SOCIOECONOMIC STATUS, PSYCHOLOGICAL DISTRESS, AND OTHER MATERNAL RISK FACTORS FOR FETAL ALCOHOL SPECTRUM DISORDERS AMONG AMERICAN INDIANS OF THE NORTHERN PLAINS

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Abstract: The relationship of selected demographic, socioeconomic status (SES), and psychological characteristics was examined in interviews with 176 Northern Plains American Indian mothers whose children were referred to diagnostic clinics for evaluation of developmental disabilities, including fetal alcohol spectrum disorders (FASD). Thirty-nine mothers had children diagnosed with an FASD (Group 1), 107 had children who were not diagnosed with an FASD or other major disability (Group 2), and 30 additional mothers with normally performing children, matched by age, sex, and reservation with those diagnosed with an FASD, were recruited as a comparison group (Group 3). Analysis revealed statistically significant differences ($p \le .001$) in alcohol consumption among all three groups, and a statistically significant difference in the mean Total Distress score among the three groups of mothers, F(2), (176) = 9.60, p < .001, with Group 3 having a lower mean score than Groups 1 and 2. Sequential regression analysis revealed that the quantity of alcohol consumed prior to knowledge of pregnancy, when combined with SES and Total Distress, was more highly associated with having a child diagnosed with an FASD ($R^2 = .206$) than was quantity of alcohol consumed alone.

INTRODUCTION

Numerous studies on alcohol-related birth defects have concluded that maternal drinking, compounded by other risk factors, leads to fetal alcohol syndrome (FAS). FAS is a serious birth defect and the most common non-genetic cause of mental retardation (Hankin, 2002; Abel & Sokol, 1986; O'Connor, Kogan, & Findlay, 2002; May & Gossage, in press). It is unknown how much maternal alcohol consumption results in FAS or other related disorders, or why some women who drink are at substantially higher risk of giving birth to a child with alcohol-related disabilities than others (Stratton, Howe, & Battaglia, 1996). However, researchers have identified several maternal

risk factors differentially associated with FAS. These include advanced maternal age, number of pregnancies, previous births of a child with FAS, cohabitation with a male partner who drinks heavily, and low socioeconomic status (SES; May et al. 2004; 2008a; Viljoen et al., 2002).

FAS research has identified mild to severe adverse effects of prenatal alcohol exposure, many of which form a spectrum of structural anomalies, behavioral problems, and neurocognitive disabilities. This continuum of effects is referred to as *fetal alcohol spectrum disorders* (FASD; Hoyme et al., 2005). The four diagnostic categories included in the Institute of Medicine definition of the continuum are FAS, partial FAS (PFAS), alcohol-related neurodevelopmental deficits (ARND), and alcohol-related birth defects (ARBD; Stratton et al. 1996). Children with a complete phenotype at the severe end of the continuum of FASD have FAS (Hoyme et al., 2005). Estimates of the number of live births in the U.S. that meet the diagnostic criteria for FAS range from 0.3 to 3 per 1,000 (O'Connor et al., 2002; Stratton et al., 1996; Abel, 1995; May & Gossage 2001b); one more recent estimate is as high as 2 to 7 per 1,000 for FAS, and 20 to 50 per 1,000 for FASD in the general U.S. population (May et al., 2009). Children with less severe phenotypes on the FASD continuum present a diagnostic challenge, because their physical features are more subtle. All children with an FASD display a characteristic pattern of behavioral or cognitive abnormalities typical of prenatal alcohol exposure (Hoyme et al., 2005), e.g., poor executive functioning, increased activity levels, problems in inhibition, and motor and memory problems (O'Connor et al., 2002; Kodituwakku, Kalberg, & May, 2001). Therefore, maternal alcohol consumption poses a serious risk to pre- and postnatal human development.

Some of the first studies on FAS anywhere involved American Indians (AIs; May et al., 1983; May, McCloskey, & Gossage, 2002). Generally, rates of FAS and other FASD were found to be high in some tribal communities, but low to non-existent in others. In all, high-rate communities averaged 8 to 9 cases of FAS per 1,000 live births; the rate of FASD did not exceed 30 per 1,000 in any one community studied at one period of time (May et al., 2002).

This study was conducted to identify the relationship of selected demographic and behavioral characteristics to current psychological distress levels among AI mothers who have given birth to children with an FASD. Special diagnostic clinics were held in seven different sites in the Northern Plains to screen and diagnose referred children who might meet criteria for an FASD or other disabilities. Data from structured maternal interviews identified and examined mothers' marital status, age, socioeconomic status, gravidity, parity, current and past drinking patterns, and symptoms of current psychological distress. We hypothesized that these factors would be associated with higher self-reported prenatal drinking levels in mothers whose children were referred for problems and diagnosed with an FASD than among (a) mothers whose children were referred but

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not diagnosed with an FASD and (b) comparison mothers whose children were not referred and did not have problems. The assumption was that psychological distress might be associated with the birth of children with an FASD.

