Journal of Population Therapeutics and Clinical Pharmacology

INCORPORATING FETAL ALCOHOL RESEARCH

Journal de la thérapeutique des populations et de la pharmacologie clinique

Fetal Alcohol Research

DOI: 10.22374/1710-6222.25.1.3

ASSESSING MOTOR SKILLS TO INFORM A FETAL ALCOHOL SPECTRUM DISORDER DIAGNOSIS FOCUSING ON PERSONS OLDER THAN 12 YEARS: A SYSTEMATIC REVIEW OF THE LITERATURE

Bernadette Safe, BSc, ¹ Annette Joosten, PhD, ² and Roslyn Giglia, PhD³

¹Telethon Kids Institute, University of Western Australia, Perth, Western Australia. The School of Paediatrics and Child Health, the University of Western Australia, Perth, Western Australia ²School of Occupational Therapy and Social Work, Curtin University Perth, Western Australia ³Alcohol and Pregnancy and FASD, Telethon Kids Institute, University of Western Australia, Perth, Western Australia. NHMRC FASD Research Australia, Centre of Research Excellence, Telethon Kids Institute, University of Western Australia, Perth, Western Australia

Correspondence may be directed to Bernadette.safe@telethonkids.org.au

Submitted: January 19, 2018. Accepted: February 15, 2018. Published: March 1, 2018.

ABSTRACT

Background

Motor impairments are one of the difficulties present in people prenatally exposed to alcohol, and are included in the diagnostic criteria for Fetal Alcohol Spectrum Disorder.

Objectives

The aim of this review was to examine the extent and common types of motor impairment present in persons aged over 12 years prenatally exposed to alcohol as evidence for determining the skills that should be assessed and addressed in intervention.

Methods

A systematic review of current evidence using various electronic databases was conducted. Studies were appraised using a recognized clinical appraisal tool.

Results

Seven studies published between 1998 and 2014 met the inclusion criteria. There is some evidence that difficulties with fine motor skills, visual motor integration, and balance skills persist in people who have been prenatally exposed to alcohol. Most studies did not focus on adolescent or adult participants in isolation,

J Popul Ther Clin Pharmacol Vol 25(1):e25-e38; March 1, 2018. This article is distributed under the terms of the Creative Commons Attribution-Non Commercial 4.0 International License.

making it difficult to generalize results. Varied methodological designs made it difficult to compare studies as few used common standardized assessments.

Conclusion

A review of functional difficulties in each individual would be required to determine if a motor assessment is warranted. Further research is required using assessments recommended in diagnostic guidelines to determine the common motor difficulties seen in adolescents and adults.

Fetal Alcohol Spectrum Disorder (FASD) is a diagnostic term used to describe the lifelong neurological disability directly linked with exposure to alcohol in utero. A recently published systematic review estimated that the global prevalence of FASD in the general population was 7.7 per 1000 people. In Australia FASD is thought to be underdiagnosed and reported; but estimates of FASD (with associated facial features) range from between 0.01 to 1.7 per 1000 live births. The first Australian prevalence study conducted in 2009 in a remote Western Australian community with known high levels of alcohol abuse, found 12% of the children assessed met criteria for Fetal Alcohol Syndrome.

People with FASD can exhibit a range of neurological impairments, which are linked to difficulties including poor school completion, increased contact with the criminal justice system, decreased independent living, mental health issues, and poor peer relationships. 4,5 The Australian criteria for a FASD diagnosis has been modified, and like the Canadian guidelines with which it harmonizes, uses "FASD" as a singular umbrella term.^{6,7} The currently used Australian Guide to the Diagnosis of FASD characterizes FASD as: confirmed prenatal alcohol exposure; the presence (or absence) of three facial features; and impairments across at least three neurocognitive domains of brain structure/neurology, motor skills, cognition, language, academic achievement, memory, attention, executive functions, affect regulation, adaptive behaviour, social skills or social communication. A multidisciplinary team of health professionals is required to assess all domains. Previous diagnostic terms under the FASD umbrella were often related to the number of facial features seen, and included Fetal Alcohol Syndrome (FAS), partial Fetal Alcohol Syndrome (pFAS), Prenatal Exposure to Alcohol (PEA), Alcohol Related

Neurodevelopmental Disorder (ARND), and Fetal Alcohol Effects (FAE). In the *Diagnostic and Statistical Manual of Mental Disorders Fifth Edition (DSM 5)*, FASD currently falls under the diagnostic category of "Other specified neurodevelopmental disorder." In this review, all terms will be used interchangeably depending upon those used in the reported study.

