Screening for Fetal Alcohol Spectrum Disorder (FASD) in Western Australia: Policy and Practice Recommendations

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Title: Screening for Fetal Alcohol Spectrum Disorder (FASD) in Western Australia: Policy and Practice Recommendations

Using the term Aboriginal

Within Western Australia, the term Aboriginal is used in preference to Aboriginal and Torres Strait Islander, in recognition that Aboriginal people are the original inhabitants of Western Australia. Aboriginal and Torres Strait Islander may be referred to in the national context and Indigenous may be referred to in the international context. No disrespect is intended to our Torres Strait Islander colleagues and community.

Citation

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Contents

Supplementary material: GRADE evidence report

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We would like to acknowledge the contribution of the members of the Advisory Group, Project Group and Working Group in this project (see list of contributors on page 2). We would also like to thank Professor Carol Bower for her invaluable advice given in the development of this report.

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Commonly used abbreviations

Abbreviation	Full description
ARND	Alcohol related neurodevelopmental disorder
FAE	Fetal alcohol effects
FAS	Fetal alcohol syndrome
FASD	Fetal alcohol spectrum disorder
GRADE	Grading of Recommendations Assessment, Development and Evaluation
ND-PAE	Neurobehavioral disorder associated with prenatal alcohol exposure
NST	Neurobehavioral Screening Test
PAE	Prenatal alcohol exposure
PFAS	Partial fetal alcohol syndrome
PICO	Patient, intervention, comparison, outcome
QUADAS-2	Quality Assessment of Diagnostic Studies-2
TREIN	Tallying Reference Errors in Narrative

Executive summary

Fetal alcohol spectrum disorder (FASD) is a neurodevelopmental disorder caused by prenatal exposure to alcohol. Individuals with FASD experience a range of severe neurodevelopmental impairments and may also display facial anomalies and differences in physical development. The impacts of FASD on individuals, families and communities are significant, and thus strategies to screen for FASD are warranted to provide appropriate support and management for FASD. However, there is currently no validated standardised screening tool for FASD in use in Western Australia.

The aims of the current report were (1) to provide an overview of FASD screening tools available internationally; (2) to provide recommendations on the implementation of available FASD screening tools in Western Australia; and (3) to provide recommendations on FASD screening approach in Western Australia to underpin implementation of FASD screening in the future.

The first aim was addressed through a systematic review of studies reporting the properties of different FASD screening tools. The systematic review included 14 articles, reporting on eight different FASD screening tools. All the articles originated from the United States and Canada; none of the articles were from Australia. An evidence report was produced for each screening tool and was used to guide the formulation of the recommendation.

The second aim was addressed through a systematic formulation of recommendations using the Grading of Recommendations, Assessment, Development and Evaluation (GRADE) approach.[1] An Advisory Group was established, including clinicians, researchers, and others with expertise in FASD, to provide strategic advice during the formulation of the recommendations. The final recommendation for each FASD screening tool was decided based on the evidence report and agreed by consensus within the Advisory Group. None of the eight available FASD screening tools was recommended for use as a universal screening tool in Western Australia.

The third aim was addressed through a rapid review of the existing literature and national and state policies, direction and strategic plan regarding FASD

screening approaches and models of care. The main findings corresponding to each specific research question are described in the Executive Summary Table.

Executive Summary Table. Summary of the main findings. This is a replication of Table 13 presented in the Discussion section in Chapter 9.

What FASD screening tools are available internationally?
The systematic review included 14 articles, examining eight different FASD screening tools: ▪ The Neurobehavioral Screening Tool ▪ Eye movement behaviour assessment via machine learning ▪ Tally Reference Errors in Narrative Task ▪ Dysmorphic examination via photographs ▪ Physical and dysmorphic examination ▪ Craniofacial measurements approach ▪ The FAS Screen ▪ The FAS diagnostic checklist The quality of evidence for the eight screening tools was variable, ranging from moderate to very low.
Which FASD screening tools are recommended for use in Western Australia?
These recommendations apply only in the context of *universal* FASD screening in Australia. The recommendations were assessed as 'strong recommendation' or 'conditional recommendation' (depending on screening context) according to the GRADE approach.[1] **Strong recommendations:** ▪ against the use of the Tallying Reference Errors in Narrative task to screen for individuals at risk of FASD. ▪ against the use of the dysmorphic examination via photographs to screen for individuals at risk of FASD. ▪ against the use of the craniofacial measurement approach to screen for individuals at risk of FASD.

- against the use of the FAS Screen to screen for individuals at risk of FASD.
- against the use of the FAS diagnostic checklist to screen for individuals at risk of FASD.

Conditional recommendations:

- against the use of the Neurobehavioral Screening Test to screen for individuals at risk of FASD.
- against the use of the eye movement behaviour assessment via machine learning to screen for individuals at risk of FASD.
- against the use of the physical and dysmorphic examination to screen for individuals at risk of FASD.

What is the recommended approach for FASD screening in Western Australia?

Even though no FASD screening tools are recommended for use in Western Australia, the following recommended approach to FASD screening has been formulated with the view that appropriate screening tools are required and will be developed in the future. All recommendations relate to the screening approach for FASD (with and without sentinel facial features).

Recommendations regarding FASD screening approaches

Three types of screening approaches have been identified for use in Western Australia, that together will support the likelihood that individuals with FASD will be identified in a timely fashion.

- Universal screening:
  - Implement prenatal alcohol exposure (PAE) screening approaches in the entire population, regardless of risk.
- Targeted screening:
  - Implement neurodevelopmental screening with individuals who have PAE, show signs of a neurodevelopmental disorder, such as developmental delay, have a sibling who has been diagnosed with FASD, have facial anomalies associated with FASD, have observed problems with behaviour, or where there is parental concern that a child may have FASD. Refer these individuals to

Child Developmental Services or private practitioners for further evaluation, and, if necessary, a comprehensive diagnostic assessment.

- Selective screening:
  - Implement neurodevelopmental and PAE screening in selected populations known to report a higher prevalence of FASD. For example, children referred to Child Development Services or Child and Adolescent Mental Health Services; children with mothers who access alcohol treatment services; children in state/foster care; and youth in justice settings. Refer these individuals to Child Developmental Services or private practitioners for further evaluation and, if necessary, a comprehensive diagnostic assessment.

Recommendations regarding FASD Model of Care

A review of the existing FASD Model of Care 2013-2018 is warranted. The development of additional strategies to address the following areas of concern are recommended:

- Improve coordinated information sharing regarding PAE between relevant health care services and organisations supporting children at risk of FASD through the Purple book, a personalised child health record.
- Support parents with children aged between ages 12 months and 2 years to take up community child health services (for example, the Purple book appointments) to aid early identification of concerns in their child's development.
- Support parents with children aged between 4 and 5 years to take up community child health services (for example, the School Entry Health Assessment) to aid early identification of concerns in their child's development.
- Support relevant systems and organisations in the integration of neurodevelopmental and PAE screenings in general health assessments conducted with populations at high risk of FASD (for example, children referred to Child Development Services, children with mothers who

receive alcohol treatment services, children in state/foster care, and young people in justice settings).

- Review existing child health referral pathways to improve coordinated referral of children with poor outcomes to Child Developmental Services or private practitioners for further evaluation and, if necessary, a comprehensive diagnostic assessment.

This report incorporates a comprehensive review of the evidence regarding FASD screening tools. The findings provide key insights into the research evidence relevant to the development of a FASD screening approach in Western Australia. This information is critical to inform clinical and policy decisions regarding the implementation of FASD screening approach most appropriate for at-risk individuals in Western Australia. The absence of a recommended FASD screening tool reinforces the need for further development of FASD screening tools and approach. Notably, the absence of FASD screening tools appropriate for the culturally and linguistically diverse populations highlights an urgent research priority. The findings of the systematic reviews will change as further scientific evidence emerges, and it is recommended that this report is updated on a regular basis.

1 Introduction

1.1 What is fetal alcohol spectrum disorder?

Fetal alcohol spectrum disorder (FASD) is a diagnostic term used to describe a condition involving neurodevelopmental abnormalities in at least three of ten specified domains of central nervous system structure or function because of prenatal exposure to alcohol.[2,3] Categorisation of FASD varies internationally. In Australia and Canada, FASD is classified into two sub-categories[3,4]: FASD with three sentinel facial features and FASD with less than three sentinel facial features. However, in the United States, FASD is an umbrella term for sub-categories such as fetal alcohol syndrome (FAS), partial FAS (PFAS), alcohol related neurodevelopmental disorder (ARND), fetal alcohol effects (FAE), and neurobehavioral disorder associated with prenatal alcohol exposure (ND-PAE).[5]

Recent reviews have estimated the global prevalence of FASD and FAS at 7.7 per 1000 population and 1.5 per 1000 population, respectively.[6,7] Notably, the prevalence of FASD in special subpopulations, for instance, children in care, youth justice system, special education, specialised clinical, and Aboriginal populations, was found to be 10 to 40 times higher compared with the global prevalence in the general population.[8]

1.2 The impact of FASD

Individuals with FASD experience a range of neurodevelopmental impairments and may also display facial anomalies and differences in physical development.[3,9] Together, the physiological and neurocognitive difficulties experienced by individuals with FASD can adversely impact daily function at home, school, and work.[10,11] Many individuals with FASD also develop mental health problems that persist into adulthood.[12] Besides mental health problems, adults with FASD are more likely to have difficulty finding employment and are more reliant on social welfare when compared with typically developing adults.[13,14] Furthermore, there is growing recognition of the involvement of people with FASD in the justice system; the prevalence of FASD among young people in the Canadian youth justice system has increased from 12% in 2005 to 21% in 2013 and in Western Australia the prevalence of FASD in youth detention was 36%.[15,16] Furthermore,

in one Canadian study, individuals with FASD were found to have a life expectancy of 34 years, 42% lower than that of the general population.[17]

FASD has also been shown to have tremendous impacts on families and society. Caregivers have reported a wide range of needs, concerns and high levels of stress when caring for individuals with FASD.[18] The time taken to care for individuals with FASD usually comes at the cost of reduced working time, and thus leads to productivity and salary loss within the family.[19] Additionally, there are huge economic consequences associated with FASD. A study conducted in Sweden revealed that at the societal level the annual direct and indirect costs for all individuals with FASD were estimated to be around €1.6 billion.[19] Another study conducted in the United States found that the annual cost for all persons with FASD, inclusive of direct and indirect costs, was found to range from USD$74.6 million to USD$3.9 billion.[20] Overall, the impact of FASD is pervasive at the individual, family, and societal level.

1.3 FASD context in Western Australia

Available data suggest a growing prevalence of FASD in Western Australia. A study based on the Western Australian Register of Developmental Anomalies data reported a twofold increase in FASD notifications in Western Australia over the past 30 years, in both Aboriginal and non-Aboriginal children.[21] The prevalence of FASD in Aboriginal children born between 1980 and 1989 was 3 per 1000 births but increased to 6 per 1000 births for those born between 2000 and 2010.[21] Children with FASD were reported to be predominately born to mothers with a rural place of residence.[21] Due to inconsistencies and limitations in the way prenatal alcohol exposure (PAE) has been measured and reported over this period, it is not possible to determine whether this is due to increasing prevalence of PAE/FASD or increased awareness and training of health professionals in FASD identification. However, given the considerable efforts in recent years to deliver professional education about FASD to health professionals, it is likely that this contributes, at least in part, to rising prevalence figures.

Another population-based study found that the prevalence of FASD in children born in 2002 and 2003 in a remote community in Western Australia was 120 per

1000 births,[22] of which 98% (106/108) of the children were Aboriginal. Thirteen of these children received a formal diagnosis of FASD for the first time at 7 to 10 years of age.[22] A research by Telethon Kids Institute also reported 36% (36/99) of the young people who participated in a FASD prevalence study at a youth justice setting in Western Australia were diagnosed with FASD for the first time at 13 to 17 years of age.[16] Among the young people in detention, 74% (73/99) were Aboriginal, and 51% lived in metropolitan communities while 49% lived in rural, regional, or remote communities.[16] The findings from these studies highlight the increasing prevalence of FASD in Western Australia in recent years and a significantly higher prevalence of FASD in some settings in Western Australia compared to the global prevalence of the disorder.

The rising prevalence of FASD in Australia has been recognised by the Australian Government. Since 2014, the Australian Government has indicated its commitment to reduce the impact of FASD in Australia.[23] In the Portfolio Budget Statement 2016-2017,[24] funds were allocated to support FASD health service delivery and education initiatives to reduce the prevalence and impact of FASD, as well as promote responsible alcohol consumption.[24] The National FASD Strategic Action Plan 2018-2028 was also introduced, detailing specific priorities and opportunities to improve the prevention, diagnosis, and management of FASD in Australia.[25] The National FASD Strategic Action Plan aimed to reduce prevalence of FASD, reduce the associated impact of FASD, and improve the quality of life for people living with FASD.[25]

More recently, the Sustainable Health Review report to the Western Australian Government was published; one of its strategies was to provide pregnant women with support and children with the best start over the first 1000 days of life.[26] The focus on support during pregnancy and the first 1000 days of life is significant because FASD is preventable during pregnancy.[26] Recommendations from the Sustainable Health Review report pertaining to FASD included the introduction and evaluation of further targeted, cross-agency approaches to reduce the incidence of alcohol consumption during pregnancy, and for screening and management of the impairments of FASD, as well as the review of the FASD Model of Care 2010, which outlined approaches to addressing FASD.[27]

Implementation of the recommendations from the Sustainable Health Review report are not without challenges. One key resource required for FASD screening is a short, accurate, and cost-effective screening tool that can identify at-risk individuals. Some desired features of a FASD screening tool include:[28,29] (1) simple, safe, and precise; (2) validated to identify features associated with FASD; (3) acceptable to the population; and (4) when appropriate leads to further evaluation, diagnostic investigation, and inclusion in a management plan. However, a FASD screening tool with these features has yet to be identified for use in Australia.[30] Additionally, given that a FASD diagnosis relies on PAE, FASD screening efforts are closely linked to the need for reliable and consistent screening and reporting of PAE.

Historically, screening for FAS has largely focused on identification of the characteristic facial anomalies associated with this condition (now referred to as FASD with three facial features) - short palpebral fissures, smooth philtrum and thin upper lip.[31] Facial photographs to detect these anomalies have been effectively used in high-risk populations in North America.[32] However, screening for FASD is more difficult as the majority of individuals with FASD do not have these facial features.[30] Furthermore, the neurobehavioral features associated with FASD are not unique to this disorder, making it difficult to differentiate between individuals with FASD and those that have developmental delays and neurodevelopmental disorders with different aetiology.[33]

Another challenge is that FASD screening needs to sit within an appropriate model of care which guides the delivery of best practice supports and services to the intended population. Best practice encompasses understanding and responding to the health needs of the population, providing equitable and integrated care, and ensuring effective evaluation of health services and quality improvements.[34,35] Overall, the identification of a FASD screening tool and a review of the FASD Model of Care are warranted.

In response to these needs, the *Screening for FASD in Western Australia: Policy and Practice Recommendations* report seeks to provide policymakers, health-care providers, individuals, families, and communities affected by FASD with clear, objective, and independent and up-to-date recommendations on FASD screening tools and best practice principles for implementation in Western

Australia. These recommendations are intended to inform decisions about the feasibility and costs of integrating evidence into policy and practice, as well as the need for workforce training and ongoing evaluation.

2 Objectives, target audience and scope

The aims of the current report were (1) to provide an overview of FASD screening tools available internationally; (2) to provide recommendations on the implementation of available FASD screening tools in Western Australia; and (3) to provide recommendations on FASD screening approach in Western Australia to underpin implementation of FASD screening in the future.

The primary target audiences of the current report are policymakers and health-care providers. The secondary target audiences are individuals, families and communities affected by FASD. While screening for PAE is closely linked to screening for FASD, the scope of the current report does not include recommendations regarding tools for screening of alcohol biomarkers in pregnant women or newborns, or alcohol use behaviours in pregnant women. The following questions were addressed:

i. What FASD screening tools are available internationally?

ii. Which FASD screening tools are recommended for use in Western Australia?

iii. What are the recommended best practices and guiding principles for implementation of FASD screening approaches in Western Australia?

iv. What constitutes "best practice principles" for models of care integrating FASD screening in Western Australia?

3 Development process

3.1 Review groups

FASD Screening Working Group: Members of the Working Group were staff from the Department of Health Western Australia, and staff from the FASD Research Australia Centre of Research Excellence, Telethon Kids Institute. The Working Group contributed to the planning and oversight of the processes involved in the report development, reviewed the research questions, advised on the establishment of the FASD Screening Advisory Group and ensured that all the processes were carried out with objectivity and independence.

