Research in Developmental Disabilities 122 (2022) 104168

Contents lists available at ScienceDirect

Research in Developmental Disabilities

journal homepage: www.elsevier.com/locate/redevdis

Review article

Fetal alcohol spectrum disorders screening tools: A systematic review

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ARTICLE INFO

Number of reviews completed is 2

Keywords:
Fetal alcohol syndrome
Alcohol
Pregnancy
Developmental disability
Biomarker

ABSTRACT

Background: Screening facilitates the early identification of fetal alcohol spectrum disorder (FASD) and prevalence estimation of FASD for timely prevention, diagnostic, and management planning. However, little is known about FASD screening tools.
Aims: The aims of this systematic review are to identify FASD screening tools and examine their performance characteristics.
Methods: Four electronic databases were searched for eligible studies that examined individuals with FASD or prenatal alcohol exposure and reported the sensitivity and specificity of FASD screening tools. The quality of the studies was assessed using the Quality Assessment of Diagnostic Studies-2 tool.
Results: Sixteen studies were identified, comprising five fetal alcohol syndrome (FAS) and seven FASD screening tools. They varied in screening approach and performance characteristics and were linked to four different diagnostic criteria. FAS screening tools performed well in the identification of individuals at risk of FAS while the performance of FASD screening tools varied in the identification of individuals at risk of FASD.
Conclusion and implications: Results highlight the vast differences in the screening approaches performance characteristics, and diagnostic criteria linked to FASD screening tools. More research is needed to identify biomarkers unique to FASD to guide the development of accurate FASD screening tools.

What does this paper add?

Identification of FASD enables at-risk individuals to receive timely management and support. Screening can facilitate the identification of FASD. However, there are several challenges to screening for FASD, including a limited understanding of FASD screening tools. This systematic review is the first to report synthesised information about FASD screening tools, including their performance characteristics, interpretation of screen test results, and scalability. This review provided new knowledge about the wide differences in the screening approaches, performance characteristics and diagnostic criteria linked to FASD screening tools.

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https://doi.org/10.1016/j.ridd.2021.104168
Received 1 August 2021; Received in revised form 27 December 2021; Accepted 28 December 2021
Available online 4 January 2022

1. Introduction

Fetal alcohol spectrum disorder (FASD) involves severe neurodevelopmental impairments associated with prenatal alcohol exposure (PAE) (Bower et al., 2016; Chudley et al., 2005). Established diagnostic categories of FASD include fetal alcohol syndrome (FAS), partial FAS (pFAS), alcohol-related neurodevelopmental disorder (ARND), alcohol-related birth defects (ARBD), and neurobehavioural disorder associated with prenatal alcohol exposure (ND-PAE) (American Psychiatric Association, 2013; Bower et al., 2016; Hoyme et al., 2016). These neurodevelopmental impairments experienced by individuals with FASD impact many aspects of their cognitive and social functioning (Bower et al., 2016; Millar et al., 2017; Millians, 2015; Pei, Denys, Hughes, & Rasmussen, 2011). The impact of FASD also extends to families of individuals with FASD and the community; some families experience stigma associated with FASD (Corrigan et al., 2019) and some communities bear significant societal costs linked with FASD, which in Canada estimated to cost $5.3 billion annually (Popova, Stade, Bekmuradov, Lange, & Rehm, 2011).

Identification of FASD is important for at-risk individuals to receive timely management and support, and to aid the estimation of FASD prevalence to guide prevention, screening, diagnosis, and management services (Popova, Lange, Probst, Gmel, & Rehm, 2017; Watkins, Elliott, Halliday, O'Leary et al., 2013; Watkins, Elliott, Halliday, Mutch et al., 2013). Screening is the presumptive identification of disease by the application of tools or examinations (Wilson & Junger, 1968). The screening process begins with establishing at-risk populations to undergo FASD screening then applying tools sensitive to presume identification of FASD in a short time (Dobrow, Hagens, Chafe, Sullivan, & Rabeneck, 2018; Wilson & Junger, 1968). Screening tools do not require the accuracy of a diagnostic evaluation as individuals with positive screen results would be referred for comprehensive diagnostic evaluation (Dobrow et al., 2018; Wilson & Junger, 1968).

Screening for FASD is challenging for several reasons. First, approaches to screening are linked directly to diagnostic criteria of FASD, which have been known to differ across countries as several diagnostic criteria of FASD have been produced internationally to help clinicians make a diagnosis of FASD (Hemingway et al., 2019; Watkins, Elliott, Wilkins, Mutch et al., 2013). Thus, the lack of international consensus on diagnostic criteria contributes to differences in approaches to FASD screening (Watkins, Elliott, Halliday, O'Leary et al., 2013). Second, there is a lack of awareness of FASD among clinicians to recommend FASD screening for at-risk populations (Lange et al., 2017). Third, recognized screening tools for FAS are not appropriate for the screening for FASD. Screening tools for FAS usually contain items to assess growth and facial features; however, not all individuals with FASD present with facial features associated with FAS (Watkins, Elliott, Halliday, O'Leary et al., 2013). The established facial features associated with FAS include short palpebral fissure, smooth philtrum, and thin upper lip (Watkins, Elliott, Halliday, O'Leary et al., 2013). Fourth, FASD screening is difficult as no biomarkers or unique neurodevelopmental impairments associated with FASD have been identified (Watkins, Elliott, Halliday, O'Leary et al., 2013). Many neurodevelopmental impairments associated with FASD overlap with neurodevelopmental disorders, such as attention deficit hyperactivity disorder (McLennan, 2015). Fifth, the stigma associated with FASD could discourage and delay FASD screening and diagnostic engagements (Corrigan et al., 2019; Hamilton et al., 2020; Helgesson et al., 2018). Sixth, diagnostic evaluation of FASD post-screening is complex and access to this service is limited in many countries (Brown, Bland, Jonsson, & Greenshaw, 2018; Chudley, Kilgour, Cranston, & Edwards, 2007; Lange et al., 2017; Peadon, Fremantle, Bower, & Elliott, 2008).