The reported motor impairments associated with prenatal alcohol exposure include difficulties with speed, precision, tremor, postural stability, reaction time, bimanual coordination, visual motor integration, ataxia, and sensorimotor deficits. 9-12 A diagnosis of cerebral palsy is reported to be a comorbidity in 2–10% of people diagnosed with FAS. 13 Motor delays in children with FASD are thought to occur in isolation from the intellectual delays commonly reported. ¹⁴ Motor difficulties have been related to neurological damage to the corpus callosum, cerebellum, motor cortex, and the peripheral nervous system caused by prenatal alcohol exposure. 15 Previous literature reviews have examined these motor difficulties in younger children, including Doney et al. 16 who reviewed fine motor skills in primary school aged children prenatally exposed to alcohol, and found that more complex motor skills including visual motor integration were more impaired, but that functional fine motor skills need further assessment and research. A meta-analysis by Lucas, et al., 17 reviewed the research concerning gross motor skills in children from birth to 18 years of age who were prenatally exposed to alcohol. They found that gross motor skills including balance, coordination, and ball skills were significant affected by prenatal alcohol exposure.

Motor skill development can be affected by multiple factors. A review found that prenatal alcohol was a risk factor for delayed childhood motor performance. ¹⁸ They also reported on studies, which used mixed reliability,

factors affecting motor development, including hereditary factors; exposure to toxic trace metals during pregnancy; prenatal drug use including cannabis; stress during delivery; preterm delivery, low birth weight or perinatal complications. When considering a FASD diagnostic assessment in an adolescent or adult, the drug use of the young person, or any history of head injuries also needs to be considered as a compounding factor impacting on neurological deficits. 7

Studies into the motor impairments seen in those diagnosed with Developmental Coordination Disorder (DCD), indicate that the daily impact of motor difficulties seen in childhood persist into adolescence and adulthood in approximately 50% of cases. ¹⁹ It is thought that some people may have improved motor skills as a result of intervention in childhood, or after extensive practice. They may also employ compensatory strategies including avoiding challenging tasks, or modifying the environment to suit their needs (e.g., using a computer rather than handwriting). ²⁰ It is unknown if this premise would be similar in people who have motor impairment as a result of prenatal alcohol exposure.

Motor impairments among adolescents have been correlated with secondary concerns including social skill problems, lower school achievement, decreased independence in daily living skills, reduced participation in sport and recreation, reduced employment rates, higher anxiety, and reduced self-esteem. ^{21–23} Therefore, motor skills are important to assess.

In recent times, the profile of FASD been raised with growing awareness of its impact and prevalence. More funding is being invested into prevalence and intervention research and into establishing diagnostic clinics in Australia. As a result, it is possible that many people who previously could not access a diagnostic assessment, are now adolescents or adults. Therefore, it is important to know the level of motor impairment expected in these older populations to help determine whether an assessment is warranted. This information can help guide clinicians working in diagnostic teams, or who are providing intervention, about which assessments should be included in the therapeutic process.

This review will investigate the extent and common types of motor impairment reported in persons

aged over 12 years prenatally exposed to alcohol. A systematic review method was chosen to review and critically appraise the articles as it offers a transparent approach for minimizing bias when selecting, appraising and extracting data from articles.²⁵

METHODS

Search Strategy

An initial search was conducted and included the following search terms: motor impairment, motor delay, motor coordination, fine motor, balance, gross motor, visual motor AND fetal alcohol spectrum, fetal alcohol disorder, FAS*. FAS* was used to include FAS and FASD in the search. Many irrelevant articles were discovered, and so the search strategy was refined in Medline and adopted for the other databases which included Medical Subject Heading (MeSH) terms. All publication dates were considered without a time restriction. Age ranges were included in the original search strategy, but were eliminated to allow more studies to be included in the review. All studies were reviewed manually, and references were cross-checked, with some relevant papers followed up and added.

Databases that were accessed included Web of Knowledge, Ovid (Medline in-process, other non-indexed citations; Embase), EBSCOhost (Cumulative Index to Nursing and Allied Health Literature), The Cochrane Library, Google Scholar, and OT Seeker over August and September 2016. Hand searching was also conducted from references used in selected articles. The search was re-run in September 2017, and no new articles were found.

Study Selection

Titles and abstracts were reviewed by the first author to determine their eligibility (see Table 1 for inclusion and exclusion criteria). Full text articles that were identified as eligible or unclear were retrieved and reviewed in full.

The PRIMSA checklist was used to guide the literature review. A total of 215 articles were found across databases; 83 were duplicates and removed. The remaining 132 (plus three that were added from reference lists of articles) were screened, and assessed for inclusion criteria based on the title, abstract or after reviewing the article in entirety. Articles were commonly

FIG. 1 Example of Search Strategy used in Medline

1.	Fetal Alcohol Spectrum Disorders/
2.	fetal alcohol syndrome.mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
3.	1 or 2
4.	(motor impairment or motor skills or motor delay or fine motor or gross motor or visual motor).mp. [mp=title, abstract, original title, name of substance word, subject heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier]
5.	Motor Skills Disorders/
6.	apraxias/ or apraxia, ideomotor/ or gait apraxia/
7.	4 or 5 or 6
8.	3 and 7
9.	limit 8 to (english language and humans)

