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Characteristics of Mothers of Children with Fetal Alcohol Syndrome in the Western Cape Province of South Africa: A Case Control Study*

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ABSTRACT. *Objective*: Factors associated with alcohol consumption during pregnancy and with fetal alcohol syndrome (FAS) births were examined as part of a larger epidemiologic study of FAS in a community in the Western Cape Province of South Africa. *Method*: Using retrospective case-control methodology, 31 mothers who had given birth to FAS children 6 to 9 years previously were compared with 31 matched controls on a variety of demographic, socioeconomic, drinking, family and maternity variables. Descriptive analyses were utilized to determine major differential characteristics between the two groups. *Results*: In this community with a very high rate of FAS and rather uniform low socioeconomic status, the two groups were found to be comparable with respect to age, annual income, ethnic background, age of initiation of regular drinking, age at birth of the index child, gravidity and parity. However, mothers of FAS children reported initiating drinking at an earlier age, as well as reporting higher rates of heavy alcohol consumption in their extended family, current use of alcohol, drinking before and during pregnancy, and smoking of tobacco (percentage who smoke) during each trimester of the pregnancy. Mothers of FAS children had lower educational attainment and reported lower religiosity than control mothers. *Conclusions*: This study in South Africa draws upon the experience of mothers of 31 children with FAS to confirm many of the same high-risk variables identified in maternal risk studies in the United States and Europe. Some factors associated with less maternal alcohol abuse in this high-risk population were also identified, which may be helpful for implementing prevention in this region as well as in other developing countries. (*J. Stud. Alcohol* **63**: 6-17, 2002)

SINCE THE FIRST recognition and documentation of fetal alcohol syndrome (FAS) as a distinct birth defect (Jones and Smith, 1973), the search for unique maternal characteristics, risk factors and protective factors has been a goal of researchers, both for determining the specific etiology of FAS and for prevention. Epidemiologic studies of mothers who drink during pregnancy have identified traits, in addition to drinking alcohol heavily during pregnancy, that are strongly associated with FAS births. Women who were older, multigravidas, not currently married, and who smoked cigarettes and used other drugs were more likely to have children with FAS (Sokol et al., 1980). Later studies identified a greater risk for FAS among women characterized by advanced maternal age, high parity, low socioeconomic status (SES), African-American race and with severe drinking patterns, most particularly heavy episodic use (Abel, 1995; Abel and Hannigan, 1995; Abel and Sokol, 1986; Darrow et al., 1992; Sokol et al., 1986). Studies of mothers who had produced children with what was then diagnosed and referred to as "fetal alcohol effects" (FAE; a lesser degree and consistency of symptoms than the full syndrome) provided further understanding of maternal risk factors.

Alcohol, in a dose response effect, is likely to produce not only FAS, but such individual features as growth and developmental delay, neurobehavioral defects, microcephaly and craniofacial anomalies in children born to women who are older and of low socioeconomic status (Day et al., 1991, 1999; Ernhart et al., 1987; Jacobson et al., 1996). FAE is now referred to as alcohol-related birth defects (ARBD) and alcohol-related neurodevelopmental disorder (ARND) (Stratton et al., 1996). These lesser effects, when combined with the prevalence of FAS, are believed to affect approximately 1% of all births in the United States (Sampson, et al., 1997).

The influence of socioeconomic status on the characteristics and rates of FAS, ARBD and ARND has been a particularly common finding in various studies, and the association appears to hold even in one retrospective control study. Bingol et al. (1987b) compared two groups of alco-

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holic women of different social strata who drank an average of 6 ounces of absolute alcohol a day (12 drinks) and who had begun drinking during their teens. The rate of FAS and alcohol-related effects in this study was 16 times greater among women of low SES than among the upper middle-class sample (4.6% vs 70.9%). There was variance between the two groups in family histories of alcoholism, percentage of college graduates, percent married and other variables; however, the women's current SES was highly influential (Bingol et al., 1987b). Abel's (1995) summary of the worldwide epidemiology literature also demonstrates a consistently elevated rate of FAS births among African-American women of low SES, when coupled with drinking heavily.

In the United States and other developed countries, between 16% and 32% of women who are pregnant drink during gestation (Centers for Disease Control and Prevention, 1997; Day et al., 1993; Serdula et al., 1991). In some other western countries, maternal drinking is substantially higher, reaching over 50% (Godel et al., 1992; Waterson and Murray-Lyon, 1989). Obvious variations in this percentage of at-risk drinkers exist throughout the world, with subpopulations of high-risk drinkers contained within the overall category of female drinkers.

Studies of alcohol dependence and alcohol misuse have reported that these behaviors do run in families, therefore implying heritability or a genetic influence in susceptibility. Twin studies have estimated that the heritability for susceptibility to alcoholism in women is in the range of 50% to 60% (Kendler et al., 1992). Among men, those who came from families with a large number of alcoholics have been shown to have more alcohol misuse than are those from families with a low density of alcoholics (McCaul et al., 1991; Schuckit, 1998; Schuckit and Smith, 1996). Siblings with a history of alcohol-related illness and frequent drinking with family members were associated in one study with women who continued to drink throughout pregnancy (Smith et al., 1987). Epidemiologic studies of the occurrence of FAS have also shown familial clustering (Abel, 1988; May et al., 1983; Pierog et al., 1979). Families with FAS children, particularly those with multiple FAS cases, present a tremendous challenge both to clinicians and prevention programs (Davis and Lipson, 1984). Influences on maternal drinking are complex and originate from a combination of factors: biological, familial, social and psychological (Gomberg, 1993). Prevention of FAS is thus a multifaceted enterprise (May, 1996).

