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ORIGINAL ARTICLE

Prevalence of fetal alcohol syndrome in a population-based sample of children living in remote Australia: The Lililwan* Project

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Aim: Aboriginal leaders concerned about high rates of alcohol use in pregnancy invited researchers to determine the prevalence of fetal alcohol syndrome (FAS) and partial fetal alcohol syndrome (pFAS) in their communities.

Methods: Population-based prevalence study using active case ascertainment in children born in 2002/2003 and living in the Fitzroy Valley, in Western Australia (April 2010–November 2011) (n = 134). Socio-demographic and antenatal data, including alcohol use in pregnancy, were collected by interview with 127/134 (95%) consenting parents/care givers. Maternal/child medical records were reviewed. Interdisciplinary assessments were conducted for 108/134 (81%) children. FAS/pFAS prevalence was determined using modified Canadian diagnostic guidelines.

Results: In 127 pregnancies, alcohol was used in 55%. FAS or pFAS was diagnosed in 13/108 children, a prevalence of 120 per 1000 (95% confidence interval 70–196). Prenatal alcohol exposure was confirmed for all children with FAS/pFAS, 80% in the first trimester and 50% throughout pregnancy. Ten of 13 mothers had Alcohol Use Disorders Identification Test scores and all drank at a high-risk level. Of children with FAS/pFAS, 69% had microcephaly, 85% had weight deficiency and all had facial dysmorphology and central nervous system abnormality/impairment in three to eight domains.

Conclusions: The population prevalence of FAS/pFAS in remote Aboriginal communities of the Fitzroy Valley is the highest reported in Australia and similar to that reported in high-risk populations internationally. Results are likely to be generalisable to other age groups in the Fitzroy Valley and other remote Australian communities with high-risk alcohol use during pregnancy. Prevention of FAS/pFAS is an urgent public health challenge.

Key words: Aboriginal Australian; alcohol-related disorder; fetal alcohol spectrum disorder (FASD); fetal alcohol syndrome (FAS); prevalence study.

What is already known on this topic

1 Although national guidelines advise against drinking during pregnancy, half of Australian women reported drinking during pregnancy in the recent (2010) national household survey.
2 Indigenous women are less likely than non-Indigenous women to drink during pregnancy, but many who do so drink frequently and at high-risk levels.
3 Alcohol exposure in pregnancy is a leading preventable cause of intellectual disability, developmental delay and birth defects and can cause fetal alcohol syndrome (FAS) and partial FAS (pFAS).

What this paper adds

1 This paper provides Australia's first population-based data on the prevalence of FAS and pFAS.
2 This paper identifies the significant burden of FAS and pFAS in remote Aboriginal communities in the Fitzroy Valley in north Western Australia and the need for prevention strategies.
3 This paper is an exemplar of research initiated and led by Aboriginal communities.

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*Lililwan is a Kimberley Kriol word meaning 'all the little ones'.

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Alcohol is a teratogen that may cause fetal alcohol spectrum disorders (FASD), including fetal alcohol syndrome (FAS) and fetal alcohol syndrome with partial physical features (pFAS).[1] FAS and pFAS are characterised by facial dysmorphology, structural or functional abnormalities of the central nervous system (CNS) and, in some cases (FAS), growth deficiency.[1] FASD are leading causes of preventable intellectual disabilities, costing up to $5.4 billion per year in health and education services in the USA.[2] Australia has among the highest alcohol consumption in the world (10.4 litres of pure alcohol per annum per person ≥15 years old),[3] and between 51% and 60% of Australian women report alcohol use in pregnancy.[4,5] Indigenous Australian women are less likely than non-Indigenous women to drink in pregnancy but are more likely to drink at high-risk levels.[4,6]