TABLE 1 Inclusion and Exclusion Criteria

	Inclusion Criteria	Exclusion Criteria
Time	No restriction	Search concluded September 2016 and rerun September 2017
Age of participants	Studies that included participants 12 years and older (no upper limit on age was set)	Studies with only children younger than 12 years old
Language	English	All other languages
Participants	Humans	Animals
Design	Randomized Control Trials Cohort Studies Case-Control studies	Descriptive Reviews Case studies
Prenatal alcohol	Confirmed	Not confirmed
Motor assessment	Direct assessment by clinician Published, commonly used standardised assessments Parent or other informant report	Observational data Non-standardised or uncommonly used assessments (e.g., computer or robotic)

excluded due to reasons of: absence of prenatal alcohol exposure; animal studies; literature reviews; intervention studies, studies referring to other prenatal drug exposures; assessments used that are not commonly available to clinicians (e.g., non-standardized robotic or computerized tasks); and participants not included in the age range for this review. The original inclusion criteria were solely participants of any age over 12 years-old; however, this only generated one study. As a result, the criteria were widened to include studies with participants older than 12 years but who also had some participants under this age. These studies were sorted manually. Following exclusion, seven articles remained that met the inclusion criteria.

Data Analysis

The seven articles that met the inclusion criteria were read and key data were extracted into tables. The primary outcomes that were measured included any type of motor function such as fine or gross motor coordination, bilateral coordination, balance, and motor speed. The National Health and Medical Research Council (NHMRC) Evidence Hierarchy was used to guide the strength of the evidence. ²⁶ The McMaster University Critical Review Form- Quantitative Studies, ²⁷ was used as the quality rating tool as

is freely available, used across varied research areas to evaluate a range of research designs (e.g.²⁸). The data from the included articles was reviewed by an additional assessor.

RESULTS

The search identified a total of 215 articles. Using the inclusion and exclusion criteria, seven articles were eligible for review. Participants across studies ranged from 5–39 years-old.

Details about each article were recorded including author, sample size, type of study, age of participants, NHMRC level of evidence, assessment tools used, and findings. Full details are presented in Table 2.

All studies apart from one used matched controls based on age, gender and ethnicity. One study aimed to determine if intellectual ability had an effect on motor performance, so the control group was matched for Intellectual Quotient (IQ) levels.²⁹ Usually IQ was reported as lower in the prenatal alcohol exposed group; which is in line with research findings that on average, children with FASD have IQ ability in the low-average range.³⁰

One study was a longitudinal study and compared motor performance in the same person when they were

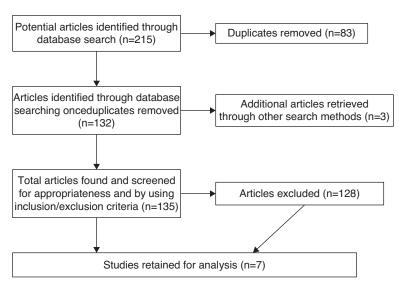


FIG. 2 PRISMA Flow Diagram

J Popul Ther Clin Pharmacol Vol 25(1):e25-e38; March 1, 2018. This article is distributed under the terms of the Creative Commons Attribution-Non Commercial 4.0 International License.

 TABLE 2 Review of Articles

Criteria	Yes	Yes
Outcome	Hand Dynamometer (dominant); Finger Tapping (dominant); VMI; GPB (dominant);	Clinical Strength, Range of Motion, and Posture Screen; MABC-2; SPM; Pediatric Clinical Test of Sensory Interaction for Balance-2; MABC-2 Checklist; DGI; MultiModal Balance Entrainment Response system
Diagnoses	ARND (based on the Institute of Medicine Criteria -PAE and Significant impairment in a minimum of two areas)	FASD (FAS, pFASD, static encephalopathy/alcohol-exposed, and neurobehavioral disorder/alcohol-exposed)
Matched	no, but norms used	yes
Was age ranges separated?	yes	no
Age range	5-17:9 years	8.0-15.9 years
Sample	117	20
Motor	visual motor; fine motor	balance
Prenatal alcohol confirmation	yes	yes
NHMRC Level of evidence	V	III-3
Reference	Tamana, Pei, Massey, Massey, Rasmussen ³²	Jirikowic, McCoy, Lubetzky- Vilnai, Price, Ciol, Kartin, Hsu, Gendler, Astley ⁴¹

Criteria	Yes	Yes	Yes
Outcome measures	VMI; GPB	GPB	DNCT; FS; HST; DB
Diagnoses studied	Matched studied studied studied studied studied Search FAS and PAE and pFAS (modified Institute of Medicine Criteria usednumber of number of neurological impairments required for diagnosis was not stated)		FAS, FAE, ARND
Matched			yes
Was age ranges separated?	ranges separated?		All older age range
Age range	6-16 years	7-17 years	Longitudinal study: 25-27 years; Clinical sample: 18-37 years
Sample size	Sample size 110		1) Longitudinal study = 402.2) Clinical sample = 90
Motor Category	Motor Category visual motor; fine motor visual motor; fine motor		fine motor, balance
Prenatal alcohol confirmation	Prenatal alcohol confirmation yes yes; but also 20% reported some and infrequent alcohol use in the control group		yes
NHMRC Level of evidence	NHMRC Level of evidence III-3 III-3		III-3 and II
Reference	Vaurio, Riley, Mattson ²⁹	Aragon, Kalberg, Buckley, Barela- Scott, Tabachnick, May ⁴⁰	Connor, Sampson, Streissguth, Bookstein, Barr ³¹