The general literature regarding alcohol misuse among women (i.e., that which does not specifically address prenatal drinking) contributes additional information for understanding maternal drinking and risk factors for FAS (Stratton et al., 1996). Women at high risk for alcohol-related problems and for continued drinking have some of the following traits: cohabitation with men when not married, part-time employment, frequent sexual dysfunction and living with others (usually male) who drink heavily (Wilsnack, 1989). Alcohol misuse is frequently found among young women (ages 21 to 34) who are the daughters of alcoholics (either mother or father), and who report regular drinking early in life (Wilsnack, 1991; Wilsnack and Beckman, 1984; Wilsnack et al., 1991). Furthermore, these women commonly use and misuse other drugs, including tobacco (Serdula et al., 1991; Sokol et al., 1986). Their social context is characterized by many alcohol-use or drug-use situations that are socially condoned, sanctioned and/or at least not disapproved of by significant others (Shore and Batt, 1991; Shore and Pieri, 1992). In addition, women who misuse alcohol report multiple social and psychological risk factors, particularly low self-efficacy, low purpose in life, depression and feelings of powerlessness (Baily, 1990; Schlesinger et al., 1990).

Whereas the above variables may imply relative helplessness in certain contexts, some authors have identified protective factors (e.g., cultural support and unique personality traits) that assist particular individuals in avoiding alcohol misuse (Blume, 1990). Further explication and clarification of protective factors are needed for evidence-based prevention and intervention initiatives (Hanna et al., 1993; Stratton et al., 1996; Jacobson et al., 1991; Schimidt et al., 1990), especially in unstudied and unique populations.

The study setting

This study attempted to identify specific risk factors for drinking heavily during pregnancy and for FAS, in a community in the Western Cape Province of South Africa. The population of the community was enumerated in 1996 as 45,225 (35,364 urban and 9,861 rural) (Republic of South Africa, 1997). The majority of the population is "Cape coloured" (mixed race), with less than 25% white and a few black Africans. The "coloured" population is polyethnic and polygenetic, with influences from several indigenous South African tribes and European and Asian admixture. This community is similar to others in grape-growing and wine-producing regions, in which problem-drinking practices and patterns have existed among the agriculture laborers for multiple generations. For many years, alcohol was supplied to workers on a daily basis as partial payment for labor, a system of payment referred to as the "Dop system." Although outlawed today, vestiges of the system still exist in patterns of frequent and severe episodic drinking (Crome and Glass, 2000). It is still apparent today that alcohol is a favored, valued and expected commodity among many of the local population of workers, who receive low pay and who live in very humble circumstances.

A high rate of problematic drinking behavior has been documented in the region. As measured by standard screening instruments (e.g., CAGE and MAST) and serum GGT

levels, alcohol misuse was found to affect 83% of the male fruit-farm workers (London et al., 1995). This was, by far, the highest rate in any South African population studied by that time (Parry and Bennetts, 1998). A more recent study classified 87% of the farm workers in the region as problem drinkers; half consume more alcohol per week than is considered safe (>210 g) and 9.3% consume amounts considered dangerous (>490 g per week) (London, 2000).

In this study community and others like it, a number of commercial sources of alcohol exist. Although alcohol may be consumed on a daily basis (through "Dop") on some farms, it is quite common for a substantial segment of the population to participate in regular and extended drinking parties on the weekends and, occasionally, on weekday evenings. Wine and beer are the beverages of choice and are relatively inexpensive. Drinking at "shebeens" (informal bars), in the home and in other venues is a major form of recreation for a substantial subculture of the population.

Drinking during pregnancy has been reported to be frequent in parts of the Western Cape, even among prenatal clinic patients. Croxford and Viljoen (1999) reported that 34.4% of prenatal patients in the large metropolitan areas of the Cape drank during pregnancy, as did 46.1% to 50.8% in the rural Cape regions (Croxford, 1998). Concern about FAS has grown in recent years in parts of South Africa (National Institute on Alcohol Abuse and Alcoholism, 1996, 1998).

The study of the characteristics of Western Cape mothers who have FAS children is useful for understanding their lifestyle, and maternal drinking in this region, among women in general and, more particularly, in other parts of the developing world. Since the current study was the first ever undertaken on the epidemiology of FAS in a South African community, no specific hypotheses were formulated.

Method

Participants

The data in this article are an integral part of a larger epidemiologic study of the prevalence and characteristics of FAS in the Western Cape (May et al., 2000). Subjects in this study of maternal characteristics were chosen based on the characteristics of their children. Children in Sub-A (first grade) classrooms (N = 992) received an initial brief (Tier I) screening for height, weight and head circumference. After local growth and development norms were established, those who were below the 10th percentile for head circumference and/or both height and weight were also given full dysmorphology evaluations by a team of international experts on FAS. Dysmorphologists working in blinded teams utilized established active-case ascertainment methodologies, including a quantified checklist for a preliminary diagnosis of FAS or not FAS. Fifty-three children were identified with a preliminary diagnosis of FAS or possible FAS. Each of these 53 children received developmental and life-skills testing (Griffiths Intelligence and Developmental Test; Griffiths, 1984), and their mothers were interviewed extensively on various risk factors (Adnams et al., 2001). After all screening, testing and information gathering were completed by the blinded, independent examiners from each domain (dysmorphology, developmental and maternal), a case conference of the examiners determined the final diagnosis. Institute of Medicine (IOM; Stratton et al., 1996) criteria for FAS were followed: evidence of pre- and postnatal growth retardation, dysmorphology consistent with prenatal alcohol exposure and evidence of developmental delay. Maternal drinking data were used as confirmatory as per IOM criteria, and only one child was diagnosed with FAS in its absence.