Most FASD prevalence studies report only FAS/pFAS as these diagnoses are most strongly associated with prenatal alcohol exposure. A recent report published by the Alberta Institute of Health Economics included a meta-analysis of cross-sectional and cohort studies, and reports pooled prevalence rates of 0.006–3.0 per 1000 for FAS, and 0.5–5.3 per 1000 for pFAS in the general population.[7] Higher prevalence rates are observed in populations at high risk of prenatal alcohol exposure, including children in foster care (pooled prevalence 210.0 per 1000 for FAS). No pooled prevalence data are available for pFAS in foster care as there are only three small heterogeneous studies available.[7] Based on a small number of studies, prevalence rates are also high for youth in the juvenile justice system (10.4 per 1000 for FAS; 180.0 per 1000 for pFAS).[7] Pooled prevalence rates in Indigenous communities in Canada, the USA and Australia are 2.0 per 1000 for FAS and 1.3 to 39.0 per 1000 for pFAS.[7] High prevalence rates have also been documented in a South African community where high-risk drinking is common. FAS or pFAS was diagnosed in 104.6–160.6 per 1000 school-aged children.[8] In Australia, it is likely that FAS/pFAS prevalence, currently reported as 0.01–0.68 per 1000 births, has been underestimated.[9–12] The variation in prevalence rates and low rates currently reported in Australia may be due to under-diagnosis, under-reporting, lack of prenatal alcohol exposure data, inconsistent use of diagnostic criteria, differing study methods, lack of active case ascertainment and under-representation of high-risk populations.

Alcohol use in pregnancy and FASD occur in both non-Indigenous and Indigenous communities; however, there are communities in which rates of drinking are high and high FASD prevalence might be expected. In the very remote Aboriginal communities of the Fitzroy Valley in the Kimberley region of north Western Australia, high-risk alcohol use is common.[13] Aboriginal community leaders became concerned that learning and behavioural problems caused by prenatal alcohol exposure might disrupt intergenerational transfer of language and culture. To better understand this problem, they invited researchers to collaborate in Australia's first population-based study of FASD prevalence, the Lililwan Project.[14] 'Lililwan' is a Kimberley Kriol word meaning 'all the little ones'.

The study objective was to determine the prevalence of FAS and pFAS in all children born in 2002 and 2003 and living in the Fitzroy Valley during the study period.

Methods

Design, setting and participants

A population-based study was conducted using active case ascertainment to determine FAS/pFAS prevalence (see published protocol).[14] The study involved structured interviews of parents/care givers and case note review (stage 1) and interdisciplinary health and developmental assessments of all children (stage 2). STROBE guidelines for reporting observational studies were used.[15] The study was conducted in the Fitzroy Valley, 2500 km north of Perth, Western Australia, comprising Fitzroy Crossing town and 45 very remote communities located within 200 km of the town.[16] Approximately 80% of the population of 4500 is Aboriginal.[16] All children born in 2002 or 2003, living in the Fitzroy Valley between April 2010 and November 2011, were eligible (n = 134). Children were identified from local population and health databases.[14] Of 134 eligible children 127 (95%) parents/care givers consented to participate in stage 1 and 108 children (81%) participated in stage 2 (Fig. 1).

Data collection and outcome measures

During stage 1 (April–November 2010), 127 parents/care givers were interviewed using a structured, reliable questionnaire[17] developed by paediatricians and local Aboriginal language experts. Antenatal and child health records were reviewed. Information collected included demographics, language group, schooling, family and community characteristics, prenatal

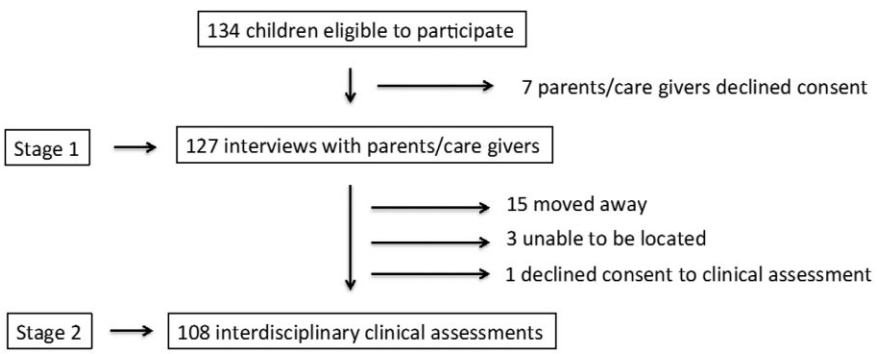

Fig. 1 Participant flowchart in a population-based cohort of children born in 2002/2003 and living in the Fitzroy Valley in 2010/2011.

