developmental Test of Visual-Motor Integration, including its associated subtests of Visual Perception and Fine Motor Coordination, in a cohort of predominantly Australian Aboriginal children (7.5 to 9.6 years, *n* = 108) in remote Western Australia to explore whether PAE adversely affected test performance. Cohort results were reported, and comparisons made between children i) without PAE; ii) with PAE (no FASD); and iii) FASD. The prevalence of moderate (≤ 16th percentile) and severe (≤ 2nd percentile) impairment was established.

Results: Mean VMI scores were 'below average' (M = 87.8±9.6), and visual perception scores were 'average' (M = 97.6±12.5), with no differences between groups. Few children had severe VMI impairment (1.9%), but moderate impairment rates were high (47.2%). Children with FASD had significantly lower fine motor coordination scores and higher moderate impairment rates (M = 87.9±12.5; 66.7%) than children without PAE (M = 95.1±10.7; 23.3%) and PAE (no FASD) (M = 96.1±10.9; 15.4%).

Conclusions: Aboriginal children living in remote Western Australia have poor VMI skills regardless of PAE or FASD. Children with FASD additionally had fine motor coordination problems. VMI and fine motor coordination should be assessed in children with PAE, and included in FASD diagnostic assessments.

What this paper adds

This study is the first to report VMI, visual perception, and fine motor coordination skills in a population-based cohort of Aboriginal children in a remote region of Australia. It is also the first to consider whether visual perception or fine motor coordination impairments are present, which could account for VMI difficulties in children with PAE or FASD. The outcomes identified that many children in remote Australia have VMI impairment, even those without PAE or FASD, suggesting that PAE is just one of many neurodevelopmental risk factors which may cause VMI impairment. Children with FASD had significantly lower fine motor coordination scores, and higher rates of moderate impairment. It seems that, for children with FASD, they either have a fine motor coordination impairment which exists independently of the VMI impairment, or alternatively, that their problems contribute, in part, to observed VMI difficulties. However, given the poor VMI score of children without PAE, it is likely that other factors also contributed to VMI difficulties in children with FASD in our cohort. The study shows that many children in the region require therapeutic support for VMI impairment, and it is important to assess both VMI and fine motor coordination in children with PAE and/or FASD.

4.2.2 Introduction

Alcohol consumption during pregnancy may result in a range of irreversible, clinically distinguishable, lifelong conditions, collectively called Fetal Alcohol Spectrum Disorder (FASD) (Centers for Disease Control and Prevention, 2005). FASD is an umbrella term which includes the diagnoses of Fetal Alcohol Syndrome (FAS); partial FAS (pFAS); and Alcohol Related Neurodevelopmental Disorder (ARND), also known as Neurodevelopmental Disorder – Prenatal/Alcohol Exposed (ND-PAE/ND-AE) (Astley & Clarren, 2000; Chudley et al., 2005). Individuals diagnosed with FAS or pFAS have characteristic dysmorphic facial features and/or growth impairment, while those with ARND/ND-AE diagnoses do not necessarily have these impairments. However, individuals with any of the FASD diagnoses have significant neurological damage which can cause mild to severe deficits in cognition, executive function, memory, language, attention, social and adaptive function, and soft neurological signs, including fine and gross motor skills and visual-motor integration (Chudley et al., 2005). These deficits can lead to social and adaptive problems at home, school or work, and in society (Streissguth et al., 2004).

Visual-motor integration (VMI) is the ability to use input from the visual perceptive system (which includes visual acuity, accommodation, binocular fusion, stereopsis,

and convergence/ divergence) to coordinate fine motor skills (which require the use of the smaller muscles of the wrist and hand, including dexterity, precision, coordination, and manual control) (Schneck, 2010). VMI underpins many everyday functions such as handwriting and drawing, catching a ball, dressing, eating, and driving (Tomchek & Schneck, 2006). There are two types of VMI: i) constructional VMI, which includes tasks such as using blocks to build a 3D shape, and ii) graphomotor VMI, which includes paper and pencil tasks such as drawing a series of lines to form geometric shapes. Different neural processes underpin constructional and graphomotor VMI, so they should be considered separate skill sets (Benton & Tranel, 1993). Impairment of graphomotor VMI can be due to underlying problems with visual perception or fine motor skills, but impairment can also exist when visual perception and fine motor skills are intact, meaning the problem lies with the integration of these skills (Beery & Beery, 2010; Kulp, 1999; Milner, 2006).

In Australia, students in Years 3, 5, 7, and 9 annually complete the National Assessment Program – Literacy and Numeracy (NAPLAN) assessment, which assesses reading, writing, language, and numeracy skills. Children in very remote areas of Western Australia, including the Fitzroy Valley, consistently under-perform compared to national averages (Australian Curriculum, Assessment and Reporting Authority, 2015). Although the NAPLAN does not specifically assess VMI or handwriting skills, other studies have shown that VMI impairment is associated with poor handwriting performance (Kulp, 1999; Weil & Cunningham Amundson, 1994), and students with poor handwriting skills often receive lower grades on written assessments despite adequate content (Chase, 1986). Therapeutic interventions which aim to improve handwriting, especially those which are integrated into the classroom, have successfully improved VMI and handwriting skills (Case-Smith, 2002).

Neurological damage resulting from prenatal alcohol exposure (PAE) may affect brain regions involved in VMI. Neuroimaging studies have shown that the corpus callosum, basal ganglia, cerebral cortex, and cerebellum may all be damaged by PAE (Riley, Infante, & Warren, 2011; Sowell et al., 2002). Within the cerebral cortex, the parietal and temporal lobes are particularly affected by PAE (Archibald et al., 2001; Sowell et al., 2002). Children with FASD with reduced parietal lobe white matter have been shown to have reduced VMI abilities (Sowell et al., 2008). Optic nerve hypoplasia (Stromland, 2004) has been reported following PAE, which may also impair visual perception, as have skeletal malformations (Jones et al., 2010), atypical muscle development (David & Subramaniam, 2005), tremor (Marcus, 1987),

and impaired nerve conductivity (de los Angeles Avaria et al., 2004) which may affect fine motor skills.

In a recent systematic review of fine motor skills in children with PAE or FASD we concluded that complex skills, including VMI, were more likely to be impaired than basic skills such as grip strength (Doney et al., 2014). VMI was the most commonly assessed skill and the Beery-Buktenica Developmental Test of Visual-Motor Integration (Beery VMI) (Beery & Beery, 2010) was the most commonly used assessment tool. The Beery VMI is commonly used by occupational therapists, psychologists, and other health and educational professionals because it reports outcomes as standard scores and percentile ranks of VMI abilities; has well-established reliability and validity; is relatively quick to administer; can be used across a wide range of ages; and is suitable for use with people from diverse educational and linguistic backgrounds (Beery & Beery, 2010). VMI skills can be predictive of handwriting performance (Daly, Kelley, & Krauss, 2003), which may be adversely affected in children with FASD (Duval-White, Jirikowic, Rios, Deitz, & Olson, 2013). The Beery VMI includes two optional subtests of Visual Perception and Fine Motor Coordination which can be used to determine whether VMI deficits are due to underlying problems with these skills. In all but one study in which the Beery VMI was used, significant differences were reported between children with and without PAE or FASD, but no studies reported outcomes from the Visual Perception or Fine Motor Coordination subtests.

To date, no studies have reported graphomotor VMI skills in a population-based cohort of Australian Aboriginal children with high levels of PAE, and so the prevalence of impairment in these populations is unknown. The only other population-based study of VMI skills in children with PAE or FASD found that first-grade children in a Midwestern US city with FASD (n = 36) had lower Beery VMI percentile rank scores than the control group (n = 98) (May et al., 2014). Only one study has examined whether children with FASD make different types of errors than children without PAE when copying shapes (Uecker & Nadel, 1996), and this information can provide insight into specific areas of difficulty, and thus guide therapeutic interventions (Beery & Beery, 2010). The Beery VMI subtests examine whether impairment in VMI could be due to underlying problems with visual perception and/or fine motor coordination (Beery & Beery, 2010), but to date, no studies have reported data from the Visual Perception or Fine Motor Coordination subtests in relation to children with PAE or FASD.

Conflicting evidence exists for impairment of visual perception (Aronson, Kyllerman, Sabel, Sandin, & Olegard, 1985; Janzen, Nanson, & Block, 1995; Uecker & Nadel, 1996) or fine motor coordination skills (Doney et al., 2014) in children with PAE or FASD, which may be due to the wide range of assessment tools used in different studies. The advantage of using the Beery VMI subtests to assess visual perception and fine motor coordination is that they use the same shapes as the core component of the Beery VMI, which the authors claim 'makes comparisons between performances on all three tests as valid as possible' (Beery & Beery, 2010, p.99).

Understanding whether impairment in VMI could be due to underlying visual perception or fine motor deficits, or whether these skills are intact and the problem lies with their integration, will assist in developing a more accurate neurological profile for children with PAE and FASD, and inform therapeutic interventions which target specific deficits. Additionally, the subtests are timed, and this information may be useful to determine if sustained visual attention, processing speed, or slower motor response time, which have previously been reported as impaired in children with FASD (Mattson, Calarco, & Lang, 2006; Simmons, Wass, Thomas, & Riley, 2002) may affect VMI performance.

The aims of our study were to:

1. Compare VMI, visual perception, and fine motor coordination, including types of errors and time to complete subtests, in 7 to 9 year old children in the Fitzroy Valley, i) without PAE; ii) with PAE who did not meet criteria for one of the FASD diagnoses; and iii) with FASD.

2. Investigate whether VMI deficits, if any, could be due to visual perception or fine motor coordination impairment.

3. Determine the prevalence of VMI, visual perception, and fine motor coordination impairments in children without PAE; with PAE but no FASD diagnosis; and with FASD.

Based on the literature, we hypothesized that children with FASD would have significantly lower VMI, visual perception, and fine motor coordination scores; would take longer to complete subtests; and would make different types of errors than children without PAE. We also anticipated that children with FASD would have a significantly higher prevalence of impairment than children without PAE.

4.2.3 Methods

Setting

In this paper we report VMI data from the Lililwan Project, a population-based study of FASD prevalence in the remote Fitzroy Valley region of Western Australia. The Fitzroy Valley has a population of 4,500 people living in approximately 45 communities within a 200km radius of the main service town of Fitzroy Crossing (Morphy, 2010). Approximately 80% of the population identify as being of Australian Aboriginal descent, representing five Aboriginal cultural groups. As previously published, Stage 1 of the Lililwan Project (2010) involved interviews with parents or caregivers of children born in 2002 or 2003 (*n* = 127; 95% participation) regarding antenatal risk factors and childhood exposures which may affect development, including exposure to alcohol, nicotine, and marijuana *in-utero*, socioeconomic factors such as employment and household structure, and environmental exposures such as nutrition and early life trauma (Fitzpatrick et al., 2013). PAE was based on retrospective self-report, in most cases by the birth mother, but in some instances by family members who were living with the birth mother during her pregnancy. Where possible, information was corroborated by review of maternal medical records. PAE data were deemed to be 'very reliable' in most instances (Fitzpatrick et al., 2015). PAE levels were classified as 'no exposure', 'low risk', 'risky', 'high risk', or 'uncertain risk' (when PAE was confirmed, but levels were unknown) according to the frequency, pattern, and amount of alcohol consumption as defined within the Alcohol Use Disorders Identification Test – Consumption (AUDIT-C), which is a standardized and validated measure of PAE (Bush, Kivlahan, McDonell, Fihn, & Bradley, 1998; Fitzpatrick et al., 2015). Stage 2 of the Lililwan Project (2011) involved a comprehensive neurodevelopmental assessment, including of VMI skills, for FASD by a multidisciplinary team including a pediatrician, physiotherapist, psychologist, occupational therapist, and speech pathologist (*n* = 108; 81% participation) (Fitzpatrick et al., 2012). FASD diagnoses were assigned according to modified Canadian FASD diagnostic guidelines (Chudley et al., 2005), which require severe impairment in a minimum of three (out of ten) neurodevelopmental domains. A detailed study protocol outlining procedures, assessments, and criteria for diagnosing FAS, pFAS, or ND-AE has been published (Fitzpatrick et al., 2012). Non-participation in Stage 2 was due to families moving away from the Fitzroy Valley after Stage 1 (*n* = 15); withdrawal of consent (*n* = 1); and being unable to locate the child for assessment (*n* = 3).

The Fitzroy Valley is located in a very remote region of Western Australia. General practitioners, a midwife, and a Child Health Nurse are located in Fitzroy Crossing (the main town site), but allied health and pediatric services are provided via an outreach, visiting service from Derby, 260km (162 miles) to the west. Remote communities may only receive visits from pediatric and allied health services once or twice a year, thus diagnostic and regular therapeutic services are extremely limited. One of the goals of the Lililwan Project was to highlight the needs of children in the area, and campaign for improved services. Prior to the Lililwan Project, only a small number of children had been reported as having a type of Fetal Alcohol Spectrum Disorder, and none had received a comprehensive neurodevelopmental assessment and formal diagnosis made by a multidisciplinary team.

The Lililwan Project provided both a clinical diagnostic service and enabled a population-prevalence study (Fitzpatrick et al., 2012). Each child and family received a comprehensive assessment report and therapy plan, including FASD diagnoses when relevant. Reports were provided to local health services and schools with parental consent. Referrals were made for ongoing therapy if warranted. Families whose child was diagnosed with FASD were offered support from an Aboriginal FASD educator and social worker.

Ethics

Ethical approval for the Lililwan Project was provided by the University of Sydney Human Research Ethics Committee; Western Australian (WA) Aboriginal Health and Information Ethics Committee; WA Country Health Services Board Research Ethics Committee; and Kimberley Aboriginal Health Planning Forum Research Sub-committee. Analysis and publication of VMI data was approved by the Curtin University Human Research Ethics Committee and the Western Australian (WA) Aboriginal Health and Information Ethics Committee. The Lililwan Project was initiated by local Aboriginal leaders, and extensive community consultation and engagement utilized through all stages of the project). Caregivers were provided with study information in written and verbal English and/or local language, and provided signed consent for participation of themselves and their child. Families and children could withdraw from the project at any stage without repercussions.

Participants

The majority of children (98.1%) identified as Australian Aboriginal, and were aged 7.5 to 9.6 years at the time of assessment (M = 8.7 years) (Table 4.4). Most children with PAE (*n* = 60) were exposed at risky (AUDIT-C score of 4 or 5; *n* = 4) or high

risk (AUDIT-C score 6 to 12; *n* = 47) levels according to AUDIT-C criteria (Bush et al., 1998). FASD diagnoses made during the project (*n* = 21) included FAS (*n* = 1), pFAS (*n* = 12), and ND-AE (*n* = 8) (Table 4.4). No children were identified with strabismus, amblyopia, or other visual defects detected other than the reduced visual acuity reported in Table 4.4. Aside from marijuana, no other prenatal illicit drug use was reported.

Most children (*n* = 107) lived with either their biological mother or father (72.9%), but less than half lived with both biological parents (42.1%). Only half of the primary carers (*n* = 106) were involved in full-time employment (51.9%), with the remainder employed either on a part-time basis (10.4%) or unemployed (16.0%). Many children lived in overcrowded households (M = 6.1, range 2 -16), and many had lived in more than four homes since birth (*n* = 17, 15.8%). Education level of the birth mother (*n* = 86) included lower high school (Year 10 or less) (46.5%); upper high school (Year 11 or 12) (50.0%), or 'unknown' (3.5%). No birth mother attained further or tertiary education.

Clinical Psychologists assessed cognitive abilities using the Universal Non-verbal Intelligence Test (UNIT) (Bracken & McCallum, 1998). Similar to other neurodevelopmental assessment tools, including the Beery VMI, no normative data are available for Australian Aboriginal children. The UNIT was chosen as being the most appropriate measure of cognitive abilities for our cohort because it does not rely on verbal, language, or hearing abilities. UNIT full-scale standard scores did not differ between children with and without PAE and/or FASD (No PAE M = 89.9 (*SD* 8.5); PAE, no FASD M = 89.4 (*SD* 9.1); FASD M = 85.0 (*SD* 12.3); *p* = .329). Two children (both subsequently diagnosed with a type of FASD) had a UNIT standard score below 70, and thus met the criteria for an intellectual disability.

Table 4.4

Cohort characteristics

	Total Cohort[a] *N* = 108		No PAE *n* = 43		PAE (no FASD) *n* = 39		FASD *n* = 21	
	n	(%)	*n*	(%)	*n*	(%)	*n*	(%)
Gender								
Male	57	(52.8)	24	(55.8)	18	(46.2)	13	(61.9)
Female	51	(47.2)	19	(44.2)	21	(53.8)	8	(38.1)
Australian Aboriginal	106	(98.1)						
Handedness								
Right	101	(93.5)	41	(95.3)	38	(97.4)	19	(90.5)
Left	7	(6.5)	2	(4.7)	1	(2.6)	2	(9.5)
Visual acuity[b,c]								
Normal	88	(81.5)	37	(86.0)	31	(79.5)	16	(76.2)
Reduced	11	(10.2)	4	(9.3)	2	(5.1)	4	(19.0)
Missing	9	(8.3)	2	(4.7)	6	(15.4)	1	(4.8)
Hearing[d]								
Normal	42	(38.9)	16	(37.2)	14	(35.9)	10	(47.6)
Mild loss	38	(35.2)	15	(34.9)	13	(33.3)	7	(33.3)
Moderate loss	13	(12.0)	7	(16.3)	3	(7.7)	3	(14.3)
Missing	15	(13.9)	5	(11.6)	9	(23.1)	1	(4.8)
Prenatal nicotine exposure[e]								
No	34	(31.5)	25	(58.1)	6	(15.4)	3	(14.3)
Yes	67	(62.0)	18	(41.9)	32	(82.1)	15	(71.4)
Unknown	7	(6.5)	0	(0.0)	1	(2.6)	3	(14.3)
Prenatal marijuana exposure[e]								
No	88	(81.5)	41	(95.3)	28	(71.8)	18	(85.7)
Yes	13	(12.0)	2	(4.7)	10	(25.6)	1	(4.8)
Unknown	7	(6.5)	0	(0.0)	1	(2.6)	2	(9.5)
PAE risk levels[f]								
No exposure	43	(100.0)	0	(0.0)	0	(0.0)	0	(0.0)
Low (1-3)	4	(3.7)	0	(0.0)	4	(10.3)	0	(0.0)
Risky (4-5)	4	(3.7)	0	(0.0)	3	(7.7)	1	(4.8)
High risk (≥ 6)	46	(42.6)	0	(0.0)	29	(74.4)	17	(81.0)
PAE, uncertain risk	6	(5.6)	0	(0.0)	3	(7.7)	3	(14.3)
Unknown PAE	5	(4.6)	0	(0.0)	0	(0)	0	(0.0)

[a] 'Total cohort' includes *n* = 5 children with unknown PAE who are not included in the No PAE, PAE (no FASD), or FASD groups

[b] Reduced visual acuity defined as ≤6/9 in one or both eyes

[c] Not all children completed audiology and ophthalmology testing

[d] Mild hearing loss 26 – 40dB; moderate hearing loss 41 – 55dB

[e] Some prenatal exposure information not available, either due to the primary carer not knowing, or the birth mother choosing not to disclose this information

[f] Risk level according to AUDIT-C scoring criteria

Assessment procedures

Children in the Lililwan Project were assessed in their local community or school in the presence of a local Aboriginal facilitator. Children were screened for hearing and visual impairments by an audiologist and ophthalmologist, and these results were reviewed prior to VMI assessment so accommodations could be made if required, such as ensuring the child was wearing prescribed glasses, or that task requirements were demonstrated if the child had a hearing impairment. The Beery VMI was administered in a single sitting and scored by the primary author (RD), who is a qualified occupational therapist with experience working with children in the Fitzroy Valley and other Kimberley communities. The assessor was blinded to PAE and maternal and child history during the assessment. The multidisciplinary team remained blinded to PAE until consensus was reached regarding whether the child met FASD diagnostic criteria.

The Beery-Buktenica Developmental Test of Visual-Motor Integration

The Beery VMI was used to assess VMI, visual perception, and fine motor coordination (Beery & Beery, 2010). The Beery VMI is a standardized, norm-referenced assessment of graphomotor VMI suitable for ages 2 to 100 years. Since its development in 1961, the Beery VMI has consistently demonstrated strong validity, including concurrent validity with the Bender-Gestalt (mean r = .56), the Developmental Test of Visual Perception (r = .62 to .75), and the drawing subtest of the Wide Range Assessment of Visual Motor Abilities (r = .52) (Beery & Beery, 2010). It also has been shown to have sound content reliability, test-retest reliability, and inter-rater reliability (Beery & Beery, 2010). The Beery VMI norms were used, which are based on data from 1737 US children and adolescents aged 2 to 18 years, and 1021 adults aged 19 to 100 years (Beery & Beery, 2010). Although Beery VMI norms do not exist for Australian Aboriginal children, the authors of the Beery VMI claim it is suitable for use with children from different backgrounds, because it does not require alphabetical or numerical knowledge (Beery & Beery, 2010). Tasks include copying a series of 24 increasingly complex geometric shapes. Scoring ceases after three consecutive errors. Shapes are scored as ‘correct’ or ‘incorrect’ according to detailed criteria, with higher scores indicating better performance. Scores are reported as standard scores, descriptive categories, and percentile ranks, and range from: < 70 (‘very low’, < 2nd percentile); 70 to 79 (‘low’, 2nd to 7th percentile); 80 to 89 (‘below average’, 8th to 16th percentile); 90 to 109 (‘average’, 17th to 65th percentile); 110 to 119 (‘above average’, 66th to 72nd percentile); 120 to 129 (‘high’, 73rd to 97th percentile); and > 129 (‘very high’, > 98th percentile). The different types of errors

made by each child were counted and assigned one point per error. Errors were classified as ratio or spatial (if the shape was copied disproportionately or oriented incorrectly on the page); corner difficulties (rounded or open); 'dog-earing' (insertion of an extra line or angle due to difficulty turning a corner); reversal or directionality confusion; errors that may indicate difficulties with crossing the midline; and gross distortions in which the original shape was difficult to discern (*Figure 4.3*).

The Beery VMI Visual Perception and Fine Motor Coordination subtests were also administered to all children. These subtests can be used to identify whether VMI difficulties could be due to underlying problems with either visual perception or fine motor coordination (Beery & Beery, 2010). The Visual Perception subtest is a relatively motor-free measure of visual perception skills, requiring the child to point at the shape they identify as matching a stimulus. The Visual Perception subtest is timed, and scoring ceases after 3 minutes, or once the child makes three consecutive errors. The Fine Motor Coordination subtest requires the child to trace the outline of shapes while staying within double-lined borders. It includes the same shapes as the core Beery VMI test and the Visual Perception subtest. The Fine Motor Coordination subtest minimizes visual perception requirements by using examples, starting dots, and paths as visual guides (Beery & Beery, 2010). The Fine Motor Coordination subtest is timed and scoring ceases after 5 minutes, but the number of errors allowed is unlimited. Each subtest uses the same shapes as the core component of the Beery VMI, which increases the validity of comparisons between the core test and the subtests (Beery & Beery, 2010).

Statistical analysis

Means (M), standard deviations (SD), and confidence intervals for the mean (CI) were calculated from the standard scores from the Beery VMI and the Visual Perception and Fine Motor Coordination subtests; counts of error types made by each child were summed; and the number of subtest items completed in the timeframe recorded. Results were reported for the total cohort, and scores compared between children without PAE ('No PAE' group); PAE (including low, risky, high-risk, and uncertain risk levels) who were not diagnosed with any type of FASD because they did not have significant impairment in a minimum of three neurodevelopmental domains ('PAE (no FASD)' group); and a FASD diagnosis ('FASD' group). The FASD group included all diagnoses (FAS; pFAS; and ND-AE) as there were insufficient numbers in each diagnostic grouping to perform separate comparisons. The small numbers in each group made statistical adjustment for potential confounders infeasible. Data were assessed for normality using the

Shapiro-Wilk test. Normally distributed data were analyzed using a one-way between groups analysis of variance (ANOVA) to compare scores between groups, and Tukey's Honestly Significant Difference test (HSD) was used to determine which groups differed from each other. Non-parametric data were analyzed using the Kruskal-Wallis test. Effect sizes (eta^2) were calculated, with 0.01 being considered a small effect size; 0.06 a medium effect size; and 0.14 a large effect size (Portney & Watkins, 2000). Significance was set at $p < 0.05$. The prevalence of impairment in VMI, visual perception, and fine motor coordination in each group was reported at two different levels: i) 'moderate' impairment (one or more *SD* below the mean; or ≤ 16th percentile); and ii) 'severe' impairment (two or more *SD* below the mean; or ≤ 2nd percentile). Chi-square analysis was used to determine if prevalence rates differed between groups. Statistical analysis was completed using IBM SPSS Statistics for Windows version 21.0 (Armonk, NY: IBM Corp.).

4.2.4 Results

Visual-motor integration

The mean VMI scores were in the 'below average' range for the total cohort (M = 87.8), and for children without PAE (M = 89.6); PAE (no FASD) (M = 87.0); and FASD (M = 84.2), and differences between groups were not significant (F (2,100) = 2.5; p = 0.091) (Table 4.5). Ratio/ spatial errors were the most common error type when copying shapes for all groups (Table 4.6). Children with PAE (no FASD) or FASD were two to three times more likely than children without PAE to draw shapes that were distorted, reversed, or had 'dog earing' errors (Table 4.6; and *Figure 4.3*), but there were no significant differences in VMI scores or types of errors between groups (Table 4.5 and Table 4.6).

Visual perception

The mean Visual Perception scores were in the 'average' range for the cohort (M = 97.6), and also for all groups, and differences between groups were not significant (F (2,100) = 1.5; p = 0.239) (Table 4.5). There were no significant differences between groups in the visual perception scores (Table 4.5), including the number of tasks completed in the allotted timeframe (No PAE M = 29.0 (SD 1.7); PAE (no FASD) M = 29.0 (SD 2.0); FASD M = 28.6 (SD 2.8), p = 0.861).

Fine motor coordination

The Fine Motor Coordination subtest scores for the overall cohort were in the 'average' range, but were 'below average' and significantly lower in children with

FASD (M = 87.9, p = .020) than those without PAE (M = 95.1, HSD p = 0.046) and with PAE (no FASD) (M = 96.1; F (2,100) = 4.1; HSD p = 0.020) (Table 4.5). There were no differences between groups regarding the number of items completed in the timeframe (No PAE M = 28.1 (SD 2.0); PAE (no FASD) M = 28.3 (SD 2.1); FASD M = 27.5 (SD 3.1), p = 0.628).

Prevalence of impairments

Few children in the cohort had severely impaired VMI (*n* = 2; 1.9%), visual perception (*n* = 1; 0.9%), or fine motor coordination (*n* = 3; 2.8%) (Table 4.7). However, nearly half of the children in the cohort had moderate VMI impairment (47.2%). Although children with FASD had the highest rates of moderate VMI impairment (66.7%), rates were also high for children without PAE (41.9%) and with PAE (no FASD) (43.6%), and differences between groups were not statistically significant. Prevalence rates for moderate impairment in visual perception in our cohort (17.6%) were similar to norms, and similar between groups. Rates of moderate impairment in fine motor coordination were slightly higher (26.9%) in the total cohort than expected based on Beery VMI norms (16.0%), but were significantly higher in children with FASD (52.4%) compared to children without PAE (23.3%, *p* = 0.007) (Table 4.7).

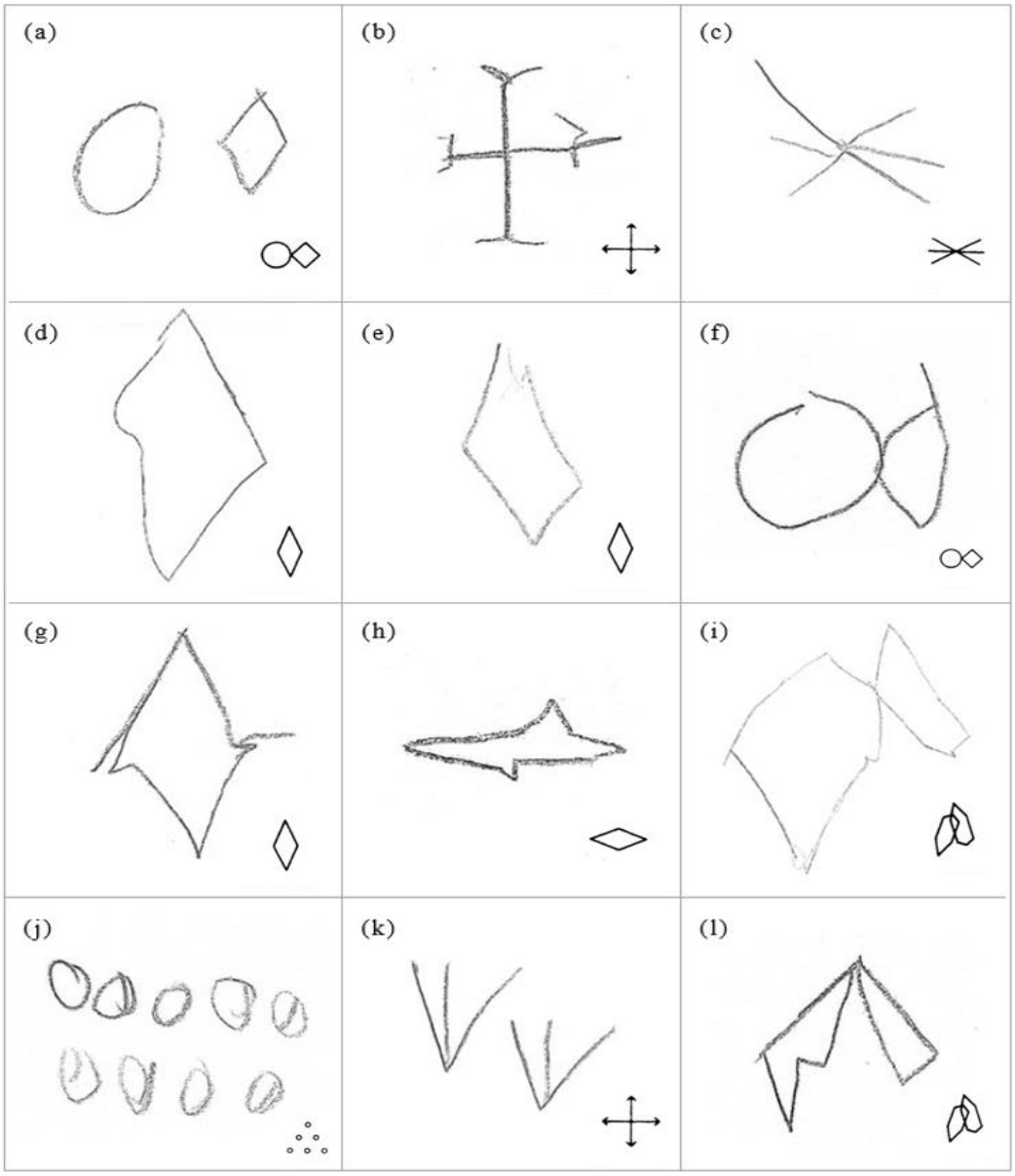

Figure 4.3 Examples of errors made when copying Beery VMI shapes

a = spatial error; b = reversal/ directionality confusion; c = crossing the midline error; d-f = corner difficulties (d = rounded corner; e = open corner; f = both rounded and open corner); g-i = dog-earing errors; j-l = distortions. The correct shape is shown in the bottom right corner of each box.

Table 4.5

Beery VMI, visual perception, and fine motor coordination standard scores for the cohort, and according to PAE and FASD status

	Total Cohort[a] *N* = 108			No PAE *n* = 43			PAE (no FASD) *n* = 39			FASD *n* = 21			Effect size[b] (eta^2)	ANOVA[c] (*p*)
	M	(*SD*)	95% CI	M	(*SD*)	95% CI	M	(*SD*)	95% CI	M	(*SD*)	95% CI		
Beery VMI[d]	87.8	(9.6)	86.0 – 89.6	89.6	(10.4)	86.4 - 92.8	87.0	(8.3)	84.3 - 89.6	84.2	(8.3)	80.4 - 88.0	0.05	0.091
Visual Perception[d]	97.6	(12.5)	95.2 –100.0	97.9	(12.7)	94.0 - 101.8	99.4	(11.9)	95.5 - 103.2	93.7	(13.1)	87.7 - 99.6	0.03	0.239
Motor Coordination[d]	93.9	(11.6)	91.6 – 96.1	95.1	(10.7)	91.8 - 93.8	96.1	(10.9)	92.6 - 99.6	87.9	(12.5)	82.2 - 93.6	0.08	0.020*[e]

* $p < 0.05$

[a] 'Total Cohort' includes $n = 5$ children with unknown PAE who are not included in the No PAE, PAE (no FASD), or FASD groups

[b] 0.01 = a small effect size; 0.06 = medium effect size; 0.14 = large effect size

[c] Between group differences for No PAE; PAE (no FASD); and FASD

[d] The Beery VMI, Visual Perception, and Motor Coordination standard scores have a M = 100, $SD = 15$

[e] Tukey's Honestly Significant Difference test: No PAE > FASD ($p = .046$); and PAE (no FASD) > FASD ($p = 0.020$)

Table 4.6

Beery VMI Error types for the cohort, and according to PAE and FASD status

	Total Cohort[a] *N* = 108			No PAE *n* = 43			PAE (no FASD) *n* = 39			FASD *n* = 21			Kruskal-Wallis (*p*)
	M	(*SD*)	95% CI	M	(*SD*)	95% CI	M	(*SD*)	95% CI	M	(*SD*)	95% CI	
Error type													
Ratio/spatial	3.3	(1.3)	3.1 – 3.6	3.7	(1.4)	3.3 - 4.2	3.1	(1.4)	2.6 - 3.6	3.0	(1.0)	2.5 - 3.4	0.052
Corner (rounded/open)	0.4	(0.6)	0.2 – 0.5	0.4	(0.7)	0.2 - 0.6	0.3	(0.6)	0.2 - 0.5	0.3	(0.5)	0.1 - 0.5	0.974
Dog earing	0.1	(0.4)	0.1 – 0.2	0.1	(0.2)	0.0 - 0.1	0.2	(0.5)	0.0 - 0.4	0.2	(0.4)	0.0 - 0.4	0.159
Reversal/directionality	0.3	(0.5)	0.2 – 0.3	0.1	(0.4)	0.0 - 0.3	0.4	(0.5)	0.2 - 0.5	0.2	(0.4)	0.0 - 0.4	0.070
Crossing the midline	0.1	(0.4)	0.1 – 0.2	0.1	(0.0)	0.0 - 0.2	0.1	(0.0)	0.0 - 0.2	0.2	(0.4)	0.0 - 0.4	0.801
Distortions	0.3	(0.6)	0.2 – 0.4	0.2	(0.6)	0.0 - 0.4	0.4	(0.5)	0.2 - 0.6	0.5	(0.7)	0.2 - 0.8	0.065

[a] 'Total Cohort' includes *n* = 5 children with unknown PAE who are not included in the No PAE, PAE (no FASD), or FASD groups

Table 4.7

Prevalence of visual-motor integration, visual perception, and fine motor coordination impairments for the cohort and according to PAE and FASD status

	Total Cohort[a] *N* = 108		No PAE *n* = 43		PAE (no FASD) *n* = 39		FASD *n* = 21		Chi-square *(p)*
	n	(%)	*n*	(%)	*n*	(%)	*n*	(%)	
Beery VMI									
≥ -2*SD*[b]	2	(1.9)	1	(2.3)	1	(2.6)	0	(0.0)	0.768
≥ -1 *SD*[c]	51	(47.2)	18	(41.9)	17	(43.6)	14	(66.7)	0.144
Visual Perception									
≥ -2*SD*[b]	1	(0.9)	1	(2.3)	0	(0.0)	0	(0.0)	0.494
≥ -1*SD*[c]	19	(17.6)	9	(20.9)	4	(10.3)	5	(23.8)	0.309
Motor Coordination									
≥ -2*SD*[b]	3	(2.8)	1	(2.3)	0	(0.0)	2	(9.5)	0.107
≥ -1*SD*[c]	29	(26.9)	10	(23.3)	6	(15.4)	11	(52.4)	0.007*

* $p < 0.05$

[a] 'Total Cohort' includes $n = 5$ children with unknown PAE who are not included in the No PAE, PAE (no FASD), or FASD groups

[b] ≥ - 2*SD* = ≤ 2nd percentile

[c] ≥ - 1*SD* = ≤ 16th percentile

4.2.5 Discussion

This study is the first in-depth report of VMI, visual perception, and fine motor coordination skills in a population-based cohort of Aboriginal children living in remote Australia. The population had 'below average' VMI scores, regardless of the presence or absence of PAE or FASD, but had 'average' Visual Perception and Fine Motor Coordination (except in children with FASD), which may indicate that the children had difficulties coordinating or integrating these two skill sets (Beery & Beery, 2010).