The protocols and consent forms used for human subjects were approved by the University of New Mexico Medical School (HHRC 96-209) and the College of Arts and Sciences (01-93-86-9908). The methodology was also reviewed and approved by the University of Cape Town Ethics Committee, the Office of Protection from Research Risks (OPRR) of the National Institutes of Health, and by a single site assurance committee in the Western Cape community. All participating mothers provided active consent for inclusion in the study, and the parents and guardians also consented to FAS screening for their child.

FAS, as diagnosed by strict IOM criteria, was the focus of the study, and not ARBD or ARND. In the first-grade population, 46 children received a final diagnosis of FAS, yielding a very high (world record) in-school FAS rate of 46 per 1,000 (May et al., 2000). Because of the high-risk lifestyle of the mothers of FAS children, only 35 were accessible for follow-up study. Reasons for the others' inaccessibility, and the effect of the reduced sample, are presented in the Results section and in Table 1.

The control group consisted of the mothers of the control children in the epidemiology study. One control child was selected from the same grade and school as each FAS child, matched for age (within 1 year), gender and rural urban residence. Of the 35 FAS children whose mothers were both alive and interviewed, we were able to match 31 to controls, exactly by gender, rural/urban school attendance and age (mean [SD] difference = 3.8 [3.5] months). Because of age differences (some FAS children were older than other first graders), perfect matches were not possible for four FAS children. Four mothers of FAS children and 20 controls were excluded as a result of the strict case-control matching process used in this study.

Most studies of maternal risk factors have studied women who drink alcohol during pregnancy; very few of these have had or will have children with FAS. This article focuses on the characteristics of mothers who have had FAS children (i.e., the highest risk mothers). Some researchers have re-

ported that a previous child with FAS is the single best indicator of maternal risk (Abel, 1988; May, 1995).

Measures

The interview questions utilized in this study were drawn from questionnaires first developed among populations in the southwestern United States. They were adapted specifically for the South-African population and pilot tested in the target community. There are 114 items covering such basic demographics as residence; religion, and ethnicity; social and economic variables; family health; diet; and quantity, frequency, variability and social context of drinking. Questions focused on reconstructing the general characteristics of the women's lives, including history of drinking during the pregnancy of the index child. To establish rapport, nonthreatening questions were asked first, regarding where they were born, grew up, worked, their parents' lives and deaths, education, diet, child bearing and general health. Alcohol consumption responses have been found to be more accurate when included in such a format, especially in the context of dietary questions (King, 1994).

In the alcohol section of the questionnaire, respondents were first asked about the drinking habits of their parents, siblings and friends. The context and details of the mother's current drinking then were explored orally via a 1-week, day-by-day log methodology. Drinks were measured in standard ethanol units, in which one unit was a 340 ml can of beer (10.2 oz of 5% ethanol), 120 ml of wine (4 oz of 11.5% ethanol) or 44 ml of distilled spirits (1.5 oz of 86 proof). Respondents were shown pictures of standardized containers utilizing local brands and vessels familiar to them. Questions on current drinking were then used as benchmarks, for reconstructing drinking patterns during the period of gestation of the index child. This order refreshed the respondents' recall of drinking and life context before moving to sensitive and distant questions about life and pregnancy some years earlier.

Smoking was measured more directly. Local informants indicated that smoking purchases and practices were less stigmatized, more regular, more stable and memorable due to cost, lack of sharing of individual cigarettes and the need to pause from the daily routine to "roll" cigarettes. Respondents were asked how much tobacco (for hand-rolled) or packaged cigarettes they usually bought in a week (most were paid and went to town once a week) and smoked each day, currently and during the index pregnancy. Questions on other drugs utilized both open-ended and fixed-response items to assess the quantity, frequency and type of drugs used.

Interviews were performed in Afrikaans, the primary language of the area. All of the respondents, both mothers of FAS children and controls, were South African "coloured."

The analyses utilize comparisons of the two groups. Statistical tests (two-tailed) are used to assess significance (at <0.05) and, where appropriate, odds ratios (OR) are provided.

Results

Of the 46 mothers of FAS children in the larger epidemiology study, 35 were interviewed (May et al., 2000). Six (13%) of the mothers were dead at the time of the study: four from violent death, one from a house fire and one from pulmonary tuberculosis. Five (10.9%) were so nomadic that either repeated attempts to contact them were unsuccessful, or they had permanently moved from the area. Thus, 11 mothers could not be reached for participation in the maternal characteristics study. In Table 1, the effect of missing mothers from the FAS sample is described via data from the FAS children in the study. There are only two significant differences, overall, between the FAS children of the interviewed mothers and the FAS offspring of mothers not available. The children of the missing mothers had significantly lower performance IQ scores and higher mean dysmorphology scores (more physical anomalies consistent with prenatal drinking). However, all of the nonsignificant

TABLE 1. Physical and mental development of FAS children by mother's availability for interview

	FAS children		
Growth and development, mean (SD)	Mothers interviewed ($n = 35$)	Mothers not available for interview ($n = 11$)	Test, significance
Height percentile	5.2 (4.15)	3.0 (1.41)	$t = 1.695$, $p = .097^a$
Weight percentile	11.4 (14.03)	3.8 (2.32)	$t = 1.760$, $p = .085^a$
Occipitofrontal circumference (%)	9.2 (17.45)	7.1 (8.11)	$t = 0.391$, $p = .697^a$
Mental age score	78.8 (13.39)	73.7 (13.11)	$t = 1.101$, $p = .277^a$
Performance IQ score	76.0 (10.79)	68.3 (10.53)	$t = 2.084$, $p = .043$
Verbal IQ score	72.4 (12.42)	69.3 (12.82)	$t = 7.300$, $p = .469^a$
Dysmorphology score[b]	9.9 (4.25)	13.0 (4.52)	$t = 2.060$, $p = .044$

[a]Not significant. [b]The higher the dysmorphology score the higher the frequency and severity of structural anomalies.

