

# Identifying Maternal Self-Reported Alcohol Use Associated With Fetal Alcohol Spectrum Disorders

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**Background:** The incidence of Fetal Alcohol Syndrome (FAS) has been estimated at 1 to 3 per 1000 live births. Fetal Alcohol Spectrum Disorders (FASD) (which include FAS) are estimated to occur in about 1 in 100 births. Cessation of drinking during pregnancy can improve the outcome even if the unborn child is already affected. For individuals born with FASD, an early diagnosis appears to be a protective factor against secondary disabilities. A quick screening tool to identify newborn children at risk has been elusive.

**Methods:** A simple descriptive presentation is offered that shows where 36 individuals with FASD were found from among the many patterns and amounts of prenatal alcohol use that were reported by a sample of 1439 pregnant women whose offspring were later examined within the first 7 years of life.

**Results:** Individuals with FASD (i.e., those with FAS, fetal alcohol effects, alcohol-related neurodevelopmental disorder) were found within two aggregates of alcohol scores that together recommend a set of three to four alcohol questions. Within this derivation sample, one scoring of the questions yields almost 78% sensitivity and 97% specificity for FASD. Another scoring of the same instrument yields 100% sensitivity with 90% specificity.

**Conclusions:** These new data may facilitate early identification of offspring who may be most in need of early intervention, namely those born with FASD.

**Key Words:** Prenatal Alcohol Exposure, Pregnancy Risk, Fetal Alcohol Syndrome, Fetal Alcohol Effects, Screening.

ALCOHOL IS THE most commonly used teratogen in the western world, and many of the dose-response effects of prenatal alcohol on the fetus are well established. In general, the higher the dose of alcohol, the more pronounced the effects. There is no known low dose that appears to be safe for every pregnancy (Jacobson and Jacobson, 1994; Sampson et al., 2000; Streissguth et al., 1993).

At high doses, the most long lasting and debilitating effects characterizing this birth defect are on the central nervous system (CNS) and are manifest in behavior and functional disturbance. People with the most identifiable forms of the birth defect have been given various diagnoses such as Fetal Alcohol Syndrome (FAS), Fetal Alcohol Effects (FAE), Alcohol-related Neurodevelopmental Disorder (ARND), or static encephalopathy, alcohol exposed. These forms of the Fetal Alcohol Spectrum Disorders (FASD) are differentiated primarily by the facial features present or absent, but they share developmental abnormal-

ities in adaptive functioning, attention and memory problems, distractibility, learning problems, poor judgment, and fine and gross motor difficulties (Streissguth and O'Malley, 2000). Among all individuals who are prenatally exposed to alcohol, those with FASD are often most in need of developmental, medical, and community interventions.

The estimated prevalence of FASD from a population-based study in Seattle found a conservative rate of FASD (FAS and ARND) to be nearly 1 per 100 births (Sampson et al., 1997). The earlier in pregnancy a woman can stop drinking, the better the outcome (Little et al., 1984); the younger the age at which an affected child is identified, the lower the frequency of secondary disabilities (Streissguth et al., 1996). Early detection of affected newborns is thus paramount for intervention in the course of this pervasive developmental disability.

A quick screening tool for identification of newborns most at risk has been elusive. Even in a large sample of patients with FASD (specifically FAS and FAE) only about 14% are mentally retarded (IQ below 70) (Streissguth et al., 1996). Only 4 of 34 subjects (11.8%) from a birth cohort who were later identified as FASD (specifically FAS and ARND) had low birthweight (<2500 g) (Sampson et al., 1994). These common markers of a child at risk have poor sensitivity for these alcohol-related birth defects.

Alcohol clears from the system too quickly to allow laboratory monitoring of alcohol use across pregnancy to be practical. Verbal self-report is important for screening

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for alcohol use during pregnancy and at delivery. An early set of questions designed to help clarify alcohol abuse from self-report, the Alcohol Problems Scale deriving from Rimmer (1971), identified less than 1% of women and not all of them were currently drinking (Streissguth et al., 1977). A review of newer screens such as the T-ACE (tolerance, annoyed, cut down, eye-opener) and TWEAK (tolerance, worried, eye opener, amnesia, cutdown) (Russell, 1994) finds them to be sensitive to the riskiest prenatal drinking as defined by consuming over 1 oz of alcohol per drinking day before pregnancy (Hankin et al., 2000). Various criteria such as ">1 oz per drinking day," "7 or more drinks per week," or "5 or more drinks on an occasion" have been suggested as defining heavy/moderate drinking for FAS research (Hankin and Sokol, 1995; Jacobson et al., 1993; Stratton et al., 1996). Yet many screens for risk level drinking during pregnancy ask questions only about alcohol abuse where "abuse" is defined by the respondent. Additionally, Kaskutas and Graves (2000, in press) find that women who report frequent drinking, underestimate quantities of alcohol consumption with the highest error. Whaley and O'Connor (1999) reported that many health care workers felt uncomfortable asking their clients about alcohol use. Adding to the discomfort of instruments such as the TWEAK and T-ACE are questions that some people might consider to be judgmental such as, "Have you ever had a drink first thing in the morning to steady your nerves or get rid of a hangover (eye-opener)?"