The FAS-specific literature and the general literature on women and alcohol provide insight into maternal drinking during pregnancy. Mothers who drink alcohol during pregnancy are less responsive and less stimulating in their parent/child interactions, and are often under stress. These factors may result in their children's insecure attachment (Kelly, Day, & Streissguth, 2000). The importance of screening pregnant women and new mothers for stress is underscored by the fact that many mothers (40%), especially those from low SES groups, meet clinical criteria for depression (Anhalt, Telzrow, & Brown, 2007). Depressed mothers who are less sensitive and engaged with their children than nondepressed mothers reportedly interact with their infants in a more negative manner (McCarty & McMahon, 2003).

Research on alcohol use and abuse trends among AIs is valuable for a general understanding of the study population. Although there is a lower overall prevalence of drinking among some AI tribes than among the general U.S. population, some AI women who drink are heavy drinkers (May, 1996). A study of a random sample of 1,436 enrolled members from four tribes in the northern U.S. described the quantity, frequency, and variability of their alcohol and other substance use. On a typical day, abstinence from alcohol was the pattern for both men and women (May & Gossage, 2001b). However, those who did drink drank large amounts over a short period of time, resulting in high blood alcohol levels. The mean quantity of drinks consumed, reported by males, was 9 drinks per drinking day; for females, it was more than 5 drinks per drinking day. Frequency of drinking was reported as 4.7 drinking days in the past month for males, and an average of 2.1 days for females. Therefore, on most days, respondents consumed no alcohol at all, but drinking days were characterized by heavy drinking. Regarding variability in drinking behavior, males reported having 5 or more drinks (binge drinking) on 3 days in the prior month, whereas females reported 1.3 days of heavy drinking in the past month (May & Gossage, 2001b). Therefore, one major risk factor for FASD among Northern Plains women is the quantity of alcohol consumed per occasion, rather than the frequency of drinking.

METHODOLOGY

Recruitment Strategies

Participants in the current study were, by design, mothers from several AI tribal backgrounds whose children were referred to special diagnostic/developmental clinics in six rural AI community sites and one urban site in the Northern Plains. The participating mothers were the biological mothers

of the children participating in the clinics. The mothers were recruited from a database maintained for a larger study, which was an ongoing Fetal Alcohol Syndrome Epidemiology research project to determine (via the diagnostic/developmental clinics) the prevalence of FASD in AI communities in the Northern Plains.

A total of 985 children were referred to the clinics. Their ages ranged from 2 months to 17 years. Children were referred either by school or health officials, child protection agencies, or parents because of (1) concerns with inadequate growth, (2) behavior or learning problems, (3) a previous diagnosis of an FASD, (4) a previous diagnosis of a sibling with an FASD, or (5) concerns about the mother's possible drinking during pregnancy.

The diagnostic process utilized all four of the fetal alcohol spectrum diagnoses: FAS, PFAS, ARND, and ARBD (Hoyme et al., 2005). All domains of the diagnostic criteria were considered in the diagnosis: physical growth and dysmorphology, psychological traits, behavior, and maternal risk factors. Final diagnoses were made via a formal, structured case conference with data from clinicians who performed the diagnostic tests for each domain, a pediatric dysmorphologist, an educational diagnostician, and a maternal interviewer.

A total of 293 mothers were included in the larger study database and accounted for the 985 referred children; all but 9.2% were AI. For the current study, mothers were contacted by a project staff member who resided in the same community, and consent for the interviews was requested. Virtually no mothers refused the interview; most of those who did not participate had either moved from the area, had lost custody of their children, or were deceased.

A total of 176 mothers participated in the current study. From this sample, for the purpose of analysis, if her child had been diagnosed with an FASD, the mother was placed in Group 1. If the child had been referred for one of the above problems but not diagnosed with an FASD, the mother went into Group 2. A variety of methods, described below, was used to recruit control mothers for Group 3.

It is important to note that this study and the findings reported here utilized a case control perspective. The comparison mothers are considered controls because their children did not have the study condition, an FASD. The terms "control" and "comparison" are used interchangeably in this article.

To recruit control mothers, notices were placed in newspapers and posted at health facilities and on public bulletin boards seeking typically developing children and their mothers to participate in a developmental clinic. While recruiting controls, project staff asked families if their children had any known developmental disabilities, or were receiving special education services; if so, the children and their mothers were excluded from the study. Control children were matched by tribal community, school, age, and gender with the children who had been diagnosed with an FASD. The mothers of the control children, who became the subjects of this study, had no drinking history during pregnancy and expressed no concerns regarding their children's growth and development; their children were confirmed normal in the diagnostic clinics described above.