Children with FASD had significantly lower fine motor coordination scores and higher rates of moderate impairment in fine motor coordination than children without PAE. Only one other study has investigated whether VMI difficulties in children with PAE could be due to visual perceptual or fine motor deficits (Janzen at al., 1995). In that small study ($n = 20$), 3.5 to 5 year old children with FAS ($n = 10$) had significantly lower VMI scores than a matched control group but, similar to our findings, visual perception (assessed using the Recognition-Discrimination Test from the Florida Kindergarten Screening Battery; Satz & Fletcher, 1982) was unaffected. When considered along with other fine motor outcomes from the study, the authors concluded that the impairment in VMI was more likely due to impaired fine motor

skills than visual perception. However, in our study, even children without PAE or FASD had poor VMI performance, so it is inconclusive as to whether the fine motor coordination problems experienced by children with FASD contributed to their VMI problems, but it is likely they had some impact on VMI performance.

Few children in our study had severe impairment in VMI, visual perception, or fine motor coordination. However, rates of moderate impairment in VMI were very high across the cohort, particularly in children with FASD, who had VMI impairment more than four times higher (66.7%) than expected based on published norms (Beery & Beery, 2010). Rates of moderate impairment in Visual Perception were similar to norms for all groups. More than half of the children with FASD had moderate impairment of fine motor coordination (52.4%), which was significantly higher than children without PAE (23.3%) and children with PAE (no FASD) (15.4%). Our results have important implications for FASD diagnoses and therapeutic interventions. The Canadian FASD Diagnostic Guidelines propose that for the purpose of FASD diagnosis only impairments below the 2nd percentile (or when there is a discrepancy of 1.5 to 2 *SD* between subtests) should contribute towards a FASD diagnosis (Chudley et al., 2005). However the CDC FASD diagnostic guidelines suggest that impairments below the 16th percentile should contribute to a FASD diagnosis, citing evidence that only a quarter of children with FAS (the most severe type of FASD diagnosis) have neurodevelopmental impairment below the 2nd percentile, and that using a cut-off of 2 *SD* would preclude many affected children from a diagnosis (Centers for Disease Control and Prevention, 2005). The more conservative cut-off of 1 *SD* has also been proposed for use when diagnosing ND-PAE within the DSM-5 guidelines (Doyle & Mattson, 2015). Only two children in our study had impairment in VMI below the 2nd percentile, which seems conservative given the very high levels of PAE, FASD, and other neurodevelopmental risk factors evident in the cohort. Conversely, the cut-off for impairment proposed by the CDC guidelines may have over-estimated rates of impairment in VMI impairment in children with PAE or FASD. This issue warrants further consideration and investigation.

The lack of significant differences in VMI between children with PAE or FASD compared to children without PAE in our study is inconsistent with other studies. The Beery VMI has detected impairments in children with moderate to high PAE (>10 drinks/ week) (Korkman, Kettunen, & Autti-Rämö, 2003), as well as in children with FASD (Astley et al., 2009). In another study significant differences in VMI scores were found in children with FAS, but not heavy PAE without FAS (Mattson, Riley, Gramling, Delis, & Jones, 1998). In most studies children with PAE or FASD

were compared to typically developing children. It may be that our non-significant findings are due to our relatively small sample size. Additionally, our non-significant findings may reflect the multitude of neurodevelopmental risk factors experienced by the children in our cohort, which is similar to many Aboriginal children in remote regions. Even children without PAE may have been exposed to intergenerational and early life trauma along with loss of land and culture, restricted access to therapeutic and early childhood services, overcrowded living conditions and poor nutrition (Australian Health Ministers' Advisory Council, 2012). Many children in our cohort, even those without PAE, had high levels of prenatal nicotine exposure. Some studies have shown prenatal nicotine exposure may adversely affect VMI skills (Cornelius, Ryan, Day, Goldschmidt, & Willford, 2001; Willford, Chandler, Goldschmidt, & Day, 2010), although these studies have used assessment tools other than the Beery VMI and it remains uncertain whether graphomotor VMI skills are affected by prenatal nicotine exposure. Prenatal marijuana exposure was also high, particularly in the group with PAE who were not diagnosed with FASD, but similarly no studies have examined the impact on graphomotor VMI skills. Many children in the Fitzroy Valley participate in multiple outdoor and cultural activities, such as hunting and recreational sports (Lucas et al., 2016), which may promote visual perception skills. Indoor activities which promote VMI and fine motor skills in the early years, such as arts and crafts, are less common and may also have contributed to the below average VMI skills in our cohort.

We also examined the types of errors children made when copying VMI shapes on the core test component, and the time taken to complete the Visual Perception and Fine Motor Coordination subtests. Ratio or spatial errors were the most common type of error made by all groups. Children with PAE or FASD were up to four times more likely to draw shapes which were distorted, reversed, or had 'dog earing' errors, although differences between groups were not significant. The types of Beery VMI errors have been investigated in one other group of children with FASD (Uecker & Nadel, 1996). These researchers found that children with FAS (n = 15) made more dog-earing errors and drew shapes which were grossly distorted compared to a control group, although, similar to our study, differences between groups were not statistically significant. 'Dog earing' and reversal type errors can indicate immature VMI development (Beery & Beery, 2010), which has been associated with difficulties with handwriting and other academic skills including reading, writing, and spelling (Kulp, 1999; Volman, van Schendel, & Jongmans, 2006; Weil & Cunningham Amundson, 1994).

There were no differences between groups in the number of tasks completed in the allotted time for the Visual Perception or Fine Motor Coordination subtests, suggesting that children with PAE or FASD neither worked more slowly, nor were more impulsive, than children without PAE. Other studies have shown that children with heavy PAE have difficulty sustaining visual attention (Mattson et al., 2006), as well as delayed central processing and motor response time (Simmons et al., 2002). Accordingly, it was anticipated that the children in our study with PAE or FASD would take longer to complete the Visual Perception or Fine Motor Coordination subtests, but this hypothesis was not supported.

Strengths

This is the first population-based report of VMI skills in Aboriginal children in a remote region of Australia using a standardized, norm-referenced assessment tool. The study had a very high participation rate, and assessed almost two entire age cohorts of children in the Fitzroy Valley. The choice of assessment tools was unique in that we used the optional Beery VMI Visual Perception and Fine Motor Coordination subtests, which have not been reported in other studies of children with FASD. Use of the Beery VMI subtests, rather than different assessment tools, increases the likelihood that any observed difficulties are actual deficits rather than due to variation between different assessments (Beery & Beery, 2010).

Limitations

This study had several limitations. First, it was conducted in a remote area of Western Australia, so results may not be applicable to other populations with different demographics and social conditions. Conversely, outcomes may be applicable to other children living in similar remote communities. Second, PAE was reported retrospectively by parents or caregivers, which may introduce recall bias. However, information was corroborated by review of medical records and reports from direct observers, such as family members. There is some evidence that retrospective reporting is accurate up to 14 years after birth (Alvik, Haldorsen, Groholt, & Lindemann, 2006). Third, although the study included two entire age cohorts of children in the region and participation rates were high, the relatively small sample size meant that we could not statistically control for potential confounders, including prenatal nicotine and marijuana exposure, and future studies should consider the impact of these and other factors on neurodevelopment, including VMI. Fourth, the Beery VMI has not been validated for use with Australian Aboriginal children, and norms are based on a US normative sample. The low

overall VMI scores may indicate that the Beery VMI was invalid for use with our cohort, and there are concerns about the cross-cultural validity of neurodevelopmental tests developed for, and normed with, populations with different linguistic or cultural backgrounds (Thorley & Lim, 2011). However, the Beery VMI was selected because the test developers claim it is 'virtually culture-free' and does not require alphabet or numerical knowledge (Beery & Beery, 2010), and is endorsed as an appropriate FASD diagnostic assessment tool (Chudley et al., 2005). Additionally, children in our study had been attending school for a number of years, and thus should have been familiar with completing similar tasks to those of the Beery VMI. Fifth, this paper reports VMI data from a population-based prevalence study. Children with FASD were more likely to have VMI impairments, because significant impairment in VMI could contribute to the FASD diagnosis. However, FASD diagnoses were only made in the presence of confirmed PAE and at least three domains of severe impairment, which may or may not include VMI.

Future directions

Future studies should examine potential confounders, other than PAE, which may account for poor VMI skills observed in our cohort. Consideration should also be given to what degree of impairment should be considered 'significant' in terms of FASD diagnosis. Studies in other populations have shown the effectiveness of therapeutic interventions to improve handwriting proficiency, including in children with poor VMI skills (Case-Smith; 2002) and as a collaborative approach between Occupational Therapists and classroom teachers (Case-Smith, Weaver, & Holland, 2014). Trialling similar programs for children in the Fitzroy Valley may be of benefit.

Conclusions

The VMI skills of Aboriginal children in the Fitzroy Valley region in our cohort were below average, indicating that many children in the region require therapeutic support regardless of whether they have PAE or FASD. Visual perception and fine motor coordination performance were within normal ranges for the cohort except in those with FASD, who had poorer fine motor coordination skills than children without PAE. Our findings indicate that both VMI and fine motor coordination should be assessed in children with PAE, and as part of the FASD diagnostic process, to give a more accurate profile of neurological impairment. This knowledge can be used to guide the development of therapeutic interventions which target specific areas of impairment.

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4.3 Graphomotor Skills

This section is the peer-reviewed, published paper that reports results from clinical observations, the ETCH (Amundson, 1995) and M-FUN (Miller, 2006).

> Doney, R., Lucas, B. R., Jirikowic, T., Tsang, T. W., Watkins, R. E., Sauer, K., Howat, P., Latimer, J., Fitzpatrick, J. P., Oscar, J., Carter, M., & Elliott, E. J. (2016). Graphomotor skills in children with prenatal alcohol exposure and Fetal Alcohol Spectrum Disorder: A population-based study in remote Australia. *Australian Occupational Therapy Journal*, *64*(1), 68-78.

4.3.1 Abstract

Background/ Aim: Few studies have examined graphomotor skills in children with prenatal alcohol exposure (PAE) or Fetal Alcohol Spectrum Disorder (FASD).

Methods: Graphomotor skills were assessed in 108 predominantly Australian Aboriginal children aged 7.5 to 9.6 years in remote Western Australia using clinical observations (pencil grasp; writing pressure) and standardised assessment tools (the Evaluation Tool of Children's Handwriting; and the Miller Function and Participation Scales – The Draw-a-Kid Game). Skills were compared between children i) without PAE; ii) PAE but not FASD; and iii) FASD.

Results: Most children used a transitional pencil grasp and exerted heavy handwriting pressure (83.3% and 30.6% of the cohort). The percentage of letters (M = 62.9%) and words (M = 73.3%) written legibly was low. Children with FASD were more likely than children without PAE to use a cross-thumb grasp ($p = 0.027$); apply heavy writing pressure ($p = 0.036$); be unable to write a sentence ($p = 0.041$); and show poorer word legibility ($p = 0.041$). There were no significant differences between groups for drawing outcomes, although some children with FASD drew pictures which appeared delayed for their age. There were no significant differences between children without PAE and those with PAE but who were not diagnosed with FASD.

Conclusions: Overall, graphomotor skills were poor in this cohort, but children with FASD performed significantly worse than children without PAE. Findings suggest the need for improved occupational therapy services for children in remote regions, and evaluation of graphomotor skills in children with PAE.

4.3.2 Introduction

Graphomotor skills include handwriting and drawing, and involve the reproduction of letters, figures, pictures, or plans either from memory or by copying, onto paper or another writing surface using a pencil or other writing implement (Ziviani & Wallen, 2006). Graphomotor skills facilitate the recording of information, thoughts and events; are a tool for communication; and allow expression of feelings and ideas (Tomchek & Schneck, 2006). In children, successful graphomotor performance is essential for participation in numerous classroom activities to demonstrate learning, as well as recreational and play activities. Previous studies indicated that up to 60% of the school day is spent in handwriting and other fine motor tasks (McHale & Cermak, 1992). Despite recent advancements in and increased use of computers and other technology to complete academic tasks (Cahill 2009), students with poor handwriting are also likely to have difficulty with keyboarding skills (Connelly, Gee, & Walsh, 2007). Further, handwriting proficiency can influence the quality of academic work (Baker, Gersten, & Graham, 2003), and students with illegible handwriting are more likely to receive lower grades regardless of written content (Chase, 1986). Handwriting and drawing are complex developmental skills which require a complex interaction of biomechanical, psychomotor, cognitive, and linguistic abilities (Benbow, 2006).

Although research on graphomotor skills in children with prenatal alcohol exposure (PAE) is limited, PAE can disrupt the development of many neural regions which are involved in graphomotor skills, including the cerebellum, basal ganglia, corpus callosum, and motor cortex (Norman, Crocker, Mattson, & Riley, 2009; Xie, Yang, Chappell, Li, & Waters, 2010). PAE can also impair nerve conduction (de los Angeles Avaria et al., 2004), and cause skeletal malformations (Jones et al., 2010) and atypical muscle development (David & Subramaniam, 2005) which may also affect graphomotor proficiency. Individuals with PAE and significant, multiple neurodevelopmental impairments may be diagnosed with one of the Fetal Alcohol Spectrum Disorders (FASD). This umbrella term includes the diagnoses of Fetal Alcohol Syndrome (FAS), in which individuals have characteristic facial dysmorphology and significant growth impairment; partial FAS (pFAS), with fewer dysmorphic facial features and normal growth; and Alcohol Related Neurodevelopmental Disorder (ARND) or Neurodevelopmental Disorder – Alcohol Exposed (ND-AE), with few or no dysmorphic facial features and normal growth. All diagnoses require significant neurodevelopmental impairment in at least three domains of function, which may include hard and soft neurologic signs (including

sensory-motor impairment), cognition, communication, academic achievement, memory, executive functioning, attention deficit/ hyperactivity, or adaptive skills and social communication (Chudley et al., 2005). Individuals with PAE may have some degree of impairment but not at a level sufficient to be diagnosed with a type of FASD (Astley et al., 2009).

Children with FASD often have impaired fine and visual motor skills (Adnams et al., 2001; Barr, Streissguth, Darby, & Sampson, 1990; Mattson et al., 2010). However, despite anecdotal reports of graphomotor impairment in children with FASD (Clarren, 2004), few studies have reported the quality of handwriting or drawing skills in children with FASD. Those studies have either included only a small, exploratory sample (n = 20) (Duval-White, Jirikowic, Rios, Deitz, & Olson, 2013), or have not assessed human figure drawing skills within a motor performance framework (Aronson, Kyllerman, Sabel, Sandin, & Olegard, 1985; Urban et al., 2008). Functional assessments of graphomotor skills in children with PAE may assist identification of fine motor impairment during the FASD diagnostic process; improve knowledge of the functional implications of PAE; and guide therapeutic interventions.

Study aims

The purpose of this study was to describe graphomotor performance of children in the remote Fitzroy Valley region of northern Western Australia. We aimed to assess:

1. Pencil grasp, writing pressure, and ability to write their name and a short sentence, using clinical observation
2. Handwriting legibility in terms of percentage of letters and words formed correctly when writing their name and a short sentence, using the Evaluation Tool of Children's Handwriting (Amundson, 1995)
3. Drawing abilities in terms of motor accuracy and body awareness, using the Miller Function and Participation Scales (Miller, 2006)

The differences in graphomotor skills between children without PAE; children with PAE but who were not diagnosed with FASD; and children with FASD were determined. This is the first comprehensive description of graphomotor skills in Aboriginal children in remote Australia, and the first to examine whether graphomotor skills differ between children with PAE or FASD.

In accordance with the teratogenic nature of PAE on neural regions associated with graphomotor skills, it was anticipated that a) children with PAE or FASD would have poorer handwriting and drawing skills than children without PAE, and b) children with FASD would be most impaired.

4.3.3 Methods

Background and setting

The children completed graphomotor assessments as part of the Lililwan Project, which was Australia's first active case ascertainment population-based study of FASD prevalence (Fitzpatrick et al., 2012). The Lililwan Project formed part of the Marulu strategy, which is an initiative developed by local Aboriginal leaders in response to their concerns about the impact of high levels of alcohol misuse in the region, including consumption of alcohol during pregnancy. In 2010, families of children born in 2002 or 2003 who were currently living in the Fitzroy Valley were invited to participate in the Lililwan Project (*n* = 127, 95% participation). Parents or caregivers completed in-depth verbal questionnaires with 'community navigators', who were local Aboriginal people who worked with the Lililwan Project clinicians to ensure cultural safety of procedures. Families provided information regarding prenatal and postnatal exposures, including health, developmental, and socioeconomic circumstances which may have impacted on the child's development (Fitzpatrick et al., 2013). PAE was scored according to a standardised measure of alcohol consumption (the Alcohol Use Disorders Identification Test – Consumption (AUDIT-C) (Bush, Kivlahan, McDonell, Fihn, & Bradley, 1998). In 2011, the children – who were then aged from 7 to 9 years - completed approximately six hours of health and neurodevelopmental assessments with an audiologist, occupational therapist, ophthalmologist, paediatrician, physiotherapist, psychologist, and speech pathologist. Assessors were blinded to PAE and other neurodevelopmental risk factors, such as early life trauma. Clinicians conducted comprehensive case conferences for each child to determine if they met FASD diagnostic criteria. Children were assigned FASD diagnoses according to Canadian FASD Diagnostic Guidelines (Chudley et al., 2005) which were modified to suit the cultural context. A detailed study protocol has been published (Fitzpatrick et al., 2012).

As part of the multidisciplinary neurodevelopmental assessments, children completed approximately one hour of assessments, including graphomotor skills, with a qualified occupational therapist (RD) who was experienced in working with Aboriginal children in the region.

Participant consent and ethical approval

The Lililwan Project was conducted in accordance with National Health and Medical Research Council's guidelines for ethical conduct in Aboriginal and Torres Strait Islander health research (National Health and Medical Research Council, 2003).

Local Aboriginal leaders in the Fitzroy Valley conceived and designed the protocols for the Lililwan Project. Extensive community consultation occurred in the communities prior to, and throughout, the project. Families received study information verbally and written information about the study in English or their local language if preferred. Families or children could withdraw from the study or assessment process at any stage without repercussions. 'Community navigators', who were local Aboriginal people, assisted clinicians in administering assessments and interpreting results, and if requested by the family, could be present when results from neurodevelopmental assessments were provided to families.

Ethics approval was provided for the Lililwan Project by the Kimberley Aboriginal Health Planning Forum Research Sub-committee; University of Sydney Human Research Ethics Committee; Western Australian Aboriginal Health and Information Ethics Committee; and the Western Australian Country Health Services Board Research Ethics Committee. The Curtin University Human Research Ethics Committee and the Western Australian Aboriginal Health and Information Ethics Committee provided separate approval related to the fine motor, including graphomotor, aspects of the Lililwan Project.

Outcome measures

Clinical observations

Observations recorded during graphomotor tasks included i) hand dominance; ii) writing pressure, which was ranked as 'light'; 'light to appropriate'; 'appropriate'; 'appropriate to heavy'; or 'heavy'; and iii) pencil grasp, which was classified according to Schneck and Henderson's (1990) criteria as either 'primitive' (digital pronate; radial cross palmar; palmar supinate; digital pronate; brush; or extended fingers grasps); 'transitional' (cross thumb; static tripod; or four fingers grasps); or 'mature' (lateral tripod; or dynamic tripod grasps).

Evaluation Tool of Children's Handwriting

Children were asked to write their name and a short sentence of their choice. The Evaluation Tool of Children's Handwriting (ETCH) Task VI – Sentence Composition (Amundson, 1995) scoring guidelines were applied to evaluate letter legibility (name and sentence) and word legibility (sentence only). The ETCH is a criterion-referenced, standardised measure of handwriting ability suitable for primary-school aged children (Amundson, 1995). It has moderate to high intra-rater, inter-rater, and test-retest reliability for letter and word legibility, and good discriminant and concurrent validity (Duff & Goyen, 2010). Children could choose whether to use a

cursive or manuscript handwriting style, because local schools teach both styles. Handwriting samples were evaluated for correct letter formation, spacing, size, and alignment. The ETCH scores represent the percentage of i) letters which are legible in their name; ii) letters which are legible in a sentence; and iii) words which are legible in a sentence, with higher percentages indicating better performance.

The Miller Function and Participation Scales: The Draw-a-Kid Game

Children were asked to draw a picture of themselves, a friend, or family member. They were instructed to 'make it the best drawing you can'. The Miller Function and Participation Scales (M-FUN): Draw-a-Kid Game (Miller, 2006) scoring guidelines were applied. Drawings were scored according to (i) Body Awareness (possible score range 0 to 6) of the drawn figure and (ii) Motor Accuracy (possible score range 0 to 9), which were summed to give (iii) a Total Score (possible score range 0 to 15). Higher scores represent better performance. Although normative data are not available for individual tasks - including The Draw-a-Kid Game - this task forms part of the M-FUN's Visual Motor subgroup, which has been demonstrated to have good internal consistency, excellent inter-rater reliability (Miller, 2006), and strong concurrent and construct validity (Diemand & Case-Smith, 2013). Although M-FUN Visual Motor norms are only available for children aged 2.6 to 7.11 years, developers of M-FUN advise that tasks, including the Draw-a-Kid Game, are suitable for use with older children (Miller, 2006).

Statistical analysis

Graphomotor skills were assessed as part of the neurodevelopmental assessments conducted during the Lililwan Project. Clinical observations were recorded by the occupational therapist during assessment of graphomotor and other fine motor tasks. Drawing and handwriting samples were scored retrospective to the Lililwan Project by two Occupational Therapists who were blinded to the child's PAE and FASD status. Inter-rater reliability was calculated using weighted kappa (κ) with quadratic weighting for ordinal M-FUN data, and intra-class correlation coefficients (ICC: 2-way mixed model; single measures) for continuous ETCH data. Strength of agreement was interpreted as follows: 0.81 to 1.00 = excellent agreement; 0.61 to 0.80 = substantial agreement; 0.41 to 0.60 = moderate agreement; 0.21 to 0.40 = fair agreement; 0.00 to 0.20 = slight agreement; and <0.00 = poor agreement (Landis & Koch, 1977).

Descriptive statistics were derived for clinical observations of hand dominance; pencil grasp; writing pressure; and the ability of children to write their name and a short sentence. Outcomes were reported for the total cohort, and also according to

whether children i) did not have PAE ('No PAE' group); ii) had PAE but did not meet criteria for one of the FASD diagnoses ('PAE, no FASD)' group); and iii) had PAE and were diagnosed with a type of FASD ('FASD' group). Children with unknown PAE (*n* = 5) were excluded from the between-groups analysis. Drawings which were not of a human figure (*n* = 11) were excluded from the M-FUN analysis. Results from clinical observations were compared between groups using chi-square tests. Drawing (M-FUN) and handwriting (ETCH) data had non-normal distributions, so a non-parametric test (Kruskal-Wallis) was used to examine differences between groups. Statistical analysis was completed using IBM SPSS Statistics for Windows, version 21.0 (Armonk, NY: IBM Corp.).

4.3.4 Results

Cohort characteristics

Children (*n* = 108) were aged from 7.5 to 9.6 years (M = 8.7) at the time of assessment, and the majority identified as being Australian Aboriginal. Many children lived in overcrowded households (M = 6.1, range 2 -16), and many had lived in more than four homes since birth (*n* = 17, 15.8%). Children had attended an average of 1.8 schools (range one to five schools), and most children (82.4%) attended 4 to 5 days per week (Table 4.8).

Cognitive abilities were assessed by Clinical Psychologists using the Universal Non-verbal Intelligence Test (UNIT) (Bracken & McCallum, 1998). UNIT full-scale standard scores were similar between exposure groups (No PAE M = 89.9 (*SD* 8.5); PAE, no FASD M = 89.4 (*SD* 9.1); FASD M = 85.0 (*SD* 12.3); *p* = 0.329).

Clinical Observations

The majority of the children in the cohort were observed to be right handed (93.5%) and used a transitional style pencil grasp (43.5% cross-thumb; 29.6% static tripod; or 10.2% four fingers grasp). Many children exerted non-optimal writing pressure, including 'appropriate to heavy' (22.2%) or 'heavy' (30.6%). While most children could write their first name (97.2%), some were unable to write their surname (15.7%) or a short sentence (6.5%) (Table 4.9).

Children with PAE (no FASD) did not differ significantly from other groups for any of the clinical observations. Children with FASD were more likely to use a cross-thumb pencil grasp than children without PAE (*p* = 0.027); more likely to exert heavy pressure when writing (*p* = 0.036); and less likely to be able to write a sentence (*p*=0.041) (Table 4.9).

Table 4.8

Cohort characteristics

	Total Cohort[a] *N* = 108		No PAE *n* = 43		PAE (no FASD) *n* = 39		FASD *n* = 21	
	n	(%)	*n*	(%)	*n*	(%)	*n*	(%)
Australian Aboriginal	106	(98.1)						
Gender								
Male	57	(52.8)	24	(55.8)	18	(46.2)	13	(61.9)
Handedness								
Right	101	(93.5)	41	(95.3)	38	(97.4)	19	(90.5)
Hearing[b,c] (*n*=93)								
Normal	42	(38.9)	16	(37.2)	14	(35.9)	10	(47.6)
Mild loss	38	(35.2)	15	(34.9)	13	(33.3)	7	(33.3)
Moderate loss	13	(12.0)	7	(16.3)	3	(7.7)	3	(14.3)
Missing	15	(13.9)	5	(11.6)	9	(23.1)	1	(4.8)
Prenatal nicotine exposure[d]								
No	34	(31.5)	25	(58.1)	6	(15.4)	3	(14.3)
Yes	67	(62.0)	18	(41.9)	32	(82.1)	15	(71.4)
Unknown	7	(6.5)	0	(0.0)	1	(2.6)	3	(14.3)
Prenatal marijuana exposure[d]								
No	88	(81.5)	41	(95.3)	28	(71.8)	18	(85.7)
Yes	13	(12.0)	2	(4.7)	10	(25.6)	1	(4.8)
Unknown	7	(6.5)	0	(0.0)	1	(2.6)	2	(9.5)
PAE risk levels[e]								
No exposure	43	(100.0)	0	(0.0)	0	(0.0)	0	(0.0)
Low (1-3)	4	(3.7)	0	(0.0)	4	(10.3)	0	(0.0)
Risky (4-5)	4	(3.7)	0	(0.0)	3	(7.7)	1	(4.8)
High risk (≥ 6)	46	(42.6)	0	(0.0)	29	(74.4)	17	(81.0)
PAE, uncertain risk	6	(5.6)	0	(0.0)	3	(7.7)	3	(14.3)
Unknown PAE	5	(4.6)	0	(0.0)	0	(0.0)	0	(0.0)

[a] 'Total cohort' includes *n* = 5 children with unknown PAE who are not included in the No PAE, PAE (no FASD), or FASD groups.

[b] Not all children completed audiology testing.

[c] Mild hearing loss 26 – 40dB; moderate hearing loss 41 – 55dB.

[d] Some prenatal exposure information not available, either due to the primary carer not knowing, or the birth mother choosing not to disclose this information.

[e] Risk level according to AUDIT-C scoring criteria.

Handwriting

Letter and word legibility scores were relatively low across the cohort. The proportion legible words in a sentence was higher (M = 73.3%) than legible letters in the name (M = 60.8%) or sentence samples (M = 62.9%) (Table 4.10).

According to ETCH scoring criteria, letter legibility in their name (No PAE M = 62.5%; PAE, no FASD M = 60.9%; FASD M = 56.1%) or a sentence (No PAE M = 62.0%; PAE, no FASD M = 63.6%; FASD M = 60.1%) was low for all groups, and differences between groups were not significant (Table 4.10). However, for children with FASD, on average only half of the words in a sentence were written legibly (M = 50.0%) which was significantly less than children without PAE (M = 81.0%) and

children with PAE, no FASD (M =73.9%, p = 0.008) (Table 4.10). Many of the children with FASD had handwriting difficulties which were characterised by difficulties with letter and word formation in comparison to children without PAE (Figure 4.4).

Drawing

Most children scored towards the higher performance upper limits of the M-FUN Draw-a-Kid Game (possible score range 0 – 6) for Body Awareness (M = 5.2), but the score for Motor Accuracy (possible score range 0 – 9) was somewhat lower than the ceiling score (M = 7.6), as was the Total Score (possible score range 0 – 15) (M = 12.8) (Table 4.10).

M-FUN Draw-a-Kid Game scores (Body Awareness; Motor Accuracy; and Total Score) were similar for all children regardless of PAE or FASD, and there were no statistical differences between groups (Table 4.10). However, some of the drawings done by children with PAE and/or FASD showed evidence of developmental immaturity and poor pencil control, especially in comparison to children without PAE (Figure 4.5). Inter-rater reliability for the ETCH and M-FUN Draw-a-Kid Game is reported in Table 4.11.

Table 4.9

Clinical observations for the cohort, and according to PAE and FASD status

	Total Cohort[a] *N* =108		No PAE *n* =43		PAE (no FASD) *n* = 39		FASD *n* = 21		
Clinical Observations	n	(%)	n	(%)	n	(%)	n	(%)	*p* [b]
Hand dominance									
Right	101	(93.5)	41	(95.3)	38	(97.4)	19	(90.5)	0.487
Left	7	(6.5)	2	(4.7)	1	(2.6)	2	(9.5)	0.487
Pencil grasp									
Primitive grasps	0	(0.0)	0	(0.0)	0	(0.0)	0	(0.0)	-
Transitional grasps									
Cross-thumb	47	(43.5)	12	(27.9)	18	(46.2)	13	(61.9)	0.027*
Static tripod	32	(29.6)	12	(27.9)	15	(38.5)	4	(19.0)	0.271
Four fingers	11	(10.2)	7	(16.3)	3	(7.7)	1	(4.8)	0.280
Mature grasps									
Lateral tripod	3	(2.8)	2	(4.7)	0	(0)	1	(4.8)	0.390
Dynamic tripod	15	(13.9)	10	(23.3)	3	(7.7)	2	(9.8)	0.104
Writing pressure (*n* = 107)									
Light	0	(0.0)	0	(0.0)	0	(0.0)	0	(0.0)	-
Light – Appropriate	1	(0.9)	0	(0.0)	1	(2.6)	0	(0.0)	0.442
Appropriate	49	(45.4)	18	(41.9)	22	(56.4)	8	(38.1)	0.310
Appropriate – Heavy	24	(22.2)	12	(27.9)	8	(20.5)	2	(9.5)	0.218
Heavy	33	(30.6)	12	(27.9)	8	(20.5)	11	(52.4)	0.036*

Clinical Observations	Total Cohort[a] *N* =108		No PAE *n* =43		PAE (no FASD) *n* = 39		FASD *n* = 21		*p* [b]
	n	(%)	n	(%)	n	(%)	n	(%)	
Handwriting ability									
Unable to write first or surname	3	(2.8)	1	(2.3)	1	(2.6)	1	(4.8)	0.851
Unable to write surname	17	(15.7)	8	(18.6)	4	(10.3)	5	(23.8)	0.358
Unable to write a sentence	7	(6.5)	2	(4.7)	1	(2.6)	4	(19.0)	0.041*

* $p < 0.005$

[a] Total Cohort includes n =5 children with unknown PAE who were excluded from the group analysis.

[b] Significance tested for No PAE; PAE (no FASD); and FASD groups.

Table 4.10

Drawing (M-FUN Draw-a-Kid Game) and handwriting (ETCH) outcomes for the cohort, and according to PAE and FASD status

Outcomes	Total Cohort[a] *N* =108		No PAE *n* =43		PAE (no FASD) *n* = 39		FASD *n* = 21		*p* [b]
	M	*(SD)*	M	*(SD)*	M	*(SD)*	M	*(SD)*	
Drawing (M-FUN) (*n* = 97)									
Body Awareness[c]	5.2	(1.0)	5.3	(1.1)	5.1	(1.0)	5.1	(1.2)	0.565
Motor Accuracy[d]	7.6	(1.6)	7.7	(1.6)	7.6	(1.4)	7.1	(2.0)	0.419
Total Score[e]	12.8	(2.6)	13.0	(2.6)	12.6	(2.3)	12.2	(3.1)	0.522
Handwriting (ETCH)									
Name: Letter legibility[f] (*n* = 91)	60.8	(26.0)	62.5	(27.1)	60.9	(24.2)	56.1	(28.0)	0.729
Sentence: Letter legibility[f] (*n* = 101)	62.9	(22.3)	62.0	(24.4)	63.6	(21.7)	60.4	(21.5)	0.872
Sentence: Word legibility[f] (*n* = 101)	73.3	(29.1)	81.0	(22.1)	73.9	(28.6)	50.0	(37.0)	0.008**

** $p < 0.001$

[a] Total Cohort includes n =5 children with unknown PAE who were excluded from the group analysis.

[b] Significance tested for No PAE; PAE (no FASD); and FASD groups.

[c] Possible score ranges 0 to 6; [d] 0 to 9; and [e] 0 to 15.

[f] Scores indicate percentage of legible letters or words.

Table 4.11

Inter-rater reliability

	Weighted Kappa[1]	Confidence Interval	Strength of agreement[2]
M-FUN			
Body Awareness			
Number of parts	0.84	0.75 – 0.93	Excellent
Overall impression	0.44	0.30 – 0.58	Moderate
Motor Accuracy			
Number of parts	0.62	0.43 – 0.81	Substantial
Overall impression	0.16	-0.003 – 0.32	Slight
ETCH: Name	ICC[3]		
Letter legibility	0.90		Excellent
ETCH: Sentence			
Letter legibility	0.90		Excellent
Word legibility	0.91		Excellent

[1]Weighted kappa with quadratic weights

[2] Strength of agreement based on Landis & Koch (1977) criteria

[3] ICC = Intra-class correlation coefficient

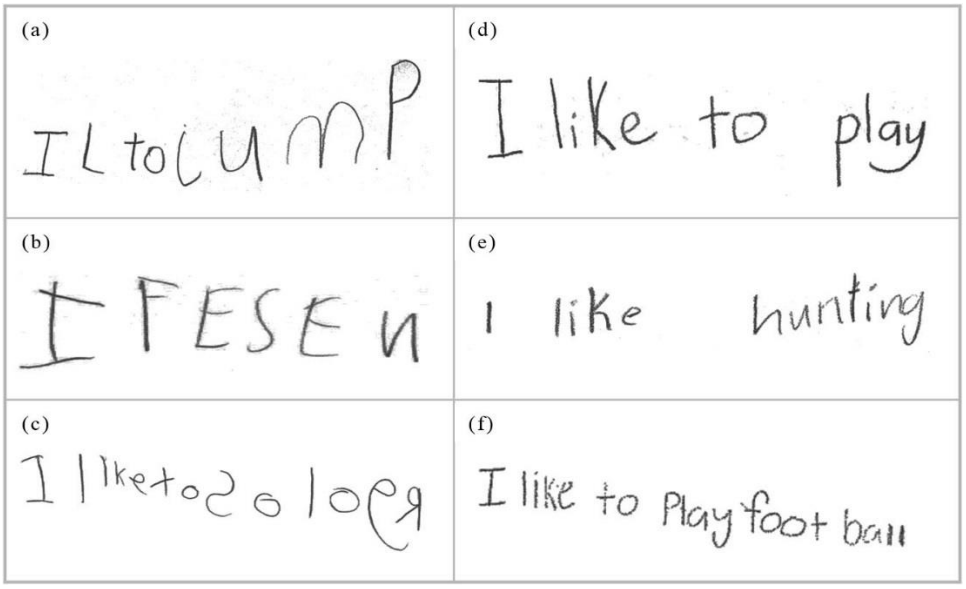

Figure 4.4 Handwriting samples from The Evaluation Tool of Children's Handwriting Sentence Writing Task

Completed by children with (a) PAE (high exposure); no FASD; 7.11 years; IQ = 82 ('I like to jump'); (b) ND-AE; 8.5 years; IQ = 76 ('I like fishing'); (c) pFAS; 7.11 years; IQ = 61 ('I like to colour'); (d) No PAE; 9.5 years; IQ = 80; (e) No PAE; 9.2 years; IQ = 91; (f) No PAE; 9.5 years; IQ = 88. IQ = Universal Nonverbal Intelligence Test (UNIT) Full Scale Standard Score. The UNIT has a normative M=100.0, SD=15.