In the 1970s, Majewski found that severity of damage from prenatal alcohol exposure among people with FAS seemed to be correlated to the mother's stage of alcoholism (Majewski, 1981; Seidenberg and Majewski, 1978). Jacobson et al., (1998) report data that indicate higher risk of ARND for offspring of older mothers compared with offspring of younger mothers. Two conceptions about FAS that have arisen based on these types of findings are: (1) that young mothers cannot have a child with FAS, and (2) that the more children a drinking mother has, the more likely she is to have a child with FAS.

This report uses the Seattle Longitudinal Prospective Study cohort (Streissguth et al., 1993) to explore maternal age, parity, and selected sets of alcohol scores from the 1974–1975 prenatal interview for their properties regarding the early identification of individuals who were later identified as having FASD. The goal was to provide information that will help future projects to make decisions about a family's risk of FASD from self-reported prenatal alcohol-use data that can be gathered either at delivery or during pregnancy.

## METHODS

### *The Sample*

During 1974–1975, 1529 consecutive women who received prenatal care at one of two Seattle area hospitals were interviewed regarding a broad spectrum of prenatal health habits including use of alcohol, cigarettes, marijuana, caffeine, street drugs and medications, nutrition, and basic demographic characteristics. The interviews were conducted during

the 5th month of pregnancy. At that time, alcohol questions were asked regarding two time periods: during this target pregnancy (D), and retrospectively to the month or so prior to recognition of pregnancy (P; the month or so prior to pregnancy or pregnancy recognition). From these 1529 pregnancies, 1439 singleton, live-born were located at birth. As the babies delivered, a cohort of about 500 were chosen for a longitudinal prospective follow-up study in which all heavily alcohol-exposed infants were selected along with some unexposed. Details of the study design can be found in Streissguth et al., (1993).

### *Assessment of FASD*

Each member of the cohort was examined at least once for diagnosable FASD (FAS, FAE, ARND): (1) blind examination at birth by a dysmorphologist (Hanson et al., 1978), (2) blind examination at 4 years of age by a dysmorphologist (Graham et al., 1988), (3) blind examination of photographs at 7 years of age by experienced observers (Clarren et al., 1987), and/or (4) behavioral phenotype assessed from blind-collected neurobehavioral data at birth through 7 years driven by a study of the relationship of prenatal alcohol to neurobehavioral outcomes (Streissguth et al., 1993). Not every member of the birth cohort participated in each of these four exams. The 36 subjects detected by one or more of these four examinations will be considered "true" FASD in the analyses that follow. Persons detected by one or more of these examinations who were "unexposed" according to the alcohol scores were considered non-FASD for this project. A total of 36 subjects in the cohort have been identified as FASD from one or more of these four exams.

### *The Alcohol Questions*

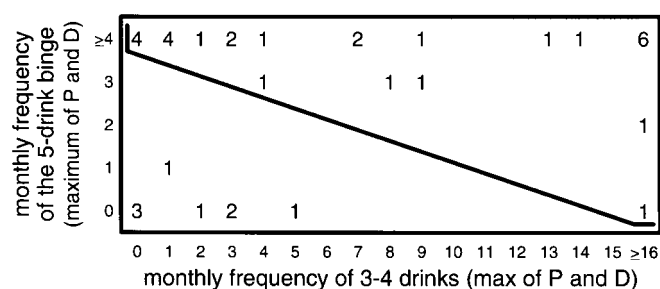
The alcohol questions included the Quantity-Frequency-Variability interview (Cahalan et al., 1969; Jessor et al., 1968). Each mother was asked the absolute frequency per month with which she consumed each given beverage (wine, beer, liquor). For each beverage type she was asked for the relative frequency with which she consumed various quantities (1–2, 3–4, and 5 or more drinks) when she drank. Summary scores were assessed for each of the two gestational time periods P and D. The selection of alcohol scores for the current project is restricted to summaries that could be obtained from a few simple questions, even if only as a first stage screening. Alcohol scores include number of intoxications, binge scores (5 or more drinks, the 5-drink binge; and 3–4 drinks), frequency of alcohol use (even if not in large quantities), over 1 oz per drinking day, number of drinks per week, and the Alcohol Problems Scale (Rimmer et al., 1971).