Effective screening depends on the availability of reliable and accurate screening tools. However, little is known about FASD screening tools as evidence relating to FASD screening tools has not been synthesised. This systematic review was undertaken to synthesise information on FASD screening tools, including the identification of FASD screening tools, examination of their performance characteristics, evaluation of how they are used to identify early indicators of risk and to inform care, and examination of the scalability of the screening tools.

2. Methods

This systematic review follows the Preferred Reporting Items for Systematic Reviews and Meta-Analyses 2020 guideline (Page et al., 2021). This review was not registered but no methodological changes were made between study conception and completion.

2.1. Eligibility criteria

Empirical studies were included if they (i) involved individuals with FASD or PAE; (ii) examined tools designed to screen for FASD (including diagnostic categories of FAS, pFAS, ARND, and ARBD); (iii) reported the sensitivity and specificity of the tools; and (iv) were peer-reviewed. Studies were excluded if they (i) examined tools designed to screen for alcohol use behaviours in pregnant women; (ii) examined tools designed to screen for alcohol exposure in pregnant women or individuals via pharmacodynamic biomarkers; or (iii) were case reports, reviews, conference proceedings, dissertations, and book chapters.

2.2. Information sources and search strategy

Literature searches were conducted in four electronic databases from inception to 27 January 2021: Cumulative Index to Nursing and Allied Health Literature (CINAHL), Embase, MEDLINE and PsycINFO. The search strategy included a combination of terms relating to FASD, screening, and screening tools. Reference lists from relevant articles were also hand-searched for eligible studies. See the appendix for full search strategies of all databases.

2.3. Study selection

Two independent reviewers (Y.H.L. and N.R.K.) each screened 100 % of the search results by applying the eligibility criteria on the titles and abstracts of identified references. The same process was conducted for full-text screening. Disagreements were resolved through discussion to achieve a final consensus on included studies.

2.4. Data extraction

One reviewer (Y.H.L.) extracted data from the included studies using a standardized data extraction form that was developed based on the screening principles presented by Dobrow et al. (2018). These screening principles provide a comprehensive method to characterize screening tools, which include (a) screening tool performance characteristics; (b) interpretation of screening tool results; and (c) post-screening test procedures (Dobrow et al., 2018). Data extracted comprised the screening tool, authors, country of the study, study design, proportion of consent obtained, participant characteristics, setting, screening method, training for screeners, informant, screening tool performance characteristics (sensitivity, specificity, positive predictive value, and negative predictive value), interpretation of screen test results (cut-off points), time to complete the screening tool, cost of screening tool, post-screening test procedures, and diagnostic criteria used for diagnostic assessment. We abstracted or calculated sensitivity, specificity, positive and negative predictive values wherever possible using MedCalc Version 15.0 (2020).

Tools designed to screen for FAS were reported and analysed separately from tools designed to screen for all diagnostic categories of FASD because the approaches used to screen for FAS and FASD are different (Denny, Coles, & Blitz, 2017).

2.5. Risk of bias of included studies

The risk of bias and applicability of included studies were independently evaluated by three reviewers (Y.H.L., A.F-J., and R.E.W.) using the Quality Assessment of Diagnostic Studies-2 (QUADAS-2) tool (Whiting et al., 2011). The QUADAS-2 tool examined the level of bias concerning participant selection, screening test execution and interpretation, diagnostic criteria execution and interpretation, flow and timing of screening test and diagnostic criteria execution, as well as the applicability of participant selection, screening test, and diagnostic criteria. All discrepancies relating to the risk of bias assessment were resolved through discussion.

2.6. Data synthesis and analysis

A narrative synthesis was used to synthesize data across all studies.

3. Results

The search strategy resulted in 820 references after duplicates were removed. An additional four references were added from the manual screening of identified references. After title and abstract screening as well as full-text screening, 16 records were included in the systematic review (Fig. 1).

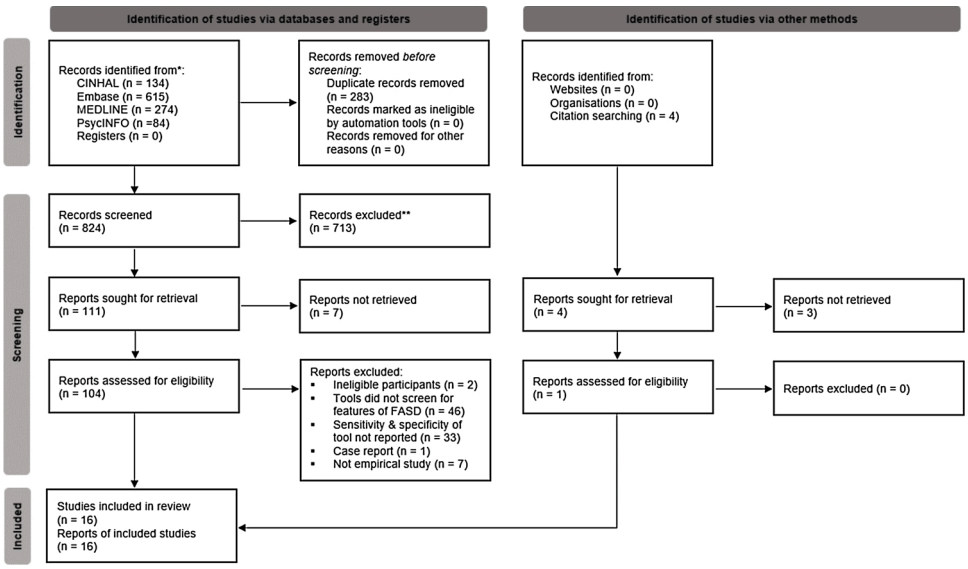

Fig. 1. Preferred Reporting Items for Systematic Reviews and Meta-Analyses 2020 flow diagram of search and selection process.