TABLE 2. Demographic, socioeconomic and maternity variables for mothers of children with FAS (n = 31) and control mothers (n = 31)

	Mothers of children with FAS (SD)	Control mothers (SD)	Test,significance
Demographic and socioeconomic			
Age on day of interview (mean)	34.3 (7.7)	33.8 (7.2)	$t = 0.259, p = .797^a$
Residence (% rural)	64.5	64.5	$\chi^2 = 0.00$, 1 df, $p = 1.000^a$
Educational attainment (years)	4.4 (3.2)	6.1 (2.6)	$t = 2.269, p = .027$
Religion (% practicing)	80.6	87.1	$\chi^2 = 0.48$, 1 df, $p = .489$, OR = 1.62^a
Religiosity (church attendance or prayer)(%)			
Never or not very often	54.8	41.9	
Often	38.7	16.1	$\chi^2 = 4.239$, 1 df, $p = .040^b$
Very often	6.5	41.9	$t = 2.402, p = .019$
Usual occupation (%)			
Factory worker	16.7	13.8	
Farm worker	50.0	34.5	
Office worker or other (i.e., cook, etc.)	33.3	51.7	$\chi^2 = 2.09$, 2 df, $p = .351^a$
Employment status (%)			
Full-time	38.7	41.9	
Part-time	6.5	12.9	
Seasonal	29.0	16.1	
Unemployed	16.1	19.4	
Not employed/not looking for work	9.7	9.7	$\chi^2 = 1.94$, 4 df, $p = .747^{a\,b}$
Income (weekly in Rand)(mean)	99.8 (91.5)	135.5 (158.3)	$t = 1.083, p = .283^a$
Maternity			
Gravidity (mean)	3.9 (1.7)	3.1 (1.4)	$t = 1.953, p = .055^a$
Parity (pre- and full-term)(mean)	3.5 (1.5)	2.9 (1.2)	$t = 1.788, p = .078^a$
Miscarriages (mean)	0.5 (0.9)	0.3 (0.4)	$t = 1.085, p = .282^a$
Living children (mean)	3.3 (1.3)	2.8 (1.2)	$t = 1.527, p = .132^a$
Age at birth of the index child (mean)	26.7 (7.6)	26.3 (7.2)	$t = 0.233, p = .816^a$
Birth order of index child (mean)	2.9 (1.7)	2.3 (1.4)	$t = 1.479, p = .145^a$
Marital status during pregnancy with index child (%)			
Married	12.9	25.8	
Single	29.0	35.5	
Unmarried, living with partner	58.1	38.7	$\chi^2 = 2.73$, 2 df, $p = .255^{a\,b}$

[a]Not significant. [b]Cell(s) <5 detract(s) from significance.

differences also document greater severity of FAS traits in the children of the missing mothers. Therefore, although the entire sample of 35 FAS mothers contacted and interviewed (none refused) was very high risk, they may not represent all of the very highest risk mothers of the FAS children. The results that follow present a comparison only of the two groups of the 31 matched cases and controls.

The mothers of FAS children and controls in this sample (Table 2) did not differ significantly by age, current rural/urban residence, profession of a religion, usual occupation, current employment status or weekly income, although the indicators were generally poorer for the FAS mothers. This provides validity to matching via the case control design. By South African standards, they were of relatively similar background and SES. By U.S. standards, respondents had a 1997 weekly income of $22 to $30; all, therefore, had very humble economic resources. Two variables in this category were significant: educational attainment and religiosity. Mothers of FAS children had significantly less formal education (1.7 years [28%] less), and were less likely to practice regular religious behaviors (measured by church attendance and prayer). Therefore, in spite of the two groups being relatively equal in SES, two specific social risk factors were identified in this category of SES and demographic variables.

Table 2 also highlights the maternity variables for the two groups. FAS mothers did not differ from controls on any of the standard measures commonly used in studies of this type: gravidity, parity, miscarriage, number of living children, age at birth of index child, birth order of index child or marital status during pregnancy of index child. All of the differences between the two groups, however, are in the direction predicted by the extant literature (e.g., higher gravidity, parity, miscarriage, etc. for FAS mothers). If one-tailed test criteria were used for gravidity and parity, they would be significantly different between groups. The lack of significant difference in maternal age is addressed in the Discussion section.

Details of the drinking habits of the family and friends reported by the maternal informants are presented in Table

TABLE 3. Reported drinking habits of family and friends of mothers of children with FAS (n = 31) and control mothers (n = 31)

