

A Multiple Source Methodology for the Surveillance of Fetal Alcohol Syndrome—The Fetal Alcohol Syndrome Surveillance Network (FASSNet)

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INTRODUCTION

Describing the prevalence of fetal alcohol syndrome (FAS) in the United States and monitoring the trends in its occurrence remain elusive undertakings. Although FAS has been recognized as a public health problem and preventable birth defect since 1973, no surveillance system specifically designed to monitor its occurrence existed until the development of the U.S. Fetal Alcohol Syndrome Surveillance Network (FASSNet) in 1997. Measuring the occurrence (incidence or prevalence) of FAS and monitoring trends in its rates in population subgroups are necessary to understand and identify vulnerable populations; target prevention and treatment resources; and evaluate the strengths and limitations of various prevention, intervention, and treatment strategies.

Prevalence rates for FAS reported in the literature vary widely from 0.2 to 120.0 per 1,000 live births (CDC, '95b; Robinson et al., '87). Estimates from case registries, some of which are population-based, also vary (Table 1). These variations reflect differences in case definitions, methods of case ascertainment, and populations examined. Developing an epidemiologic surveillance system for FAS presents unique challenges that cannot be met by traditional surveillance systems designed to monitor infectious diseases or even by current birth defects monitoring programs (BDMPs). Most communicable, infectious, and chronic disease surveillance models rely primarily on notifiable diseases or required reporting from health-care providers, laboratories, hospitals, or vital records. Most

BDMPs focus primarily on the first year of life for case finding and diagnosis. FAS is not easily identified, and there is no definitive laboratory test or single characteristic (beyond the cluster of facial dysmorphic features, which is often recognizable only to expert clinicians) specific to the diagnosis of FAS. A FAS diagnosis is based primarily on clinical examination and the application of diagnostic criteria in each of the following three categories: 1) prenatal or postnatal growth retardation; 2) central nervous system (CNS) abnormalities, which may manifest as developmental delays in childhood; and 3) characteristic abnormal facial features (Aase, '94; Institute of Medicine, '96). To further complicate case finding and diagnosis, application of clinical criteria requires expertise in recognizing dysmorphic features and differentiating the condition from other syndromes and malformations that may manifest similar features. FAS becomes easier to diagnose with increasing age of the child, at least until puberty, because some of the cardinal facial features and CNS abnormalities are not apparent during the first 1–2 years of life (Aase, '94; Jones, '99; Stoler and Holmes, '99). Finally, even when the clinical manifestations are present and recognized, some physicians are reluctant

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TABLE 1. Registry-based studies reporting a prevalence of fetal alcohol syndrome

Method	Population	Age at diagnosis	Rate per 1000 children by race or year of birth	
BDMP ¹ (Chavez et al., '88)	United States 1981–1986	Newborn	0.6 0.08 2.99 0.03 0.09	Blacks Hispanics American Indians Asians Whites
BDMP (Chavez et al., '88; CDC, '93b; CDC, '95a)	United States 1979–1993	Newborn	0.1 0.37 0.67 0.22	1979 1992 1993 1979–1993
BDMP (Cordero et al., '94)	United States 1992	Newborn	0.52	1992
MACDP ² (CDC, '95b; Cordero et al., '94)	5-County Metropolitan Atlanta 1989–1992	Birth–1 year	0.23 0.33	1989–1992 1992
Linked MACDP and MADDSP ³ (CDC, '97)	5-County Metropolitan Atlanta 1981–1989	Birth–10 years	0.1 0.16	1981–1989 observed 1981–1989 projected
Multiple Sources (Egeland et al., '98)	Alaska 1977–1992	3–18 years	0.8	1977–1992

¹Birth Defects Monitoring Program.

²Metropolitan Atlanta Congenital Defects Program.

³Metropolitan Atlanta Developmental Disabilities Surveillance Program.

to make the diagnosis of FAS because of the potential social stigma associated with the diagnosis and the perceived lack of resources and services to support treatment for the child and family (Morse et al., '92).

