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Issues in Estimating the Prevalence of Fetal Alcohol Syndrome: Examination of 2 Counties in New York State

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ABSTRACT

OBJECTIVE. Two demographically similar counties included in the New York Fetal Alcohol Syndrome Surveillance Network had very different prevalence rates. This study examined the components of the surveillance in an attempt to discover the reasons for this discrepancy.

METHODS. Erie County and Monroe County were the 2 most populous counties included in the New York Fetal Alcohol Syndrome Surveillance Network. Erie County includes Buffalo, the second largest city in New York State, and Monroe County includes Rochester, the third largest city. Multiple sources of ascertainment included birth defect surveillance systems, genetic clinics, and early intervention programs. The case definition was based on the Institute of Medicine criteria of an abnormality in each of the following 3 areas: facial features, central nervous system, and growth.

RESULTS. Children born in Erie County or Monroe County between 1995 and 1999 were included. The fetal alcohol syndrome prevalence rates in these 2 counties were 0.90 cases per 1000 births and 0.21 cases per 1000 births, respectively. The 2 counties were demographically similar and had similar rates of binge drinking among women of childbearing age. There was less participation in the surveillance system by sources in Monroe County. Erie County had a very active clinician with a specialized fetal alcohol syndrome clinic.

CONCLUSIONS. The participation of clinicians in one county, especially one with expertise in fetal alcohol syndrome, was the most likely explanation for the differences in prevalence rates between the counties.

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Dr Druschel and Ms Fox had full access to all of the data in the study and take responsibility for the integrity of the data and the accuracy of the data analysis.

Key Words

fetal alcohol syndrome, prevalence, birth defects, statistical estimation, data collections, facial features, diagnostic criteria

Abbreviations

FAS—fetal alcohol syndrome
FASSNet—Fetal Alcohol Syndrome Surveillance Network
IOM—Institute of Medicine
CNS—central nervous system

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WIDE VARIATIONS in fetal alcohol syndrome (FAS) prevalence rates have been found in various studies. The results range from ~0.2 cases per 1000 newborns in passive systems to ~1.0 case per 1000 births in some clinic-based studies^{1,2} and 3.0 cases per 1000 children in a study of first-grade children in one county in Washington State.³ Even higher prevalence rates have been found in studies of specific communities. A prevalence rate of 65.2 to 74.5 cases per 1000 was found for school-aged children in a wine-growing community in South Africa.⁴

There are many factors that may account for this variation. Disparities in FAS prevalence rates based on demographic features such as socioeconomic status and race/ethnicity are well known.^{2,5} Abel⁶ found that prevalence rates were generally ~10 times higher for black individuals. Variation could result from different levels of alcohol intake or different drinking patterns, but differences in ascertainment methods or age groups included could also be contributing factors.^{2,5,7} FAS is best diagnosed in children 2 to 11 years of age; facial features may be difficult to distinguish in some newborns and become easier to recognize over time.^{4,8–12} Newborns may not be well examined for FAS.^{11,13} Therefore, prevalence rates may seem lower if only newborns or younger children are included.³ Other reasons for disparities include differences in case definitions, the expertise and willingness of clinicians to make the diagnosis, and diligence in searching for cases.^{8,9,13}

The Institute of Medicine (IOM) report compared surveillance methods for FAS, pointing out that active surveillance systems find more cases and the diagnoses are “more likely to be valid and reliable.”¹⁴ The report pointed out that the difficulties with active surveillance include the expense and, because of population selection, resulting prevalence rates may not be generalizable.¹⁴ Alaska performed multiple-source FAS surveillance and found that 65% of the cases were identified through active screening and referral programs to specialty diagnostic clinics, “[illustrating] the magnitude of under-reporting by all of the passive reporting data systems.”¹⁵ Through active screening, Clarren et al³ found a prevalence rate of 3.1 cases per 1000 children, one of the higher prevalence rates found for a general population in the United States. May and Gossage² demonstrated similarly that “active case ascertainment generally yields the highest number of cases and rates of FAS for a particular population.”