Selected information for this paper was obtained from a 230-item maternal questionnaire that covered basic demographic information; general health and medical history; amount and frequency of alcohol consumption at different times in the mother's life; and family and close friends' drinking history.

Alcohol Intake and Demographic Measures

Responses to the following interview questions on the quantity, frequency, and timing of alcohol use prior to knowledge of pregnancy were used to assess alcohol intake: (1) On the days you drank alcohol, how many drinks did you usually drink? (2) How often did you drink this amount? and (3) How far along were you (in your pregnancy) when you found out that you were pregnant? Demographic measures, such as age, education, occupation, and marital status, were included in this same questionnaire.

Psychological Distress

Psychological distress was assessed in the questionnaire with the use of the Self-Report Symptoms Checklist (SSCL-51), developed by Uhlenhuth et al. (1983) to measure common psychological symptoms. It is a slightly modified version of the 58-item Hopkins Symptom Checklist by Derogatis et al. (1974). To our knowledge, this checklist had not been used previously with an AI/AN population; however, it was chosen because it consists of general (i.e., not culturally specific) statements about individual traits. The total score represents a self-report of how much distress a person has experienced in the past month. Symptom subscale scores indicate the ways and the extent to which the person is distressed (e.g., not at all, a little, or a lot). The symptoms are categorized into nine dimensions: somatic anxiety, decreased energy and interest, depressed mood, hostility, anxious mood, panic/phobia, impaired cognitive functions, sleep disturbance, and appetite disturbance. Derogatis et al. (1974) reported that the internal consistency reliability coefficients of the dimensions ranged from .84 to .87. Item-total score correlations were calculated for each dimension, and all were above .50, with most at about .70, indicating substantial shared common variance among the items in the original 58-item checklist.

Data Analysis

All data were entered, and statistical analyses performed, using SPSS for Windows, version 14.0. Initial internal analyses for each measure were conducted, providing descriptive statistics for each defined item, score, and category for the sample subgroups. Intercorrelations of scores within and across variables were conducted for meaning as well as to determine specific measures for subsequent analyses. Correlations, analysis of variance (ANOVA), and regression were used. Descriptive statistics provided a check on whether specific scores met the analytic assumptions.

RESULTS

Selected Maternal Background Characteristics

Selected background characteristics are found in Table 1. The sample consisted of 176 mothers who lived in one of the seven sites, and were categorized into three groups. Of the referred mothers (n = 146), 39 had children diagnosed with an FASD (Group 1), and 107 mothers had children without an FASD diagnosis (Group 2); 30 mothers were selected as comparisons/controls (Group 3).

Table 1 Maternal Background Information: Referred Mothers and Comparison Mothers (N = 176)									
	Group 1: Mothers of children with FASD (n = 39)	Group 2: Mothers of children without FASD (n = 107)	Group 3: Control/ comparison mothers (n = 30)	<i>F</i> -test	p				
Age									
Age on day of interview, mean (SD)	33.1 (6.24)	30.4 (7.75)	31.3 (5.83)		ns				
Age at birth of target child, mean (SD)	25.8 (6.72)	23.7 (5.78)	24.2 (4.60)						
Marital Status									
Married (%)	25.6	22.4	30.0						
Divorced (%)	7.7	0.0	3.3		ns				
Separated (%)	2.6	2.8	3.3						
Single (%)	25.6	33.6	20.0						
Living w/partner (%)	38.5	41.1	43.3						

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Table 1, Continued Maternal Background Information: Referred Mothers and Comparison Mothers (N = 176)									
	Group 1: Mothers of children with FASD (n = 39)	Group 2: Mothers of children without FASD (n = 107)	Group 3: Control/ comparison mothers (n = 30)	<i>F-</i> test	p				
SES									
Business managers (%)	0.0	1.9	3.3						
Administrative (%)	2.6	3.7	26.7						
Clerical/Sales (%)	7.7	6.5	26.7						
Skilled labor (%)	0.0	0.0	3.3	F(2, 174) = 14.17	p < .001				
Semi-skilled (%)	30.8	15.9	10.0						
Unskilled/unemployed (%)	43.6	43.9	16.7						
Homemaker (%)	2.6	6.5	3.3						
Student/disabled (%)	10.3	20.6	10.0						
Educational Level									
Less than 7th grade (%)	0.0	0.9	0.0						
7th to 9th grade (%)	25.6	8.4	0.0						
10th to 11th grade (%)	20.5	31.8	0.0	<i>F</i> (2, 173) = 12.67	р < .001				
HS diploma or GED (%)	25.6	38.3	40.0		P ····				
Some college (%)	25.6	16.8	53.3						
College graduate (%)	2.6	2.8	3.3						
Graduate school (%)	0.0	0.9	3.3						
Parity, mean (SD)	4.1 (1.7)	3.5 (1.8)	2.8 (1.3)	F(2, 176) = 4.88	p < .006				
Gravidity, mean (SD)	5.1 (1.9)	4.4 (2.2)	3.7 (1.5)		ns				

