

The Wisconsin Fetal Alcohol Syndrome Screening Project

*Marianne Weiss, DNSc, RN; Christine E. Cronk, ScD; Sandra Mahkorn, MD, MPH;
Randall Glysch, MS; Sara Zirbel, MSN, RN*

ABSTRACT

Problem: Fetal Alcohol Syndrome (FAS) is preventable, under-diagnosed, and under-reported. Wisconsin rates for alcohol use and binge drinking in childbearing-age women exceed the national average. FAS prevalence in Wisconsin has not previously been systematically evaluated.

Methods: The Wisconsin Fetal Alcohol Syndrome Screening Project (WFASSP) used a multi-stage, multi-source prospective population-based screening methodology to identify children born in 1998-1999 in Southeast Wisconsin who met a surveillance case definition for FAS. The 4-stage methodology used screening of electronic birth files, abstraction of neonatal medical records, and direct assessment of facial features, growth, and development at age 2 to 3 years.

Results: The FAS prevalence rate was 0.23 per 1000 births. Children directly evaluated had fewer demographic, pregnancy, and maternal substance use risk factors than lost-to-follow-up children. Thirty-two percent of children with weight and head circumference below the 10th percentile at birth were developmentally delayed and 47% had at least one physical growth delay.

Conclusions: The WFASSP methodology identified

children who had not previously been diagnosed with FAS. Using the combination of weight and head circumference below the 10th percentile at birth is a useful methodology for identifying children at substantial risk for growth and developmental delays from FAS or other unspecified etiologies.

INTRODUCTION

Fetal Alcohol Syndrome (FAS) is a leading cause of preventable birth defects and developmental disabilities¹ that results in long-term physical, cognitive and developmental deficits.² While the prevalence of any alcohol use among pregnant women has decreased from 16.3% in 1995 to 12.8% in 1999, the rates of binge drinking (2.9% in 1995 and 2.7% in 1999) and frequent drinking (3.5% in 1995 and 3.3% in 1999) reported by pregnant women has remained largely unchanged.³ Data from the Behavioral Risk Factor Surveillance System indicate that binge drinking (defined as 5 or more drinks on an occasion, 1 or more times per month) rates for Wisconsin women are consistently well above the national average and have increased from 1990 to 2002 (Wisconsin: 11.2% to 15.5%; United States: 6.5 to 8.1%).⁴ While women typically reduce alcohol consumption during pregnancy,⁵ these rates are alarming.

Current population-based systems in Wisconsin (Birth certificate data, Birth Defects Outcomes Monitoring Program) underestimate the prevalence of alcohol consumption in pregnant women and the number of children with FAS. In 2002, only 1.1% of women responded affirmatively to drinking during pregnancy on a self-report question on the birth certificate form. The Wisconsin electronic birth file reports only to the first decimal for International Classification of Disease (ICD) codes.⁶ Yet, for 1998 and 1999, only 0.03% and 0.02% of birth records included ICD-9 code 760.7, which captures noxious influences affecting the fetus from alcohol consumption, substance use, or other sources.

FAS is the most serious of a constellation of possible

The Wisconsin Fetal Alcohol Syndrome Surveillance Project (WFASSP) was funded through Cooperative Agreement Award # U50/CCU514563 awarded by the Centers for Disease Control and Prevention to the Wisconsin Bureau of Health Information, Sandra Mahkorn, Principal Investigator. Dr Weiss is an associate professor at St Joseph Regional Medical Center and Sr Rosalie Klein Professor of Women's Health at Marquette University College of Nursing. Dr Cronk is an associate professor in pediatrics at the Medical College of Wisconsin. Doctor Mahkorn is medical director for the Wisconsin Division of Health Care Financing. Mr Glysch is a research scientist for the Wisconsin Bureau of Community Health Promotion and was the program coordinator for the WFASSP. Ms Zirbel is a nurse practitioner at St Ben's Clinic for the Homeless in Milwaukee and was the project's research assistant. Address correspondence to Marianne Weiss, DNSc, RN, Marquette University College of Nursing, PO Box 1881, Milwaukee, WI, 53201-1881; e-mail marianne.weiss@marquette.edu.