exposures (including alcohol, cigarettes and other drugs), pregnancy complications, birth and neonatal history, early life trauma, growth, health, development, and educational outcomes.[17] The consumption subset of the Alcohol Use Disorders Identification Test (AUDIT-C) was used as a proxy measure of fetal alcohol exposure risk.[18] AUDIT-C scores are derived from the frequency and quantity of alcohol intake and occurrences of 'binge' drinking. The maximum possible AUDIT-C score is 12, and risk categories include low risk (score 0–3), risky (score 4–5) and high risk (score ≥6). An AUDIT-C score of 0 indicates no alcohol consumption. Minor modifications were made to the wording of AUDIT-C items in our questionnaire so that they were easily understood by participants with English as a second or subsequent language.

In stage 2 (May–November 2011), interdisciplinary assessments of each child were conducted over three non-consecutive days. The clinical team included experienced paediatricians, child/school psychologists, speech and language pathologists, an occupational therapist, and paediatric physiotherapist, trained to use the Canadian FASD diagnostic guidelines.[1] The choice of assessment tools was informed by literature review and consultation with Aboriginal community members, local clinicians and national and international researchers.[14]

Growth impairment was defined as weight and/or height ≤10th percentile for age that was not attributable to nutrition or other known causes. Impairment in CNS domains was based on conservative cut-offs (≥2 standard deviations (SD) from the mean, or 3rd/97th percentile) when standardised scores were available (Table 1). When standardised scores were not available, or when multiple tests were used to assess a domain, or when there was variability between results of sub-tests, a conservative approach to assigning impairment was taken. All data from assessments were subsequently reviewed by clinicians to ensure adherence to predetermined criteria for impairment.

Clinicians were blinded to prenatal alcohol exposure until the presence/absence of facial characteristics, growth restriction and CNS abnormalities was determined and reported during an interdisciplinary case conference for each child. The Canadian FASD diagnostic guidelines, modified for use in remote Aboriginal communities,[1,14] were then used to assign the diagnosis of FAS or pFAS (Table 1). Differential diagnoses[1] and potential confounders (central auditory processing disorder, chronic suppurative otitis media, hearing loss, poor school attendance and early life trauma) were considered during the clinical assessment and diagnostic process.[14,17]

Feedback to participants included a report outlining assessment results, diagnoses and a management plan including home and school-based strategies. This was provided in writing and explained to each participant's family and, with their consent, provided to relevant health professionals and teachers. Follow-up support from a FASD educator and social worker/counsellor was provided for children and families in whom a diagnosis of FAS/pFAS was made. Immediate treatment was provided for children with acute medical problems such as otitis media, skin sores and respiratory infections. Treatment and/or referral were provided for all other diagnoses made during assessments. Professional development training about FASD was provided to teachers at participating community schools and local health service staff.

Table 1 Case definitions for fetal alcohol syndrome (FAS) and partial FAS (pFAS)[1]

Fetal alcohol syndrome
- Growth deficiency (weight or height ≤10th percentile at any age) *and*
- Three characteristic FAS facial features: short palpebral fissure length (≥2SD below the mean using the Hall charts),[19] thin upper lip and smooth philtrum† (rank 4 or 5 using the University of Washington lip-philtrum guide)[20] *and*
- Evidence of abnormality/impairment‡ in three or more of the central nervous system domains listed below
- With or without confirmed alcohol exposure

Partial fetal alcohol syndrome
- Two of the characteristic FAS facial features described above† *and*
- Evidence of abnormality/impairment‡ in three or more of the central nervous system domains listed below *and*
- Confirmed alcohol exposure

Central nervous system domains assessed
- CNS structure including microcephaly or other structural CNS abnormality§
- Hard and/or soft neurological signs; seizure disorder; gross and/or fine motor functioning (± articulation, phonology and motor speech)
- Cognition – cognitive delay and/or uneven cognitive profile
- Memory – auditory and/or visual
- Executive functioning and abstract reasoning
- Communication – expressive and/or receptive language
- Attention deficit/hyperactivity (± other behavioural problems ± sensory function)
- Visual motor integration¶
- Adaptive behaviour/social skills/social communication
- Academic achievement