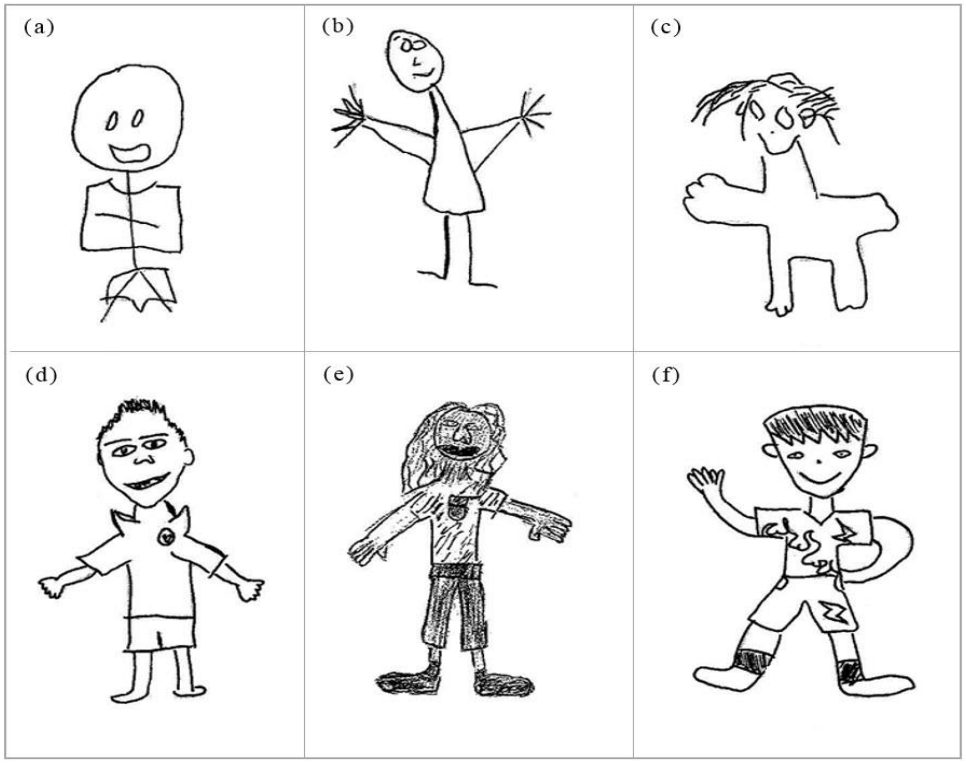

Figure 4.5 Human figure drawings from the Miller Function and Participation Scales Draw-a-Kid Game

Drawn by children with (a) pFAS: 7.9 years; IQ = 93; (b) ND-AE: 8.8 years; IQ = 91; (c) pFAS: 8.1years; IQ = 82; and (d) No PAE: 9.2 years; IQ = 86; (e) No PAE: 9.6 years; IQ = 98; (f) No PAE: 9.2 years; IQ = 90. IQ = Universal Nonverbal Intelligence Test (UNIT) Full Scale Standard Score. The UNIT has a normative M=100.0, SD=15.

4.3.5 Discussion

This is the first comprehensive description of graphomotor skills of 7.5 to 9.6 year old children living in a remote area of Australia, most of whom were Aboriginal, and the first to examine whether these skills differed between children with and without PAE or FASD. Many children in this cohort had poor graphomotor skills, including a delayed pencil grasp and application of excessive pressure through their pencil, and showing reduced functional writing and handwriting legibility. Children with FASD were significantly more likely to have handwriting difficulties than children without PAE, including using a cross-thumb pencil grasp which was immature for their age, applying heavy pressure through their pencil during graphomotor tasks, being unable to write a sentence, and writing fewer legible words in sentence writing tasks. Drawing skills, which were evaluated for motor accuracy and body awareness, were similar between children with and without PAE or FASD.

The handwriting abilities of children in the Fitzroy Valley are concerning. In a previous study of 320 children aged 3 to 6.11 years in the US, transitional grasps were used until about 6 years of age, but by 6.11 years, 72.5% of children used a dynamic tripod grasp (Schneck & Henderson, 1990). These findings contrasted to our cohort, in which only 13.9% of children used a dynamic tripod grasp. The children in the Lililwan Project were older than those in Schneck and Henderson's study, and thus would be expected to have a greater, not lesser, proportion of children using a mature pencil grasp. Although some researchers have failed to find a relationship between use of a dynamic pencil grasp and handwriting performance (Schwellnus et al., 2012), it is generally acknowledged that transitional grasps are inefficient and can cause muscle fatigue and cramping and lead to poorer graphomotor output (Tseng & Cermak, 1993). An immature pencil grasp can also indicate problems with proprioception, sensory processing, and the sensory-motor feedback loop (Benbow, 2006), as can exerting excessive pressure through the pencil during graphomotor tasks (Levine, 1987).

Many academic and recreational tasks require proficiency in graphomotor skills, and poor performance of these skills can impair the ability to participate in many classroom activities and communicate learned knowledge (Chase, 1986; Tomchek & Schneck, 2006). There are limited data directly related to graphomotor skills of Australian Aboriginal children. However, the National Assessment Program – Literacy and Numeracy (NAPLAN), which is an annual assessment of reading, writing, language, and numeracy skills completed annually by Australian students, has highlighted that many Aboriginal students perform below national academic

benchmarks (Australian Curriculum Assessment and Reporting Authority, 2015). The Australian Early Developmental Index (AEDI), which is based on teachers' evaluation of student competence in their first year of school, reports that 20.6% of students in Fitzroy Crossing were below the 10th percentile for fine and gross motor skills (The Royal Children's Hospital Melbourne, 2012). In addition, some aspects of local Aboriginal culture, such as painting and boab nut carving, require sound fine motor skills. Addressing graphomotor and other fine motor impairments is likley to have a positive flow-on effect to many aspects of a child's occupational performance.

Other groups have evaluated handwriting legibility using the ETCH. In a study of 31 children in Grade 1 in the US, an average of 78% to 80% letter legibility was found when writing a sentence (Diekema, Deitz, & Amundson, 1998). Similarly, a study evaluating 26 children in Grades 2 and 3 in Canada, found a mean letter legibility of 73.3% and word legibility of 70.6% (Brossard-Racine, Mazer, Julien, & Majnemer, 2012). Both these studies reported much higher letter legibility rates than for the children in the Lililwan Project (62.9%). Although the cohort's mean word legibility (73.3%) was similar to the children in Bossard-Racine's study, the children in our cohort were older and therefore we expected them to have higher rates of legibility. The common use of immature pencil grasps and application of excessive writing pressure, along with fine motor (Doney et al., 2017) and visual motor integration difficulties (Doney et al., 2016) observed in our cohort, likely contributed to the reduced handwriting legibility for many of the children.

No other publications report outcomes from the Draw-a-Kid Game, so findings cannot be compared to other studies. This assessment tool was chosen because of its unique properties of evaluating motor accuracy in the context of human figure drawing rather than developmental maturity. The cohort's mean Total Score (M = 12.8) was lower than the maximum possible score (15), as were the Motor Accuracy scores (M = 7.6; maximum possible = 9). These scores are noteworthy as the Draw-a-Kid Game is designed for younger children (4.0 to 7.11 years) than those in the cohort (7.5 to 9.6 years), and hence most children in the cohort should have scored at the upper limits. However, normative data are not available for the M-FUN Draw-a-Kid Game, so results should be interpreted cautiously.

Children with FASD had more difficulties with graphomotor skills than children without PAE. Some children with FASD had particular patterns of handwriting difficulties, including letter reversals, inconsistent letter size, missing or incorrect letter choice, and a lack of spacing between words. Typical examples are shown in Figure 4.4. The findings are consistent with those of Duval-White et al. (2013) who

found that most children with FASD in their study (*n* = 20) scored in the 'well-below average' range for letter legibility.

Children with PAE or FASD had similar drawing scores to children without PAE, which was an unexpected finding. The lack of significant differences between groups on the Total Score of the M-FUN may be due to low inter-rater reliability for the 'Motor Accuracy: Overall Impression' score, but this is unlikely because substantial to excellent reliability was achieved for all other scores which contributed to the Total Score. Similar to handwriting, some children with FASD had characteristic styles of drawing (*Figure* 4.5) which possibly reflects general developmental delay, rather than specific Body Awareness or Motor Accuracy difficulties.

In contrast to the drawing outcomes for the children in the Lililwan Project, human figure drawings from children with FASD were evaluated in two other studies and significant impairment was identified. One study of 142 Grade 1 South African children with pFAS or FAS reported significantly lower drawing scores than children without PAE (Urban et al., 2008), although these children were younger than those in the Lililwan Project. Another study of 28 Swedish children with and without PAE evaluated human figure drawings and concluded that perceptual difficulties accounted for poorer drawing abilities in children with PAE (Aronson et al., 1985). However, in these studies the drawings were not evaluated within a motor skills framework, so comparisons with the findings in the present study are difficult.

Limitations and future directions

Despite representing almost two entire age cohorts for the region, the sample size (*n* = 108) was relatively small, but still larger than the only other published study of graphomotor performance in children with FASD (Duval-White et al., 2013). The cohort was mostly of Australian Aboriginal descent and living in a remote region of Australia, and while results may be similar to children in other remote regions with comparable demographics, results should not be generalised to other populations.

Validated measures of graphomotor and other fine motor skills do not exist for Australian Aboriginal children, and caution should be used when using assessment tools which have been developed for different populations, especially those with differing cultural contexts (Thorley & Lim, 2011). However, the children in the Lililwan Project had been attending primary school for several years, and should have been familiar with the graphomotor requirements of the ETCH and M-FUN.

Many factors contribute to graphomotor performance other than motor skills, including cognition, language, attention, hearing, school attendance, and early exposure to fine motor skills (Benbow, 2006; Tomchek & Schneck, 2006). The cohort had high levels of other prenatal exposures and socioeconomic risk factors which may have affected performance. Future studies should explore the impact of these factors and explore whether they differ in children with PAE or FASD.

Implications for occupational therapy practice

This study provides evidence that many children in the region, regardless of PAE, have graphomotor impairment which could interfere with academic performance and participation in cultural and recreational activities, and may benefit from occupational therapy input. Letter legibility rates of less than 76.0%, and word legibility rates of less than 75.0%, indicate the need for therapeutic treatment (Brossard-Racine et al., 2012). This recommendation is based on younger children (Grades 2 and 3) than those in the Lililwan Project, but nevertheless indicates that 37.6% (based on word legibility) to 68.3% (based on letter legibility) of children may benefit from handwriting intervention. Handwriting intervention has shown improvement in skills in other populations (Case-Smith, Weaver, & Holland, 2014), and may be of benefit to children in the Fitzroy Valley. The Fitzroy Valley has a population of 4,500 people and is currently serviced by two occupational therapists, who provide services across the lifespan via a fortnightly outreach service from Derby, 260km to the West. Given that this study only included children from two age groups, it is evident that occupational therapy services in the Fitzroy Valley are severely under-resourced. It is recommended that i) therapeutic services, with a focus on early fine motor skill development, particularly handwriting instruction, would be of benefit to children in the Fitzroy Valley with graphomotor impairment; and ii) functional graphomotor skills should be assessed along with standardised measures of fine motor skills for children with PAE or suspected FASD.

Conclusions

In this study it was identified that many children in the Fitzroy Valley had poor graphomotor skills, which likely reflects the multitude of neurodevelopmental risk factors, including PAE and FASD, experienced by children in the region. Children with FASD had significantly poorer graphomotor skills than children without PAE, including delayed pencil grasp, heavy writing pressure, being unable to write a sentence, and having reduced word legibility. This study adds new evidence to the functional impairments experienced by children with FASD, including those which

may impact on successful school performance and function in the classroom. Based on these findings it is recommended that graphomotor skills should be assessed in populations with high levels of PAE, in addition to other fine motor skills, as they are important components of occupational performance. Graphomotor skills are critical for successful performance of many academic, recreational, and cultural activities, and identifying performance challenges will help guide appropriate therapeutic interventions which remediate or accommodate the fine motor impairment associated with PAE.

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4.4 Prevalence of Significant Domains of Fine Motor or Visual-motor Integration Impairment

This section provides additional results that were not reported in the published papers.

4.4.1 Introduction

The Lililwan Project diagnostic protocol defined fine motor skills and visual-motor integration as separate domains of impairment (Fitzpatrick et al., 2012; Fitzpatrick et al., 2017). To be considered a significant impairment, and thus count as a domain of impairment, children had to meet criteria as outlined in 3.1.5.2.

4.4.2 Results

Overall, rates of fine motor and visual-motor integration impairment were very high in the cohort. In a population, approximately 3% of children should score 2SD below the mean. In the children involved in the Lililwan cohort, nearly three times as many children (8.3%) had a fine motor impairment at this level, and more than eight times as many (25.0%) had a significant visual-motor integration impairment (Table 4.12). Interestingly, fewer children with FASD (4.8%) had a fine motor impairment compared to children without PAE (9.3%), but rates of visual-motor integration impairment were much higher (38.1%) in children with FASD compared to those without PAE (23.3%)

Table 4.12

Children with significant fine motor and visual-motor integration impairments according to the Lililwan Project diagnostic criteria

	Total Cohort *N* =108		No PAE *n* =43		PAE (no FASD) *n* = 39		FASD *n* = 21	
Domain	n	(%)	n	(%)	n	(%)	n	(%)
Fine motor skills	9	*(8.3)*	4	*(9.3)*	3	*(7.7)*	1	*(4.8)*
Visual-motor integration	27	*(25.0)*	10	*(23.3)*	9	*(23.1)*	8	*(38.1)*

[1] See (insert cross-reference for section on diagnostic criteria here)
[2] 'Total Cohort' includes *n* = 5 children with unknown PAE who are not included in the No PAE, PAE (no FASD), or FASD group

4.4.3 Discussion

It is unclear why fewer children with FASD had a significant fine motor impairment compared to children without FASD. One explanation may be that this skill is not impaired in children with FASD, although this is unlikely considering that PAE is known to damage CNS structures associated with fine motor skills (Autti-Rämö et al., 2002; Miller, 2007; Ramadoss et al., 2007). An alternative explanation is that the criteria developed during the Lililwan Project for determining a significant fine motor impairment was either too stringent, or did not account for specific types of fine motor impairment. A significant impairment was generally required to be 2SD below the mean across several domains, however as outlined in section 4.1, few children had impairment on the BOT-2 at this level. This supports the literature which has shown that CNS damage from PAE can be specific, and that grouping them together as 'fine motor skills' (or grouping them with gross motor skills for an overall 'motor' impairment) can be problematic (Adnams et al., 2001; O'Leary & Bower, 2009). It may be that specific aspects of fine motor skills are impaired by PAE, and indeed the results from the BOT-2 support this suggestion. The BOT-2 is an appropriate tool for assessment of fine motor skills in children with suspected FASD, however consideration should be given to the individual types of fine motor skills which may be impaired as there is a risk that stronger skills will mask specific fine motor deficits.The results also show that visual-motor integration skills are impaired amongst children of the Fitzroy Valley, but even more so in children with FASD. This supports the inclusion of the Beery VMI as an appropriate assessment tool for use in FASD diagnosis.

Chapter 5 Discussion

This chapter provides a summary of key fine motor findings from the Lililwan Project. A detailed discussion of the findings from each of the assessment tools in relation to existing literature is included in the discussion sections of each of the published papers (4.1 Fine Motor ; 4.2 Visual-motor ; 4.3 Graphomotor).

5.1 Key Findings

This chapter provides an overview of results from the published papers, strengths and limitations of the study, and an overall summary and integrated recommendations based on the key findings.

5.1.1 Fine motor skills (BOT-2)

Across the cohort, children had 'average' BOT-2 Fine Motor Composite scores compared to norms reported in the assessment manual. However, consistent with the findings from the systematic review, this 'average' score appeared to mask specific areas of strengths and deficits. Upper-Limb Coordination scores approached 'above-average', but Fine Motor Integration scores were close to 'below average'. This profile of strengths and weaknesses supports the use of the BOT-2 as a fine motor assessment tool in the region, because it includes a range of different fine motor skills and reports scores as both composite and discrete types of fine motor skills.

Children with FASD had significantly lower Fine Motor Composite and Manual Coordination scores than children without PAE. This adds further evidence that the BOT-2 is a useful assessment tool for FASD diagnosis. The BOT-2 detected overall fine motor impairment but also highlighted specific areas of difficulty. These attributes are useful both diagnostically, and clinically to guide clinicians to develop therapeutic interventions which target specific difficulties. Children with PAE didn't score significantly lower than children without PAE in any BOT-2 fine motor composite or subtest.

Only one child (who was diagnosed with FASD) had a 'severe' (below the 2nd percentile) impairment on any BOT-2 fine motor composite or subtest, but rates of 'moderate' (below the 16th percentile) impairment were very high, especially for the Fine Manual Control composite (and the contributing Fine Motor Precision and Fine Motor Integration subtests). In contrast, rates of moderate impairment were low for

the Manual Coordination composite (and the contributing Manual Dexterity and Upper-Limb Coordination subtests). Children with FASD had the highest rates of 'moderate' impairment across all fine motor composites and subtests (aside from Upper-Limb Coordination).

5.1.2 Visual-motor integration (Beery VMI)

Beery VMI scores were 'below average' across the cohort, regardless of whether children had no PAE or PAE and/or FASD. Visual Perception scores were 'average' for all groups. Fine Motor Coordination scores were 'average' in children without PAE and those with PAE (no FASD), but` significantly lower and 'below average' in children with FASD.

Similar to BOT-2 outcomes, very few children had 'severe' visual-motor integration, visual perception, or fine motor coordination impairments, but rates of 'moderate' impairments were very high, especially in children with FASD.

5.1.3 Graphomotor (ETCH; M-FUN)

Many children in the cohort had impaired graphomotor skills, which was expected given the fine motor and visual-motor integration impairments detected using the BOT-2 and Beery VMI. Many children used a pencil grip which was immature for their age, exerted heavy pressure through their pencil, had reduced handwriting legibility and were unable to write their full name. Consistent with BOT-2 and Beery VMI outcomes, children with PAE did not have significantly more graphomotor problems compared to children without PAE, but children with FASD had specific difficulties, including being more likely to use a cross-thumb pencil grasp, exert excessive pressure through their pencil, write less words legibly, and be less likely to be able to write a sentence. The scores for human-figure drawings, which were evaluated for body awareness and fine motor accuracy according to M-FUN criteria, were similar between children with and without PAE and/or FASD. However, some drawings by children with FASD were delayed for their age and showed poor fine motor control. As such, the drawings were of value because they supported clinical judgements of fine motor ability and added evidence to the functional impact of fine motor impairment, even though the M-FUN did not discriminate differences in the quality of the drawings.

5.1.4 Prevalence of significant domains of fine motor or visual-motor integration

According to the FASD diagnostic criteria used in the Lililwan Project, more children without PAE had a significant fine motor impairment than children with PAE (no FASD) and children with FASD (Table 4.12).This was an unexpected finding, and highlights the complexity of neurodevelopmental risk factors aside from PAE, such as low socioeconomic status, overcrowded housing, poor nutrition, and high levels of childhood illness including otitis media, which are experienced by many children in the cohort and may adversely affect fine motor development.

Significant visual-motor integration impairment was three times higher in the cohort than fine motor impairment, which may indicate either that children had more difficulties with this complex skill, or that the Beery VMI was more sensitive to detecting these impairments. Visual-motor integration was 1.5 times more likely to be impaired in children with FASD than children without PAE and PAE.

It may be that the FASD diagnostic criteria used in the Lililwan Project for determining fine motor and visual-motor integration impairment were too conservative. However, this is a complex issue, particularly in a population with many health and socioeconomic factors which could affect neurodevelopment. Revising the FASD diagnostic criteria used in the Lililwan Project requires careful consideration, because rates of impairment were also very high in children without PAE (albeit not as high as in children with FASD). This complex issue would benefit from further research both in Australia and internationally.

5.2 Strengths

This thesis provides the first comprehensive profile of fine motor abilities of Aboriginal children living in a remote region of Australia, and the first in an Australian population-based cohort with high levels of PAE and FASD. It is unique because the candidate has used a combination of standardised and functional assessment tools to develop a detailed profile of fine motor skills. The study used fine motor assessment tools which are internationally endorsed as being appropriate for FASD diagnostic assessment and commonly used by occupational therapists in Australia, as well as assessing a range of different types of fine motor skills. Assessments were conducted by a qualified occupational therapist (the doctoral candidate) with experience working with children in the region.

Clinicians were blinded to PAE and other prenatal and childhood exposures during the assessment and diagnostic phase. In contrast to previous studies of fine motor skills in children with FASD, results were compared to children without PAE in the same population, many of whom experienced similar neurodevelopmental risk factors other than PAE which may have adversely impacted on fine motor abilities. This increases the likelihood that any observed impairments were due to PAE. Stringent FASD diagnostic cut-offs were applied, thus reducing the likelihood of over-diagnosis.

The Lililwan Project was Australia's first population-based prevalence study of FASD and had very high participation rates. The study was initiated by local Aboriginal leaders, and community consent and feedback was rigorously applied at all stages of the project, and was commended by the Australian Aboriginal and Torres Strait Islander Commissioner as a good example of research in Aboriginal communities (Latimer et al., 2010).

5.3 Limitations

This study was conducted in the remote Fitzroy Valley with predominantly Aboriginal children, so while results may be comparable to other populations with similar demographics, results should not be generalised.

Although the sample size was relatively large compared to other studies of fine motor skills in children with PAE or FASD, and captured almost two entire age cohorts, it was not large enough to statistically control for a range of potentially confounding factors. Many other factors may have adversely affected fine motor performance, including other prenatal and early childhood exposures, socioeconomic conditions, health factors, and other neurodevelopmental skills including cognition, language, and behavioural and emotional factors.

In the Lililwan Project, alcohol consumption in pregnancy was reported retrospectively and may have been subject to recall bias. However, information was provided either by the birth mother or close family members living in the household, and corroborated by review of maternal and child medical records.

No fine motor assessment tools have been developed specifically for Aboriginal children, nor normed for children living in remote regions of Australia. Although the BOT-2 was established as having good to excellent reliability with our cohort (Lucas et al., 2013), validity was not established due to time, geographical, and resource constraints. However, the BOT-2 and the Beery VMI were chosen due to their recommended suitability and validity for children from diverse cultures. Additionally,

children in the cohort had been attending school for several years and thus should have been familiar with testing requirements. However, some health, cultural and lifestyle factors may have had an impact on fine motor outcomes. Many families in the Fitzroy Valley are highly mobile between communities (Morphy, 2010), and it is common for children to stay with extended family members which can result in disrupted schooling and poor school attendance. When a community member dies, their name cannot be used for a period, and anyone in the community with the same name must use a different name. Surnames may also be used interchangeably to reflect different kinship ties. These factors may account for some of the difficulties children had when writing their name.

Children in the Fitzroy Valley live an 'outdoors' lifestyle with an emphasis on many gross motor activities; less time is given to indoor activities with a fine motor focus. Children in the study had strong gross motor skills in comparison to fine motor skills (Lucas et al., 2016). Thus, a lack of early exposure to fine motor skills may also account for some of the difficulties experienced by children in our cohort.

5.4 Summary and Recommendations

This section provides recommendations based on the fine motor outcomes

1. Children in the Fitzroy Valley had relatively strong upper-limb coordination, but weaker fine motor precision and fine motor integration/visual-motor integration skills. They also had difficulties with many aspects of graphomotor skills, including handwriting legibility. Further, rates of moderate impairment were high across the cohort for many types of fine motor skills, indicating that many children may benefit from therapeutic interventions.

    1.1 Fine motor assessment tools, which assess a range of fine motor skills and report them as discrete types of skills, should be used in the region, and for FASD diagnosis. The BOT-2 and Beery VMI identified fine motor and visual-motor integration strengths and weaknesses in the population, and appeared to be appropriate assessment tools for use with children in the Fitzroy Valley.

    1.2 Graphomotor skills should be assessed as part of the FASD diagnostic process to determine the functional implications of identified impairments. The ETCH appeared suitable for assessing handwriting legibility, although other handwriting assessment tools that also assess speed may be appropriate. The human-figure samples provided useful insights into

developmental level and fine motor control, but the M-FUN was not suitable for detecting fine motor impairments.

1.3 Population-wide therapeutic programs with an early intervention focus, such as community playgroups with activities, which include fine motor activities, may be of benefit in the Fitzroy Valley.

1.4 Presently, allied health resources and educational services are prohibitive for one-to-one therapy programs. Classroom-wide fine motor interventions delivered by the teacher and educational assistants, under the guidance of an occupational therapist, may be the most feasible and beneficial option.

1.5 Up-skilling of classroom educational assistants, often local Aboriginal people, to deliver therapy programs to small groups of children may also be of benefit, and promote sustainability of therapeutic interventions.

2. Children with FASD had specific areas of fine motor impairment compared to children without PAE.

2.1 In keeping with recommendation 1.1, FASD diagnostic assessment tools should evaluate a range of fine motor skills, and report different aspects of fine motor skills as discrete scores.

2.2 Children with FASD would benefit from ongoing therapy delivered by multi-disciplinary teams, including occupational therapy, to improve fine motor function.

3. Few children had a ‘severe’ fine motor impairment, but rates of ‘moderate’ impairment were very high for many types of fine motor skills. Additionally, few children met the FASD diagnostic criteria used in the Lililwan Project to determine if fine motor skills should be considered impaired at a significant enough level to contribute to a FASD diagnosis.

3.1 In populations with high levels of PAE and other neurodevelopmental risk factors, such as the children in our cohort, it is difficult to conclude that identified impairments are solely the result of PAE. The amount, duration, and frequency of alcohol consumed during pregnancy should be considered when determining whether identified impairments, including fine motor impairment, is likely due to PAE. The existence of facial dysmorphology, growth impairment, and/or pervasive neurodevelopmental impairment

(across multiple domains of function) should also be considered when determining whether a fine motor impairment is likely due to PAE.

3.2 A degree of clinical judgement should be applied when determining whether fine motor skills are impaired at a significant enough level to be counted as a domain of impairment in terms of FASD diagnosis. Outcomes from both standardised assessment tools and clinical observations of functional fine motor performance, including graphomotor skills, should be considered when making a FASD diagnosis. This may be of value in populations the cohort in the Fitzroy Valley, and many others in Australia, where validity of existing assessment tools has not been established.

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Appendices

Appendix A Map of the Fitzroy Valley

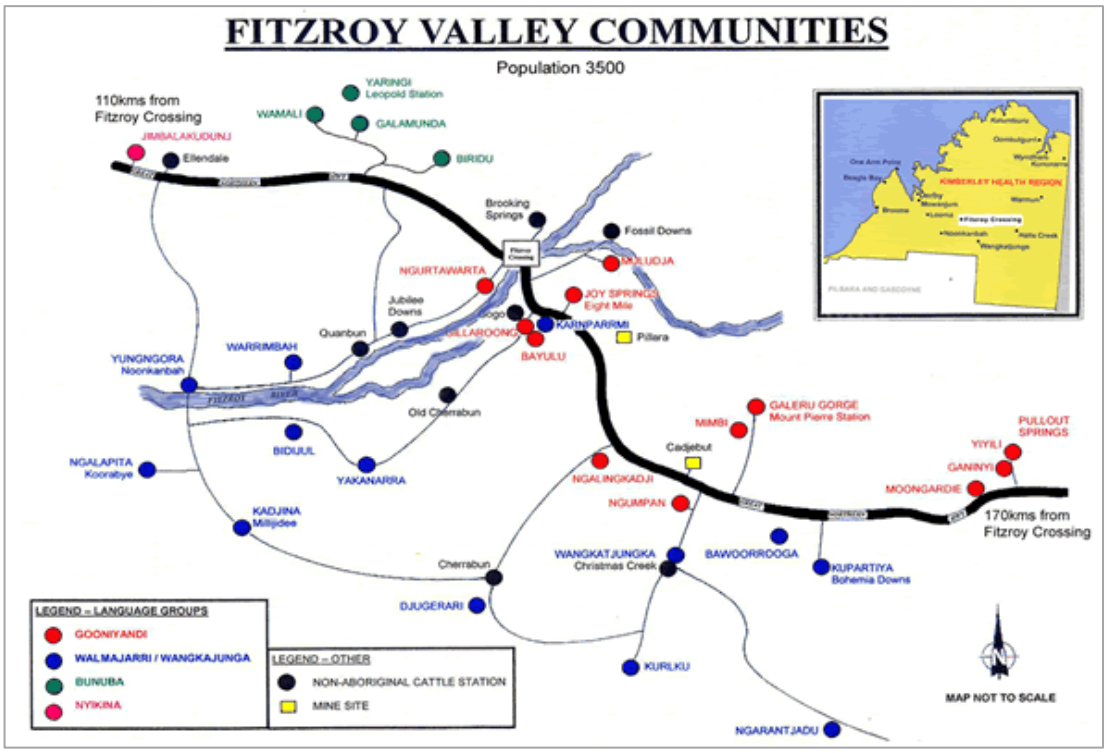

Figure A.1 Map of the Fitzroy Valley

Appendix B Literature Review Data Extraction Table

Table B.1

Studies of fine motor skills in children with PAE or FASD

Study	Country (ethnicity*)	Sample (n)	Age (years)	PAE/ FASD/ other *(n)*	PAE ascertainment	FASD diagnostic system	Research design (matching criteria)	FM assessment/ results: mean (SD/SE)	
Adnams et al., 2001	South Africa (South African colored or mixed ancestry)	68	6.33-8.00	FAS (34)	Maternal interview	Process/ criteria described	MCC (age, sex, first language, family income, school)	**GMDS:** Eye-hand coordination Controls: 93.77(1.58) FAS: 84.41(1.77) *F* = 14.83 *p* < .001	
Aragón et al., 2008	USA (Native American)	56	7-17	pFAS (14) FAS (10)	Maternal/ adoptive parent interview	Revised IOM	MCC (age, ethnicity)	**GPT:** *Dominant hand:* Controls: 0.15(1.07) pFAS: 0.35(1.30) FAS: 0.43(1.11) *F* = 0.307 *p* = .737	*Non-dominant hand:* Controls: 0.07(0.84) pFAS: 0.52(0.88) FAS: 0.54(0.64) *F* = 2.09 *p* = .133
Aronson et al., 1985	Sweden	42 (GMDS: 26)	1.5-9	Maternal alcohol dependency PAE (no diagnosis) (11) FAS (10)	Nd	Nd	MCC (sex, age, birth weight, gestational age, living area)	**GMDS:** Eye-hand coordination Controls: 109.1(9) PAE: 88.5(12) *p* < .001	

Study	Country (ethnicity*)	Sample (n)	Age (years)	PAE/ FASD/ other *(n)*	PAE ascertainment	FASD diagnostic system	Research design (matching criteria)	FM assessment/ results: mean (SD/SE)
Astley et al., 2009	USA	81 (BVMI: 78)	8-15.9	Drinks/occasion (mean(SD)): ND/AE 11.7(7.3) SE/AE 14.1(8.9) FAS 11.6 (7.1) ND/AE (20) SE/AE (24) FAS (18)	Retrospective parent interview and/ or record review	UW FAS DPN	MCC (age, gender, race)	**BVMI:** Controls: 102.7(12.9) ND/AE: 90.9(11.8) SE/AE: 81.4(9.2) FAS/pFAS: 76.2(12.7) *F* (overall)= 17.18 *F* (A-priori) = 51.6 $p < .05$ (between all groups except FAS/pFAS and SE/AE)

Study	Country (ethnicity*)	Sample (n)	Age (years)	PAE/ FASD/ other *(n)*	PAE ascertainment	FASD diagnostic system	Research design (matching criteria)	FM assessment/ results: mean (SD/SE)	
Barr et al., 1990	USA (Caucasian)	449 (427-449 WFSMB; HRNB)	4.3	*Authors define 1.0oz alcohol=2 drinks* Average ounces of absolute alcohol/day: 0.5-1.5oz (i.e. 1-3 drinks/day; or 7-21 drinks/week) a) AAP: early pregnancy (prior to pregnancy recognition) b) AAD: mid-pregnancy	Maternal interview during pregnancy		NCC	**WFMSB:** <u>Vertical/ Horizontal Groove Board; Grooved Maze Board; Resting Steadiness Hole Board</u> *AAP (AAD not reported)* *Errors* *F* = 6.47 *p* = .011 <u>Grooved Pegboard:</u> *AAP (AAD not reported)* *Time to complete* *F* = 5.68 *p* = .018 **HRNB:** <u>Finger tapping</u> *AAD (AAP not reported)* *F* = 5.86 *p* = .016 <u>Grip strength</u> *AAD (AAP not reported)* *p* Not significant	*Latency to correct* *F* = 15.25 *p* = .000 *(AAD not reported)* *Peg drop count* *p* Not significant

Study	Country (ethnicity*)	Sample (n)	Age (years)	PAE/ FASD/ other *(n)*	PAE ascertainment	FASD diagnostic system	Research design (matching criteria)	FM assessment/ results: mean (SD/SE)
Bay et al., 2012	Denmark	685	5.1-5.3	Average standard (12g alcohol) drinks/ week: a) 0 (325) b) 1-4 (267 c) 5-8 (82) d) >9 (11) (max=14 drinks/week)	Self-report during pregnancy	Na	NCC	**M-ABC:** Manual dexterity tasks *Mean difference in scores(CI)* 0: Reference group 1-4: 0.24(-0.51-0.99) 5-8: -0.35(-1.52-0.83) >9: -0.02(-2.43-2.38) *p* = .79
Chiodo et al., 2009	USA (African-American)	75	3.93-5.63	a) Absolute and average alcohol at conception b) 2 weeks before first antenatal visit c) across pregnancy d) CAGE; T-ACE; MAST; MSAC. e) Above measures combined: 'At risk alcohol exposure' (ARAE)	Maternal interview throughout pregnancy	Na	NCC (age, gender)	**PENTB:** Visual motor integration *ARAE* *r*=0.25*; β*=0.21 *p* < .025; < .05 **BARS:** PPT *ARAE* *Total pegs* *r*=0.23; *β*=0.23 *p* < .025; < .025 *Total pegs - both hands* *r*=0.25; *β*=0.25 *p* < .025; < .025 *Results reported for six different measures of PAE. Mixed results dependent on measure used* *Total pegs: left hand* *r*=0.24; *β*=0.24 *p* < .025; < .025 *Total pegs: non-dominant hand* *r*=0.25; *β*=0.24 *p* < .025; < .025

Study	Country (ethnicity*)	Sample (n)	Age (years)	PAE/ FASD/ other *(n)*	PAE ascertainment	FASD diagnostic system	Research design (matching criteria)	FM assessment/ results: mean (SD/SE)
Coles et al., 1997	USA (African-American)	149	7-8.5	Absolute alcohol/week (mean(SD)): PAE: non-dysmorphic 6.67(6.48) (i.e. average 13.34 drinks/week) FAS/FAE 12.50(11.52) (i.e. average 25 drinks/week) PAE: non-dysmorphic (62) FAS/FAE (25) ADHD (27)	Maternal/ caregiver interview	Dysmorph-ology examination	NCC	**BVMI:** Controls: 97.82(10.31) ADHD: 89.67(10.95) PAE(non-dysmorphic): 92.79(11.37) FAS/FAE: 87.84(13.90) $F = 4.41$ $p < .005$

Study	Country (ethnicity*)	Sample (n)	Age (years)	PAE/ FASD/ other *(n)*	PAE ascertainment	FASD diagnostic system	Research design (matching criteria)	FM assessment/ results: mean (SD/SE)
Conry, 1990	Canada (Native American)	38	5.2-18.5	FAS/FAE (19)	Retrospective maternal interview; hospital records; community sources	Research Society on Alcoholism criteria	MCC (sex, age)	**BVMI:** Controls: 9.4 FAE: 7.3 FAS: 3.0 *F* = 18.33 *p* < .001 (significant between Controls and FAS; and FAS and FAE) <u>Finger tapping:</u> *Dominant hand:* Controls: 52.5 FAE: 50.7 FAS: 41.4 *F* = 6.10 *p* = .005 (significant between Controls and FAS; and Controls and FAE) *Non-dominant hand:* Controls: 53.7 FAE: 47.0 FAS: 42.5 *F* = 5.86 *p* = .006 (significant between Controls and FAS) <u>Grip strength:</u> *Dominant hand:* Controls: 53.0 FAE: 40.7 FAS: 41.2 *F* = 9.35 *p* < .001 (significant between Controls and FAS; and Controls and FAE) *Non-dominant hand:* Controls: 54.7 FAE: 42.9 FAS: 42.3 *F* = 9.35 *p* < .001 (significant between Controls and FAS; and Controls and FAE)