### *Statistical Analysis*

An exploratory presentation will show where individuals with FASD are found with respect to selected maternal self-report alcohol scores, age, and parity. Because almost all heavily exposed offspring are in the follow-up cohort, it will be assumed that all the cases of FASD in the screening sample of 1439 are in the smaller follow-up cohort. All 1439 will be used for the denominators of percentages. Three descriptive statistics will help the reader to compare characteristics of the alternative alcohol criteria: sensitivity is the percent of true FASD who meet the criteria; specificity is the percent of the non-FASD who (appropriately) do not meet the criteria; and accuracy within the criterion group is the percent of the group who are true FASD. Ideally, all three of these statistics would be 100%.

## RESULTS

Figure 1 presents a scatterplot of monthly frequency of 5-drink binge against monthly frequency of 3–4 drinks (excluding 5 or more) just for the 36 mothers of the individuals with FASD. The digits on the scatterplot indicate the number of individuals with FASD who were exposed to the reported frequencies of binges. Twenty-eight of the 36



**Fig. 1.** Frequency of binge drinking self-reported by 36 mothers whose children were later considered FASD. The digits on this scatterplot show how many of the 36 mothers reported each combination of scores: times per month (frequency) of 5-drink binge, by times per month (frequency) of 3–4 drinks on an occasion. The eight mothers in the lower left reported daily (or, almost daily) drinking.

FASD cases (77.8%) lie on or above a line empirically determined by the weighted sum: “frequency of 5 or more + 1/4 of the frequency of 3–4”  $\geq 4$ . This boundary is drawn on the chart. Scores above this line will be considered positive for risk of FASD on a new alcohol score: the Binge Alcohol Rating Criterion (BARC).

The remaining eight individuals with FASD fall in the lower left quadrant—fairly separate from the other 28 cases. What these eight individuals have in common is mothers who report 27 or more drinking occasions a month (this is daily or almost daily) in conjunction with as little as 1 to 2 drinks per occasion.

Based on the above observations, the 1439 mothers were sorted into three groups based on self-report prenatal drinking. Table 1 defines the groups and shows the observed frequency of FASD among these three groups of mothers. BARC (criterion 1 on Table 1) yields almost 78% sensitivity with 97% specificity for FASD within this sample. If the screening uses instead the union of these first two criteria, then 100% sensitivity is achieved with about 90% specificity. This union of criterion 1 and 2 will be called the Frequency-Binge Aggregate Score (F-BAS).

For each of seven original alcohol criteria, Table 2 pre-

**Table 2.** Sensitivity, Specificity, and Accuracy of Criteria ( $N = 1439$ )

Alcohol-defined criteria	Sensitivity/ specificity for FASD	Accuracy of criterion
Alcohol Problems Scale, one or more	0%/99%	0%
Intoxication(s), one or more	44%/90%	10%
Binge Alcohol Rating Criterion (BARC)	78%/97%	38%
5-drink binge (even once)	81%/85%	12%
$\geq 7$ drinks per week	97%/83%	13%
$>1$ oz per drinking day	100%/68%	7%
Frequency-Binge Aggregate Score (F-BAS)	100%/90%	21%

Note: These seven criteria are not mutually exclusive.

**Table 3.** Questions Proposed for Alcohol Screening for Risk of FASD

	Month or so before pregnancy	During this pregnancy
Please estimate the following. Write “0” if it never happened:		
Number of times per month that you drank 5 or more drinks on an occasion	# _____	# _____
Number of times per month that you consumed 3–4 drinks on an occasion	# _____	# _____
Number of times per month that you consumed just 1–2 drinks on an occasion	# _____	# _____
Did you drink alcohol almost every day, even if only a small quantity? (circle)	yes no	yes no

Note: The fourth question is redundant, but is suggested as a check on the three first questions, and seems to reflect the tone of many of the respondents represented in Table 1, Criterion 2.

sents the sensitivity and specificity scores observed from this sample of 1439 pregnancies and the accuracy within criterion. They are ordered by increasing sensitivity for FASD. The single scale for alcohol abuse (Alcohol Problems Scale) did not identify even one case of FASD in this sample. Two alcohol-defined criteria identify all 36 of the people with FASD, but 1 of these ( $>1$  oz per drinking day) has much lower specificity than the other (F-BAS). The column labeled “accuracy of criterion” may be helpful to projects needing to identify as many individuals with FASD as possible within the smallest sample possible. The new BARC score yields the highest “accuracy of criterion,” with offspring of more than 1/3 of the mothers reporting positive BARC scores later being identified as FASD.

In this sample, 9 of the 36 individuals identified as FASD (25%) were born to teenage mothers, women who had not yet reached their 20th birthday; 16 of the 36 (44%) were first born children. Filtering for older multiparous mothers would not improve classification of children for presumed risk of FASD in this sample.