Table 1
Characteristics of Included Studies.

Screening Tool	Author	Country	Study Design	Proportion of Consent Obtained	Sample size	Diagnostic criteria	Age Range, y	Setting
FAS Screening Tool								
Craniofacial Measurements	Moore et al. (2001)	United States	Case-control	131/131	FAS (n = 41), PFAS (n = 59), and control (n = 31)	Institute of Medicine criteria	0–40	Research centres, FAS support centres and orthodontic pre-screening clinics
FAS Facial Photographic Screening Tool	Astley and Clarren (1996)	United States	Case-control	NR	FAS (n = 42) and control (n = 84)	Expert opinion	0–27	FAS clinic
	Astley et al. (2002)	United States	Cross-sectional	600/600	FAS (n = 8), with and without PAE (n = 592)	4-Digit Diagnostic Code	0–12	Out-of-home care programme
FAS Diagnostic Checklist	Burd et al. (2003)	United States	Case-control	NR	FAS (n = 140), PFAS (n = 134), and control (n = 78)	Institute of Medicine criteria	0–18	Medical genetic outreach clinics
FAS Screen	Burd et al. (1999)	United States	Cross-sectional	1013/1481	With and without PAE (n = 1013)	Gestalt method	4–8	Schools
	Poitra et al. (2003)	United States	Cross-sectional	NR	With and without PAE (n = 1384)	Gestalt method and Institute of Medicine criteria	5–10	Schools
FAS Screening Tool	Astley and Clarren (1995)	United States	Cross-sectional	NR	PAE (n = 194)	Gestalt method	0–10	FAS clinic
FASD Screening Tool								
Eye movement behaviour tasks	Tseng et al. (2013)	Canada	Case-control	31/31	FASD (n = 13) and control (n = 18)	NR	10–12	Not reported
	Zhang et al. (2019)	Canada	Case-control	207/207	FASD (n = 91) and control (n = 116)	Canadian Guidelines	5–18	FASD research centre
FASD Brief Screen Checklist	McLachlan et al. (2020)	Canada	Cross-sectional	41/145	With and without PAE (n = 41)	NR	Mean = 39, SD = 13	Outpatient forensic mental health programme
FASD Risk Assessment Questions	McLachlan et al. (2020)	Canada	Cross-sectional	47/145 plus data from discharged patients:104/104	With and without PAE (n = 151)	NR	Mean = 39, SD = 13	Outpatient forensic mental health programme
FASD Screening and Referral Tool for Youth Probation Officers	McLachlan et al. (2020)	Canada	Cross-sectional	47/145 plus data from discharged patients:104/104	With and without PAE (n = 151)	NR	Mean = 39, SD = 13	Outpatient forensic mental health programme
Life History Screen	Grant et al. (2013)	United States	Case-control	NR	FASD (n = 25), PAE (n = 61), and control (n = 463)	NR	24–27	Parent-child assistance programme
	McLachlan et al. (2020)	Canada	Cross-sectional	41/145	With and without PAE (n = 41)	NR	Mean = 39, SD = 13	Outpatient forensic mental health programme
Neurobehavioral Screening Test	Breiner et al. (2013)	Canada	Case-control	NR	FASD (n = 17) and control (n = 25)	Canadian Guidelines	4–6	FASD research clinic
	LaFrance et al. (2014)	United States	Case-control	NR	FASD (n = 48) and control (n = 32)	4-Digit Diagnostic Code; Canadian Guidelines	6–17	FASD clinical services program, FASD research centres and clinics, schools, and community centres
	Nash et al. (2006)	Canada	Case-control	60/60	FASD (n = 30) and control (n = 30)	Institute of Medicine criteria	6–16	FAS research clinic
	Nash et al. (2011)	Canada	Case-control	NR	FASD (n = 56) and control (n = 50)	Canadian Guidelines	6–18	FASD research clinic
Tallying Reference Errors in Narrative Task	Thorne (2017)	United States	Case-control	138/138	FASD (n = 69) and control (n = 69)	4-Digit Diagnostic Code	7–12	FASD clinic and schools

Note. FAS, fetal alcohol syndrome; FASD, fetal alcohol spectrum disorder; NR, not reported; PAE, prenatal alcohol exposure; PFAS, partial fetal alcohol syndrome; SD, standard deviation; y, years.

3.1. Study characteristics

Table 1 shows the characteristics of the 16 included studies, featuring five unique FAS screening tools and seven unique FASD screening tools. The included studies were conducted in the United States (10 studies) and Canada (six studies) and were published between 1995 and 2020. Of the 16 studies, 11 used a case-control design and five used a cross-sectional design. Participants in the studies were aged between newborn and 40 years; only four out of the 16 studies recruited participants above 18 years of age. Sample sizes ranged from $n = 31$ to $n = 1384$. The proportion of consent obtained for children to participate in studies examining FAS and FASD screening tools (68–100 %) was higher in comparison to adult participants (28 %). Eight studies recruited participants through diagnostic clinics and research centres, two through schools or the community, and two used a combination of these recruitment settings. A further study recruited participants through an out-of-home care program, another through a case-management intervention program, and one through an outpatient forensic mental health programme. One study did not report its recruitment strategy.

All studies, except four (Astley & Clarren, 1996; Grant et al., 2013; McLachlan, Amlung, Vedelago, & Chaimowitz, 2020; Tseng et al., 2013), employed diagnostic criteria to guide the diagnosis of FASD. Among studies that examined FAS screening tools, the most frequently used diagnostic criteria were the Institute of Medicine criteria (Hoyme et al., 2005) and Gestalt method (Sokol & Clarren, 1989), followed by the 4-Digit Diagnostic Code (Astley & Clarren, 2000), and expert opinion. For studies that examined FASD screening tools, the most frequently used diagnostic criteria were the Canadian Guidelines (Cook et al., 2016), followed by the 4-Digit Diagnostic Code (Astley, 2013), then the Institute of Medicine criteria (Hoyme et al., 2016).