The basic criteria for diagnosis of FAS were originally laid out by Jones and Smith¹⁶ and, with some modification, are still in use.¹⁴ In addition to maternal alcohol use, they include criteria in 3 areas, namely, central nervous system (CNS) abnormalities, such as microcephaly or developmental delays; growth retardation (prenatal and postnatal); and specific abnormal facial features, such as short palpebral fissures, flat philtrum,

or thin upper lip. The facial features are the most specific factors in the diagnosis that differentiate FAS from other syndromes or conditions that result in growth retardation. However, each feature “represents a minor anomaly or variant of normal,”¹⁰ and features may be overlooked unless clinicians search specifically for them.

In 1997, the Centers for Disease Control and Prevention provided funding for 5 states (Alaska, Arizona, Colorado, New York, and Wisconsin) to develop the Fetal Alcohol Syndrome Surveillance Network (FASSNet). This project was described in detail elsewhere.¹⁷ Results from 4 of the states were published, and some variation in prevalence rates was demonstrated.¹⁸ In examining regional data from the New York FASSNet system, differences were noted in the prevalence of FAS in 2 of the largest counties in the surveillance region. In this article, possible reasons for the differences, including demographic factors, alcohol use, and sources for case ascertainment, are examined.

METHODS

FASSNet

The FASSNet project objectives were to enhance or to develop a multiple-source surveillance system, to generate population-based surveillance data, to establish relationships with diagnostic and service programs, to evaluate the completeness of the surveillance system method, and to implement provider training. Four of the participating sites (Alaska, Arizona, Colorado, and New York) used the same general methods, including a common case definition, a common data collection form, and multiple sources for identification of cases. These sources included hospitals, birth defect registries, genetic clinics, developmental clinics, and early intervention programs. No direct examinations of the children were performed for the surveillance. All information was abstracted from medical records with the standardized form.

The case definition for FASSNet surveillance was based on the IOM criteria,¹⁴ which were adapted for the project by a FASSNet committee of experts, including a dysmorphologist, a geneticist, and a developmental psychologist. The IOM criteria provide no specific parameters for each of the criteria, although there has been a recent effort to do so.¹⁹ The criteria were quantified by the FASSNet committee to be uniform and specific for use across sites. The FASSNet surveillance case definition criteria are described in Table 1.

To meet the criteria for growth retardation (ie, weight, height, or weight for height), a child's growth measures (at any age) needed to be ≤10th percentile for age. Standard growth curves for both intrauterine and postnatal growth were selected for uniform evaluation of growth.¹⁷ To meet the criteria for CNS abnormality, either structural (ie, head circumference) or functional

TABLE 1 FASSNet Surveillance Case Definition Categories

Case Definition Category	Phenotype Positive		
	Facial Features	CNS	Growth
Confirmed FAS phenotype, with or without maternal alcohol exposure ^a	Abnormal facial features consistent with FAS, as reported by physician Two of the following: short palpebral fissures, abnormal philtrum, or thin upper lip	Fronto-occipital circumference of ≤ 10 th percentile at birth for any age Standardized measure of intellectual function ≤ 1 SD below the mean Standardized measure of developmental delay ≤ 1 SD below the mean Developmental delay or mental retardation diagnosed by qualified examiner (eg, psychologist or physician) Attention-deficit disorder diagnosed by qualified evaluator	Intrauterine weight or height corrected for gestational age ≤ 10 th percentile Postnatal weight or height of ≤ 10 th percentile for age Postnatal weight for height of ≤ 10 th percentile
Probable FAS phenotype, with or without maternal alcohol exposure ^a	Required same as for confirmed category	Must meet either CNS or growth criteria as outlined for confirmed category	

^a Maternal alcohol exposure indicates documentation in the records of some level of maternal alcohol use during the index pregnancy.