Criteria met		
Criteri	Yes	Yes
Outcome measures	Outcome measures WMI; GPB (dominant); computerized version of the Bimanual Coordination Test	
Diagnoses studied	PAE and FAS	PAE and FAS
Matched	yes	yes
Was age ranges separated?	no	no
Age range	10-19 years	5-16 years
Sample size	38	50
Motor Category	bilateral coordination; visual motor; fine motor	visual motor; fine motor
NHMRC Prenatal Level of alcohol evidence confirmation	yes	yes
NHMRC Level of evidence	III-3	III-3
NHMRC Reference Level of evidence	Roebuck- Spencer, Mattson, Marion, Brown, Riley ¹²	Mattson, Riley, Gramling, Delis, Jones ³⁸

 $VMI = Beery\ Buktenica\ Developmental\ Test\ of\ Visual-Motor\ Integration;\ GPB = Grooved\ Peg\ Board\ Test;\ MABC-2 = Movement\ Battery\ for\ Children-2nd\ edition;\ SPM = Sensory\ Processing\ Measure;\ DGI = Dynamic\ Gait\ Index\ Test;\ DNCT = Denckla\ Neurological\ Coordination\ Test;\ FS = Finger\ Sequencing\ Test;\ HST = Hand\ Steadiness$ Test; DB = dynamic balance.

NHMRC Levels of Evidence²³: IV = cross-sectional study; III-3 = case-control study; II = prospective cohort.

a child, and again when they were an adult.³¹ Five studies were case-control studies; and the remaining one was a cross-sectional design using normative data from standardized assessments.³² There was only one study that focused solely on adolescent or adult participants. Six of the studies included younger children outside of the original inclusion criteria age range, and only one of these six analyzed the participants for any age-related differences.

Although some studies stated that information about additional prenatal toxic exposures was gathered during assessment, they did not report the prevalence or discuss this as a factor in the results. Only one study reported that the control group was exposed to cigarettes (11%) and marijuana (4%) during pregnancy.²⁹

Most commonly used motor skill assessments included the Beery-Buktenica Test of Visual Motor Integration (VMI)³³ and Grooved Peg Board (GPB).³⁴ The Sensory Processing Measure,³⁵ Movement Assessment Battery for Children, Second Edition (MABC-2) - Parent Checklist,³⁶ and Dynamic Gait Index (DGI)³⁷ were used in one study. Often the research reported on motor assessments as secondary results, with the primary outcome focusing on other neurocognitive areas.

Two of the studies found that only those with facial features commonly seen in people with FASD did significantly worse on the VMI and fine motor tasks compared to the control group. However, other studies that separated the FASD group into different diagnostic categories depending on the number of facial features, found similar motor results between the different groups. Results are reported for fine motor skills, VMI and balance.

Fine Motor Skills

Mattson et al.³⁸ reported that when 50 children and adolescents were assessed using the GPB (dominant hand), results were not significantly different between the control group and those diagnosed with PEA or FAS. However, when assessed using the GPB (nondominant hand), results showed a significant reduction in speed in both PEA and FAS groups compared with the control group. Aragon et al.⁴⁰ had a similar sample size and age range, and also used the GPB to assess motor skills. Although GPB (non-dominant hand)

scores were lower in the FASD group compared to the controls, the difference was not statistically significant. Important to note is that in this study approximately 20% of the control group's mothers reported some alcohol consumption during pregnancy, which may have had an effect on the control participant's neurological profiles. In a larger sample size (n=110) of 6 to 16-year-olds with IQ matched controls, the GPB test results showed no significant difference between the FASD group and the control group's performance.²⁹ All three of the aforementioned studies included both younger children and adolescents with no indication of the number of children in each age group, or separation of any age groups for statistical analysis.

Tamana et al.³² completed a cross-sectional study of 117 young people diagnosed with FASD, comparing groups within three different age ranges (5–8, 9–12, 13-17 years). No control group was included, with results compared with normative assessment data. Participants in the older group performed poorer than younger participants on the hand dynamometer (to assess hand strength), finger tapping (to assess fine motor speed), and GPB tasks. Connor et al.³¹ completed a longitudinal study of 402 participants (25-27 years-old), alongside a clinical study of participants aged between 18 and 37 years. In the clinical study, three-quarters of the subjects with FASD demonstrated more difficulties with motor function compared to the controls. People with FASD were less accurate and slower on a hand steadiness task. In the longitudinal sample, only subjects who had been identified in childhood as possibly having FAS (FASD with all three associated facial features) had difficulties with motor tasks, relative to comparison subjects. They concluded that those who were exposed to high levels of alcohol prenatally, and had other neurocognitive impairments, had motor deficits that persisted into adulthood.