Table 1, Continued

Referred and comparison mothers were similar in age at interview and at the birth of the target children. The data show that 38.5% of mothers in Group 1, 41.1% in Group 2, and 43.3% in Group 3 were unmarried and living with a partner, indicating no significant difference in marital status.

Mothers' SES was defined by using the one-step method for the Hollingshead scores (Hollingshead, 1957), based on occupational status (e.g., unemployed, homemaker, student, disabled) and type of occupation, if any. The majority of Group 1 and 2 mothers (43.6% and 43.9%, respectively) reported that they were in the occupational category of unemployed/unskilled, compared to 16.7% of Group 3 mothers. Overall ANOVA was not significant; however, post hoc analysis

revealed significantly higher SES in the control group than in the two referred groups. Among the three groups, 46.1% of Group 1 mothers, 41.1% of Group 2 mothers, and 0% of Group 3 mothers reported that they had not completed high school. Similarly, overall ANOVA was not significant and post hoc analysis revealed significance between the control group and the two referred groups.

Parity (number of live births) was highest among mothers of children with an FASD (Group 1) and differed significantly compared to controls (Group 3; p < .006). Post-hoc analysis revealed no significant difference in mean parity between the two groups of referred mothers. Mean gravidity (total number of pregnancies) also was highest among Group 1 mothers. Group 2 mothers had fewer pregnancies, but post-hoc analysis revealed no significant differences in gravidity among the three groups.

Children

From the total maternal sample (N = 176), 306 children were screened: 143 females (46.7%) and 163 males (53.3%), with an age range from 2 months to 17 years. Of these children, 94 (30.7%) were from referred mothers who had at least one child diagnosed with an FASD (Group 1); 50% were female, with a mean age of 78.34 months (SD = 49.58). One hundred seventy-seven (57.8%) were from referred mothers whose children did not have an FASD diagnosis (Group 2); 44.6% were female, and the mean age was 79 months (SD = 49.1). Thirty-five (11.4%) children were born to control mothers (Group 3); 48.6% were female, and the mean age was 82.86 months (SD = 44.23).

Alcohol Consumption Prior to Known Pregnancy

In Table 2, alcohol consumption is reported as the number of drinks consumed per month prior to knowledge of pregnancy. Of the Group 1 mothers, 30 out of 39 (76.9%) reported drinking before they knew they were pregnant, and provided the amount of alcohol consumed. Nine mothers (23.1%) reported drinking "unknown" amounts of alcohol. Of the Group 2 mothers, 99 out of 107 (93%) reported drinking and provided the amount of alcohol consumed; 8 others (7%) reported "unknown" amounts of alcohol.

Mean and Standard Deviation of Reported Number of Drinks per Month Prior to Knowledge of Pregnancy for the 3 Groups of Mothers (N = 159)									
	Group 1 (<i>n</i> = 30)		Grou (<i>n</i> =		Group 3 (<i>n</i> = 30) ANO		ANOVA	р	
	X	SD	X	SD	X	SD			
Drinking amount per month	173.58*	256.68	59.65**	130.37	16.19	52.43	F(2, 159) = 8.917	p < .001	

Table 2

Asterisks indicate significant post-hoc pairwise comparison between groups adjusted using Bonferroni techniques

*Group 1 (children diagnosed with FASD) was found to differ significantly from both Group 2 (no diagnosis of FASD) and Group 3 (comparison group)

**Group 2 was significantly different from Group 3 (comparison group)

ANOVA results revealed significant differences in reported alcohol consumption among the three groups, especially between Group 1 mothers and mothers in the other two groups. Group 1 mothers reported the highest alcohol consumption and gave birth to 47 affected children (an average of 1.2 affected children per mother). There were 21 children diagnosed with FAS, 20 with PFAS, 5 with ARND, and 1 with ARBD.

Psychological Distress

Using the SSCL-51 to assess mothers' overall levels of distress and the incidence of common psychological symptoms of distress, a Total Distress score (range, 0-102) was obtained. Nine symptom subscale scores indicate type and extent of distress. Symptoms were scored from 0 (*not at all*) to 2 (*a lot*). Table 3 presents the mean scores, standard deviations, and multivariate ANOVA (MANOVA) for the average Total Distress score and symptom subscale scores among the three groups of mothers. Experimentwise alpha = .05 was achieved by setting alpha for each test at .005, adjusted for the number of scales.