FASD Screening Project Group: Members of the Project Group were staff from the FASD Research Australia Centre of Research Excellence, Telethon Kids Institute. The Project Group synthesised the evidence from empirical studies and produced the evidence-based recommendations.

FASD Screening Advisory Group: Members of the Advisory Group were invited in their individual capacities. They represent different key stakeholder representatives with expertise and experience in the field of FASD, including researchers, clinicians, policymakers, and consumers (refer to List of contributors on page 2). The Advisory Group was involved in the provision of advice on the evidence-based recommendations (refer to Terms of reference for the Advisory Group in Appendix A).

Chair of the FASD Screening Advisory Group: The chair of the Advisory Group facilitated discussions on methodological and content issues during the discussion of the evidence-based recommendations.

3.2 Management of conflict of interest

All members of the Advisory Group declared no conflict of interest in relation to the report development. All documents circulated within the Advisory Group were considered strictly confidential during the report development process.

3.3 Decision-making

The members of the Working Group drafted the current report, refined the PICO (patient, intervention, comparison, outcome) questions and identified possible outcomes (Table 1). The Advisory Group provided input on the evidence synthesis and on the direction of the recommendations during the Advisory Group consensus meetings. An evidence-to-recommendations decision tool was used to guide the decision-making process (Appendix B). The recommendations were agreed by consensus. This means that recommendations were accepted when the majority of the Advisory Group members agreed with them and there was no major objection to acceptance.

Table 1. PICO question for the evidence review.

Patients	Intervention	Comparison	Outcomes	Study design
Individuals at risk of FASD	Screening and management of FASD	No screening and management of FASD	▪ True positive ▪ False negative ▪ False positive ▪ True Negative	Empirical studies

3.4 Review of the evidence for FASD screening tools

The Project Group synthesised the evidence and prepared the evidence profiles according to the Grading of Recommendations Assessment, Development and Evaluation (GRADE) framework.[1] The evidence was synthesised using a systematic review design. The review included only empirical studies that (i) involved individuals with PAE or FASD; (ii) examined tools designed for screening for the associated features of FASD; (iii) reported at least the sensitivity and specificity of the tool; and (iv) were peer-reviewed. Online databases were searched from the inception of each database up to May 2020 for relevant studies published in English language. The four online databases included Cumulative Index to Nursing and Allied Health Literature (CINAHL), Embase, MEDLINE and PsycINFO. Additional references were identified by reviewing bibliographies of identified studies. The search strategy for the systematic review is displayed in Appendix C.

Two members of the Project Group independently screened titles and abstracts and the full text of relevant studies; extracted data about participant's characteristics, diagnostic test accuracy and reference standard; and appraised the quality of the eligible studies using the Quality Assessment of Diagnostic Studies-2 (QUADAS-2) tool. A third investigator was involved to resolve any disagreements.

Three members of the Project Group evaluated the quality of evidence using the GRADE framework[1] and presented the evidence and its quality in the GRADE evidence report (see Supplementary material: GRADE evidence report). The quality of the evidence was assessed as high, moderate, low, or very low, according to the GRADE criteria (Table 2). The GRADE evidence report uses the evidence-to-recommendation form to facilitate decision-making for recommendations (see Appendix B). The form includes a summary of the evidence (benefits and harms), an assessment of the quality of the evidence, relevant patient values and preference, and any implication for use of resource, acceptability, and feasibility.

Table 2. Definitions for rating of the quality of evidence (GRADE).

Ratings	Implications of a strong recommendation	Implications of a conditional recommendation
High	This research provides a very good indication of the likely effect. The likelihood that the effect will be substantially different* is low.	This evidence provides a very good basis for deciding about whether to implement the intervention. Impact evaluation and monitoring are unlikely to be needed if it is implemented.
Moderate	This research provides a good indication of the likely effect. The likelihood that the effect will be substantially different* is moderate.	This evidence provides a good basis for deciding about whether to implement the intervention. Monitoring of the impact is likely to be needed and impact evaluation may be warranted if it is implemented.
Low	This research provides some indication of the likely effect. However, the likelihood that it will be substantially different* is high.	This evidence provides some basis for deciding about whether to implement the intervention. Impact evaluation is likely to be warranted if it is implemented.

Ratings	Implications of a strong recommendation	Implications of a conditional recommendation
Very low	This research does not provide a reliable indication of the likely effect. The likelihood that the effect will be substantially different* is very high.	This evidence does not provide a good basis for deciding about whether to implement the intervention. Impact evaluation is very likely to be warranted if it is implemented.

* Substantially different: large enough difference that it might influence a decision.

3.5 Development of recommendations regarding FASD screening tools

The Advisory Group and Project Group met to discuss the recommendations. The meeting was led by the chair of the Advisory Group. Members of the Project Group presented the GRADE evidence report to the Advisory Group. Members of the Advisory Group provided input on the acceptability and feasibility sections of the GRADE evidence report via an online survey (refer to Appendix D for the compilation of comments from the Advisory Group). The results of those discussions are documented in the evidence-to-recommendation tables for each recommendation (see Supplementary material: GRADE evidence report). The Advisory Group also identified key research gaps, which are recorded in Appendix D.

The recommendations were assessed as 'strong recommendation' or 'conditional recommendation' (depending on screening context) according to the GRADE approach.[1] Of the eight recommendations in the current report, five were rated as strong recommendations.

3.6 Development of recommendations regarding implementation of FASD screening approaches and principles for a FASD Model of Care

The Project Group conducted a rapid review to synthesise the evidence regarding best practice FASD screening approaches and principles for a FASD Model of Care. Two comprehensive systematic searches were performed. The review for best practice FASD screening approaches included evidence that (i)

involved individuals with PAE or FASD; (ii) involved community child health practice; (iii) examined screening approaches; and (iv) were peer-reviewed. The review for principles for a FASD Model of Care included evidence that (i) involved community child health practice; (ii) examined model of care; and (iii) were peer-reviewed or grey literature. Online databases searched were limited to articles published in the English language between 2010 and November 2020. The three online databases included Cumulative Index to Nursing and Allied Health Literature, Embase and Medline. Additional references were identified by reviewing bibliographies of identified studies. The search strategy for the literature reviews on FASD screening approaches is displayed in Appendix C.

3.7 Recommendation review and approval process

The current report underwent the following peer review processes during development.

- The protocol for the systematic review of FASD screening tools was circulated to the Advisory Group. Evidence identified through the systematic review, the evidence profiles and the evidence-to-recommendation outcomes were discussed with the Advisory Group.
- Additional recommendations regarding best practice for FASD screening in Western Australia were noted throughout the Advisory Group meetings and evidence review process.
- The final recommendation draft was circulated among the members of the Advisory Group for review before submission to the Department of Health Western Australia.

4 Evidence for FASD Screening Tools

This section summarises the main findings of the systematic review and of the evidence profile. The search of evidence resulted in 778 citations of which 14 studies were included in the systematic review (Figure 1). Of the 14 studies, eight used a case-control study design and six used a cross-sectional study design (see Appendix C for the reference list of all studies included in the systematic review).

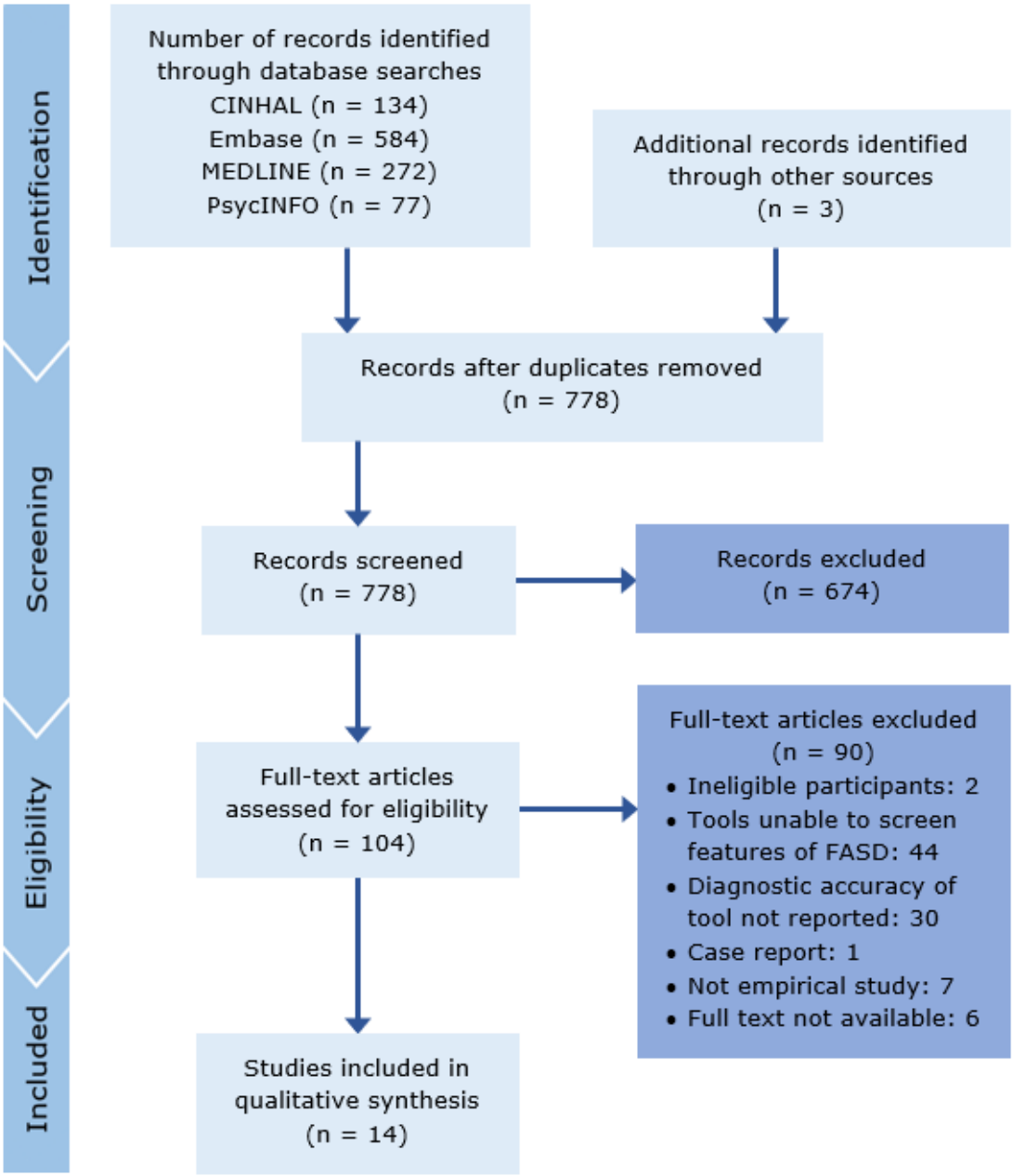

Figure 1. PRISMA flow diagram of search and selection process.

Participant

Across the studies reviewed, a total of 4,483 participants were included, comprising individuals with FASD and typically developing individuals. Five of the screening tools identified were tested with individuals with FAS. Most of the participants were aged 0 to 18 years; only two studies included participants above 18 years old.[36,37] Nine of the studies were conducted in the United States and five were conducted in Canada. Studies were conducted in various settings including FASD clinics, FASD support centres, genetics outreach clinics, government agencies, and in the community (Table 3).

Table 3. Characteristics of the screening tools.

Screening tool	Case	Case prevalence	Reference standard	Setting	Psychometric properties (%)	Administration time and cost
NST	FASD	-	Canadian Guidelines and 4-Digit Diagnostic Code	FASD Clinic	Sensitivity range: 63-98 Specificity range: 42-100	22 minutes, CAD$20[a]
Eye movement behaviour assessment via machine learning	FASD	-	Canadian Guidelines	Community	Sensitivity range: 73-77 Specificity range: 79-91	17-20 minutes, CAD$50
TREIN task	FASD	-	4-Digit Diagnostic Code	FASD Clinic	Sensitivity: 54 Specificity: 96	Not reported
Dysmorphic examination via photographs	FAS	10-15 per 1000 sample population	Expert opinion and 4-Digit Diagnostic Code	FAS Clinic and Division of Children and Family Services	Sensitivity: 100 Specificity: 100	30 minutes, cost not reported
Physical and dysmorphic examination	FAS	200 per 1000 sample population	Gestalt method	FAS Clinic	Sensitivity: 100 Specificity: 89	Not reported

Screening tool	Case	Case prevalence	Reference standard	Setting	Psychometric properties (%)	Administration time and cost
Craniofacial measurements approach	FAS and PFAS	-	IOM criteria	Research centre and FAS support centre	Sensitivity: 98 Specificity: 90	Not reported
FAS Screen	FAS and PFAS	4.3 per 1000 sample population	Gestalt method and IOM criteria	School	Sensitivity: 100 Specificity range: 94-95	8-15 minutes, USD$8-13
FAS diagnostic checklist	FAS and PFAS	-	IOM criteria	Genetics Outreach Clinic	Sensitivity: 89 Specificity: 72	Not reported

[a], information derived from Berrigan (2019); CAD, Canadian dollars; FAS, fetal alcohol syndrome; FASD, fetal alcohol spectrum disorder; IOM, Institute of Medicine; NST, Neurobehavioral Screening Test; TREIN, Tallying Reference Errors in Narrative; USD, United States dollar.

Screening tool

Among the 14 studies, eight different FASD screening tools were identified: (1) The Neurobehavioral Screening Tool; (2) Eye movement behaviour assessment via machine learning; (3) Tally Reference Errors in Narrative Task; (4) Dysmorphic examination via photographs; (5) Physical and dysmorphic examination; (6) Craniofacial measurements approach; (7) The FAS Screen; and (8) The FAS diagnostic checklist (Table 3). All screening tools were administered and scored by trained assessors and/or medical professionals, except for the eye movement behaviour assessment via machine learning and dysmorphic examination via photographs screening tools, which were processed using computer software. Sensitivities of the screening tools ranged from 53.6% to 100% and specificities ranged from 55% to 100%.

The eight screening tools assessed between one and eight of the 12 features/functions identified in the diagnostic criteria for FASD (Table 4). Notably, only the FAS diagnostic checklist assessed eight of these features/functions.

Reference standard

Across the 14 studies, each used either one or two reference standards to diagnose individuals with FASD. The most frequently employed reference standards were the Canadian Guidelines (4 studies) and the 4-Digit Diagnostic Code (4 studies), followed by the Gestalt method (3 studies), the Institute of Medicine (IOM) criteria (3 studies) and expert opinion (1 study). Only one study did not report the reference standard used. The 4-Digit Diagnostic Code, Gestalt method and IOM criteria were used to diagnose individuals with FAS, PFAS and ARND; these diagnostic terms are used in the United States but not in the Australian Guide to the diagnosis of FASD.

Study quality

The quality of the studies in the review was mainly rated at high risk of bias due to the methodology around participant selection and flow and timing (Figure 2). Participant selection was considered at high risk of bias in 9 studies, largely due to the use of case-control study design. Ten studies had study flow issues that were rated at high risk of bias, either due to a failure to include all participants in the analysis or a failure to apply the same reference standard to all participants. There were concerns about the applicability of the studies predominantly around the reference standard used in relation to the Australian Guide to the diagnosis of FASD. Six studies received high concerns as the target conditions as defined by the reference standard were FAS, PFAS and ARND, and not FASD (with and without sentinel facial features) (Figure 2).

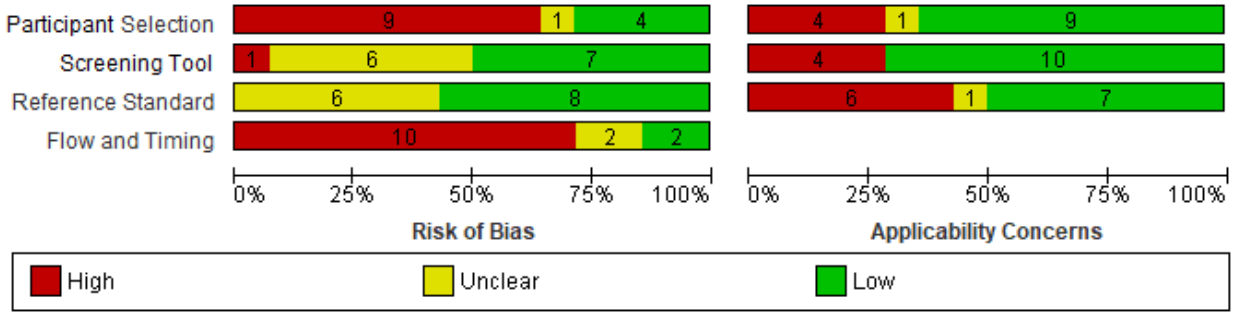

Figure 2. QUADAS-2 risk of bias and applicability concern graph of the reviewers' judgement of each domain presented as percentage across the included studies.