Study	Country (ethnicity*)	Sample (n)	Age (years)	PAE/ FASD/ other *(n)*	PAE ascertainment	FASD diagnostic system	Research design (matching criteria)	FM assessment/ results: mean (SD/SE)	
								DTLA: <u>The Motor Speed and Precision Test</u> Controls: 51.5 FAE: 47.9 FAS: 39.6 *F* = 7.49 *p* = .002 (significant between Controls and FAS only)	
Fried & Watkinson, 1990	Canada	130 (4 year olds)	Assessed at 3 and 4 years (only 4 year olds met review age inclusion criteria)	*Authors define 0.86=1 drink* a) Light/no use: <0.14 oz absolute alcohol/day (i.e. approx. <1 drink/week) b) Moderate/ heavy use: >0.14 oz absolute alcohol/day (i.e. approx. >1 drink/week) Light/no PAE (59) Moderate/ heavy PAE (71)	Maternal interview during pregnancy (each trimester)	Na	NCC	**GPT:** *(Time; Errors)* *4yo data:* *Dominant hand:* Light/none: 69.9; 1.5 Moderate/heavy: 72.8; 1.3	*Non-dominant hand:* Light/none: 74.2; 1.6 Moderate/heavy: 77.5; 1.8 *p* Not significant (either hand)

Study	Country (ethnicity*)	Sample (n)	Age (years)	PAE/ FASD/ other *(n)*	PAE ascertainment	FASD diagnostic system	Research design (matching criteria)	FM assessment/ results: mean (SD/SE)
Henry et al., 2007	USA	274	6-16	Trauma/FASD (type not specified) (161) Trauma/no FASD (274)	Nd	UW FAS DPN	CC	**PEEX/PEERAMID:** <u>Fine motor</u> *Percentage with moderate/major delays:* Trauma/no FASD: 48 Trauma/FASD: 60 *p* Not significant <u>Graphomotor</u> *Percentage with moderate/major delays:* Trauma/no FASD: 48 Trauma/FASD: 60 *p* Not significant
Irner et al., 2012	Denmark	163 (GMDS: 101)	Assessed at 0-3; and 3-7 years (only 3-7 year olds met age criteria for current review)	Missing data, but at least half exposed to high PAE (>15 drinks/week)	Maternal interview 'several times' during pregnancy	Na	NCC	**GMDS:** <u>Eye-hand coordination</u> *3-7 yo:* *During pregnancy* No PAE/ no Polydrug: n=0 No PAE/ Polydrug: 94.8(14) PAE/ no Polydrug: 87.1(11) PAE/ Polydrug: 94.9(15) *p* = .30 *At time of birth* No PAE/ no Polydrug: 94.9(12) No PAE/ Polydrug: 95.7(12) PAE/ no Polydrug: 80.6(19) PAE/ Polydrug: n=1 (excluded from analysis) *p* = .03

Study	Country (ethnicity*)	Sample (n)	Age (years)	PAE/ FASD/ other *(n)*	PAE ascertainment	FASD diagnostic system	Research design (matching criteria)	FM assessment/ results: mean (SD/SE)	
Janzen et al., 1995	Canada (Native American)	20	3.5-5	Maternal alcohol dependency FAS (10)	Maternal history of alcohol exposure; social service agency records; medical records of pregnancy	Revised Research Society on Alcoholism criteria	MCC (age, sex, race)	**BVMI:** Controls: 95 FAS: 54 *p* =.004 **WFMSB** <u>GPT:</u> *Number placed in 1 minute* *Right hand:* Controls: 8.5 FAS: 4.3 *p* = .011	 *Left hand:* Controls:6.7 FAS: 4.3 *p* = .047
Jirikowic et al., 2008	USA	51	5-8.5	High: 60% ARND (20) FAS (5)	Nd	UW FAS DPN	CC	**NEPSY:** <u>Sensorimotor</u> Controls: 107.1(10.9) FASD: 84.6(16.1) *p* = .001	
Kooistra et al., 2009	Canada	116	7-10	ARND (various subtypes) (30) ADHD (47)	Review of medical records if PAE unknown	UW FAS DPN	CC	**M-ABC:** <u>Manual dexterity tasks</u> Controls: 2.5(3.1) ADHD: 7.9(3.8) FASD: 6.1(3.4) *F* = 25.71 *p* < .001	

Study	Country (ethnicity*)	Sample (n)	Age (years)	PAE/ FASD/ other *(n)*	PAE ascertainment	FASD diagnostic system	Research design (matching criteria)	FM assessment/ results: mean (SD/SE)
Korkman et al., 1998	Finland	72	5.5 -9.2	Moderate to high (>10 drinks/week) a)1st trimester only (16) b)1st&2nd trimesters only (16) c) All trimesters (14) PAE – no diagnosis (12) PAE-microcephaly; no diagnosis (14) FAE (13) FAS (7)	Self-report in pregnancy; or clinical/ blood sample indicating intoxication during pregnancy	Criteria described	NCC	**BVMI:** Controls: -0.3(0.7) 1st trimester: -0.9(1.2) 1st&2nd trimester: -1.1(0.9) All trimesters: -1.5(1.2) *F* = 2.89 *p* < .05 **NEPSY:** <u>Visuomotor precision</u> Controls: 0.0(0.0) 1st trimester: -0.3(0.8) 1st&2nd trimester: -0.4(0.6) All trimesters: -0.5(1.0) *F* = 1.54 *p* Not significant <u>Kinesthetic praxis - position of hands</u> Controls: -0.5(0.7) 1st trimester: -0.3(0.7) 1st&2nd trimester: -0.2(0.4) All trimesters: -0.1(0.4) *F* = 0.69 *p* Not significant <u>Dynamic praxis - manual movement series</u> Controls: 0.2(0.5) 1st trimester: 0.1(0.5) 1st&2nd trimester: -0.1(0.2) All trimesters: -0.2(0.4) *F* = 4.08 *p* < .001

Study	Country (ethnicity*)	Sample (n)	Age (years)	PAE/ FASD/ other *(n)*	PAE ascertainment	FASD diagnostic system	Research design (matching criteria)	FM assessment/ results: mean (SD/SE)	
Laforce et al., 2001	Canada	10	8-14	Heavy FAS/ FAE (5)	Nd	Nd	MCC (age, IQ)	**PPT:** *Both hands:* Controls: 73.3(15.6) FAS/FAE: 92.7(19.5) *p* Not significant	
Mattson et al., 1998	USA	50	5-16	High PEA: not FAS (10) FAS (15)	Retrospective maternal self-report and/ or medical records	Physical/ dysmorph-ology examination	MCC (age, sex, ethnicity)	**BVMI:** Controls: 96 PEA: 84 FAS: 80 *F* = 9.09 *p* < .001 (FAS<Controls) **GPT:**	
								Dominant hand: Controls: 108 PEA: 96 FAS: 102 *F* = 2.58 *p* Not significant	*Non-dominant hand:* Controls: 105 PEA: 93 FAS: 96 *F* = 5.62 *p* < .01 (FAS, PEA < Controls

Study	Country (ethnicity*)	Sample (n)	Age (years)	PAE/ FASD/ other *(n)*	PAE ascertainment	FASD diagnostic system	Research design (matching criteria)	FM assessment/ results: mean (SD/SE)	
Mattson et al., 2010	USA and Finland	Analysis 1: 87 Analysis 2: 98	7-21	Heavy (>4 drinks/ occasion at least once/week; or >13 drinks/ week) Analysis 1: FAS (41) Analysis 2: PAE (not FAS) (38)	Review of records; maternal report	Physical/ dysmorph-ology examination	CC	**BVMI:** *Analysis 1* Controls: 91.39(14.57) AE/FAS: 83.27(14.51) $p < .05$ **GPT:** *Dominant hand:* *Analysis 1* Controls: -0.42(0.78) AE/FAS: 0.56(1.12) $p < .05$ *Analysis 2* Controls: -0.38(0.71) AE/Not FAS: 0.62(1.14) $p < .05$	*Analysis 2* Controls: 91.00(13.81) AE/Not FAS: 79.37(11.97) $p < .05$ *Non-dominant hand:* *Analysis 1* Controls: -0.28(0.74) AE/FAS: 0.58(1.38) $p < .05$ *Analysis 2* Controls: -0.22(0.69) AE/Not FAS: 0.93(1.80) $p < .05$

Study	Country (ethnicity*)	Sample (n)	Age (years)	PAE/ FASD/ other *(n)*	PAE ascertainment	FASD diagnostic system	Research design (matching criteria)	FM assessment/ results: mean (SD/SE)
Russell et al., 1991	USA	164	6	Prior to pregnancy recognition (in the year prior); ounces of absolute alcohol per day *Authors define 1oz=2 drinks* a) abstainer Nil b) light/moderate 0-1 (i.e. 0-2 drinks/day) c) heavy PPAA: 1-3.5(i.e. 2-7 drinks/day) d) very heavy PPAA: >3.5(i.e. >7 drinks/day) a) abstainer (46) b) light/moderate (111) c) heavy (13) d) very heavy (5) e) Indications of problem drinking (IPD) (<1 (161); >1 (14))	Self-administered questionnaire during pregnancy	Growth and dysmorph-ology examination	MCC (age, race, education, child's gender)	**BVMI:** Abstainer: 39.5(3.72) Light/moderate: 40.3(2.26) Heavy: 29.5(7.06) Very heavy: 32.4(11.72) IPD < 1: 39.5(1.85) IPD>1: 35.8(6.51) *p* Not significant **CATB:** <u>Draw-a-line slowly</u> Abstainer: 35.7(2.76) Light/moderate: 32.1(1.68) Heavy: 28.3(5.24) Very heavy: 26.2(8.69) IPD < 1: 32.9(1.37) IPD>1: 28.8(4.82) *p* Not significant
Sowell et al., 2008	USA	36	7-15	Heavy ARND (8) pFAS (5) FAS (4)	Maternal report; reliable collateral report; medical/ legal records	UW FAS DPN	CC	**BVMI:** Controls: 24(3.873) FASD: 19.824(3.729) *t* = 3.294 *p* = .002

Study	Country (ethnicity*)	Sample (n)	Age (years)	PAE/ FASD/ other *(n)*	PAE ascertainment	FASD diagnostic system	Research design (matching criteria)	FM assessment/ results: mean (SD/SE)
Uecker & Nadel, 1996	USA (Native American)	30	7-12	FAS (15)	Nd	Physical/ dysmorph-ology examination ; CNS assessment	MCC (age, gender)	**BVMI:** Controls: 8.47(2.80) FAS: 5.13(2.20) *F* = 13.15 *p* < .001
Vaurio et al., 2011	USA	110	6-16	Heavy (>4 drinks/ occasion at least once/week; or >14 drinks/ week throughout pregnancy) FAS (18) PAE, no diagnosis (33) Not assessed (4)	Retrospective review of medical records; social service records; adoption agency records; or maternal report	Physical/ dysmorph-ology examination	MCC (IQ)	**BVMI:** Controls: 89.60(12.93) FASD: 85.85(12.90) *Cohen's d* = 0.29 *p* = .022 **GPT:** Controls: 97.70(11.45) FASD: 101.45(10.96) *Cohen's d* = -0.33 *p* = .051
Zhou et al., 2011	Canada	66 (NEPSY: 19)	6-30 (only 6-12 year olds completed NEPSY)	FASD (type unspecified (9) NBD/AE (12) pFAS (2) FAS (3)	Nd	UW FAS DPN	MCC (age, sex, hand-edness)	**NEPSY:** Visuomotor precision *(FASD compared to assessment norms)* FASD: 8.3(3.0) *p* = .027

***Note**:* *Ethnicity provided only for homogenous/ majority ethnicities
PAE: Prenatal alcohol exposure; FASD: Fetal Alcohol Spectrum Disorder; CC: Case control; MCC: Matched case control; NCC: Nested case control. Nd: Not described; Na: Not applicable.
Assessment abbreviations: BARS: Behavioral Assessment and Research System; BVMI: Beery Buktenica Test of Visual Motor Integration; CATB: The Cincinnati Autonomy Test Battery; DTLA: Detroit Tests of Learning Aptitude; GMDS: Griffiths Mental Development Scales; GPT: Grooved Pegboard Test; HRNB: Halstead Reitan Neuropsychological Battery; M-ABC: Movement Assessment Battery for Children; NEPSY: Developmental Neuropsychological Examination; PENTB: Pediatric Environmental Neurobehavioral Test Battery; PEERAMID: Pediatric Examination of Educational Readiness at Middle Childhood; PEEX: Pediatric Early Entry Examination; PPT: Purdue Pegboard Test; WFMSB: Wisconsin Fine Motor Steadiness Battery

Appendix C Thesis Publications

C.1 Publication 1

Doney, R., Lucas, B. R., Jones, T., Howat, P., Sauer, K., & Elliott, E. J. (2014). Fine motor skills in children with prenatal alcohol exposure or Fetal Alcohol Spectrum Disorder. *Journal of Developmental and Behavioral Pediatrics, 35*(9), 598-609. doi:10.1097/dbp.0000000000000107

Fine Motor Skills in Children With Prenatal Alcohol Exposure or Fetal Alcohol Spectrum Disorder

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ABSTRACT: ***Objective:*** **Prenatal alcohol exposure (PAE) can cause fetal alcohol spectrum disorders (FASD) and associated neurodevelopmental impairments. It is uncertain which types of fine motor skills are most likely to be affected after PAE or which assessment tools are most appropriate to use in FASD diagnostic assessments. This systematic review examined which types of fine motor skills are impaired in children with PAE or FASD; which fine motor assessments are appropriate for FASD diagnosis; and whether fine motor impairments are evident at both "low" and "high" PAE levels.** ***Methods:*** **A systematic review of relevant databases was undertaken using key terms. Relevant studies were extracted using a standardized form, and methodological quality was rated using a critical appraisal tool.** ***Results:*** **Twenty-four studies met inclusion criteria. Complex fine motor skills, such as visual-motor integration, were more frequently impaired than basic fine motor skills, such as grip strength. Assessment tools that specifically assessed fine motor skills more consistently identified impairments than those which assessed fine motor skills as part of a generalized neurodevelopmental assessment. Fine motor impairments were associated with "moderate" to "high" PAE levels. Few studies reported fine motor skills of children with "low" PAE levels, so the effect of lower PAE levels on fine motor skills remains uncertain.** ***Conclusions:*** **Comprehensive assessment of a range of fine motor skills in children with PAE is important to ensure an accurate FASD diagnosis and develop appropriate therapeutic interventions for children with PAE-related fine motor impairments.**

*(J Dev Behav Pediatr 35:*598–609, 2014) **Index terms:** fetal alcohol spectrum disorders, motor skills, psychomotor performance, child development.

Prenatal alcohol exposure (PAE) can result in a range of lifelong neurological impairments in offspring termed fetal alcohol spectrum disorders (FASD).[1–3] Diagnoses on the spectrum include fetal alcohol syndrome (FAS), which includes characteristic dysmorphic facial features and growth impairments; partial FAS, with some facial and growth impairments; and alcohol-related neurodevelopmental disorder, in which individuals do not have facial or growth changes. All diagnoses have significant neurological impairments, which can include impaired cognition, executive function, memory, language, attention, social and adaptive skills, and motor skills.[2]

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Prenatal Alcohol Exposure and Motor Skills

Prenatal alcohol exposure can affect both fine motor (FM) and gross motor (GM) skills.[1] Early reports noted that infants with FAS had motor impairments,[4] and subsequently, orthopedic and structural defects were also recorded, including clinodactyly (fixed laterally curved fifth finger), camptodactyly (fixed finger flexion), impaired upper limb pronation/supination, tapering of the distal phalanges, and resting and kinetic hand tremors.[5–7] In animal studies, PAE has been associated with impaired myelination of spinal and peripheral nerves,[8,9] and impaired motor coordination, response, speed, activity, reflexes, and tone.[10,11] Neuroimaging studies of individuals with PAE or FASD have identified damage to specific brain regions. Damaged regions that may affect motor skills include the cerebellum,[12] basal ganglia,[13] corpus callosum,[14] and hippocampus.[12] PAE can also damage neural circuits, including projections which extend into motor and premotor cortices.[15] There are limited studies of the motor cortex in relation to PAE. However, one study of adolescents with PAE concluded that observed motor impairments likely were due to damage to the motor cortex.[16]

Fine Motor Skills in Fetal Alcohol Spectrum Disorder Diagnosis

Fine motor skills require the use of small hand muscles and include speed, accuracy, control, coordination, dexterity, visual motor skills, and eye-hand coordination.[17] FM skills are important in children because they facilitate increasing independence in self-care tasks such as dressing, eating, brushing hair, and cleaning teeth; academic skills including handwriting, drawing, and using scissors; and participation in play and social activities.[18] Parents and teachers of primary school-aged children with FASD often report that they have difficulty with many of these functional tasks.[19]

Fetal alcohol spectrum disorders diagnostic guidelines universally advise that motor skills should be assessed in children with PAE, but variation exists regarding whether FM skills are differentiated from GM skills and which assessment tools are recommended. The Canadian Guidelines recommend specific tools, such as the Movement Assessment Battery for Children (M-ABC), the Bruininks-Oseretsky Test of Motor Proficiency, and the Beery-Buktenica Developmental Test of Visual-Motor Integration.[2] The UW 4-Digit Diagnostic Code guidelines do not recommend specific assessment tools but advise that they should be standardized and validated.[20] The Institute of Medicine and Centers for Disease Control guidelines offer no advice about which assessment tools to use.[1,3]

It has been suggested that generalized neurodevelopmental assessments may not detect subtle and specific impairments resulting from PAE.[21,22] Many neurodevelopmental assessment tools used to assess children with PAE are problematic in terms of FM assessment: some, such as the Wechsler Intelligence Scale for Children,[23] do not include a motor skills component; others, such as the McCarthy Scales of Children's Abilities,[24] provide an overall motor score which is a composite of FM and GM skills, which may mask specific areas of FM or GM impairment[25]; whereas others, such as the Griffiths Mental Development Scales,[26] assess only limited types of FM skills.

Fine Motor Skills in Children with Prenatal Alcohol Exposure or Fetal Alcohol Spectrum Disorders

Three systematic reviews have examined the relationship between PAE and motor skills. The first review concluded that only high levels of PAE (10–30 drinks per week) were associated with impaired motor function, although findings were not consistent between studies.[27] However, this study did not explore whether different types of motor skills were more likely to be impaired or if FM skills were affected independently of GM skills. The second systematic review and meta-analysis found that GM skills were 2.9 times more likely to be impaired in children aged 0 to 18 years with "moderate" to "high" PAE compared with children without PAE, but this review did not include FM skills or lower levels of PAE within the scope of the review.[28] The third study of neurological impairments included 13 studies of "visual and motor" skills and concluded that impairments were not associated with "mild," "moderate," or "binge" PAE, but none of the included assessed children were older than 5 years.[29]

Aims and Hypotheses

Although FASD diagnostic guidelines recommend assessing motor skills in children with PAE, it is unclear which types of FM skills are most likely to be impaired or which assessment tools are best to use. It is also uncertain whether impaired FM skills account for the functional difficulties reported by caregivers and teachers of children with FASD. This knowledge is essential to ensure accurate and timely FASD diagnosis, contribute to the development of an FASD neurological profile, perform objective measurement of FM skills over time, and develop therapeutic programs which promote independence by enhancing FM strengths and supporting areas of difficulty.

This systematic review examined FM skills in primary school-aged children (4–12 years) with PAE or FASD and aimed to:

1. Establish which types of FM skills are impaired after PAE;
2. Identify which FM assessment tools are commonly used with children with PAE or FASD; and
3. Investigate whether different levels of PAE are associated with FM impairments.

Given the diversity of neural regions affected by PAE, we hypothesized that a range of FM skills would be impaired in children with PAE or FASD, especially those which are more complex and involve multiple neural regions and connectivity. We further hypothesized that FM impairments would be more evident among children with high levels of PAE and that neurodevelopmental assessments which assessed FM skills as part of a generalized assessment battery would not identify FM impairments.

METHODS

Literature Search

A search strategy was used to systematically search peer-reviewed journals in the Web of Knowledge, Ovid (Medline, PsychInfo, Maternity and Infant Care, Embase, and Amed), EBSCOhost (Cumulative Index to Nursing and Allied Health Literature), ProQuest, SciVerse, The Cochrane Library, Emerald, InformIT, OT Seeker, and PEDro databases. The following key words were combined to identify relevant articles: alcohol*, fetal, maternal, prenatal*, in utero, and fine motor. No articles were identified that specifically reported fine motor (FM) skills in relation to prenatal alcohol exposure (PAE), so search terms were broadened to include motor, neuro*, development*, sensor*, and visu*. Experts in the field were contacted to identify unpublished studies, and reference lists of relevant articles were manually searched.

Inclusion Criteria and Selection Process

Full texts were obtained for articles that met the study inclusion criteria (Table 1). Included FM skills were control, precision, speed, or accuracy; visual-motor integration or visuomotor precision; in-hand manipulation; hand grasp and release; manual dexterity or coordination; and foundational skills including praxis, grip strength, and finger tapping. Functional FM skills included pencil grasp and writing pressure, handwriting or drawing skills, and adaptive skills with an FM component such as tying shoelaces or doing up buttons. Studies were excluded if they only reported neuromuscular performance such as range of motion, reflexes, or skeletal deformities; gross motor (GM) skills (walking, running, strength, or balance); visual perception (acuity, convergence, tracking, and eye-blink conditioning); visual cognitive skills (visual discrimination, visual memory, form constancy, spatial skills, figure ground, or visual closure); or sensory processing skills.

Data Extraction and Quality Assessment

There were 24 studies which met all the inclusion criteria. These were summarized by 2 authors (R.D. and T.J.) using a data extraction form (Table 2 and Supplemental Digital Content 1, http://links.lww.com/JDBP/A65). A 10-point critical appraisal tool was developed based on STROBE and Health Evidence guidelines to assess the methodological quality and relevance (Table 3). The studies were independently rated by 2 authors (R.D. and T.J.) and consensus agreement reached. Studies were classified as having either "low" (0–4), "moderate" (5–7), or "strong" (8–10) methodological quality. Studies that quantified PAE levels as ounces of absolute alcohol per day or week were converted to drinks per day or drinks per week (1 oz absolute alcohol = 28.35 g; 14 g approximates 1 standard drink). A meta-analysis was not conducted because of the heterogeneity of FM skills and assessment tools, and the variability of PAE levels and FASD diagnoses between studies.

RESULTS

Literature Search

We identified 6259 studies after removal of duplicates (Fig. 1). An additional 10 studies were identified by manually searching reference lists. Unpublished studies were not identified from experts in the field. A total of 6173 studies were excluded because they did not meet inclusion criteria, and a further 72 studies were excluded for not meeting fine motor (FM)-related criteria. This resulted in 24 studies eligible for inclusion.

Study Characteristics

Characteristics of the 24 included studies, including prenatal alcohol exposure (PAE) levels and fetal alcohol spectrum disorder (FASD) diagnoses when provided, are outlined in Supplemental Digital Content 1 (http://links.lww.com/JDBP/A65). Included studies were published between 1985 and 2012. Sample sizes ranged from 10 to 685 and included 8 small ($n < 50$), 7 medium ($n = 50$–100), and 9 large ($n > 100$) studies.

Table 1. Inclusion and Exclusion Criteria

	Inclusion Criteria	Exclusion Criteria
Time	Any time until December 2012	January 2013 onwards
Language	English	All other languages
Design	RCTs	Descriptive studies
	Cohort studies	Case studies
	Case–control studies	Reviews
Publications	Original research	Non-peer–reviewed articles
	Peer-reviewed journals	Conference abstracts/posters
		Postgraduate theses
Participants	Humanistic studies	Animal studies
	4–12 yrs (all or some participants within this range)	Babies, infants, adolescents, adults
PAE	FASD diagnoses	Nil
	PAE (including low-high, binge, any trimester)	
FM Assessment	Direct assessment of skills by clinician/therapist	Parental or teacher report
	Published, commonly available, standardized assessments	Observational data, e.g., handedness; tremor
	Assessments using standardized equipment, e.g., dynamometer; mechanical finger tapper	Assessments not commonly available to clinicians (e.g., robotic or computerized tasks)
FM results	Reported separately to GM and other neurological results	Composite GM/FM outcomes (e.g., McCarthy Scales of Children's Abilities)
	Statistical significance of results reported	

FASD, fetal alcohol spectrum disorder; FM, fine motor; GM, gross motor; PAE, prenatal alcohol exposure; RCT, randomized control trial.

Table 2. Fine Motor Assessments and Outcomes

	Visual Motor Skills			Dexterity		Foundational FM Skills				Functional FM Skills	
Study	**VMI**	**Eye-Hand**	**VM Precision**	**Speed and Precision**	**Manual Dexterity**	**Grip Strength**	**Finger Tapping**	**Praxis**	**Sensorimotor**	**Graphomotor**	**Motor Inhibition**
Undefined; or low, moderate and high PAE											
Barr et al[30]				±*GPT*[a]	±*WFMSB*[b]	−*Dyna*	+*HRNB*				
Bay et al[31]					−*M-ABC*						
Coles et al[32]	+*BVMI*										
Chiodo et al[33]	±*PENTB*[c]			±*PPT*[d]							
Fried and Watkinson[34]				−*GPT*							
Irner et al[35]		±*GMDS*[c]									
Russell et al[36]	−*BVMI*										−*CATB*
Moderate to high PAE; or pFAS/FAS											
Adnams et al[21]		+*GMDS*									
Aragón et al[37]				−*GPT*							
Aronson et al[38]		+*GMDS*									
Janzen et al[39]	+*BVMI*			−*GPT*[f]							
Jirikowic et al[40]									+*NEPSY*[g]		
Korkman et al[41]	+*BVMI*		−*NEPSY*[h]					±*NEPSY*[i,j]			
Laforce et al[42]				−*PPT*							
Mattson et al[43]	±*BVMI*[k]			±*GPT*[l]							
Mattson et al[44]	+*BVMI*			+*GPT*							
Sowell et al[45]	+*BVMI*										
Uecker and Nadel[46]	+*BVMI*										
Vaurio et al[47]	+*BVMI*			−*GPT*							
FASD (PAE levels unspecified)											
Astley et al[48]	±*BVMI*[m]										
Conry[49]	±*BVMI*[n]			±*DTLA*[o]		±*Sphyg*[p]	±[q]				

(*Table continues*)

Table 2. Continued

Study	Visual Motor Skills			Dexterity		Foundational FM Skills			Functional FM Skills		
	VMI	Eye-Hand	VM Precision	Speed and Precision	Manual Dexterity	Grip Strength	Finger Tapping	Praxis	Sensorimotor	Graphomotor	Motor Inhibition
Henry et al[50]								−*PEEX*		−*PEEX*	
Kooistra et al[51]					+*M-ABC*						
Zhou et al[52]			+*NEPSY*[b]								

[a]Significant ($p = .018$) impairments in time to complete for children with early pregnancy PAE (time to complete); peg drop count outcomes and midpregnancy PAE outcomes not reported. [b]Significant ($p = .011$; $p = .000$) number of errors and latency to correct for children with early pregnancy PAE; midpregnancy PAE outcomes not reported. "Significant ($p < .025$) for "at-risk alcohol measure" only; "Significant ($p < .05$) for children with PAE "at time of birth," but nonsignificant ($p = .59$) for children with PAE "during pregnancy"; "NB comparisons for "during pregnancy" made between children with PAE, no polydrug, polydrug/no PAE, and polydrug PAE groups only). [c]Nonsignificant number of pegs placed in 1 minute, left ($p = .011$) or right ($p = .047$) hands; authors have used conservative p values ($p < .01$). +NEPSY, Sensorimotor domain (includes visuomotor precision, finger tapping, imitating hand positions, and manual motor series [praxis] subtests). [d]NEPSY, Visuomotor precision subtest. "Significant ($p < .0001$) for dynamic praxis; nonsignificant for kinesthetic praxis. +NEPSY, Manual motor series (dynamic praxis) and imitating hand positions (kinesthetic praxis) subtests. "Significant ($p < .001$) between FAS and controls; nonsignificant between PAE and controls, or FAS and PAE. "Significant ($p < .01$) nondominant hand; nonsignificant dominant hand. "Significant ($p < .05$) between controls, pFAS, FAS, neurodevelopmental disorder/alcohol exposed, and SE/AE; nonsignificant between pFAS/FAS and SE/AE. "Significant ($p < .001$) between controls and FAS, and FAS and PAE; nonsignificant between controls and PAE. "Significant ($p = .002$) between controls and FAS; nonsignificant between controls and PAE, and FAS and PAE. "Significant ($p < .001$) dominant and nondominant hands between controls and FAS, and controls and PAE; nonsignificant between FAS and PAE. "Significant ($p = .005$) dominant hand between controls and FAS, and controls and PAE; nonsignificant between FAS and PAE. "Significant ($p = .006$) nondominant hand between controls and FAS; nonsignificant between controls and PAE, and FAS and PAE. +, Significant difference between children with PAE/FASD and a control group; +, Both significant and nonsignificant outcomes, for example, between FAS and controls, but not FAE and controls; or for the dominant hand but not the nondominant hand; −, Nonsignificant difference. BVMI, Beery-Buktenica test of visual motor integration; CATB, The Cincinnati Autonomy Test Battery (Draw-a-line slowly test); Dyna, Dynamometer; DTLA, Detroit Tests of Learning Aptitude (The motor speed and precision test); FAE, fetal alcohol effect; FAS, fetal alcohol syndrome; GMDS, Griffiths Mental Development Scales (Eye and hand coordination subscale); GPT, Grooved Pegboard test; HRNB, Halstead-Reitan Neuropsychological Battery (Finger tapping test); M-ABC, Movement Assessment Battery for Children (Fine motor subarea); NEPSY, Developmental Neuropsychological Examination; PAE, prenatal alcohol exposure; PEATB, Pediatric Environmental Neurobehavioral Test Battery (Visual-motor integration test); PFEX, Pediatric Early Entry Examination or Pediatric Examination of Educational Readiness at Middle Childhood (fine motor/graphomotor test); pFAS, partial fetal alcohol syndrome; PPT, Purdue Pegboard test; SE/AE, static encephalopathy/alcohol exposed; Sphyg, Sphygmomanometer; VM, visual motor; VMI, visual-motor integration; WPMSB, Wisconsin Fine Motor Steadiness Battery (vertical/horizontal groove board; grooved maze board; resting steadiness hole board).

Risk of Bias and Quality Assessment

Studies were rated using the critical appraisal tool (Table 3). Most of the studies had moderate (58.3%) or strong (16.7%) methodological quality. Half of the studies (50%) blinded assessors to PAE levels or FASD diagnoses as a strategy to reduce bias. All studies discussed potential confounding factors, but variation existed regarding which confounding factors were considered relevant, and whether these were controlled for statistically. Most studies used at least 1 assessment tool that specifically assessed motor skills, as opposed to generalized neurodevelopmental assessments with an FM component (75.0%), but less than half of the studies assessed more than 1 type of FM skill (41.7%).

Fetal Alcohol Spectrum Disorders and Prenatal Alcohol Exposure

Most studies included children with FASD diagnoses (70.8%), but 7 of these included only children with the more severe diagnoses of partial fetal alcohol syndrome (pFAS) or fetal alcohol syndrome (FAS) (29.2%). Only 3 studies reported FM outcomes for children with "no/low" PAE <0.14 oz per day (<2 drinks per week),[34] low PAE (1–4 drinks per week),[31] or light/moderate PAE (0–1 oz per day or 0–14 drinks per week).[36] A further 3 studies included children with low PAE but did not stratify FM outcomes by PAE levels.[30,33,35] Thirteen studies only included children with moderate or high PAE or children with pFAS or FAS (who likely had "moderate to high" PAE as these diagnoses include the most severe effects of PAE). Not all studies quantified PAE levels, but those that did variously defined moderate and high PAE as moderate (5–8 drinks per week),[31] moderate to high/heavy (>10 drinks per week),[41] >0.14 oz per day (or >2 drinks per week),[34] high/heavy (9–14 drinks per week),[31] 0.5 to 1.5 oz per day (or 7–21 drinks per week),[30] >13 or >14 drinks per week or >4 drinks per occasion,[44,47] 1 to 3.5 oz per day (or 14–49 drinks per week),[36] and "very heavy" (>3.5 oz per day or >49 drinks per week).[36] A further 4 studies included children with a range of FASD diagnoses but did not specify PAE levels.[49–52]

Fine Motor Assessments and Outcomes

The most common types of FM skills assessed were visual motor skills and manual dexterity. Researchers used a range of different FM assessment tools (Table 2).

Visual Motor Skills (n = 17)

Studies of visual motor skills included visual-motor integration (VMI), eye-hand coordination, and visuomotor precision. Assessments included a combination of drawing and functional tasks such as threading beads or posting coins.

Beery-Buktenica Developmental Test of Visual-Motor Integration (Beery VMI) (n = 11)

The Beery VMI[53] was the most commonly used assessment in this review. The Beery VMI requires children to copy increasingly complex geometric shapes. All but

Table 3. Study Quality and Relevance

	Sample				Assessment				Bias		
Study	Age[a]	Defined Sample[b]	PAE/ FASD[c]	Matching[d]	Size[e]	FM Depth[f]	Psychometrics[g]	FM Specificity[h]	Blinding[i]	Confounders[j]	Total/ 10
Adnams et al[21]	✓	✓	✓	✓	✗	✗	✗	✗	✓	✓	6
Aragón et al[37]	✗	✓	✓	✓	✗	✗	✓	✓	✗	✓	6
Aronson et al[38]	✗	✗	✗	✓	✗	✗	✗	✗	✗	✓	2
Astley et al[48]	✗	✓	✓	✓	✓	✗	✗	✓	✓	✓	7
Barr et al[30]	✓	✓	✓	✗	✗	✓	✗	✓	✓	✓	7
Bay et al[31]	✓	✓	✓	✓	✗	✗	✓	✓	✓	✓	8
Chiodo et al[33]	✓	✓	✓	✓	✗	✓	✓	✓	✓	✓	9
Coles et al[32]	✓	✓	✓	✓	✗	✗	✗	✓	✓	✓	7
Conry[49]	✗	✓	✓	✓	✗	✓	✓	✓	✓	✓	8
Fried and Watkinson[34]	✓	✓	✓	✗	✗	✗	✗	✓	✓	✓	6
Henry et al[50]	✗	✗	✓	✗	✗	✓	✓	✗	✗	✓	4
Irner et al[35]	✗	✗	✓	✓	✗	✗	✗	✗	✗	✓	3
Janzen et al[39]	✗	✓	✓	✓	✗	✓	✓	✓	✗	✓	7
Jirikowic et al[40]	✓	✓	✓	✗	✗	✗	✓	✗	✓	✓	6
Kooistra et al[51]	✓	✓	✓	✗	✗	✗	✓	✓	✓	✓	7
Korkman et al[41]	✓	✓	✓	✗	✗	✓	✓	✓	✗	✓	7
Laforce et al[42]	✗	✓	✗	✓	✗	✗	✗	✓	✗	✓	4
Mattson et al[43]	✗	✓	✓	✓	✗	✓	✗	✓	✗	✓	6
Mattson et al[44]	✗	✓	✓	✗	✗	✓	✓	✓	✗	✓	6
Russell et al[36]	✓	✓	✓	✓	✗	✓	✗	✓	✓	✓	8
Sowell et al[45]	✗	✓	✓	✓	✗	✗	✓	✓	✗	✓	6
Uecker and Nadel[46]	✗	✗	✓	✓	✗	✗	✗	✓	✗	✓	4
Vaurio et al[47]	✗	✓	✓	✓	✗	✓	✗	✓	✓	✓	7
Zhou et al[52]	✗	✓	✓	✓	✗	✗	✗	✗	✗	✓	4

[a]Entire sample aged 4 to 12 years. [b]Cohort studies: selection process provided; case–control studies: inclusion/exclusion criteria provided. [c]Method of PAE ascertainment provided (e.g., interview; review of medical records); or FASD diagnostic criteria provided. [d]Cohort studies: sample comparable with FM assessment normative data; or case–control studies: matched cases and control groups. [e]Sample size/power calculation; or justification provided for sample size (e.g., population study). [f]More than 1 FM skill reported, either using 2 different assessments; or 1 assessment which assesses multiple types of FM skills. [g]Psychometrics for assessment provided; or justification for assessment choice. [h]FM-specific assessment (i.e., not part of general neuropsychological assessment). [i]Assessors blinded to PAE or FASD status at time of assessment. [j]Consideration given to potential confounding factors. ✓, criterion met; ✗, criterion not met/ uncertain; FASD, fetal alcohol spectrum disorder; FM, fine motor; PAE, prenatal alcohol exposure.