(ie, mental retardation, developmental delay, or attention-deficit disorder) criteria were used. A standard head circumference growth curve was chosen for evaluation of both birth and postnatal head circumference. A child whose head circumference was ≤ 10 th percentile for age met the CNS structural criteria for FAS. To measure functional delays (ie, developmental delay, mental retardation, and other intellectual deficits), a list of acceptable standardized tests was developed in consultation with developmental psychologists. Tests scores of ≥ 1 SD below the mean for a child of similar age were considered to meet the criteria for CNS abnormality. Alternatively, a diagnosis of developmental delay, mental retardation, or attention-deficit disorder by a qualified clinician met the CNS criteria. More than 95% of the New York FASSNet case subjects who met the CNS criteria did so on the basis of a head circumference of ≤ 10 th percentile. To meet the facial dysmorphic criteria, 3 facial anomalies (ie, short palpebral fissures, abnormal philtrum, and thin upper lip) were chosen as case definition criteria, on the basis of the characteristics that have been shown to discriminate best between children who have FAS and those who do not.²⁰

Case status was determined electronically by using a computer algorithm to evaluate all of the data abstracted for the child. A case was classified as “definite” if the criteria in each of the 3 categories were met. A “probable” case classification met the criteria for facial features and either growth criteria or CNS criteria.

New York FASSNet

The surveillance area for the New York FASSNet consisted of 9 counties in western New York, which in-

cluded both urban and rural areas. The 2 largest and most urban counties are Erie County and Monroe County. Erie County includes Buffalo, the second largest city in New York State, and Monroe County includes Rochester, the third largest city. Case ascertainment began in 1998 and was retrospective to birth year 1995. Birth certificates were used to define maternal residence at birth and other demographic variables. Birth years 1995 through 1999 were included, and definite and probable cases were combined and used to calculate the FASSNet prevalence. Maternal alcohol use was not used as a criterion, although $>90\%$ of the children confirmed as having FAS had a notation of maternal alcohol use in their medical records.

In the New York FASSNet, cases were ascertained from multiple sources, including genetic clinics, early intervention programs, hospital discharge data, birth defect surveillance systems, developmental clinics, vital records, and other clinics, including a special clinic for mothers with substance abuse problems and their children. Some sources were directly accessible by FASSNet staff members; these included hospital discharge data, birth certificate data, and data from 2 birth defect surveillance systems (the statewide Congenital Malformations Registry, which relies on hospital reporting, and a regional surveillance system in which staff members review records of children identified with birth defects). Other sources, such as genetic or developmental clinics and early intervention programs, required that potential cases be identified to FASSNet staff members. Although these sources were visited regularly by FASSNet staff members, the clinics varied in how they identified cases.

Some had computerized records, whereas others kept lists as cases were identified.

Frequently, all of the information needed to meet the case definition was not available from the ascertainment source; therefore, a child would be evaluated with other sources, to obtain more information for confirmation. For each child, the FASSNet database tracked the initial source(s) of ascertainment and any follow-up sources. Therefore, it can be determined which sources identified children and which provided the specific information for the case definition items, such as facial features.

RESULTS

FASSNet Prevalence Rates According to County

All prevalence rates presented were calculated per 1000 live births and were based on maternal residence at birth in Erie County or Monroe County. The prevalence rates in Erie County and Monroe County were 0.90 and 0.21 cases per 1000 live births, respectively. FAS prevalence rates according to race/ethnicity for 1995 to 1999 are presented in Table 2. The prevalence rate among white individuals in Monroe County was approximately one half of that in Erie County; the prevalence rate among black individuals in Monroe County was slightly more than 10% of that in Erie County. In Erie County, the prevalence rate among black individuals was 10 times that among white individuals; in Monroe County, the prevalence rate among black individuals was twice that among white individuals.

Demographic Factors

For the study period, Erie County had ~18% more live births than Monroe County (58 435 vs 47 901). Maternal and infant characteristics are shown in Table 3. In general, the characteristics were very similar in the 2 counties; however, Monroe County had a slightly higher percentage of minority births.