Table 4. Features/functions assessed by the screening tools that are related to the scope of the Australian Guide to the diagnosis of FASD.

Features/functions	**Screening tools**							
	NST	Eye movement behaviour via machine learning	TREIN task	Dysmorphic examination via photographs	Physical and dysmorphic examination	Craniofacial measurements approach	FAS Screen	FAS diagnostic checklist
Prenatal alcohol exposure								✓
Brain structure (HC)/ neurology					✓	✓		✓
Motor skills		✓						
Cognition							✓	✓
Language			✓				✓	✓
Academic achievement								✓
Memory								
Attention	✓	✓					✓	✓
Executive function, including impulse control and hyperactivity	✓						✓	✓
Affect regulation								
Adaptive behaviour, social skills, or social communication	✓							
Sentinel facial features including short palpebral fissure, smooth philtrum, and thin upper lip				✓	✓	✓	✓	✓

FAS, fetal alcohol syndrome; HC, head circumference; NST, Neurobehavioral Screening Test; TREIN, Tallying Reference Errors in Narrative

Evidence profile

The evidence for diagnostic accuracy of the screening tools is presented in the GRADE evidence profile (Appendix E, Supplementary tables 1 to 8). Across the eight screening tools, the evidence for diagnostic accuracy of the screening tools was rated as very low (2 screening tools), low (3 screening tools) and moderate (3 screening tools).

5 Recommendations regarding FASD screening tools

5.1 Important considerations that apply to all FASD screening recommendations

Population targeted by the recommendations

The recommendations apply to screening of individuals at risk for FASD who are between ages 0 to 18 years. While these recommendations were formulated for Western Australia, they may be extrapolated to other jurisdictions in Australia. We considered the appropriateness of the screening tool for universal screening, as well as targeted screening for subgroups with whom elevated prevalence of FASD has been previously reported, including individuals in out-of-home care, justice-involved individuals, and individuals receiving special education.[8]

Considerations for screening tools

The recommendations relate to eight screening tools designed to screen for FASD or FAS: (1) The Neurobehavioral Screening Tool; (2) Eye movement behaviour assessment via machine learning; (3) Tally Reference Errors in Narrative Task; (4) Dysmorphic examination via photographs; (5) Physical and dysmorphic examination; (6) Craniofacial measurements approach; (7) The FAS Screen; and (8) The FAS diagnostic checklist. The Dysmorphic examination via photographs, Physical and dysmorphic examination, Craniofacial measurements approach, FAS Screen and FAS diagnostic checklist include the evaluation of sentinel facial features. It should be noted that all recommendations relate to the use of these tools to screen for FASD (with and without sentinel facial features).

5.2 FASD screening tool recommendations

It is crucial to note that the following recommendations were formulated based on comparing the *specified FASD screening tool* with *no FASD screening tool* (refer to section 3.3, Decision-making on page 17). Hence, some of the summary of the evidence entries in the GRADE framework are similar across all eight screening tools. For example, the potential benefits of using a FASD screening tool versus no FASD screening tool and the potential harms of using a FASD screening tool versus no FASD screening tool (see Supplementary material: GRADE evidence report for details). The general potential benefits and harms of FASD screening proposed by the Advisory Group and evidence from the literature[38] are presented in Table 5. It is also essential to note that some of the screening tools identified in the systematic review are useful in certain settings and can be used in situations where at-risk individuals require further evaluation for FASD. For example, some of the tools are useful to screen for FASD with facial features only but should not be used as part of a broader screening approach that aims to identify people with FASD with and without sentinel facial features. Overall, the following recommendations should be interpreted based on the context of the current report.

Recommendation 1. Conditional recommendation against the use of the Neurobehavioral Screening Test (NST) to screen for individuals at risk of FASD in Western Australia.

Summary of the evidence: Four studies evaluated the diagnostic accuracy of the NST. This is a 10-item caregiver reported questionnaire that identifies at-risk individuals through the measurement of attention, executive function, adaptive behaviour, and social skills. The diagnostic test accuracy of the NST is variable. The quality of evidence of the diagnostic test accuracy, management effects and effects of the NST in the screening of FASD overall is low. The potential benefits of using the NST for FASD screening include early detection of FASD and early access to management strategies. The potential harms due to false positives with the NST are presented in Table 5. The evidence concerning the resources required to implement the NST in Australia is limited. The NST was not considered by the

Advisory Group to be acceptable and feasible for use in Western Australia. Research to determine the accuracy and cost-effectiveness of the NST as a targeted screening tool in Western Australia is warranted.

Recommendation 2. Conditional recommendation against the use of the eye movement behaviour assessment via machine learning to screen for individuals at risk of FASD in Western Australia.

Summary of the evidence: Two studies evaluated the diagnostic accuracy of the eye movement behaviour assessment via machine learning. This approach uses eye- tracking data, collected while individuals watch video clips, and a machine learning algorithm, to identify at-risk individuals through the measurement of visual motor skills and attention. The diagnostic test accuracy of the eye movement behaviour assessment via machine learning is inaccurate. The quality of evidence of the diagnostic test accuracy, management effects and effects of the eye movement behaviour assessment via machine learning in the screening of FASD overall is low. The potential benefits of using the eye movement behaviour assessment via machine learning for FASD screening are presented in Table 5. The potential harms of using an inaccurate screening tool that could give rise to false positive and negative screen results are also presented in Table 5. The evidence concerning the resources required to implement the eye movement behaviour assessment via machine learning in Australia is limited. While the eye movement behaviour assessment via machine learning was considered potentially acceptable by the Advisory Group for use in Western Australia, they did not consider this screening tool feasible for use in Western Australia.

Recommendation 3. Strong recommendation against the use of the Tallying Reference Errors in Narrative (TREIN) task to screen for individuals at risk of FASD in Western Australia.

Summary of the evidence: One study evaluated the diagnostic accuracy of the TREIN task. The TREIN task uses story narration data from individuals to identify at-risk individuals through the measurement of language skills. The diagnostic test accuracy of the TREIN task is very inaccurate. The quality of evidence of the

diagnostic test accuracy, management effects and effects of the TREIN task in the screening of FASD overall is low. The potential benefits of using the TREIN task for FASD screening are presented in Table 5. The potential harms of using an inaccurate screening tool that could give rise to false positive and negative screen results are also presented in Table 5. The evidence concerning the resources required to implement the TREIN task in Australia is limited. The TREIN task was not considered by the Advisory Group to be acceptable and feasible for use in Western Australia.

Recommendation 4. Strong recommendation against the use of the dysmorphic examination via photographs to screen for individuals at risk of FASD in Western Australia.

Summary of the evidence: Two studies evaluated the diagnostic accuracy of the dysmorphic examination via photographs. This involves a 3-item examination of the palpebral fissure, philtrum and upper lip using a person's headshot photograph to identify at-risk individuals through the measurement of facial features. The diagnostic test accuracy of the dysmorphic examination is very accurate for the screening of individuals with sentinel facial features only. The quality of evidence of the diagnostic test accuracy, management effects and effects of the dysmorphic examination in the screening of FASD overall is very low. The potential benefits of using the dysmorphic examination for FASD screening include early detection of FASD with sentinel facial features and thus early access to management strategies (Table 5). The potential harms include not able to detect FASD without sentinel facial features. Potential harms due to false positives with the dysmorphic examination are presented in Table 5. The evidence concerning the resources required to implement the dysmorphic examination in Australia is limited. The dysmorphic examination was not considered by the Advisory Group to be acceptable and feasible for use in Western Australia.

Recommendation 5. Conditional recommendation against the use of the physical and dysmorphic examination to screen for individuals at risk of FASD in Western Australia.

Summary of the evidence: One study evaluated the diagnostic accuracy of the physical and dysmorphic examination. It is a 16-item physical examination checklist that identifies at-risk individuals through the measurement of facial features and brain structures (head circumference). The diagnostic test accuracy of the physical and dysmorphic examination is accurate for the screening of individuals with sentinel facial features only. The quality of evidence of the diagnostic test accuracy, management effects and effects of the physical and dysmorphic examination in the screening of FASD overall is low. The potential benefits of using the physical and dysmorphic examination for FASD screening include early detection of FASD with sentinel facial features and thus early access to management strategies (Table 5). The potential harms include not able to detect FASD without sentinel facial features. Potential harms due to false positives are presented in Table 5. The evidence concerning the resources required to implement the physical and dysmorphic examination in Australia is limited. The physical and dysmorphic examination was not considered by the Advisory Group to be acceptable and feasible for use in Western Australia.

Recommendation 6. Strong recommendation against the use of the craniofacial measurement approach to screen for individuals at risk of FASD in Western Australia.

Summary of the evidence: One study evaluated the diagnostic accuracy of the craniofacial measurement approach. It is a 22-item examination checklist that identifies at-risk individuals through the measurements of facial features and brain structures (head circumference). The diagnostic test accuracy of the craniofacial measurement approach is accurate for the screening of individuals with sentinel facial features only. The quality of evidence of the diagnostic test accuracy, management effects and effects of the craniofacial measurement approach in the screening of FASD overall is very low. The potential benefits of using the craniofacial measurement approach for FASD screening include early detection of FASD with sentinel facial features and thus early access to management strategies (Table 5). The potential harms include not able to detect FASD without sentinel facial features. Potential harms due to false positives are presented in Table 5. The

evidence concerning the resources required to implement the craniofacial measurement approach in Australia is limited. The craniofacial measurement approach was not considered by the Advisory Group to be acceptable and feasible for use in Western Australia.

Recommendation 7. Strong recommendation against the use of the FAS Screen to screen for individuals at risk of FASD in Western Australia.

Summary of the evidence: Two studies evaluated the diagnostic accuracy of the FAS Screen. It is a 30-item examination checklist that identifies at-risk individuals through the measurements of cognition, language, attention, executive function, and facial features. The diagnostic test accuracy of the FAS Screen is accurate for the screening of individuals with sentinel facial features only. The quality of evidence of the diagnostic test accuracy, management effects and effects of the FAS Screen in the screening of FASD overall is low. The potential benefits of using the FAS Screen for FASD screening include early detection of FASD with sentinel facial features and thus early access to management strategies (Table 5). The potential harms include not able to detect FASD without sentinel facial features. Potential harms due to false positives with the FAS Screen are presented in Table 5. The evidence concerning the resources required to implement the FAS Screen in Australia is limited. The FAS Screen was not considered by the Advisory Group to be acceptable and feasible for use in Western Australia.

Recommendation 8. Strong recommendation against the use of the FAS diagnostic checklist to screen for individuals at risk of FASD in Western Australia.

Summary of the evidence: One study evaluated the diagnostic accuracy of the FAS diagnostic checklist. It is a 62-item examination checklist that identifies at-risk individuals through the measurements of PAE, brain structure, cognition, language, attention, executive function, and facial features. The diagnostic test accuracy of the FAS diagnostic checklist is accurate for the screening of individuals with sentinel facial features only. The quality of evidence of the diagnostic test accuracy, management effects and effects of the FAS diagnostic checklist in the screening of

FASD overall is low. The potential benefits of using the FAS diagnostic checklist for FASD screening include early detection of FASD with sentinel facial features and thus early access to management strategies (Table 5). The potential harms include not able to detect FASD without sentinel facial features. Potential harms due to false positives with the FAS diagnostic checklist are presented in Table 5. The evidence concerning the resources required to implement the FAS diagnostic checklist in Australia is limited. The FAS diagnostic checklist was not considered by the Advisory Group to be acceptable and feasible for use in Western Australia.

Table 5. General potential benefits and harms of FASD screening.

Potential benefits
▪ True positive screen result enables early detection and diagnosis of FASD. ▪ True negative screen result provides assurance that the individual is not affected by FASD. ▪ Diagnosis of FASD enables early access to healthcare and management services and supports for individuals and their families, which may prevent serious adverse outcomes associated with FASD (e.g., mental illness, early death via youth suicide, and committing crimes). ▪ Diagnosis of FASD enables high cost savings for the individual, family and society with appropriate diagnosis, education and supports. ▪ Diagnosis of FASD enables the individual to connect with FASD-related support organisations for help. ▪ Diagnosis of FASD provides explanation for difficulties that the individual and family may encounter and an end to their search for explanations. ▪ Diagnosis of FASD enables the calculation of prevalence and incidence to inform FASD prevention, screening, diagnostic, and management strategies. ▪ Diagnosis of FASD may help to prevent subsequent alcohol exposed pregnancies through targeted prevention approaches.
Potential harms
▪ False negative screen result may prevent individuals from receiving appropriate diagnostic assessment and management of FASD. ▪ False positive screen result may lead to burden on caregivers in relation to time/resource burden and emotional burden. ▪ FASD-related evaluation may result in anxiety and distress associated with FASD in individuals and their families. ▪ FASD-related evaluation and management may result in high resource use and financial cost. ▪ The identification of FASD may result in negative stigmatisation, blame and shame associated with FASD in individuals, their families, and communities. ▪ The identification of FASD may draw attention away from other significant issues of the individual. ▪ The identification of FASD may result in the perception of disability in the individuals that may limit his/her potential to live beyond his/her disorder. ▪ The identification of FASD may lead to the infringement of personal autonomy and disempowerment in cases where government may, for instance, decided to limit access to alcohol certain populations or areas.

6 Review of principles to guide implementation of FASD screening

This section summarises the main findings of the literature review on best practice principles underlying FASD screening. The search of evidence resulted in 21 citations, of which four studies were included in the review. The studies were conducted in various countries, including South Africa (1 study), the United States (1 study), Canada (1 study) and Australia (1 study). Two studies used a qualitative/report study design, one used a cross-sectional study design, and one used a mixed-methods study design. Only one study reported consensus regarding recommendations for FASD screening; a Delphi study by Watkins and colleagues[30] used information from the literature as well as from Australian health professionals with experience or expertise in FASD screening or diagnosis to inform their recommendations for FASD screening.

6.1 FASD screening approaches

Three types of FASD screening approaches are possible – universal, targeted, and selective (Table 6). The universal approach refers to the screening of the entire population, or particular age groups; for example, early childhood.[39] The selective approach refers to the screening of selected populations known to have a high risk of developing a disease.[39] Lastly, the targeted approach refers to the screening of people at risk; for example, people who show signs of a disorder.[39] The type of screening approaches employed by the studies differ based on the country of study origin and populations involved in the study.

Table 6. FASD screening approaches.

Screening approach	Description
Universal	Screening approaches are implemented in the entire population, regardless of risk.
Targeted	Screening approaches are implemented with individuals showing signs of a disorder (e.g., where there is parental concern that a child may have FASD).
Selective	Screening approaches are implemented for select populations known to report a higher prevalence of FASD (e.g., juvenile justice settings).

Universal screening approach

In the studies identified in our review, one study[40] used a universal approach to screen for FASD. The study by O'Connor and colleagues[40] originated from South Africa, a country with a high prevalence of FASD at 29-290 per 1000 births. Notably, the prevalence of FASD in South Africa is 4 to 36 times more than the global prevalence of FASD.[41] O'Connor and colleagues[40] applied the universal screening approach in communities in South Africa to identify 18-month-old children at-risk of FASD. The identification of at-risk children was conducted through a two stage screening and diagnostic method; the FASD screening process was performed by trained nonmedical community workers and the FASD diagnostic process was performed by trained paediatricians. The screening process in stage I included the evaluation of key features of FAS: (1) growth restriction, (2) two of the three sentinel facial feature of FAS, (3) central nervous system dysfunction including small occipitofrontal head circumference, and (4) prenatal alcohol exposure. All screen-positive children were referred for a more comprehensive stage II examination and diagnosis. Of the 14 children screened positive for referral for further diagnostic evaluation, 93% (13/14) received an FASD diagnosis.[40] Considerations noted in this study relevant to FASD screening approaches included: (1) the employment of trained nonmedical community workers of the same cultural/ethnic background of individuals with FASD and families to be involved in FASD screening;[40] and (2) the need for specialised services for both individuals with FASD and their families.[40]

Targeted screening approach

A Delphi study by Watkins and colleagues[30] used information from the literature as well as from Australian health professionals with experience or expertise in FASD screening or diagnosis to inform their recommendations for FASD screening. The study recommended the use of targeted approach for FASD screening in individuals who present with features associated with FASD, such as parental concerns that their child might have FASD, developmental delay, structural and functional central nervous system abnormalities, facial anomalies, or problems with behaviour.[30] Other considerations noted in this study relevant to FASD screening approaches included: (1) the need to screen for PAE; (2) screening for FASD during childhood rather than at birth; (3) the need for increased service capacity for FASD screening; and (4) ensuring adequate early diagnostic and specialised intervention services to support individuals with FASD and their families.[30]

Selective screening approach

Three studies[30,42,43] made recommendations regarding selective FASD screening approaches for high risk populations in the United States, Canada, and Australia. At-risk individuals, such as children in foster/state care and children of mothers who received alcohol treatment services, were recommended to be screened for FASD via neurodevelopmental screening assessments, which can take place as early as at birth.[30,42,43] Additional considerations noted in these studies relevant to FASD screening approaches included: (1) the need to implement routine screening for all mothers and children entering foster care system;[42] (2) training staff and healthcare professionals to be comfortable asking questions related to problematic alcohol or substance use;[42,43] (3) the need for community partnership across all systems and services supporting individuals with FASD and their families;[42] and (4) ensuring adequate early intervention services to support individuals with FASD and their families.[30]

Overall, FASD screening approaches adopted will depend on the community context. The synthesised evidence suggests that there is no "one size fits all approach" to FASD screening as it is influenced by the prevalence of FASD and the

prevalence of high-risk populations in the community/country of interest. In addition to FASD screening, the evidence highlights the importance of PAE screening, the need for increased coordination of information sharing and management care for both individuals with FASD and their families, as well as the need for adequate specialised early intervention services to support individuals with FASD (Refer to Table 7 for the summary of the considerations for different FASD screening approaches).