The challenges to developing a simple and efficient state-based surveillance system for FAS, one that can derive national prevalence rates and track trends in the occurrence of FAS in the population, have been described elsewhere (Cordero et al., '94). Surveillance of FAS cannot depend on any one service delivery system or other single data source for complete case ascertainment. Lessons from previous efforts indicate that a multiple-source FAS surveillance method yields more promising data (Egeland et al., '98). A multiple-source method may include such data sources as birth defects monitoring programs, developmental disabilities or special needs registries, early intervention programs, hospital discharge data, Medicaid data, vital statistics, private providers, special diagnostic or genetic clinics, and other population-based systems (Miller et al., '95; Egeland et al., '98; CDC, '93a). The theoretical basis for a multiple-source approach to case finding is that, because of the nature of the health and developmental problems associated with the condition, children with FAS are likely to encounter one or more of these providers for services at some point in early childhood. Often, however, the diagnosis of FAS is not made at the time of such encounters. In addition, documentation in patients' medical or other source records is often insufficient to support a clinical diagnosis of FAS. Thus, ensuring more comprehensive documentation of the diagnostic features in medical records and educating and training providers to recognize FAS-

1. Enhance or develop a multiple-source surveillance system;
2. Generate population-based surveillance data;
3. Establish relationships with diagnostic and service programs;
4. Evaluate the completeness of the surveillance system methodology;
5. Implement provider training and education on FAS.

Fig. 1. Major goals of the state-based Fetal Alcohol Syndrome Surveillance Network (FASSNet)

affected children and refer them for diagnosis would improve the success of the multiple-source methodology.

In 1997, the Centers for Disease Control and Prevention (CDC) announced the availability of funds for 5-year, cooperative agreement programs for states to establish or enhance population-based FAS surveillance (DHHS, Federal Register, '97). States were asked to develop a multiple-source surveillance system by establishing relationships with diagnostic and service programs to generate FAS prevalence rates in defined geographic areas (Fig. 1). Five states—Alaska, Arizona, Colorado, New York, and Wisconsin—were awarded cooperative agreements and now make up FASSNet. Four of these states (Alaska, Arizona, Colorado, New York) follow the multiple-source approach described in this report; Wisconsin's alternative meth-

TABLE 2. The geographic region covered by FASSNet and the average annual live births, by state 1995–1997

State	Geographic region	Average annual live births	
		Number	Percentage of state population
Alaska	Statewide	10,073	100%
Arizona	Statewide	74,348	100%
Colorado	Six counties in the Denver-Boulder Metropolitan Statistical Area	31,712	57.0%
New York	Nine-county region in western New York	29,597	12.0%
Total Four-State Area		145,730	

odology will be described elsewhere. In this article, we describe the methods used by FASSNet, including the multiple-source methodology, surveillance case definition, data collection variables, and the record abstraction process. Prevalence rates for FAS, applicability of this methodology to larger-scale FAS surveillance, and issues concerning staffing and budget requirements will be addressed in future papers.

METHODS

Population

The four-state FASSNet catchment area includes approximately 145,730 average annual live births for the initial target birth years 1995–1997, representing all live births in the states of Alaska and Arizona and select regions of Colorado and New York (Table 2). The population data used for determining prevalence rates are estimated from each individual state’s vital statistics department. The racial composition is diverse. Approximately 62% of the children in the four-state study area are white (non-Hispanic), 6% are black, 6% are American Indian or Alaska Native, 2% are Asian or Pacific-Islander, and <1% are classified as “other” or “unknown” race. Approximately 23% are Hispanic.

To be included in FASSNet, a child must have been born to a resident of the study area on or after January 1, 1995. To balance the need for a timely reporting system with the need to allow ample opportunity for a child with FAS to enter the health-care delivery system, we initially focused case finding and abstraction activities on children in the birth years 1995–1997. This allows a minimum of two years for children with FAS to come to the attention of the health-care delivery systems and expands the window of case finding beyond that of traditional birth defects monitoring programs.

Surveillance case definition

The FASSNet surveillance case definition was developed by a committee of physicians (including dysmorphologists), psychologists familiar with FAS, experts in clinical anthropometry, epidemiologists, and other experts in public health surveillance of FAS and other birth defects. This definition was developed before beginning record abstraction. Initially, committee members examined the clinical case definition for FAS presented in the 1996 Institute of Medicine (IOM) report