Mean Scores, Standard Deviations, and MANOVAs for the SSCL-51 Total Distress Score and Symptom Subscale Scores									
	Group 1 (<i>n</i> = 39)			oup 2 107)		up 3 30)	<i>F</i> -test (2, 176)	р	
	X	SD	X	SD	X	SD			
Total Distress	31.74	(20.42)	27.24	(22.83)	11.00	(8.90)	9.60	<.001*	
<u>Symptoms</u>									
Somatic Anxiety	.461	(.38)	.361	(.45)	.162,3	(.21)	4.42	.013	
Decreased Energy	.71 ¹	(.58)	.57	(.54)	.33 ³	(.30)	4.47	.013	
Decreased Mood	.72 ¹	(.46)	.681	(.58)	.25 ^{2,3}	(.29)	9.05	<.001*	
Hostility	.701	(.49)	.621	(.49)	.25 ^{2,3}	(.27)	9.43	<.001*	
Anxious Mood	.65 ¹	(.45)	.56 ¹	(.53)	.262,3	(.27)	6.29	.002*	
Panic/Phobia	.431	(.47)	.401	(.49)	.042,3	(.08)	8.54	<.001*	
Impaired Cognition	.621	(.53)	.471	(.48)	.10 ^{2,3}	(.14)	11.3	<.001*	
Sleep Disturbance	.70	(.68)	.67	(.66)	.40	(.51)	2.57	.079	
Decreased Appetite	.641	(.51)	.43	(.51)	.33 ³	(.37)	3.34	.038	

Table 3

*p < .005 (for the SSCL-51 symptom subscales, the adjusted alpha = .005, 2-tailed test)

^{1,2,3}Indicate significant post-hoc pairwise comparison between groups using Bonferroni adjustment of significance (1 = Comparison group, 2 = No FASD group, 3 = FASD group)

Total Distress Score

As shown in Table 3, the referred mothers (Groups 1 and 2) had higher distress than the comparison mothers (Group 3). Group 1 mothers had the highest level of distress (31.74). MANOVA and post-hoc analysis revealed a significant difference in Total Distress between the comparison mothers (Group 3) and the referred mothers (Groups 1 and 2), but not between Group 1 and 2 mothers.

Distress Symptom Subscale Scores

Table 3 also provides the mean score (0, 1, or 2) for each symptom subscale for the three groups, indicating the frequency of each stress symptom. Sleep disturbance was most commonly reported. Group 1 mothers reported that distress occurred with decreased mood, decreased energy, hostility, and sleep disturbance. Group 2 mothers reported that distress occurred with decreased mood, sleep disturbance, and hostility. Group 3 mothers cited distress with sleep disturbance, decreased energy, and decreased appetite.

The correlation of each symptom subscale score with other symptom subscale scores and with the Total Distress score was moderately high to high. Table 4 provides data on the correlations between the symptom subscale scores and with the Total Distress score. Decreased mood and anxious mood correlated most highly with Total Distress, and sleep disturbance the lowest.

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Table 4 Correlation of the SSCL-51 Symptom Subscale Scores and the Total Distress Score (N = 176)										
	Somatic anxiety	Decreased energy	Decreased mood	Hostility	Anxious Mood	Panic/ Phobia	Impaired Cognition	Sleep Disturbance	Decreased Appetite	Total Distress
Somatic Anxiety	1.00	.714*	.678*	.625*	.741*	.727*	.679*	.554*	.645*	.854*
Decreased Energy		1.00	.787*	.646*	.740*	.691*	.713*	.516*	.592*	.872*
Decreased Mood			1.00	.709*	.802*	.690*	.710*	.590*	.607*	.899*
Hostility				1.00	.656*	.606*	.685*	.464*	.497*	.802*
Anxious Mood					1.00	.780*	.716*	.613*	.618*	.899*
Panic/ Phobia						1.00	.748*	.492*	.535*	.856*
Impaired Cognition							1.00	.530*	.595*	.859*
Sleep Disturbance								1.00	5.04*	.662*
Decreased Appetite									1.00	.704*
Total Distress										1.00

*Correlations significant at the .001 level (2-tailed)

With the high intercorrelations in mind, we examined whether mothers of children diagnosed with an FASD (Group 1) obtained significantly higher scores than did mothers in Groups 2 and 3 on the nine separate subscales assessed on the SSCL-51. Symptom subscale scores showed a consistent pattern of an average increase in the extent of distress reported by the Group 1 mothers, compared to Group 2 and Group 3 mothers. An omnibus ANOVA and post-hoc analysis (using p < .05) indicated significantly higher scores for Group 1 mothers on eight of the nine symptom

subscales when compared to Group 3 mothers, and for six of the nine symptom subscales when compared to Group 2 mothers. Sleep disturbance was the major exception, as it was distributed almost equally across all groups. Group 2 mothers did not receive significantly higher scores than Group 3 mothers in decreased energy and decreased appetite. Group 1 and 2 mothers did not differ significantly in the extent of their symptoms on any of the scales; therefore, all referred mothers (Groups 1 and 2) were essentially equally distressed.