Measures of alcohol consumption	Mothers of children with FAS	Control mothers	Test, significance
Drinking habits of woman's father (%)			
Nondrinker or light drinker	27.6	22.2	
Occasional or moderate drinker	6.9	25.9	
Frequent or heavy drinker	48.3	33.3	χ^2 = 4.08, 3 df, p = .253[a,b]
Has had problems with alcohol	17.2	18.5	χ^2 = 1.08, 1 df, p = .298, OR = 1.76[a,c]
Drinking habits of woman's mother (%)			
Nondrinker or light drinker	51.6	77.4	
Occasional or moderate drinker	12.9	6.5	
Frequent or heavy drinker	25.8	9.7	χ^2 = 4.74, 3 df, p = .192[a,b]
Has had problems with alcohol	9.7	6.5	χ^2 = 3.03, 1 df, p = .082, OR = 2.86[a,c]
Drinking habits of woman's brothers (%)			
N	57	83	
Nondrinker or light drinker	35.1	43.4	
Occasional or moderate drinker	10.5	21.7	
Frequent or heavy drinker	43.9	24.1	χ^2 = 7.14, 3 df, p = .067[a]
Has had problems with alcohol	10.5	10.8	χ^2 = 5.22, 1 df, p = .022, OR = 2.22[c]
Drinking habits of woman's sisters (%)			
N	70	86	
Nondrinker or light drinker	57.1	72.1	
Occasional or moderate drinker	5.7	19.8	
Frequent or heavy drinker	30.0	4.6	χ^2 = 23.46, 3 df, p = .000[b]
Has had problems with alcohol	7.1	3.5	χ^2 = 19.46, 1 df, p = .000, OR = 6.67[c]
Drinking habits of father of index child during index pregnancy (%)			
Nondrinker or light drinker	6.5	37.9	
Occasional or moderate drinker	3.2	20.7	
Frequent or heavy drinker	77.4	34.5	χ^2 = 16.08, 3 df, p = .001[b]
Has had problems with alcohol	12.9	6.9	χ^2 = 16.15, 1 df, p = .000, OR = 13.22[c]
Drinking habits of woman's best friend (%)			
Does not have a best friend	38.7	19.4	
Nondrinker or light drinker	29.0	71.0	
Occasional or moderate drinker	12.9	6.5	
Frequent or heavy drinker	19.4	3.2	χ^2 = 11.69, 3 df, p = .009[b]

[a]Not significant. [b]Cell(s) < 5 detract(s) from significance. [c]χ^2 calculated from data collapsed to 2 × 2 table to maximize possibility of significance. The two categories of nondrinker or light drinker and occasional or moderate drinker were collapsed together and compared to the other two categories that were also collapsed together.

3. Many controls and mothers of FAS children reported fathers and mothers who drank heavily, but more of the maternal grandmothers of FAS children were heavy drinkers, as indicated in Table 3. This comparison approached significance when items were aggregated into a 2 × 2 configuration by collapsing and comparing the two lowest-drinking categories (nondrinkers through moderate drinkers) with the two problem-drinking categories (p = .08; odds ratio [OR] = 2.86). All the other variables are significantly different. Three were significant when tested in either the 4 × 2 or 2 × 2 configuration described above. The heavy-drinking pattern of the fathers of FAS children (p = .000; OR = 13.22) is worth noting. Mothers of FAS children more frequently had brothers, sisters, male partners and friends who drank heavily. The mothers of FAS children live in an environment full of heavy drinkers. Yet social isolation is also an issue, for the last variable in Table 3 indicates that FAS women reported not having a "best friend" more frequently than did controls; and when they do have friends, those friends are more likely to be heavy drinkers (p = .009).

The only two substances used by women in this Western Cape region were tobacco and alcohol; virtually none of the respondents reported use of any other drugs currently or during pregnancy. The two groups did differ significantly (Table 4) in the mean age at which they first drank alcohol (p < .05), but not in the age at which they commenced regular drinking. All other variables were significant. Current use of alcohol, use prior to the pregnancy of the index child and use during all trimesters of the pregnancy were reported to be higher for the mothers of FAS children. Virtually all of the mothers of FAS children (81%) are current drinkers, compared with 45% of controls. Beer and wine constitute the vast majority of all alcoholic beverages consumed (not shown). When all respondents are considered, the average FAS mother currently drinks a mean (SD) of 13.0 (12.6) standard drinks per week compared to 2.8 (5.3) for the control sample. When current abstainers are eliminated from the analysis (six of the mothers of FAS children have quit), the mean drinks per week is 16.1 (12.1) for FAS mothers and 6.3 (6,4) for controls. Heavy drinking (5+ standard drinks per day) is by far the most prevalent

TABLE 4. Drinking and smoking behaviors of mothers of children with FAS (n = 31) and control mothers (n = 31)

	Mothers of children with FAS (SD)		Control mothers (SD)		Test, significance
Drinking variables					
Age first drank alcohol (mean)	18.3 (3.3)		20.9 (6.2)		t = 2.025, p = .047
Age began drinking regularly (mean)	20.5 (4.7)		22.9 (6.9)		t = 1.425, p = .161[a]
Current drinker (%)	80.6		45.2		χ^2 = 8.36, 1 df, p = .004
	Whole sample (n = 31)	Drinkers only (n = 25)	Whole sample (n =31)	Drinkers only (n =14)	
Current use of alcohol (mean drinks per week)	13.0 (12.6)	16.1 (12.1)	2.8 (5.3)	6.3 (6.4)	Whole sample, t = 4.125, p = .000 Drinkers only, t = 2.804, p = .008
Current consumption on weekends					
Friday (mean)	3.4	4.2	0.6	1.3	
Saturday (mean)	7.2	8.9	1.9	4.3	
Sunday (mean)	2.0	2.5	0.2	0.4	
Total for weekends (mean)	12.6	15.6	2.7	6.0	Whole sample, t = 3.92, p = .000
Percentage on weekends	(97.7%)	(96.9%)	(51.3%)	(93.8%)	Drinkers only, t = 2.86, p = .007
Drinking in months before pregnancy with index child (%)					
Drank about the same (as current use)	19.4		48.4		
Drank less (than current use)	22.6		38.7		
Drank more (than current use)	58.1		12.9		χ^2 = 14.08, 2 df, p = .000[b]
Drinking during 1st trimester of pregnancy with index child (%)					
Drank about the same (as current use)	19.4		48.4		
Drank less (than current use)	19.4		38.7		
Drank more (than current use)	61.3		12.9		χ^2 = 15.64, 2 df, p = .000[b]
Drinking during 2nd trimester of pregnancy with index child (%)					
Drank about the same (as current use)	16.1		51.6		
Drank less (than current use)	22.6		38.7		
Drank more (than current use)	61.3		9.7		χ^2 = 18.71, 2 df, p = .000[b]
Drinking during 3rd trimester of pregnancy with index child (%)					
Drank about the same (as current use)	16.1		51.6		
Drank less (than current use)	29.0		41.9		
Drank more (than current use)	54.8		6.5		χ^2 = 18.33, 2 df, p = .000[b]
Smoking variables					
Current smoker (%)	83.9		48.4		χ^2 = 8.71, 1 df, p = .003
Smoked during index pregnancy (%)	83.9		45.2		χ^2 = 10.15, 1 df, p = .001, OR = 6.33
	Whole sample (n = 31)	Smokers only (n = 26)	Whole sample (n = 31)	Smokers only (n =15)	
Current use of tobacco (grams per week)(mean)	50.4 (44.6)	60.0 (42.2)	29.5 (43.9)	61.1 (45.6)	Whole sample, t = 1.851, p = .069 Smokers only, t = 0.073, p = .942[a]
Grams of tobacco used per week					
1st trimester (mean)	43.7 (37.9)	52.1 (35.6)	25.7 (39.0)	53.1 (41.2)	Whole sample, t = 1.843, p = .070 Smokers only, t = 0.083, p = .934[a]
2nd trimester (mean)	44.7 (38.3)	53.3 (35.9)	22.2 (33.9)	49.1 (35.2)	Whole sample, t = 2.451, p = .017 Smokers only, t = 0.358, p = .722[a]
3rd trimester (mean)	42.1 (38.1)	50.2 (36.3)	19.7 (32.6)	43.6 (36.5)	Whole sample, t = 2.482, p = .016 Smokers only, t = 0.540, p = .592[a]