†Lip-philtrum guide 1 (Caucasian norms) was used for non-Aboriginal participants; Lip-philtrum guide 2 (African American norms) for Aboriginal participants. In contrast to the University of Washington FASD 4-Digit Diagnostic Code, the Canadian guidelines accept two of three characteristic facial features in the diagnosis of pFAS. ‡Impairment in functional areas in parentheses alone was not sufficient to be deemed a 'domain of impairment'. Where standardised data were available, ≥2SD from the mean or 3rd/97th percentile was the cut-off for impairment. When standardised scores were not available, or when multiple tests were used to assess a domain, or when there was variability between results of sub-tests, a conservative approach to assigning impairment was taken. §Microcephaly defined as head circumference ≤3rd percentile at any age. In accordance with the Canadian FASD diagnostic guidelines, CNS structural abnormality or microcephaly constituted only a single domain of abnormality/impairment. ¶In this study, impairment in visual motor integration was sufficient to be deemed a 'domain of impairment'.

Statistical analysis

Descriptive analyses were performed to obtain frequencies, means and prevalence estimates using (IBM SPSS Statistics for Windows, Version 20.0.2 Armonk, NY, USA). Valid percentages are reported. Given the small sample size, only predetermined, biologically plausible or clinically relevant associations were

investigated. Chi-squared distributions were used to examine associations between hearing impairment and FAS/pFAS diagnosis.

Ethics

Ethics approval was granted by the University of Sydney Human Research Ethics Committee (No. 12527); the Western Australian Aboriginal Health Information and Ethics Committee (No. 271-01/10); the Western Australian Country Health Service Board Research Ethics Committee (No. 2010:01); and the Kimberley Aboriginal Health Planning Forum Research Sub-committee (No. 2010-001). Written informed consent was obtained from all participants at three time points during the study.

Community consultation

This study was initiated and led by Aboriginal community leaders who comprised almost half the project team, two as chief investigators. Extensive consultation was undertaken prior to the study to gain community consent,[21] and Aboriginal leaders provided cultural guidance to the research team throughout the study. Kimberley Kriol was the preferred language, but most participants also spoke Australian English and one or more local Aboriginal languages. Assessment tools were administered in consultation with local Aboriginal people to minimise bias by culture and language. Some non-verbal assessments were used, and assessment of communication took into account the multilingual and cultural context.

Results

Characteristics of index pregnancy and birth

Of 127 mothers, 95% were Indigenous; the mean age during the index pregnancy was 25.5 ± 6.7 years (range: 14–43 years), most lived in very remote communities and 55% drank alcohol during the index pregnancy (Table 2). An AUDIT-C score was calculated for 115/127 pregnancies. Of the 60 women with an AUDIT-C score who drank alcohol in pregnancy, 87% drank at a high-risk level.

Table 2 Characteristics of the index pregnancy and birth of children born in 2002/2003 and living in the Fitzroy Valley 2010/2011

Pregnancy characteristics	Study cohort *n* = 127 *n* (%)
Maternal age during pregnancy (*n* = 127) – mean years ± SD (range)	25.5 ± 6.7 (14–43)
Indigenous (mother)† (*n* = 127)	121 (95)
Place of residence during pregnancy (*n* = 114)	
Major town/inner regional/outer regional/remote‡	13 (11)
Very remote‡§	101 (89)
Antenatal care received (*n* = 107)	107 (100)
Maternal alcohol use during pregnancy (*n* = 122)	67 (55)
High-risk alcohol use in pregnancy (*n* = 60 with AUDIT-C ≥ 1)	52 (87)
Other exposures (*n* = 119)	
Cigarettes	76 (64)
Marijuana	16 (13)
Both cigarettes and marijuana	16 (13)
Birth characteristics	
Preterm birth (<37 weeks) (*n* = 118)	15 (13)
Birthweight (*n* = 105)	
Very low birthweight (<1500 g)	3 (3)
Low birthweight (<2500 g)	18 (17)
≤3rd percentile¶	2 (2)
≤10th percentile¶	11 (11)
Birth length (*n* = 98)	
≤3rd percentile¶	2 (2)
≤10th percentile¶	5 (5)

†Aboriginal and/or Torres Strait Islander and/or South Sea/Pacific Islander. ‡Australian Bureau of Statistics remoteness categories based on road distance measurements from over 12 000 populated localities to the nearest service centres. §Fitzroy Valley Population Project (2010).[16] ¶Corrected for sex and gestational age. For birthweight and birth length, ≤10th percentile includes those ≤3rd percentile.