outcomes associated with prenatal alcohol exposure that have recently been aggregated under the umbrella term Fetal Alcohol Spectrum Disorder (FASD).⁷ FASD includes the range of outcomes associated with all levels of prenatal alcohol exposure and includes FAS, alcohol-related birth defects (ARBD), and alcohol-related neurodevelopmental disorder (ARND). Risk factors associated with FASD in addition to alcohol dose, pattern of intake, and timing of exposure during pregnancy include co-exposure to other teratogens and environmental contaminants (eg smoke, lead), high parity, low socioeconomic status, race, poor nutrition, poor health, and increased stress. It is clear that the influences are multifactorial and interdependent.⁸⁻⁹

Prevalence estimates for FAS vary widely reflecting the methodologic challenges in case-finding, sampling, differing study designs, inconsistencies in diagnosis, and variation in birth defects reporting. Previous studies have used a variety of passive surveillance and active case ascertainment methods to estimate the prevalence of FAS in general and selected high-risk populations.^{8,10} Under-reporting and inaccuracy are major concerns due to variation in the level of provider training to identify FAS, inconsistent criteria applied to case definition, and biases related to diagnostic labeling and stigmatization.^{8,11} Birth defects registries often have a preponderance of cases enrolled from the first year after birth. Diagnosis of FAS is most accurate between ages 3 and 12¹⁰ and often is not diagnosed or documented in newborns even in the presence of identifying characteristics.¹² Overestimation of general population prevalence may occur if the study cohort represents a high-risk population. Active case ascertainment is a population-based approach that uses active review of records from a variety of sources (medical, school, etc) and direct methods of diagnosis to identify children with case characteristics. More cases are detected than with passive surveillance due to its multi-source methodology.¹³ Published prevalence estimates have ranged from less than 1 per 1000 births for general birth cohorts to more than 10 per 1000 births for high-risk populations.^{8,10,14} The estimated prevalence rate for the United States is between 0.5 and 3.0 per 1000 live births.^{8,10} The rate for all measurable effects (FAS, ARBD, ARND) is estimated to be 1% or more.¹⁰

In 1997, the Centers for Disease Control and Prevention (CDC) awarded funds to 5 states under a cooperative agreement to develop population-based FAS surveillance systems using a multi-source methodology.¹⁵ FASSNet (Fetal Alcohol Syndrome Surveillance Network), a standardized, multi-source surveillance

method was developed for use in data collection. Alaska, Arizona, Colorado, and New York used a multi-source retrospective record-based case review approach. Prevalence ranged from 0.3 to 0.4 per 1000 births in 3 states and 1.5 per 1000 births in Alaska for a combined prevalence estimate of 0.4 per 1000 births.¹⁶

METHODS

The Wisconsin Fetal Alcohol Syndrome Surveillance Project used a prospective, multi-source, multi-stage active case ascertainment system to identify children who met diagnostic criteria for fetal alcohol syndrome. Each successive screening stage assessed one of the project's case selection criteria. The case selection criteria for the WFASSP and FASSNet were both based on the 1996 Institute of Medicine (IOM) Report on FAS.⁸ The IOM FAS diagnostic criteria, FASSNet case definition, and the WFASSP case selection criteria for each screening stage are presented in Table 1.

The birth cohort for the project consisted of infants born in 1998 and 1999 in 22 birth hospitals to mothers resident in an 8-county southeast region of Wisconsin. This region included urban, suburban, and rural households. In the first screening stage, cases were selected using the electronic birth file from the Wisconsin Bureau of Health Information. Cases selected in Screen 1 were small for gestational age, defined as birth weight below the 10th percentile (BWT<10th) based on sex and gestational age-specific reference values from the 1991 US Live Birth File.¹⁷ To adjust for the expected lower birth weight values for low risk African American infants,¹⁸ 10th percentile cut-offs for African American infants (based on maternal race category on the birth certificate) were 200 g less than those for other infants.

In Screen 2, neonatal medical records were abstracted for birth head circumference (BHC), gestational age, evidence of maternal alcohol use or other FAS case criteria, and contact information (for use in the next screening stage). Medical records for children who were adopted or had died were not abstracted. Newborns with BHC less than gestational age-specific 10th percentile (BHC<10th)¹⁹ were retained for the next screen level.