Visual Motor Integration

Mixed results were reported for visual motor integration skills using the VMI assessment. Vaurio et al.²⁹ found a significant difference between results among the FASD group compared to cognitive matched controls. This aligned with results from Tamana et al.³² who also found that the VMI difficulties in participants

with FASD were independent of cognitive functioning. In this study, the participants in the older age range who had FASD had more difficulty completing the VMI compared to younger participants. They concluded that deficits in visual motor abilities may become more obvious with age. However, as this was a cross-sectional, rather than longitudinal study these results need to be interpreted with caution. Mattson et al.³⁸ found that VMI results in the FAS group were significantly poorer than those of the control group. The PEA group did not perform as well as the controls but there was no statistically difference. They concluded that visual motor difficulties were poorer in those diagnosed with FAS. Roebuck-Spencer et al. 12 assessed 10 to 19-year-olds and found that children with FASD were slower than the controls to compete the VMI assessment, but were equally accurate on basic visual motor tasks. They found no statistical differences between the age groups during analysis.

Balance

Jirikowic et al.41 assessed balance skills in 20 participants aged 8-16 years-old. In standardized caregiver questionnaires (Sensory Processing Measure and Movement Assessment Battery for Children (MABC-2) - Parent Checklist), carers who had a child diagnosed with FASD identified more functional balance concerns. During a clinical balance test, children with FASD were more reliant on vision to support balance. There was a significant difference during the Dynamic Gait Index (DGI) test, with the children with FASD achieving lower scores; however, as this tool is not normed on this age range, it is unknown if the scores were clinically meaningful. No significant difference between groups was seen during the clinical balance tasks of the MABC-2. As the proportion of participants in different age groups was not documented, it is expected that the sample of adolescents was very small, and results cannot be generalized.

Connor et al.,³¹ in a larger, longitudinal study of adults, used the Dynamic Balance Test. They found that participants with FAE and FAS had poorer balance compared to the control group in the clinical study. FAE and FAS groups had similar balance scores. These results demonstrate that balance difficulties may persist into adulthood in those prenatally exposed to alcohol.

DISCUSSION

The aim of this systematic review was to explore the available evidence for motor skill impairment and functioning in older children or adults who were prenatally exposed to alcohol. Although the focus was on adolescents or adults, only two of the seven studies analyzed the results of this group in isolation from results for children. As a result, it is difficult to make conclusions about motor skills in an adolescent or adult population. Most studies, aside from one, excluded any prenatal alcohol exposure from control groups. Factors including additional prenatal drug exposures. personal drug misuse, and head trauma histories, are confounding factors for motor performances across the lifespan.^{7,18} However, no study adequately captured or discussed additional risk factors for motor impairment during the prenatal period, birth or other lived experiences. Many FASD umbrella terms were considered. The varied diagnostic criteria and terms used between studies meant that comparing results to make definitive conclusions was difficult. Most studies relied on maternal disclosure of prenatal alcohol consumption quantity. The reliability of this information is likely questionable given the passage of time since pregnancy and the stigma associated with drinking alcohol during pregnancy leading to underreporting. 42 Details about alcohol quantity and timing during the prenatal period were not disclosed in the studies, so conclusions relating to this are not made in this review.

The VMI and GPB were commonly used in the studies that were analyzed. The VMI is commonly used clinically by occupational therapists or psychologists and is recommended in the Australian Guide to the Diagnosis of FASD.⁷ The GPB test is available to clinicians, but norms are outdated, it is rarely used in isolation, and it is not recommended in the diagnostic guidelines. Along with the VMI, other assessment recommended by The Australian Guide to the Diagnosis of FASD include the MABC-2, or the Bruininks-Oseretsky Test of Motor Proficiency, Second Edition. No studies were found which used either of these assessments to determine a motor functioning score (with the exception of one which used the balance subtests of the MABC-2⁴¹). The motor tasks used

and analyzed in the identified studies would not be sufficient to assess a person for a motor impairment using these guidelines,⁷ so the level of impairment may be underrepresented.

One article included caregiver questionnaires to determine any difficulties with functional motor tasks. 41 Otherwise, no studies directly assessed functional motor tasks such as handwriting. Functional ramification of motor skill difficulties in adolescence or adults are poorly researched and understood. The Australian Guide to the Diagnosis of FASD, 7 recommends occupational therapists or physiotherapist be involved in the assessment of motor skills. However, no articles discussed whether these professionals were involved in the assessment; which questions the quality of the assessment and credibility of the reported result's ability to draw conclusions about motor skills.

Most studies assessed motor skills as part of a larger neurological assessment therefore weakening the statistical power of these studies to detect differences in motor functioning between the matched control groups. This limits the ability to make conclusions about clinical differences in motor functioning between people who are prenatally exposed to alcohol, from those who have no exposure.

There is some evidence that adolescents with FASD have difficulties with more complex motor tasks compared to control groups. For example, the non-dominant GPB test was more challenging, and difficulties with complicated shape copying during the VMI was seen in adolescent participants in some studies. 12,38,40 This could explain why older children did not perform as well on the VMI as reported by Tamana et al.³² Visual motor integration skills require aspects of executive functioning such as planning and problem solving. Executive functioning is thought to become more difficult for a person with prenatal alcohol exposure as they move into their adolescent years, 43 so changes in VMI may be in line with this finding. However, in another study, participants with FASD required longer to complete visual motorbased tasks, but were equally accurate as the control group. 12 Research by Connor et al., 31 and Tamana et al.³² showed that difficulties with fine motor speed and accuracy were still present in adolescents and adults, and therefore should be routinely assessed during a FASD assessment. When analyzed, motor skill difficulties are seen independently of intellectual abilities, so should be tested in addition to cognitive assessment.