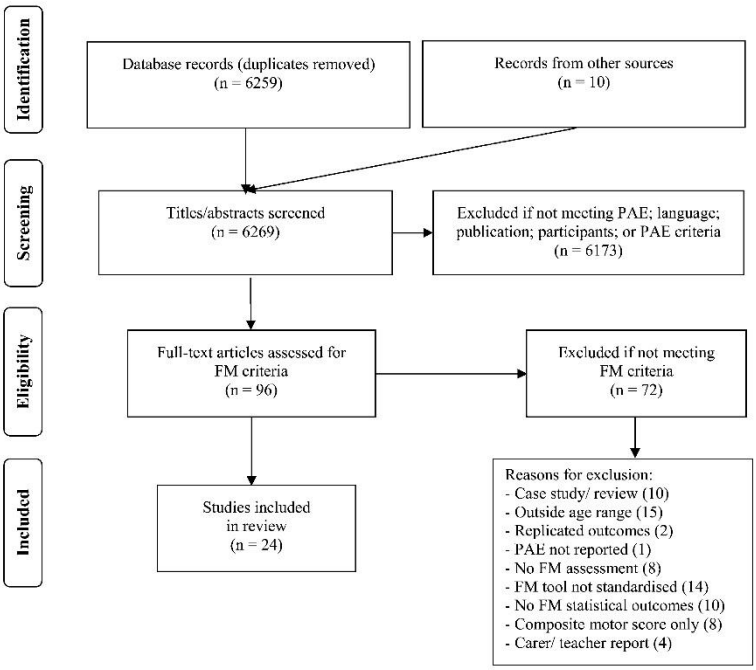

Figure 1. Flowchart for article inclusion. FM, fine motor; PAE, prenatal alcohol exposure.

one study[36] that used this assessment tool reported significantly impaired VMI skills, including in children with a range of FASD diagnoses.[32,35,47–49] One study identified VMI impairments in children with moderate to high PAE (>10 drinks per week) in the first trimester, first and second trimesters, and all 3 trimesters.[41] In contrast, another study with 5- to 16-year-old children with FAS had impaired VMI skills, but these findings did not extend to children with "heavy" PAE (unquantified levels) but no FASD diagnosis.[43] However, a subsequent larger multisite study by the same researchers did identify significant VMI impairment in children with heavy PAE (>13 drinks per week; or >4 drinks per occasion) but no FASD diagnosis.[44] Only one study did not report VMI impairments when these skills were assessed using the Beery VMI.[36] This study included 6-year-old children with "none"; "light to moderate" (0–2 drinks per day); heavy (2–7 drinks per day); or very heavy (>7 drinks per day) PAE early in pregnancy. In contrast to other studies, PAE in this study included any alcohol consumption in the year leading up to pregnancy, as the authors considered that this was the most accurate proxy for PAE early in the first trimester.

Pediatric Environmental Neurobehavioral Test Battery (PENTB): Visual-Motor Integration Test (n = 1)

The PENTB includes a VMI component, which consists of copying line drawings.[54] Only one study assessed VMI skills using the PENTB.[33] When VMI skills were analyzed using the newly developed "at-risk alcohol exposure" measure, impairments remained significant after adjustment for potential confounding factors.

Griffiths Mental Development Scales (GMDS): Eye and Hand Coordination Subscale (n = 3)

Three studies used the eye and hand coordination subscale of the GMDS, which assesses eye-hand coordination by having the child copy geometric figures, as well as functional tasks such as threading beads.[26] Two studies found significant eye and hand coordination impairments, including in children with FAS[21] and children of alcohol-dependent mothers.[38] The third study of children aged 3 to 7 years, at least half of whom had high PAE (>15 drinks per week), identified significant eye-hand coordination impairments.[35]

A Developmental Neuropsychological Assessment (NEPSY): Visuomotor Precision Subtest (n = 3)

The NEPSY visuomotor precision subtest assesses FM speed and accuracy through a timed pencil and paper maze.[55] One study found no significant visuomotor precision impairments in 5- to 9-year-old children with moderate to high PAE (>10 drinks per week) in either the first, first and second, or all trimesters.[41] In contrast, the second study found significant visuomotor precision impairments in 6- to 12-year-old children with FASD. PAE levels were unspecified in this study.[52] The third study

used the sensorimotor domain of NEPSY, which includes visuomotor precision, finger tapping, imitating hand positions, and manual motor series subtests, to assess 5- to 8.5-year-old children with FASD, of whom 60% had high PAE.[40] This study did not report visuomotor precision findings independently of the other subtests but found that the overall sensorimotor domain scores were significantly impaired.

Fine Motor Dexterity (Speed and Coordination) (n = 13)

Fine motor dexterity was reported in 13 studies using 4 different assessment tools.

Pegboard Tests (n = 9)

Pegboard tests assess a range of FM skills, including manual dexterity, precision, speed, and accuracy. Nine studies used pegboard tests and reported varying outcomes. Assessment tools included the Purdue Pegboard Test, the Grooved Pegboard Test, and pegboard tests that comprise part of generalized neurodevelopmental assessment tools, such as the Wisconsin FM Steadiness Battery (WFMSB). Three studies reported nonsignificant results in children with pFAS or FAS (unspecified PAE levels),[37] FAS or fetal alcohol effects (a term previously used to describe FASD diagnoses other than FAS) with heavy PAE (unquantified levels),[42] and children with heavy PAE (>14 drinks per occasion or >14 drinks per week).[47] One study reported nonsignificant results among children with "none/light" PAE (<0.14 oz of absolute alcohol per day or <2 drinks per week) compared with children with "moderate/heavy" PAE (>0.14 oz of absolute alcohol per day or >2 drinks per week).[34] Another study reported nonsignificant results in left or right hands in children with FAS born to mothers with alcohol dependency, although conservative *p*-values were used, which may have been considered significant in other studies.[39] At 4 years, children with PAE (7–21 drinks per week) early in pregnancy, and children who were exposed to an average of more than 1 drink per day throughout pregnancy, took longer to complete the pegboard but did not make more errors than children without PAE.[30] A different study examined whether different ways of quantifying PAE affected the findings.[33] This study found that a dichotomous measure of x at-risk alcohol exposure was associated with fewer pegs being placed accurately, but these findings were not significant when other measures of PAE were used. A further study reported some significant results when comparing different FASD diagnoses, but findings were not consistent between dominant and nondominant hands.[43] In contrast, a larger multisite study by the same authors found participants with FAS and heavy PAE (>13 drinks per week or >4 drinks per occasion) had impairments in both their dominant and nondominant hands.[44]

Movement Assessment Battery for Children (M-ABC): Manual Dexterity Subarea (n = 2)

The M-ABC manual dexterity component includes items such as posting coins, threading beads, and completing a paper and pencil drawing trail.[56] The M-ABC was used in 2 studies to assess manual dexterity skills and reported contrasting results. The first study found no significant manual dexterity impairments in 5-year-old children stratified by different PAE levels.[31] This study mostly included "low to moderate" PAE (≤8 drinks per week), with only 1.6% (n = 11) of the sample being exposed to more than 9 drinks per week, and no children being exposed to more than 14 drinks per week. The second study found that manual dexterity skills were significantly impaired among 7- to 10-year-old children with various FASD diagnoses, as well as children with attention-deficit hyperactivity disorder.[51] This study did not report levels of PAE among the children with FASD.

Detroit Tests of Learning Aptitude (DTLA): Timed Motor and Precision Test (n = 1)

One study[49] used the DTLA Timed Motor and Precision Test, which assesses FM speed and precision by rapidly drawing crosses in circles of decreasing size.[57] This study found significant differences in FM speed and precision skills among children aged 5 to 18 years with FAS, but not children with fetal alcohol effect (FAE). PAE levels were not defined in this study.

Wisconsin Fine Motor Steadiness Battery (WFMSB) (n = 1)

One study used the FM tests from the WFMSB[58] to assess 4-year-old children with no PAE, children with PAE only in early pregnancy, and children with PAE throughout pregnancy.[30] This study reported that the children with "moderate to heavy" PAE (7–21 drinks per week) early in pregnancy made significantly more errors and were slower to self-correct their errors compared with children with no PAE. These impairments were significant for children with PAE early in pregnancy as well as children with PAE (average ≥1 drink per day) throughout pregnancy. These difficulties remained significant after adjustment for IQ and other confounding factors.

Foundational Fine Motor Skills: Grip Strength, Finger Tapping, and Praxis

Grip Strength (n = 2)

Two studies reported grip strength findings with mixed results. The first study identified no significant differences in grip strength among 4-year-old children with PAE early or throughout pregnancy.[30] PAE levels in this study ranged from 0.5 to 1.5 oz of absolute alcohol per week (7–21 drinks per week). The second study identified that 5- to 18-year-old children with FAS or FAE had significant impairments when using both their dominant and nondominant hands.[49] PAE levels were not defined for the children in this study.

Finger Tapping (n = 3)

Finger tapping was assessed in 3 studies with mixed results. Two studies used a mechanical finger tapper to record how many times the child could tap their finger while holding the other fingers stationary, and the third study measured finger tapping as part of the NEPSY

sensorimotor domain. One study reported a significant finger tapping impairment in 5- to 18-year-old Native Americans with FAS or FAE in both their dominant and nondominant hands.[49] In contrast, the second study found that 4-year-old children with moderate to heavy PAE in early pregnancy, but not midpregnancy, had impaired finger tapping skills.[30] The third study assessed finger tapping skills in 5- to 8.5-year-old children with FASD, of whom 60% had high PAE (unquantified levels), but finger tapping results were not reported independently of other tasks in the sensorimotor domain.[55]

Praxis (n = 2)

One study assessed kinesthetic and dynamic praxis using the "Imitating Hand Positions" and "Manual Motor Series" subtests from the sensorimotor domain of the NEPSY.[55] These tests require imitation of static hand positions and rhythmic hand movements. This study found that dynamic praxis, but not kinesthetic praxis, was affected in 5- to 9-year-old Finnish children with moderate to high PAE (≥10 drinks per week).[41] Dynamic praxis was impaired in children who had been exposed to moderate to high PAE in the first, first and second, and all 3 trimesters of pregnancy.

Combined Foundational Fine Motor Skills (n = 2)

Two studies reported combined outcomes of different types of foundational hand skills. The first study reported outcomes from the sensorimotor domain of NEPSY, which includes dynamic and kinesthetic praxis, finger tapping, and visuomotor precision.[40] These authors found that children with FASD (60% with high levels of PAE) had impaired sensorimotor skills. The second study assessed FM skills in 6- to 16-year-old children with FASD, who had also experienced significant trauma, using the Pediatric Early Elementary Examination (PEEX-2) or the Pediatric Examination of Educational Readiness at Middle Childhood (PEERAMID-2).[50] Tasks included lateral preference, imitative finger movements, finger tapping, and sequential finger opposition. These authors did not identify significant differences between children with or without FASD. PAE levels were not defined in this study.

Functional Fine Motor Skills

Two studies included assessment of functional FM skills as part of a generalized neurodevelopmental assessment and neither found significant impairments. The first study assessed graphomotor skills using the PEEX-2 or the PEERAMID-2 with 6- to 16-year-old children, who had experienced trauma and compared outcomes between children with and without FASD. Tasks included pencil control, pencil speed, and writing the alphabet.[50] The second study used the "Draw a Line Slowly" test from The Cincinnati Autonomy Test Battery to assess motor inhibition skills. No impairments were identified in 6-year-old children with none, light to moderate (0–2 drinks per day), heavy (2–7 drinks per day), or very heavy (>7 drinks per day) levels of PAE early in pregnancy.[36]

DISCUSSION

This review identified 24 studies which assessed fine motor (FM) skills in primary school–aged children with prenatal alcohol exposure (PAE) or fetal alcohol spectrum disorders (FASD). A range of FM skills, including visual motor skills (visual-motor integration [VMI], visuomotor precision, and eye-hand coordination), manual dexterity (speed, coordination, and precision), foundational FM skills (grip strength, finger tapping, and praxis), and graphomotor (handwriting, drawing, and motor inhibition) skills were assessed using a variety of FM-specific and generalized neurodevelopmental assessments.

Complex FM skills, such as VMI, were consistently impaired in children with moderate to high PAE and with pFAS or fetal alcohol syndrome (FAS) diagnoses. These findings support the rationale that complex skills, which are controlled by several neural regions and involve multiple neural pathways, are more likely to be impaired after PAE.[59] VMI skills were impaired in most studies which used the Beery VMI, but findings were less consistent when visual motor skills were assessed using generalized neurodevelopmental assessments such as the pediatric environmental neurobehavioral test battery or Griffiths Mental Development Scales. It may be that generalized neurodevelopmental assessments do not assess visual motor skills in sufficient detail or alternatively that the constructional visual motor skills are less sensitive to PAE than graphomotor visual motor skills. VMI deficits were evident in both young children and adolescents and also across different trimesters of PAE exposure, making this an important FM skill to assess in children with PAE for both diagnostic and therapeutic purposes.

Pegboard tests were used to assess manual dexterity, precision, speed, and coordination, which are also relatively complex FM skills. The Purdue Pegboard Test requires placement of pegs into round holes, whereas the Grooved Pegboard Test requires pegs to be rotated for accurate placement, and thus may be considered a more complex task. However, despite the differences in complexity, studies which used pegboard tests as an FM outcome measure did not consistently identify impairments, despite most studies only including children with moderate to high PAE, who would be most likely to show impairments. It is possible that the pegboard tasks may be too simple to adequately detect subtle and complex FM impairments resulting from PAE.

Few studies investigated foundational FM skills, such as grip strength, finger tapping, praxis, and kinesthesia, and each used different assessment tools, so outcomes were difficult to compare between studies. Only 2 studies reported grip strength and finger tapping skills, which are relatively basic FM skills, and reported varying results.[30,49] The contrasting outcomes may be due to age differences of the children in each study, which is consistent with the theory that PAE-related impairments become more pronounced with maturity.[60] Although it is important to assess foundational

FM skills in individual children with PAE as they may be affected and therefore warrant therapeutic intervention and support, evidence suggests that they do not provide a reliable marker for FASD presentation and diagnosis.

Only 2 studies reported functional outcomes of FM skills, and both used assessments of handwriting or drawing as a small part of a generalized neurodevelopmental assessment.[36,50] One study included children with exposure to trauma and compared outcomes between those with and without FASD with undefined levels of PAE.[50] The second study included children with various levels of PAE, which included alcohol consumption in the year before pregnancy as a proxy measure for PAE early in pregnancy.[36] The demographic and methodological differences between these 2 studies make it difficult to compare outcomes, but neither study identified impaired graphomotor (handwriting or drawing) skills.

Only 3 studies stratified FM outcomes specific to low PAE, and these studies did not identify FM impairments. Low PAE was variously defined as <2 drinks per week,[34] 1 to 4 drinks per week,[31] and 0 to 14 drinks per week in the year leading up to pregnancy (as a proxy measure for early first trimester PAE).[36] Each of these studies reported different types of FM skills (VMI, motor inhibition, and manual dexterity), and each used different assessment tools, so outcomes were difficult to compare. The effect of low PAE on specific type of FM skills remains uncertain.

Studies of Functional Fine Motor Skills Which Did Not Meet Inclusion Criteria

Several other studies of functional FM skills were identified that did not meet inclusion criteria for this review. However, impaired functional FM skills are frequently reported by parents and teachers of children with FASD, and these studies warrant mentioning. Caregivers of 5- to 8-year-old children reported difficulties with adaptive function, many of which require FM skills, including name writing, fastening clothing, and brushing teeth.[40] In another study, only 7% of the children with FAS had adaptive FM skills in the "adequate" range, with the remainder scoring in the low or "moderately low" range.[61] One study assessed handwriting abilities in 20 primary school-aged children with FASD and found that they scored well-below average in handwriting legibility, speed, and visuomotor precision. This was an exploratory descriptive study, and no control group was used for comparison.[62]

Neuroanatomical Explanations for Fine Motor Impairments

Damage to the cerebellum,[12] basal ganglia,[13] corpus callosum,[14] and parietal lobes[12] may account for some FM impairments. The cerebellum is commonly associated with balance but also incorporates information from the visual system to control oculomotor function,[63] which may contribute toward VMI impairments. The basal ganglia modulates emotional responses but also has connections to motor areas, including in the frontal cortex and thalamus. Neuroimaging studies have shown that the corpus callosum may be reduced, misplaced, or missed in individuals with FASD,[14] and the role of the corpus callosum in relaying communications between hemispheres may explain bimanual coordination and manual dexterity deficits. One study identified that children with FASD had corpus callosum damage, which may have resulted in difficulties with complex VMI tasks which required interhemispheric interaction.[64] The parietal lobe is involved with sensorimotor integration and visual perception, and damage to these regions may also contribute to VMI deficits observed in children with FASD.[46] Furthermore, neural myelination can also be damaged by PAE,[45] which may explain why more complex FM skills, which require input from multiple areas as well as effective connectivity between regions, are more likely to be affected after PAE.

Limitations

This review had several limitations. Studies had significant methodological differences regarding PAE levels and FASD diagnoses, which hindered the comparability of FM outcomes. Studies were included only if they used standardized assessments commonly available to clinicians. Consequently, novel assessments of FM skills, such as computer- and robotic-based studies, were excluded, as were observational assessments, such as finger-nose touching, tremors, and finger localization. Some studies assessed clock and person drawing, and although these tasks require FM skills, they were excluded because outcomes were reported as visual-perceptual and visual-spatial skills.[38,46] The comparability of outcomes between studies was problematic because of the varying requirements of FM skills and assessment tools used as outcome measures, and also the use of different sets of normative data, comparison groups, and outcome criteria. For example, some studies used unmatched or matched control groups as a comparison, whereas others compared outcomes to varying sets of normative data. Studies that used pegboard tests variously reported outcomes based on dominance, laterality, or a combined score. More than half of the studies included children older or younger than the target age range of 4 to 12 years. This reduced certainty that the conclusions are specific to primary school-aged children, although FM impairments were still evident in studies that included younger children and adolescents. As with all systematic reviews, there is a risk of positive publication bias. This risk was minimized by including studies, which reported a range of neurodevelopmental outcomes, not just studies which reported FM skills, and by contacting experts in the field to identify unpublished studies.

Future Directions

Fetal alcohol spectrum disorders diagnostic guidelines should be updated to advice clinicians that a range of FM skills, particularly those which are more complex, may be affected by PAE and that these should be assessed separately to other motor skills using FM-specific assessment tools. Future studies should clarify what degree of FM impairment should contribute toward an FASD diagnosis. Establishing whether there is a relationship between specific FM skills and functional performance at school and home in children with PAE would be beneficial to those involved in FASD diagnosis, as well as clinicians, caregivers, and teachers of children with PAE.

CONCLUSION

This review identified that fine motor (FM) skills are impaired in children with moderate to high prenatal alcohol exposure (PAE) and children with a pFAS or FAS diagnosis. It remains uncertain if low PAE affects FM skills because few studies reported FM skills in children with PAE, and those that did assessed different types of FM skills, and each used different assessment tools. Consistent with our hypothesis, complex FM skills, such as visual-motor integration (VMI), were more likely to be impaired after PAE than basic FM skills such as grip strength. Furthermore, specific FM assessment tools, such as the Beery VMI, were more likely to detect impairments than generalized neurodevelopmental assessments. However, pegboard tests, although they are a specific FM assessment tool, and manual dexterity is a relatively complex FM skill, did not consistently detect manual dexterity impairments. It is uncertain whether pegboard tests are too basic to detect manual dexterity impairments resulting from PAE or if this type of skill is not affected by PAE. In all likelihood, it is probably a combination of both of these factors. However, the heterogeneity of the FM skills assessed and variability among assessment tools make it difficult to draw definite conclusions. Comprehensive assessment of FM skills in children with PAE or fetal alcohol spectrum disorder (FASD) is essential to contribute toward an accurate FASD diagnosis, and inform therapeutic interventions which support specific areas of FM difficulties and enhance individual strengths.

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C.2 Publication 2

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Fine motor skills in a population of children in remote Australia with high levels of prenatal alcohol exposure and Fetal Alcohol Spectrum Disorder

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Abstract

Background: Many children in the remote Fitzroy Valley region of Western Australia have prenatal alcohol exposure (PAE). Individuals with PAE can have neurodevelopmental impairments and be diagnosed with one of several types of Fetal Alcohol Spectrum Disorder (FASD). Fine motor skills can be impaired by PAE, but no studies have developed a comprehensive profile of fine motor skills in a population-based cohort of children with FASD. We aimed to develop a comprehensive profile of fine motor skills in a cohort of Western Australian children; determine whether these differed in children with PAE or FASD; and establish the prevalence of impairment.

Methods: Children ($n = 108$, 7 to 9 years) were participants in a population-prevalence study of FASD in Western Australia. Fine motor skills were assessed using the Bruininks-Oseretsky Test of Motor Proficiency, which provided a Fine Motor Composite score, and evaluated Fine Manual Control (Fine Motor Precision; Fine Motor Integration) and Manual Coordination (Manual Dexterity; Upper-Limb Coordination). Descriptive statistics were reported for the overall cohort; and comparisons made between children with and without PAE and/or FASD. The prevalence of severe (≤ 2nd percentile) and moderate (≤16th percentile) impairments was determined.

Results: Overall, Fine Motor Composite scores were 'average' ($M = 48.6 \pm 7.4$), as were Manual Coordination ($M = 55.7 \pm 7.9$) and Fine Manual Control scores ($M = 42.5 \pm 6.2$). Children with FASD had significantly lower Fine Motor Composite ($M = 45.2 \pm 7.7$ $p = 0.046$) and Manual Coordination scores ($M = 51.8 \pm 7.3$, $p = 0.027$) than children without PAE (Fine Motor Composite $M = 49.8 \pm 7.2$; Manual Coordination $M = 57.0 \pm 7.7$). Few children had severe impairment, but rates of moderate impairment were very high.

Conclusions: Different types of fine motor skills should be evaluated in children with PAE or FASD. The high prevalence of fine motor impairment in our cohort, even in children without PAE, highlights the need for therapeutic intervention for many children in remote communities.

Keywords: Fetal Alcohol Spectrum Disorder, Psychomotor performance, Motor skills, Indigenous population

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Background

Local Aboriginal leaders in the remote Fitzroy Valley region of Western Australia introduced alcohol restrictions in 2007 because they were concerned about the social and health effects of chronic alcohol misuse. These concerns included the potential harm caused by alcohol consumption during pregnancy, which can cause Fetal Alcohol Spectrum Disorder (FASD). In 2009 local leaders initiated 'The Lililwan Project' ('Lililwan' is Kimberley Kriol for 'all the little ones') to determine the prevalence of FASD [1]. Diagnoses on the FASD spectrum include Fetal Alcohol Syndrome (FAS) and partial Fetal Alcohol Syndrome (pFAS), both with characteristic facial anomalies and impaired growth; and Alcohol-Related Neurodevelopmental Disorder (ARND) or Neurodevelopmental Disorder – Prenatal/Alcohol Exposed (ND-PAE/ND-AE) with neurodevelopmental impairment in the absence of physical features [2, 3].

PAE can affect the development and function of the corpus callosum [4], cerebellum [5], basal ganglia [6], and motor cortex [7], and children with FASD may have skeletal malformations [8], abnormal muscle development [9], tremor [10], and impaired nerve conductivity [11]. All these factors may impair fine motor performance. Fine motor skills include basic skills such as grip strength, and more complex skills including visual (or fine) motor integration, manual dexterity, and upper-limb coordination. These skills underpin many self-care, academic, and recreational activities, including handwriting, dressing, and ball sports. Fine motor skills are particularly important in primary school aged children, who can spend more than half of their day completing tasks which require fine motor skills [12]. Handwriting quality can be affected by poor fine motor skills, and students with poor handwriting often receive poorer grades [13]. Teacher reports indicate that 20.6% of first year students at Fitzroy Fitzroy Crossing are below the Australian population 10th percentile for fine and gross motor skills [14]. Many Australian Aboriginal students perform below-average on the National Assessment Program – Literacy and Numeracy (NAPLAN), which is conducted annually with students in Years 3, 5, 7, and 9 [15].

Few studies of children with PAE or FASD have reported whether they have a motor impairment, and of those that do, many report a motor score that is a combination of fine motor and gross motor skills [16–18], or a score based on subtests of generalised developmental assessment tools [19], such as the Eye and Hand Coordination subscale from the Griffith's Mental Development Scales [20]. Individuals with FASD can have subtle neurological impairment, and researchers have highlighted the importance of assessing a range of specific areas of function rather than reporting amalgamated scores [18, 19]. Motor scores that are an average of fine and gross motor skills provide little insight into deficits, which is essential for understanding the child's neurological profile and developing appropriate therapy goals.

Several studies have assessed a range of fine motor skills in children with PAE or FASD [21–24], but each has used varying assessment tools and none report data from an entire population age-cohort. Motor skills in children with PAE or FASD are summarised in three systematic reviews. In one review, 'visual and motor' skills were not associated with mild, moderate, or binge PAE, however, none of the included studies assessed children older than 5 years [25]. Another review found an association between motor impairment and levels of PAE, but did not differentiate between fine and gross motor skills [26]. We reviewed fine motor skills in primary school aged children with PAE or FASD [27], and found that complex fine motor skills, such as visual-motor integration, were more likely to be impaired than basic skills, such as grip strength. We identified a range of assessment tools used to assess fine motor skills in children with PAE or FASD, but few that comprehensively assessed a range of different skills.

Study hypotheses

Fine motor proficiency and prevalence of impairment amongst children in the remote Fitzroy Valley, Western Australia were evaluated. We hypothesised that rates of fine motor impairment would be high due suspected high rates of neurodevelopmental and socioeconomic risk factors, including PAE. We also hypothesised that children with PAE, particularly those with FASD, would have the most impairment due to the teratogenic effect of alcohol on the central and peripheral nervous systems involved in performance of fine motor skills.

Study aims

1. Assess and evaluate fine manual control (fine motor precision and fine motor integration) and manual coordination (manual dexterity and upper-limb coordination) in a cohort of children in the Fitzroy Valley.
2. Compare fine motor skills of children (i) without PAE; (ii) with PAE but not FASD; and (iii) with FASD.
3. Determine the prevalence of moderate (≤ 16th percentile) and significant (≤ 2nd percentile) fine motor impairments in the cohort.

Methods

Setting

We evaluated fine motor data from the Lililwan Project, a population-based study of FASD prevalence in the Fitzroy Valley in the West Kimberley region of northern Western Australia. The Fitzroy Valley has a population of 4500 people living in communities across a 200 km radius, 80% of whom identify as being Australian Aboriginal [28].

Procedures

All children born in 2002 or 2003 and living in the Fitzroy Valley during 2010 and 2011 were eligible for inclusion. In Stage 1 of the study parents and carers of 127 children (95% participation) provided information about prenatal and childhood exposures, including PAE, antenatal drug exposures, nutrition, living conditions, and exposure to early life trauma [29]. The Alcohol Use Disorders Identification Test – Consumption (AUDIT-C) was used to classify PAE as 'low', 'risky', or 'high risk' [30].

In Stage 2, 108 of the children completed comprehensive neurodevelopmental assessments by qualified paediatricians and allied health practitioners. Attrition occurred because families moved out of the Fitzroy Valley ($n = 15$); we were unable to locate families or children ($n = 3$); or clinical assessment was declined ($n = 1$).

Assessors were blinded to alcohol and other pre and postnatal exposures. Adapted Canadian FASD Diagnostic Guidelines were used to assign FASD diagnoses, including FAS, pFAS, and ND-AE. To be diagnosed with one of the FASD diagnoses, a child was required to have 'significant' impairment (defined as ≥2 *SD* below the mean, or clinically significant variability between subtests on standardised assessments) in a minimum of 3 of 10 neurodevelopmental domains. The diagnoses of pFAS or FAS additionally required evidence of characteristic facial features or growth impairment. A study protocol detailing assessment tools and diagnostic criteria has been published [1]. Children were referred to local health services for medical or therapeutic treatment if required. Families whose child had a FASD diagnosis were referred to a Social Worker and an Indigenous Support Worker with extensive experience working with families affected by FASD. Fine motor skills were assessed in a one hour session by the primary author (RD), an Occupational Therapist with experience working with children in the Fitzroy Valley. Overall motor proficiency and gross motor skills were assessed by a Paediatric Physiotherapist (BRL), and have been reported [31, 32].

Instrumentation

The Bruininks-Oseretsky test of motor proficiency (second edition)

The Bruininks-Oseretsky Test of Motor Proficiency (BOT-2) is a standardised, norm-referenced tool suitable for motor assessment in children and young adults aged 4–21 years [33]. Complete (53 tasks) and short versions (14 tasks) are available. The complete version of the BOT-2 was chosen for use in our study because it evaluates a diverse range of fine motor skills; is frequently used in Australia [34] and international FASD diagnostic clinics [35]; and is recommended in the Canadian FASD Diagnostic Guidelines [3]. The BOT-2 provides a Fine Motor Composite score, which is an overall measure of fine motor proficiency. The Fine Motor Composite score is derived from the Fine Manual Control and Manual Coordination composite scores, which in turn are derived from Fine Motor Precision (which assesses precise hand and finger control through paper and pencil tasks, folding paper, and scissor skills), Fine Motor Integration (which assesses ability to reproduce a series of eight geometric shapes), Manual Dexterity (which assess reaching, grasping, and bimanual control through timed tasks such as stringing blocks and placing pegs in a pegboard), and Upper-Limb Coordination (which assesses coordinated arm and hand movement in terms of catching, throwing, and dribbling a tennis ball) subtest scores (Fig. 1). Composites are reported as standardised scores (mean (M) = 50.0, standard deviation (*SD*) = 10.0), and subtest scores are reported as scale scores (M = 15.0, *SD* = 5.0). Descriptive categories are defined as 'well-above average'(standard score ≥ 70; scale score ≥ 25; ≥ 98th percentile); 'above average' (standard score 60 to 69; scale score 20 to 24; 84th to 97th percentile); 'average'(standard score 41 to 59; scale score 11 to 19; 18th to 83rd percentile); 'below average'(standard score 31 to 40; scale score 6 to 10; 3rd to 17th percentile); and 'well-below average' (standard score ≤ 30; scale score ≤ 5; ≤ 2nd percentile) [33].

BOT-2 tasks are designed to be novel for all children, including those from diverse cultural backgrounds, regardless of familiarity with the tasks, and the composites and subtests have well-established internal consistency and test-retest reliability [33]. The BOT-2 Short Form was trialled in a subset of children from the Lililwan project and we found it to have excellent inter-rater reliability (0.88 to 0.92) and fair to good test-retest reliability (0.62 to 0.73) in this population [35]. The BOT-2 is endorsed as a suitable measure of motor skills in FASD diagnostic assessment [3].

Statistical analysis

Data were scored using the sex-specific norms of the BOT-2 ASSIST scoring software. The Fine Motor Composite score was calculated using the online Q-global™ scoring system. Means and standard deviations were obtained for all BOT-2 fine motor composite standardised scores and subtest scale scores. Fine motor scores were assessed for normality and analysed using a one-way between groups analysis of variance (ANOVA). Children with unconfirmed or unknown PAE ($n = 5$) were excluded from the between-groups analysis. Group differences were analysed using ANOVA between children without PAE ('No PAE' group); children with PAE who did not have multiple, significant neurodevelopmental impairments and were therefore not diagnosed with a type of FASD ('PAE (no FASD)' group); and children with confirmed PAE plus FASD ('FASD' group). Significance was set at $p < 0.05$. Effect sizes (eta^2) were calculated, with 0.01 being deemed a small effect size; 0.06 a medium effect size; and 0.14 a large effect size

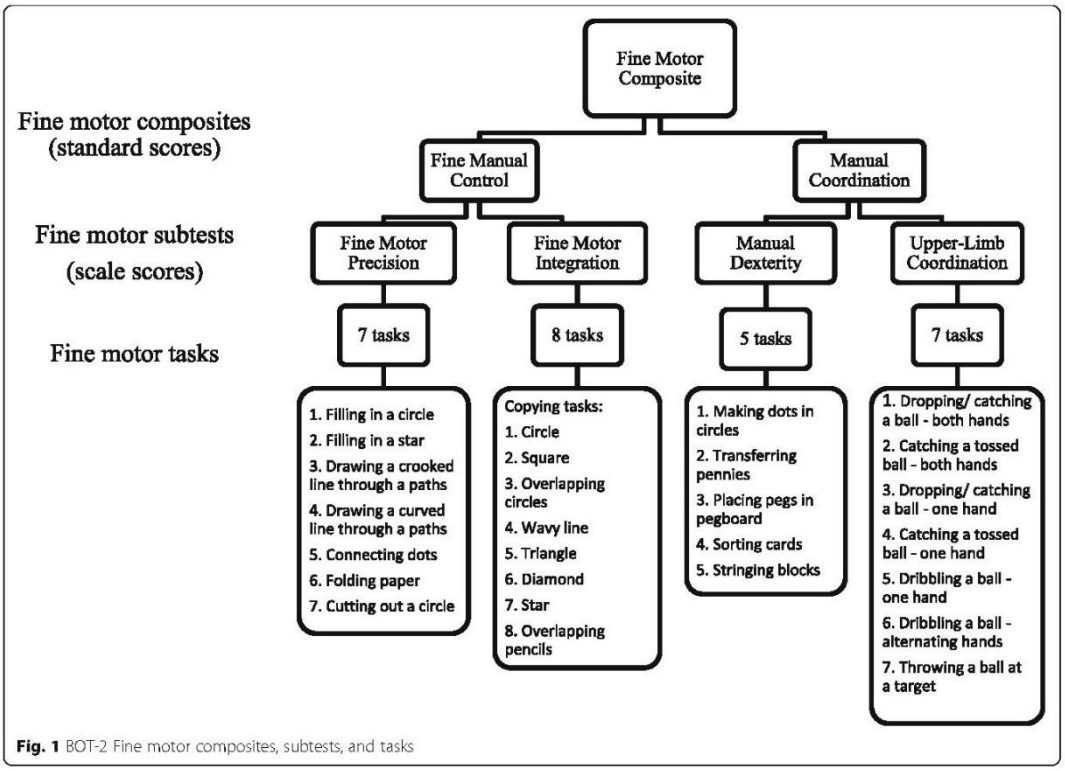

Fig. 1 BOT-2 Fine motor composites, subtests, and tasks

[36]. Tukey's Honestly Significant Difference (HSD) test was utilised as a post-hoc test to determine which groups differed. Prevalence of severe (≥ 2 *SD* below the mean; ≤ 2nd percentile) and moderate (≥ 1 *SD* below the mean; ≤ 16th percentile) impairment was reported for each fine motor composite and subtest for the cohort, and also by exposure group. Statistical analysis was completed using IBM SPSS Statistics for Windows, version 21.0 (Armonk, NY: IBM Corp.).

Results

Participants

Participants were aged between 7.5 to 9.6 years (M = 8.7 years) at assessment. The majority were of Australian Aboriginal descent (Table 1). Of the children with PAE (n = 60, 55.6%), most (95%) were exposed to 'risky' or 'high risk' levels according to AUDIT-C criteria [37]. Children who participated in Stage 1 only (n = 15) were slightly less likely to have PAE (36.8%) than children who participated in both Stage 1 and 2 (55.6%) but were otherwise similar. Children with and without PAE were born at similar weeks of gestation, and the incidence of pre-term births were also similar [37]. The Universal Non-Verbal Intelligence Test [38] formed part of the assessment battery during the Lililwan Project and was used to evaluate cognitive abilities. Full-scale standard scores were similar between groups with and without PAE or FASD (No PAE M = 89.9, *SD* = 8.5; PAE, no FASD M = 89.4, *SD* = 9.1; FASD M = 85.0, *SD* = 12.3; p = 0.329).