Table 7. Summary of the considerations for different FASD screening approaches.

Screening approach	Consideration
Universal	▪ Used in countries with high prevalence of FASD. ▪ Early childhood FASD screening, as early as 18 months. ▪ Employment of trained nonmedical community workers of the same cultural/ethnic background of individuals with FASD and families to be involved in FASD screening. ▪ Need for specialised services for both individuals with FASD and their families.
Targeted	▪ Used in individuals who present with features associated with FASD. ▪ Need to screen for PAE. ▪ Screen for FASD during childhood rather than at birth. ▪ Need for increased service capacity for FASD screening. ▪ Ensure adequate early diagnostic and specialised intervention services to support individuals with FASD and their families.
Selective	▪ Used in individuals from populations at high risk of FASD. ▪ Need to implement routine screening for all mothers and children – FASD screening via neurodevelopmental screening can be as early as at birth. ▪ Train staff and healthcare professionals to be comfortable asking questions related to problematic alcohol or substance use. ▪ Need for community partnership across all systems and services supporting individuals with FASD and their families. ▪ Ensure adequate early intervention services to support individuals with FASD and their families.

7 Recommendations regarding best practices and guiding principles for FASD screening approaches in Western Australia

It is important to note that even though no FASD screening tools are recommended for use in Western Australia, the following recommendations for FASD screening approach have been formulated with the view that appropriate screening tools are required and will be developed in the future. It should be noted that all recommendations relate to the screening approach for FASD (with and without sentinel facial features).

According to the evidence found in the review on FASD screening approaches, recommendations regarding best practice FASD screening approaches in Western Australia should be tailored to the screening context. Information relating to FASD in Western Australia, while limited, shows a growing prevalence of FASD in Western Australia (see FASD context in Western Australia on page 11). Even though the prevalence of FASD in Western Australia at 6 per 1000 births is comparable to the global prevalence of FASD at 8 per 1000 births,[21,41] the prevalence of FASD is significantly increased in high risk populations, such as in remote communities and the youth justice system.[16,22] Given the varying prevalence of FASD across different population groups in Western Australia and the high prevalence of alcohol use during pregnancy among Australian women,[44] a combination of FASD and PAE screening approaches is recommended.

The potential benefits and harms of different screening approaches must also be considered when making recommendations regarding FASD screening in Western Australia (see Advisory Group comments in Appendix D). For example, universal screening reduces the risk of missed cases and can enable earlier diagnosis and support.[30] However, universal screening is also resource-intensive and may not be cost-effective. Another essential consideration is the opinions of Australian healthcare professionals when making recommendations regarding FASD screening approaches in Western Australia. A study reported that while there was no

consensus support for universal FASD screening in Australia, over 95% of healthcare professionals supported selective or targeted screening for FASD.[30] Circumstances in which selective or targeted FASD screening were supported included where there is parental concern that their child may have FASD; where there is evidence of sentinel facial features associated with FASD; where there is presentation of developmental delay in children; where there is a sibling who has been diagnosed with FASD, and where the birth mother has an alcohol use disorder during pregnancy.[30] Furthermore, the *Fetal Alcohol Spectrum Disorder Model of Care 2010*[27] and the *Western Australian interagency, state-wide Implementation Plan for the Fetal Alcohol Spectrum Disorder Model of Care 2013*[45] reports recommend the implementation of alcohol consumption in pregnancy screening for all woman in the antenatal period and further screening of FASD in at-risk children with poor outcomes from screening by community child health service providers.

7.1 Recommendations regarding FASD/PAE screening approaches

The recommendations regarding FASD/PAE screening approaches in Western Australia presented in Table 8 are guided by the evidence base, several national and state policies, directions and strategic plans, and consultation with health and community services staff.

Table 8. Recommended FASD/PAE screening approaches.

Screening approach	Description	Consideration
Universal	Implement PAE screening approaches in the entire population, regardless of risk.[27,45]	▪ Need for coherent health policies to ensure standardised manner of alcohol screening during antenatal care.[46] ▪ Need for ongoing capacity building within the health system to support the implementation of antenatal alcohol screening, education on the risk of alcohol use in pregnancy and provision of appropriate support for pregnant women who use alcohol.[46]
Targeted	Implement FASD screening approaches with individuals who: have PAE, show signs of a neurodevelopmental disorder, such as developmental delay, have a sibling who has been diagnosed with FASD, have facial anomalies associated with FASD, have observed problems with behaviour, or where there is parental concern that a child may have FASD,.[30]	▪ Need for PAE data to be linked to the child's health record to support a more accurate screening and diagnostic outcome for individuals with FASD.[47] ▪ Staff in child health services need to be trained on FASD, identification of children with developmental delay and behaviour problems, as well as referring at-risk children for further evaluation and management.
Selective	Implement FASD screening approaches in selected populations known to report a higher prevalence of FASD; for example, children referred to Child Development Services or Child and Adolescent Mental Health Services, children with mothers who receive alcohol treatment services, children in state/foster care, youth in justice settings.[30]	▪ Staff need to be trained to ask questions about alcohol use in a non-stigmatising way that encouraged open communication.[48] ▪ Provision of training on FASD identification to equip staff to screen for at-risk individuals accurately and without judgement. ▪ Staff need to be trained on cultural and language differences in relation to FASD screening.[49,50]

7.2 Recommended guiding principles for FASD screening approaches

The following principles were adapted from the *Consolidated principles for screening based on a systematic review and consensus process* by Dobrow and colleagues[51] to guide FASD screening approaches (Table 9). These guiding principles can be used to guide future development, validation, and implementation of FASD screening tools in Western Australia.

Table 9. Recommended guiding principles for FASD screening approaches.

Principle	Description
Target population for screening	The target population for screening should be clearly defined (e.g., with an appropriate target age range), identifiable and able to be reached.
Screening test performance characteristics	Screening test performance should be appropriate for the purpose, with all key components specific to the test (rather than the screening program) being accurate (e.g., in terms of sensitivity, specificity and positive predictive value) and reliable or reproducible. The test should be acceptable to the target population and it should be possible to perform or administer it safely, affordably, and efficiently.
Interpretation of screening test results	Screening test results should be clearly interpretable and determinate (e.g., with well-defined and agreed cut-off points) to allow identification of the screening participants who should (and should not) be offered diagnostic assessment and other post screening care.
Post screening test options	There should be an agreed course of action for screening participants with positive screening test results that involves diagnostic assessment, management, and follow-up care that will modify the natural history and clinical pathway for FASD; that is available, accessible, and acceptable to those affected; and that results in improved outcomes (e.g., increased functioning or quality of life, decreased cause-specific mortality). The burden of testing on all participants should be understood and acceptable, and the effect of false-positive and false-negative tests should be minimal.
Screening program infrastructure	There should be adequate existing infrastructure (e.g., financial resources, health human resources, information technology, facilities, equipment, and test technology), or a clear plan to develop adequate infrastructure, that is

Principle	Description
	appropriate to the setting to allow for timely access to all components of the screening program*
Screening program coordination and integration	All components of the screening program* should be coordinated and, where possible, integrated with the broader health care system (including a formal system to inform, counsel, refer and manage the treatment of screening participants) to optimise care continuity and ensure no screening participant is neglected.
Screening program acceptability and ethics	All components of the screening program* should be clinically, socially, culturally, and ethically acceptable to screening participants, health professionals and society, and there should be effective methods for providing screening participants with informed choice, promoting their autonomy, and protecting their rights.
Screening program benefits and harms	The expected range and magnitude of potential benefits (e.g., increased functioning or quality of life, decreased cause-specific mortality) and harms (e.g., overdiagnosis and overtreatment) for screening participants and society should be clearly defined and acceptable, and supported by existing high-quality scientific evidence (or addressed by ongoing studies) that indicates that the overall potential benefit of the screening program outweighs its potential harms.
Economic evaluation of screening program	An economic evaluation (e.g., cost-effectiveness analysis, cost–benefit analysis and cost–utility analysis) of the screening program, using a health system or societal perspective, should be conducted (or a clear plan to conduct an economic evaluation) to assess the full costs and effects of implementing, operating and sustaining the screening program while clearly considering the opportunity costs and effect of allocating resources to other potential non-screening alternatives (e.g., primary prevention, improved treatments and other clinical services) for managing FASD.
Screening program quality and performance management	The screening program should have clear goals or objectives that are explicitly linked to program planning, monitoring, evaluating, and reporting activities, with dedicated information systems and funding, to ensure ongoing quality control and achievement of performance targets.
*Components of a screening program include recruitment, testing, information access, diagnosis, referral, management, follow-up, patient education and support, staff training and program management and evaluation.	

8 Recommendations regarding the integration of FASD screening approaches into existing models of care in Western Australia

Besides identifying best practices FASD/PAE screening approaches in Western Australia, it is necessary to also identify how and when to integrate FASD (with and without sentinel facial features)/PAE screening approaches into the existing models of care of community child health services in Western Australia. This section provides a summary of the existing community child health services in Western Australia and recommendations on how FASD/PAE screening approaches can be integrated into these existing models of care.

8.1 Existing models of care of community child health services

There are several community child health services in Western Australia. For example, the Child and Adolescent Health Service in Perth Metropolitan, and the Western Australia County Health Services and Ord Valley Aboriginal Health Service in rural and remote Western Australia. However, only three of the child health services have provided recommended time points for children to be in contact with health professionals. An overview of these three community child health services is provided below.

1. Purple book appointments

The Purple book appointment is a free child health service provided by the Child and Adolescent Health Service and the Western Australia Country Health Service to all children born in Western Australia.[52,53] This community child health service includes five child health visits by the community child health nurse and a free child health record book that is parent-held. The book helps parents keep a record of their child's health and development from birth to school entry, in partnership with child health nurses and other health professionals. The nurses conduct neurodevelopmental screenings on the children at their homes at recommended time points: 0-14 days; 8 weeks; 4 months; 12 months; and 2 years.[52]

2. School Entry Health Assessment

The School Entry Health Assessment is a free child health service provided by the Child and Adolescent Health Service and the Western Australia Country Health Service to all school age children.[53,54] This community child health service includes the assessments of vision and hearing; growth (including height, weight and Body Mass Index); and oral health. The nurses conduct these assessments on the children in school at the recommended time points: 4-5 years.[54]

3. National Immunisation Program

The National Immunisation Program is a free health program to provide Australians a series of immunisations throughout specific times of their life in order to protect them against harmful diseases.[55] Children are recommended to follow a childhood immunisation schedule at designated immunisation providers.[56] The recommended time points for immunisations are: birth to less than 7 days (all children); 6 weeks (all children); 4 months (Aboriginal children only); 6 months (Aboriginal children or children with a medical risk condition); 18 months (Aboriginal children only); 4 years (all children).[56] In addition, adolescents are recommended to follow a school-based immunisation program in school.[57] The recommended time points for immunisations are: 12-13 years and 14-16 years.[55]

8.2 Recommendations regarding the integration of targeted FASD screening into existing models of care

The three community child health services provide approximately 11-14 points of contact between children aged 0 to 16 years and health professionals (see Table 10). The age range in which the children have contact with health professionals, mainly community child health nurses, is ideal for the integration of targeted FASD screening.[30] Notably, FASD screening can be implemented at birth for newborns presented with microcephaly or sentinel facial features of FASD.[3,58] However, FASD screening is often recommended to be conducted in early childhood as central nervous system dysfunction, such as executive function or adaptive behavioural problems, is often not apparent in very young children.[30,59] Hence, the Purple book

appointments at 12 months and 2 years as well as the School Entry Health Assessment at 4-5 years are recommended time points to integrate FASD screening.

A component of the Purple book appointments at 12 months and 2 years involves a neurodevelopmental screening via the Ages and Stages Questionnaires.[60] The Ages and Stages Questionnaire, Third Edition (ASQ-3™),[61] the Ages and Stages Questionnaire: Social-Emotional, Second Edition (ASQ:SE-2™), and the ASQ-TRAK (for use with Aboriginal clients), are caregiver-reported assessments, containing components of language, motor, cognitive, executive functioning, self-care, affect regulation, attention, social skills, and socio-communication skills, to screen and monitor for developmental delays in children. In addition, a component of the School Entry Health Assessment includes a neurodevelopmental screening via the Parents' Evaluation of Developmental Status.[54] The Parents' Evaluation of Developmental Status[62] is a caregiver-reported assessment, containing 10 questions about the child's language, motor, adaptive behaviour, social, socio-communication, self-care and cognitive skills.[54]

Information from the Ages and Stages Questionnaires and the Parents' Evaluation of Developmental Status provides a useful guide for health professionals to refer children with poor outcomes to Child Developmental Services or private practitioners for further evaluation, including, if indicated a comprehensive diagnostic assessment. An important aspect of this recommendation is to ensure that PAE information is considered and communicated when a child is referred for further evaluation. The recommendation regarding the referral pathways for the neurodevelopmental assessments is adapted from the *Ages and Stages Questionnaires and referral pathway table*[60] created by the Department of Health, Western Australia and is presented in Figure 3.

Another important aspect of this recommendation is to determine best practice for supporting parents to take up the indicated community child health services to aid early identification of concerns in their child's development. There is currently a low up take rate of child health services in Australia. According to recent data from the

Australian Institute of Family Studies on the use of health care services in Australia, only 66% of 0-1 year olds received consultation with a maternal and child health nurse. Furthermore, child health service participation rates decrease as children approach school age, from 32% of those 2-3 years old to 12% of those 4-5 years old.[63] Therefore, there is a need to identify and overcome barriers to the use of child health services in Australia.

Table 10. Summary of the existing community child health services in Western Australia and the recommended time points to participate in these services.

Recommended age for children to participate in child health services	**Community child health services**		
	Purple book appointment	School Entry Health Assessment	National Immunisation Program
0-14 days	✓		✓
6 weeks			✓
8 weeks	✓		
4 months	✓		✓*
6 months			✓*
12 months	✓		
18 months			✓*
2 years	✓		
4-5 years		✓	✓
12-13 years			✓
14-16 years			✓

✓ applicable; * additional immunisations for Aboriginal children.