Other studies have shown that simple motor task difficulties seen in earlier childhood may diminish in adolescence. 44 Although animal studies were not considered in the inclusion criteria for studies in this review, this is supported by some rat studies that have shown that motor training can support neuroplasticity, and diminish the effects of prenatal alcohol exposure on motor skills. 45,46 Further studies in children using non-standardized computer assessment have also shown that motor timing and motor reach accuracy difficulties diminish as a child gets older. 46,47 Adolescents with FASD required additional time to process increasing amounts of information, but the time required to elicit a motor response was similar to the performance of the control group; and older children were able to make straighter reaches than younger children compared with age matched control groups.⁴⁷ Motor skills, particularly balance, has been linked to poor sensorimotor processing (visual, vestibular, and somatosensory) for people affected by prenatal alcohol exposure. This aspect of motor function needs further exploration about whether these difficulties play a role in older childhood.

Like research into DCD, it may be that the functional motor impairments seen after prenatal alcohol exposure become less problematic due to motor practice or utilization of compensatory strategies. As adolescents have had years of exposure to motor skill practice through everyday activities at school and recreationally, simple motor challenges seen in early childhood may be reduced. However, this is not consistently seen across all studies, and highlights the gaps in knowledge about the impact of age of motor skill development.

Limitations

There are limitations in this systematic literature review which make generalizing the results to older children or adult cohorts with prenatal alcohol exposure difficult. These include the fact that only seven studies met the inclusion criteria, and sample sizes were usually small. As only one study was found that focused solely on children over 12 years of age,

it was necessary to expand the inclusion criteria to also include studies that included participants who were younger than 12 years of age. Most studies did not separate an older childhood group from younger participants, so it is difficult to make conclusions for adolescents and adults. Study participants may have been exposed to additional confounders affecting motor skills, which were not reported. As the research into FASD is relatively new, knowledge is evolving regularly, so comparing studies across the years is challenging. For example, the varied FASD diagnostic criteria employed.

The fact that no study assessed functional motor skills, except one which included a caregiver questionnaire; and that the motor assessments were often not standardized, suggests that motor impairments may be underestimated. Larger cohort studies would be required to follow up young people with prenatal alcohol exposure and determine how motor impairments develop through their lifetime.

CONCLUSION

This systematic review of the research literature regarding the motor profile of persons aged over 12 years with FASD found limited reliable evidence to indicate what skills should be routinely assessed as part of a FASD assessment. The differences in study designs, varied diagnostic criteria and participant age ranges; as well as assessment tools with poor clinical application, meant that making definitive conclusions about motor skills in older children with a history of prenatal alcohol exposure is impossible. There is conflicting research regarding whether the motor impairments commonly seen after prenatal alcohol exposure diminish or become more obvious with increasing age in childhood. Evidence suggests that VMI, fine motor and balance skills may continue into adolescence, but potentially only when functional motor concerns are identified.

As funding sources and public knowledge about FASD grows, many people may seek a diagnosis later in life, so an understanding of their expected motor profile is important to ensure accurate assessment and diagnosis. More research is needed including assessing motor skills in older children and adults using recommended standardized assessments by relevant

disciplines. As motor skills may affect a young person's success at school, employment, recreation, and their social abilities, early assessment and intervention is critical.

ACKNOWLEDGEMENTS

Professor Carol Bower for her encouragement and for sharing her wealth of research knowledge, and the clinical team on the FASD Banksia Hill Detention Centre research project. This work was supported by the NHMRC FASD Research Australia Centre of Research Excellence (#1110341).

REFERENCES

- Lange S, Probst C, Gmel G, et al. Global prevalence of fetal alcohol spectrum disorder among children and youth: a systematic review and meta-analysis. JAMA Pediatr 2017;171:10:948–56. doi: 10.1001/ jamapediatrics.2017.1919.
- Burns L, Breen C, Bower C, O' Leary C, Elliott EJ. Counting fetal alcohol spectrum disorder in Australia: the evidence and the challenges. Drug Alcohol Rev 2013;32:461–7. doi: 10.1111/dar.12047.
- 3. Fitzpatrick JP, Latimer J, Carter M, Oscar J, Ferreira ML, Carmichael Olson H, et al. Prevalence of fetal alcohol syndrome in a population-based sample of children living in remote Australia: The Lililwan Project. J Paediatr Child Health 2015;51:450–7. doi: 10.1111/jpc.12814.
- Clark E, Lutke J, Minnes P, Ouellette-Kuntz H. Secondary disabilities among adults with fetal alcohol spectrum disorder in British Columbia. J FAS Int 2004;2:1–12.
- Streissguth A, Bookstein F, Barr H, Sampson P, O'Malley K, Young J. Risk factors for adverse life outcomes in fetal alcohol syndrome and fetal alcohol effects. J Dev Behav Pediatr 2004;25:228–38.
- Cook JL, Green CR, Lilley CM, Anderson SM, Baldwin ME, Chudley AE, et al. Fetal alcohol spectrum disorder: a guideline for diagnosis across the lifespan. CMAJ 2016;188:191. doi: 10.1503/cmaj.141593.
- Bower C, Elliott EJ. On behalf of the Steering Group. Report to the Australian Government Department of Health: Australian Guide to the diagnosis of Fetal Alcohol Spectrum Disorder (FASD)."2016. Available at: https://alcoholpregnancy.telethonkids.org.au/contentas sets/6bfc4e8cd1c9488b998d50ea4bff9180/australianguide-to-diagnosis-of-fasd all-appendices.pdf.