Characteristics of children undergoing clinical assessment

Of 108 children assessed, most were Indigenous (98%), 53% were male and the mean age at assessment was 8.7 ± 0.6 years (range: 7.5–9.6 years) (Table 3). Weight deficiency (≤10th percentile) at any age was documented in 25% and height deficiency (≤10th percentile) in 13%. Microcephaly (≤3rd percentile) was documented in 15% of children. A history of chronic suppurative otitis media (44%) and any hearing loss (55%) was common. A diagnosis of FAS ($n = 1$) or pFAS ($n = 12$) was made in 13 of 108 children assessed, giving a prevalence of 120 per 1000 (95% confidence interval (CI) 70–196).

Characteristics of children with FAS/pFAS

All 13 children with FAS/pFAS were prenatally exposed to alcohol. First trimester alcohol exposure (which may also include exposure in other trimesters) was reported in 80%, and 50% were exposed in all trimesters. AUDIT-C scores could be calculated for mothers of 10 children with FAS/pFAS, indicating all were drinking at a high-risk level. The median AUDIT-C score was 10 (range: 8–12). One mother drank daily/almost daily, five mothers drank two to three times per week and four mothers drank once per week or once every 2 weeks. On a typical drinking day, nine of the 10 mothers consumed at least 14 standard drinks, and one consumed seven or more standard drinks. Seven reported drinking more than six drinks on an occasion at least weekly during the pregnancy.

Of the 13 children with FAS/pFAS, gestation was known for 11, of whom 36% were preterm. Weight deficiency (85%) or height deficiency (46%) at any age was common (Table 3). One child had all three FAS facial features, and the rest had two features. Microcephaly was recorded in 69%. The mean number of domains of abnormality/impairment in CNS structure and/or function was 4.7 ± 1.8 (range: 3–8). The most common functional impairments were attention deficit hyperactivity disorder (ADHD) with or without sensory dysfunction (69%), academic

Table 3 Clinical assessment outcomes of 108 children born in 2002/2003 and living in the Fitzroy Valley 2010/2011

Assessment outcome	All children assessed *n* = 108 *n* (%)	FAS/pFAS group *n* = 13 *n* (%)	Non-FAS/pFAS group *n* = 95 *n* (%)
Child's age at assessment – mean years ± SD (range)	8.7 ± 0.6 (7.5–9.6)	8.5 ± 0.6 (7.6–9.6)	8.7 ± 0.6 (7.5–9.6)
Indigenous (child)†	106 (98)	13 (100)	93 (98)
Sex (male)	57 (53)	7 (54)	50 (53)
Place of residence at time of assessment			
Very remote‡	108 (100)	13 (100)	95 (100)
Remote town (pop. 1000–9999)	34 (32)	3 (23)	31 (33)
Outer suburbs (within 30 km of remote town)	21 (19)	3 (23)	18 (19)
Sub-regional hub (pop. 200–999)	27 (25)	3 (23)	24 (25)
Remote satellite community (pop. <200)	26 (24)	4 (31)	22 (23)
Weight deficiency at any age (*n* = 108)			
≤3rd percentile	12 (11)	6 (46)	6 (6)
≤10th percentile	27 (25)	11 (85)	16 (17)
Height deficiency at any age (*n* = 108)			
≤3rd percentile	6 (6)	3 (23)	3 (3)
≤10th percentile	14 (13)	6 (46)	8 (8)
Microcephaly (≤3rd percentile) (*n* = 108)	16 (15)	9 (69)	7 (7)
Cardinal facial features of FAS (*n* = 108)			
Short palpebral fissures (PFLs) ≤2SD§	23 (21)	7 (54)	16 (17)
Smooth philtrum – rank 4 or 5§	33 (31)	9 (69)	24 (25)
Thin upper lip – rank 4 or 5§	39 (36)	11 (85)	28 (30)
Number of FAS facial features			
0	43 (40)	0 (0)	43 (45)
1	36 (33)	0 (0)	36 (38)
2	28 (26)	12 (92)	16 (17)
3	1 (1)	1 (8)	0 (0)
Documented ear/hearing/auditory processing problem¶			
Chronic suppurative otitis media (*n* = 103)	45 (44)	8 (62)	37 (41)
Any hearing loss (*n* = 93)	51 (55)	7 (54)	44 (55)
Mild hearing loss	38 (41)	6 (46)	32 (40)
Moderate hearing loss	13 (14)	1 (8)	12 (15)
Central auditory processing disorder (*n* = 84)	10 (12)	0 (0)	10 (13)
Reduced visual acuity†† (*n* = 99)	11 (11)	3 (25)	8 (9)
FAS or pFAS diagnosis (*n* = 108)	13 (12)	13 (100)	0 (0)