Table 3 presents a set of questions that are suggested by these data analyses as a short screen to identify newborn children at risk for FASD.

## DISCUSSION

Teratogenic effects depend on exposure and individual factors in the mother and child. Not everyone who is exposed prenatally to alcohol above the levels suggested by these questions displays FASD. On the other hand, indi-

**Table 1.** Frequency of FASD by Maternal Self-Report of Prenatal Alcohol Use From 1439 Singleton Live Born Children

Criteria	Prenatal alcohol report	N	FASD n (%)
1.	Monthly frequency of 5 or more drinks + 1/4 monthly frequency of 3–4 drinks $\geq 4$ (P and/or D)	73	28 (38.4)
2.	Daily or almost daily drinking without meeting criteria 1 above (24 or more drinking occasions per month) (P and/or D)	99	8 (8.1)
3.	Everyone else	1267 (~500)*	0 (0)

Note: Criterion 1 includes exactly the alcohol reports that lie in the area above the diagonal line on the Figure 1. Criterion 1 also defines the Binge Alcohol Rating Criterion (BARC). The Frequency-Binge Aggregate Score (F-BAS) is positive for anyone having a positive score for criteria 1 or 2.

\* A total of about 500 offspring were individually examined. Because the 500 examined include almost all of the heaviest drinkers from the original screening sample, it is assumed that all individuals with FASD were identified.



viduals not identified as "at risk" by the suggested screen are not necessarily unaffected by prenatal alcohol exposure. It is the nature of dose-response relationships that effects from low dose are small.

Characteristics of seven new or historical criteria for identifying newborns at risk of FASD have been compared. None achieved 100% sensitivity and 100% specificity within this derivation sample. Two criteria (">1 oz per drinking day," and the F-BAS) yield 100% sensitivity, meaning that every individual in this sample with FASD was reportedly exposed to these patterns. Specificities are only 68% and 90% respectively, showing that many individuals without FASD also were exposed to these levels of alcohol. The TWEAK and the T-ACE were unfortunately not available at the time this study began.

It is hoped that this presentation of characteristics of new and old self-report alcohol scores, evaluated here within a common sample, will be helpful to researchers and clinicians needing to make decisions about individuals. For one study, poorer specificity may be tolerated if it means greater success of identifying every individual with FASD; for another study, poor specificity may be too expensive and a sample with high accuracy of the criterion for FASD is desirable. Two new scores, the BARC and F-BAS, appear to offer better specificity than many alternatives. The degree to which they should be accompanied with detailed personal interviews depends on the goals of the initial screenings.

The literature on dose-response effects of prenatal alcohol exposure mandates caution if extrapolating these findings to demographically different samples. For example, the usefulness of these guidelines to groups with a higher minority sample or less affluent samples needs further research.

The observation that individuals with FASD in this sample were found within two distinctly different alcohol-use domains (Fig. 1) may interplay with the variety of outcomes associated with prenatal alcohol exposure. The mothers of FASD who reported daily drinking with low or zero frequency of binges were also among the oldest mothers of FASD offspring. The combination "older age plus daily drinking" may reflect an age effect; but underestimation of drinks per day when there have been so many occasions to remember, as found by Kaskutas and Graves (2000, in press) in their careful study of women who drank frequently, is also a compelling interpretation. For detection of affected children, clearly those associated with a report of daily drinking of any quantity need to be considered to be at risk.

The good performance of several alcohol criteria reviewed here demonstrates the efficacy of quick screens that may not measure true alcohol dose but that, nonetheless, capture an essence of drinking practice from brief maternal report that is related to FASD in the offspring. Diagnosis of FASD cannot be made from self-reported alcohol use alone. The guidelines suggested by these data identify in-

dividuals who appear to be at highest risk of FASD, and who therefore benefit the most from full evaluation for FASD. This is not a screen of defective children. It is a screen for individuals at risk of FASD. Subsequent evaluation of the offspring should focus on developmental and behavioral measurements known from the animal and human literature to be affected by prenatal alcohol. Short screening shows promise of an ability to identify offspring who may be most in need of early intervention, i.e., individuals born with FASD. It also shows promise of a short screen to identify mothers who may benefit from additional interviews.

In 1981, the Surgeon General of the United States published a warning to abstain from alcohol during pregnancy and when planning a pregnancy (Food and Drug Administration, 1981). The current report should not be interpreted as modifying, in any way, this important health warning that has subsequently been endorsed by other Surgeons General and many professional organizations. The CDC reports that drinking among women of child bearing age generally declined during the 1980s, but that the frequency seems to have risen again during the 1990s (CDC, 1997). Identification of newborn children most in need of intervention for prenatal alcohol exposure remains a serious medical and public health concern.

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