In addition, to prevent alpha slippage and to utilize a more discriminating level of significance, we conducted a MANOVA analysis using a probability level of .005; this analysis indicated statistical significance for five of the nine symptom subscales among the three groups of mothers: decreased mood, hostility, anxious mood, panic/phobia, and impaired cognition.

Combined Alcohol Consumption, SES, and Total Distress

The ultimate goal of this study was to examine potential risk factors (i.e., selected personal and demographic characteristics of mothers) that, in addition to quantity of alcohol consumed prior to knowledge of pregnancy, could increase the likelihood of an FASD diagnosis in offspring. Reported level of distress and SES were selected as the potential risk factors to consider. As shown in Table 5, the quantity of alcohol consumed prior to knowledge of pregnancy, Total Distress score, and SES, taken together, were more highly associated with diagnosis of an FASD than was the single variable of quantity of alcohol consumed. Knowing the quantity of alcohol consumed prior to knowledge of pregnancy produced a 9.6% reduction of error in predicting the birth of a child with an FASD. Knowing both alcohol consumption amount and SES reduced error in predicting an FASD by 16.9%, and adding Total Distress Score reduced error by 20.6%. The combined variables provided a significant increase in predicting which mothers were likely to have children with an FASD.

Drinking Prior to Pregnancy, SES, and Total Distress Score as Predictors of Mothers Having a Child with FASD: Sequential Regression ($N = 176$)									
Variables	R	R Square	Adjusted <i>R</i> Square	F	Sig.				
Alcohol	.309	.096	.090	16.365	< .001				
SES	.411	.169	.158	15.627	< .001				
Total Distress	.453	.206	.190	13.194	< .001				

Table 5

DISCUSSION

This study examined, individually and in combination, the relationship of the quantity of alcohol consumed prior to knowledge of pregnancy and selected personal and background characteristics to the birth of children with an FASD in a sample of 176 mothers. Analyses utilized three groups of mothers: those whose children were diagnosed with an FASD (Group 1), those whose children were not diagnosed with an FASD (Group 2), and controls (Group 3).

Limitations

First, because this study involved questions that were retrospective in nature, that is, requiring the mother to recall alcohol consumption approximately five years after the fact, recall accuracy may at first pose a concern. But the reliability of long-term (5-year) self-report of drinking has been documented through correlations of original and retrospective data, in that heavy drinking may be more accurately reported retrospectively than during pregnancy (May et al., 2008a; Viljoen et al., 2002; Alvik et al., 2006; May et al., 2005; Czarnecki et al., 1990). In fact, a recent study has found that retrospective data collected as long as 14 years after pregnancy are more accurate than similar data collected in the prenatal period (Hannigan et al., 2009).

Second, the present study would have benefited in assessing the validity of the information obtained by using a test-retest method to determine reliability. The maternal interviewer independently conducted an internal audit of the responses after each interview (see Appendix 1). Concerning the veracity of the responses, 5% were believed to be grossly misrepresented due to contradictory information found in the child's chart or obtained from reliable sources. The sampling technique used in this study was nonprobability sampling (specifically, purposive sampling) and the data are cross-sectional and time bound. External validity addresses the question of whether this sample represents the larger population of mothers whose children have an FASD. Because the referred mothers in this study were suspected of drinking during pregnancy or already had at least one child diagnosed with an FASD, they appear likely to be representative of the larger population of mothers whose children have been exposed to alcohol in utero in these Northern Plains communities.

Third, the comparison group is a small, matched, convenience sample and, thus, may not be considered representative of all AI mothers who meet the same group criteria. However, their children were matched by age, sex, and residence to children with an FASD to ensure appropriate comparisons. Furthermore, most of the variables addressed (e.g., SES of most individuals in the participating communities) tended to be robust over time. This finding does not ensure that the need to care for children with an FASD did not cause reduced employment and a subsequent reduction in SES level. Similarly, low SES and psychological distress may also negatively influence drinking style and alcohol use. But given the cross-sectional nature of the study, we cannot completely speak to these possibilities.