Many children lived in overcrowded households (M = 6.1, range 2–16), and many had lived in more than four homes since birth (n = 17, 15.8%). Most children (n = 89, 82.4%) attended school 4 to 5 days a week, with only one child (who did not have FASD) not attending school at all. Approximately half (53.3%) of the children's biological mothers had studied beyond secondary education. These socioeconomic factors were similar between children with and without FASD [39].

Fine motor composites and subtests

For the total cohort, all fine motor composite and subtest scores were in the 'average' range (Table 2). Children with FASD had significantly lower Fine Motor Composite scores and Manual Coordination scores than children without PAE (Fine Motor Composite eta^2 = 0.06, Tukey's HSD p = 0.038; Manual Coordination eta^2 = 0.07, Tukey's HSD p = 0.024) (Table 2). There were no other significant differences between groups, but the mean scores of the

Table 1 Cohort characteristics

	Total Cohort[a] N = 108	No PAE n = 43	PAE (no FASD) n = 39	FASD n = 21
	n (%)	n (%)	n (%)	n (%)
Australian Aboriginal	106 (98.1)			
Gender				
Male	57 (52.8)	24 (55.8)	18 (46.2)	13 (61.9)
Handedness				
Right	101 (93.5)	41 (95.3)	38 (97.4)	19 (90.5)
Hearing[b,c] (n = 93)				
Normal	42 (45.2)	16 (37.2)	14 (35.9)	10 (47.6)
Mild loss	38 (40.9)	15 (34.9)	13 (33.3)	7 (33.3)
Moderate loss	13 (14.0)	7 (16.3)	3 (7.7)	3 (14.3)
Missing	15 (13.9)	5 (11.6)	9 (23.1)	1 (4.8)
Prenatal nicotine exposure[d]				
Yes	67 (62.0)	18 (41.9)	32 (82.1)	15 (71.4)
Unknown	7 (6.5)	0 (0)	1 (2.6)	3 (14.3)
Prenatal marijuana exposure[d]				
Yes	13 (12.0)	2 (4.7)	10 (25.6)	1 (4.8)
Unknown	7 (6.5)	0 (0)	1 (2.6)	2 (9.5)
PAE risk levels[e]				
No exposure	43 (100.0)	0 (0)	0 (0)	0 (0)
Low (1–3)	4 (3.7)	0 (0)	4 (10.3)	0 (0)
Risky (4–5)	4 (3.7)	0 (0)	3 (7.7)	1 (4.8)
High risk (≥ 6)	46 (42.6)	0 (0)	29 (74.4)	17 (81.0)
PAE, uncertain risk	6 (5.6)	0 (0)	3 (7.7)	3 (14.3)
Unknown PAE	5 (4.6)	0 (0)	0 (0)	0 (0)

[a] 'Total cohort' includes n = 5 children with unknown PAE who are not included in the No PAE, PAE (no FASD), or FASD groups
[b] Not all children completed audiology testing
[c] Mild hearing loss 26 – 40 dB; moderate hearing loss 41 – 55 dB
[d] Some prenatal exposure information not available, either due to the primary carer not knowing, or the birth mother choosing not to disclose this information
[e] Risk level according to AUDIT-C scoring criteria

PAE (no FASD) and FASD groups were consistently lower than in children without PAE in almost all composites and subtests (aside from the Upper-Limb Coordination subtest), and the scores of children with FASD were lower again (Fig. 2).

Prevalence of fine motor impairment

Prevalence of severe impairment (range 0 to 0.9%) was low in all composites and subtests (Table 3). Prevalence of moderate impairment for the Fine Motor Composite (14.8%) was derived from a high prevalence of moderate impairment in the Fine Manual Control composite (38.9%), and low prevalence in the Manual Coordination composite (1.9%) (Table 3). Only one child with PAE (who had FASD) had severe impairment in any fine motor composite or subtest (Table 3). Prevalence of moderate impairment in the Fine Motor Composite was slightly lower than BOT-2 norms for children without PAE (11.6%) and PAE (no FASD) (7.7%), but much higher in children with FASD (28.6%). Moderate impairment was very high in the Fine Manual Control composite (and its associated subtests) for all exposure groups, but highest in children with FASD (47.6%). Moderate impairment was less than expected in the Manual Coordination composite for all exposure groups (range 0–4.8%), but this composite was an amalgamation of the Manual Dexterity subtest, which had high rates of moderate impairment, particularly for children with FASD (23.8%), and the Upper-Limb Coordination subtest, in which few children had moderate impairment (range 4.7 to 5.1%).

Discussion

This is the first study to comprehensively assess fine motor skills in a population-based cohort of predominantly Aboriginal children in Australia. Many children in our study had high levels of PAE and were diagnosed

Table 2 BOT-2 Fine motor composite standardised scores and subtest scale scores in children with no PAE; PAE (no FASD); and FASD

	Total Cohort $n = 108$[a]		No PAE $n = 43$		PAE (no FASD) $n = 39$		FASD $n = 21$		ANOVA		
	M (*SD*)	95% CI	M (*SD*)	95% CI	M (*SD*)	95% CI	M (*SD*)	95% CI	df	F	*p*
FINE MOTOR COMPOSITE	48.6 (7.4)	47.2–50.0	49.8 (7.2)	47.6–52.0	48.8 (6.2)	46.8–50.8	45.2 (7.7)	41.7–48.7	2100	3.17	0.046*[d]
Fine Manual Control[b]	42.5 (6.2)	41.3–43.6	43.4 (6.2)	41.4–45.3	41.9 (5.3)	40.2–43.6	41.1 (7.3)	37.8–44.5	2100	1.10	0.336
Fine Motor Precision[c]	12.3 (3.3)	11.7–12.9	12.7 (3.4)	11.7–13.8	11.9 (2.6)	11.0–12.7	11.8 (4.0)	10.0–13.6	2100	0.94	0.393
Fine Motor Integration[c]	11.0 (2.9)	10.5–11.6	11.3 (2.7)	10.4–12.1	11.2 (2.9)	10.3–12.2	10.1 (3.0)	8.8–11.5	2100	1.29	0.279
Manual Coordination[b]	55.7 (7.9)	54.2–57.2	57.0 (7.7)	54.6–59.4	56.2 (7.0)	53.9–58.5	51.8 (7.3)	48.4–55.1	2100	3.74	0.027*[d]
Manual Dexterity[c]	14.9 (3.7)	14.2–15.6	15.4 (3.5)	14.3–16.4	15.1 (3.1)	14.1–16.1	13.2 (4.0)	11.4–15.0	2100	2.97	0.056
Upper-Limb Coordination[c]	19.6 (4.4)	18.7–20.4	19.8 (4.4)	18.5–21.2	20.0 (4.5)	18.5–21.5	18.0 (3.8)	16.3–19.7	2100	1.64	0.200

* $p < 0.05$
[a] 'Total Cohort' includes $n = 5$ children with unknown PAE who are not included in the No PAE, PAE (no FASD), or FASD groups
[b] BOT-2 norms M = 50, *SD* = 10
[c] BOT-2 norms M = 15, *SD* = 5. Lower scores represent poorer performance in composites and subtests
[d] Tukey's HSD: No PAE > FASD

with FASD. The cohort's mean BOT-2 Fine Motor Composite scores were in the 'average' range, an unexpected finding given the high levels of PAE and other neurodevelopmental risk factors in our cohort. However, in keeping with our hypothesis, children with FASD had poorer fine motor skills than children without PAE. Manual coordination skills, including fine motor speed, manual precision, and coordinated arm and hand movement were specific areas of difficulty for children with FASD. Few children had severe impairment (below the 2nd percentile), but rates of moderate impairment (below the 16th percentile) were very high.

Other studies of fine motor impairment in children with PAE or FASD have also reported a mixed profile of strengths and difficulties. A range of assessment tools have been used to evaluate fine motor skills in children with PAE or FASD, including the Visuomotor Precision subtest from the Developmental Neuropsychological

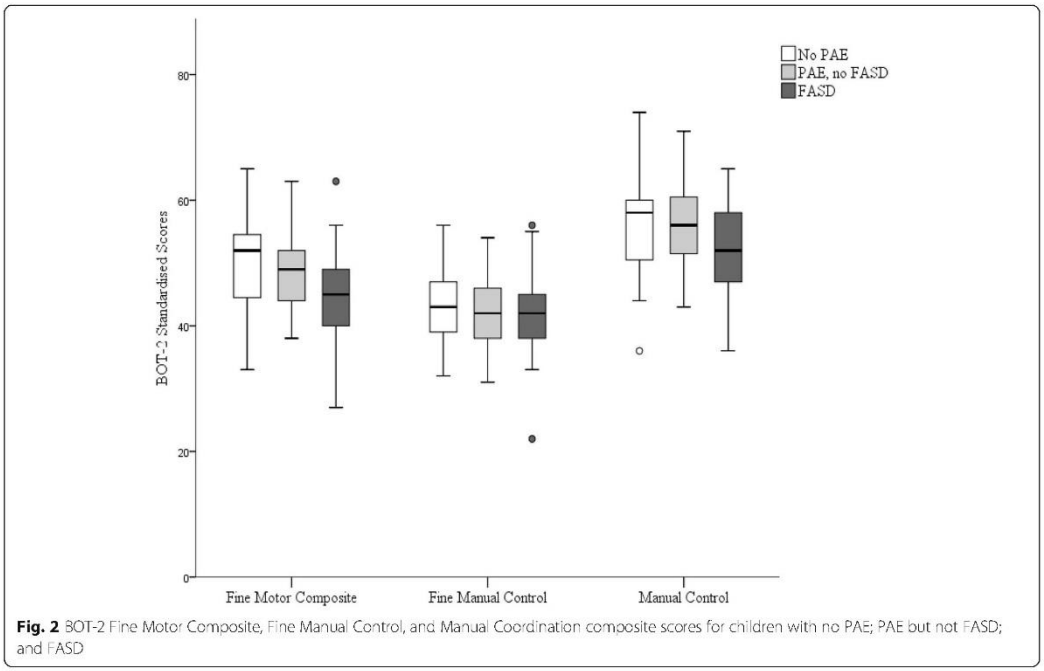

Fig. 2 BOT-2 Fine Motor Composite, Fine Manual Control, and Manual Coordination composite scores for children with no PAE; PAE but not FASD; and FASD

Table 3 Prevalence of severe (≥ −2*SD*) and moderate (≥ −1*SD*) fine motor impairment in children with no PAE; PAE (no FASD); and FASD

	Total Cohort $n = 108^a$	No PAE $n = 43$	PAE (no FASD) $n = 39$	FASD $n = 21$
	n (%)	*n* (%)	*n* (%)	*n* (%)
Fine Motor Composite				
- ≥ 2*SD*	1 (0.9)	0 (0)	0 (0	1 (4.8)
- ≥ 1*SD*	16 (14.8)	5 (11.6)	3 (7.7)	6 (28.6)
Fine Manual Control				
-≥ 2*SD*	1 (0.9)	0 (0)	0 (0)	1 (4.8)
-≥ 1*SD*	42 (38.9)*	16 (37.2)*	14 (35.9)*	10 (47.6)**
Fine Motor Precision				
- ≥ 2*SD*	1 (0.9)	0 (0)	0 (0)	1 (4.8)
- ≥ 1*SD*	33 (30.6)	12 (27.9)	11 (28.2)	9 (42.9)*
Fine Motor Integration				
- ≥ 2*SD*	1 (0.9)	0 (0)	0 (0)	1 (4.8)
- ≥ 1*SD*	48 (44.4)*	17 (39.5)*	15 (38.5)*	13 (61.9)**
Manual Coordination				
- ≥ 2*SD*	0 (0)	0 (0)	0 (0)	0 (0)
- ≥ 1*SD*	2 (1.9)	1 (2.3)	0 (0)	1 (4.8)
Manual Dexterity				
- ≥ 2*SD*	1 (0.9)	0 (0)	0 (0)	1 (4.8)
- ≥ 1*SD*	11 (10.2)	5 (11.6)	0 (0)	5 (23.8)
Upper-Limb Coordination				
- ≥ 2*SD*	0 (0)	0 (0)	0 (0)	0 (0)
- ≥ 1*SD*	5 (4.6)	2 (4.7)	2 (5.1)	1 (4.8)

- ≥ 2*SD* = ≤ 2nd percentile; − ≥ 1*SD* = ≤ 16th percentile
* = at least twice, and ** = at least three times, the rate of BOT-2 norms
[a] 'Total Cohort' includes *n* = 5 children with unknown PAE who are not included in the No PAE, PAE (no FASD), or FASD group

Evaluation (NEPSY) [40], the Movement Assessment Battery for Children (M-ABC) [41], and The Beery Buktenica Developmental Test of Visual-Motor Integration (Beery VMI) [42]. Other studies have reported mixed findings for fine motor precision [24, 43] and manual dexterity [44, 45] skills, which weren't impaired in children with PAE or FASD in our study. Ball skills were also not impaired, which is consistent with other reported findings [44–46]. We found that visual-motor integration (termed 'fine motor integration' in the BOT-2) wasn't impaired, but this contradicts other studies which commonly report visual-motor integration impairment in children with FASD [47–49]. This may be due to the limited number of tasks used to evaluate this skill in the BOT-2 ($n = 8$), compared to the more commonly used Beery VMI ($n = 30$). The Beery VMI formed part of the neurodevelopmental assessment battery in the Lililwan Project, and we reported that the Fine Motor Coordination subtest of the Beery VMI was significantly lower in children with FASD [50].

Only one other study group [17] has published motor outcomes in children with FASD using the BOT. These authors used an earlier version of the BOT (1st edition), which does not include a Fine Motor Composite score. The authors reported that the motor score (an amalgamation of fine and gross motor skills) was not significantly different in children with FASD (M = 49.1) compared to 'typically developing' (M = 57.7, $p = 0.36$) children. These non-significant findings may result from areas of stronger skills masking fine motor impairments, in much the same way that children in our cohort with FASD had an 'average' Fine Motor Composite score (M = 45.2), which was derived from relatively stronger Manual Coordination (M = 51.8) and weaker Fine Manual Control scores (M = 41.1).

Implications of prevalence rates

The very low prevalence of severe fine motor impairment in our cohort has implications for FASD diagnosis. The University of Washington 4-digit Diagnostic Code [51] and the Canadian FASD Diagnostic Guidelines [3] each advise that scores 2 *SD* below the mean (≤ 2nd percentile) indicate impairment when diagnosing FASD. In contrast, 1 *SD* below the mean (≤ 16th percentile) indicates impairment according to the Centers for Disease Control (CDC) [2]. Other authors have also proposed a 1 *SD* cut-off for identifying impairment for ND-PAE [52]. Only one child in our cohort (who had FASD) had fine motor scores below the 2nd percentile, which seems conservative given the high levels of PAE and other neurodevelopmental risk factors in our cohort. This issue warrants further consideration and investigation.

Strengths

This study is the first comprehensive, population-based study of fine motor skills in Aboriginal children in Australia. It is also the first to use a standardised fine motor assessment to develop a comprehensive profile of fine motor skills in children with PAE and/or FASD.

Limitations

Most children in our study identified as Australian Aboriginal and all were living in remote communities, and so the results should not be generalised. Nevertheless, outcomes may be relevant to other populations with similar demographics. Although the study involved almost two entire age cohorts and had a high participation rate (%), the sample size was too small to statistically control for potentially confounding factors. However, many risk factors, such as early life trauma and low socioeconomic status, were common to almost all children in our study. Many children without PAE also had a moderate level of fine motor impairment, and thus impairments cannot be solely attributed to PAE. However, the high proportion of children in our cohort with "risky" or "high risk" levels of

PAE make it likely that PAE contributed, at least in part, to the identified fine motor impairment.

Recommendations and future directions

This study highlights the importance of comprehensively assessing a range of fine motor skills in children with PAE or suspected FASD. Other researchers have expressed concerns that composite scores may not be sensitive enough to detect subtle neurological impairment in children with FASD [18, 19]. Our findings support these concerns. We recommend that a range of fine motor skills be assessed in children with PAE, and outcomes not be amalgamated with other fine or gross motor scores, because an averaged 'motor' score could mask specific difficulties, resulting in inaccurate diagnoses and missed opportunities for therapeutic support.

Conclusions

Children in our cohort had Fine Motor Composite scores in the 'average' range. Upper-limb coordination (ball skills) was a strength, while fine motor integration skills (copying complex shapes) were an area of weakness. Children with FASD had significantly lower Fine Motor Composite and Manual Coordination scores than children without PAE. These outcomes highlight the importance of reporting specific types of fine motor skills, rather than an amalgamated 'motor' or even 'fine motor' score. The very high levels of impaired fine motor precision and fine motor integration skills highlight the need for therapeutic intervention for many children in the Fitzroy Valley, regardless of PAE, to encourage successful participation in self-care, academic, and recreational activities.

Abbreviations

ARND: Alcohol-Related Neurodevelopmental Disorder; AUDIT-C: Alcohol Use Disorders Identification Test – Consumption; BOT-2: Bruininks-Oseretsky Test of Motor Proficiency; FAS: Fetal Alcohol Syndrome; FASD: Fetal Alcohol Spectrum Disorder; HSD: Tukey's Honestly Significant Difference test; NAPLAN: National Assessment Program – Literacy and Numeracy; ND-AE: Neurodevelopmental Disorder – Alcohol Exposed; ND-PAE: Neurodevelopmental Disorder – Prenatal Alcohol Exposed; PAE: Prenatal alcohol exposure; SD: Standard deviation

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Availability of data and materials

Data from the Lililwan Project is stored at The University of Sydney, Sydney, Australia. It is not publicly available as it contains sensitive information related to individual participants.

Authors' contributions

RD conceptualised and designed the fine motor assessments for the Lililwan study; applied for ethics approval relevant to the fine motor aspects of the Lililwan Project; completed the BOT-2 fine motor assessments; analysed and interpreted the BOT-2 fine motor data; and drafted, revised, and finalised the manuscript. BRL conceptualised, designed and completed the gross motor assessments for the Lililwan study, including the Upper-Limb Coordination BOT-2 subtest; assisted with analysing BOT-2 fine motor data; and assisted with drafting and finalising the manuscript. REW and TWT performed the statistical analysis and interpreted the data; and assisted with drafting and finalising the manuscript. KS and PH assisted with conceptualisation and design of the fine motor aspects of the study; assisted with interpretation of data; and assisted with drafting and finalising the manuscript. JL, JPF, JO, MC, and EJE conceptualised and designed the Lililwan study; assisted with interpretation of data; and assisted with drafting and finalising the manuscript. All authors have approved of the final version of the manuscript for publication and have agreed to be accountable for all aspects of the work.

Authors' information

Robyn Doney is an Occupational Therapist and PhD student. She has extensive clinical experience working with children in the Kimberley, including the Fitzroy Valley.

Ethics approval and consent to participate

The Lililwan Project was conceived, designed, and approved by local leaders in the Fitzroy Valley, who also consented to publication of results. Families were provided with verbal and written information about the study in English and their local language if preferred. Parents or guardians provided signed consent, and families or children could withdraw from the study at any stage without consequences. Ethics approval was provided by the Curtin University Human Research Ethics Committee; Kimberley Aboriginal Health Planning Forum Research Sub-committee; University of Sydney Human Research Ethics Committee; Western Australian Aboriginal Health and Information Ethics Committee; and the Western Australian Country Health Services Board Research Ethics Committee.

Consent for publication

Not applicable.

Competing interests

The authors declare that they have no competing interests.

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Visual-motor integration, visual perception, and fine motor coordination in a population of children with high levels of Fetal Alcohol Spectrum Disorder

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ABSTRACT

Background: Visual-motor integration (VMI) skills are essential for successful academic performance, but to date no studies have assessed these skills in a population-based cohort of Australian Aboriginal children who, like many children in other remote, disadvantaged communities, consistently underperform academically. Furthermore, many children in remote areas of Australia have prenatal alcohol exposure (PAE) and Fetal Alcohol Spectrum Disorder (FASD), which are often associated with VMI deficits.

Methods: VMI, visual perception, and fine motor coordination were assessed using The Beery-Buktenica Developmental Test of Visual-Motor Integration, including its associated subtests of Visual Perception and Fine Motor Coordination, in a cohort of predominantly Australian Aboriginal children (7.5–9.6 years, $n = 108$) in remote Western Australia to explore whether PAE adversely affected test performance. Cohort results were reported, and comparisons made between children i) without PAE; ii) with PAE (no FASD); and iii) FASD. The prevalence of moderate (≤16th percentile) and severe (≤2nd percentile) impairment was established.

Results: Mean VMI scores were 'below average' ($M = 87.8 \pm 9.6$), and visual perception scores were 'average' ($M = 97.6 \pm 12.5$), with no differences between groups. Few children had severe VMI impairment (1.9%), but moderate impairment rates were high (47.2%). Children with FASD had significantly lower fine motor coordination scores and higher moderate impairment rates ($M = 87.9 \pm 12.5$; 66.7%) than children without PAE ($M = 95.1 \pm 10.7$; 23.3%) and PAE (no FASD) ($M = 96.1 \pm 10.9$; 15.4%).

Conclusions: Aboriginal children living in remote Western Australia have poor VMI skills regardless of PAE or FASD. Children with FASD additionally had fine motor coordination problems. VMI and fine motor coordination should be assessed in children with PAE, and included in FASD diagnostic assessments.

What this paper adds

This study is the first to report VMI, visual perception, and fine motor coordination skills in a population-based cohort of Aboriginal children in a remote region of Australia. It is also the first to consider whether visual perception or fine motor coordination impairments are present, which could account for VMI difficulties in children with PAE or FASD. The outcomes identified that many children in remote Australia have VMI impairment, even those without PAE or FASD, suggesting that PAE is just one of many neurodevelopmental risk factors which may cause VMI impairment. Children with FASD had significantly lower fine motor coordination scores, and higher rates of moderate impairment. It seems that, for children with FASD, they either have a fine motor coordination impairment which exists independently of the VMI impairment, or alternatively, that their problems contribute, in part, to observed VMI difficulties. However, given the poor VMI score of children without PAE, it is likely that other factors also contributed to VMI difficulties in children with FASD in our cohort. The study shows that many children in the region require therapeutic support for VMI impairment, and it is important to assess both VMI and fine motor coordination in children with PAE and/or FASD.

1. Introduction

Alcohol consumption during pregnancy may result in a range of irreversible, clinically distinguishable, lifelong conditions, collectively called Fetal Alcohol Spectrum Disorder (FASD) (Centers for Disease Control and Prevention, 2005). FASD is an umbrella term which includes the diagnoses of Fetal Alcohol Syndrome (FAS); partial FAS (pFAS); and Alcohol Related Neurodevelopmental Disorder (ARND), also known as Neurodevelopmental Disorder – Prenatal/Alcohol Exposed (ND-PAE/ND-AE) (Astley & Clarren, 2000; Chudley et al., 2005). Individuals diagnosed with FAS or pFAS have characteristic dysmorphic facial features and/or growth impairment, while those with ARND/ND-AE diagnoses do not necessarily have these impairments. However, individuals with any of the FASD diagnoses have significant neurological damage which can cause mild to severe deficits in cognition, executive function, memory, language, attention, social and adaptive function, and soft neurological signs, including fine and gross motor skills and visual-motor integration (Chudley et al., 2005). These deficits can lead to social and adaptive problems at home, school or work, and in society (Streissguth et al., 2004).

Visual-motor integration (VMI) is the ability to use input from the visual perceptive system (which includes visual acuity, accommodation, binocular fusion, stereopsis, and convergence/divergence) to coordinate fine motor skills (which require the use of the smaller muscles of the wrist and hand, including dexterity, precision, coordination, and manual control) (Schneck, 2010). VMI underpins many everyday functions such as handwriting and drawing, catching a ball, dressing, eating, and driving (Tomchek & Schneck, 2006). There are two types of VMI: i) constructional VMI, which includes tasks such as using blocks to build a 3D shape, and ii) graphomotor VMI, which includes paper and pencil tasks such as drawing a series of lines to form geometric shapes. Different neural processes underpin constructional and graphomotor VMI, so they should be considered separate skill sets (Benton & Tranel, 1993). Impairment of graphomotor VMI can be due to underlying problems with visual perception or fine motor skills, but impairment can also exist when visual perception and fine motor skills are intact, meaning the problem lies with the integration of these skills (Beery & Beery, 2010; Kulp, 1999; Milner, 2006).

In Australia, students in Years 3, 5, 7, and 9 annually complete the National Assessment Program – Literacy and Numeracy (NAPLAN) assessment, which assesses reading, writing, language, and numeracy skills. Children in very remote areas of Western Australia, including the Fitzroy Valley, consistently under-perform compared to national averages (Australian Curriculum, Assessment and Reporting Authority, 2015). Although the NAPLAN does not specifically assess VMI or handwriting skills, other studies have shown that VMI impairment is associated with poor handwriting performance (Kulp, 1999; Weil & Cunningham Amundson, 1994), and students with poor handwriting skills often receive lower grades on written assessments despite adequate content (Chase, 1986). Therapeutic interventions which aim to improve handwriting, especially those which are integrated into the classroom, have successfully improved VMI and handwriting skills (Case-Smith, 2002).

Neurological damage resulting from prenatal alcohol exposure (PAE) may affect brain regions involved in VMI. Neuroimaging studies have shown that the corpus callosum, basal ganglia, cerebral cortex, and cerebellum may all be damaged by PAE (Riley, Infante, & Warren, 2011; Sowell et al., 2002). Within the cerebral cortex, the parietal and temporal lobes are particularly affected by PAE (Archibald et al., 2001; Sowell et al., 2002). Children with FASD with reduced parietal lobe white matter have been shown to have reduced VMI abilities (Sowell et al., 2008). Optic nerve hypoplasia (Stromland, 2004) has been reported following PAE, which may also impair visual perception, as have skeletal malformations (Jones et al., 2010),

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atypical muscle development (David & Subramaniam, 2005), tremor (Marcus, 1987), and impaired nerve conductivity (de los Angeles Avaria et al., 2004) which may affect fine motor skills.

In a recent systematic review of fine motor skills in children with PAE or FASD we concluded that complex skills, including VMI, were more likely to be impaired than basic skills such as grip strength (Doney et al., 2014). VMI was the most commonly assessed skill and the Beery-Buktenica Developmental Test of Visual-Motor Integration (Beery VMI) (Beery & Beery, 2010) was the most commonly used assessment tool. The Beery VMI is commonly used by Occupational Therapists, Psychologists, and other health and educational professionals because it reports outcomes as standard scores and percentile ranks of VMI abilities; has well-established reliability and validity; is relatively quick to administer; can be used across a wide range of ages; and is suitable for use with people from diverse educational and linguistic backgrounds (Beery & Beery, 2010). VMI skills can be predictive of handwriting performance (Daly, Kelley, & Krauss, 2003), which may be adversely affected in children with FASD (Duval-White, Jirikowic, Rios, Deitz, & Olson, 2013). The Beery VMI includes two optional subtests of Visual Perception and Fine Motor Coordination which can be used to determine whether VMI deficits are due to underlying problems with these skills. In all but one study in which the Beery VMI was used, significant differences were reported between children with and without PAE or FASD, but no studies reported outcomes from the Visual Perception or Fine Motor Coordination subtests.

To date, no studies have reported graphomotor VMI skills in a population-based cohort of Australian Aboriginal children with high levels of PAE, and so the prevalence of impairment in these populations is unknown. The only other population-based study of VMI skills in children with PAE or FASD found that first-grade children in a Midwestern US city with FASD ($n=36$) had lower Beery VMI percentile rank scores than the control group ($n=98$) (May et al., 2014). Only one study has examined whether children with FASD make different types of errors than children without PAE when copying shapes (Uecker & Nadel, 1996), and this information can provide insight into specific areas of difficulty, and thus guide therapeutic interventions (Beery & Beery, 2010). The Beery VMI subtests examine whether impairment in VMI could be due to underlying problems with visual perception and/or fine motor coordination (Beery & Beery, 2010), but to date, no studies have reported data from the Visual Perception or Fine Motor Coordination subtests in relation to children with PAE or FASD.

Conflicting evidence exists for impairment of visual perception (Aronson, Kyllerman, Sabel, Sandin, & Olegard, 1985; Janzen, Nanson, & Block, 1995; Uecker & Nadel, 1996) or fine motor coordination skills (Doney et al., 2014) in children with PAE or FASD, which may be due to the wide range of assessment tools used in different studies. The advantage of using the Beery VMI subtests to assess visual perception and fine motor coordination is that they use the same shapes as the core component of the Beery VMI, which the authors claim 'makes comparisons between performances on all three tests as valid as possible' (Beery & Beery, 2010; p.99).

Understanding whether impairment in VMI could be due to underlying visual perception or fine motor deficits, or whether these skills are intact and the problem lies with their integration, will assist in developing a more accurate neurological profile for children with PAE and FASD, and inform therapeutic interventions which target specific deficits. Additionally, the subtests are timed, and this information may be useful to determine if sustained visual attention, processing speed, or slower motor response time, which have previously been reported as impaired in children with FASD (Mattson, Calarco, & Lang, 2006; Simmons, Wass, Thomas, & Riley, 2002) may affect VMI performance.

The aims of our study were to:

1. Compare VMI, visual perception, and fine motor coordination, including types of errors and time to complete subtests, in 7–9 year old children in the Fitzroy Valley, i) without PAE; ii) with PAE who did not meet criteria for one of the FASD diagnoses; and iii) with FASD
2. Investigate whether VMI deficits, if any, could be due to visual perception or fine motor coordination impairment
3. Determine the prevalence of VMI, visual perception, and fine motor coordination impairments in children without PAE; with PAE but no FASD diagnosis; and with FASD

Based on the literature, we hypothesized that children with FASD would have significantly lower VMI, visual perception, and fine motor coordination scores; would take longer to complete subtests; and would make different types of errors than children without PAE. We also anticipated that children with FASD would have a significantly higher prevalence of impairment than children without PAE.

2. Materials and methods

2.1. Setting

In this paper we report VMI data from the Lililwan Project, a population-based study of FASD prevalence in the remote Fitzroy Valley region of Western Australia. The Fitzroy Valley has a population of 4500 people living in approximately 45 communities within a 200 km radius of the main service town of Fitzroy Crossing (Morphy, 2010). Approximately 80% of the population identify as being of Australian Aboriginal descent, representing five Aboriginal cultural groups. As previously published, Stage 1 of the Lililwan Project (2010) involved interviews with parents or caregivers of children born in 2002 or 2003 ($n=127$; 95% participation) regarding antenatal risk factors and childhood exposures which may affect development, including exposure to alcohol, nicotine, and marijuana *in-utero*, socioeconomic factors such as employment and household

structure, and environmental exposures such as nutrition and early life trauma (Fitzpatrick et al., 2013). PAE was based on retrospective self-report, in most cases by the birth mother, but in some instances by family members who were living with the birth mother during her pregnancy. Where possible, information was corroborated by review of maternal medical records. PAE data were deemed to be 'very reliable' in most instances (Fitzpatrick et al., 2015). PAE levels were classified as 'no exposure', 'low risk', 'risky', 'high risk', or 'uncertain risk' (when PAE was confirmed, but levels were unknown) according to the frequency, pattern, and amount of alcohol consumption as defined within the Alcohol Use Disorders Identification Test – Consumption (AUDIT-C), which is a standardized and validated measure of PAE (Bush, Kivlahan, McDonell, Fihn, & Bradley, 1998; Fitzpatrick et al., 2015). Stage 2 of the Lililwan Project (2011) involved a comprehensive neurodevelopmental assessment, including of VMI skills, for FASD by a multidisciplinary team including a Pediatrician, Physiotherapist, Psychologist, Occupational Therapist, and Speech Pathologist (n = 108; 81% participation) (Fitzpatrick et al., 2012). FASD diagnoses were assigned according to modified Canadian FASD diagnostic guidelines (Chudley et al., 2005), which require severe impairment in a minimum of three (out of ten) neurodevelopmental domains. A detailed study protocol outlining procedures, assessments, and criteria for diagnosing FAS, pFAS, or ND-AE has been published (Fitzpatrick et al., 2012). Non-participation in Stage 2 was due to families moving away from the Fitzroy Valley after Stage 1 (n = 15); withdrawal of consent (n = 1); and being unable to locate the child for assessment (n = 3).

The Fitzroy Valley is located in a very remote region of Western Australia. General practitioners, a midwife, and a Child Health Nurse are located in Fitzroy Crossing (the main town site), but Allied Health and Pediatric services are provided via an outreach, visiting service from Derby, 260 km (162 miles) to the west. Remote communities may only receive visits from Pediatric and Allied Health services once or twice a year, thus diagnostic and regular therapeutic services are extremely limited. One of the goals of the Lililwan Project was to highlight the needs of children in the area, and campaign for improved services. Prior to the Lililwan Project, only a small number of children had been reported as having a type of Fetal Alcohol Spectrum Disorder, and none had received a comprehensive neurodevelopmental assessment and formal diagnosis made by a multidisciplinary team.

The Lililwan Project provided both a clinical diagnostic service and enabled a population-prevalence study (Fitzpatrick et al., 2012). Each child and family received a comprehensive assessment report and therapy plan, including FASD diagnoses when relevant. Reports were provided to local health services and schools with parental consent. Referrals were made for ongoing therapy if warranted. Families whose child was diagnosed with FASD were offered support from an Aboriginal FASD educator and social worker.

2.2. Ethics

Ethical approval for the Lililwan Project was provided by the University of Sydney Human Research Ethics Committee; Western Australian (WA) Aboriginal Health and Information Ethics Committee; WA Country Health Services Board Research Ethics Committee; and Kimberley Aboriginal Health Planning Forum Research Sub-committee. Analysis and publication of VMI data was approved by the Curtin University Human Research Ethics Committee and the Western Australian (WA) Aboriginal Health and Information Ethics Committee. The Lililwan Project was initiated by local Aboriginal leaders, and extensive community consultation and engagement utilized through all stages of the project. Caregivers were provided with study information in written and verbal English and/or local language, and provided signed consent for participation of themselves and their child. Families and children could withdraw from the project at any stage without repercussions.

2.3. Participants

The majority of children (98.1%) identified as Australian Aboriginal, and were aged 7.5–9.6 years at the time of assessment (M = 8.7 years) (Table 1). Most children with PAE (n = 60) were exposed at risky (AUDIT-C score of 4 or 5; n = 4) or high risk (AUDIT-C score 6–12; n = 47) levels according to AUDIT-C criteria (Bush et al., 1998). FASD diagnoses made during the project (n = 21) included FAS (n = 1), pFAS (n = 12), and ND-AE (n = 8) (Table 1). No children were identified with strabismus, amblyopia, or other visual defects detected other than the reduced visual acuity reported in Table 1. Aside from marijuana, no other prenatal illicit drug use was reported.

Most children (n = 107) lived with either their biological mother or father (72.9%), but less than half lived with both biological parents (42.1%). Only half of the primary carers (n = 106) were involved in full-time employment (51.9%), with the remainder employed either on a part-time basis (10.4%) or unemployed (16.0%). On average, children had lived in two homes since birth, but some children had lived in as many as 10 homes. Education level of the birth mother (n = 86) included lower high school (Year 10 or less) (46.5%); upper high school (Year 11 or 12) (50.0%), or 'unknown' (3.5%). No birth mother attained further or tertiary education.

Clinical Psychologists assessed cognitive abilities using the Universal Non-verbal Intelligence Test (UNIT) (Bracken & McCallum, 1998). Similar to other neurodevelopmental assessment tools, including the Beery VMI, no normative data are available for Australian Aboriginal children. The UNIT was chosen as being the most appropriate measure of cognitive abilities for our cohort because it does not rely on verbal, language, or hearing abilities. UNIT full-scale standard scores did not differ between children with and without PAE and/or FASD (No PAE M = 89.9 (*SD* 8.5); PAE, no FASD M = 89.4 (*SD* 9.1); FASD M = 85.0

Table 1
Cohort characteristics.