[a]Not significant. [b]Cell(s) <5 detract(s) from significance.

pattern of consumption. These bouts are characteristic of heavy episodic drinking, as the mothers of FAS children and the drinking controls consume 97% and 94%, respectively, of all of their alcohol on the weekends. In open-ended comments, mothers of FAS children commonly characterized the index pregnancies as extremely stressful and particularly trying times in their lives, which they linked to heavier drinking. A source of stress cited frequently was a poor relationship with a heavy-drinking man. During the trimesters of the pregnancy, control mothers generally (87% to 94%) continued to drink the same amount of alcohol or less, compared with their current level. The majority (55% to 64%) of the mothers of FAS children, however, reported drinking the same amount or substantially more, during the index pregnancy. The average daily alcohol consumption of the mothers of FAS children was 2.3 standard drinks per day (i.e., 1.2 oz of absolute alcohol per day). It is of particular importance that the mothers of FAS children who

are currently drinking are consuming eight standard drinks (4 oz of absolute alcohol) on the typical drinking day. This is the most meaningful measure of drinking risk, in this population that drinks episodically.

Maternal smoking data (Table 4) reveal that mothers of FAS children were more likely than controls to be current smokers (84% vs 48%) and to have smoked during the index pregnancy (84% vs 45%). There is little change in percent smoking, over time, in either group. Also, the number of cigarettes smoked per week or daily did not differ significantly between the individuals in the two groups either currently or during any trimester of the index pregnancy. The current smoking levels are calculated for both the entire sample and for smokers only (Table 4). Among current smokers, mothers of FAS children consume 60 grams (1 gram = 1 hand-rolled cigarette in South Africa) of tobacco per week, and controls 61.1 grams. This translates to 8.6 cigarettes per day for FAS mothers and 8.7 per day for controls. Therefore, control mothers who smoke report smoking quantities similar to those of FAS mothers for both current and index pregnancy. Both groups report slightly lower levels of smoking during the index pregnancy, particularly in the last two trimesters of the control pregnancies. However, since the differences between the smoking quantity of smokers in the two groups is not significant, either currently or in any of the trimesters, smoking seems less important than alcohol as a teratogenic risk. Women of both groups smoke only a moderate amount per day (by U.S. standards). This is likely due to a lack of disposable income for purchase and because "rolling one's own" takes more time, concerted action and thought than using prerolled cigarettes. Whereas the *quantity* of cigarettes smoked seems to present neither a major differential nor overall influence on the smoking pregnancies across groups, smoking is twice as common among the mothers who have had FAS children.

Discussion

This study has examined South-African data on known maternal risk factors that have been described in other studies in the United States. The samples were drawn from an active case ascertainment study of FAS in a predominantly lower-SES population, from a developing region dominated by over 200 years of grape and wine production. The characteristics of maternal alcohol use described here, quite specific to the Western Cape population, may also have implications for other mixed-ancestry, low-SES groups in South Africa and elsewhere. This study, most importantly, has raised questions relevant to low SES and to developing areas, throughout the world, in which unique lifestyles and episodic drinking patterns combine to produce high blood alcohol concentrations in a substantial number of pregnant women.

The mothers of FAS children and controls were matched to represent similar cultural, racial and specific community backgrounds. The mothers of FAS children had lower educational attainment and less regular practice of religion, but were not significantly lower on other SES variables. An additional demographic risk factor that had been identified in our overall epidemiological study of this region was rural residence. This variable was controlled here, however, by case-control matching techniques. We reported previously (May et al., 2000) that FAS cases in this community were found more frequently in rural areas than in the overall population. Instead of 26% of the FAS cases originating in the rural areas as predicted by the population distribution, 61% were from rural environs ($\chi^2 = 8.96$, $p = .003$; OR = 14.8). Because of the residence matching in this study, the rural/urban variable is controlled, leaving educational attainment and religiosity as the major social differences.

The mothers of FAS children were not found to be of significantly higher gravidity or parity, or less likely to be married at the conception and birth of the index child; this is unlike the results of U.S. studies. Small sample size may be partly responsible for these results; however, it should also be noted that marriage rates were low in the control groups as well. As in other studies (Wilsnack et al., 1991), there is some tendency for the FAS mothers to be cohabitating while single.