A fourth limitation was the overall small sample size. Because of this, a targeted case control design was used to compare the results for the three groups first via analysis of variance, appropriate for these data. As the results for the comparisons of SES, education level, and drinking behavior were significant and powerful for the three groups, and also for SSCL-51 scores between categories, moving on to more sophisticated analysis was warranted.

Despite these possible limitations, the authors believe that the study findings are consistent with axiomatic expectations and provide an accurate examination of the relationship of maternal characteristics to the birth of children with an FASD.

Selected Maternal Demographics

In general, the findings from this study are similar to previous findings on associations with FASD: high parity, single/cohabitation status, and low SES (May et al., 2005). As reported in previous studies of maternal risk for FASD, mothers of children with an FASD in this study had higher parity rates than the other two groups (Kvigne, et al., 2003; May, et al., 2005, 2008a) and were more likely to be single or living with a partner than were controls (May et al., 2008a; Kivgne et al., 1998); low education level was also present as a risk factor for FASD (O'Connor et al., 2002; May et al., 2005, May et al., 2008a).

Abel & Hannigan (1995) have identified low SES, which can lead to poor nutrition and stress, as a primary risk factor for FASD. They also noted examples of FASD occurring in predominantly low-SES populations, regardless of race. The first cases of children diagnosed with FAS by Jones and Smith (1973) involved mothers receiving welfare. Other studies of children with an FASD, where the participants were equally divided by race, also were characterized by poverty, with family income averaging less than \$400/month (Abel & Hannigan, 1995). It is possible that low-SES populations are more often targeted for FASD research; however, other studies comparing low- and high-SES groups show similar results. One study by Bingol et al. (1987) compared two groups of women with alcoholism from different social strata. Each group of women drank an average of 6 ounces of absolute alcohol a day (12 drinks), yet the women of low SES were 16 times more likely to give birth to children with an FASD than were the upper middle-class women. Findings from South Africa also illustrate that mothers of children with an FASD had lower incomes than controls, and suffered poorer nutrition and lower body mass index (May et al., 2005, 2008a; Viljoen et al., 2002). All of the above findings are consistent with this study and suggest that low SES, along with co-factors such as stress and poor nutrition, is significantly associated with FASD.

When combined with heavy drinking, advanced maternal age has been reported as a risk factor for FASD (Hankin, 2002; May et al., 1983). In this study, the mean age of referred mothers whose children had an FASD, although slightly older at the time of delivery than that of comparison mothers, did not differ significantly from that of referred mothers whose children did not have an FASD. The comparison mothers, however, reported consuming less alcohol.

Alcohol Consumption

Drinking prior to knowledge of pregnancy proved to be a useful measure of alcohol consumption. Mothers whose children had an FASD reported drinking significantly more alcohol prior to knowing they were pregnant than mothers in the other two groups, which suggests a high likelihood of continuing an increased level of drinking during pregnancy. Group 1 mothers reported a mean of 9.1 weeks before they knew that they were pregnant, compared to 7.6 and 5.8 weeks, respectively, for the Group 2 and Group 3 mothers. Thus, even if mothers quit drinking once they learned they were pregnant, the children with an FASD were exposed to alcohol longer in utero. We also know from other questionnaire items that most of the Group 1mothers did not quit, but drank throughout pregnancy.

Psychological Distress

Mothers of children with an FASD had higher Total Distress and symptom subscale scores. Maternal psychological distress may increase drinking and/or otherwise negatively influence the mother-child relationship, placing children at risk for later developmental and emotional problems. Previous findings indicate that children whose mothers reported moderate to heavy alcohol consumption during pregnancy had significantly higher levels of depressive symptoms than children of light drinkers or abstainers (O'Connor & Paley, 2006). Also, mothers with depression lasting for years after giving birth may place their children at risk for lower cognitive and language functioning and behavioral problems during formative years (Anhalt, Telzrow, & Brown, 2007). In one study of 100 randomly selected AI adults, Parker et al. (1997) found that 18% seeking primary care at an urban Indian Health Service clinic met full criteria for a mental health disorder, and 17% met the subthreshold for a mental disorder. The most frequently diagnosed disorders were alcohol abuse/dependence, major depressive disorders, and generalized anxiety disorders. With the findings regarding comorbidity in this study, lack of diagnosis and treatment for depression may place future children at risk for an FASD.