The New York Behavioral Risk Factor Surveillance System survey provided county-level data on binge drinking (defined as ≥ 5 drinks at 1 occasion) for the years 1995, 1997, and 1999. Women in Monroe County reported somewhat higher levels of binge drinking than did women in Erie County (16.6% vs 12.3%). Rates for both counties were higher than reported rates of binge drinking for the state as a whole for the same time

period, which ranged from 6.3% to 8.2%. The small numbers available did not allow more-detailed analysis.

Ascertainment Sources for New York FASSNet

To gain a better understanding of how children were identified and how the criteria for the case definition were met, the ascertainment sources for each county were examined. This evaluation included not only children confirmed as having FAS but also all cases ascertained by the FASSNet system, both confirmed and unconfirmed. The comparison of initial ascertainment sources according to county is presented in Table 4. The totals for the sources are more than the total number of children in each category because a case might have been ascertained independently by >1 source.

All Identified Children

Overall, as illustrated in Table 4, Erie County had more than twice as many initial ascertainment abstractions in the system as Monroe County (542 and 222, respectively) and twice as many children (420 and 208, respectively). Sources directly accessible by FASSNet staff members accounted for 89% of Monroe County ascertainment, compared with 66% of Erie County referrals. In Erie County, 75 children (18% of total children) were referred from genetic clinics, compared with 5 children (2.5% of total children) in Monroe County. The percentages of children identified by the system who were black were similar (67.9% in Erie County and 63.0% in Monroe County).

Children Confirmed as Having FAS

Comparisons of children confirmed as having FAS were more difficult, because the numbers in Monroe County were small. As shown in Table 4, 13% of the children from Erie County who were identified through FASSNet were confirmed as meeting the FAS case definition (53 of 420 children), whereas 5% from Monroe County were confirmed (10 of 208 children). In Erie County, many of the children confirmed as having FAS (36 of 53 children; 68%) were identified initially through genetic clinics, compared with only 20% (2 of 10 children) in Monroe County identified through that source. In both counties, approximately one half of the children referred from genetic clinics met the FAS case definition (in Erie County: 36 of 75 children; 48%; in Monroe County: 2 of 5 children; 40%). Children who were black accounted for 68% of confirmed cases in Erie County and 40% of confirmed cases in Monroe County.

Identification of Facial Features

Because the description of facial features, which depends on active clinical recognition, is the critical element in the surveillance case definition, the sources providing information on abnormal facial features used for case definition, regardless of FAS case status, were examined.

TABLE 2 FAS Prevalence According to Race/Ethnicity in Erie and Monroe Counties, in New York FASSNet (1995–1999)

	FAS Prevalence, Cases per 1000 Live Births (n)	
	White, Non-Hispanic	Black
Erie	0.34 (15)	3.31 (36)
Monroe	0.18 (6)	0.40 (4)

Values were based on maternal residence at birth.

TABLE 3 Maternal and Infant Characteristics of Live Births in Erie and Monroe Counties, in New York FASSNet (1995–1999)

Characteristics	Erie County (n = 58 435)	Monroe County (n = 47 901)
Maternal age, mean (range), y	28.1 (12–55)	28.2 (11–49)
Mother's race/ethnicity, n (%)		
White, non-Hispanic	44 200 (75.6)	32 807 (68.5)
Black	10 731 (18.4)	9870 (20.6)
Hispanic	1846 (3.2)	3357 (7.0)
Asian/Pacific Islander	1092 (1.9)	1500 (3.1)
Other/unknown	200 (0.3)	281 (0.6)
American Native	366 (0.6)	86 (0.2)
Mother's educational level, n (%)		
Never completed high school	7418 (12.7)	7798 (16.3)
Only completed high school	16 749 (28.7)	12 860 (26.8)
Some college	17 079 (29.2)	11 235 (23.5)
College graduate or more	15 837 (27.1)	15 149 (31.6)
Missing	1352 (2.3)	859 (1.8)
Primary payer for birth, n (%)		
Medicaid	15 314 (26.2)	12 756 (26.6)
Health maintenance organization	34 574 (59.2)	30 964 (64.6)
Private insurance	7725 (13.2)	3712 (7.7)
Self-pay	601 (1.0)	425 (0.9)
Missing	221 (0.4)	44 (0.1)
Prenatal care, n (%)		
Yes	55 627 (95.2)	46 183 (96.4)
No	1472 (2.5)	1543 (3.2)
Unknown	1336 (2.3)	175 (0.4)
Birth weight, n (%)		
<1500 g	709 (1.2)	541 (1.1)
1500–2499 g	3615 (6.2)	2840 (5.9)
≥2500 g	54 099 (92.6)	44 519 (92.9)
Missing	12 (0.0)	1 (0.0)