(IOM, '96) and used this as a framework for developing the FASSNet surveillance case definition. Though the general clinical criteria for FAS (i.e., growth retardation, CNS abnormalities, and characteristic abnormal facial features) are well established (IOM '96), the specific items that may be found in a medical or psychological record that fulfill the criteria are not. Therefore, several general guidelines were followed when determining which specific items would fulfill the criteria for the FASSNet surveillance case definition. First, for items that could be quantified, the use of a measure was selected over a qualitative description. For example, one criterion in the IOM report was “decreased cranial size at birth” which was operationalized as an actual head circumference measurement $\leq 10^{\text{th}}$ centile at birth or any age. Second, to remain consistent across sites and err on the side of inclusion, in cases for which conflicting information was present, information that met the case definition was selected over information that did not. For example, if one qualified examiner (an examiner with the appropriate license or degree and deemed qualified in the field of interest) diagnosed a child as having developmental delay and another qualified examiner documented no developmental delay, the positive diagnosis was recorded (FASSNet Abstractor’s Manual, '00). The source and expertise of the examiner were also considered when determining the acceptability of conflicting information. For example, head circumference measurements made by a physician in a genetics clinic would be accepted over a head circumference measurement recorded in an early intervention program record by an unknown examiner.

To meet the facial dysmorphic criteria, three facial anomalies (i.e., short palpebral fissures, abnormal philtrum, and thin upper lip) were chosen based on the characteristics that have been shown to best discriminate between children who have FAS and those who do not (Hymbaugh et al., '93; Astley and Clarren, '95). In addition, a broader criterion of a physician diagnosis of abnormal facial characteristics consistent with FAS was included.

To meet the criteria of CNS abnormality, either structural (i.e., head circumference) or functional (i.e., mental retardation, developmental delay, or attention deficit disorder) criteria were included. A standard head circumference growth curve was chosen for both birth and postnatal head circumference. (See section on

DIAGNOSTIC CATEGORY	PHENOTYPE POSITIVE		
	FACE	CENTRAL NERVOUS SYSTEM	GROWTH
Confirmed FAS Phenotype <u>With or Without Documentation*</u> In utero Alcohol Exposure	Abnormal facial features consistent with FAS as reported by a physician Or Two of the following: - short palpebral fissures - abnormal philtrum - thin upper lip	At least one structural or functional anomaly <u>STRUCTURAL</u> - Head Circumference \leq 10th centile at birth or any age Or <u>FUNCTIONAL</u> - Standardized measure of intellectual function \leq 1 std dev below the mean Or - Standardized measure of developmental delay \leq 1 std dev below the mean Or - Developmental delay or mental retardation diagnosed by a qualified examiner (e.g., psychologist and physician) Or - Attention-Deficit/Hyperactivity Disorder (ADHD) diagnosed by a qualified examiner	Growth delay indicated in at least one of the following: <u>INTRAUTERINE</u> - weight or height corrected for gestational age \leq 10 th centile Or <u>POSTNATAL</u> - weight or height \leq 10 th centile for age Or - weight for height \leq 10 th centile
Probable FAS Phenotype <u>With or Without Documentation*</u> In utero Alcohol Exposure	Required Same as Confirmed FAS Phenotype above	Must meet <u>either</u> CNS or GROWTH criteria as outlined in the Confirmed FAS Phenotype above	
Suspect	All children referred into the surveillance system including all children with ICD-9 codes 760.71, provider referrals, children identified by abstractors who meet predetermined criteria from the specific referral source, newborn nursery logs, etc.		

* Determined from the availability of documentation in the records of some level of maternal alcohol use during the index pregnancy.

Fig. 2. FASSNet surveillance case-definition categories

standardization of growth measures data.) A child whose head circumference is \leq 10th centile for age (at any age) meets the CNS structural criteria. To measure functional delays (e.g., developmental delay, mental retardation, and other intellectual deficits), a list of acceptable standardized tests was developed in consultation with developmental psychologists. Tests were considered to meet the criteria for CNS abnormality if the score was at least one standard deviation below the mean for a child at any age. In addition to test results, a diagnosis of developmental delay, mental retardation, or attention-deficit/hyperactivity disorder (ADHD) by a qualified clinician met the CNS criteria.

To meet the criteria of growth retardation (i.e., weight, height, or weight for height), any child whose growth measures fell \leq 10th centile for age (at any age) was included. Standard growth curves for both intra-

uterine and postnatal growth were selected. (See section on standardizing the growth measures.)