Families of children with positive findings (BWT and BHC<10th) were contacted when the child was between 2 and 3 years old. Following IRB approvals and after securing a Certificate of Confidentiality from the National Institute of Alcohol Abuse and Alcoholism, parents were contacted by letter of introduction and subsequently by telephone to discuss participation in

Table 1. Institute of Medicine, Fetal Alcohol Syndrome Surveillance Network and Wisconsin Fetal Alcohol Syndrome Screening Project Case Definitions

*Institute of Medicine FAS Diagnostic Criteria for Category 2: FAS without confirmed maternal alcohol exposure	†FASSNET Case Definition Criteria: Confirmed FAS phenotype with or without maternal alcohol exposure‡	WFASSP Case Selection Criteria
<p>Evidence of growth retardation, as in at least one of the following:</p> <ul style="list-style-type: none"> a. Low birth weight for gestational age b. Decreasing weight over time not due to nutrition c. Disproportional low weight for height 	<p>Growth criteria:</p> <p>Intrauterine weight or height corrected for gestational age \leq10th percentile</p> <p style="text-align: center;">Or</p> <p>Postnatal weight or height \leq10th percentile</p> <p style="text-align: center;">Or</p> <p>Postnatal weight for height \leq10th percentile</p>	<p>Screen #1:</p> <p>Birth weight $<$10th percentile corrected for gestational age and race</p>
<p>Evidence of CNS neurodevelopmental abnormalities, as in at least one of the following:</p> <ul style="list-style-type: none"> a. Decreased cranial size at birth b. Structural brain abnormalities (eg microcephaly, partial or complete agenesis of the corpus colosum, cerebellar hypoplasia) c. Neurological hard or soft signs (as age appropriate) such as impaired motor function skills, neurosensory hearing loss, poor tandem gait, poor eye-hand coordination 	<p>Central Nervous System (CNS) criteria:</p> <p>Frontal-occipital circumference \leq10th percentile at birth or at any age</p> <p style="text-align: center;">Or</p> <p>Standardized measure of intellectual function $<$1 standard deviation (SD) below the mean</p> <p style="text-align: center;">Or</p> <p>Standardized measure of developmental delay $<$1 standard deviation (SD) below the mean</p> <p style="text-align: center;">Or</p> <p>Developmental delay or mental retardation diagnosed by a qualified medical examiner (eg psychologist or physician)</p> <p style="text-align: center;">Or</p> <p>Attention deficit disorder diagnosed by a qualified evaluator</p>	<p>Screen # 2:</p> <p>Gestational-age specific frontal-occipital circumference $<$10th percentile at birth</p>
<p>Evidence of a characteristic pattern of facial abnormalities that includes:</p> <ul style="list-style-type: none"> a. Features such as short palpebral fissures, and b. Abnormalities in the premaxillary zone (e.g. flat upper lip, flattened philtrum, and flat midface) 	<p>Face criteria:</p> <p>Abnormal facial features consistent with FAS as reported by a physician</p> <p style="text-align: center;">Or</p> <p>Two of the following:</p> <ul style="list-style-type: none"> • Short palpebral fissures • Abnormal philtrum • Thin upper lip 	<p>Screen # 3:</p> <p>Two of the following:</p> <ul style="list-style-type: none"> • Short palpebral fissures • Abnormal philtrum • Thin upper lip

*Source: Institute of Medicine⁹

†Source: Hymbaugh et al¹⁵

‡ Documentation in the records of some level of maternal alcohol use during the index pregnancy.

the direct assessment stage. Screen 3 consisted of assessment of facial features of the FAS phenotype, and measurements of growth (weight, height, head circumference) and development (Denver Developmental Screening Test II [DENVER II]). All assessments in this screen level were carried out by graduate nursing students at Marquette University College of Nursing, and completed either at the Marquette Infant Assessment Lab (44%) or at the family's home (56%). Assessors had received 16 hours of training in procedures used for subject contact and enrollment, growth

measurements, developmental assessment, and evaluation of facial phenotype.