†Aboriginal and/or Torres Strait Islander and/or South Sea/Pacific Islander. ‡Australian Bureau of Statistics remoteness categories based on road distance measurements from over 12 000 populated localities to the nearest service centres. Remote town, outer suburbs, sub-regional hub and satellite communities classified according to the Fitzroy Valley Population Project (2010).[16] §Short PFL (≥2SD below the mean using the Hall[19] charts). Smooth philtrum and thin upper lip (rank 4 or 5 using the University of Washington[20] lip-philtrum guide). ¶Conditions documented in medical records (prior history) or detected at the time of clinical assessment. Chronic suppurative otitis media defined as perforated tympanic membrane and otorrhoea for ≥2 weeks. Mild hearing loss 26–40 dB, Moderate hearing loss 41–55 dB. Auditory processing assessed using the LISN-S test.[22] ††Reduced visual acuity defined as ≤6/9 in one or both eyes using culturally appropriate optotypes. FAS, fetal alcohol syndrome; pFAS, fetal alcohol syndrome with partial physical features.

achievement (62%), communication (54%), cognition (50%), memory (50%) and executive functioning (50%).

Characteristics of children without FAS/pFAS

In 95 children without FAS/pFAS, microcephaly, weight deficiency and height deficiency were less common than in children with FAS/pFAS. The mean number of domains of CNS abnormality/impairment was 1.7 ± 1.5 (range: 0–7). Similar rates of hearing loss (mild or moderate) were recorded in children with (54%) and without (55%) a FAS/pFAS diagnosis (P = 0.94). Of the 95 children, 45% had no FAS facial features, 38% had one feature and 17% had two features.

Discussion

FAS/pFAS was diagnosed in 120 per 1000 (95% CI 70–196) children in the Fitzroy Valley born in 2002 and 2003, among the highest prevalence world-wide. The high FAS/pFAS prevalence is consistent with alcohol consumption during pregnancy, which was documented in all children diagnosed with FAS/pFAS for whom maternal AUDIT-C scores could be calculated. This

prevalence rate, in a geographically isolated population, is considerably higher than previously reported in the general population or other Indigenous communities in Australia[9–12] and is similar to rates previously reported in high-risk groups internationally.[7,8] Findings are likely to be generalisable to other age cohorts within this community born prior to alcohol-restrictions being introduced and to other populations across Australia in which there are similarly high rates of prenatal alcohol exposure.

Comparison of prevalence between international studies is difficult due to use of a number of different diagnostic criteria and different study designs.[1,8,20] In this study, the Canadian FASD diagnostic guidelines were applied following minor modifications, because they provide a practical approach to diagnosis using well-defined case definitions.[1] The cut-offs used in this study for functional impairment and palpebral fissure length (PFL)/microcephaly (≥2SD from the mean, or 3rd/97th percentile, or an approach appropriate to the defining data) are generally conservative relative to those used in other studies. By comparison, high prevalence rates for FAS/pFAS obtained in South Africa (104.6–160.6 per 1000)[8] and Italy (45.1 per 1000)[23] were derived using a 10th percentile cut-off for PFL and microcephaly. Application of the ≤10th percentile cut-off for PFL and microcephaly in the 108 children in our study would add six children with 'microcephaly' and 26 children with 'short PFL'. Furthermore, the screening process of selecting children for comprehensive assessment in some studies may have biased the reported prevalence.

