Burden of fetal alcohol syndrome in a rural West Coast area of South Africa

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Background. Fetal alcohol syndrome (FAS) is common in parts of South Africa; rural residence is a frequently cited risk factor. We conducted a FAS school prevalence survey of an isolated rural community in a West Coast village of Western Cape Province, so obtaining the first directly measured rate, focusing specifically on a South African rural area, of FAS and partial FAS (PFAS).
Methods. The study area (Aurora village), a community of about 2 500 people in a grain-producing region, has one primary school. All learners were eligible for study inclusion. Initial anthropometry screening was followed by a diagnostic stage entailing examination by a dysmorphologist for features of FAS, neurodevelopmental assessment, and an interview assessing maternal alcohol consumption.
Results. Of 160 learners screened, 78 (49%) were screen-positive, of whom 63 (81%) were clinically assessed for FAS. The overall FAS/PFAS rate among the screened learners was 17.5% (95% confidence interval 12.0 - 24.2%), with 16 (10.0%) children having FAS and 12 (7.5%) PFAS. High rates of stunting, underweight and microcephaly were noted in all learners, especially those with FAS or PFAS. Five (18%) mothers of affected children were deceased by the time of assessment.
Conclusion. We describe very high rates of FAS/PFAS in an isolated rural part of the Western Cape that is not located in a viticultural region. Our study suggests that the prevalence of FAS may be very high in isolated communities, or in particular hot-spots. It adds to the growing evidence that FAS/PFAS is a significant, and underestimated, health problem in South Africa. Expanded screening and surveillance programmes, and preventive interventions, are urgently needed.

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Fetal alcohol syndrome (FAS) and the less severe fetal consequences of maternal drinking, collectively described as fetal alcohol spectrum disorders (FASD) than FAS/partial FAS (PFAS), are endemic in impoverished communities of the Western and the Northern Cape provinces of South Africa (SA).[1-4] Over the past decade, levels of FAS/PFAS ranging between 40.5 and 119.4 per 1 000 have been documented in several parts of these provinces.[1,4] A recent study examined the prevalence of FASD (which is a broader and less well defined group) in an area of the Western Cape, and estimated the prevalence at between 135.1 and 207.5 per 1 000.[5]

Heavy alcohol intake during pregnancy, usually in a binge-drinking pattern, occurs in over 20% of women in the Western Cape.[6] Drinking during pregnancy remains a problem despite considerable media attention, efforts by the national government to limit alcohol harm,[7] and isolated initiatives to increase universal and other prevention activities in these provinces.[8] Heavy drinking has been socially entrenched and 'normalised' through a history including, but not restricted to, the dop system. For many women, the underlying social determinants of heavy alcohol consumption remain unchanged, and include poor socio-economic conditions, single-parent families, low levels of maternal education, concomitant use of tobacco and other substances, low religiosity and lack of alternative recreational opportunities.[6,9]

Most surveys have been conducted in small towns, and have consistently shown that FAS/PFAS is more common where mothers were resident in a rural area at the time of pregnancy.[1-3] To date, no studies have directly documented FAS/PFAS rates in entirely rural or remote parts of the country. In 2008, community leaders of a rural area in the West Coast region of the Western Cape invited our research team to assess levels of FAS/PFAS, as well as substance use, in pregnancy. The study therefore aimed to document the prevalence and risk factors for FAS/PFAS in this isolated rural community, and to provide information to assist the community to advocate for increased resources to counter substance use and related problems in the area.

Methods

The study site is a rural village (Aurora) of approximately 2 500 inhabitants. They work mostly as seasonal labourers at surrounding potato and grain farms. There is one school in the area (grades 0 - 7), at which the parents/guardians of all learners were invited to enrol in the study. Workshops were held with approximately 220 people to generate awareness of the study, with involvement of community and local government leaders, including social workers, police officers, health workers and educators from the school. The school principal and educators also attended a course on how to manage children with FASD in the classroom.