The mothers of FAS children were from families with more extensive histories of heavy drinking and alcohol abuse than families of controls (Pascoe et al., 1995; Smith et al., 1987). This difference was more prominent among maternal grandmothers than maternal grandfathers, and more significant among sisters than brothers. Furthermore, as in the U.S. literature, the partners of women who had FAS children were more likely to drink heavily. The current and past drinking patterns of the mothers of FAS children were confirmed to be significantly heavier than those of the controls for all data points; this was notably true for continuing to drink heavily and/or increasing drinking throughout the gestation of the index child. The control mothers reported significantly lower current drinking levels, which generally declined during pregnancy. Episodic drinking on weekends was the modal pattern for both groups, with "bingeing" (5+ drinks) normative on 2 consecutive days during the weekend for the FAS mother group. More mothers of FAS children smoked than did controls and smoking quantities were identified for smokers of both groups. Members of neither group smoked high numbers of cigarettes (by U.S. standards) or reported cutting tobacco use substantially during pregnancy.

Because it was retrospective, this study had to confirm alcohol intake during the index pregnancy while dealing with problems related to recall of alcohol consumption many years earlier. Measurement of drinking during pregnancy has presented problems in studies of maternal behavior in

both prospective and retrospective studies. In prenatal settings, recall and self report of drinking 3 and 5 months after first report was considered "moderately reliable," with correlations for average daily volume between 0.61 and 0.53 (Robles and Day, 1990). A summary of reliability studies indicated that "research has failed to establish the parameters of reliable recall of alcohol consumption, including the type of information reliably obtained (e.g., quantity, frequency, specific beverages), the relationship between current drinking practices, retrospective reliability, and the reliability of reporting over longer time intervals" (Czarnecki et al., 1990, p. 69). However, a test of the reliability of long-term (5-year), self-report of maternal alcohol consumption concluded that original and retrospective data were highly correlated (Czarnecki et al., 1990). Studies also indicate that *frequency* of drinking is recalled better than *quantity*, that the heaviest drinkers tend to report disproportionately greater consumption when questioned 5 years after the pregnancy, and that "considerable confidence can be placed in retrospective reports of total alcohol consumption by nonalcoholic women over a relatively long-term interval" (Czarnecki et al., 1990, p. 68). Heavy drinking may be reported more accurately retrospectively than when current measurements are taken during pregnancy (Czarnecki et al., 1990; Jacobson et al., 1991). Some mothers may tend to understate their actual levels of drinking during pregnancy due to stigma, low self-esteem and depression (Jacobson et al., 1991).

Recall of exact quantities of alcohol consumed 6 to 9 years earlier in a low SES population in which the modal drinking pattern is a heavy episodic pattern could be problematic and inaccurate. Individuals in such populations experience a greater frequency of blackouts and memory loss, most likely related to quantity of consumption. In this exploratory, community-based study in a unique social and cultural setting, the research team chose to concentrate on current recall of drinking and on ordinal measures of retrospective maternal drinking (e.g., more than or less than) rather than assume that long-term recall or reporting of exact quantities of fetal alcohol exposure was going to be accurate. Stigma could have had an influence on validity in this community; furthermore, we could not assume that methods established in prenatal clinic settings in lower-risk, western populations were directly applicable. As these maternal interviews were tied to the larger study, they were designed to accomplish three tasks: to rule out confounding factors (e.g., smoking and other drugs) that might be of significance for the diagnosis of FAS, to define the general social and biological risk factors extant among these subjects and to provide confirmation of alcohol consumption during pregnancy (as per Stratton et al., 1996, criteria). The prenatal drinking questions were, therefore, asked in a unique, less threatening manner, and we based our quantitative measures on current drinking recall before moving to simple but meaningful questions regarding the pregnancies 6 to 9 years earlier. The interdisciplinary research team developed this conservative approach as the questionnaires were drafted, piloted and finalized.

We have described a population in which the identified current and past high levels of episodic drinking and other maternal risk factors stand out among the mothers of FAS children, although a number of high-risk factors are found in both controls and mothers of FAS children. The percentages of smoking and drinking, for example, are higher for both groups than are reported rates for women in prenatal studies in the United States. Prenatal surveys in three nearby locales in the Western Cape indicated that 42.8% drank alcohol and 45.6% smoked during pregnancy (Croxford, 1998; Croxford and Viljoen, 1999). These percentages are virtually identical to the levels reported by our control group. In this study, however, the case control comparisons have identified some other variables, lifestyle patterns and levels of alcohol misuse that appear to be associated with births of FAS children.

Important questions remain

The amount of alcohol currently consumed by the mothers of FAS children is not dramatically high by most standards (16.1 drinks per week). But the data clearly indicate that episodic drinking is the norm among all drinkers in this community, and "binge" drinking may be the norm for mothers of FAS children. Therefore, high blood alcohol concentrations during episodes and "binges" elevate the likelihood of fetal damage for at least 2- to 3-day periods per week. Furthermore, women may not be reporting (or be able to report) all of the alcohol that they consume. In piloting the study questionnaire, women were found to emphasize alcohol that they had purchased and to possibly overlook or "skip" reporting alcohol distributed to them by others (e.g., fellow workers, spouses or employers). Attempts were made by the research team to remedy this by piloting several revisions of the questionnaire, using pictures of standard alcohol containers of various sizes, and vigilance and persistence by the interviewer. We still believe, however, that alcohol distributed and consumed under group-sharing practices or the vestiges of the Dop system remains underreported, at least among those who live on farms where it is still available in spite of illegality. Furthermore, there are likely to be biologic and genetic variables that exacerbate fetal damage, which were not included in this study. We have reported a very substantial difference in alcohol consumption between subjects and controls and documented the propensity for heavy episodic drinking among mothers of FAS children. A further question raised is: Are these mothers likely to produce FAS children when consuming relatively lower quantities of alcohol than has been shown in studies elsewhere? The physiological char-

acteristics of the female population in this study (e.g., small body size, relatively poor nutrition and diet, and possibly unique genetic traits) suggest that researching other variables (e.g., metabolism and body composition) may eventually elucidate whether there is a particularly high propensity in this population to produce high blood alcohol concentrations per unit of alcohol consumed and, therefore, elevate the overall risk for FAS births.