3.2. Risk of bias of included studies

The quality of the included studies was rated at high risk of bias primarily due to the methods used in the selection of participants and the flow of screening tests and diagnostic criteria execution (Table 2). Where participant selection was rated high risk of bias, it was due to the use of case-control design and inadequate blinding of the examiner during the screening test and diagnostic criteria execution. The flow of screening test and diagnostic criteria execution was also rated high risk of bias because some participants did not receive diagnostic evaluation and some studies did not assess participants using the same diagnostic criteria.

3.3. Screening tool performance characteristics

Table 3 shows the performance characteristics of the screening tools, which were administered via a variety of methods. Most studies provided information to interpret screen test results except for one screening tool - eye movement behaviour tasks (Tseng et al., 2013; Zhang et al., 2019). In contrast, many studies did not provide information about the cost of administration except for three screening tools - FAS Screen, eye movement behaviour tasks, and Neurobehavioral Screening Test (Berrigan, Andrew, Reynolds, & Zwicker, 2019; Burd et al., 1999; Zhang et al., 2019).

3.4. FAS screening tools

3.4.1. Craniofacial measurements

One study examined craniofacial measurements of participants (Moore et al., 2001). The measurements comprised 21 different

Table 2
QUADAS-2 Assessment of Included Studies.

Study	Risk of bias				Applicability		
	Participant selection	Screening test	Diagnostic criteria	Study flow	Participant selection	Screening test	Diagnostic criteria
Astley and Clarren (1995)	Low	Unclear	Low	Low	High	Low	High
Astley and Clarren (1996)	High	Unclear	Low	Unclear	High	High	High
Astley et al. (2002)	Low	Unclear	Unclear	High	High	High	High
Breiner et al. (2013)	High	Unclear	Unclear	High	Low	Low	Low
Burd et al. (1999)	Unclear	Low	Unclear	Unclear	Low	High	High
Burd et al. (2003)	Low	Unclear	Low	Low	Unclear	Low	High
Grant et al. (2013)	High	Unclear	Unclear	High	Low	Low	Low
LaFrance et al. (2014)	High	Unclear	Unclear	High	Low	Low	Unclear
McLachlan et al. (2020)	Low	Unclear	Unclear	High	Low	Low	Unclear
Moore et al. (2001)	High	High	Low	High	Low	Low	Low
Nash et al. (2006)	High	Low	Unclear	High	Low	Low	Low
Nash et al. (2011)	High	Low	Low	High	Low	Low	Low
Poitra et al. (2003)	Low	Low	Low	High	Low	High	High
Thorne (2017)	High	Low	Low	High	Low	Low	Low
Tseng et al. (2013)	High	Low	Unclear	High	Low	Low	Unclear
Zhang et al. (2019)	High	Low	Low	High	Low	Low	Low

Table 3
Screening Tool Performance Characteristics.

Screening Tool	Screening Method	Training for Screeners	Informant	Sensitivity (%)	Specificity (%)	PPV (95 % CI)	NPV (95 % CI)	Cut-off Point	Time to Complete Screening Tool, min	Cost of Screening Tool
FAS Screening Tool										
Craniofacial Measurement	Head and facial measurement	NR	Nil	100	100	NR	NR	D-score ≤0.20	NR	NR
FAS Facial Photographic Screening Tool	Computer and photograph	Yes	Nil	100	100	85.7[a]	100[a]	D-score >0.7; Palpebral fissure lengths >2 SD above the mean of normal physical measurements; philtrum at Likert rank 4 or 5; vermilion border of upper lip at Likert rank 4 or 5	30	NR
FAS Diagnostic Checklist	Face-to-face Interview	NR	Caregiver; physician	89	72	68	91	Score ≥14.5 (FAS)	NR	NR
FAS Screen	Face-to-face Interview	Yes	Caregiver; physician	100	94–95	9[e] (7, 11)	100[e]	Score ≥20	8–15	USD$13
FAS Screening Tool	Physical and facial measurement	NR	Nil	100	89	70[b] (59, 78)	100[b]	D-score ≥1.5	NR	NR
FASD Screening Tool										
Eye movement behaviour tasks	Computer and eye tracking device	Yes	Nil	73–77	79–91	NR	NR	NR	17–20	CAD$50
FASD Brief Screen Checklist	Face-to-face Interview	Yes	Participant	25 (1, 81)	100 (91, 100)	92 (87, 95)	93 (80, 98)	> 10 on Behavioural Indicator, >2 on Historical Indicator, and presence of maternal alcohol use in childhood on Maternal Indicator	10–15	NR
FASD Risk Assessment Questions	Chart review of existing personal and medical history records	Yes	Existing personal and medical history records	64 (31, 89)	77 (69, 84)	18 (11, 27)	96 (92, 98)	≥ 5 "Yes" out of 9 items	5	NR
FASD Screening and Referral Tool for Youth Probation Officers	Chart review of personal and medical history records	Yes	Existing personal and medical history records	91 (59, 100)	71 (63, 79)	20 (15, 26)	99 (94, 100)	1 Social item and ≥2 Personal items; or ≥3 Personal items	10–15	NR
Life History Screen – 11 items	Face-to-face Interview	Yes	Participant	75–81	66–73	23 (12, 39)	96 (83, 99)	≥5 "Yes" out of 11 items	10	NR
Neurobehavioral Screening Test	Paper and pencil	NR	Caregiver	63–98	42–100	100 [a]	64[c] (55, 72)	≥5–6 'Yes' out of 7 items and ≥3 'Yes' in indicated items	22	CAD$20[d]
Tallying Reference Errors in Narrative Task	Story narration	Yes	Nil	54	96	NR	NR	Grammatical error rate of 2 SD above the mean of the control group	NR	NR