FAS diagnostic process

Prevalence of FAS and PFAS was determined by active case ascertainment using a validated two-tier screening method.[1] This entails an initial screening stage, followed by a comprehensive diagnostic stage (clinical evaluation, neurocognitive assessment and maternal interview). In the first stage, pupils were screened by a professional nurse who measured height, weight and head circumference (HC), using standard methods. Children ≤10th

percentile of the National Center for Health Statistics charts for height and weight, or ≤10th percentile for HC, were considered screen-positive and invited to the second stage.

The diagnostic stage entailed assessment by an experienced dysmorphologist (DV) for clinical features of FAS.[10] The clinical diagnosis of FAS/PFAS was substantiated by a battery of neurodevelopmental tests. Positive evidence of neurological abnormality required a HC ≤10th percentile, the presence of 'hard' signs on neurological examination, or significant abnormalities on neurocognitive assessment (below average in more than 4 of 10 scales) using primarily the Griffiths Mental Development Scale.[11]

Trained interviewers completed a semi-structured questionnaire with the mother/guardian of screen-positive children. Interviews were in the home language of participants and focused on drinking habits during the index pregnancy, which were elicited using a timeline follow-back method.[12]

The final diagnosis, based on criteria published by Hoyme *et al.* (2005),[10] encompassed all data from the clinical evaluation, the neurodevelopmental profile and maternal history. Final diagnostic categories were FAS and PFAS. The clinical diagnosis of FAS, and with less certainty PFAS, is considered distinctive even in the absence of a history of maternal alcohol use.

Study ethics and data management

Permission was obtained from the Department of Education (provincial and district) to evaluate learners and informed consent was given by the parents or legal guardians of each child. If consent was given but the child was not present for clinical or neurodevelopmental assessment, 2 further attempts were made to assess the child. Similarly, 3 attempts were made to secure maternal interviews.

A trained counsellor communicated the FASD diagnosis to the parents or guardians, as well as its implications and a proposed intervention tailored to each child. Children identified as having FAS/PFAS were referred to local government services, including speech and hearing therapists, occupational therapists, physiotherapists and social workers. With parental permission, school personnel were informed of each child's FASD status to facilitate educational support and further remedial assessment. Children with other medical conditions were referred to local and regional medical services, as required.

Intercooled Stata 12.1 (Stata Corporation, College Station, USA) was used for statistical analysis. For analysis of categorical variables, the chi-square test was used, while for continuous variables we used an unpaired Student's *t*-test or Mann-Whitney U-test for normally and non-normally distributed data, respectively.

Results

Consent for participation in the study was sought for all 171 learners in the school, of whom 160 consenting learners were available for anthropometric screening. Of the 160 screened learners, 78 (49%) were screen-positive on anthropometry, with 60 (38%) falling below the 10th percentile for both height and weight. Status regarding FAS/PFAS diagnosis was determined for 63 children (81% of screen-positives) (Table 1). Fifteen learners could not be clinically assessed because they were absent from school on all follow-up visits made to assess them.

The screened group, who were all of coloured ethnicity, were aged 4.8 - 16.4 years. About 9% (14/160) of learners were older than 14 years. Exactly half the pupils were male, though females predominated in grades 0 and 1 (62%, 32/52; p=0.04).

In the group of 160 screened learners, 16 FAS and 6 PFAS cases (13.8%; 95% CI 8.8 - 20.0) were ascertained based on full clinical assessment Mean dysmorphology score for the 16 confirmed FAS children was 17.1 (SD)±2.2, and 9.1±4.5 for children with PFAS. A further 6 were suspected of having PFAS, based on facial features