- American Psychiatric Association. DSM-5 Diagnostic Classification. Diagnostic and Statistical Manual of Mental Disorders. DSM Library. Arlington, VA.: American Psychiatric Association; 2013.
- Roebuck TM, Simmons RW, Richardson C, Mattson SN, Riley EP. Neuromuscular responses to disturbance of balance in children with prenatal exposure to alcohol. Alcohol Clin Exp Res 1998;22:1992–7.
- Jirikowic T, Hsu LY, McCoy SW, et al. Clinical balance responses to sensorimotor training to affect balance for childrenwith fetal alcohol spectrum disorders. Alcohol Clin Exp Res 2014;38:179A.
- 11. Adnams CM, Kodituwakku PW, Hay A, et al. Patterns of cognitive-motor development in children with fetal alcohol syndrome from a community in South Africa [corrected] [published erratum appears in Alcoholism 2001 Aug;25(8):1187]. Alcohol Clin Exp Res 2001;25:557–62 6p.
- 12. Roebuck-Spencer TM, Mattson SN, Marion SD, Brown WS, Riley EP. Bimanual coordination in alcohol-exposed children: Role of the corpus callosum. J Int Neuropsychol Soc 2004;10:536–48.
- 13. Abel EL. Cerebral palsy and alcohol consumption during pregnancy: Is there a connection? Alcohol Alcohol 2010;45:592–4. doi: 10.1093/alcalc/agg063.
- 14. Kalberg WO, Provost B, Tollison SJ, et al. Comparison of motor delays in young children with fetal alcohol syndrome to those with prenatal alcohol exposure and with no prenatal alcohol exposure. Alcohol Clin Exp Res 2006;30:2037.
- 15. Bookstein FL, Streissguth AP, Sampson PD, Connor PD, Barr HM. Corpus callosum shape and neuropsychological deficits in adult males with heavy fetal alcohol exposure. Neuroimage 2002;15:233–51.
- Doney R, Lucas BR, Jones T, et al. Fine motor skills in children with prenatal alcohol exposure or fetal alcohol spectrum disorder. J Dev Behav Pediatr. 2014;35:598–609. doi: 10.1097/DBP.000000000000107.
- 17. Lucas BR, Latimer J, Pinto RZ, et al. Gross motor deficits in children prenatally exposed to alcohol: a meta-analysis. Pediatrics 2014;134:192–209. doi: 10.1542/peds.2013-3733.
- 18. Golding J, Emmett P, Iles-Caven Y, Steer C, Lingam R. A review of environmental contributions to childhood motor skills. J Child Neurol 2014;29:1531–47. doi: 10.1177/0883073813507483.

- 19. Cousins M, Smyth MM. Developmental coordination impairments in adulthood. Hum Mov Sci 2003;22:433–59.
- 20. Kirby A, Edwards L, Sugden D. Emerging adulthood in developmental co-ordination disorder: Parent and young adult perspectives. Res Dev Disabil 2011;32:1351-60. doi: 10.1016/j.ridd.2011.01.041.
- 21. Hands B, Licari M, Piek J. A review of five tests to identify motor coordination difficulties in young adults. Res Dev Disabil 2015;41–42:40-51. doi: 10.1016/j. ridd.2015.05.009.
- Skinner RA, Piek JP. Psychosocial implications of poor motor coordination in children and adolescents. Hum Mov Sci 2001;20:73–94.
- 23. Eggleston M, Hanger N, Frampton C, Watkins W. Coordination difficulties and self-esteem: A review and findings from a New Zealand survey. Aust Occup Ther J 2012;59:456-62. doi: 10.1111/1440-1630.12007.
- 24. Foundation for Alcohol Research and Education. National FASD Clinic and Clinical Network Plan. 2015. Available at: http://fare.org.au/2015/11/push-for-national-fasd-clinic/
- 25. Adam K, Peters S, Chipchase L. Knowledge, skills and professional behaviours required by occupational therapist and physiotherapist beginning practitioners in work-related practice: A systematic review. Aust Occup Ther J 2013;60:76–84. doi: 10.1111/1440-1630.12006.
- 26. National Health and Medical Research Council. NHMRC Levels of Evidence and Grades for Recommendations for Developers of Guidelines. 2009. Available at: https:// www.nhmrc.gov.au/_files_nhmrc/file/guidelines/developers/nhmrc_levels_grades_evidence_120423.pdf.
- 27. Law M, Stewart D, Pollock N, Letts L, Bosch J, Westmorland M. Critical Review Form Quantitative Studies: McMaster University; 1998. Available at: https://www.unisa.edu.au/Global/Health/Sansom/Documents/iCAHE/CATs/McMasters_Quantitative review.pdf.
- 28. Eagers J, Franklin R, Broome K, Yau M. A review of occupational therapy's contribution to and involvement in the work-to-retirement transition process: An Australian perspective. Aust Occup Ther J 2016;63:277–92. doi: 10.1111/1440-1630.1230.
- 29. Vaurio L, Riley E, Mattson S. Neuropsychological Comparison of Children with Heavy Prenatal Alcohol Exposure and an IQ-Matched Comparison Group. J Int Neuropsychol Soc 2011;17:463–73. doi: 10.1017/S1355617711000063.
- 30. Streissguth A. Offspring Effects of prenatal alcohol exposure from birth to 25 years: The Seattle Prospective