Note. [a], data from Astley et al. (2002); [b], data from Astley and Clarren (1995); [c], data from LaFrance et al. (2014); [d], data from Berrigan et al. (2019); [e], data from Burd et al. (1999); CAD, Canadian dollars; FAS, fetal alcohol syndrome; FASD, fetal alcohol spectrum disorder; NPV, negative predictive value; NR, not reported; PPV, positive predictive value; SD, standard deviation; USD, United States dollars.

sentinel facial features of FAS and head circumference. The evaluation of participants using head circumference and bigonial breadth measurements yielded 100 % sensitivity and specificity to discriminate individuals with and without FAS. In addition, the evaluation of participants using six measurements, consisting of minimal frontal breadth, bigonial breadth, midfacial depth, palpebral fissure length, head circumference, and maxillary, yielded 98 % sensitivity and 90 % specificity to discriminate individuals with and without PAE.

3.4.2. FAS facial photographic screening tool

Two studies examined facial measurements of participants using photographs (Astley & Clarren, 1996; Astley, Stachowiak, Clarren, & Clausen, 2002). The study by Astley and Clarren (1996) used a computer programme to measure four sentinel facial features of FAS from photographs, including palpebral fissure length, inner canthi distance, philtrum smoothness, and upper lip thinness. The study by

Table 4
Post-screening Procedures.

Screening Tool	Author	Results Discussed	Referral Offered	Intervention Delivered	Diagnostic Criteria	Description of Follow-up
FAS Screening Tool						
Craniofacial Measurements	Moore et al. (2001)	–	–	–	Institute of Medicine criteria	NR
FAS Facial Photographic Screening Tool	Astley and Clarren (1996)	–	–	–	Expert opinion	NR
	Astley et al. (2002)	✓	✓	✓	4-Digit Diagnostic Code	Children with positive results were referred to a social worker, and for a diagnostic evaluation and management planning.
FAS Diagnostic Checklist	Burd et al. (2003)	–	–	–	Institute of Medicine criteria	NR
FAS Screen	Burd et al. (1999)	✓	✓	NR	Gestalt method	Children with positive results were referred for a diagnostic evaluation; results of the evaluation were discussed with caregivers.
	Poitra et al. (2003)	✓	✓	✓	Gestalt method and Institute of Medicine criteria	Children with positive results were referred for a diagnostic evaluation; results of the evaluation were discussed with the child's physician and school for management planning.
FAS Screening Tool	Astley and Clarren (1995)	–	–	NR	Gestalt method	NR
FASD Screening Tool						
Eye movement behaviour tasks	Tseng et al. (2013)	–	–	–	NR	NR
	Zhang et al. (2019)	–	–	–	Canadian Guidelines	NR. The authors recommended participants classified as high risk be referred for a diagnostic evaluation.
FASD Brief Screen Checklist	McLachlan et al. (2020)	–	–	–	NR	NR
FASD Risk Assessment Questions	McLachlan et al. (2020)	–	–	–	NR	NR
FASD Screening and Referral Form for Youth Probation Officers	McLachlan et al. (2020)	–	–	–	NR	NR
Life History Screen	Grant et al. (2013)	–		–	NR	NR
	McLachlan et al. (2020)	–	–	–	NR	NR
Neurobehavioral Screening Test	Breiner et al. (2013)	–	–	–	Canadian Guidelines	NR
	LaFrance et al. (2014)	–	–	–	4-Digit Diagnostic Code; Canadian Guidelines	NR
	Nash et al. (2006)	–	–	–	Institute of Medicine criteria	NR
	Nash et al. (2011)	–	–	–	Canadian Guidelines	NR
Tallying Reference Errors in Narrative Task	Thorne (2017)	–	–	–	4-Digit Diagnostic Code	NR

Note. -, not applicable; ✓, applicable; NR, not reported.

Astley et al. (2002) used a computer programme to measure three sentinel facial features of FAS from photographs, including palpebral fissure length, philtrum smoothness, and upper lip thinness. The evaluation of participants using the FAS Facial Photographic Screening Tool yielded 100 % sensitivity and 100 % specificity to differentiate individuals with and without FAS.

3.4.3. FAS diagnostic checklist

One study examined the FAS Diagnostic Checklist (Burd, Martsolf, Klug, & Kerbeshian, 2003). The 62-item checklist included assessment of PAE, head circumference, growth impairments, brain dysfunction, and sentinel facial features of FAS (Burd & Martsolf, 1989). The evaluation of participants using the FAS Diagnostic Checklist yielded 89 % sensitivity and 72 % specificity to differentiate individuals with and without FAS.

3.4.4. FAS screen

Two studies examined the FAS Screen (Burd et al., 1999; Poitra et al., 2003). The 30-item screening form included assessment of growth impairments, neurologic dysfunction, and sentinel facial features of FAS. The evaluation of participants using the FAS Screen yielded 100 % sensitivity and 94–95 % specificity to discriminate children with and without FAS.

3.4.5. FAS screening tool

One study examined the FAS Screening Tool (Astley & Clarren, 1995). The 16-item screening tool comprised measurements of 12 different FAS sentinel facial features, hockey-stick palmar creases, head circumference, height, and weight. The evaluation of participants using three measurements, consisting of palpebral fissure length, smooth philtrum, and thin upper lip, yielded 100 % sensitivity and 89 % specificity to discriminate individuals with and without FAS.

3.5. FASD screening tools

3.5.1. Eye movement behaviour tasks

Two studies examined the eye behaviour of participants to assess for deficits in attention and visual-motor function (Tseng et al., 2013; Zhang et al., 2019). The assessment involved the collection of participants' eye movement data using an eye-tracking device when looking towards visual targets and at a series of video clips. A machine-learning algorithm was used to learn about the eye movement data of those with FASD to identify at-risk individuals. The assessment of eye movement behaviour yielded 73–77 % sensitivity and 79–91 % specificity to differentiate individuals with and without FASD.

