

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

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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 ($p = .027$); apply heavy writing pressure ($p = .036$); be unable to write a sentence ($p = .041$); and show poorer word legibility ($p = .041$). 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.

Keywords

Fetal Alcohol Spectrum Disorders; Handwriting; Motor Skills; Psychomotor Performance; Indigenous Population

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 advancements 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 dysmorphism 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.

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 processes 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

1. 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).

2. 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.

3. 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.).

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 (Table 1). Many children had a hearing impairment, and many lived in overcrowded housing and had poor school attendance. The age and proportion of boys and girls in each of the study groups was similar (No PAE age $M = 8.75$ (range 7.11 to 9.70), boys = 55.8%; PAE, no FASD age $M = 8.55$ (range 7.10 to 9.60), boys = 46.2%; FASD $M = 8.40$ (range 7.11 to 9.70), boys = 61.9%). 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 $M = 85.0$ ($SD 12.3$); $p = .329$).

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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 (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 to 16 people). 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.

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); $p = .329$).

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Inter-rater reliability

~~Excellent inter-rater reliability was achieved for the M-FUN Draw a Kid Game ‘Body Awareness: Number of parts’ ($\kappa = 0.84$, 95% Confidence Interval (CI) 0.75–0.93); moderate agreement for ‘Body Awareness: Overall impression’ ($\kappa = 0.44$, CI 0.30–0.58); substantial agreement for ‘Motor Accuracy: Number of parts’ ($\kappa = 0.62$, CI 0.43–0.81); and slight agreement for Motor Accuracy: Overall impression ($\kappa = 0.16$, CI 0.003–0.32).~~

~~For ETCH data, excellent inter-rater reliability was achieved for the scoring of letter legibility in name (ICC = 0.90) and sentence writing tasks (ICC = 0.90), as well as for the scoring of word legibility in the sentence writing task (ICC = 0.91).~~

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 ($p = .027$); more likely to exert heavy pressure when writing ($p = .036$); and less likely to be able to write a sentence ($p = .041$) (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 FASD ($M = 73.9\%$, $p = .008$) (Table 2). Many of the children with FASD (Figure 2, (a) – (c)) had handwriting difficulties which were characterised by difficulties with letter and word formation in comparison to children without PAE (Figure 2, (d) – (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 1, (a) – (c)) showed evidence of developmental immaturity and poor pencil control, especially in comparison to children without PAE (Figure 1, (d) – (f). Inter-rater reliability for the ETCH and M-FUN Draw a Kid Game is reported in Table 3. *(Delete the above sentence if the editor chooses to retain the data in text (results section) rather than table form)*

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 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 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., submitted manuscript-b) and visual motor integration difficulties (Doney et al., submitted manuscript-a) 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 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.

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 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 4500

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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Figure captions

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 $M = 100.0$, $SD = 15$

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 $M = 100.0$, $SD = 15$

Table 1. Cohort characteristics (*ORIGINAL VERSION*)

Characteristics (N = 108)	n (%)
Australian Aboriginal	106 (98.1)
Mean age at assessment (range)	8.7 (7.5-9.6)
Male	57 (52.8)
Right handed	101 (93.5)
Hearing (<i>n</i> = 93) ¹	
Normal	42 (45.2)
Mild loss	38 (40.9)
Moderate loss	13 (14.0)
CSOM (<i>n</i> = 103) ²	45 (44)
CAPD (<i>n</i> = 84) ³	10 (12)
Prenatal nicotine exposure ⁴	
No	34 (31.5)
Yes	67 (62.0)
Unknown	7 (6.5)
Prenatal marijuana exposure ⁴	
No	88 (81.5)
Yes	13 (12.0)
Unknown	7 (6.5)
Prenatal alcohol exposure (PAE) ⁴	
No	43 (39.8)
Yes	60 (55.6)
Unknown	5 (4.6)
Audit-C PAE levels (<i>n</i> = 97) ⁵	
Low (0 - 3)	46 (42.6)
Risky (4 - 5)	4 (3.7)
High risk (≥ 6)	47 (43.5)
FASD	21 (19.4)
FAS	1 (0.9)
pFAS	12 (11.1)
ND-AE	8 (7.4)
Number of homes since birth	
1-3	88 (81.5)
4-5	14 (13.0)
6-10	3 (2.8)
Unknown	3 (2.8)
Number of people in household mean (range)	6.1 (2-16)
Number of schools attended mean (range)	1.8 (1-5)
Frequency of school attendance	
4 – 5 days/ week	89 (82.4)
2 – 3 days/ week	16 (14.8)
1 day/ week	2 (1.9)
Not at all	1 (0.9)

¹ Not all children completed audiology testing

² CSOM: Chronic suppurative otitis media

³ CAPD: Central auditory processing disorder

⁴ Unknown prenatal exposures were due to the primary carer being unaware, or birth mother choosing not to disclose this information

⁵ Risk level according to Alcohol Use Disorders Identification Test – Consumption criteria