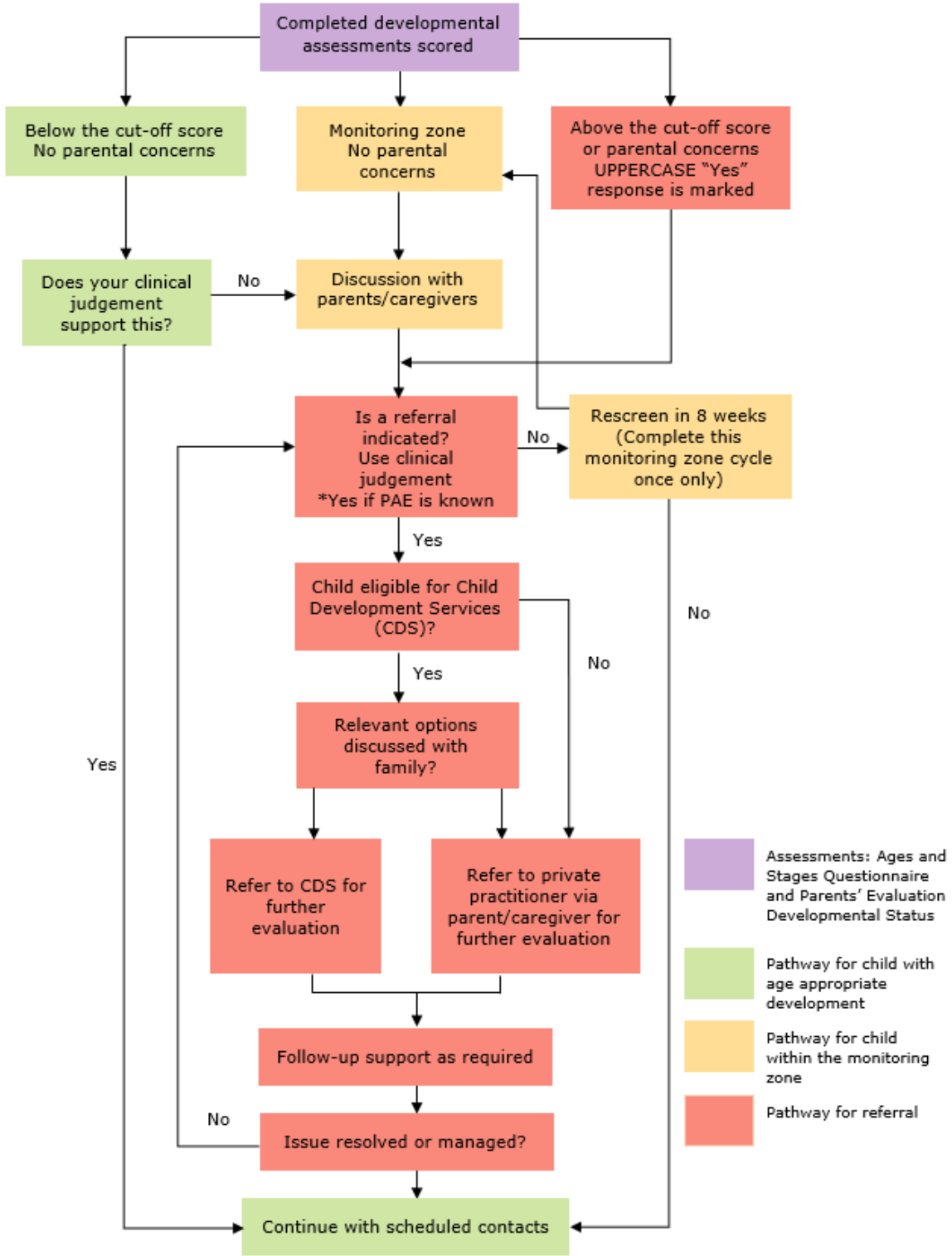

Figure 3. Referral pathway for targeted FASD screening approach at routine community child health neurodevelopmental screening at 12 months, 2 years, and 4-5 years. Note: *PAE, prenatal alcohol exposure.*

8.3 Existing health care services in systems and organisations supporting high risk populations

There are several health care services in systems and organisations supporting selected populations with a higher prevalence of FASD; for example, children referred to child development services, children with mothers who receive alcohol treatment services, children in state/foster care, and young people in justice settings.[30] An overview of five systems and organisations in Western Australia supporting high risk populations is described below.

1. Child Development Service

The Child Development Service is a free child health service provided by the Child and Adolescent Health Service and the Western Australia Country Health Service for children with developmental delay or difficulty that impacts on function, participation, and/or parent-child relationship.[64] This service includes a range of assessment, early intervention, and treatment services via a multidisciplinary team of doctors, nurses, social workers, speech pathologists, occupational therapists, physiotherapist, audiologists, and clinical psychologists.[64]

2. Child and Adolescent Mental Health Services

The Child and Adolescent Mental Health Services is a free child health service provided by the Child and Adolescent Health Service to children and adolescents (0-18 years) with severe, complex and persistent emotional, psychological, behavioural, social, and/or mental health problems.[65] This service include a range of assessment, case coordination and multidisciplinary treatment services for children led by psychiatrists, clinical psychologists, social workers, family therapists, community mental health nurses, occupational therapists, speech pathologists, and child care leaders.[65]

3. Women and Newborn Drug and Alcohol Service

The Women and Newborn Drug and Alcohol Service provides free postnatal services to women with drug and alcohol dependence and their babies up to 3 months old.[66]

These services include a multidisciplinary team of doctors, nurses, social workers, dietitians, mental health professionals and parent educators.[66]

4. Foster/state care

The Department of Communities Child Protection and Family Services refers children (0-18 years) who are new to care to a general practitioner or paediatrician for an initial medical assessment, and subsequently to the Child and Adolescent Health Service – Community Health or Western Australia Country Health Services for neurodevelopmental screening, if necessary.[67] The initial medical assessment has to be completed within 30 business days of admission into care.[67] In addition, children undergo a comprehensive health assessment and health care planning as well as a 12 month health care plan review led by general practitioner or paediatrician to identify and address any health and development concerns.[67]

5. Youth Justice System

The Department of Corrective Services provides health screening checks to youth offenders (10-18 years), which are conducted by a multidisciplinary team of doctors, nurses, mental health and addiction specialist, and visiting health specialists including psychiatrists, dentist and allied health specialists.[16,68] The health screening check has to be completed within 28 days of admission into the youth justice system.[68]

8.4 Recommendations regarding the integration of selective FASD screening into existing models of care

The five systems and organisations supporting populations at high risk of FASD provides health assessments that are conducted by a multidisciplinary team of health professionals. The age range in which the populations at high risk of FASD have contact with health professionals is between 0-18 years. These children and adolescents ideally receive health assessments within approximately one month upon entry into the systems and organisation. Therefore, an integrated neurodevelopmental and PAE screenings is recommended during these health

assessments. Following the general health screenings, clinical judgement should be considered for the referral of the child for further evaluation, including, if indicated, a comprehensive diagnostic assessment. If the child/adolescent is screened positive, timely and appropriate follow-up to diagnosis assessment and management care and support should be provided during their care under the systems/organisations.

8.5 Recommendations on obtaining information about PAE

A crucial aspect of FASD screening and diagnosis is to determine whether individuals have been exposed to alcohol prenatally. The *Fetal Alcohol Spectrum Disorder Model of Care 2010*[27] recommended routine screening of women of child-bearing age and during pregnancy for alcohol consumption to prevent FASD and to provide early intervention for pregnant women with alcohol problems and children diagnosed with FASD. Information about PAE can be obtained through health professional's interaction with pregnant women or though self-reported questionnaires,[69] such as CAGE,[70] Tolerance, Annoyance, Cut down attempts, Eye opener (T-ACE),[71] Tolerance, Worried, Eye-opener, Amnesia, K/Cut down attempts (TWEAK),[72] Michigan Alcoholism Screening Test (MAST),[73] and Alcohol Use Disorders Identification Test – Consumption (AUDIT-C)[74]. The AUDIT-C is the recommended PAE screening tool to use in the Australian Guide to the diagnosis of FASD.[3] Despite the availability of these screening tools, the Advisory Group has indicated a lack of coordinated information sharing regarding PAE between relevant health care services and organisations supporting children at risk of FASD.

The National Institute for Health and Care Excellence recently published an extensive list of recommendations[47] that were produced via a consultation with stakeholders to develop quality standards on the assessment and diagnosis of FASD in the United Kingdom. One of the recommendations that was proposed to resolve the problem of uncoordinated information sharing between relevant health care services was to transfer PAE information onto personalised digital child health records. The potential benefit of this recommendation would be improved information sharing on maternal alcohol history between primary care and

secondary care providers. As access to accurate antenatal health records is essential for screening and diagnosis of FASD, this would then facilitate timely management and support services for individuals with FASD and their families. The potential harms of this recommendation would be the loss of medical confidentiality, and stigma and anxiety in biological parents. However, it is important to weigh the potential benefits and harms of any recommendation; in this case, the potential benefits of the having a timely diagnosis and management support for individuals with FASD and their families may outweigh the potential harms of medical confidentially loss, stigma, and anxiety.

At present, personalised child health records in Australia are detailed in the Purple book that is parent-held. The transfer of maternal health at pregnancy and alcohol exposed pregnancy information onto the Purple book would provide health professionals easy access to relevant information to guide their clinical judgement on the need to refer the child for further evaluation without compromising medical confidentially. Therefore, it is recommended that all pregnant women be screened for alcohol use during pregnancy and for primary care providers to transfer PAE information into the child's individualised Purple book to aid accurate FASD screening and diagnosis processes.

8.6 Guiding principles for FASD Model of Care

The following principles were taken from the *Fetal Alcohol Spectrum Disorder Framework to guide and support* the *Western Australian FASD Implementation Plan for the Model of Care 2013-2018*[75] to guide the way in which government departments and community agencies develop and apply strategies for the prevention of and to reduce the incidence of FASD (Table 11).

Table 11. Guiding principles for FASD Model of Care.

Principles	Descriptions
FASD is preventable	The priority is primary and secondary prevention.
Across sector responsibility and accountability	Due to the multifaceted nature of FASD, a holistic approach is required, across the continuum of care and inclusive of sectors beyond the health service delivery sectors.
Coordinated interventions	Stakeholders will collaborate and work in partnership to maximise the best use of resources and opportunities for access to services.
Accessibility and equitability	Screening approaches will be responsive to the needs of people from all cultural and linguistic backgrounds, in particular Aboriginal populations, and all socio-economic and educational backgrounds residing in communities across Western Australia.
Public health principles	Based on the World Health Organization's Ottawa Charter for public health, health is the result of interactions between people's personal behaviours, social, economic, political, and physical environmental factors.
Consumer and carer focussed	Screening approaches will be consumer and carer focussed. Consumers and carers will be consulted and informed about prevention, screening, diagnosis, and management of FASD and the relevance to them, their families, and communities.
Responsive to emerging evidence-based policy and practice	Through the governance framework and reporting structure, WA strategies will be included in the national planning and policy development.
Cultural diversity	When working with Aboriginal people and communities, ensure an approach that respects the rights, values, and beliefs of Aboriginal people. Aboriginal leadership, community consultation, direction and involvement form an essential part of this process as does working in partnership with Aboriginal communities.

8.7 Inclusion criteria for the development of strategies within FASD Model of Care

The inclusion criteria were taken from the *Fetal Alcohol Spectrum Disorder Framework to guide and support* the *Western Australian FASD Implementation Plan for the Model of Care 2013-2018*[75] and comprise of the following criteria (Table 12).

Table 12. Inclusion criteria for the development of strategies within FASD Model of Care.

Criteria	Description
Based on evidence and best practice model	Where available, the strategies are based on evidence and best practice, adaptable to the current context.
Align with public health principles	The strategies aim to develop personal skills, create supportive environments, strengthen community action, reorientate health services and build responsive public policy.
Cause no more harm	The strategies aim to minimise harm and consideration has been given to potential unintended consequences.
Measurable and have been and/or can be evaluated	Measurable outcomes have been aligned with the strategies to begin developing an evidence base for the future.
Accessible for at-risk groups	The strategies aim to provide the best outcomes for the most people, are culturally appropriate and take other access barriers, such as cost, and/or geographical location into consideration.
Sustainable	The strategies do not rely on the provision of new resources.

9 Discussion

The current report provides an overview of FASD screening tools available internationally, evidence-based recommendations for the implementation of available FASD screening tools in Western Australia and recommendations on best practice principles for FASD screening approach and models of care in Western Australia. It should be noted that all recommendations relate to the use of these tools to screen for FASD (with and without sentinel facial features). The broad summary of the findings is presented in Table 13.

Table 13. Summary of the main findings of the report.

What FASD screening tools are available internationally?
The systematic review included 14 articles, examining eight different FASD screening tools: ▪ The Neurobehavioral Screening Tool ▪ Eye movement behaviour assessment via machine learning ▪ Tally Reference Errors in Narrative Task ▪ Dysmorphic examination via photographs ▪ Physical and dysmorphic examination ▪ Craniofacial measurements approach ▪ The FAS Screen ▪ The FAS diagnostic checklist The quality of evidence for the eight screening tools was variable, ranging from moderate to very low.

Which FASD screening tools are recommended for use in Western Australia?

These recommendations apply only in the context of *universal* FASD screening in Australia. The recommendations were assessed as 'strong recommendation' or 'conditional recommendation' (depending on screening context) according to the GRADE approach.[1]

Strong recommendations:

- against the use of the Tallying Reference Errors in Narrative task to screen for individuals at risk of FASD.
- against the use of the dysmorphic examination via photographs to screen for individuals at risk of FASD.
- against the use of the craniofacial measurement approach to screen for individuals at risk of FASD.
- against the use of the FAS Screen to screen for individuals at risk of FASD.
- against the use of the FAS diagnostic checklist to screen for individuals at risk of FASD.

Conditional recommendations:

- against the use of the Neurobehavioral Screening Test to screen for individuals at risk of FASD.
- against the use of the eye movement behaviour assessment via machine learning to screen for individuals at risk of FASD.
- against the use of the physical and dysmorphic examination to screen for individuals at risk of FASD.

What is the recommended approach for FASD screening in Western Australia?

Even though no FASD screening tools are recommended for use in Western Australia, the following recommended approach to FASD screening has been formulated with the view that appropriate screening tools are required and will

be developed in the future. All recommendations relate to the screening approach for FASD (with and without sentinel facial features).

Recommendations regarding FASD screening approaches

Three types of screening approaches have been identified for use in Western Australia, that together will support the likelihood that individuals with FASD will be identified in a timely fashion.

- Universal screening:
  - Implement prenatal alcohol exposure (PAE) screening approaches in the entire population, regardless of risk.
- Targeted screening:
  - Implement neurodevelopmental screening with individuals who have PAE, show signs of a neurodevelopmental disorder, such as developmental delay, have a sibling who has been diagnosed with FASD, have facial anomalies associated with FASD, have observed problems with behaviour, or where there is parental concern that a child may have FASD. Refer these individuals to Child Developmental Services or private practitioners for further evaluation, and, if necessary, a comprehensive diagnostic assessment.
- Selective screening:
  - Implement neurodevelopmental and PAE screening in selected populations known to report a higher prevalence of FASD. For example, children referred to Child Development Services or Child and Adolescent Mental Health Services; children with mothers who access alcohol treatment services; children in state/foster care; and youth in justice settings. Refer these individuals to Child Developmental Services or private practitioners for further evaluation and, if necessary, a comprehensive diagnostic assessment.

Recommendations regarding FASD Model of Care

A review of the existing FASD Model of Care 2013-2018 is warranted. The development of additional strategies to address the following areas of concern are recommended:

- Improve coordinated information sharing regarding PAE between relevant health care services and organisations supporting children at risk of FASD through the Purple book, a personalised child health record.
- Support parents with children aged between ages 12 months and 2 years to take up community child health services (for example, the Purple book appointments) to aid early identification of concerns in their child's development.
- Support parents with children aged between 4 and 5 years to take up community child health services (for example, the School Entry Health Assessment) to aid early identification of concerns in their child's development.
- Support relevant systems and organisations in the integration of neurodevelopmental and PAE screenings in general health assessments conducted with populations at high risk of FASD (for example, children referred to Child Development Services, children with mothers who receive alcohol treatment services, children in state/foster care, and young people in justice settings).
- Review existing child health referral pathways to improve coordinated referral of children with poor outcomes to Child Developmental Services or private practitioners for further evaluation and, if necessary, a comprehensive diagnostic assessment.

None of the existing FASD screening tools available in the literature are recommended for use in Western Australia. Each of the existing tools have various limitations that affected its acceptability and feasibility in the context of Western Australia's population and geography. Western Australia has people who are culturally and linguistically diverse as well as has areas that range from urban to

rural and remote. The design of all the screening tools displayed a lack of consideration for the cultural and linguistic context of their screening populations, especially in cross-cultural communities. Several of the screening tools were designed to assess only one associated feature of FASD or the sentinel facial features associated with FASD, which are not always presented in individuals at risk of FASD. Therefore, further development and trial of a FASD screening tool that fits Western Australia context is warranted.

Another shortcoming of the current evidence base of FASD screening tools is a lack of consideration in areas where health systems are not robust, resources are limited, or quality assurance is not maintained. Quality assurance to ensure accurate and reproducible results during FASD screening requires intensive human and financial resources; most of the studies did not report the human and financial resources required of the screening processes. Given that economic evaluation of a screening program is one of the identified guiding principles for FASD screening approach (see section 7.2, Recommended guiding principles for FASD screening approaches on page 42-43), future evaluation of the full costs and effects of implementing, operating, and sustaining the screening process is required.