3.5.2. FASD brief screen checklist

One study examined the FASD Brief Screen Checklist (McLachlan et al., 2020). The self-reported checklist contained a total of 36 behavioural, historical, and maternal indicators associated with FASD (MacPherson, Chudley, & Grant, 2011; McLachlan et al., 2020). The evaluation of participants using the FASD Brief Screen Checklist yielded 25 % sensitivity and 100 % specificity to discriminate individuals with and without a high risk of having FASD.

3.5.3. FASD risk assessment questions

One study examined the FASD Risk Assessment Questions (McLachlan et al., 2020). The nine questions in the FASD Risk Assessment Questions were considered to be useful indicators of FASD risk (Kellerman, 2005; Substance Abuse & Mental Health Services Administration, 2014). The evaluation of participants using the FASD Risk Assessment Questions yielded 64 % sensitivity and 77 % specificity to differentiate individuals with and without high risk of having FASD.

3.5.4. FASD screening and referral tool for youth probation officers

One study examined the FASD Screening and Referral Tool for Youth Probation Officers (McLachlan et al., 2020). The checklist comprised five social factors and five personal factors that were indicators of FASD risk (Conry & Asante, 2010). The evaluation of participants using the FASD Screening and Referral Tool for Youth Probation Officers yielded 91 % sensitivity and 71 % specificity to differentiate individuals with and without high risk of having FASD.

3.5.5. Life history screen

Two studies examined the Life History Screen (Grant et al., 2013; McLachlan et al., 2020). The semi-structured instrument comprised 27 items, relating to personal and family history, education level, employment history, criminal history, mental health, and day-to-day behaviours (Grant et al., 2013; McLachlan et al., 2020). The study by McLachlan et al. (2020) administered 25 items due to uncertain scoring criteria of two items "In what grade did you start using alcohol or drugs?" and "If you did not finish school, why did you leave?". The evaluation of participants based on a score of 10 or higher on the 25-item Life History Screen yielded 100 % sensitivity and 51 % specificity to differentiate individuals with and without a high risk of having FASD. The study by Grant et al. (2013) administered 11 items that were correlated to items from the Addiction Severity Index, a standardized assessment of substance abuse treatment (McLellan, Luborsky, Woody, & O'Brien, 1980). The evaluation of participants based on a score of five or higher on the 11-item Life History Screen yielded 81 % sensitivity and 66 % specificity to differentiate individuals with and without a high risk of having FASD.

3.5.6. Neurobehavioral screening test

Four studies examined the Neurobehavioral Screening Test (Breiner, Nulman, & Koren, 2013; LaFrance et al., 2014; Nash et al., 2006; Nash, Koren, & Rovet, 2011). The caregiver-reported questionnaire included nine to 10 items relating to attention, adaptive behaviour, executive functioning, and social skills. Test items were extracted from the Child Behaviour Checklist, a standardized caregiver reported assessment of behaviour problems in children (Achenbach, 1999). The Neurobehavioral Screening Test screening tool has been tested in children with FASD ages four to 18 years. The evaluation of participants using the Neurobehavioral Screening Test yielded 63−98 % sensitivity and 42−100 % specificity to differentiate individuals with and without FASD.

3.5.7. Tally reference errors in narrative task

One study examined the Tally Reference Errors in Narrative Task to assess for language impairment (Thorne, 2017). The task involved the collection of participants' narratives based on a wordless storybook that was provided to them. The evaluation of participants using the Tally Reference Errors in Narrative Task yielded 54 % sensitivity and 96 % specificity to discriminate individuals with and without FASD.

3.6. Post-screening follow-up procedure

Table 4 shows the various post-screening follow-up procedures from the included studies. Three studies reported follow-up procedures after screening for FASD (Astley et al., 2002; Burd et al., 1999; Poitra et al., 2003). The procedure included the discussion of screening results with caregivers, the participant's physicians, and schools and an offer of referral for a comprehensive diagnostic evaluation. Of these three studies, one that involved children from an out-of-home program made an additional referral to a social worker for children with positive screen results (Astley et al., 2002). Only two studies reported the delivery of management interventions to children upon the confirmation of an FASD diagnosis (Astley et al., 2002; Poitra et al., 2003). The rest of the studies did not report follow-up procedures as they employed a case-control study design.

4. Discussion

In the present review, we identified five tools that were designed to screen for FAS and seven tools that were designed to screen for all diagnostic categories of FASD. These screening tools consisted of various screening approaches, methods, performance characteristics, and are linked to different diagnostic criteria. FAS screening tools demonstrated high accuracy in identifying individuals at risk of FAS while FASD screening tools demonstrated limited accuracy in identifying individuals at risk of FASD. In comparison with the adult population, the present review revealed a higher proportion of consent from the children population to receive FAS or FASD screening. Most of the screening tools provided well-defined instructions for administration and interpretation of screen test results; however, information regarding the resources required to administer the screening tool, and post-screening follow-up procedures were not well-reported across all included studies. Therefore, there is a limited understanding of the feasibility and scalability of these tools.

Studies that examined FAS screening tools were published between 1995 and 2003 (Astley & Clarren, 1995, 1996; Astley et al., 2002; Burd et al., 1999, 2003; Moore et al., 2001; Poitra et al., 2003). These tools were linked to three diagnostic criteria: The Institute of Medicine criteria (Hoyme et al., 2005), Gestalt method (Sokol & Clarren, 1989), and 4-Digit Diagnostic Code (Astley & Clarren, 2000). The main approach of these tools was to assess for sentinel facial features associated with FAS, which had been found to be an accurate biomarker in the identification of individuals at risk of FAS (89–100 % sensitivity and 72–100 % specificity). Besides the assessment of sentinel facial features, four of the five tools also screened for head circumference, growth impairments, brain dysfunction, and neurologic dysfunction (Astley & Clarren, 1995; Burd et al., 1999, 2003; Moore et al., 2001; Poitra et al., 2003).