Facial features were assessed by 2 independent observers using the Astley-Clarren philtrum and lip chart.²⁰ Scores of 4 or 5 were considered positive for features characteristic of the FAS phenotype. Palpebral fissure length was measured using a clear plastic ruler following a technique described by Hall et al.²¹ Two measurements of each eye were taken by each of 2 observers. The child was considered positive for the feature if the smallest of the measurements was below the

Table 2. Demographic, Pregnancy, and Substance Use Risk Factors for WFASSP Birth Cohort and Screening Levels

	Birth Cohort	Screen 1: BWT <10th Percentile	Screen 2: BWT & BHC <10th Percentile	Screen 3: Seen	Screen 3: LTFU
N	56,257	3291	615	177	438
Smoking	16%	33%	31%	21%	32%
Alcohol Use	2%	3%	4%	7%	13%
Illicit Drug Use	2%	4%	6%	5%	13%
Alcohol or Drug	NA	6%	8%	10%	19%
African American	21%	19%	35%	21%	40%
Milwaukee County	53%	56%	63%	57%	66%
Unmarried	36%	45%	54%	40%	60%
Mother < 20 Years	13%	17%	18%	14%	20%
< High school Graduate	20%	26%	29%	17%	34%
Inadequate Prenatal Care	4%	8%	9%	3%	11%
≤36 Weeks Gestation	11%	15%	19%	22%	18%

BWT=birth weight; BHC=birth head circumference; LTFU =lost to follow up; NA =not available.

10th percentile.²¹ The child met the case criteria for FAS if 2 of the 3 facial criteria were positive (philtrum, lip, eyes).

Growth measurements and developmental assessments were also completed for all children seen at Screen 3. Anthropometric measurement followed methods described by Lohman and coworkers.²² Weight was measured using a calibrated scale and recorded to the nearest 0.1 kilogram. Recumbent length was measured to the nearest millimeter (mm) using a recumbent length board. Head circumference was measured to the nearest mm using a non-flexible tape. All measurements were repeated twice by 2 observers, and the 2 closest measurements were averaged. The DENVER II was completed by 2 observers and followed procedures outlined in the DENVER II training manual.²³ Children with 2 FAS facial characteristics were referred for dysmorphology evaluation, further developmental testing, and assessment of maternal alcohol use (Screen 4).

In addition to the prospective screening methodology described above, a retrospective case ascertainment approach similar to the methodology of the other 4 FASSNet state projects¹⁵ was used to evaluate whether the WFASSP screening methodology missed cases documented in other sources. Records for all children in the birth cohort who had received inpatient or outpatient services at Children's Hospital of Wisconsin were screened for the study's case selection criteria. ICD-9 codes for FAS, microcephaly, and maternal substance use were used as the initial search strategy. Records were then abstracted for the same case selection criteria as the WFASSP prospective methodology.

RESULTS

The birth cohort for the project consisted of 56,247 in-

fants. A total of 3291 (6% of the birth cohort) met Screen 1 criteria and were selected for Screen 2 medical record abstraction. Of the records reviewed for Screen 2, weight or gestational age for 38 neonates (1.2%) had been misreported to the state vital records office, and did not meet the criterion of BWT<10th. Of the remaining records, a total of 615 cases met Screen 2 criteria (19% of Screen 1 cases, representing 1% of the birth cohort) and were eligible for Screen 3 assessment. Of cases eligible for participation in Screen 3, 177 (29%) agreed to participate (referred to as Screen 3 Seen), and 438 (71%) were lost to follow-up (referred to as Screen 3 LTFU). Three hundred and six children (50%) could not be located, 130 (21%) parents refused, and 18 (0.3%) were excluded for other reasons (died, moved). Screen 3 assessments were completed at an average of 28 months of age (range 21-41 months).

Demographic and pregnancy risk factors were obtained from the electronic birth file and used to compare the characteristics of children at each screening stage. Compared with the birth cohort, the Screen 2 (BWT and BHC<10th) children were more likely to have been exposed to smoking, alcohol use, and illicit drug use in utero, and had higher rates of demographic and pregnancy risk factors than the birth cohort (Table 2). Children evaluated (Seen) in Screen 3 had fewer demographic and pregnancy risk factors than those who were lost to follow-up and a lower rate of smoking, alcohol, and illicit drug use.

