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Prevalence and characteristics of fetal alcohol syndrome and partial fetal alcohol syndrome in a Rocky Mountain Region City

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ABSTRACT

Background: The prevalence and characteristics of fetal alcohol syndrome (FAS) and partial FAS (PFAS) in the United States (US) are not well known.
Methods: This active case ascertainment study in a Rocky Mountain Region City assessed the prevalence and traits of children with FAS and PFAS and linked them to maternal risk factors. Diagnoses made by expert clinical dysmorphologists in multidisciplinary case conferences utilized all components of the study: dysmorphology and physical growth, neurobehavior, and maternal risk interviews.
Results: Direct parental (active) consent was obtained for 1278 children. Averages for key physical diagnostic traits and several other minor anomalies were significantly different among FAS, PFAS, and randomly-selected, normal controls. Cognitive tests and behavioral checklists discriminated the diagnostic groups from controls on 12 of 14 instruments. Mothers of children with FAS and PFAS were significantly lower in educational attainment, shorter, later in pregnancy recognition, and suffered more depression, and used marijuana and methamphetamine during their pregnancy. Most pre-pregnancy and pregnancy drinking measures were worse for mothers of FAS and PFAS. Excluding a significant difference in simply admitting drinking during the index pregnancy (FAS and PFAS = 75% vs. 39.4% for controls), most quantitative intergroup differences merely approached significance. This community's prevalence of FAS is 2.9–7.5 per 1000, PFAS is 7.9–17.7 per 1000, and combined prevalence is 10.9–25.2 per 1000 or 1.1–2.5%.
Conclusions: Comprehensive, active case ascertainment methods produced rates of FAS and PFAS higher than predicted by long-standing, popular estimates.

1. Introduction

The rate of fetal alcohol syndrome (FAS) in the United States (US) was estimated for many years, and believed by many, to be 0.33–3.0 per 1000 children (Abel, 1998; Abel and Sokol, 1987, 1991; CDC, 1995, 1997; Fox et al., 2015; May and Gossage, 2001; Stratton et al., 1996). However, given the lack of active case ascertainment studies (which seek to identify cases in a general population) of any fetal alcohol spectrum disorders (FASD) in the US and other developed countries (May et al., 2009), many people believed that the above estimates were substantial under estimates. FASD are rarely recognized or diagnosed in the general population or in general clinical

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settings (Chasnoff et al., 2015). Therefore, surveillance and clinic-based studies are unlikely to produce a true prevalence (Fox et al., 2015). Active case ascertainment studies undertaken in schools have proven to be accurate and to produce high rates of FASD (May et al., 2009). In-school study methods for large communities have been employed mostly in South Africa and Italy (May et al., 2000, 2006, 2007, 2011, 2013a; Urban et al., 2008; Viljoen et al., 2005).

Prior to 2014, only three articles had been published that utilized active case ascertainment methods in schools in the US general population. Clarren et al. (2001) reported a rate of 3.1 FAS cases per 1000 in a county in Washington State. In that study, only one of the seven FAS cases found in the schools had been diagnosed previously. Burd et al. (1999) and Poitra et al. (2003) have reported on active case ascertainment methods used in multiple years in Head Start classrooms using a screening tool and follow up dysmorphology exam. Burd et al. (1999) reported six cases of FAS out of 1013 children, 5.9 per 1000. Poitra et al. (2003) included more children from the same community and 4.3 children per 1000 had FAS. The authors do mention that some of the children with FAS identified in the Head Start studies had been diagnosed prior to the study, but this community benefited from an ongoing active dysmorphology consultation service. Many undiagnosed cases of FAS and other FASD exist in the US population, and active case ascertainment studies are designed to find them and to estimate the true prevalence of FASD.

1.1. Purpose

The purpose of this study was to: (1) determine the feasibility of using these particular active case ascertainment methods in a US community to identify children with FAS and partial fetal alcohol syndrome (PFAS) in public and private schools; (2) ascertain maternal risk factors for FASD in a general US population; and (3) determine the prevalence and characteristics of children with FAS and PFAS in this community. Data were collected in three samples of first grade students in this Rocky Mountain Region City (RMRC). This study was initiated at the invitation of the city/county health department, endorsed by school administrators, and approved by the Board of Trustees of the City Schools. The studies utilized research methods pioneered in South Africa and Italy.