To indicate positive maternal alcohol exposure, any medical record documentation of alcohol use by the mother during the index pregnancy (other than hearsay reported by a third party) met the criteria. Though substantial alcohol use is necessary to cause full FAS, information in medical records is usually insufficient to determine the level of use. However, this information is collected when available.

Initially, four surveillance case-definition categories will be used (Fig. 2), including 1) confirmed FAS phenotype **with** maternal alcohol exposure, 2) confirmed FAS phenotype **without** maternal alcohol exposure, 3) probable FAS phenotype **with** maternal alcohol exposure, and 4) probable FAS phenotype **without** maternal alcohol exposure. A fifth category, "suspect," is used

TABLE 3. Number of sources, by major source type for each of the FASSNet states

Sources	Alaska	Arizona	Colorado	New York	TOTAL
Hospitals	18	49	19	28	114
Specialty Clinics (e.g., Genetic and Developmental)	3	3	6	4	16
Private Physician Group	4			4	8
Early Intervention	1			8	9
Birth Defects Surveillance Program	1	1	1	2	5
Birth Certificates	1	1	1	1	4
Other Vital Records	1		1		2
Medicaid	1		1		2
Hospital Discharge Data		1	1	1	3
Other	6	1	2	9	18
TOTAL	36	56	32	57	181

for all other children referred to the system. Information about maternal alcohol use during the index pregnancy is the only factor that differentiates the two “confirmed FAS phenotype” and the two “probable FAS phenotype” categories. To be included in either confirmed category 1 or 2, the child must manifest abnormalities in each of the three areas described earlier (i.e., facial dysmorphism, central nervous system abnormality, and growth retardation). In the probable categories 3 and 4, the child must manifest the same facial characteristics as in the confirmed FAS phenotype but is only required to meet one of the other criteria—CNS or growth criteria. All other children identified (see case-finding procedures below) are categorized as “suspect.”

The case-definition categories may be modified pending further experience with their application. By the nature of the methodology we are using, some cases in the surveillance network undoubtedly fall in the categories of alcohol-related birth defects commonly referred to as alcohol-related neurodevelopmental disorders (ARND), alcohol-related birth defects (ARBD) or fetal alcohol effects (FAE). Analysis of the data on probable and suspect cases, along with maternal exposure, could reveal further insights into these categories. However, the intent of FASSNet was to develop a specific case definition of FAS for surveillance and use this as the indicator against which any suspected case was to be measured. Therefore, these data will ultimately provide a method for determining prevalence of the syndrome rather than a measure of the problem of alcohol-related birth defects in its broader context.

Description of the multiple data sources and case-finding procedures

The case-finding methodology used by FASSNet relies on both passive reporting and active record review of source records. All states use birth defects monitoring programs (BDMP) as a source for FAS case finding. In some FASSNet states (i.e., Alaska, Colorado, and New York), FAS is reportable by legislation (in Alaska and New York, FAS is one among numerous birth defects reported) or by board of health regulation (Colorado requires physician reporting) to the state BDMP. FAS reporting is not mandatory in Arizona.

Building on existing resources and developing collaborative relationship, each of the four FASSNet sites identified additional sources for conducting surveillance activities (Table 3). These additional sources include the following: 1) hospital discharge data sets; 2) genetics clinics; 3) developmental clinics; 4) neonatology clinics; 5) private physicians; 6) state Medicaid programs; 7) early intervention programs; 8) vital records (birth and death certificates); and 9) miscellaneous other sources. Although case-finding sources varied, each state included hospitals, genetics clinics, and developmental clinics as major source categories. In addition, each state can link to birth certificate data.

Various methods are used to identify potential FAS cases at the different sources. Examples of these methods include the following: 1) identification of children with an *ICD-9* code of 760.71 (newborn affected by alcohol via placenta or breast milk) or 742.1 (microcephaly) in a hospital discharge data set; 2) manual review of all records at a genetic clinic to identify children referred for prenatal alcohol exposure or maternal substance abuse during pregnancy; 3) identification of children whose birth weight is at or below the 10th percentile for gestational age from a specialty clinic; and 4) referral of children with the clinical features of FAS by private providers to the state FASSNet program (Fig. 3). Specific details on the case-finding procedures at each source were recorded at the beginning of the project. These data will be critical in the analysis of variation in case yields among sources within a state and for comparisons across states.