Besides the need for an appropriate FASD screening tool, there is an equal importance to translate policy into practice. At present, few of the FASD policies have been implemented in practice. For example, there is still a lack of coordinated information sharing between relevant systems/organisations and management care and support for both the individuals with FASD and their families despite existing FASD policies that have been produced to improve support and services for those affected by FASD.[25,27,45,75] An attitude change within the child health services and systems and organisations supporting high risk populations has been argued to be fundamental in translating policies into practice.[76] One effective way of helping to change attitudes and encourage new practices would be to implement training that includes both health professionals and consumers who are involved in delivering the training.[76] Another way would be to empower individuals with FASD and their families to act as agents of change within their communities.[76] Strategies to

improve attitude change in systems and organisations supporting those affected by FASD are warranted.

Furthermore, there is a need to review the FASD Model of Care. The existing Western Australian FASD Model of Care 2013-2018 has provided guidelines for the delivery of best practice to the intended population in Western Australia.[27,45,75] However, with the advancement in research relating to FASD prevention, screening, diagnosis, and management, there also needs to be a continuous review of the FASD Model of Care in order to understand and respond to the health needs of the population, provide equitable and integrated care, and ensure evaluation of health services and quality improvements in an effective and timely manner.[34,35] Future research regarding FASD prevention, screening, diagnosis, and management in Western Australia is recommended to be conducted in tandem with a review of the FASD Model of Care.

10 References

1. GRADE Working Group. *GRADE Handbook.* Canada: McMaster University; 2013. ISBN.
2. Chudley AE, Conry J, Cook JL, et al. Fetal alcohol spectrum disorder: Canadian guidelines for diagnosis. *CMAJ.* 2005;172(5 Suppl):S1-S21.
3. Bower C, Elliott EJ, and 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)".* Australian Government Department of Health; 2016. ISBN: 978-0-6481297-4-5.
4. Cook JL, Green CR, Lilley CM, et al. Fetal alcohol spectrum disorder: a guideline for diagnosis across the lifespan. *CMAJ.* 2016;188(3):191-197.
5. Hoyme HE, Kalberg WO, Elliott AJ, et al. Updated clinical guidelines for diagnosing fetal alcohol spectrum disorders. *Pediatrics.* 2016;138(2).
6. Lange S, Probst C, Gmel G, Rehm J, Burd L, Popova S. Global prevalence of fetal alcohol spectrum disorder among children and youth: a systematic review and meta-analysis. *JAMA Pediatr.* 2017;171(10):948-956.
7. Popova S, Lange S, Probst C, Gmel G, Rehm J. Global prevalence of alcohol use and binge drinking during pregnancy, and fetal alcohol spectrum disorder. *Biochem Cell Biol.* 2017;96(2):237-240.
8. Popova S, Lange S, Shield K, Burd L, Rehm J. Prevalence of fetal alcohol spectrum disorder among special subpopulations: a systematic review and meta-analysis. *Addiction.* 2019;114(7):1150-1172.
9. Riley EP, Infante MA, Warren KR. Fetal alcohol spectrum disorders: an overview. *Neuropsychol Rev.* 2011;21(2):73.
10. Millar JA, Thompson J, Schwab D, et al. Educating students with FASD: linking policy, research and practice. *J Res Spec Educ Needs.* 2017;17(1):3-17.
11. Millians MN. Educational needs and care of children with FASD. *Curr Dev Disord Rep.* 2015;2(3):210-218.
12. Pei J, Denys K, Hughes J, Rasmussen C. Mental health issues in fetal alcohol spectrum disorder. *J Ment Health.* 2011;20(5):473-483.
13. Rangmar J, Hjern A, Vinnerljung B, Stromland K, Aronson M, Fahlke C. Psychosocial outcomes of fetal alcohol syndrome in adulthood. *Pediatrics.* 2014;135(1):e52-e58.
14. Duquette C, Orders S. On fitting a triangle into a circle: a study on employment outcomes of adults with fetal alcohol spectrum disorder who attended postsecondary institutions. *Intl J Alcohol Drug Res.* 2013;2(3):27.
15. Hughes N, Clasby B, Chitsabesan P, Williams H. A systematic review of the prevalence of foetal alcohol syndrome disorders among young people in the criminal justice system. *Cogent Psychology.* 2016;3(1):1214213.
16. Bower C, Watkins RE, Mutch RC, et al. Fetal alcohol spectrum disorder and youth justice: a prevalence study among young people sentenced to detention in Western Australia. *BMJ Open.* 2018;8(2):e019605.
17. Thanh NX, Jonsson E. Life expectancy of people with fetal alcohol syndrome. *J Popul Ther Clin Pharmacol.* 2016;23(1):e53-59.

18. Bobbitt SA, Baugh LA, Andrew GH, et al. Caregiver needs and stress in caring for individuals with fetal alcohol spectrum disorder. *Res Dev Disabil.* 2016;55:100-113.
19. Ericson L, Magnusson L, Hovstadius B. Societal costs of fetal alcohol syndrome in Sweden. *Eur J Health Econ.* 2017;18(5):575-585.
20. Popova S, Stade B, Bekmuradov D, Lange S, Rehm J. What do we know about the economic impact of fetal alcohol spectrum disorder? A systematic literature review. *Alcohol Alcohol.* 2011;46(4):490-497.
21. Mutch RC, Watkins R, Bower C. Fetal alcohol spectrum disorders: Notifications to the Western Australian Register of Developmental Anomalies. *J Paediatr Child Health.* 2015;51(4):433-436.
22. Fitzpatrick JP, Latimer J, Carter M, 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(4):450-457.
23. Department of Health. $7 million for Fetal Alcohol Spectrum Disorder. 2019; https://www.health.gov.au/ministers/the-hon-greg-hunt-mp/media/7-million-for-fetal-alcohol-spectrum-disorder. Accessed July 30, 2020.
24. Australian Government Department of Health. *Health Portfolio Budget Statements 2016–17.* Canberra ACT: Australian Government, Department of Health;2016.
25. Australian Government Department of Health. *National Fetal Alcohol Spectrum Disorder Strategic Action Plan 2018-2028.* Canberra ACT: Australian Government, Department of Health;2018.
26. Sustainable Health Review. *Sustainable Health Review: Final Report to the Western Australian Government.* Department of Health, Western Australia;2019.
27. Department of Health Western Australia. *Fetal Alcohol Spectrum Disorder Model of Care.* Perth: Health Network Branch, Department of Health, Western Australia;2010.
28. Richards D. Screening. *Evid Based Dent.* 2007;8(1):2-3.
29. Dawe S, Dingle G, Loxton NJ. Chapter 31 - Screening and Assessment of Comorbidity. In: Miller PM, ed. *Interventions for Addiction.* San Diego: Academic Press; 2013:299-307.
30. Watkins RE, Elliott EJ, Halliday J, et al. A modified Delphi study of screening for fetal alcohol spectrum disorders in Australia. *BMC Pediatr.* 2013;13:13-13.
31. Astley SJ, Clarren SK. A fetal alcohol syndrome screening tool. *Alcohol Clin Exp Res.* 1995;19(6):1565-1571.
32. Astley SJ, Stachowiak J, Clarren SK, Clausen C. Application of the fetal alcohol syndrome facial photographic screening tool in a foster care population. *J Pediatr.* 2002;141(5):712-717.
33. Mattson SN, Roesch SC, Fagerlund A, et al. Toward a neurobehavioral profile of fetal alcohol spectrum disorders. *Alcohol Clin Exp Res.* 2010;34(9):1640-1650.
34. Wakerman J, Humphreys JS, Wells R, Kuipers P, Entwistle P, Jones J. Primary health care delivery models in rural and remote Australia – a systematic review. *BMC Health Serv Res.* 2008;8(1):276-276.

35. Legislative Assembly Parliament of Western Australia. *Foetal Alcohol Spectrum Disorder: the Invisible Disability.* Perth: Legislative Assembly Parliament of Western Australia;2012.
36. Moore ES, Ward RE, Jamison PL, Morris CA, Bader PI, Hall BD. The subtle facial signs of prenatal exposure to alcohol: an anthropometric approach. *J Pediatr.* 2001;139(2):215-219.
37. Astley SJ, Clarren SK. A case definition and photographic screening tool for the facial phenotype of fetal alcohol syndrome. *J Pediatr.* 1996;129(1):33-41.
38. Helgesson G, Bertilsson G, Domeij H, et al. Ethical aspects of diagnosis and interventions for children with fetal alcohol Spectrum disorder (FASD) and their families. *BMC Med Ethics.* 2018;19(1):1-1.
39. Australian Institute of Health and Welfare. Prevention for a healthier future. *Australia's Health.* 2014(14). https://www.aihw.gov.au/getmedia/6c8ffb4a-a0f6-49f8-9b05-01f2157b822c/8_1-health-prevention.pdf.aspx. Accessed 18 November 2020.
40. O'Connor MJ, Rotheram-Borus MJ, Tomlinson M, Bill C, LeRoux IM, Stewart J. Screening for fetal alcohol spectrum disorders by nonmedical community workers. *J Popul Ther Clin Pharmacol.* 2014;21(3):e442-452.
41. Popova S, Lange S, Probst C, Parunashvili N, Rehm J. Prevalence of alcohol consumption during pregnancy and fetal alcohol spectrum disorders among the general and Aboriginal populations in Canada and the United States. *Eur J Med Genet.* 2017;60(1):32-48.
42. Burd L, Cohen C, Shah R, Norris J. A court team model for young children in foster care: The role of prenatal alcohol exposure and fetal alcohol spectrum disorders. *J Psychiatry Law.* 2011;39(1):179-191.
43. Hanlon-Dearman A, Green CR, Andrew G, LeBlanc N, Cook JL. Anticipatory guidance for children and adolescents with fetal alcohol spectrum disorder (FASD): practice points for primary health care providers. *J Popul Ther Clin Pharmacol.* 2015;22(1):e27-56.
44. Australian Institute of Health and Welfare. Australia's Children. 2019; https://www.aihw.gov.au/reports/children-youth/australias-children/contents/health/smoking-and-drinking-in-pregnancy. Accessed 5th October, 2020.
45. Department of Health Western Australia. *Western Australia Across Sector, Statewide Implementation Plan for the Fetal Alcohol Spectrum Disorder Model of Care 2013-2018.* Perth: Health Networks Branch, Department of Health, Western Australia;2013.
46. Finlay-Jones A. *Deeble Issues Brief No. 28: Reducing harms related to alcohol use in pregnancy – policy and practice recommendations.* Australia: Deeble Institute for Health Policy Research;2015.
47. National Institute for Health and Care Excellence. Fetal Alcohol Spectrum Disorder. In. *Health and social care directorate, quality standards, briefing paper*. United Kingdom: National Institute for Health and Care Excellence; 2019.
48. Pedruzzi RA, Hamilton O, Hodgson HHA, Connor E, Johnson E, Fitzpatrick J. 'We do what we can as soon as we can' alcohol and other drug workforce

perspectives on preventing and responding to prenatal alcohol exposure. *Drugs: Education, Prevention and Policy.* 2020:1-9.
49. Finlay-Jones A, Elliott EJ, Mayers D, et al. Community priority setting for fetal alcohol spectrum disorder research in Australia. *International Journal of Population Data Science.* 2020;5(3).
50. Gupta H, Tari-Keresztes N, Stephens D, Smith JA, Sultan E, Lloyd S. A scoping review about social and emotional wellbeing programs and services targeting Aboriginal and Torres Strait Islander young people in Australia: understanding the principles guiding promising practice. *BMC Public Health.* 2020;20(1):1625.
51. Dobrow MJ, Hagens V, Chafe R, Sullivan T, Rabeneck L. Consolidated principles for screening based on a systematic review and consensus process. *CMAJ.* 2018;190(14):E422-E429.
52. Child and Adolescent Health Service Western Australia. Child Health. 2020; https://www.cahs.health.wa.gov.au/Our-services/Community-Health/Child-Health. Accessed 20 November, 2020.
53. WA Country Health Service. Child Health. 2011; http://www.wacountry.health.wa.gov.au/index.php?id=childhealth. Accessed 20 November, 2020.
54. Child and Adolescent Health Service Western Australia. *Universal Contact School Entry Health Assessment.* Western Australia: Department of Health, Western Australia;2020.
55. Department of Health Western Australia. National Immunisation Program Schedule. 2020; https://www.health.gov.au/health-topics/immunisation/immunisation-throughout-life/national-immunisation-program-schedule. Accessed 20 November, 2020.
56. Department of Health Western Australia. Childhood Immunisation Schedule. n.a.; https://healthywa.wa.gov.au/Articles/A_E/Childhood-immunisation-schedule. Accessed 20 November, 2020.
57. Child and Adolescent Health Service Western Australia. School Health. 2020; https://www.cahs.health.wa.gov.au/Our-services/Community-Health/School-Health. Accessed 20 November, 2020.
58. Jones K, Smith D. Recognition of the fetal alcohol syndrome in early infancy. *The Lancet.* 1973;302(7836):999-1001.
59. CDC. *Fetal Alcohol Syndrome: Guidelines for Rreferral and Diagnosis* USA: National Center on Birth Defects and Developmental Disabilities, Centers for Disease Control and Prevention, Department of Health and Human Services 2004.
60. Child and Adolescent Health Service Western Australia. *Ages and Stages Questionnaires Guideline.* Western Australia: Department of Health Western Australia;2020.
61. Squires J, Brickers D, Twombly E. *Ages and Stages Questionnaires User's Guide.* Baltimore, MD: Paul H. Brookes Publishing Co., Inc.; 2009. ISBN.
62. PEDStest.com. Parents' Evaluation of Developmental Status 2018; https://pedstest.com/index.html. Accessed 23 November, 2020.
63. Warren D. Children's use of health care services. 2018; https://growingupinaustralia.gov.au/research-findings/annual-statistical-

report-2017/childrens-use-health-care-services. Accessed 18 December, 2020.
64. Child and Adolescent Health Service Western Australia. Child Development Service. 2020; https://www.cahs.health.wa.gov.au/Our-services/Community-Health/Child-Development-Service. Accessed 18 December, 2020.
65. Child and Adolescent Health Service Western Australia. Community CAMHS clinics. 2020; https://cahs.health.wa.gov.au/Our-services/Mental-Health/Community-CAMHS-clinics. Accessed 18 December, 2020.
66. Department of Health. Women and Newborn Drug and Alcohol Service (WANDAS). n.a.; https://healthywa.wa.gov.au/Articles/U_Z/Women-and-Newborn-Drug-and-Alcohol-Service-WANDAS. Accessed 23 November, 2020.
67. Child and Adolescent Health Service Western Australia. Children in Care - Conducting an Assessment. In. Western Australia: Department of Health, Western Australia; 2019.
68. Department of Corrective Services. *Assessment of Clinical Service Provision of Health Services of the Western Australian Department of Corrective Services.* Western Australia: Department of Corrective Services, Western Australia;2010.
69. DeVido J, Bogunovic O, Weiss RD. Alcohol use disorders in pregnancy. *Harv Rev Psychiatry.* 2015;23(2):112-121.
70. Ewing JA. Detecting alcoholism: the CAGE questionnaire. *JAMA.* 1984;252(14):1905-1907.
71. Chang G, Wilkins-Haug L, Berman S, Goetz MA, Behr H, Hiley A. Alcohol use and pregnancy: improving identification. *Obstet Gynecol.* 1998;91(6):892-898.
72. Russell M, Martier SS, Sokol RJ, et al. Screening for pregnancy risk-drinking. *Alcohol Clin Exp Res.* 1994;18(5):1156-1161.
73. Selzer ML. The Michigan alcoholism screening test: the quest for a new diagnostic instrument. *Am J Psychiatry.* 1971;127(12):1653-1658.
74. Bush K, Kivlahan DR, McDonell MB, Fihn SD, Bradley KA. The AUDIT alcohol consumption questions (AUDIT-C): an effective brief screening test for problem drinking. *Arch Intern Med.* 1998;158(16):1789-1795.
75. Department of Health Western Australia. *Fetal Alcohol Spectrum Disorder Framework to Guide and Support the Western Australian FASD Implementation Plan for the Model of Care 2013-2018.* Western Australia: Perth: Health Networks Branch, Department of Health, Western Australia;2013.
76. Department of Health Commonwealth Australia. Attitudes, Education and Training. 2006; https://www1.health.gov.au/internet/publications/publishing.nsf/Content/mental-pubs-p-natcon-toc~mental-pubs-p-natcon-sum~mental-pubs-p-natcon-sum-imp~mental-pubs-p-natcon-sum-imp-att. Accessed 23 November, 2020.