TABLE 4 Initial Ascertainment Source for All Children and Children Confirmed as Having FAS in Erie and Monroe Counties, in New York FASSNet (1995–1999)

Source	No. (%)			
	All Children		Children With Confirmed FAS	
	Erie	Monroe	Erie	Monroe
Directly accessible to FASSNet staff				
Hospital discharge records	248 (45.7)	155 (69.8)	32 (29.1)	3 (27.3)
Birth defect surveillance programs	36 (6.6)	5 (2.2)	21 (19.1)	1 (9.0)
Birth certificates ^a	73 (13.5)	37 (16.7)	8 (7.3)	0 (0.0)
Source-provided cases				
Genetic clinic	75 (13.9)	5 (2.3)	36 (32.7)	2 (18.2)
Early intervention program	65 (12.0)	12 (5.4)	9 (8.2)	2 (18.2)
Other ^b	45 (8.3)	8 (3.6)	4 (3.6)	3 (27.3)
Total initial abstractions ^c	542 (100)	222 (100)	110 (100)	11 (100)
Total black initial abstractions	384 (70.8)	140 (63.1)	79 (71.8)	4 (36.4)
Total children	420	208	53	10
Total black children	285 (67.9)	131 (63.0)	36 (67.9)	4 (40.0)

^a Birth certificates considered were those with FAS reported, with the mother reported to drink ≥10 alcoholic drinks per week during pregnancy, or for the sibling of a child with FAS.

^b Other indicates developmental clinics, other clinics, physicians, and other sources.

^c The total is greater than the number of children because some children were identified independently by >1 source.

More children in Erie County had documentation on facial features in their records, compared with Monroe County; genetic clinics were a major source of informa-

tion (Table 5). Facial feature information found through hospital discharge sources originated frequently from inpatient genetic consultations.

TABLE 5 Sources of Data on Abnormal Facial Features in Erie and Monroe Counties, in New York FASSNet (1995–1999)

Source	No. (%)	
	Erie	Monroe
Directly accessible to FASSNet staff		
Hospital discharge records	42 (37)	6 (30)
Birth defect surveillance programs	1 (1)	0 (0)
Source-provided cases		
Genetic clinic	43 (38)	3 (15)
Other ^a	27 (24)	10 (50)
Early intervention	0 (0)	1 (5)
Total children	113 (100)	20 (100)

For a child determined to have positive facial features, regardless of FAS case status, 1 of the following criteria should be positive: abnormal facial features consistent with FAS, short palpebral fissures (short palpebral fissures, small palpebral fissures, or microphthalmia/small eyes), palpebral fissures $\leq 10\%$ for age, thin/narrow upper lip (thin/narrow upper lip/vermillion), or abnormal philtrum (abnormal, absent/underdeveloped, long, smooth, hypoplastic, or flat).

^a Other indicates developmental clinics, other clinics, physicians, or other sources.

DISCUSSION

In the New York FASSNet, 2 neighboring urban counties, which are demographically similar, were found to have very different FAS prevalence rates. Given the slightly higher rates of minority births and binge drinking in Monroe County, it might be expected that the FAS prevalence rate would be similar to, if not higher than, that in Erie County. Instead, the prevalence rate was less than one third that found in Erie County. This discordance illustrates some of the problems and challenges of FAS surveillance.