- Longitudinal Study. J Clin Psychol Med Settings 2007;14:81–101. doi: 10.1007/s10880-007-9067-6.
- 31. Connor P, Sampson P, Streissguth A, Bookstein F, Barr H. Effects of prenatal alcohol exposure on fine motor coordination and balance: A study of two adult samples. Neuropsychologia 2006;44:744–51.
- 32. Tamana S, Pei J, Massey D, Massey V, Rasmussen C. Neuropsychological impairments and age-related differences in children and adolescents with fetal alcohol spectrum disorders. J Popul Ther Clin Pharmacol 2014;2:e167–e80.
- 33. Beery K, Beery N. The Beery-Buktenica Development Test of Visual-Motor Integration: administration, scoring and teaching manual. 6th ed. Minneapolis, MN: Pearson; 2010.
- 34. Matthews CG, Klove H. Instruction Manual for the Adult Neuropsychological Test Battery. Madison, WI.: University Wisconsin Medical School; 1964.
- 35. Parham L, Ecker C, Miller Kuhanek H, Glennon T. Sensory Processing Measure Manual. Los Angeles: Western Psychological Services; 2007.
- 36. Henderson A, Sugden D, Barnett A. Movement Assessment Battery for Children (Movement ABC-2). 2nd ed. London, UK.: The Psychological Corporation; 2007.
- 37. Shumway-Cook A, Woollacott M. Motor Control: Translating Research into Clinical Practice. 3 ed. Philadelphia: Lippincott, Williams, & Watkins; 2007.
- 38. Mattson SN, Riley EP, Gramling L, Delis DC, Jones KL. Neuropsychological comparison of alcohol-exposed children with or without physical features of fetal alcohol syndrome. Neuropsychology 1998;12:146–53.
- 39. Connor PD, Sampson PD, Streissguth AP, Bookstein FL, Barr HM. Effects of prenatal alcohol exposure on fine motor coordination and balance: A study of two adult samples. Neuropsychologia 2006;44:744–51.

- 40. Aragon AS, Kalberg WO, Buckley D, Barela-Scott LM, Tabachnick BG, May PA. Neuropsychological study of FASD in a sample of American Indian children: processing simple versus complex information. Alcohol Clin Exp Res 2008;32:2136–48. doi: 10.1111/j.1530-0277.2008.00802.x.
- 41. Jirikowic T, McCoy S, Lubetzky-Vilnai A, et al. Sensory control of balance: A comparison of children with fetal alcohol spectrum disorders to children with typical development. J Popul Ther Clin Pharmacol 2013;20:e212–e28.
- 42. Benz J, Rasmussen C, Andrew G. Diagnosing fetal alcohol spectrum disorder: History, challenges and future directions. Paediatr Child Health 2009;14:231–7.
- Rasmussen C, Bisanz J. executive functioning in children with fetal alcohol spectrum disorders: profiles and agerelated differences. Child Neuropsychol 2009;15:201–15. doi: 10.1080/09297040802385400.
- 44. Simmons RW, Thomas JD, Levy SS, Riley EP. Motor response programming and movement time in children with heavy prenatal alcohol exposure. Alcohol 2010;44:371–8. doi: 10.1016/j.alcohol.2010.02.013.
- 45. Klintsova AY, Matthews JT, Goodlett CR, Napper RM, Greenough WT. Therapeutic motor training increases parallel fiber synapse number per Purkinje neuron in cerebellar cortex of rats given postnatal binge alcohol exposure: preliminary report. Alcohol Clin Exp Res 1997;21:1257–63.
- 46. Klintsova AY, Goodlett CR, Greenough WT. Therapeutic motor training ameliorates cerebellar effects of postnatal binge alcohol. Neurotoxicol Teratol 2000;22:125–32.
- 47. Simmons RW, Levy SS, Riley EP, Madra NM, Mattson SN. Central and peripheral timing variability in children with heavy prenatal alcohol exposure. Alcohol Clin Exp Res 2009;33:400–7. doi: 10.1111/j.1530-0277.2008.00849.x.