The FASSNet project staff continues to work with personnel at the multiple sources for their respective states to improve specific case finding and reporting methods to the surveillance system. They also provide training on FAS to personnel at the sources (see Provider Education and Training) and continue to work to identify other potential sources for case finding in their respective target regions. FASSNet, like other surveillance programs for birth defects and developmental disabilities, examines source records rather than children. It relies on clinicians to recognize, diagnose, and document the condition and to record relevant physical and psychological findings. Not all diagnostic criteria may be found in any single record or source of referral.

- Hospital Searches:
ICD-9 code searches for 760.71 (Noxious influences affecting fetus or newborn via placenta or breast milk, specifically alcohol; including fetal alcohol syndrome); ICD-9 codes 648.4 (conditions complicating pregnancy) with 303.0 (alcohol dependence) or 305.0 (non-dependent abuse of drugs); ICD-9 code 655.4 (suspected damage to fetus from disease [alcoholism] in mother).
- Specialty clinics (e.g., genetic, developmental, and neurodevelopmental):
Suspect cases may be identified by source staff from logs, registries, or identified as children with a history of prenatal alcohol exposure.
- Early intervention programs:
Suspect cases may be identified by source staff as children with suspected maternal substance abuse, developmental delay, or a history of positive toxicology screen.
- Private providers:
Source staff based on clinical examination and physician recognition of characteristics may identify suspect cases.

Fig. 3. Examples of case ascertainment procedures, by select source types

Therefore, project abstractors abstract records from the sources blind to previous abstractions. An advantage of this approach is the ability to evaluate which sources are the “best” for case finding and documentation of information critical to the case definition. This information will be useful in the future for evaluating the sources and in streamlining the surveillance system. A disadvantage is that duplicate information (and in some cases conflicting information) may be abstracted at different sources.

Data collection and standards

Data collection instrument. Using existing state-based BDMP data-collection instruments and other state FAS data-collection tools, the FASSNet data-collection instrument was developed by a data committee made up of experts from the CDC and each FASSNet state. The instrument was pilot-tested in the field with various sources. The CDC then developed an electronic version, the FASSNet Main Application, in Microsoft ACCESS 97 software. During development of the instrument, we considered ease of use in the field, the need for specific data consistent with the FASSNet goals, ongoing evaluation of the surveillance case definition, compatibility with existing systems (e.g., BDMPs), and ease of electronic transmittal of data to CDC, reporting, and data analysis. After field testing, the data elements were discussed relative to continued inclusion. Several data elements were dropped because they were not available for most children (e.g., information about the biologic father or mother’s social history). Non-identifiable data from the four states are pooled at CDC to examine data quality and assure consistency across the four sites and to perform pooled analyses.

Types of variables. Data variables included in the FASSNet application are summarized in the following sections.

Identifiers

Personal identifiers are collected and maintained only by the local FASSNet projects in accordance with state legislative mandates for birth defects surveil-

lance and confidentiality. Personal identifiers are collected to allow multiple records of one child to be matched.

Standard demographic variables

Standard demographic variables include child’s age and date of birth, parental ages at the index birth, and race/ethnicity. When available, information is also collected on other maternal drug exposures during the index pregnancy, prenatal history, maternal education, marital status, occupation, insurance coverage, public assistance, and other information indicative of socioeconomic status. When available, information is collected on other biologic children of the mother because these data may lead to the identification of additional cases of FAS.

Variables associated with the case-definition criteria

Detailed information is also collected for each of the case-definition criteria (Fig. 2). In addition to determining case-definition status, this allows for evaluation of the case-definition categories if the criteria are modified. The selection of the data elements for each of these criteria involved considerable discussion and input from experts in the fields of dysmorphology, psychology, pediatrics, anthropometry, and public health surveillance. For those elements used for case determination, a hierarchy of sources was developed so data from the source thought to provide the best quality information (e.g., genetics or neurodevelopmental clinics) would be selected to determine case status (FASSNet Manual, ’00).

Maternal alcohol exposure information

FASSNet originally intended to collect information on amount, frequency, and timing of maternal alcohol use. Data were first collected in a format similar to the one in the Behavioral Risk Factor Surveillance System (BRFSS) (CDC, ’98). However, during field testing, the information in records was found to be incompatible with the BRFSS categories designed for interview surveys. Thus, the FASSNet form was revised to capture the more frequent qualitative statements found in medical records as well as detailed information when available.