1.2. The study community

The population of RMRC is 59,000 in a county of 85,000. The composition of the population is 88.5% White, 5.0% American Indian, 3.8% mixed race, 1.1% Black, and 3.4% of Hispanic ethnicity (US Census, 2015). Compared to the US population, the study community was more White (+10.8%), less Black (−12.1%), more American Indian (+3.8%) and less Hispanic (−13.7%). RMRC residents are predominantly middle class, with average economic indicators similar to small cities and counties in the region and state. The averages in the county are slightly below US averages on a number of economic indicators: per capita income is $24,100 (US average is $28,155), median household income is $43,800 (US = $53,000), and 16.5% are below the poverty level (US = 15.4%) (US Census, 2015). Per capita alcohol consumption in this state was 2.99 gallons (11.3 l) of ethanol per year in 2009 compared to the US average of 2.3 gallons (8.7 l) (LaVallee and Yi, 2011). The overall health rank of this state by the United Health Foundation (2014) is well in the upper middle tier, between 20 and 25 of 50 states. County data from the CDC Behavioral Risk Factor Surveillance System (BRFSS) indicates less binge drinking overall (13.3%) than in either the state (16.9%) or the US (15.5%); however among adults ages 18–44, 18.3% binge drink which compares to 24.5% for the state ("Western" County, 2011). Heavy drinking was reported for this county at 4.9%, compared to 5.9% for the state, and 5.1% for the US ("Western" County, 2011). Among adults 18–44 years, heavy drinking was 5.6% in the County and 6.9% for the State ("Western" County, 2011). Binge drinking among women (four or more drinks per occasion) was lower at 14.3% in this state than among males (five or more drinks per occasion) at 27.3% ("Western State" BRFSS, 2014), but bingeing among women was 2.2% higher than US averages (12.5%) (CDC, 2013). Therefore, this site has slightly lower economic status than US averages, overall good health, but indicators of heavy drinking and binge drinking are lower than the home state but higher than US averages.

2. Methods

The diagnostic criteria used for FAS and PFAS (see Fig. 1) were originally set forth by the FAS advisory group of the U.S. Institute of Medicine (Stratton et al., 1996), and operationalized and updated from clinical experience (Hoyme et al., 2005). The classification of the children is based on: (1) specific facial dysmorphology, (2) diminished physical stature and/or weight, (3) defined intellectual, developmental, social, and neuropsychological assessments, and (4) multiple measures of maternal alcohol consumption. Data for all of the diagnostic domains were collected and analyzed by members of a multidisciplinary clinical team, each member (physicians, psychologists, and interviewers) working blinded to any prior knowledge of the diagnostic findings in other parts of the study and the child and family. The data and findings were summarized for each child, and structured case conferences were held among the specialists. Highly experienced pediatric dysmorphologists/clinical geneticists assigned final diagnoses after considering all of the relevant data and the opinions of team members involved in the examination, testing, and interview process. The diagnoses considered in this study were: FAS, PFAS, normal/not FAS, another known disorder, or deferred for further testing. Due to limited resources and a short time frame, the team focused on only FAS and PFAS diagnoses, as these conditions exhibit the most clearly defined dysmorphic features in the FASD continuum. Detailed numbers of sampling and the flow of the study are found in Fig. 2.

2.1. Child data

The overall study population was all children enrolled in first grade ($n = 2377$) in all of the 17 elementary schools (15 public and 2 private) during school years beginning in 2007, 2008, and 2009. Children who participated in the study did so via active (direct written) parental consent. While almost 60% of the parents returned the consent forms for the combined three samples ($n = 1421$), 143 checked the box that declined participation; so the total consented into the study was 1278 (53.8%) (see Fig. 2). The return of forms was lowest in the first sample (53%), better in the second (57%), and extra effort by the staff (multiple mailings and phone calls) in sample 3 led to a 70% return rate.

Data on the growth, development, and dysmorphology of the children were collected via a three-tier method. In Tier I all consented children were screened for height, weight, and occipitofrontal (head) circumference (OFC) in their respective schools by project staff. Then, if the child had measurements $\leq$10th centile on OFC and/or both height and weight on the Centers for Disease Control and Prevention (CDC) growth charts, he/she was referred for Tier II screening, a full dysmorphology exam ($n = 588$). Because children with FASD are often not recognized or identified (Chasnoff et al., 2015), special referrals of children experiencing problems in school were also accepted from the teachers ($n = 10$). For the dysmorphology exams, all qualifying children were seen by the research team in their respective schools (in specially arranged rooms). Three pediatric clinical geneticists/dysmorphologists who are highly trained in and experienced with FASD diagnoses provided the exams. Each child was examined individually and independent of any prior knowledge (e.g. reason for entry to the study). Each physician-led team collected independent measures of growth, physical development, and dysmorphology and recorded them on a structured inventory form for child data salient to FASD and other known birth defects. This form includes a weighted FAS dysmorphology score (Hoyme et al., 2005). In the first sample of this study, each child received identical exams from two dysmorphologists; in samples 2 and 3, only one exam. At the end of each day, after the dysmorphology assessments, a preliminary diagnosis of FAS, PFAS, deferred, another diagnosis, or not FASD was assigned by the examining physicians. Those children who received a preliminary diagnosis of a FASD or deferred were advanced to cognitive and behavioral testing as part of Tier III of the study.

2.2. Normal controls

Candidates for normal comparison/control children were picked by random methods (without replacement) from the school rolls. By drawing the children from the same first grade population, the final control children were well matched with the subject children by age, sex, and general environment. Identical physical exams and cognitive/behavioral testing were performed on the control candidates. One