Studies that examined FASD screening tools were published between 2005 and 2020 (Breiner et al., 2013; Grant et al., 2013; LaFrance et al., 2014; McLachlan et al., 2020; Nash et al., 2006, 2011; Thorne, 2017; Tseng et al., 2013; Zhang et al., 2019). These tools were linked to three diagnostic criteria: The Canadian Guidelines (Cook et al., 2016), 4-Digit Diagnostic Code (Astley, 2013), and Institute of Medicine criteria (Hoyme et al., 2016). Various approaches were used to screen for FASD, including the assessment of cognition, academic ability, adaptive behaviour, attention, memory, affect regulation, executive function, visual motor, language, and social skills. However, the accuracy in the identification of FASD using these neurodevelopmental features (25–98 % sensitivity and 42−100% specificity) was significantly variable. Despite the varying accuracy of these seven screening tools, three of them showed high accuracy in the identification of individuals at risk of FASD (above 80 % sensitivity and above 70 % specificity): The FASD Screening and Referral Tool for Youth Probation Officers (McLachlan et al., 2020), Life History Screen – 11 items (Grant et al., 2013; McLachlan et al., 2020), and Neurobehavioral Screening Test (Breiner et al., 2013; LaFrance et al., 2014; Nash et al., 2006, 2011). Given the wide variability in the accuracy of these tools, further understanding of neurodevelopmental profiles or unique biomarkers of FASD is required to facilitate the development of accurate FASD screening tools.

Few studies provided adequate information on the acceptability of the screening tools to the population. Among studies that reported the proportion of consent obtained from participants to undergo screening, one study that involved adult participants enrolled in a forensic mental health programme received a response rate of 28 %. In contrast, seven studies that involved child participants in various community settings (clinics, research centres, out-of-home care programmes, and schools) received a response rate of 68–100 %. It might be worth noting that the lower bound response rate of 68 % correlated to screening in school (Burd et al., 1999). The higher proportion of consent for FAS or FASD screening in the child population supports the finding from one study that reported the supportiveness of most caregivers for their children to participate in screening if it allowed them to better understand their child's strengths and needs (Morelli et al., 2014).

Few studies also provided adequate information on resources required to administer the screening tool to determine their feasibility and scalability. Information about resources, especially training requirements of screeners, time to complete screening, and cost of administrating the tool, are also important considerations to guide policy and practice decisions about implementation (Dobrow et al., 2018). Screening tools that require screeners to undergo extensive training, take a long time to administer, or are costly to administer may be obstacles to the scalable implementation of the tool. Future studies are recommended to include information about resource requirements of FAS or FASD screening tools.

Only three studies reported the post-screening follow-up procedures for participants who received positive screen test results. Some reasons for the lack of post-screening follow-up reporting included one study that did not conduct diagnostic evaluations for their participants and most of the studies used a retrospective case-control study design where diagnostic evaluation and management plans had already been implemented. An essential component of post-screening follow-up for participants with positive results involves diagnostic evaluation, and thus it is crucial to note that in the present review a total of four different diagnostic criteria were identified to confirm the diagnosis of FASD. The use of different diagnostic criteria may lead to disagreements in the diagnosis of FASD. A comparison study, not included in this review, applied four different FASD diagnostic criteria - the 4-Digit Diagnostic Code, Canadian Guidelines, Australian Guidelines, and Institute of Medicine criteria - to the same patient cohort and reported discrepancies in FASD diagnostic outcomes (Hemingway et al., 2019). For example, the proportion of patients diagnosed with FASD using the 4-Digit Diagnostic Code and the Canadian Guidelines was 79 % and 16 %, respectively (Hemingway et al., 2019). One of the key factors contributing to this discrepancy was the difference in the diagnostic criteria regarding the inclusion of moderate dysfunction as an outcome of PAE (Hemingway et al., 2019). For instance, an individual who presents with one or two neurodevelopmental abnormalities that are two or more standard deviations below the mean (moderate dysfunction) and with confirmed PAE could meet the criteria for FASD using the 4-Digit Diagnostic Code; however, the same individual would not meet the criteria for FASD using the Canadian Guidelines. This finding was consistent with another review that reported discrepancies in FASD diagnosis were due to the disagreements in the identification and definition of features associated with FASD in different diagnostic criteria (Brown et al., 2018). The implications of discrepancies in FASD diagnosis include misdiagnoses or missed diagnoses, inappropriate management plans for individuals, increased risk of secondary disabilities in individuals, and even difficulties establishing consistent estimates of FASD prevalence across countries (Brown et al., 2018). Therefore, an internationally consistent FASD diagnostic criteria would be beneficial to guide the development of a tool that can screen for all diagnostic categories of FASD and the subsequent post-screening follow-up procedures.

Little is known about the use of the available FAS and FASD screening tools in subpopulations with an identified high prevalence of FASD, for example, special education and First Nation populations (Popova, Lange, Shield, Burd, & Rehm, 2019). Many of the included studies also did not report on cultural considerations in the design and administration of the screening tool. Only one study accounted for a flat mid-face profile, which is commonly found in the Asian and American Indian races, replacing that facial measurement with palpebral fissure length (Astley & Clarren, 1995). Prior studies have found that cultural adaptation of a screening tool improved its acceptability within the targeted population as well as clarity and comprehension of concepts relating to the tool (Kaiser et al., 2019; Soto et al., 2014). More research regarding the cultural adaptation of FASD screening tools is warranted.

This review has several limitations. Firstly, the validity of the conclusions drawn from this review is limited by the quality of the included studies, which was rated at high risk of bias. Secondly, a meta-analysis of diagnostic accuracy of screening tools was not performed as many included studies did not provide 2×2 contingency tables.