Thirteen children evaluated in Screen 3 met the case criteria for FAS, yielding a prevalence estimate of 0.23 per 1000 births. Three of these children had evidence of alcohol exposure during pregnancy in the electronic birth record or neonatal medical record. None of the children meeting the FAS case definition had documen-

Table 3. FAS Diagnostic Criteria and Follow-up Growth and Development Delays Present in Children (n=13) Who Met the FAS Case Definition

Subject #	Birth Measurements		Measurements at Age 2-3 Years					Alcohol Use in Pregnancy		
	BWT <10th	BHC <10th	WT <10th	HC <10th	Lip	Philtrum	Palpebral Fissures	DENVER II Delay	Reported on Birth Certificate	Reported in Other Source *
1	+	+	+	+	+	+	+	+		
2	+	+	+	+	+	+	+	+		+
3	+	+			+	+	+	+		
4	+	+	+		+	+	+	+		+
5	+	+	+	+		+	+	+	+	+
6	+	+	+		+	+	+	+		
7	+	+			+		+	+		
8	+	+	+		+		+	+		
9	+	+	+	+	+	+	+	+		
10	+	+			+		+			
11	+	+			+	+	+			
12	+	+		+	+	+				
13	+	+	+	+		+	+			

* Maternal/neonatal medical record or verbal report at Screen 3 Evaluation; BHC=Birth head circumference; BWT=Birth weight.

tation of microcephaly in the neonatal medical record or in the electronic birth file, and none had been previously diagnosed with FAS. Table 3 displays findings for case selection criteria and follow-up growth and development assessment for the 13 children meeting the FAS case definition. These children were referred to a dysmorphologist for Screen 4 assessment and diagnosis.

Among the children evaluated in Screen 3, nearly a third (31.6%) were classified as suspect (i.e. delayed) on the DENVER II. The most common deficit subscale was language, followed by gross motor, personal-social, and fine motor skills (Figure 1). Nearly two-thirds (66%) were delayed on 2 or more subscales. Forty-seven percent of the children seen for Screen 3 evaluation had at least 1 physical growth measurement less than the age- and sex-specific 10th percentile. Weight (36.9%) was the most commonly restricted physical growth parameter. Of children with developmental delays, 62% also exhibited at least 1 physical growth delay. Z scores for height, weight, head circumference, and weight for height were all significantly lower for children with developmental delays than for children without evidence of delay on the DENVER II (Table 4).

In the retrospective case review, 23 children with a clinical diagnosis of FAS were identified. Of these, 7 met the Screen 1 criteria, 2 met the Screen 2 criteria but could not to be located for Screen 3 follow-up, and none met the criteria for the FAS case definition.

DISCUSSION

Using a multistage screening ascertainment system, 13

cases of FAS were identified out of a birth cohort of 56,247 (prevalence estimate 0.23 per 1000 births). This rate is similar to rates reported in previous studies, but lower than those reported by the majority of other states in the FASSNet project that used a retrospective record-based multi-source methodology (Table 5).¹⁵ The Wisconsin rate was lower than 3 of the 4 FASSNet states but was within the 95% confidence interval of 1 of the states (Colorado).

The prevalence rate documented by the WFASSP is likely a conservative estimate of the true prevalence of FAS in Wisconsin. The screening methodology using BWT and BHC<10th as the first 2 screening parameters identified a subset of children with a higher risk profile for FAS as well as growth and development deficits when compared with the birth cohort. A selection bias was evident in the children seen for Screen 3 evaluation. The Screen 3 Seen children had fewer demographic, pregnancy, and maternal substance use risk factors than the children who were lost to follow-up. It is likely that the rates of FAS, physical growth delay, and developmental delay would be higher if the entire group of children eligible for Screen 3 assessment were evaluated.

A number of important differences between Wisconsin and the other 4 states in the FASSNet surveillance project may account for differences in prevalence estimates. The WFASSP used more restrictive case selection criteria than the other FASSNet states. The Wisconsin case criteria used BWT and BHC <10th while the FASSNet case definition adopted by the

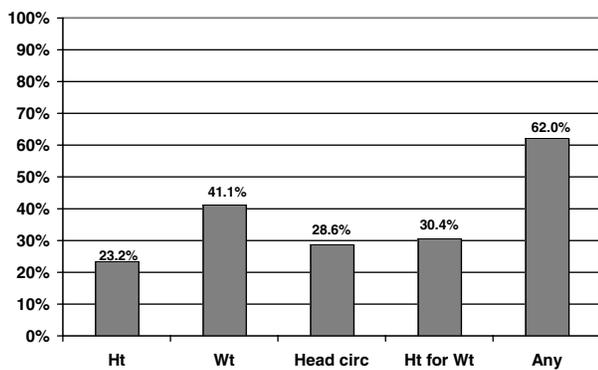


Figure 1. Percent of developmentally delayed children (N=56) with co-occurring growth measurements less than 10th percentile at age 2-3 years.