BIRTH MEASURES**Birth Weight** by gestational age and sex using:Alexander GR, Himes JH, Kaufman RB, Mor J, Kogan M. A United States National Reference for Fetal Growth. *Obstetrics & Gynecology*, 87(2):163-168, February 1996.**Birth Length and Head Circumference** by gestational age using:Lubchenco LL, 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*, 37(3):403-408, March 1966.**POSTNATAL MEASURES****Weight and Height** by gender and chronological or corrected age* using:

Anthropometric values (centiles, z-scores, percent of median, BMI) generated by EpiInfo 2000 for both the WHO/NCHS 1977 and the CDC/NCHS 2000 Reference Standards (Dibley et al., '87). Initially the WHO/NCHS 1977 Reference will be used to determine case status.

Head Circumference by sex and chronological or corrected age* using:

WHO/NCHS 1977. Tenth centile cutpoints are programmed. EpiInfo 2000 can generate CDC/NCHS 2000 centiles and z-scores.

Palpebral Fissures by chronological or corrected age* using:Thomas IT, Gaitantzis YA, Firas JL. Palpebral Fissure Length From 29 Weeks Gestation to 14 Years. *The Journal of Pediatrics: Clinical and Laboratory Observations* 111(2):267-268, August 1987. Cutpoints for 10th centile for age are programmed.

* Corrected age applied when gestational age <37 weeks' gestation and chronological age <24 months.

Fig. 4. FASSNet references for standardizing growth measures**Program evaluation and follow-up data elements**

All abstracted information is linked to the source from which it was obtained. Because information is compiled from many sources for any individual child, the multiple source approach will provide the opportunity to evaluate the productivity of the various sources relative to case finding, quality of data, and contribution of data for the assignment of the case-definition category. Information on other potential sources (e.g., programs where a child is referred for services) is also collected. This follow-up information can lead to new sources, additional data, changes in case-definition, and identification of additional cases.

Standardization of growth measurements. All available growth measurements (along with dates of each measurement) for weight, height, head circumference, and palpebral fissure length are abstracted and entered into the FASSNet Application. Because measurement practices in the United States vary (between customary and metric), all growth measures are converted to metric for ease of calculations. Standard growth references for age and sex were identified for both prenatal (Alexander et al., '96; Lubchenco et al., '66) and postnatal (Dibley et al., '87) growth (Fig. 4). Because references for palpebral fissure measurements are scarce, a group of clinicians with extensive experience in FAS and dysmorphology selected the standard used for palpebral fissure length (Thomas et al., '87). For all growth measures, growth retardation is indicated when the value is at or below the 10th centile.

All growth measures for birth are corrected for the recorded gestational age. When conflicting gestational ages are found, a hierarchy reflecting the most reliable source of data is used. For postnatal growth measures,

chronological age is determined by taking the date of exam minus the date of birth. When the recorded gestational age is <37 weeks, prematurity is corrected up to age 24 months. CDC's EpiInfo 2000 Nutrition software program is used to assign anthropometric values including percentile for age, z-scores, and percent of median (Dean et al., '00). Both the World Health Organization (WHO)/National Center for Health Statistics (NCHS) 1977 and the new CDC/NCHS 2000 Reference Standards are generated in the FASSNet program. Initially, the WHO/NCHS 1977 Reference will be used as it was the standard during the target birth years.

Data abstraction. Standard data abstraction procedures were developed collaboratively. The FASSNet Abstractors Committee reviewed appropriate abstractors' manuals and used them as guides to develop the FASSNet Abstractors' Manual. The Committee also manages continuing development of the manual and oversees training procedures for new abstractors. Abstractors were initially trained at the CDC and are evaluated periodically for quality assurance.

After identification of suspect cases by one of the methods previously described, the records are reviewed and abstracted on site and data are entered into the FASSNet Main Application via laptop computers. Data abstraction takes 15 minutes – 2 hours, depending on the type and amount of information found. Drop-down menus and fields in the abstraction database were designed to provide uniform coding and reflect commonly used medical records terminology.

Data quality control/quality assurance. Several methods are used to ensure data quality. First, the database was programmed so that many of the fields

have range checking at data entry. Second, the data manager at each FASSNet site evaluates specific data fields for missing and incongruent data. This is done manually and by the use of an automated "Consistency Check" computer program. The "Consistency Check" program reviews key variables associated with the case definition for missing or inaccurate dates. Third, each FASSNet site plans to re-abstract 10 – 15% of the records for additional quality assurance.