Table 1. Cohort characteristics*(ALTERNATIVE TABLE AS SUGGESTED BY REVIEWER 2)*

	Total Cohort ¹	No PAE	PAE (no FASD)	FASD
	<i>N</i> = 108	<i>n</i> = 43	<i>n</i> = 39	<i>n</i> = 21
	<i>n</i> (%)	<i>n</i> (%)	<i>n</i> (%)	<i>n</i> (%)
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 ^{2,3} (<i>n</i> = 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 ⁴				
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 ⁴				
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 ⁵				
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)

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 Not all children completed audiology testing

³ Mild hearing loss 26 – 40dB; moderate hearing loss 41 – 55dB

⁴ Some prenatal exposure information not available, either due to the primary carer not knowing, or the birth mother choosing not to disclose this information

⁵ Risk level according to AUDIT-C scoring criteria

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

	Total Cohort¹ <i>N</i> =108		No PAE <i>n</i> =43		PAE (no FASD) <i>n</i> = 39		FASD <i>n</i> = 21		
CLINICAL OBSERVATIONS	<i>n</i>	(%)	<i>n</i>	(%)	<i>n</i>	(%)	<i>n</i>	(%)	<i>p</i> ²
Hand dominance									
Right	101	(93.5)	41	(95.3)	38	(97.4)	19	(90.5)	.487
Left	7	(6.5)	2	(4.7)	1	(2.6)	2	(9.5)	.487
Pencil grasp									
<i>Primitive grasps</i>	0	0	0	0	0	0	0	0	-
<i>Transitional grasps</i>									
Cross-thumb	47	(43.5)	12	(27.9)	18	(46.2)	13	(61.9)	.027*
Static tripod	32	(29.6)	12	(27.9)	15	(38.5)	4	(19.0)	.271
Four fingers	11	(10.2)	7	(16.3)	3	(7.7)	1	(4.8)	.280
<i>Mature grasps</i>									
Lateral tripod	3	(2.8)	2	(4.7)	0	(0)	1	(4.8)	.390
Dynamic tripod	15	(13.9)	10	(23.3)	3	(7.7)	2	(9.8)	.104
Writing pressure (<i>n</i> = 107)									
Light	0	(0)	0	(0)	0	(0)	0	(0)	-
Light – Appropriate	1	(0.9)	0	(0)	1	(2.6)	0	(0)	.442
Appropriate	49	(45.4)	18	(41.9)	22	(56.4)	8	(38.1)	.310
Appropriate – Heavy	24	(22.2)	12	(27.9)	8	(20.5)	2	(9.5)	.218
Heavy	33	(30.6)	12	(27.9)	8	(20.5)	11	(52.4)	.036*
Handwriting ability									
Unable to write first or surname	3	(2.8)	1	(2.3)	1	(2.6)	1	(4.8)	.851
Unable to write surname	17	(15.7)	8	(18.6)	4	(10.3)	5	(23.8)	.358
Unable to write a sentence	7	(6.5)	2	(4.7)	1	(2.6)	4	(19.0)	.041*
DRAWING (M-FUN) (<i>n</i> = 97)									
	M	(SD)	M	(SD)	M	(SD)	M	(SD)	
Body Awareness ³	5.2	(1.0)	5.3	(1.1)	5.1	(1.0)	5.1	(1.2)	.565
Motor Accuracy ⁴	7.6	(1.6)	7.7	(1.6)	7.6	(1.4)	7.1	(2.0)	.419
Total Score ⁵	12.8	(2.6)	13.0	(2.6)	12.6	(2.3)	12.2	(3.1)	.522
HANDWRITING (ETCH)									
	M	(SD)	M	(SD)	M	(SD)	M	(SD)	
Name: Letter legibility ⁶ (<i>n</i> = 91)	60.8	(26.0)	62.5	(27.1)	60.9	(24.2)	56.1	(28.0)	.729
Sentence: Letter legibility ⁶ (<i>n</i> = 101)	62.9	(22.3)	62.0	(24.4)	63.6	(21.7)	60.4	(21.5)	.872
Sentence: Word legibility ⁶ (<i>n</i> = 101)	73.3	(29.1)	81.0	(22.1)	73.9	(28.6)	50.0	(37.0)	.008**

¹Total Cohort includes *n* =5 children with unknown PAE who were excluded from the group analysis.

²Significance tested for No PAE; PAE (no FASD); and FASD groups.

³ Possible score ranges 0 to 6; ⁴ 0 to 9; and ⁵ 0 to 15.

⁶ Scores indicate percentage of legible letters or words.

* *p* < .005; ** *p* < .001

Table 3: Inter-rater reliability*(NEW TABLE AS PROPOSED BY REVIEWER 2)*

	Weighted Kappa¹	Confidence Interval	Strength of agreement²
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	ICC³		
Letter legibility	0.90	-	Excellent
ETCH: Sentence			
Letter legibility	0.90	-	Excellent
Word legibility	0.91	-	Excellent

¹Weighted kappa with quadratic weights²Strength of agreement based on Landis & Koch (1977) criteria³ ICC = Intra-class correlation coefficient

Figure 1: Drawing samples



(a)



(b)



(c)



(d)



(e)



(f)

Figure 2: Handwriting samples

(a) I like to play

(b) I like to play

(c) I like to play

(d) I like to play

(e) I like hunting

(f) I like to play football