Table 4. Comparison of Z Scores for Growth Measurements in Children With and Without Developmental Delay

GM	DD	N	Z	SD	t	df	p
Height	No	113	-0.42	0.88	2.36	163	0.02
	Yes	52	-0.78	0.92			
Weight	No	116	-0.66	1.09	3.10	168	0.002
	Yes	54	-1.22	1.09			
Head Circumference	No	112	-0.29	0.88	3.40	161	0.001
	Yes	51	-0.84	1.14			
Weight for Height	No	113	-0.43	1.05	2.67	162	0.009
	Yes	51	-0.91	1.07			

GM=growth measure; DD=developmental delay; Z=mean Z score; SD=standard deviation

Table 5. Prevalence of FAS (per 1000 Births, ± 95% Poisson Confidence Interval) by State

	# Cases	# Births	Rate per 1000 Births	Lower CI	Upper CI
Alaska*	45	30,284	1.49	1.05	1.92
Arizona*	75	223,043	0.34	0.26	0.41
Colorado*	25	95,136	0.26	0.16	0.37
New York*	40	88,789	0.45	0.31	0.59
Wisconsin	13	56,257	0.23	0.11	0.36

*Source: CDC¹⁶

other 4 states used BWT and BHC equal to or less than the 10th percentile. In addition, the other states permitted inclusion as a case if 1 of several growth or central nervous system (CNS) parameters met the case definition criteria at any age in the surveillance period. Because 1 of the goals of the Wisconsin project was to test a multi-stage screening approach that might promote earlier detection of FAS, inclusion as a case required that the growth and CNS criteria be met at birth. This, however, excluded any children who exhib-

ited normal weight or head circumference at birth but had subsequent below-normal growth patterns. The Wisconsin approach evaluated children at 2 to 3 years of age. In the other states in the project, data were collected for a 1995 to 1997 birth cohort with a 2- to 4-year post-birth period for case-finding,¹⁵ giving more opportunity for case identification. For these reasons, it is not unexpected that the prevalence rate for Wisconsin is at the lower end of the range for FASSNet states.

None of the Wisconsin children identified through the prospective screening methodology had been previously diagnosed. In the retrospective case review for additional cases not identified by the screening methodology, 23 cases were found, with no duplication from the prospective screening method. While the retrospective method identified children with an established clinical diagnosis, the Wisconsin method identified children who had not been referred for evaluation of FAS. Thus, these 2 ascertainment methods identify different children. Combining the multi-source detection strategies offered by the Wisconsin prospective ascertainment approach and the records-based surveillance of the other FASSNet states would provide a comprehensive strategy for prevalence estimation and population-based surveillance of FAS. A clear advantage of the prospective approach is that it provides an opportunity to combine case finding for surveillance purposes with early detection and initiation of intervention.

The children who screened positive in Screen 3 met the case definition for FAS without confirmed maternal alcohol exposure (IOM Category 2).⁸ The source data for maternal alcohol use was the State of Wisconsin electronic birth file and abstracted neonatal medical records. Only 3 out of 13 cases had evidence of alcohol exposure documented in these sources. Maternal alcohol exposure is substantially underreported in birth records.²⁴ In the study sample, 3% of mothers in Screen 1 (BWT<10th) and 4% in Screen 2 (BWT and BHC<10th) reported any alcohol use during the pregnancy. These data indicate substantial underreporting of the prevalence of prenatal alcohol exposure compared to BRFSS data for Wisconsin and the nation.