There is scope for research to further understand biomarkers unique to FASD to guide the development of accurate FASD screening tools as well as to link FASD screening tools to an internationally consistent FASD diagnostic criteria. Future studies are also recommended to include more information on risks and harms of screening, and resource requirements of FASD screening tools that will be useful to determine the feasibility and scalability of these tools. Further knowledge about the attitude of individuals with FASD and caregivers on screening tools and programmes would be beneficial to evaluate the acceptability of the tools, guiding policy and practice decisions. More research on cultural considerations of FASD screening tools is recommended. The administration of an FASD screening tool might create additional burdens on healthcare systems when developmental screening has already been performed during scheduled child health checks, and thus future works need to consider the additional burdens and identify opportunities to overcome them.

5. Conclusion

We identified five FAS and seven FASD screening tools. These screening tools assessed a variety of features associated with FASD, employed different screening approaches, had different performance characteristics and were linked to different diagnostic criteria. FAS screening tools showed high accuracy in the identification of at-risk individuals with sentinel facial features of FASD while the accuracy of FASD screening tools was varied. FAS and FASD screening tools were found to be acceptable to the children population but information regarding the feasibility and scalability of the tools cannot be determined due to inadequate reporting of data on resources required for screening. The diagnostic criteria used for the diagnosis of FASD also differed across the included studies, impacting the comparability of study findings. This review highlights the vast differences in the screening approaches, performance characteristics, and diagnostic criteria linked to FASD screening tools.

Funding source

This work was supported by the FASD Research Australia Centre of Research Excellence (National Health and Medical Research

Council); and the Department of Health Western Australia.

Contributors' statement page

Dr Lim contributed to the conceptualization of the study, conducted the literature searches for the systematic review, reviewed articles for inclusion, performed data extraction, reviewed articles methodology by using the quality appraisal tool, analysed the data, contributed to the interpretation of results, drafted the initial manuscript, and revised the manuscript.

Dr Finlay-Jones and Dr Watkins contributed to the conceptualization of the study, reviewed articles methodology by using the quality appraisal tool, analysed the data, contributed to the interpretation of results, and reviewed and revised the manuscript.

Ms Jones conceptualized and contributed to the study design, contributed to the interpretation of results, and reviewed and revised the manuscript.

Ms Kippin reviewed articles for inclusion, contributed to the interpretation of results, and reviewed and revised the manuscript.

All authors approved the final manuscript as submitted and agree to be accountable for all aspects of the work.

Declaration of Competing Interest

None.

Appendix A

Search strategy for FASD screening tools

Database	Search strategy
CINAHL	S1: fetal alcohol spectrum disorder S2: fetal alcohol syndrome S3: Prenatal alcohol expos* S4: alcohol and (birth defects or neurodevelopmental disorder) S5: fetal alcohol effects S6: static encephalopathy alcohol exposed S7: neurobehavioral disorder alcohol exposed S8: FASD or PAE or ND-PAE S9: S1 OR S2 OR S3 OR S4 OR S5 OR S6 OR S7 OR S8 S10: screening S11: screening tool S12: screening test S13: clinical examination S14: biomarkers S15: S10 OR S11 OR S12 OR S13 OR S14 S16: S9 AND S15
Embase	1. exp fetal alcohol syndrome/ 2. F?etal alcohol spectrum disorder.tw. 3. F?etal alcohol syndrome.tw. 4. Prenatal alcohol expos*.tw. 5. (alcohol and (birth defects or neurodevelopmental disorder)).tw. 6. fetal alcohol effects.tw. 7. static encephalopathy alcohol exposed.tw. 8. neurobehavioral disorder alcohol exposed.tw. 9. (FASD or PAE or ND-PAE).tw. 10. 1 or 2 or 3 or 4 or 5 or 6 or 7 or 8 or 9 11. exp screening/ 12. Screening tool.tw. 13. screening test.tw. 14. exp clinical examination/ 15. exp biological marker/ 16. 11 or 12 or 13 or 14 or 15 17. 10 and 16 18. limit 17 to animals 19. 17 not 18 20. limit 19 to English language
MEDLINE	1. exp Fetal Alcohol Spectrum Disorders/ 2. F?etal alcohol spectrum disorder.tw. 3. F?etal alcohol syndrome.tw. 4. Prenatal alcohol expos*.tw. 5. (alcohol and (birth defects or neurodevelopmental disorder)).tw. 6. fetal alcohol effects.tw.

(*continued on next page*)

(*continued*)

Database	Search strategy
PsycINFO	7. static encephalopathy alcohol exposed.tw. 8. neurobehavioral disorder alcohol exposed.tw. 9. (FASD or PAE or ND-PAE).tw. 10. 1 or 2 or 3 or 4 or 5 or 6 or 7 or 8 or 9 11. exp Screening/ 12. Screening tool.tw. 13. screening test.tw. 14. clinical examination.tw. 15. exp Biomarkers/ 16. 11 or 12 or 13 or 14 or 15 17. 10 and 16 18. limit 17 to animals 19. 17 not 18 20. limit 19 to English language 1. exp Fetal Alcohol Syndrome/ 2. F?etal alcohol spectrum disorder.tw. 3. F?etal alcohol syndrome.tw. 4. Prenatal alcohol expos*.tw. 5. (alcohol and (birth defects or neurodevelopmental disorder)).tw. 6. fetal alcohol effects.tw. 7. static encephalopathy alcohol exposed.tw. 8. neurobehavioral disorder alcohol exposed.tw. 9. (FASD or PAE or ND-PAE).tw. 10. 1 or 2 or 3 or 4 or 5 or 6 or 7 or 8 or 9 11. exp Screening/ 12. Screening tool.tw. 13. exp Screening Tests/ 14. clinical examination.tw. 15. exp biological markers/ 16. 11 or 12 or 13 or 14 or 15 17. 10 and 16 18. limit 17 to animal 19. 17 not 18 20. limit 19 to English language

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