Although the county-level Behavioral Risk Factor Surveillance System data on binge drinking were not specific to pregnant women, the data do demonstrate that reported binge drinking among women of child-bearing age is somewhat higher in Monroe County. Heavy alcohol use before pregnancy is highly predictive of continued use in pregnancy.²¹ Even if a woman reduces her drinking at pregnancy recognition, the fetus can be affected very early in pregnancy, before the woman realizes she is pregnant.^{22,23} It is unlikely that the time of pregnancy recognition varied greatly between Monroe County and Erie County.

Overall, in Monroe County, fewer children were identified, a lower percentage of the children were confirmed as having FAS, and a lower percentage of black children were confirmed as having FAS. There was less “active” participation by Monroe County sources, especially by the clinical sources. Most cases were ascertained from sources accessed directly by FASSNet staff members, and few were ascertained from diagnostic clinics. Diagnostic clinics have been an important source of cases in other surveillance systems.^{2,15} In Erie County, genetic clinics were an important source of case ascertainment and information on facial features; in Monroe County, the numbers ascertained and confirmed through genetic clinics were lower. The lower percentage confirmed in Monroe County, compared with Erie County, may indi-

cate difficulty in obtaining the needed documentation of facial features. The geneticists in Monroe County have expressed a willingness to make the diagnosis, but clinicians in that county may be more reluctant to refer patients to geneticists. Geneticists, through both clinic records and inpatient consultations, provided most of the facial feature information. The limited information on facial features made it more difficult for the FASSNet system to identify or to confirm potential cases in Monroe County. In addition, most of the FASSNet sources are biased toward the newborn period and younger children, but older children could be identified through genetic clinics. Thirty percent of New York FASSNet cases were diagnosed after 2 years of age, and the majority of them were ascertained through genetic clinics.

For a diagnosis of FAS, it is important that clinicians think about the possibility of FAS and how to identify the facial features. For surveillance purposes, the facial features need to be recorded in the medical record. FAS is approximately as common as Down syndrome (~ 1.3 cases per 1000).²⁴ However, a diagnosis of FAS is more complicated. There is no diagnostic test specific for FAS. A child may need to be monitored after birth, because FAS facial features may not be present at birth and growth and CNS deficiencies may not be detected until the child is older.^{4,8–12}

It is important that clinicians understand that there are reasons to make a diagnosis other than improving FAS surveillance. There can be direct benefits to the child and the family. Although some clinicians do not want to “label” the child, they overlook an opportunity to intervene with the family. In addition, studies have shown that children who are diagnosed earlier fare better, because they may receive specific interventions.²⁵ The study of first-grade children in Washington found that, of the children who screened positive for FAS, only 1 of 7 had been diagnosed previously and only one half were receiving special services.³ There may be many children with undiagnosed FAS in communities. Not all children with FAS are mentally retarded, but they have serious learning problems that may not be recognized; therefore, the children may not receive appropriate interventions and may experience failure in school. There needs to be an awareness that, although these children have problems, diagnosis allows for the use of programs that can help the children be more successful in life.^{26,27}

Much of the success of FASSNet in Erie County seems to be attributed to the involvement of a knowledgeable clinician, a dysmorphologist. He has expertise in FAS and was involved actively with FASSNet, regularly reporting cases to the FASSNet staff. He documented facial features (both normal and abnormal) routinely in the record. He interacted with several of the FASSNet sources in Erie County and worked to educate health care providers in Erie County to be aware of the diagnosis, as well as the procedures for making the diagnosis.

CONCLUSIONS

The prevalence of FAS is highly variable among populations. It is frequently difficult to know what accounts for these differences. In this article, the components of a surveillance project were examined in detail, in an attempt to discover the reasons behind the large differences in FAS prevalence between 2 neighboring and demographically similar counties. The willingness and cooperation of clinicians in one county, especially one with expertise in FAS, represent the most likely explanation.

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