The Combined Effect of Alcohol Consumption, SES, and Level of Distress

The sequential regression analysis revealed that the combination of mothers' higher prenatal alcohol consumption, low SES, and higher Total Distress scores was more likely to be associated with children's FASD diagnoses than was mothers' prenatal alcohol consumption alone—11% more. All three of these variables were found to significantly influence the birth of children with an FASD and to explain 20.6% of the variance in FASD diagnosis. The findings suggest that low SES and high distress are influential in combination with prenatal drinking. Thus, it appears important for future research and clinical care to include screening for psychological distress during pregnancy, especially in individuals with low SES and other known maternal risk factors. Information about pregnancy, depression, and drinking might be provided to youths on a routine basis, perhaps in schools, community centers, and health clinics, so that early awareness might lead to prevention.

SUGGESTIONS FOR FUTURE RESEARCH

Despite ongoing prevention efforts, recent studies have indicated that 13% of women continue to drink during pregnancy. This information suggests that at least 1 in 8 fetuses is exposed to alcohol (Hankin, 2002; "Alcohol Use," 2009; O'Conner, & Whaley, 2007). The findings from this study suggest that it is vital for future research to focus on identifying characteristics of prenatal drinking, such as psychological distress, other behavioral components, and SES, to provide a means for appropriate prevention. Once these factors are identified, the directives for prevention and intervention are to change attitudes, lifestyles, stress levels, and behavior during pregnancy. It has been demonstrated that prevention efforts delivered via case management have benefited AI women who have a history of alcohol abuse in pregnancy, and such efforts can be highly effective (May et al., 2008b). The focus of case management is to promote a healthy pregnancy outcome via multiple strategies: establish trust and confidence between mothers and case managers, provide a support network for transportation and health care appointments, ensure proper prenatal care, reduce stress, and motivate women to change (May et al., 2008b). Research that identifies specific personal, behavioral, and social influences on maternal drinking is valuable. Once these factors are identified, the directions for prevention and intervention are clear. FASD is theoretically 100% preventable. This study shows that successful alcohol abuse prevention among AI women must consider their mental health status.

Why some women continue to drink during pregnancy is not fully understood. A vast majority of Northern Plains AI women (88-94%) know about FAS, but many still do not, or cannot, abstain from alcohol when pregnant (May & Gossage, 2001a). Therefore, ongoing studies, especially those that identify children with an FASD, are vital for assessing the characteristics of mothers for

appropriate prevention measures. Future research also should focus on comparing mothers at both extremes of SES and depression—those with low SES and high SES and those with and without depressive symptomotology—to examine further the potential protective factors that influence personal well-being. This current study provides additional evidence that mothers who drink during pregnancy are placing their unborn children at risk of developing FASD.

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APPENDIX 1 – INTERNAL AUDIT QUESTIONS

Sensitive questions regarding quantity, frequency, and timing of alcohol consumption during the pregnancy of the target child, as well as other personal, behavioral, and social factors, were included to help assess the validity of the responses.

1. Was the information given distorted by the woman's misrepresentation?

Factors considered to measure the accuracy of the responses were the following: Was the information contradictory to the child's chart, such as documentation of intoxication at birth; was the information contradictory with information given from a reliable source; and were there contradictions within the interview?

2. Was the information given distorted by the risk category of the woman?

Factors that were considered to answer this question were: Is the woman currently drinking and pregnant; is the woman currently drinking and not using birth control; has the woman been abstinent less than one year, but her partner is still drinking and she has not made any positive changes in her social situation; and has the woman had surgical sterilization?

ACKNOWLEDGEMENTS

This project was funded by the National Institute on Alcohol Abuse and Alcoholism (NIAAA) under grants R01 AA9440, R01-UO1 AA11685. Multiple constituents have collaborated closely with the project, including the tribal councils and health officials of six Northern Plains tribes, the Indian Health Service, and local public health officials. Many individuals at NIAAA have long advocated for FAS prevention, including Jan Howard, Ph.D., Faye Calhoun, D.P.A., Kenneth Warren, Ph.D., Enoch Gordis, M.D., Michael Hilton, Ph.D., Marcia Scott, Ph.D., and T-K Li, M.D. Without their support for the tribal communities of the Northern Plains and the research team they funded, this project would not exist. The Fetal Alcohol Syndrome Epidemiology Research (FASER) Prevention field team, which was involved in many areas of FASD prevention and was most influential in recruiting mothers for interviews, included Irene Lake, Rose Maestas, the late Joan Alvord, Renee Parker, Lorinda Beck, Mary White Country, Karen Goodhart, Whitney Renville, Mabel Bad Moccasin-Granados, Rene Fasthorse, Sherlynn Herrera, and Jill Plumage. The University of New Mexico-based FASER team members who carried out the clinics and have maintained all of the child and maternal data include Wendy Kalberg, Alfredo Aragon, David Buckley, Jan Gossage, Eugene Hoyme, Luther Robinson, and Melanie Manning. Each has contributed greatly to this study, and we express our appreciation to each of them.