Appendix A:
Terms of Reference for the
FASD Screening Advisory Group

Background

For effective implementation of FASD screening Western Australia, it is necessary to understand best practice nationally and internationally. This includes synthesising evidence on the psychometrics of available tools, reviewing clinical and implementation issues, and considering community and ethical perspectives on screening. To achieve this, there is a need to review and critically appraise current evidence and discuss the feasibility and costs of integrating the evidence into policy and practice, as well as the need for workforce training and ongoing evaluation.

The *Screening for FASD in Western Australia: Policy and Practice Recommendations* will undertake the first two stages and provide recommendations regarding stages three and four.

1. Project scoping
2. Best practice recommendations for implementation of screening tool/s in WA
3. Feasibility
4. Implementation

The aims of this recommendation are to:

- provide evidence-based recommendations regarding screening for FASD in Western Australia
- examine current FASD screening recommendations internationally and nationally and consider which time-points during a child's development a screening tool could be used; for example, which settings would be most appropriate and how screening may be implemented
- consider the economic and social impact of screening and referral for early assessment and diagnosis for FASD and early intervention

Funder

This project is funded by the Department of Health Western Australia

Purpose of the FASD Screening Advisory Group

The FASD Screening Advisory Group is a time-limited group that brings together key stakeholder representatives with expertise and experience in their related fields to provide strategic advice on the implementation of the project.

Roles and responsibilities

Advisory Group members will:

- understand the aim and intended outcomes of the recommendation
- have a genuine interest in the recommendation development and the outcomes that are intended
- act as a sounding board for the Working Group to discuss and advance any complex elements of the proposed recommendations and implementation that are relevant to their area of expertise or experience
- provide high level advice and expertise on aspects of the recommendation relevant to their experience
- review & comment on proposed recommendations
- actively participate in meetings through attendance, discussion and review of meeting notes, papers, and other materials

Guiding principles

- Contributions from the members will be valued and considered without fear or favour
- All members will have an equal voice and should feel free to express their views at meetings
- Where possible decisions will be reached by consensus and the chair will conclude a final decision when a consensus cannot be reached
- Meetings are strictly confidential
- Members are expected to respond to communications within the agreed requested timeframe
- Members are expected to declare any conflicts of interest

Chair will:

- clarify the purpose of the Advisory Group
- provide guidance on expectations and positive reinforcement to the Advisory Group ensure the Advisory Group fulfils its functions
- ensure meeting agendas and papers are appropriate for the Advisory Group
- facilitate effective meetings
- ensure the Advisory Group is focused on matters relevant to their function and consider each matter with appropriate care and propriety
- ensure the Advisory Group arrives at clear decisions
- ensure decisions are implemented appropriately and outstanding actions are monitored
- ensure all members have the opportunity to participate

Membership

The Advisory Group will comprise a group of individuals with significant experience and expertise who can contribute to and provide leadership, and strategic direction on a range of activities. They reflect the diverse interests and concerns of a range of stakeholders. Overall, the Advisory Group will consist representatives from the following stakeholder organisations or sectors:

- Department of Health Western Australia – Child Health Nurse (1)
- Department of Education/Health Western Australia – School Nurse or School Psychologist (1)
- Western Australia Country Health Service (1)
- Western Australia Primary Health Alliance (1)
- Paediatrician with experience in assessment & diagnosis (1)
- Representative from the group currently reviewing the Australian Guide to the diagnosis of FASD (1)
- Community representatives – preference for a parent who has been involved in screening or assessment process, and must include one Aboriginal and Torres Strait Islander person (1)

- Representatives from the Department of Health Western Australia project coordination team (2)

Meeting frequency & duration

Three meetings will be held between July and October 2020. Each meeting will last about 60 minutes. Meetings will take place by teleconference or Zoom (or similar apps).

Payment

Group membership is voluntary and there is no fee paid to members. An honorarium will be offered to community representative members.

Appendix B:
Evidence-to-recommendation template

Evidence-to-recommendations template adapted from the GRADEpro Guideline Development Tool[1] evidence-to-recommendations framework

PICO Questions				
Problem: **Background:** **Intervention:** **Comparison:** **Setting:**		**Background:**		
	Criteria	**Judgement**	**Research evidence**	**Additional consideration**
Problem	Is the problem a priority?	☐ No ☐ Probably no ☐ Uncertain ☐ Probably yes ☐ Yes ☐ Varies		
Test accuracy	How accurate is the test?	☐ Very inaccurate ☐ Inaccurate ☐ Accurate ☐ Very accurate ☐ Varies ☐ Don't know		
	What is the overall certainty of the evidence of test accuracy	☐ Very low ☐ Low ☐ Moderate ☐ High ☐ No included studies		
Benefits & harms of the options	How substantial are the desirable anticipated effects?	☐ Trivial ☐ Small ☐ Moderate ☐ Large ☐ Varies ☐ Don't know		
	How substantial are the undesirable anticipated effects?	☐ Large ☐ Moderate ☐ Small ☐ Trivial ☐ Varies ☐ Don't know		
	What is the overall certainty of the evidence for any critical or important direct benefits, adverse effects, or burden of the test?	☐ Very low ☐ Low ☐ Moderate ☐ High		

[1] The GRADE Working Group. The Evidence-to-Decision framework. In: Schünemann H, Brożek J, Guyatt G, Oxman A, eds. *GRADE handbook for grading quality of evidence and strength of recommendations.*2013.

		□ No included studies		
	What is the overall certainty of the evidence of effects of the management that is guided by the test results?	□ Very low □ Low □ Moderate □ High □ No included studies		
	How certain is the link between test results and management decisions?	□ Very low □ Low □ Moderate □ High □ No included studies		
	What is the overall certainty of the evidence of effects of the test?	□ Very low □ Low □ Moderate □ High □ No included studies		
	Is there important uncertainty about or variability in how much people value the main outcomes?	□ Important uncertainty or variability □ Possibly important uncertainty or variability □ Probably no important uncertainty or variability □ No important uncertainty or variability		
Resource use	How large are the resources requirement (costs)?	□ Large costs □ Moderate costs □ Negligible costs and savings □ Moderate savings □ Large savings □ Varies □ Don't know		
	What is the certainty of the evidence of resource requirements (costs)?	□ Favours the comparison □ Probably favours the comparison □ Does not favour either the intervention or the comparison □ Probably favours the intervention □ Favours the intervention □ Varies □ No included studies		
Equity	What would be the impact on health equity?	□ Reduced □ Probably reduced		

[1] The GRADE Working Group. The Evidence-to-Decision framework. In: Schünemann H, Brożek J, Guyatt G, Oxman A, eds. *GRADE handbook for grading quality of evidence and strength of recommendations.*2013.

		□ Probably no impact □ Probably increased □ Increased □ Varies □ Don't know		
Acceptability	Is the intervention acceptable to key stakeholders?	□ No □ Probably no □ Uncertain □ Probably yes □ Yes □ Varies □ Don't know		
Feasibility	Is the intervention feasible to implement?	□ No □ Probably no □ Uncertain □ Probably yes □ Yes □ Varies □ Don't know		
Balance of effects	Does the balance between desirable and undesirable effects favour the intervention or the comparison (no screening)?	□ Favours the comparison □ Probably favours the comparison □ Does not favour either the intervention or the comparison □ Probably favours the intervention		
Types of recommendation	□ Strong recommendation against the intervention □ Conditional recommendation against the intervention □ Conditional recommendation for either the intervention or the comparison □ Conditional recommendation for the intervention □ Strong recommendation for the intervention			
Conclusion				
Recommendation				
Justification				
Considerations				
Monitoring and evaluation				
Research priorities				

[1] The GRADE Working Group. The Evidence-to-Decision framework. In: Schünemann H, Brożek J, Guyatt G, Oxman A, eds. *GRADE handbook for grading quality of evidence and strength of recommendations.*2013.

Appendix C:
Supplementary material for systematic review

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

Database	Search strategy
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. 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
PsycINFO	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

Reference list for all studies include in the systematic review of studies examining FASD screening tools

1. Astley SJ, Clarren SK. A fetal alcohol syndrome screening tool. *Alcohol. Clin. Exp. Res*. 1995;19(6):1565-1571.
2. Astley SJ, Clarren SK. A case definition and photographic screening tool for the facial phenotype of fetal alcohol syndrome. *J. Pediatr*. 1996;129(1):33-41.
3. Astley SJ, Stachowiak J, Clarren SK, Clausen C. Application of the fetal alcohol syndrome facial photographic screening tool in a foster care population. *J. Pediatr*. 2002;141(5):712-717.
4. Breiner P, Nulman I, Koren G. Identifying the neurobehavioral phenotype of fetal alcohol spectrum disorder in young children. *J Popul Ther Clin Pharmacol.* 2013;20(3):e334-339.
5. Burd L, Cox C, Poitra B, et al. The FAS Screen: a rapid screening tool for fetal alcohol syndrome. *Addict. Biol.* 1999;4(3):329-336.
6. Burd L, Martsolf JT, Klug MG, Kerbeshian J. Diagnosis of FAS: a comparison of the fetal alcohol syndrome diagnostic checklist and the Institute of Medicine Criteria for fetal alcohol syndrome. *Neurotoxicol Teratol.* 2003;25(6):719-724.
7. LaFrance MA, McLachlan K, Nash K, et al. Evaluation of the neurobehavioral screening tool in children with fetal alcohol spectrum disorders (FASD). *J Popul Ther Clin Pharmacol.* 2014;21(2):e197-210.
8. Moore ES, Ward RE, Jamison PL, Morris CA, Bader PI, Hall BD. The subtle facial signs of prenatal exposure to alcohol: an anthropometric approach. *J. Pediatr.* 2001;139(2):215-219.
9. Nash K, Koren G, Rovet J. A differential approach for examining the behavioural phenotype of fetal alcohol spectrum disorders. *J Popul Ther Clin Pharmacol.* 2011;18(3):e440-453.
10. Nash K, Rovet J, Greenbaum R, Fantus E, Nulman I, Koren G. Identifying the behavioural phenotype in fetal alcohol spectrum disorder: sensitivity, specificity, and screening potential. *Arch Women Ment Health.* 2006;9(4):181-186.
11. Poitra BA, Marion S, Dionne M, et al. A school-based screening program for fetal alcohol syndrome. *Neurotoxicol Teratol.* 2003;25(6):725-729.
12. Thorne JC. Accentuate the negative: grammatical errors during narrative production as a clinical marker of central nervous system abnormality in school-aged children with fetal alcohol spectrum disorders. *J Speech Lang Hear Res.* 2017;60(12):3523-3537.
13. Tseng PH, Cameron IG, Pari G, Reynolds JN, Munoz DP, Itti L. High-throughput classification of clinical populations from natural viewing eye movements. *J Neurol.* 2013;260(1):275-284.
14. Zhang C, Paolozza A, Tseng PH, Reynolds JN, Munoz DP, Itti L. Detection of children/youth with fetal alcohol spectrum disorder through eye movement, psychometric, and neuroimaging data. *Front Neurol.* 2019;10:80.

Search strategy for FASD screening approaches

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: community child health practice or child health services or delivery of health care S12: S9 AND S10 AND S11 Limit to English language and yr="2010-Current"
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. child health care/ or community child health practice.mp. or community care/ 13. health care delivery/ 14. 12 or 13 15. 10 and 11 and 14 16. limit 15 to (English language and yr="2010-Current")
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. 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. screening.mp. or exp Mass Screening/ 12. community child health practice.mp. or Child Health Services/ or "delivery of health care"/ 13. 10 and 11 and 12 14. limit 13 to (English language and yr="2010 -Current")

Reference list for all studies include in the systematic review of studies examining FASD screening approaches

1. Burd L, Cohen C, Shah R, Norris J. A court team model for young children in foster care: The role of prenatal alcohol exposure and fetal alcohol spectrum disorders. *J Psychiatry Law*. 2011;39(1):179-191.
2. Hanlon-Dearman A, Green CR, Andrew G, LeBlanc N, Cook JL. Anticipatory guidance for children and adolescents with fetal alcohol spectrum disorder (FASD): practice points for primary health care providers. *J Popul Ther Clin Pharmacol.* 2015;22(1):e27-56.
3. O'Connor MJ, Rotheram-Borus MJ, Tomlinson M, Bill C, LeRoux IM, Stewart J. Screening for fetal alcohol spectrum disorders by nonmedical community workers. *J Popul Ther Clin Pharmacol.* 2014;21(3):e442-452.
4. Watkins RE, Elliott EJ, Halliday J, et al. A modified Delphi study of screening for fetal alcohol spectrum disorders in Australia. *BMC Pediatr.* 2013;13:13-13.

Appendix D:
Compilation of comments from the Advisory Group

	Concern	Comment from the Advisory Group
1	What are the potential benefits and harms of FASD screening?	Potential benefits: ▪ Early identification and diagnosis of children at risk of neurodevelopmental impairment leading to access to early intervention, family support and improved developmental outcomes for children. ▪ Prevention of subsequent PAE pregnancies through counselling and educating families for future pregnancies. ▪ Possible prevention of serious adverse outcomes such as youth suicide and crime. ▪ Limited benefit of FASD specific screening unless screening tools are effective and definitive, as well as being able to screen individuals at an early age. Potential harms: ▪ Screening needs to lead to diagnostic assessment and support services which are currently inadequate in the public sector. It is not ethical to do screening if screen positive individuals have no follow-on services or have to wait a very long time such as more than 3 months for assessment. This has potential psychological harm in causing anxiety and distress and shame. However, if there were increased resources for assessment and treatment, this would reduce the potential harm of screening activity. ▪ Incorrect diagnosis as symptoms correlate closely with other neurodevelopmental disorders could result in negative stigmatisation. PAE screening needs to be administered very early to get universal uptake e.g., by maternity services or first 8 weeks postnatally when engagement with services is at its highest. ▪ FASD screening is not yet able to be done with sufficient fidelity as the diagnostics and purported 'gold standard' is problematic. With this as the background it can be expected that screening for FASD is likely to disproportionately identify currently disproportionately disadvantaged minority groups. This will contribute to an overly simplistic understanding of the causes of developmental delay and the solutions, leading to an overshadowing of the actual determinants of health outcomes. This could lead to potential blame and shame for a mother, greater family discord, an unhelpful attitude towards the dysfunction that an individual may experiences, poorly spent public health money, and potentially unjustifiable and draconian infringements upon personal autonomy and freedoms and disempowerment (in cases where government may for instance decide to limit access to alcohol etc). The

	Concern	Comment from the Advisory Group
		ongoing research for effective FASD-specific interventions means that more general and symptomatic interventions are currently likely to be most useful. ▪ FASD screening may lead to misdiagnosis of FASD and potential to miss other neurodevelopmental diagnoses. It is important to note that cultural differences in different populations may not be captured by a generic screening tool.
2	Should the diagnostic term FASD be used?	▪ There is ample international evidence of high quality to support the existence of FASD and to attribute causality to PAE for developmental delay and neurodevelopmental disability in children both with and without facial features. This does not exclude other factors having a contribution or modifying the effects of alcohol exposure, such as poverty. However, there are several studies which have controlled for other factors, such as low maternal education, quality of care giving and home environment and low socioeconomic status, but still show significant effects on child development due to PAE. ▪ The term FASD should not be used while research is still being conducted to find ways to improve the identification, treatment, and prevention of neurodevelopmental harm related to PAE. There is a need to ensure that we have a robust diagnostic process with strong governance around it before the introduction of any form of screening process. ▪ There needs to be a clearer definition of FASD and guidelines for the identification of individuals with FASD. It is not appropriate for clinicians to diagnose an individual with FASD when they are not able to rule out PAE as a contributing factor to the neurodevelopmental impairments presented in the individual. Instead, they should consider differential diagnoses that can explain the neurodevelopmental impairments because neurodevelopmental impairments could be caused by other biological or psychosocial factors (e.g., trauma) and may not be caused by PAE. The implications of diagnosing an individual presented with neurodevelopmental impairments and undetermined PAE as an individual with FASD are that it would lead to an inaccurate implicit causal link of neurodevelopmental impairments to PAE and it could lead to a misdiagnosis of FASD. In circumstances where there is low confidence of identifying PAE as the cause of neurodevelopmental impairment, it may be useful to consider the use of multiaxial Z-codes, a special group of codes