In our study, exposure to early life trauma was highly prevalent in children with and without a FAS/pFAS diagnosis. Rates of chronic suppurative otitis media and hearing loss were high, and hearing loss may affect learning and behavioural outcomes in school-aged children.[24] However, rates of hearing loss were similar between children with and without FAS/pFAS. In this study, clinicians were informed of hearing and auditory processing problems prior to assessments so accommodations could be made.

A significant strength of this study is that it was initiated and led by the Aboriginal community. High participation rates in a population-based cohort in a defined geographical area, active case ascertainment and objective quantification of alcohol use in pregnancy, including use of locally developed pictorial aids, were additional strengths.[17] Diagnoses were assigned using criteria from the Canadian guidelines following minor modification to CNS functional domains. Modifications were agreed upon after extensive consultation with international and national experts. Best practice for FASD diagnosis includes interdisciplinary assessment using standardised tests that are minimally biased by culture and language.[1,14,20] Clinicians used non-verbal and locally adapted tests when appropriate, and Aboriginal members of the research team were present to facilitate communication with participants.[17] Formal testing affirmed the reliability of selected questionnaires and assessments within the study population.[17,25] Feedback of results to participants, and opportunistic treatment, support and referral of children and families was a priority for the research team. Furthermore, training in FASD management for local education and health staff was provided as a means of developing local capacity.

Study limitations include the small sample size: this study was conducted in very remote communities and involved extremely complex logistic, cultural and climatic challenges and considerable expense. Another limitation is the absence of standardised norms for facial features in Aboriginal children. African American guides were used for lip and philtrum assessment.[20] One or more FAS facial features were documented in some children who did not meet full diagnostic criteria for FAS/pFAS. This is consistent with large international studies in which many non-alcohol-exposed children were found to have smooth philtrum (23%) or thin upper lip (28%).[8] Our findings may reflect the absence of normative data for facial features in Aboriginal children. However, many of these children were exposed to high levels of alcohol prenatally and may exhibit one or more facial features without fulfilling the diagnostic criteria for FAS/pFAS. Use of the Hall PFL charts[19] was agreed upon when the study protocol was developed, before more conservative norms for Caucasian children became available.[26] It is well documented that African American PFL is on average 1.5 mm greater than Caucasian PFL,[27] and it is likely that Australian Aboriginal PFL norms would approximate those of African Americans. Therefore, in the absence of PFL norms for Australian Aboriginal children, we believe the Hall charts to be appropriate. For CNS domains, existing norms were used for standardised tests, as collection of population-specific normative data was not feasible. These limitations are common to many of the published prevalence studies and highlight the need for collection of population-specific facial feature norms for Aboriginal children.

These are the first population-based prevalence data for FAS/pFAS from Australia. This study adds to mounting evidence that alcohol use in pregnancy and FAS/pFAS constitute a health crisis in communities with high rates of alcohol consumption. Understanding the reasons why women drink in pregnancy can inform prevention. Evidence suggests that maternal risk factors for FASD include low socio-economic status, rural residence, low educational attainment, being a single mother, having a male partner who drinks and living in a community tolerant of heavy drinking.[28] In addition to addressing these issues, FASD prevention programmes, adequately resourced mental health, drug and alcohol services and support for pregnant women and families are urgently needed.

Enhanced capacity to diagnose, manage and prevent FASD is a priority throughout Australia. Community-led restrictions on access to alcohol should be supported. This study demonstrates that research partnerships may provide immediate and enduring benefits to participants and the broader community. Echoing this sentiment, Mick Gooda, Australia's Aboriginal and Torres Strait Islander Social Justice Commissioner, said: '*This work shows what is possible when control of an issue is taken at the community level by strong Aboriginal women . . . It shows what is possible when Aboriginal leaders engage as equals with researchers . . . This work is an example of those researchers reciprocating both the spirit and intent of the community by working to address the challenges of FASD in a genuine partnership – one where research is done with the community, not just about the community*'.[21]

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