	Total Cohort[a] $N = 108$		No PAE $n = 43$		PAE (no FASD) $n = 39$		FASD $n = 21$	
	n	(%)	n	(%)	n	(%)	n	(%)
Gender								
Male	57	(52.8)	24	(55.8)	18	(46.2)	13	(61.9)
Female	51	(47.2)	19	(44.2)	21	(53.8)	8	(38.1)
Australian Aboriginal	106	(98.1)						
Handedness								
Right	101	(93.5)	41	(95.3)	38	(97.4)	19	(90.5)
Left	7	(6.5)	2	(4.7)	1	(2.6)	2	(9.5)
Visual acuity[b,c]								
Normal	88	(81.5)	37	(86.0)	31	(79.5)	16	(76.2)
Reduced	11	(10.2)	4	(9.3)	2	(5.1)	4	(19.0)
Missing	9	(8.3)	2	(4.7)	6	(15.4)	1	(4.8)
Hearing[d]								
Normal	42	(38.9)	16	(37.2)	14	(35.9)	10	(47.6)
Mild loss	38	(35.2)	15	(34.9)	13	(33.3)	7	(33.3)
Moderate loss	13	(12.0)	7	(16.3)	3	(7.7)	3	(14.3)
Missing	15	(13.9)	5	(11.6)	9	(23.1)	1	(4.8)
Prenatal nicotine exposure[e]								
No	34	(31.5)	25	(58.1)	6	(15.4)	3	(14.3)
Yes	67	(62.0)	18	(41.9)	32	(82.1)	15	(71.4)
Unknown	7	(6.5)	0	(0)	1	(2.6)	3	(14.3)
Prenatal marijuana exposure[e]								
No	88	(81.5)	41	(95.3)	28	(71.8)	18	(85.7)
Yes	13	(12.0)	2	(4.7)	10	(25.6)	1	(4.8)
Unknown	7	(6.5)	0	(0)	1	(2.6)	2	(9.5)
PAE risk levels[f]								
No exposure	43	(100.0)	0	(0)	0	(0)	0	(0)
Low (1–3)	4	(3.7)	0	(0)	4	(10.3)	0	(0)
Risky (4–5)	4	(3.7)	0	(0)	3	(7.7)	1	(4.8)
High risk (≥6)	46	(42.6)	0	(0)	29	(74.4)	17	(81.0)
PAE, uncertain risk	6	(5.6)	0	(0)	3	(7.7)	3	(14.3)
Unknown PAE	5	(4.6)	0	(0)	0	(0)	0	(0)

[a] 'Total cohort' includes $n = 5$ children with unknown PAE who are not included in the No PAE, PAE (no FASD), or FASD groups.
[b] Reduced visual acuity defined as ≤6/9 in one or both eyes.
[c] Not all children completed audiology and ophthalmology testing.
[d] Mild hearing loss 26–40 dB; moderate hearing loss 41–55 dB.
[e] Some prenatal exposure information not available, either due to the primary carer not knowing, or the birth mother choosing not to disclose this information.
[f] Risk level according to AUDIT-C scoring criteria.

(*SD* 12.3); $p = 0.329$). Two children (both subsequently diagnosed with a type of FASD) had a UNIT standard score below 70, and thus met the criteria for an intellectual disability.

2.4. Assessment procedures

Children in the Lililwan Project were assessed in their local community or school in the presence of a local Aboriginal facilitator. Children were screened for hearing and visual impairments by an Audiologist and Ophthalmologist, and these results were reviewed prior to VMI assessment so accommodations could be made if required, such as ensuring the child was wearing prescribed glasses, or that task requirements were demonstrated if the child had a hearing impairment. The Beery VMI was administered in a single sitting and scored by the primary author (RD), who is a qualified Occupational Therapist with experience working with children in the Fitzroy Valley and other Kimberley communities. The assessor was blinded to PAE and maternal and child history during the assessment. The multidisciplinary team remained blinded to PAE until consensus was reached regarding whether the child met FASD diagnostic criteria.

2.5. The Beery-Buktenica Developmental Test of Visual-Motor Integration

The Beery VMI was used to assess VMI, visual perception, and fine motor coordination (Beery & Beery, 2010). The Beery VMI is a standardized, norm-referenced assessment of graphomotor VMI suitable for ages 2–100 years. Since its development in 1961, the Beery VMI has consistently demonstrated strong validity, including concurrent validity with the Bender-Gestalt (mean $r = 0.56$), the Developmental Test of Visual Perception ($r = 0.62–0.75$), and the drawing subtest of the Wide Range

Assessment of Visual Motor Abilities ($r = 0.52$) (Beery & Beery, 2010). It also has been shown to have sound content reliability, test-retest reliability, and inter-rater reliability (Beery & Beery, 2010). The Beery VMI norms were used, which are based on data from 1737 US children and adolescents aged 2–18 years, and 1021 adults aged 19–100 years (Beery & Beery, 2010). Although Beery VMI norms do not exist for Australian Aboriginal children, the authors of the Beery VMI claim it is suitable for use with children from different backgrounds, because it does not require alphabetical or numerical knowledge (Beery & Beery, 2010). Tasks include copying a series of 24 increasingly complex geometric shapes. Scoring ceases after three consecutive errors. Shapes are scored as 'correct' or 'incorrect' according to detailed criteria, with higher scores indicating better performance. Scores are reported as standard scores, descriptive categories, and percentile ranks, and range from: <70 ('very low', <2nd percentile); 70–79 ('low', 2nd to 7th percentile); 80–89 ('below average', 8th to 16th percentile); 90–109 ('average', 17th to 65th percentile); 110–119 ('above average', 66th to 72nd percentile); 120–129 ('high', 73rd to 97th percentile); and >129 ('very high', >98th percentile). The different types of errors made by each child were counted and assigned one point per error. Errors were classified as ratio or spatial (if the shape was copied disproportionately or oriented incorrectly on the page); corner difficulties (rounded or open); 'dog-earing' (insertion of an extra line or angle due to difficulty turning a corner); reversal or directionality confusion; errors that may indicate difficulties with crossing the midline; and gross distortions in which the original shape was difficult to discern (Fig. 1).

The Beery VMI Visual Perception and Fine Motor Coordination subtests were also administered to all children. These subtests can be used to identify whether VMI difficulties could be due to underlying problems with either visual perception or fine motor coordination (Beery & Beery, 2010). The Visual Perception subtest is a relatively motor-free measure of visual perception skills, requiring the child to point at the shape they identify as matching a stimulus. The Visual Perception subtest is timed, and scoring ceases after 3 min, or once the child makes three consecutive errors. The Fine Motor Coordination subtest requires the child to trace the outline of shapes while staying within double-lined borders. It includes the same shapes as the core Beery VMI test and the Visual Perception subtest. The Fine Motor Coordination subtest minimizes visual perception requirements by using examples, starting dots, and paths as visual guides (Beery & Beery, 2010). The Fine Motor Coordination subtest is timed and scoring ceases after 5 min, but the number of errors allowed is unlimited. Each subtest uses the same shapes as the core component of the Beery VMI, which increases the validity of comparisons between the core test and the subtests (Beery & Beery, 2010).

2.6. Statistical analysis

Means (M), standard deviations (*SD*), and confidence intervals for the mean (CI) were calculated from the standard scores from the Beery VMI and the Visual Perception and Fine Motor Coordination subtests; counts of error types made by each child were summed; and the number of subtest items completed in the timeframe recorded. Results were reported for the total cohort, and scores compared between children without PAE ('No PAE' group); PAE (including low, risky, high-risk, and uncertain risk levels) who were not diagnosed with any type of FASD because they did not have significant impairment in a minimum of three neurodevelopmental domains ('PAE (no FASD)' group); and a FASD diagnosis ('FASD' group). The FASD group included all diagnoses (FAS; pFAS; and ND-AE) as there were insufficient numbers in each diagnostic grouping to perform separate comparisons. The small numbers in each group made statistical adjustment for potential confounders infeasible. Data were assessed for normality using the Shapiro-Wilk test. Normally distributed data were analyzed using a one-way between groups analysis of variance (ANOVA) to compare scores between groups, and Tukey's Honestly Significant Difference test (HSD) was used to determine which groups differed from each other. Non-parametric data were analyzed using the Kruskal-Wallis test. Effect sizes (eta^2) were calculated, with 0.01 being considered a small effect size; 0.06 a medium effect size; and 0.14 a large effect size (Portney & Watkins, 2000). Significance was set at $p < 0.05$. The prevalence of impairment in VMI, visual perception, and fine motor coordination in each group was reported at two different levels: i) 'moderate' impairment (one or more *SD* below the mean; or ≤16th percentile); and ii) 'severe' impairment (two or more *SD* below the mean; or ≤2nd percentile). Chi-square analysis was used to determine if prevalence rates differed between groups. Statistical analysis was completed using IBM SPSS Statistics for Windows version 21.0 (Armonk, NY: IBM Corp.).

3. Results

3.1. Visual-motor integration

The mean VMI scores were in the 'below average' range for the total cohort (M = 87.8), and for children without PAE (M = 89.6); PAE (no FASD) (M = 87.0); and FASD (M = 84.2), and differences between groups were not significant ($F(2100) = 2.5$; $p = 0.091$) (Table 2). Ratio/spatial errors were the most common error type when copying shapes for all groups (Table 3; and Fig. 1). Children with PAE (no FASD) or FASD were two to three times more likely than children without PAE to draw shapes that were distorted, reversed, or had 'dog earing' errors (Table 3; and Fig. 1), but there were no significant differences in VMI scores or types of errors between groups (Tables 2 and 3).

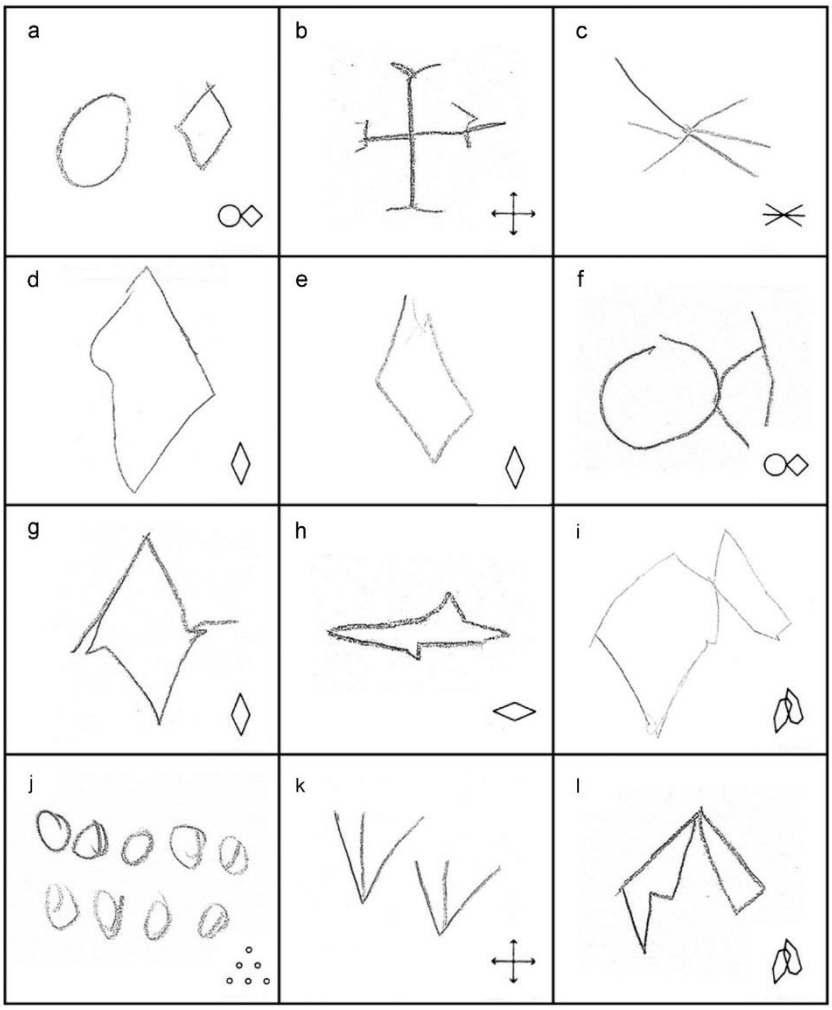

Fig. 1. Examples of errors made when copying Beery VMI shapes.
a = spatial error; b = reversal/directionality confusion; c = crossing the midline error; d–f) corner difficulties (d = rounded corner; e = open corner; f = both rounded and open corner); g–i = dog-earing errors; j–l = distortions. The correct shape is shown in the bottom right corner of each box.

Table 2
Beery VMI, visual perception, and fine motor coordination standard scores for the cohort, and according to PAE and FASD status.

	Total Cohort[a] $N = 108$			No PAE $n = 43$			PAE (no FASD) $n = 39$			FASD $n = 21$			Effect size[b]	ANOVA[c]
	M	(*SD*)	95% CI	M	(*SD*)	95% CI	M	(*SD*)	95% CI	M	(*SD*)	95% CI	(eta[b])	(*p*)
Beery VMI[d]	87.8	(9.6)	86.0–89.6	89.6	(10.4)	86.4–92.8	87.0	(8.3)	84.3–89.6	84.2	(8.3)	80.4–88.0	0.05	0.091
Visual Perception[d]	97.6	(12.5)	95.2–100.0	97.9	(12.7)	94.0–101.8	99.4	(11.9)	95.5–103.2	93.7	(13.1)	87.7–99.6	0.03	0.239
Motor Coordination[d]	93.9	(11.6)	91.6–96.1	95.1	(10.7)	91.8–93.8	96.1	(10.9)	92.6–99.6	87.9	(12.5)	82.2–93.6	0.08	0.020[*,e]

[*] $p < 0.05$.
[a] 'Total Cohort' includes $n = 5$ children with unknown PAE who are not included in the No PAE, PAE (no FASD), or FASD groups.
[b] 0.01 = a small effect size; 0.06 = medium effect size; 0.14 = large effect size.
[c] Between group differences for No PAE; PAE (no FASD); and FASD.
[d] The Beery VMI, Visual Perception, and Motor Coordination standard scores have a M = 100, *SD* = 15.
[e] Tukey's Honestly Significant Difference test: No PAE > FASD ($p = 0.046$); and PAE (no FASD) > FASD ($p = 0.020$).

Table 3
Beery VMI Error types for the cohort, and according to PAE and FASD status.

Error type	Total Cohort[a] $N = 108$			No PAE $n = 43$			PAE (no FASD) $n = 39$			FASD $n = 21$			Kruskal-Wallis
	M	(*SD*)	95% CI	M	(*SD*)	95% CI	M	(*SD*)	95% CI	M	(*SD*)	95% CI	(*p*)
Ratio/spatial	3.3	(1.3)	3.1–3.6	3.7	(1.4)	3.3–4.2	3.1	(1.4)	2.6–3.6	3.0	(1.0)	2.5–3.4	0.052
Corner (rounded/open)	0.4	(0.6)	0.2–0.5	0.4	(0.7)	0.2–0.6	0.3	(0.6)	0.2–0.5	0.3	(0.5)	0.1–0.5	0.974
Dog earing	0.1	(0.4)	0.1–0.2	0.1	(0.2)	0.0–0.1	0.2	(0.5)	0.0–0.4	0.2	(0.4)	0.0–0.4	0.159
Reversal/directionality	0.3	(0.5)	0.2–0.3	0.1	(0.4)	0.0–0.3	0.4	(0.5)	0.2–0.5	0.2	(0.4)	0.0–0.4	0.070
Crossing the midline	0.1	(0.4)	0.1–0.2	0.1	(0.0)	0.0–0.2	0.1	(0.0)	0.0–0.2	0.2	(0.4)	0.0–0.4	0.801
Distortions	0.3	(0.6)	0.2–0.4	0.2	(0.6)	0.0–0.4	0.4	(0.5)	0.2–0.6	0.5	(0.7)	0.2–0.8	0.065

[a] 'Total Cohort' includes $n = 5$ children with unknown PAE who are not included in the No PAE, PAE (no FASD), or FASD groups.

Table 4
Prevalence of visual-motor integration, visual perception, and fine motor coordination impairments for the cohort, and according to PAE and FASD status.

	Total Cohort[a] $N = 108$		No PAE $n = 43$		PAE (no FASD) $n = 39$		FASD $n = 21$		Chi-square
	n	(%)	*n*	(%)	*n*	(%)	*n*	(%)	*p*
Beery VMI									
$\geq -2SD$[b]	2	(1.9)	1	(2.3)	1	(2.6)	0	(0)	0.768
$\geq -1\ SD$[3]	51	(47.2)	18	(41.9)	17	(43.6)	14	(66.7)	0.144
Visual Perception									
$\geq -2SD$[b]	1	(0.9)	1	(2.3)	0	(0)	0	(0)	0.494
$\geq -1SD$[c]	19	(17.6)	9	(20.9)	4	(10.3)	5	(23.8)	0.309
Motor Coordination									
$\geq -2SD$[b]	3	(2.8)	1	(2.3)	0	(0)	2	(9.5)	0.107
$\geq -1SD$[c]	29	(26.9)	10	(23.3)	6	(15.4)	11	(52.4)	0.007*

* $p < 0.05$.
[a] 'Total Cohort' includes $n = 5$ children with unknown PAE who are not included in the No PAE, PAE (no FASD), or FASD groups.
[b] $\geq -2SD = \leq$ 2nd percentile.
[c] $\geq -1SD = \leq$ 16th percentile.

3.2. Visual perception

The mean Visual Perception scores were in the 'average' range for the cohort (M = 97.6), and also for all groups, and differences between groups were not significant ($F(2100) = 1.5$; $p = 0.239$) (Table 2). There were no significant differences between groups in the visual perception scores (Table 2), including the number of tasks completed in the allotted timeframe (No PAE M = 29.0 (*SD* 1.7); PAE (no FASD) M = 29.0 (*SD* 2.0); FASD M = 28.6 (*SD* 2.8), $p = 0.861$).

3.3. Fine motor coordination

The Fine Motor Coordination subtest scores for the overall cohort were in the 'average' range, but were 'below average' and significantly lower in children with FASD (M = 87.9, p = 0.020) than those without PAE (M = 95.1, HSD p = 0.046) and with PAE (no FASD) (M = 96.1; F (2100) = 4.1; HSD p = 0.020) (Table 2). There were no differences between groups regarding the number of items completed in the timeframe (No PAE M = 28.1 (SD 2.0); PAE (no FASD) M = 28.3 (SD 2.1); FASD M = 27.5 (SD 3.1), p = 0.628).

3.4. Prevalence of impairments

Few children in the cohort had severely impaired VMI ($n = 2$; 1.9%), Visual Perception ($n = 1$; 0.9%), or fine motor coordination ($n = 3$; 2.8%) (Table 4). However, nearly half of the children in the cohort had moderate VMI impairment (47.2%). Although children with FASD had the highest rates of moderate VMI impairment (66.7%), rates were also high for children without PAE (41.9%) and with PAE (no FASD) (43.6%), and differences between groups were not statistically significant. Prevalence rates for moderate impairment in visual perception in our cohort (17.6%) were similar to norms, and similar between groups. Rates of moderate impairment in fine motor coordination were slightly higher (26.9%) in the total cohort than expected based on Beery VMI norms (16.0%), but were significantly higher in children with FASD (52.4%) compared to children without PAE (23.3%, $p = 0.007$) (Table 4).

4. Discussion

This study is the first in-depth report of VMI, visual perception, and fine motor coordination skills in a population-based cohort of Aboriginal children living in remote Australia. The population had 'below average' VMI scores, regardless of the presence or absence of PAE or FASD, but had 'average' Visual Perception and Fine Motor Coordination (except in children

with FASD), which may indicate that the children had difficulties coordinating or integrating these two skill sets (Beery & Beery, 2010).

Children with FASD had significantly lower fine motor coordination scores and higher rates of moderate impairment in fine motor coordination than children without PAE. Only one other study has investigated whether VMI difficulties in children with PAE could be due to visual perceptual or fine motor deficits (Janzen et al., 1995). In that small study (n = 20), 3.5–5 year old children with FAS (n = 10) had significantly lower VMI scores than a matched control group but, similar to our findings, visual perception (assessed using the Recognition-Discrimination Test from the Florida Kindergarten Screening Battery; Satz & Fletcher, 1982) was unaffected. When considered along with other fine motor outcomes from the study, the authors concluded that the impairment in VMI was more likely due to impaired fine motor skills than visual perception. However, in our study, even children without PAE or FASD had poor VMI performance, so it is inconclusive as to whether the fine motor coordination problems experienced by children with FASD contributed to their VMI problems, but it is likely they had some impact on VMI performance.

Few children in our study had severe impairment in VMI, visual perception, or fine motor coordination. However, rates of moderate impairment in VMI were very high across the cohort, particularly in children with FASD, who had VMI impairment more than four times higher (66.7%) than expected based on published norms (Beery & Beery, 2010). Rates of moderate impairment in Visual Perception were similar to norms for all groups. More than half of the children with FASD had moderate impairment of fine motor coordination (52.4%), which was significantly higher than children without PAE (23.3%) and children with PAE (no FASD) (15.4%). Our results have important implications for FASD diagnoses and therapeutic interventions. The Canadian FASD Diagnostic Guidelines propose that for the purpose of FASD diagnosis only impairments below the 2nd percentile (or when there is a discrepancy of 1.5–2 *SD* between subtests) should contribute towards a FASD diagnosis (Chudley et al., 2005). However the CDC FASD diagnostic guidelines suggest that impairments below the 16th percentile should contribute to a FASD diagnosis, citing evidence that only a quarter of children with FAS (the most severe type of FASD diagnosis) have neurodevelopmental impairment below the 2nd percentile, and that using a cut-off of 2 *SD* would preclude many affected children from a diagnosis (Centers for Disease Control and Prevention, 2005). The more conservative cut-off of 1 *SD* has also been proposed for use when diagnosing ND-PAE within the DSM-5 guidelines (Doyle & Mattson, 2015). Only two children in our study had impairment in VMI below the 2nd percentile, which seems conservative given the very high levels of PAE, FASD, and other neurodevelopmental risk factors evident in the cohort. Conversely, the cut-off for impairment proposed by the CDC guidelines may have over-estimated rates of impairment in VMI impairment in children with PAE or FASD. This issue warrants further consideration and investigation.

The lack of significant differences in VMI between children with PAE or FASD compared to children without PAE in our study is inconsistent with other studies. The Beery VMI has detected impairments in children with moderate to high PAE (>10 drinks/week) (Korkman, Kettunen, & Autti-Rämö, 2003), as well as in children with FASD (Astley et al., 2009). In another study significant differences in VMI scores were found in children with FAS, but not heavy PAE without FAS (Mattson, Riley, Gramling, Delis, & Jones, 1998). In most studies children with PAE or FASD were compared to typically developing children. It may be that our non-significant findings are due to our relatively small sample size. Additionally, our non-significant findings may reflect the multitude of neurodevelopmental risk factors experienced by the children in our cohort, which is similar to many Aboriginal children in remote regions. Even children without PAE may have been exposed to intergenerational and early life trauma along with loss of land and culture; restricted access to therapeutic and early childhood services; overcrowded living conditions; and poor nutrition (Australian Health Ministers' Advisory Council, 2012). Many children in our cohort, even those without PAE, had high levels of prenatal nicotine exposure. Some studies have shown prenatal nicotine exposure may adversely affect VMI skills (Cornelius, Ryan, Day, Goldschmidt, & Willford, 2001; Willford, Chandler, Goldschmidt, & Day, 2010), although these studies have used assessment tools other than the Beery VMI and it remains uncertain whether graphomotor VMI skills are affected by prenatal nicotine exposure. Prenatal marijuana exposure was also high, particularly in the group with PAE who were not diagnosed with FASD, but similarly no studies have examined the impact on graphomotor VMI skills. Many children in the Fitzroy Valley participate in multiple outdoor and cultural activities, such as hunting and recreational sports (Lucas et al., Epub ahead of print), which may promote visual perception skills. Indoor activities which promote VMI and fine motor skills in the early years, such as arts and crafts, are less common and may also have contributed to the below average VMI skills in our cohort.

We also examined the types of errors children made when copying VMI shapes on the core test component, and the time taken to complete the Visual Perception and Fine Motor Coordination subtests. Ratio or spatial errors were the most common type of error made by all groups. Children with PAE or FASD were up to four times more likely to draw shapes which were distorted, reversed, or had 'dog earing' errors, although differences between groups were not significant. The types of Beery VMI errors have been investigated in one other group of children with FASD (Uecker & Nadel, 1996). These researchers found that children with FAS (n = 15) made more dog-earing errors and drew shapes which were grossly distorted compared to a control group, although, similar to our study, differences between groups were not statistically significant. 'Dog earing' and reversal type errors can indicate immature VMI development (Beery & Beery, 2010), which has been associated with difficulties with handwriting and other academic skills including reading, writing, and spelling (Kulp, 1999; Volman, van Schendel, & Jongmans, 2006; Weil & Cunningham Amundson, 1994).

There were no differences between groups in the number of tasks completed in the allotted time for the Visual Perception or Fine Motor Coordination subtests, suggesting that children with PAE or FASD neither worked more slowly, nor were more impulsive, than children without PAE. Other studies have shown that children with heavy PAE have difficulty sustaining

visual attention (Remove bold from author name and dateMattson et al., 2006), as well as delayed central processing and motor response time (Simmons et al., 2002). Accordingly, it was anticipated that the children in our study with PAE or FASD would take longer to complete the Visual Perception or Fine Motor Coordination subtests, but this hypothesis was not supported.

4.1. Strengths

This is the first population-based report of VMI skills in Aboriginal children in a remote region of Australia using a standardized, norm-referenced assessment tool. The study had a very high participation rate, and assessed almost two entire age cohorts of children in the Fitzroy Valley. The choice of assessment tools was unique in that we used the optional Beery VMI Visual Perception and Fine Motor Coordination subtests, which have not been reported in other studies of children with FASD. Use of the Beery VMI subtests, rather than different assessment tools, increases the likelihood that any observed difficulties are actual deficits rather than due to variation between different assessments (Beery & Beery, 2010).

4.2. Limitations

This study had several limitations. First, it was conducted in a remote area of Western Australia, so results may not be applicable to other populations with different demographics and social conditions. Conversely, outcomes may be applicable to other children living in similar remote communities. Second, PAE was reported retrospectively by parents or caregivers, which may introduce recall bias. However, information was corroborated by review of medical records and reports from direct observers, such as family members. There is some evidence that retrospective reporting is accurate up to 14 years after birth (Alvik, Haldorsen, Groholt, & Lindemann, 2006). Third, although the study included two entire age cohorts of children in the region and participation rates were high, the relatively small sample size meant that we could not statistically control for potential confounders, including prenatal nicotine and marijuana exposure, and future studies should consider the impact of these and other factors on neurodevelopment, including VMI. Fourth, the Beery VMI has not been validated for use with Australian Aboriginal children, and norms are based on a US normative sample. The low overall VMI scores may indicate that the Beery VMI was invalid for use with our cohort, and there are concerns about the cross-cultural validity of neurodevelopmental tests developed for, and normed with, populations with different linguistic or cultural backgrounds (Thorley & Lim, 2011). However, the Beery VMI was selected because the test developers claim it is 'virtually culture-free' and does not require alphabet or numerical knowledge (Beery & Beery, 2010), and is endorsed as an appropriate FASD diagnostic assessment tool (Chudley et al., 2005). Additionally, children in our study had been attending school for a number of years, and thus should have been familiar with completing similar tasks to those of the Beery VMI. Fifth, this paper reports VMI data from a population-based prevalence study. Children with FASD were more likely to have VMI impairments, because significant impairment in VMI could contribute to the FASD diagnosis. However, FASD diagnoses were only made in the presence of confirmed PAE and at least three domains of severe impairment, which may or may not include VMI.

4.3. Future directions

Future studies should examine potential confounders, other than PAE, which may account for poor VMI skills observed in our cohort. Consideration should also be given to what degree of impairment should be considered 'significant' in terms of FASD diagnosis. Studies in other populations have shown the effectiveness of therapeutic interventions to improve handwriting proficiency, including in children with poor VMI skills (Case-Smith, 2002) and as a collaborative approach between Occupational Therapists and classroom teachers (Case-Smith, Weaver, & Holland, 2014). Trialling similar programs for children in the Fitzroy Valley may be of benefit.

5. Conclusions

The VMI skills of Aboriginal children in the Fitzroy Valley region in our cohort were below average, indicating that many children in the region require therapeutic support regardless of whether they have PAE or FASD. Visual perception and fine motor coordination performance were within normal ranges for the cohort except in those with FASD, who had poorer fine motor coordination skills than children without PAE. Our findings indicate that both VMI and fine motor coordination should be assessed in children with PAE, and as part of the FASD diagnostic process, to give a more accurate profile of neurological impairment. This knowledge can be used to guide the development of therapeutic interventions which target specific areas of impairment.

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C.4 Publication 4

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Research Article

Graphomotor skills in children with prenatal alcohol exposure and fetal alcohol spectrum disorder: A population-based study in remote Australia

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Background/aim: *Few studies have examined graphomotor skills in children with prenatal alcohol exposure (PAE) or fetal alcohol spectrum disorder (FASD).*

Methods: *Graphomotor skills were assessed in 108 predominantly Australian Aboriginal children aged 7.5–9.6 years in remote Western Australia using clinical observations (pencil grasp; writing pressure) and standardised assessment tools (the Evaluation Tool of Children's Handwriting; and the Miller Function and Participation Scales – The Draw-a-Kid Game). Skills were compared between children (i) without PAE, (ii) PAE but not FASD and (iii) FASD.*

Results: *Most children used a transitional pencil grasp and exerted heavy handwriting pressure (83.3% and 30.6% of the cohort). The percentage of letters (M = 62.9%) and words (M = 73.3%) written legibly was low. Children with FASD were more likely than children without PAE to use a cross-thumb grasp (P = 0.027), apply heavy writing pressure (P = 0.036), be unable to write a sentence (P = 0.041) and show poorer word legibility (P = 0.041). There were no significant differences between groups for drawing outcomes, although some children with FASD drew pictures that appeared delayed for their age. There were no significant differences between children without PAE and those with PAE but who were not diagnosed with FASD.*

Conclusions: *Overall, graphomotor skills were poor in this cohort, but children with FASD performed significantly worse than children without PAE. Findings suggest the need for improved occupational therapy services for children in remote regions and evaluation of graphomotor skills in children with PAE.*

KEY WORDS *fetal alcohol spectrum disorder, handwriting, motor skill, psychomotor performance, indigenous population.*

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This research study was completed at various schools and health clinics in the Fitzroy Valley, Western Australia. The base site was at Nindilingarri Cultural Health Service, PO Box 59, Fitzroy Crossing, Western Australia 6765, Australia.

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Introduction

Graphomotor skills include handwriting and drawing and involve the reproduction of letters, figures, pictures or plans either from memory or by copying, onto paper or another writing surface using a pencil or other writing implement (Ziviani & Wallen, 2006). Graphomotor skills facilitate the recording of information, thoughts and events; are a tool for communication; and allow expression of feelings and ideas (Tomchek & Schneck, 2006). In children, successful graphomotor performance is essential for participation in numerous classroom activities to demonstrate learning, as well as recreational and play activities. Previous studies indicated that up to 60% of the school day is spent in handwriting and other fine motor tasks (McHale & Cermak, 1992). Despite recent advancements in and increased use of computers and other technology to complete academic tasks (Cahill, 2009), students with poor handwriting are also likely to have difficulty with keyboarding skills (Connelly, Gee & Walsh, 2007). Further, handwriting proficiency can influence the quality of academic work (Baker, Gersten & Graham, 2003), and students with illegible handwriting are more likely to receive lower grades regardless of written content (Chase, 1986). Handwriting and drawing are complex developmental skills which require a complex interaction of biomechanical, psychomotor, cognitive and linguistic abilities (Benbow, 2006).

Although research on graphomotor skills in children with prenatal alcohol exposure (PAE) is limited, PAE can disrupt the development of many neural regions which are involved in graphomotor skills, including the cerebellum, basal ganglia, corpus callosum and motor cortex (Norman, Crocker, Mattson & Riley, 2009; Xie, Yang, Chappell, Li & Waters, 2010). PAE can also impair nerve conduction (De los Angeles Avaria *et al.*, 2004) and cause skeletal malformations (Jones *et al.*, 2010) and atypical muscle development (David & Subramaniam, 2005), which may also affect graphomotor proficiency. Individuals with PAE and significant, multiple neurodevelopmental impairments may be diagnosed with one of the fetal alcohol spectrum disorders (FASDs). This umbrella term includes the diagnoses of fetal alcohol syndrome (FAS), in which individuals have characteristic facial dysmorphology and significant growth impairment; partial FAS (pFAS), with fewer dysmorphic facial features and normal growth; and Alcohol Related Neurodevelopmental Disorder or Neurodevelopmental Disorder Alcohol Exposed, with few or no dysmorphic facial features and normal growth. All diagnoses require significant neurodevelopmental impairment in at least three domains of function, which may include hard and soft neurologic signs (including sensory-motor impairment), cognition, communication, academic achievement, memory, executive functioning, attention deficit/hyperactivity, or adaptive skills and social communication (Chudley *et al.*, 2005). Individuals with PAE may have some degree of impairment but not at a level sufficient to be diagnosed with a type of FASD (Astley *et al.*, 2009).

Children with FASD often have impaired fine and visual motor skills (Adnams *et al.*, 2001; Barr, Streissguth, Darby & Sampson, 1990; Mattson *et al.*, 2010). However, despite anecdotal reports of graphomotor impairment in children with FASD (Clarren, 2004), few studies have reported the quality of handwriting or drawing skills in children with FASD. Those studies have either included only a small, exploratory sample ($n = 20$) (Duval-White, Jirikowic, Rios, Deitz & Olson, 2013), or have not assessed human figure drawing skills within a motor performance framework (Aronson, Kyllerman, Sabel, Sandin & Olegard, 1985; Urban *et al.*, 2008). Functional assessments of graphomotor skills in children with PAE may assist identification of fine motor impairment during the FASD diagnostic process, improve knowledge of the functional implications of PAE and guide therapeutic interventions.

Study aims

The purpose of this study was to describe graphomotor performance of children in the remote Fitzroy Valley region of northern Western Australia. We aimed to assess the following:

1. Pencil grasp, writing pressure, and ability to write their name and a short sentence, using clinical observation
2. Handwriting legibility in terms of percentage of letters and words formed correctly when writing their name and a short sentence, using the Evaluation Tool of Children's Handwriting (Amundson, 1995)
3. Drawing abilities in terms of motor accuracy and body awareness, using the Miller Function and Participation Scales (Miller, 2006)

The differences in graphomotor skills between children without PAE, children with PAE but who were not diagnosed with FASD and children with FASD were determined. This is the first comprehensive description of graphomotor skills in Aboriginal children in remote Australia and the first to examine whether graphomotor skills differ between children with PAE or FASD.

In accordance with the teratogenic nature of PAE on neural regions associated with graphomotor skills, it was anticipated that (i) children with PAE or FASD would have poorer handwriting and drawing skills than children without PAE and (ii) children with FASD would be most impaired.

Methods

Background and setting

The children completed graphomotor assessments as part of the Lililwan Project, which was Australia's first

active case ascertainment population-based study of FASD prevalence (Fitzpatrick *et al.*, 2012). The Lililwan Project formed part of the Marulu strategy, which is an initiative developed by local Aboriginal leaders in response to their concerns about the impact of high levels of alcohol misuse in the region, including consumption of alcohol during pregnancy. In 2010, families of children born in 2002 or 2003 who were currently living in the Fitzroy Valley were invited to participate in the Lililwan Project (n = 127, 95% participation). Parents or caregivers completed in-depth verbal questionnaires with 'community navigators', who were local Aboriginal people who worked with the Lililwan Project clinicians to ensure cultural safety of procedures. Families provided information regarding prenatal and postnatal exposures, including health, developmental and socioeconomic circumstances which may have impacted on the child's development (Fitzpatrick *et al.*, 2013). PAE was scored according to a standardised measure of alcohol consumption (the Alcohol Use Disorders Identification Test Consumption (Bush, Kivlahan, McDonell, Fihn & Bradley, 1998)). In 2011, the children who were then aged from seven to nine years completed approximately six hours of health and neurodevelopmental assessments with an audiologist, occupational therapist, ophthalmologist, paediatrician, physiotherapist, psychologist and speech pathologist. Assessors were blinded to PAE and other neurodevelopmental risk factors, such as early-life trauma. Clinicians conducted comprehensive case conferences for each child to determine if they met FASD diagnostic criteria. Children were assigned FASD diagnoses according to Canadian FASD Diagnostic Guidelines (Chudley *et al.*, 2005), which were modified to suit the cultural context. A detailed study protocol has been published (Fitzpatrick *et al.*).

As part of the multidisciplinary neurodevelopmental assessments, children completed approximately one hour of assessments, including graphomotor skills, with a qualified occupational therapist (R. D.) who was experienced in working with Aboriginal children in the region.