PROVIDER EDUCATION AND TRAINING

To increase awareness of FAS and improve case finding and referral, a major objective of FASSNet was to implement provider training and education. We targeted improving the ability of providers to recognize, accurately diagnose, and refer children with FAS to appropriate services, and improving documentation in the records of children suspected of having FAS. Toward this end, the FASSNet Provider Education and Training Committee developed a "charting tool" for use by healthcare providers when a child is suspected of having FAS (FASSNet Charting Tool, '00). Additional FASSNet provider education and training activities and evaluation of their impact will be described in more detail in subsequent papers.

PROGRAM EVALUATION

Another major objective of FASSNet is to evaluate the completeness of the surveillance system methodology, its ability to generate an accurate prevalence rate for FAS, and the potential for monitoring trends. Program evaluation strategies include developing methods to estimate under-ascertainment, completeness of case ascertainment by source, and availability or quality of the data to determine case status by age and source. Each FASSNet site developed specific methods to determine predictive value positive, efficiency, reproducibility, process evaluation, and quality assurance. These activities are ongoing and will be described in more detail in subsequent papers.

DISCUSSION

FASSNet is a multiple-source, collaborative, surveillance system now operating in four states. A rigorous surveillance case definition was developed using the IOM clinical case definition. The FASSNet case categories provide a standard method of case classification, can be used in a variety of clinical settings, and allow the use of information from multiple data sources. The system can adapt to changes in the clinical or surveillance case definition and continue to monitor trends in FAS occurrence. Because FASSNet collects information on all children identified as suspect cases in a geographic region, the characteristics of high-risk children who do not meet the full surveillance case definition can also be assessed. Therefore, the potential exists to evaluate the public health burden of other effects associated with prenatal alcohol exposure—

FAE, ARND, ARBD, and other nonspecific conditions. However, to monitor these other conditions would be even more challenging, if not prohibitive, since they are less specific and even less agreed upon than FAS.

The FASSNet methodology has limitations. First, even though we have established criteria for assessing completeness of case ascertainment in the four states, the true completeness of case ascertainment is unknown. However, case under-ascertainment (if present) should remain stable and, with no major changes in the methodology, allow FASSNet to monitor trends over time. Likewise, case over-ascertainment (identification of false-positive cases) may occur. Ideally, a sample of confirmed cases will be evaluated independently by a qualified examiner (e.g., dysmorphologist or clinical geneticist) to determine the false-positive rates. Finally, the complexity and variability of the expression of the phenotype and the reliance on medical record data present challenges to any FAS surveillance system.

The methodology described in this report, however, provides more complete case ascertainment than other traditional public health surveillance methods that are often limited to single sources of information and provider notification. FASSNet can also be used for special follow-up studies to monitor access to services, evaluate unmet service needs, and assess intervention programs with biological mothers. Finally, with four states contributing suspect cases, FASSNet will likely be the largest database of both children suspected of having FAS and those that meet a standardized surveillance case definition, allowing for a greater understanding of this multifaceted and challenging, yet totally preventable, birth defect.

LITERATURE CITED

- Aase JM. 1994. Clinical recognition of FAS: difficulties of detection and diagnosis. *Alcohol Health & Research World* 18(1):5–16.
- Alexander GR, Himes JH, Kaufman RB, Mor J, Kogan M. 1996. A United States national reference for fetal growth. *Obstetrics & Gynecology* 87(2):163–168.
- Astley SJ and Clarren SK. 1995. A fetal alcohol syndrome screening tool. *Alcoholism: Clinical and Experimental Research* 19(6):565–571.
- CDC. 1993a. Linking multiple data sources in fetal alcohol syndrome surveillance—Alaska. *MMWR* 42(18):312–314.
- CDC. 1993b. Fetal alcohol syndrome—United States, 1979–1992. *MMWR* 42(17):339–341.
- CDC. 1995a. Update: Trends in FAS—United States, 1979–1993. *MMWR* 44(13):249–251.
- CDC. 1995b. Birth certificates as a source for fetal alcohol syndrome case ascertainment—Georgia, 1989–1992. *MMWR* 44(13):251–253.
- CDC. 1997. Surveillance for fetal alcohol syndrome using multiple sources—Atlanta, Georgia, 1981–1989. *MMWR* 46:1118–1120.
- CDC. 1998. Behavioral Risk Factor Surveillance System (BRFSS) code book. On line at www.cdc.gov/nccdphp/brfss/surveydata/1997/CODEBK97.TXT
- Chavez GF, Cordero JF, Becerra JE. 1988. Leading major congenital malformations among minority groups in the United States, 1981–1986. *MMWR* 37(SS-3):17–24.
- Cordero JF, Floyd RL, Martin ML, Davis M, Hymbaugh K. 1994. Tracking the prevalence of FAS. *Alcohol Health & Research World* 18(1):82–85.
- Dean AG, Arner TG, Sangam S, Sanki GG, Friedman R, Latinga M, Zubieta JC, Sullivan KM, Smith DC. 2000. Epi Info 2000, a data-