Advancing age among mothers is a generally established risk factor for FAS elsewhere (see Abel, 1998, pp. 162-166 for a review). The average (SD) age of this study's mothers (26.7 [7.2]) at the birth of an FAS child is several years younger than previously reported in the United States and Western Europe (Abel and Hannigan, 1995; Abel and Sokol, 1986; Bingol et al., 1987a; Jacobson et al., 1996; May et al., 1983; Sokol et al., 1986). Neither initiation of drinking at an early age, nor early onset of regular drinking, explain this difference (Chou and Pickering, 1992). The exclusive episodic drinking pattern among the mothers of FAS children in this study (possibly combined with lifelong and current nutritional deficiency), may lead to higher blood alcohol concentrations in mother and fetus and/or a more rapid degradation of the body (e.g., the liver, leaching of trace minerals, concentration of free radicals, etc.), making FAS more likely to appear in the offspring at an earlier age (see Abel and Hannigan, 1995). More research within this population is needed to answer the question: Why do mothers of the Western Cape seem to be bearing children with FAS at a younger average age than those elsewhere?

Limitations of the study

As noted above, possible biological and genetic risk factors were not included in this study, and other variables that can adversely influence the mother's drinking behavior and alcohol processing could not be addressed. Given that maternal grandmothers tended to drink heavily, a significant number of mothers of FAS children probably were exposed themselves to alcohol prenatally. Since the results of the current study indicate that mothers of FAS children had lower educational attainment than did control mothers, a question raised is whether mothers of FAS children had more neuropsychological deficits than control mothers, leading to diminished self-regulation and alcohol abuse. There exists, also, the possibility that mothers of FAS children had more social and emotional problems than controls. The data suggest that mothers of FAS children felt socially more isolated and "more stressed" during pregnancy. The association between maternal depression, self-esteem, emotions and drinking was not assessed. Finally, small sample size limits the generalizibility of the results. Despite the above limitations, this is the first systematic study to report risk factors for prenatal alcohol effects in a large group of mothers who have borne children in a developing country.

Prevention issues

The results show that maternal characteristics and risk factors found among South African mothers were similar to those reported in the West. Also, it is encouraging that many of the factors that have been identified as possible protective mechanisms from the heaviest maternal drinking in this community are malleable or subject to change. Higher educational attainment is a primary or broad-based universal level prevention option that may well be available to the next generation of mothers in this community, given the rapid changes in South Africa today. Furthermore, the acquisition and daily practice of spiritual beliefs can be facilitated, for religiosity may be a stabilizing force in the lives of the control mothers. The question remains, however, whether these are truly protective factors. Are the lower levels of education and religiosity among the mothers of FAS children merely a result of the stifling effect of growing up and living in an alcohol-saturated family environment (see Hill et al., 1999; Rhodes and Jasinski, 1990)?

Individual and family improvement is amenable to change through both therapy and general social and economic development. Viable alternatives of selective and indicated prevention are new prenatal policies and practices, screening for early detection of alcohol problems, case management of high-risk women and treatment for alcohol misuse (Stratton et al., 1996; May, 1995).

After the initial screening for FAS was undertaken, a number of the members of the South-African and U.S. research teams, combined with local and U.S. prevention specialists, held a 2-day community workshop to plan and establish an ongoing prevention initiative. The session was attended by knowledgeable and concerned people from all over the Western Cape. The power structure of the study community (including the mayor) was well represented. The Institute of Medicine (Stratton et al., 1996) comprehensive prevention model was employed as a framework and as a menu of possible techniques. A coalition was formed that developed a strategy and a multifaceted plan. Prevention efforts are currently underway, and awareness of the FAS and alcohol abuse problems is increasing among many in the Western Cape.

Conclusion

This study, of the characteristics of 31 mothers of children with classic FAS in a unique population, has proven to be both confirming and enlightening. Socioeconomic status, race and some of the local conditions affecting pregnancy were relatively controlled by the homogeneity of the community and the matching of the case-control sample. That the sample is small, unfortunately limits statistical power. Nevertheless, compared with any single previous study of maternal risk, data have here been presented from

more mothers of "full-blown" FAS children. Confirmation of a high rate of FAS within a predominantly lower-SES population has been documented in an unique setting; even in this predominantly low-SES, high-FAS-rate community, the highest rate of FAS was found among the most socially and economically impoverished women from very chaotic extended families. Therefore, findings regarding general social class and family pathology were quite consistent with the U.S. literature; yet, the study has highlighted some potential risk and protective factors among the two groups. Control mothers have spouses who are less likely to be problem drinkers, report more regular practice of their religion, have higher educational attainment and familial networks characterized by much less drinking. The mothers of FAS children report problem drinking throughout their extended family and during all trimesters of the pregnancy, a sexual partner who drinks heavily, lack of religiosity and lower education levels. Studies such as this, which are based on samples of mothers who have had FAS children and appropriate controls, assist in accurate descriptions and general understanding. In addition, they provide contextual information to guide further research for hypothesis testing and for designing appropriate prevention initiatives useful in both the home community and elsewhere.

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