Table 1. Characteristics of grade 0 - 7 learners with fetal alcohol syndrome

Variable	All grade 0 - 7 learners (*N*=160)	Children without FAS/PFAS (*n*=132)	Children with FAS/PFAS (*n*=28)	*p*-value
Sex, *n*/*N* (%)				
Female	80/160 (50.0)	67/132 (50.8)	13/28 (46.4)	
Male	80/160 (50.0)	65/132 (49.2)	15/28 (53.6)	0.68
Age (years), *n*/*N* (%)				
4.8 - 8	59/160 (37.8)	47/132 (35.6)	12/28 (42.9)	
9 - 11	53/160 (33.6)	45/132 (34.1)	8/28 (28.6)	
12 - 16	48/160 (28.6)	40/132 (30.3)	8/28 (28.6)	0.75
School grade, *n*/*N* (%)				
0 - 1	52/160 (32.5)	40/132 (30.3)	12/28 (42.9)	
2 - 4	53/160 (33.1)	44/132 (33.3)	9/28 (32.1)	
5 - 7	55/160 (34.4)	48/132 (36.4)	7/28 (25.0)	0.37
Weight for age				
Median *Z*-score (IQR)	-1.2 (-2.1 to -0.5)	-1.0 (-1.7 to -0.4)	-2.4 (-2.9 to -1.9)	<0.001
Height for age				
Median *Z*-score (IQR)	-1.2 (-1.9 to -0.7)	-1.0 (-1.8 to -0.5)	-2.1 (-2.5 to -1.5)	<0.001
BMI for age				
Median *Z*-score (IQR)	-0.7 (-1.4 to -0.1)	-0.6 (-1.3 to 0.1)	-1.4 (-2.1 to -0.7)	0.001
Head circumference				
3rd - 10th percentile, *n*/*N* (%)	20/160 (12.6)	13/131 (9.9)	7/28 (25.0)	
≤3rd percentile, *n*/*N* (%)	30/160 (18.9)	13/131 (14.0)	17/28 (60.7)	<0.001

IQR = interquartile range; BMI = body mass index; FAS = fetal alcohol syndrom; PFAS = partial fetal alcohol syndrome

of FAS, HC <10th percentile, and a history of maternal alcohol use, but did not have a neurodevelopmental assessment. Despite the lack of this assessment, they had sufficient features to meet the Hoyme criteria for PFAS or FAS.[9] They were assigned as PFAS cases, and included as such for further analysis, bringing the total to 28 affected children (17.5%; 95% CI 12.0 - 24.2).

No associations were detected between having FAS/PFAS and gender or grade. Children with FAS/PFAS were a median 8.5 years old, while other children were a median 10.0 years (p=0.09). The median body mass index of children without FAS/PFAS was 15.8 kg/m^2 (IQR 14.6 - 17.5), compared with a median 14.7 kg/m^2 in children with FAS/PFAS (IQR 13.7 - 15.6; p=0.001). The entire cohort of children, and especially those with FAS/PFAS, showed very high levels of stunting and were underweight for age (Table 1).

Five (18%) of the mothers of the 28 subjects with FAS/PFAS had died, and in 2 instances the father was also known to have died. The study team referred 20 children to the school psychologist.

Discussion

Researchers have previously noted that levels of FAS/PFAS in parts of the Western and Northern Cape provinces are several times higher than elsewhere in the world.[3] Our data represent the first directly measured FAS/PFAS prevalence data from an entirely rural community in SA, and demonstrate levels that are high even when compared with similar local studies.

Table 2 shows FAS rates across published school prevalence studies in SA. Although direct comparisons between studies can be misleading, all these school surveys used similar methodologies, except that ours is the only study to include learners beyond first grade (there was, however, no significant difference in the prevalence of FAS across school grades).

Though the FAS rate of 10% in Aurora is very high, the true scale of overall FASD is certainly considerably higher. A further 7.5% of children were assessed as having FAS/PFAS, although this assessment was complicated by some participants not completing all study procedures, owing mostly to truancy from school (teachers suspected a high FASD rate among truant children). In addition to study-specific issues, it is likely that FASD prevalence studies inherently underestimate its prevalence because of the difficulties in diagnosing subjects without typical facial features, and in older children and adolescents as children age.[13] In this regard, the high rate of non-syndromic microcephaly found in 'unaffected' children may relate, in part, to sub-clinical cases of FASD.