No children who met the case definition completed the follow-up referral to the dysmorphologist despite active attempts to schedule appointments. Some mothers reported that they were already connected with a pediatric specialist for another medical problem. For others, costs of transportation, difficulties with child-care and work schedules, and insurance issues inter-

ferred with completion of the diagnostic process. In future early identification and intervention programs for FAS, services to assist families with the logistics of diagnosis and intervention will be necessary.

Persistent growth and development deficits have been widely reported for FAS children^{2,8} and were documented in this study. Of particular interest are the rates of growth and development delays found in children who were positive on the first 2 screening criteria (BWT and BHC<10). Without consideration of facial feature characteristics of FAS, these 2 criteria identify a group of children at high risk for growth and developmental delays that persist through age 2. Kirby²⁵ has noted the importance of recording and monitoring the co-occurrence of birth defects and developmental disabilities to better understand their overlapping nature and differentiate between perinatal outcomes, postnatal etiologies, and normative growth and development of children with birth defects. In this study we were able to assess the developmental outcomes of co-occurrence of BWT and BHC<10th, with results indicating that nearly one-half had persistent physical growth delays and one-third had developmental delays. Co-occurrence of persistent growth delays occurred in 62% of cases with developmental delays.

The etiology for the CNS (BHC<10th) and growth (BWT<10th) deficits at birth and the high rates of developmental delay, growth delay, and co-occurrence of growth and developmental delays persisting at age 2 to 3 years are unknown among this study population. It is possible that these findings reflect a range of effects from alcohol use. However, because of the incomplete documentation of maternal alcohol use, the relationship between alcohol use and the study findings cannot be evaluated adequately. Other etiologies were undoubtedly involved in developmental and growth delays in these children. Regardless of the etiologies, the group of children screened using this method represent children at substantial risk for persistent growth and development delays who would benefit from early intervention. Unfortunately, mothers of children with developmental delays in this study rarely reported referral to or participation in any kind of developmental follow-up or intervention program.

A number of factors may have limited the reliability of the prevalence estimate. The high lost-to-follow-up rate may have resulted in under-ascertainment of FAS cases. Risk factor data indicated that lost-to-follow-up children were at greater risk for FAS and postnatal growth impairment and developmental delays. The major reason for loss to follow-up was use of birth

record contact information to locate parents 2 years after the birth. Children who were adopted at birth or who were in foster care were not evaluated and may also represent a group at higher risk for FAS.¹⁴ This methodology also missed any FAS cases where growth and CNS criteria were not present at birth. Children in this study who met the Screen 3 criteria met the case selection requirements for both the IOM and FASSNet case definitions but did complete the Screen 4 clinical diagnosis stage. Systematic assessment of prenatal alcohol exposure was also a planned component of the Screen 4 evaluation. Consequently, all cases identified in this study can be classified as FAS without confirmed alcohol exposure (IOM Category 2)⁸ or confirmed FAS Phenotype without documentation of in utero alcohol exposure (FASSNet case definition).¹⁵ The difficulty in referral and completion of the clinical diagnostic process underscores the problems with obtaining accurate prevalence estimates.

The WFASSP methodology has several clear strengths for identifying FAS cases. Screens 1 and 2 used measurements routinely obtained on neonates at birth. Because these measurements also identified individuals at risk for developmental and growth delay, they could be used as a cost-effective first level screen for at-risk infants including those with FAS. Second, early identification of at-risk infants (Screens 1 and 2) allows secondary and tertiary prevention to be effectively instituted. Third, the criteria used to identify features of FAS were consistent across all cases, and all assessments had documented high reliabilities.

RECOMMENDATIONS

The results of this study suggest that using a simple screening approach beginning with identification of 2 of the FAS case definition criteria (BWT and BHC <10th percentile), which are commonly available at the time of birth to providers and public health workers, offers the opportunity to initiate a surveillance process for children at risk for FAS and/or developmental and physical growth delays. Better documentation of prenatal alcohol exposure and FAS characteristics at birth will reduce under-reporting of this birth defect and promote better case finding for early intervention.

REFERENCES

1. Floyd RL, Sidhu JS. Monitoring prenatal alcohol exposure. *Am J Med Genet Part C: Seminars Med Genet.* 2004; 127C:3-9.
2. Larkby C, Day N. The effects of prenatal alcohol exposure. *Alcohol Health Res World.* 1997;21:192-198.
3. CDC. Alcohol use among women of childbearing age – United States, 1991–1999. *MMWR.* 2002;51:273-276.