Participant consent and ethical approval

The Lililwan Project was conducted in accordance with National Health and Medical Research Council's guidelines for ethical conduct in Aboriginal and Torres Strait Islander health research (National Health and Medical Research Council, 2003). Local Aboriginal leaders in the Fitzroy Valley conceived and designed the protocols for the Lililwan Project. Extensive community consultation occurred in the communities prior to, and throughout, the project. Families received study information verbally and written information about the study in English or their local language if preferred. Families or children could withdraw from the study or assessment process at any stage without repercussions. 'Community navigators', who were local Aboriginal people, assisted clinicians in administering assessments and interpreting results, and if requested by the family, they could be present when results from neurodevelopmental assessments were provided to families.

Ethical approval was provided for the Lililwan Project by the Kimberley Aboriginal Health Planning Forum Research Sub-committee, University of Sydney Human Research Ethics Committee, Western Australian Aboriginal Health and Information Ethics Committee and the Western Australian Country Health Services Board Research Ethics Committee. The Curtin University Human Research Ethics Committee and the Western Australian Aboriginal Health and Information Ethics Committee provided separate approval related to the fine motor, including graphomotor, aspects of the Lililwan Project.

Outcome measures

Clinical observations

Observations recorded during graphomotor tasks included (i) hand dominance; (ii) writing pressure, which was ranked as 'light', 'light to appropriate', 'appropriate', 'appropriate to heavy' or 'heavy'; and (iii) pencil grasp, which was classified according to Schneck and Henderson's (1990) criteria as either 'primitive' (digital pronate, radial cross-palmar, palmar supinate, digital pronate, brush or extended fingers grasps); 'transitional' (cross-thumb, static tripod or four-fingers grasps); or 'mature' (lateral tripod or dynamic tripod grasps).

Evaluation Tool of Children's Handwriting

Children were asked to write their name and a short sentence of their choice. The Evaluation Tool of Children's Handwriting (ETCH) Task VI Sentence Composition (Amundson, 1995) scoring guidelines were applied to evaluate letter legibility (name and sentence) and word legibility (sentence only). The ETCH is a criterion-referenced, standardised measure of handwriting ability suitable for primary-school aged children (Amundson). It has moderate to high intra-rater, inter-rater and test retest reliability for letter and word legibility, and good discriminant and concurrent validity (Duff & Goyen, 2010). Children could choose whether to use a cursive or manuscript handwriting style, because local schools teach both styles. Handwriting samples were evaluated for correct letter formation, spacing, size and alignment. The ETCH scores represent the percentage of (i) letters which are legible in their name, (ii) letters which are legible in a sentence and (iii) words which are legible in a sentence, with higher percentages indicating better performance.

The Miller Function and Participation Scales: The Draw-a-Kid Game

Children were asked to draw a picture of themselves, a friend or family member. They were instructed to 'make

it the best drawing you can'. The Miller Function and Participation Scales (M-FUN): Draw-a-Kid Game (Miller, 2006) scoring guidelines were applied. Drawings were scored according to (i) body awareness (possible score range 0 6) of the drawn figure and (ii) motor accuracy (possible score range 0 9), which were summed to give (iii) a total score (possible score range 0 15). Higher scores represent better performance. Although normative data are not available for individual tasks including The Draw-a-Kid Game this task forms part of the M-FUN's Visual Motor subgroup, which has been demonstrated to have good internal consistency, excellent inter-rater reliability (Miller), and strong concurrent and construct validity (Diemand & Case-Smith, 2013). Although M-FUN Visual Motor norms are only available for children aged 2.6 7.11 years, developers of M-FUN advise that tasks, including the Draw-a-Kid Game, are suitable for use with older children (Miller).

Statistical analysis

Graphomotor skills were assessed as part of the neurodevelopmental assessments conducted during the Lililwan Project. Clinical observations were recorded by the occupational therapist during assessment of graphomotor and other fine motor tasks. Drawing and handwriting samples were scored retrospective to the Lililwan Project by two Occupational Therapists who were blinded to the child's PAE and FASD status. Inter-rater reliability was calculated using weighted kappa (κ) with quadratic weighting for ordinal M-FUN data, and intra-class correlation coefficients (ICC: two-way mixed model; single measures) for continuous ETCH data. Strength of agreement was interpreted as follows: 0.81 1.00 = excellent agreement, 0.61 0.80 = substantial agreement, 0.41 0.60 = moderate agreement, 0.21 0.40 = fair agreement, 0.00 0.20 = slight agreement and <0.00 = poor agreement (Landis & Koch, 1977).

Descriptive statistics were derived for clinical observations of hand dominance, pencil grasp, writing pressure, and the ability of children to write their name and a short sentence. Outcomes were reported for the total cohort and also according to whether children (i) did not have PAE ('No PAE' group); (ii) had PAE but did not meet criteria for one of the FASD diagnoses ('PAE, no FASD' group); and (iii) had PAE and were diagnosed with a type of FASD ('FASD' group). Children with unknown PAE ($n = 5$) were excluded from the between-groups analysis. Drawings that were not of a human figure ($n = 11$) were excluded from the M-FUN analysis. Results from clinical observations were compared between groups using chi-square tests. Drawing (M-FUN) and handwriting (ETCH) data had non-normal distributions, so a non-parametric test (Kruskal Wallis) was used to examine differences between groups. Statistical analysis was completed using IBM SPSS Statistics for Windows, version 21.0 (IBM Corp., Armonk, NY, USA).

Results

Cohort characteristics

Children ($n = 108$) were aged from 7.5 to 9.6 years (M = 8.7) at the time of assessment, and the majority were identified as being Australian Aboriginal (Table 1). Most children (81.5%) had lived in one to three homes since birth, but some (18.6%) had lived in more than four homes. Households consisted of an average of six people (range 2 16 people). Children had attended an average of 1.8 schools (range one to five schools), and most children (82.4%) attended four to five days per week.

Cognitive abilities were assessed by Clinical Psychologists using the Universal Non-verbal Intelligence Test (UNIT) (Bracken & McCallum, 1998). UNIT full-scale standard scores were similar between exposure groups (No PAE M = 89.9 (SD 8.5); PAE, no FASD M = 89.4 (SD 9.1); FASD M = 85.0 (SD 12.3); $P = 0.329$).

Clinical observations

The majority of the children in the cohort were observed to be right handed (93.5%) and used a transitional style pencil grasp (43.5% cross-thumb, 29.6% static tripod or 10.2% four-fingers grasp). Many children exerted non-optimal writing pressure, including 'appropriate to heavy' (22.2%) or 'heavy' (30.6%) (Table 2). While most children could write their first name (97.2%), some were unable to write their surname (15.7%) or a short sentence (6.5%) (Table 2).

Children with PAE (no FASD) did not differ significantly from other groups for any of the clinical observations. Children with FASD were more likely to use a cross-thumb pencil grasp than children without PAE ($P = 0.027$), more likely to exert heavy pressure when writing ($P = 0.036$) and less likely to be able to write a sentence ($P = 0.041$) (Table 2).

Handwriting

Letter and word legibility scores were relatively low across the cohort. The proportion legible words in a sentence was higher (M = 73.3%) than legible letters in the name (M = 60.8%) or sentence samples (M = 62.9%) (Table 2).

According to ETCH scoring criteria, letter legibility in their name (No PAE M = 62.5%; PAE, no FASD M = 60.9%; FASD M = 56.1%) or a sentence (No PAE M = 62.0%; PAE, no FASD M = 63.6%; FASD M = 60.1%) was low for all groups, and differences between groups were not significant (Table 1). However, for children with FASD, on average only half of the words in a sentence were written legibly (M = 50.0%) which was significantly less than children without PAE (M = 81.0%) and children with PAE, no

TABLE 1: *Cohort characteristics*

	Total cohort† (*N* = 108) *n* (%)	No PAE (*n* = 43) *n* (%)	PAE (no FASD) (*n* = 39) *n* (%)	FASD (*n* = 21) *n* (%)
Australian aboriginal	106 (98.1)			
Gender				
Male	57 (52.8)	24 (55.8)	18 (46.2)	13 (61.9)
Handedness				
Right	101 (93.5)	41 (95.3)	38 (97.4)	19 (90.5)
Hearing‡§ (*n* = 93)				
Normal	42 (45.2)	16 (37.2)	14 (35.9)	10 (47.6)
Mild loss	38 (40.9)	15 (34.9)	13 (33.3)	7 (33.3)
Moderate loss	13 (14.0)	7 (16.3)	3 (7.7)	3 (14.3)
Missing	15 (13.9)	5 (11.6)	9 (23.1)	1 (4.8)
Prenatal nicotine exposure¶				
No	34 (31.5)	25 (58.1)	6 (15.4)	3 (14.3)
Yes	67 (62.0)	18 (41.9)	32 (82.1)	15 (71.4)
Unknown	7 (6.5)	0 (0)	1 (2.6)	3 (14.3)
Prenatal marijuana exposure¶				
No	88 (81.5)	41 (95.3)	28 (71.8)	18 (85.7)
Yes	13 (12.0)	2 (4.7)	10 (25.6)	1 (4.8)
Unknown	7 (6.5)	0 (0)	1 (2.6)	2 (9.5)
PAE risk levels††				
No exposure	43 (100.0)	0 (0)	0 (0)	0 (0)
Low (1–3)	4 (3.7)	0 (0)	4 (10.3)	0 (0)
Risky (4–5)	4 (3.7)	0 (0)	3 (7.7)	1 (4.8)
High risk (≥6)	46 (42.6)	0 (0)	29 (74.4)	17 (81.0)
PAE, uncertain risk	6 (5.6)	0 (0)	3 (7.7)	3 (14.3)
Unknown PAE	5 (4.6)	0 (0)	0 (0)	0 (0)

†'Total cohort' includes *n* = 5 children with unknown PAE who are not included in the No PAE, PAE (no FASD) or FASD groups.

‡Not all children completed audiology testing.

§Mild hearing loss 26–40 dB; moderate hearing loss 41–55 dB.

¶Some prenatal exposure information not available, either due to the primary carer not knowing or the birth mother choosing not to disclose this information.

††Risk level according to AUDIT-C scoring criteria.

FASD (M = 73.9%, *P* = 0.008) (Table 2). Many of the children with FASD (Figure 2a c) had handwriting difficulties which were characterised by difficulties with letter and word formation in comparison to children without PAE (Figure 2d f).

Drawing

Most children scored towards the higher performance upper limits of the M-FUN Draw-a-Kid Game (possible score range 0 6) for body awareness (M = 5.2), but the score for motor accuracy (possible score range 0 9) was somewhat lower than the ceiling score (M = 7.6), as was the total score (possible score range 0 15) (M = 12.8) (Table 2).

M-FUN Draw-a-Kid Game scores (body awareness, motor accuracy and total score) were similar for all children regardless of PAE or FASD, and there were no statistical differences between groups (Table 2). However, some of the drawings done by children with PAE and/or FASD (Figure 1a c) showed evidence of developmental immaturity and poor pencil control, especially in comparison to children without PAE (Figure 1d f). Inter-rater reliability for the ETCH and M-FUN Draw-a-Kid Game is reported in Table 3.

Discussion

This is the first comprehensive description of graphomotor skills of 7.5- to 9.6-year-old children living in a remote area of Australia, most of whom were Aboriginal, and the first to examine whether these skills differed between children with and without PAE or FASD. Many children in this cohort had poor graphomotor skills, including a delayed pencil grasp and application of excessive pressure through their pencil, and showing reduced functional writing and handwriting legibility. Children with FASD were significantly more likely to have handwriting difficulties than children without PAE, including using a cross-thumb pencil grasp which was immature for their age, applying heavy pressure through their pencil during graphomotor tasks, being unable to write a sentence, and writing fewer legible words in sentence writing tasks. Drawing skills, which were evaluated for motor accuracy and body awareness, were similar between children with and without PAE or FASD.

The handwriting abilities of children in the Fitzroy Valley are concerning. In a previous study of 320 children aged 3 6.11 years in the United States, transitional grasps were used until about six years of age, but by 6.11 years, 72.5% of children used a dynamic tripod grasp (Schneck & Henderson, 1990). These findings contrasted to our cohort, in which only 13.9% of children used a dynamic tripod grasp. The children in the Lililwan Project were older than those in Schneck and Henderson's study, and thus would be expected to have a greater, not lesser, proportion of children using a mature pencil grasp. Although some researchers have failed to find a relationship between use of a dynamic pencil grasp and handwriting performance (Schwellnus *et al.*, 2012), it is generally acknowledged that transitional grasps are

TABLE 2: *Clinical observations, drawing (M-FUN Draw-a-Kid Game), and handwriting (ETCH) outcomes for the cohort, according to PAE and FASD status*

	Total cohort† (N = 108) n (%)	No PAE (n = 43) n (%)	PAE (no FASD) (n = 39) n (%)	FASD (n = 21) n (%)	P‡
Clinical observations					
Hand dominance					
Right	101 (93.5)	41 (95.3)	38 (97.4)	19 (90.5)	0.487
Left	7 (6.5)	2 (4.7)	1 (2.6)	2 (9.5)	0.487
Pencil grasp					
Primitive grasps	0 (0)	0 (0)	0 (0)	0 (0)	–
Transitional grasps					
Cross-thumb	47 (43.5)	12 (27.9)	18 (46.2)	13 (61.9)	0.027*
Static tripod	32 (29.6)	12 (27.9)	15 (38.5)	4 (19.0)	0.271
Four fingers	11 (10.2)	7 (16.3)	3 (7.7)	1 (4.8)	0.280
Mature grasps					
Lateral tripod	3 (2.8)	2 (4.7)	0 (0)	1 (4.8)	0.390
Dynamic tripod	15 (13.9)	10 (23.3)	3 (7.7)	2 (9.8)	0.104
Writing pressure (n = 107)					
Light	0 (0)	0 (0)	0 (0)	0 (0)	–
Light to appropriate	1 (0.9)	0 (0)	1 (2.6)	0 (0)	0.442
Appropriate	49 (45.4)	18 (41.9)	22 (56.4)	8 (38.1)	0.310
Appropriate to heavy	24 (22.2)	12 (27.9)	8 (20.5)	2 (9.5)	0.218
Heavy	33 (30.6)	12 (27.9)	8 (20.5)	11 (52.4)	0.036*
Handwriting ability					
Unable to write first or surname	3 (2.8)	1 (2.3)	1 (2.6)	1 (4.8)	0.851
Unable to write surname	17 (15.7)	8 (18.6)	4 (10.3)	5 (23.8)	0.358
Unable to write a sentence	7 (6.5)	2 (4.7)	1 (2.6)	4 (19.0)	0.041*
Drawing (M-FUN) (n = 97)	M (SD)	M (SD)	M (SD)	M (SD)	
Body awareness§	5.2 (1.0)	5.3 (1.1)	5.1 (1.0)	5.1 (1.2)	0.565
Motor accuracy¶	7.6 (1.6)	7.7 (1.6)	7.6 (1.4)	7.1 (2.0)	0.419
Total score††	12.8 (2.6)	13.0 (2.6)	12.6 (2.3)	12.2 (3.1)	0.522
Handwriting (ETCH)	M (SD)	M (SD)	M (SD)	M (SD)	
Name: Letter legibility‡‡ (n = 91)	60.8 (26.0)	62.5 (27.1)	60.9 (24.2)	56.1 (28.0)	0.729
Sentence: Letter legibility‡‡ (n = 101)	62.9 (22.3)	62.0 (24.4)	63.6 (21.7)	60.4 (21.5)	0.872
Sentence: Word legibility‡‡ (n = 101)	73.3 (29.1)	81.0 (22.1)	73.9 (28.6)	50.0 (37.0)	0.008**

*$P < 0.005$; **$p < 0.001$.

†Total cohort includes n = 5 children with unknown PAE who were excluded from the group analysis.

‡Significance tested for No PAE, PAE (no FASD) and FASD groups.

§Possible score ranges 0–6, ¶0–9 and ††0–15.

‡‡Scores indicate percentage of legible letters or words.

inefficient and can cause muscle fatigue and cramping and lead to poorer graphomotor output (Tseng & Cermak, 1993). An immature pencil grasp can also indicate problems with proprioception, sensory processing and the sensory-motor feedback loop (Benbow, 2006), as can exerting excessive pressure through the pencil during graphomotor tasks (Levine, 1987).

Many academic and recreational tasks require proficiency in graphomotor skills, and poor performance of these skills can impair the ability to participate in many classroom activities and communicate learned knowledge (Chase, 1986; Tomchek & Schneck, 2006). There are limited data directly related to graphomotor skills of Australian Aboriginal children. However, the National Assessment Program Literacy and Numeracy, which is an annual assessment of reading, writing, language and numeracy skills completed annually by Australian students, has highlighted that many Aboriginal students perform below national academic benchmarks (Australian Curriculum Assessment and Reporting

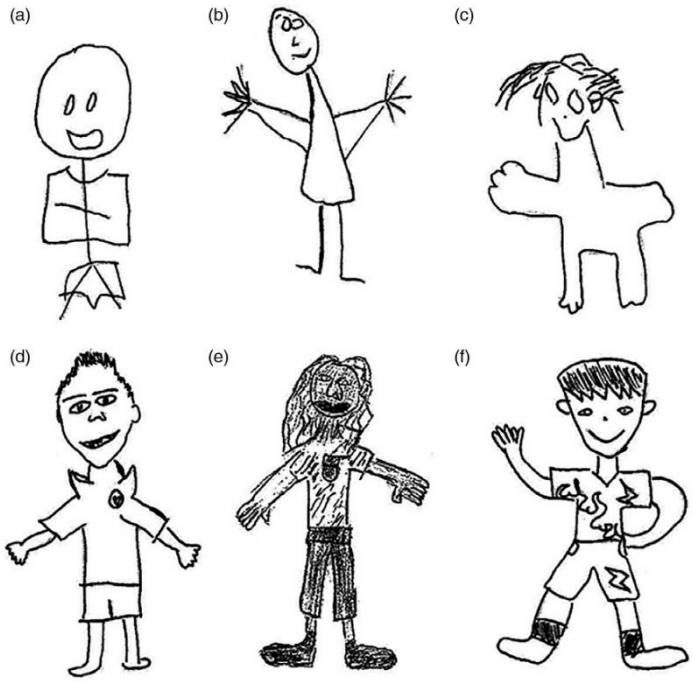

FIGURE 1: *Human figure drawings from the Miller Function and Participation Scales Draw-a-Kid Game. Drawn by children with (a) pFAS: 7.9 years; IQ = 93; (b) ND-AE: 8.8 years; IQ = 91; (c) pFAS: 8.1 years; IQ = 82; and (d) no PAE: 9.2 years; IQ = 86; (e) no PAE: 9.6 years; IQ = 98; (f) no PAE: 9.2 years; IQ = 90. IQ = Universal Nonverbal Intelligence Test (UNIT) Full Scale Standard Score. The UNIT has a normative M = 100.0, SD = 15.*

Authority, 2015). The Australian Early Developmental Index, which is based on teachers' evaluation of student competence in their first year of school, reports that 20.6% of students in Fitzroy Crossing were below the 10th percentile for fine and gross motor skills (The Royal Children's Hospital Melbourne, 2012). In addition, some aspects of local Aboriginal culture, such as painting and boab nut carving, require sound fine motor skills. Addressing graphomotor and other fine motor impairments is likely to have a positive flow-on effect to many aspects of a child's occupational performance.

Other groups have evaluated handwriting legibility using the ETCH. In a study of 31 children in Grade 1 in the United States, an average of 78 80% letter legibility was found when writing a sentence (Diekema, Deitz & Amundson, 1998). Similarly, a study evaluating 26 children in Grades 2 and 3 in Canada found a mean letter legibility of 73.3% and word legibility of 70.6% (Brossard-Racine, Mazer, Julien & Majnemer, 2012). Both these studies reported much higher letter legibility rates than for the children in the Lililwan Project (62.9%). Although the cohort's mean word legibility (73.3%) was similar to the children in Bossard-Racine's study, the children in our cohort were older, and therefore we expected them to have higher rates of legibility. The common use of immature pencil grasps and application of excessive writing pressure, along with fine motor (Doney *et al.*, submitted manuscript) and visual motor integration difficulties (Doney *et al.*, 2016) observed in our cohort, likely contributed to the reduced handwriting legibility for many of the children.

No other publications report outcomes from the Draw-a-Kid Game, so findings cannot be compared with other studies. This assessment tool was chosen because of its unique properties of evaluating motor accuracy in the context of human figure drawing rather than developmental maturity. The cohort's mean total score (M = 12.8) was lower than the maximum possible score (15), as were the motor accuracy scores (M = 7.6; maximum possible = 9). These scores are noteworthy as the Draw-a-Kid Game is designed for younger children (4.0 7.11 years) than those in the cohort (7.5 9.6 years), and hence, most children in the cohort should have scored at the upper limits. However, normative data are not available for the M-FUN Draw-a-Kid Game, so results should be interpreted cautiously.

Children with FASD had more difficulties with graphomotor skills than children without PAE. Some children with FASD had particular patterns of handwriting difficulties, including letter reversals, inconsistent letter size, missing or incorrect letter choice, and a lack of spacing between words. Typical examples are shown in Figure 1. The findings are consistent with those of Duval-White *et al.* (2013) who found that most children with FASD in their study ($n = 20$) scored in the 'well-below average' range for letter legibility.

TABLE 3: *Inter-rater reliability*

	Weighted Kappa†	Confidence interval	Strength of agreement‡
M-FUN			
Body awareness			
Number of parts	0.84	0.75–0.93	Excellent
Overall impression	0.44	0.30–0.58	Moderate
Motor accuracy			
Number of parts	0.62	0.43–0.81	Substantial
Overall impression	0.16	−0.003 to 0.32	Slight
ETCH: Name	ICC§		
Letter legibility	0.90	–	Excellent
ETCH: Sentence			
Letter legibility	0.90	–	Excellent
Word legibility	0.91	–	Excellent

†Weighted kappa with quadratic weights.

‡Strength of agreement based on Landis and Koch (1977) criteria.

§ICC = Intra-class correlation coefficient.

Children with PAE or FASD had similar drawing scores to children without PAE, which was an unexpected finding. The lack of significant differences between groups on the total score of the M-FUN may be due to low inter-rater reliability for the 'Motor Accuracy: Overall Impression' score, but this is unlikely because substantial to excellent reliability was achieved for all other scores which contributed to the total score. Similar to handwriting, some children with FASD had characteristic styles of drawing (Figure 2) which possibly reflects general developmental delay, rather than specific body awareness or motor accuracy difficulties.

In contrast with the drawing outcomes for the children in the Lililwan Project, human figure drawings from children with FASD were evaluated in two other studies and significant impairment was identified. One study of 142 Grade 1 South African children with pFAS or FAS reported significantly lower drawing scores than children without PAE (Urban *et al.*, 2008), although these children were younger than those in the Lililwan Project. Another study of 28 Swedish children with and without PAE evaluated human figure drawings and concluded that perceptual difficulties accounted for poorer drawing abilities in children with PAE (Aronson *et al.*, 1985). However, in these studies, the drawings were not evaluated within a motor skills framework, so comparisons with the findings in the present study are difficult.

Limitations and future directions

Despite representing almost two entire age cohorts for the region, the sample size ($n = 108$) was relatively

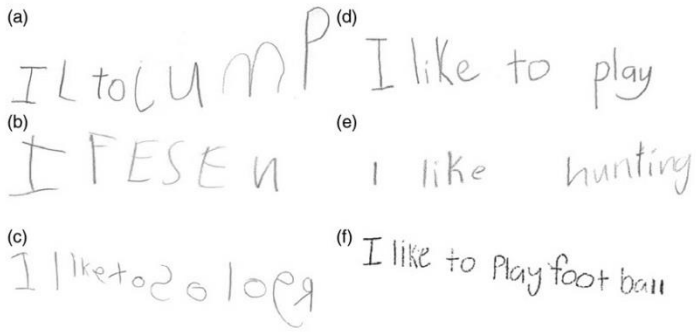

FIGURE 2: *Handwriting samples from the Evaluation Tool of Children's Handwriting Sentence Writing task completed by children with (a) PAE (high exposure), no FASD: 7.11 years; IQ = 82 ('I like to jump'); (b) ND-AE: 8.5 years; IQ = 76 ('I like fishing'); (c) pFAS: 7.11 years; IQ = 61 ('I like to colour'); (d) no PAE: 9.5 years; IQ = 80; (e) no PAE: 9.2 years; IQ = 91; (f) no PAE: 9.5 years; IQ = 88. IQ = Universal Nonverbal Intelligence Test (UNIT) Full Scale Standard Score. The UNIT has a normative M = 100.0, SD = 15.*

small, but still larger than the only other published study of graphomotor performance in children with FASD (Duval-White *et al.*, 2013). The cohort was mostly of Australian Aboriginal descent and living in a remote region of Australia, and while results may be similar to children in other remote regions with comparable demographics, results should not be generalised to other populations.

Validated measures of graphomotor and other fine motor skills do not exist for Australian Aboriginal children, and caution should be used when using assessment tools which have been developed for different populations, especially those with differing cultural contexts (Thorley & Lim, 2011). However, the children in the Lililwan Project had been attending primary school for several years and should have been familiar with the graphomotor requirements of the ETCH and M-FUN.

Many factors contribute to graphomotor performance other than motor skills, including cognition, language, attention, hearing, school attendance and early exposure to fine motor skills (Benbow, 2006; Tomchek & Schneck, 2006). The cohort had high levels of other prenatal exposures and socioeconomic risk factors which may have affected performance. Future studies should explore the impact of these factors and explore whether they differ in children with PAE or FASD.

Implications for occupational therapy practice

This study provides evidence that many children in the region, regardless of PAE, have graphomotor impairment which could interfere with academic performance and participation in cultural and recreational activities, and may benefit from occupational therapy input. Letter legibility rates of less than 76.0%, and word legibility rates of less than 75.0%, indicate the need for therapeutic treatment (Brossard-Racine *et al.*, 2012). This recommendation is based on younger children (Grades 2 and 3) than those in the Lililwan Project, but nevertheless indicates that 37.6% (based on word legibility) to 68.3% (based on letter legibility) of children may benefit from handwriting intervention. Handwriting intervention has shown improvement in skills in other populations (Case-Smith, Weaver & Holland, 2014) and may be of benefit to children in the Fitzroy Valley. The Fitzroy Valley has a population of 4500 people and is currently serviced by two occupational therapists, who provide services across the lifespan via a fortnightly outreach service from Derby, 260 km to the West. Given that this study only included children from two age groups, it is evident that occupational therapy services in the Fitzroy Valley are severely underresourced. It is recommended that (i) therapeutic services, with a focus on early fine motor skill development, particularly handwriting instruction, would be of benefit to children in the Fitzroy Valley with graphomotor impairment; and (ii) functional graphomotor skills should be assessed along with standardised measures of fine motor skills for children with PAE or suspected FASD.

Conclusions

In this study, it was identified that many children in the Fitzroy Valley had poor graphomotor skills, which likely reflects the multitude of neurodevelopmental risk factors, including PAE and FASD, experienced by children in the region. Children with FASD had significantly poorer graphomotor skills than children without PAE, including delayed pencil grasp, heavy writing pressure, being unable to write a sentence and having reduced word legibility. This study adds new evidence to the functional impairments experienced by children with FASD, including those which may impact on successful school performance and function in the classroom. Based on these findings, it is recommended that graphomotor skills should be assessed in populations with high levels of PAE, in addition to other fine motor skills, as they are important components of occupational performance. Graphomotor skills are critical for successful performance of many academic, recreational and cultural activities, and identifying performance challenges will help guide appropriate therapeutic interventions which remediate or accommodate the fine motor impairment associated with PAE.

Acknowledgments

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Fitzpatrick, Marmingee Hand, Carolyn Hartness, Genevieve Hawkes, Lorian Hayes, Dr Samantha Kaiser, Meredith Kefford, Annette Kogolo, Aimee Leong, Denise Macoun, Dr Raewyn Mutch, Juliette O'Brien, Marilyn Oscar, Trine Pedersen, Claire Salter, Charlie Schmidt, Rhonda Shandley, Stanley Shaw, Dr Gemma Sinclair, Julianne Try, Dr Angus Turner, Dr Amanda Wilkins and Harry Yungabun. Special thanks to Dr Jennifer Nash and Ms Dianne Rios for their expertise in scoring the graphomotor samples. This article has been presented, in part, at the Western Australian State Occupational Therapy Association Conference, Perth, 30 October 2015.

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Appendix D Conference and Seminar Presentations

The following presentations have been made which relate to this thesis:

- **Doney, R.** (2015, October). *Fine motor skills of children in remote Western Australia: Are they different in children with Fetal Alcohol Spectrum Disorders?* Western Australian Occupational Therapy Association Conference, Perth, Western Australia.
- **Doney, R.** (2015, September). *Fetal Alcohol Spectrum Disorders and fine motor skills: A population-based study of Aboriginal children in remote Australia.* The Mark Liveris Health Sciences Research Student Seminar, Curtin University, Perth, Western Australia.
- **Doney, R.** (2015, August). *Visual-motor integration impairment and Fetal Alcohol Spectrum Disorder: A population-based study of children in the Fitzroy Valley.* 4th Asia-Pacific Society for Alcohol and Addiction Research & 5th International Drug Abuse Research Society Conference, Sydney, New South Wales.
- **Doney, R.** (2013, November). *Diagnosing Fetal Alcohol Spectrum Disorder in remote Australia: Cross-cultural considerations for neurodevelopmental assessment and diagnosis.* Public Health Association of Australia and Foundation for Alcohol Research and Education Australasian Fetal Alcohol Spectrum Disorders Conference, Brisbane, Queensland.
- Lucas, B. R., & **Doney, R.** (2013, November). *Assessment of soft neurological signs; gross and fine motor skills, visual motor integration and sensory processing in children with pre-natal alcohol exposure or a Fetal Alcohol Spectrum Disorder.* Public Health Association of Australia and Foundation for Alcohol Research and Education Australasian Fetal Alcohol Spectrum Disorders pre-conference FASD diagnostic workshop: Lessons from the Lililwan Project, Brisbane, Queensland.
- **Doney, R.** (2012, August). *Screening of the documentary 'Tristan'.* North West Occupational Therapy Symposium, Broome, Western Australia.
- **Doney, R.** (2012, May). *The Lililwan Project: Assessing children for Fetal Alcohol Spectrum Disorders in the Fitzroy Valley*. Western Australia Developmental Occupational Therapist Group, Perth, Western Australia.

- **Doney, R.**, & Hand, M. (2011, November). *The Lililwan Project: Assessing Fetal Alcohol Spectrum Disorders in the Fitzroy Valley*. Kimberley Principals' Conference, Broome, Western Australia.
- Fitzpatrick, J. P., Carmichael-Olson, H., Salter, C., **Doney, R.**, & Lucas, B.R. (2011, September). *Fetal Alcohol Spectrum Disorders: Practical classroom strategies*. Bayulu School Teacher Professional Development, Fitzroy Valley, Western Australia.
- Fitzpatrick, J. P., Carmichael-Olson, H., Salter, C., **Doney, R.**, & Lucas, B. R. (2011, July). *Fetal Alcohol Spectrum Disorders: Practical Classroom Strategies*. Wangkatjungka School Teacher Professional Development, Fitzroy Valley, Western Australia.
- **Doney, R.** (2011, April). *Fetal Alcohol Spectrum Disorders and the Lililwan Project*. Kimberley Allied Health Forum, Broome, Western Australia.

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E.1 Publication 1

Doney, R., Lucas, B. R., Jones, T., Howat, P., Sauer, K., & Elliott, E. J. (2014). Fine motor skills in children with prenatal alcohol exposure or Fetal Alcohol Spectrum Disorder. *Journal of Developmental and Behavioral Pediatrics, 35*(9), 598-609. doi:10.1097/dbp.0000000000000107

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E.3 Publication 3

Doney, R., Lucas, B. R., Watkins, R. E., Tsang, T. W., Sauer, K., Howat, P., Latimer, J., Fitzpatrick, J. P., Oscar, J., Carter, M., & Elliott, E. J. (2016). Visual-motor integration, visual perception, and fine motor coordination in a population of children with high levels of Fetal Alcohol Spectrum Disorder. *Research in Developmental Disabilities, 55*, 346-357. doi:10.1016/j.ridd.2016.05.009

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Appendix F Statements of Contribution

F.1 Publication 1

To Whom It May Concern

I, Robyn Michelle Doney,

- Conceptualised and designed the systematic review
- Conducted the search strategy and completed the data extraction forms
- Analysed the data and interpreted the findings
- Wrote the manuscript and critically appraised the content

for the publication entitled *Fine motor skills in children with prenatal alcohol exposure or Fetal Alcohol Spectrum Disorder*.

Signature: R. Doney

I, as a Co-Author, endorse that this level of contribution by the candidate indicated above is appropriate.

Barbara R Lucas	Barbara Lucas.
Taryn Jones	Jones.
Peter Howat	PA Howat
Kay Sauer	Kathryn H Sauer
Elizabeth J Elliott	EJE 11.'09

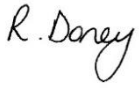
F.2 Publication 2

To Whom It May Concern

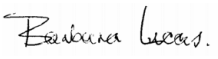
I, Robyn Michelle Doney,

- Conceptualised and designed the fine motor assessments for the Lililwan Project
- Collected the fine motor data using the BOT-2
- Analysed the data and interpreted the findings
- Wrote the manuscript and critically appraised the content

For the publication entitled *Fine motor skills in a population of children in remote Australia with high levels of prenatal alcohol exposure and Fetal Alcohol Spectrum Disorder*.

Signature: 

I, as a Co-Author, endorse that this level of contribution by the candidate indicated above is appropriate.

Barbara R Lucas	
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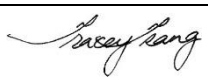
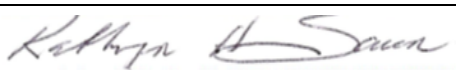
Tracey W Tsang	
Kay Sauer	
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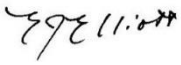
Elizabeth J Elliott	

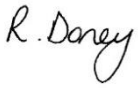
F.3 Publication 3

To Whom It May Concern

I, Robyn Michelle Doney,

- Conceptualised and designed the visual-motor integration assessments for the Lililwan Project
- Collected the visual-motor integration data using the Beery VMI
- Analysed the data and interpreted the findings
- Wrote the manuscript and critically appraised the content

for the publication entitled *Visual-motor integration, visual perception, and fine motor coordination in a population of children with high levels of Fetal Alcohol Spectrum Disorder*.

Signature: 

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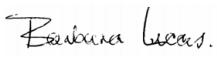
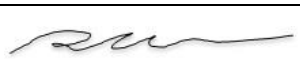
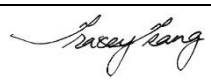
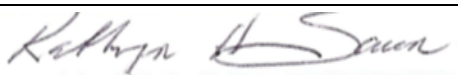
Barbara R Lucas	
Rochelle E Watkins	
Tracey W Tsang	
Kay Sauer	
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James P Fitzpatrick	
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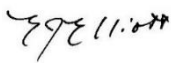
Elizabeth J Elliott	

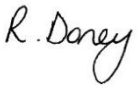
F.4 Publication 4

To Whom It May Concern

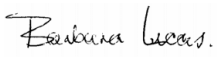
I, Robyn Michelle Doney,

- Conceptualised and designed the graphomotor assessments for the Lililwan Project
- Collected the graphomotor data using the ETCH, M-Fun, and clinical observations
- Analysed the data and interpreted the findings
- Wrote the manuscript and critically appraised the content

for the publication entitled *Graphomotor skills in children with prenatal alcohol exposure and Fetal Alcohol Spectrum Disorder: A population-based study in remote Australia*.

Signature: 

I, as a Co-Author, endorse that this level of contribution by the candidate indicated above is appropriate.

Barbara R Lucas	
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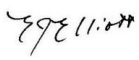
Elizabeth J Elliott	

Appendix G Ethical Approval

The Lililwan Project was conceived, designed, and approved by local leaders in the Fitzroy Valley, who also consented to publication of results. Participating families were provided with verbal and written study information in English and local language if preferred. Participants provided signed consent, and could withdraw at any stage without consequences.

The research was conducted in accordance with the National Health and Medical Research Council National Statement on Ethical Conduct in Human Research (2007) – updated March 2014. Ethics approval for the Lililwan Project was provided by the Kimberley Aboriginal Health Planning Forum Research Sub-committee; University of Sydney Human Research Ethics Committee; Western Australian Aboriginal Health and Information Ethics Committee; and the Western Australian Country Health Services Board Research Ethics Committee. Ethics approval for the research related to the fine motor aspects of the study included in this thesis was provided by the Curtin University Human Research Ethics Committee, Approval Number 172/2010.