- base and statistics program for public health professionals for use on Windows 95, 98, NT, and 2000 computers. Centers for Disease Control and Prevention, Atlanta, Georgia.
- Department of Health and Human Services. 1997. CDC, Announcement 745: Cooperative agreement for population-based surveillance of fetal alcohol syndrome; notice of availability of funds for fiscal year 1997. Federal Register 62(99):28032-28037.
- Dibley MJ, Goldsby JB, Staehling NW, Trowbridge FL. 1987. Development of normalized curves for the international growth reference: historical and technical considerations. *American Journal of Clinical Nutrition* 46:736-748.
- Egeland GM, Perham-Hester KA, Gessner BD, Ingle D, Berner JE, Middaugh JP. 1998. Fetal alcohol syndrome in Alaska, 1977 through 1992: an administrative prevalence derived from multiple data sources. *American Journal of Public Health* 88(5):781-786.
- FASSNet. 2000. Abstractor's manual, provider charting tool. www.cdc.gov/ncbddd/fas/documents/charttool.pdf.
- FASSNet. 2000. Abstractor's Manual.
- Hall JG, Froster-Iskenius UG, Allanson J. 1989. Handbook of normal physical measurements. Oxford: Oxford University Press, p 81-88. (From Nellhaus G. 1968. Head circumference from birth to eighteen years. Practical composite international and interracial graphs. *Pediatrics* 41:106-114. And Tanner JM. 1978. Physical growth and development. In: Forfar JO and Arneil GC. editors. Textbook of Pediatrics. Churchill Livingstone, Edinburgh. p 253-303.)
- Hymbaugh KJ, Boyle CA, Aase JM. 1993. Age-specific differences in physical characteristics of alcohol affected children in a special population study. Paper presented at the 1993 Research Society on Alcoholism Scientific Conference. San Antonio, Texas.
- Institute of Medicine. 1996. Fetal alcohol syndrome—diagnosis, epidemiology, prevention, and treatment. Washington, DC: National Academy Press.
- Jones KL. 1999. Early recognition of prenatal alcohol effects: A pediatrician's responsibility. *Journal of Pediatrics* 135(4):405-406.
- Lubchenco LL, Hansman C, Boyd E. 1966. Intrauterine growth in length and head circumference as estimated from live births at gestational ages from 26 to 42 weeks. *Pediatrics* 37(3):403-408.
- Miller LA, Shaikh T, Stanton C, Montgomery A, Rickard R, Keefer S, Hoffman R. 1995. Surveillance for fetal alcohol syndrome in Colorado. *Public Health Reports* 110:690-696.
- Morse BA, Idelson RK, Sachs WH, Weiner L, Kaplan LC. 1992. Pediatricians' perspective on fetal alcohol syndrome. *Journal of Substance Abuse* 4:187-195.
- Robinson GC, Conry JL, Conry RF. 1987. Clinical profile and prevalence of fetal alcohol syndrome in an isolated community in British Columbia. *Canadian Medical Association Journal* 137:203-207.
- Stoler JM and Holmes LB. 1999. Under-recognition of prenatal alcohol effects in infants of known alcohol abusing women. *Journal of Pediatrics* 135(4):430-436.
- Thomas IT, Gaitantzis YA, Firas JL. 1987. Palpebral fissure length from 29 weeks gestation to 14 years. *The Journal of Pediatrics: Clinical and Laboratory Observations* 111(2):267-268.