The high prevalence of FAS/PFAS in the village of Aurora is consistent with: (*i*) the rural-urban gradient in FAS prevalence shown in Table 2, (*ii*) studies that show a higher FAS/PFAS rate in children born in rural than urban areas,[1-4] and (*iii*) evidence that binge drinking is more prevalent in rural (20%) than urban informal (16%) and urban formal (15%) areas.[15] FASD also appears more prevalent in very isolated communities – the communities with the highest published prevalence of FAS/PFAS (Aurora and De Aar) are very isolated geographically. Conversely, high rates do not appear to relate to the presence of viticulture in an area.

Despite mounting evidence from school surveys and other sources that FASD is a significant public health problem, it remains under-recognised. For example, the Western Cape Burden of Disease study considered FASD only inasmuch as it is a cause of low birthweight, rather than as a significant cause of morbidity, mortality and disability in its own right.[16] In part because it is under-recognised, there is a dearth of local measures aimed at preventing FASD, despite international acceptance of the utility of a range of interventions to reduce alcohol-related harm, ranging from policy and community level approaches to setting-specific approaches such as 'brief interventions' in prenatal care.[17] As a local example of a successful intervention, we have reported a 30% reduction in FAS/PFAS rates associated with a community-level intervention comprising intensive 'universal prevention' measures.[8]

Interventions within the health sector that should be implemented, upgraded or urgently investigated include: (*i*) strengthened family planning services; (*ii*) improved antenatal education regarding the risks of drinking; (*iii*) identification of risky drinking in pregnancy and implementation of brief interventions to reduce it; (*iv*) strategies aimed at early identification of FASD to allow early intervention in the child and prevention of recurrence in future children; and (*v*) national-level surveillance of risky drinking and FASD rates to improve data on the problem of FASD.

Table 2 indicates that, although there is a high level of FAS in many parts of SA, the prevalence varies. A systematic approach is needed for determining overall FASD levels in the country that is both wide in scope but also provides local actionable detail in higher-risk areas. We recommend regular surveillance of a systematic sample of schools and/or antenatal clinics across the country, which would allow provincial and national baseline rates to be established and monitored over time, and would inform more intensive and cost-effective targeting of communities at highest risk.

We found that nearly 1 in 5 of mothers to children with FAS/PFAS were deceased. This phenomenon has not been studied in detail to date, although it is noteworthy that the 3 Wellington studies[1-3] reported significant maternal mortality in mothers of FAS/PFAS children. Collectively, they report 13 (7%) maternal deaths among 185 mothers of children with FASD by the time the index cases were assessed at about 7 years of age. Causes of death, where known, appeared to be potentially associated with alcohol use, including accidents such as house fires; homicide and

Table 2. FAS prevalence rates from studies among schoolchildren in diverse South African communities

Site	Site description	Viticultural area (Y/N)	FAS cases*/sample size† (cases per 1 000)
Gauteng[13]	Metropole	N	16/830 (19.3)
Wellington, Western Cape[1]	Town	Y	46/992 (46.5)
Wellington, Western Cape[2]	Town	Y	64/863 (74.2)
Wellington, Western Cape[3]	Town	Y	55/818 (67.2)
Upington, Northern Cape[4]	Town	Y	69/1 299 (53.1)
De Aar, Northern Cape[4]	Isolated town	N	54/536 (100.7)
Aurora, Western Cape	Isolated village	N	16/160 (100.0)

†To maximise comparability, data are restricted to fully assessed FAS cases divided by the number actually screened.
*Excludes PFAS cases.

other violent death; pulmonary tuberculosis; and liver disorders. These data are consistent with evidence that binge drinking, in addition to being associated with FASD, is also strongly associated with unintentional injury, interpersonal violence, unsafe sex and other negative health consequences for adults.[19,20] A case of FASD should therefore be considered as a marker for adverse maternal health consequences.

This study aims to benefit the local population, NGOs and community-based organisations, as well as inform government departments, specifically Health, Education, Social Services and Agriculture. The study heightened the community's awareness of substance abuse problems and FASD in particular. Although this district has not featured on the government's priority list for high-risk substance abuse areas and/or other health needs to date, the community specifically requested that the study findings be publicised and used to rectify this omission. Findings will also inform development of a comprehensive NGO-led intervention to address these local problems.

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