4. CDC. Behavioral Risk Factor Surveillance System. Available at www.cdc.gov/brfss/. Accessed September 3, 2004.
5. Serdula M, Williamson, DF, Kendrick, JS, Anda, RF, Byers, T. Trends in alcohol consumption by pregnant women: 1985 through 1988. *JAMA*. 1991;265:876-879.
6. State of Wisconsin. Wisconsin Birth File 2002. Available at <http://dhfs.wisconsin.gov/births/pdf/02births.pdf>. Accessed September 3, 2004.
7. Sokol RJ, Delaney-Black V, Nordstrom B. Fetal alcohol spectrum disorder. *JAMA*. 2003;290:2996-2999.
8. Stratton K, Howe C, Battaglia F, eds. *Fetal Alcohol Syndrome: Diagnosis Epidemiology, Prevention, and Treatment*. Washington, DC: National Academy Press; 1996.
9. Abel EL. An update on incidence of FAS: FAS is not an equal opportunity birth defect. *Neurotoxicol Teratol*. 1995;17:437-443.
10. May PA, Gossage JP. Estimating the prevalence of fetal alcohol syndrome. *Alcohol Res Health*. 2001;25:159-167.
11. Cordero JF, Floyd RL, Martin ML, Davis M, Hymbaugh K. Tracking the prevalence of FAS. *Alcohol Health Res World*. 1994;18:82-85.
12. Little BB, Snell LM, Rosenfeld CR, Gilstrap LC, Grant NF. Failure to recognize fetal alcohol syndrome in newborn infants. *Am J Dis Child*. 1990;144:1142-1146.
13. Egeland GM, Perham-Hester KA, Gessner BD, Ingle D, Berner JE, Middaugh JP. Fetal alcohol syndrome in Alaska, 1977 through 1992. *Am J Public Health*. 1998;88:781-786.
14. Astley SJ, Stachowiak J, Clarren SK, Clausen C. Application of the fetal alcohol syndrome facial photographic screening tool in a foster care population. *J Pediatr*. 2002;141:712-717.
15. Hymbaugh K, Miller LA, Druschel CM, et al. A multiple source methodology for the surveillance of fetal alcohol syndrome – the fetal alcohol syndrome surveillance network (FASSNet). *Teratology*. 2002;66:S41-S49.
16. CDC. Fetal alcohol syndrome—Alaska, Arizona, Colorado, and New York, 1995—1997. *MMWR*. 2002;51:433-435.
17. Alexander GR, Himes JH, Kaufman RB, Mor J, Kogan M. A United States national reference for fetal growth. *Obstet Gynecol*. 1996;87:163-168.
18. Alexander GR, Kogan MD, Himes JH, Mor JM, Goldenberg R. Racial differences in birthweight for gestational age and infant mortality in extremely-low-risk US populations. *Paediatr Perinat Epidemiol*. 1999;13:205-217.
19. Lubchenco LO, Hansman C, Boyd E. Intrauterine growth in length and head circumference as estimated from live births at gestational ages from 26 to 42 weeks. *Pediatrics*. 1966;37:403-408.
20. Astley SA, Clarren SK. A case definition and photographic screening tool for the facial phenotype of fetal alcohol syndrome. *J Pediatr*. 1996;129:33-41.
21. Hall JG, Froster-Iskenius UG, Allenson JE. *Handbook of Normal Physical Measurements*. Oxford, NY: Oxford University Press; 1989.
22. Lohman TG, Roche AF, Martorell R (eds). *Anthropometric Standardization Reference Manual*. Champaign, IL: Human Kinetics Books; 1988.
23. Frankenburg WK, Dodds J, Archer P, et al. *The Denver II Training Manual*. Denver, CO: Denver Developmental Materials, Inc.; 1992.
24. Mathis MP, Lavoie MA, Hadley C, Toomey K. Birth certificates as a source for fetal alcohol syndrome case ascertainment—Georgia, 1989-1992. *MMWR*. 1995;44:251-253.
25. Kirby RS. Co-occurrence of developmental disabilities with birth defects. *Ment Retard Dev Disabil Res Rev*. 2002;8:182-187.