	Concern	Comment from the Advisory Group
		provided in ICD-10 for the reporting of factors influencing health status and contact with health services, which minimise the misdiagnosis of the individual.
3	What are the potential benefits and harms of FASD diagnostic labels?	There is importance to weigh both the potential benefits and harms of FASD diagnosis. Potential benefits: ▪ Diagnostic labels underpin epidemiology – enable calculation of prevalence and incidence of FASD in Australia. ▪ Diagnostic labels allow research to recruit people with a specific label into epidemiological studies and clinical intervention trials and to compare results with other researchers. ▪ Diagnostic labels allow people who have that label or a family member with that label to form support groups e.g., Down syndrome support group, NOFASD and Russell Family Foundation - this breaks down the sense of isolation for sufferers and even empowers some of them. ▪ Diagnostic labels are required by our disability funding institutions e.g., NDIS - NDIS requires a diagnosis of a permanent disability. There is no funding e.g., no support for children (over 7 years) who do not have a label which is associated with permanent disability, e.g., there is no disability funding for ADHD and learning difficulties (excluding intellectual disability) or children with a background of trauma. ▪ Diagnostic labels allow people who support children with FASD to see from the child's perspective and not as labels of "misbehaving", thus resulting in frame-shifting and changing of attitudes to provide supportive environments for the children. See the works of Diane Malbin for example: http://www.fasdnetwork.org/uploads/9/5/1/1/9511748/educational_success_fas.pdf Potential harms: ▪ FASD diagnostic labels can create negative stigma. ▪ FASD diagnostic labels can draw attention away from the more significant underlying causes for developmental or behavioural challenges of the affected individual. E.g., prenatal exposure to drugs, environmental stressor - food insecurity, homelessness, attachment disorders, exposure to domestic violence,

	Concern	Comment from the Advisory Group
		sexual abuse, emotional abuse, physical abuse and neglect, chronic and recurrent infections of the skin, ears, chest and gut, or social and economic deprivation. The Advisory Group may be interested to know that a systematic review of the experiences (encompassing both negative and positive) of people with FASD and their families of the diagnostic process will be undertaken as part of the revision of the Australian FASD Assessment and Diagnostic Guideline.
4	How to reconcile the different FASD diagnostic systems and criteria in the literature and how does this impact on the formulation of recommendations for the implementation of FASD screening tool in Western Australia?	The continuum/spectrum of the FASD label could relate to the likelihood of PAE being the cause of the neurodevelopmental impairments. There is a need to define the diagnostic criteria of FASD, and the categories of FASD. In addition, it may be required to provide a clear statement that FASD can be given when there is a low probability of PAE being the primary cause.
5	How to embed the recommended FASD screening tool into a broader neurodevelopmental screening programme in Western Australia? How to integrate FASD screening into existing universal screening (e.g., Child and Adolescent Health Services, CAHS).	It is noted that the current completion rates for CAHS Community Health Birth to School Entry Assessments are: ▪ 0-14 days: 98% ▪ 8 weeks: 87% ▪ 4 months: 81% ▪ 1 year: 43% ▪ 2 Year: 27% ▪ School Entry Health Assessment (Kindy/PP): 70.6%
6	How to increase health screening coverage in children aged 2 years and above, and from diverse socioeconomic backgrounds.	There is a current CAHS 2-Year project running, examining ways to increase 2-year check uptake. The focus is currently increasing workforce capacity and engaging vulnerable groups. Also, there is a launch of Aboriginal Health Teams 1 and 2 Year Birthday cards.
7	There is a lack of cross-agency approach in the screening and management of the effects of FASD.	More information is needed from birth hospitals to community health on alcohol intake and other risk factors.
8	Is it necessary to ask about prenatal alcohol use during FASD screening/diagnostic assessment?	▪ Information on alcohol intake in pregnancy is currently not being passed to Community Health Nurses through the Stork Birth Notifications. ▪ Mothers should not be required to report their alcohol consumption in pregnancy for their child to access support.

	Concern	Comment from the Advisory Group
9	How to prevent clinicians from taking a "tick box" approach during FASD diagnosis?	• Neurodevelopmental diagnosis requires clinical judgement not just a blanket application of the diagnostic criteria. However, some clinicians do not demonstrate evidence of the application of clinical judgment in their diagnostic formulation but rather use a "tick-box approach" to the diagnosis of FASD. There may be some weaknesses in the diagnostic guideline and in the application of the diagnostic guideline which can lead to overdiagnosis of FASD. This would likely be exacerbated by the introduction of screening without improvements to the process of formal diagnosis. • Good clinical judgement is needed to reduce the risk of FASD misdiagnosis. One area of judgement would be based on accurate assessments of the neurodevelopmental domains associated with FASD, e.g., motor skills, cognition, executive functioning. Also, an area of judgement would be to determine if impairments in the neurodevelopmental domains are due to other causes other than PAE. There is a need to be cautious with the mechanistic approach to FASD diagnosis as it could lead to misdiagnosis. For example, where an individual who displays difficulties in neurodevelopmental domains, may be labelled with a FASD diagnosis, but it may not be due to PAE. • It is vital to ensure the integrity of a FASD diagnosis and that it does not become a 'catch all' condition that is applied, particularly for people in vulnerable populations. There is a need to acknowledge that individuals with FASD and their families can experience benefits from receiving a diagnosis and this should also be considered as part of the discussion. A balanced approach, through being open-minded to both potential risks and potential benefits could lead to the best outcomes for clients.
10	What are the specific trainings to screen, diagnose and manage FASD available in Australia?	▪ Very few specific training approaches are available in Australia due to lack of funding to fund research and thus build the evidence-based. ▪ It is important to consider if there is a need for further training and accreditation for FASD screening and diagnosis. Is it necessary to do a Graduate diploma to be a "FASD assessor" or would an on-line course suffice or is it something in between – accreditation to conduct a neurodevelopmental assessment? E.g., the Griffiths 3 Mental Development Scales, an evidence-based developmental assessment tool.

	Concern	Comment from the Advisory Group
		This involves an on-line course with built-in assessment questions, then a 3 day practical course, then the practitioner assesses a minimum of 5 cases and the reports are reviewed/ graded by the Griffiths tutor, before being accredited as a "Griffiths practitioner".
11	What are the implications of FASD diagnosis and the National Disability Insurance Scheme (NDIS)?	▪ Diagnostic labels are required by disability funding institutions e.g., NDIS. NDIS requires a diagnosis of a permanent disability. There is no funding e.g., no support for children (over 7 years) who do not have a diagnostic label which is associated with permanent disability, e.g., there is no disability funding for ADHD and learning difficulties (excluding intellectual disability) or children with a background of trauma. ▪ If NDIS and others disability funding institution require a specific diagnosis, then a diagnostic term that would include all of those who are currently diagnosed with FASD and those who have the same neurodevelopmental impairments but do not have PAE as a causal factor is required. ▪ Inaccurate diagnosis of FASD in order to access NDIS funding for the individual could lead to a reduction in private health insurance uptake and a greater heterogeneity within the diagnostic entities. Clinicians should be responsible to provide accurate diagnosis and not subvert the NDIS system by providing an inaccurate diagnosis so that the individual can meet the criteria to access services that the clinicians feel that the individual should receive.

Appendix E:
GRADE evidence profiles of screening tools

Supplementary table 1. Evidence profile for the Neurobehavioral Screening Test

Sensitivity	Specificity	Prevalence
0.63 to 0.98	0.42 to 1.00	0.77%

Outcome	Nº of studies (Nº of patients)	Study design	Factors that may decrease certainty of evidence					Effect per 1,000 patients tested	Quality of evidence
			Risk of bias	Indirectness	Inconsistency	Imprecision	Publication bias	pre-test probability of 0.77%	
True positives (patients with FASD)	4 studies 288 patients	case-control type accuracy study	serious[a]	not serious	serious[b]	not serious	none	5 to 8	Low
False negatives (patients incorrectly classified as not having FASD)								0 to 3	
True negatives (patients without FASD)	4 studies 288 patients	case-control type accuracy study	serious[a]	not serious	serious[b]	not serious	none	417 to 992	Low
False positives (patients incorrectly classified as having FASD)								0 to 575	

Explanations

a. Risk of bias was assessed using QUADAS-2. Studies demonstrated high risk of bias in patient selection due to the study design and high risk of bias in study flow due to only some participants received both index and reference tests.

b. Estimates of the Neurobehavioral Screening Test sensitivity and specificity were variable despite similar cut-off values and could not be explained by the quality of studies.

Supplementary table 2. Evidence profile for the eye movement behaviour assessment via machine learning

Sensitivity	Specificity	Prevalence
0.73 to 0.77	0.79 to 0.91	0.77%

Outcome	№ of studies (№ of patients)	Study design	Factors that may decrease certainty of evidence					Effect per 1,000 patients tested	Quality of evidence
			Risk of bias	Indirectness	Inconsistency	Imprecision	Publication bias	pre-test probability of 0.77%	
True positives (patients with FASD)	2 studies 259 patients	case-control type accuracy study	serious[a]	not serious	not serious	not serious	none	6 to 6	Moderate
False negatives (patients incorrectly classified as not having FASD)								2 to 2	
True negatives (patients without FASD)	2 studies 259 patients	case-control type accuracy study	serious[a]	not serious	not serious	not serious	none	784 to 903	Moderate
False positives (patients incorrectly classified as having FASD)								89 to 208	

Explanations
a. Risk of bias was assessed using QUADAS-2. Studies demonstrated high risk of bias in patient selection due to the study design and high risk of bias in study flow due to only some participants received both index and reference tests.

Supplementary table 3. Evidence profile for the Tallying Reference Errors in Narrative task

Sensitivity	Specificity	Prevalence
0.54	0.96	0.77%

Outcome	№ of studies (№ of patients)	Study design	Factors that may decrease certainty of evidence					Effect per 1,000 patients tested	Quality of evidence
			Risk of bias	Indirectness	Inconsistency	Imprecision	Publication bias	pre-test probability of 0.77%	
True positives (patients with FASD)	1 study 138 patients	case-control type accuracy study	serious[a]	not serious	not serious	not serious	none	4	Moderate
False negatives (patients incorrectly classified as not having FASD)								4	
True negatives (patients without FASD)	1 study 138 patients	case-control type accuracy study	serious[a]	not serious	not serious	not serious	none	953	Moderate
False positives (patients incorrectly classified as having FASD)								39	

Explanations
a. Risk of bias was assessed using QUADAS-2. Studies demonstrated high risk of bias in patient selection due to the study design and high risk of bias in study flow due to only some participants received both index and reference tests.

Supplementary table 4. Evidence profile for the dysmorphic examination via photographs

Sensitivity	Specificity	Prevalence
1.00 to 1.00	0.99 to 1.00	0.77%

Outcome	№ of studies (№ of patients)	Study design	Factors that may decrease certainty of evidence					Effect per 1,000 patients tested	Quality of evidence
			Risk of bias	Indirectness	Inconsistency	Imprecision	Publication bias	pre-test probability of 0.77%	
True positives (patients with FASD)	2 studies 726 patients	cohort & case-control type studies	serious[a]	very serious[b]	not serious	not serious	none	8 to 8	Very low
False negatives (patients incorrectly classified as not having FASD)								0 to 0	
True negatives (patients without FASD)	2 studies 726 patients	cohort & case-control type studies	serious[a]	very serious[b]	not serious	not serious	none	982 to 992	Very low
False positives (patients incorrectly classified as having FASD)								0 to 10	

Explanations
a. Risk of bias was assessed using QUADAS-2. Studies demonstrated high risk of bias in patient selection due to the study design and high risk of bias in study flow due to only some participants received both index and reference tests.

b. Indirectness was assessed using QUADAS-2. Studies demonstrated high risk in applicability in patient selection, screening tool and reference standard due to the target group, screening tool and reference test concerning only individuals with fetal alcohol syndrome but not those with fetal alcohol spectrum disorder.

Supplementary table 5. Evidence profile for the physical and dysmorphic examination

Sensitivity	Specificity	Prevalence
1.00 (95% CI: 0.91 to 1.00)	0.89 (95% CI: 0.83 to 0.94)	0.77%

Outcome	№ of studies (№ of patients)	Study design	Factors that may decrease certainty of evidence					Effect per 1,000 patients tested	Quality of evidence
			Risk of bias	Indirectness	Inconsistency	Imprecision	Publication bias	pre-test probability of 0.77%	
True positives (patients with FASD)	1 study 194 patients	cross-sectional (cohort type accuracy study)	not serious	very serious[a]	not serious	not serious	none	8 (7 to 8)	Low
False negatives (patients incorrectly classified as not having FASD)								0 (0 to 1)	
True negatives (patients without FASD)	1 study 194 patients	cross-sectional (cohort type accuracy study)	not serious	very serious[a]	not serious	not serious	none	883 (824 to 933)	Low
False positives (patients incorrectly classified as having FASD)								109 (59 to 168)	

Explanations
a. Indirectness was assessed using QUADAS-2. Studies demonstrated high risk in applicability in patient selection and reference standard due to the target group and reference testing concerning only individuals with fetal alcohol syndrome but not those with fetal alcohol spectrum disorder.

Supplementary table 6. Evidence profile for the craniofacial measurements approach

Sensitivity	Specificity	Prevalence
0.98 (95% CI: 0.93 to 1.00)	0.90 (95% CI: 0.74 to 0.98)	0.77%

Outcome	№ of studies (№ of patients)	Study design	Factors that may decrease certainty of evidence					Effect per 1,000 patients tested	Quality of evidence
			Risk of bias	Indirectness	Inconsistency	Imprecision	Publication bias	pre-test probability of 0.77%	
True positives (patients with FASD)	1 study 129 patients	cross-sectional (cohort type accuracy study)	very serious[a]	serious[b]	not serious	not serious	none	8 (7 to 8)	Very low
False negatives (patients incorrectly classified as not having FASD)								0 (0 to 1)	
True negatives (patients without FASD)	1 study 129 patients	cross-sectional (cohort type accuracy study)	very serious[a]	serious[b]	not serious	not serious	none	893 (737 to 972)	Very low
False positives (patients incorrectly classified as having FASD)								99 (20 to 255)	

Explanations

a. Risk of bias was assessed using QUADAS-2. Studies demonstrated high risk of bias in patient selection due to the study design, high risk of bias in screening tool due to lack of assessor blinding, and high risk of bias in study flow due to only some participants received both index and reference tests.

b. Indirectness was assessed using QUADAS-2. Studies demonstrated high risk in applicability in patient selection due to the target group concerning only individuals with fetal alcohol syndrome but not those with fetal alcohol spectrum disorder.

Supplementary table 7. Evidence profile for the FAS Screen

Sensitivity	Specificity	Prevalence
1.00 to 1.00	0.94 to 0.95	7.7%

Outcome	№ of studies (№ of patients)	Study design	Factors that may decrease certainty of evidence					Effect per 1,000 patients tested	Quality of evidence
			Risk of bias	Indirectness	Inconsistency	Imprecision	Publication bias	pre-test probability of 0.77%	
True positives (patients with FASD)	2 studies 2397 patients	cross-sectional (cohort type accuracy study)	not serious	very serious[a]	not serious	not serious	none	8 to 8	Low
False negatives (patients incorrectly classified as not having FASD)								0 to 0	
True negatives (patients without FASD)	2 studies 2397 patients	cross-sectional (cohort type accuracy study)	not serious	very serious[a]	not serious	not serious	none	934 to 947	Low
False positives (patients incorrectly classified as having FASD)								42 to 58	

Explanations
a. Indirectness was assessed using QUADAS-2. Studies demonstrated high risk in applicability in patient selection, screening tool and reference standard due to the screening tool and reference test concerning only individuals with fetal alcohol syndrome but not those with fetal alcohol spectrum disorder.

Supplementary table 8. Evidence profile for the FAS diagnostic checklist

Sensitivity	Specificity	Prevalence
0.89	0.72	0.77%

Outcome	№ of studies (№ of patients)	Study design	Factors that may decrease certainty of evidence					Effect per 1,000 patients tested	Quality of evidence
			Risk of bias	Indirectness	Inconsistency	Imprecision	Publication bias	pre-test probability of 0.77%	
True positives (patients with FASD)	1 study 352 patients	cross-sectional (cohort type accuracy study)	not serious	serious[a]	not serious	not serious	none	7	Moderate
False negatives (patients incorrectly classified as not having FASD)								1	
True negatives (patients without FASD)	1 study 352 patients	cross-sectional (cohort type accuracy study)	not serious	serious[a]	not serious	not serious	none	711	Moderate
False positives (patients incorrectly classified as having FASD)								281	

Explanations
a. Indirectness was assessed using QUADAS-2. Studies demonstrated unclear risk in patient selection due to the inclusion of only individuals with fetal alcohol syndrome and partial fetal alcohol syndrome, and high risk in applicability in reference standard due to the reference test concerning only individuals with fetal alcohol syndrome but not those with fetal alcohol spectrum disorder.