Drug and Alcohol Dependence 159 (2016) 207–218

Contents lists available at ScienceDirect

Drug and Alcohol Dependence

journal homepage: www.elsevier.com/locate/drugalcdep

Full length article

The continuum of fetal alcohol spectrum disorders in four rural communities in south africa: Prevalence and characteristics

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ARTICLE INFO

Article history:
Received 24 October 2015
Received in revised form
18 December 2015
Accepted 18 December 2015
Available online 31 December 2015

Keywords:
Fetal alcohol spectrum disorders (FASD)
Prenatal alcohol use
Alcohol abuse
Maternal risk for FASD
Prevalence
Children with FASD
South Africa

ABSTRACT

Background: Prevalence and characteristics of the continuum of diagnoses within fetal alcohol spectrum disorders (FASD) were researched in previously unstudied rural, agricultural, lower socioeconomic populations in South Africa (ZA).
Methods: Using an active case ascertainment approach among first grade learners, 1354 (72.6%) were consented into the study via: height, weight, and/or head circumference ≤25th centile and/or random selection as normal control candidates. Final diagnoses were made following: examination by pediatric dysmorphologists/geneticists, cognitive/behavioral testing, and maternal risk factor interviews.
Results: FASD children were significantly growth deficient and dysmorphic: physical measurements, cardinal facial features of FAS, and total dysmorphology scores clearly differentiated diagnostic categories from severe to mild to normal in a consistent, linear fashion. Neurodevelopmental delays were also significantly worse for each of the FASD diagnostic categories, although not as consistently linear across groups. Alcohol use is well documented as the proximal maternal risk factor for each diagnostic group. Significant distal maternal risk factors in this population are: low body weight, body mass, education, and income; and high gravidity, parity, and age at birth of the index child. In this low SES, highly rural region, FAS occurs in 93–128 per 1000 children, PFAS in 58–86, and, ARND in 32–46 per 1000. Total FASD affect 182–259 per 1000 children or 18–26%.
Conclusions: Very high rates of FASD exist in these rural areas and isolated towns where entrenched practices of regular binge drinking co-exist with challenging conditions for childbearing and child development.

1. Introduction

1.1. Diagnosing a continuum

The diagnosis of fetal alcohol spectrum disorders (FASD) has been evolving since the first criteria for fetal alcohol syndrome (FAS) were defined by Jones and Smith (1973). Children with the

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http://dx.doi.org/10.1016/j.drugalcdep.2015.12.023
0376-8716/

most dysmorphic features and cognitive and behavioral impairments were identified as FAS. Soon thereafter, less consistent, less severe patterns of dysmorphia and impairment were recognized in animal and human studies and referred to as fetal alcohol effects (FAE; Aase, 1994; Aase et al., 1995; Clarren and Smith, 1978). In 1996, four specific diagnoses were developed by a committee of the Institute of Medicine (IOM): FAS, partial FAS (PFAS), alcohol-related birth defects (ARBD), and alcohol-related neurodevelopmental disorder (ARND) (Stratton et al., 1996). These four diagnoses form a continuum from the most dysmorphic to least dysmorphic and came to be known as FASD (Streissguth and O'Malley, 2000; Warren et al., 2004). FASD are rarely diagnosed or diagnosed properly (Chasnoff et al., 2015) which colors understanding of the full spectrum of FASD. Because of this, FASD epidemiology information gathered through registries are inaccurate (Fox et al., 2015). Focused epidemiologic studies of FASD are difficult and expensive to pursue (Stratton et al., 1996). Few studies have documented the prevalence and characteristics of the full continuum of FASD, especially in the United States and Europe where FASD were believed for many years to be rare occurrences, affecting an estimated maximum of 1% of the general population (Sampson et al., 1997). But recent studies indicate that FASD prevalence is much higher, 2–5%. At least this has found to be true in the USA, Italy, Poland, and Croatia (May et al., 2009, 2011a, 2014a, 2015; Okulicz-Kozaryn et al., 2015; Petković and Barišić, 2010, 2013), and especially true in South Africa.

1.2. Epidemiology studies of FASD in South Africa

Unfortunately, some communities in South Africa (ZA) have the highest general-population rates of FASD anywhere in the world. But from an epidemiologic perspective, ZA has proven to be an excellent venue for understanding the diagnosis, prevalence, characteristics, and etiology of the FASD continuum. The prevalence and characteristics of FASD have been described by the authors of this paper in four studies of a single municipality and surrounding rural areas in the western portion of the Western Cape Province (WCP) close to the Cape Town metropolitan area (May et al., 2000, 2007, 2013a; Viljoen et al., 2005). Other researchers have completed community studies in other provinces of ZA (Oliver et al., 2013; Urban et al., 2008, 2015; Viljoen and Hymbaugh, 2003). In the previous case control studies in the WCP, physical and neurobehavioral traits were researched extensively, the diagnostic categories have been explored with thousands of children, and research has progressed from an exclusive focus on FAS to delineating and understanding all forms of FASD. In a recent WCP study publication, FAS affected 59–91 per 1000 children, PFAS 45–70 per 1000, no cases of ARBD were found, and ARND affected 31–47 per 1000. The total FASD rates were 135–208 per 1000 or 13.5–20.7% (May et al., 2013a). Quite noteworthy, the prevalence of FASD was highest in rural areas surrounding the town in all previous studies in the WCP. Multiple other findings indicated that norms and practices of regular binge drinking, low socioeconomic status (SES), insufficient nutrition, high fertility, and challenging conditions for prenatal and postnatal development combine to elevate the prevalence and severity of FASD (May et al., 2005, 2008, 2013b, 2014b; Viljoen et al., 2002).

1.3. The current study

As in previous studies of the WCP, this study utilizes active case ascertainment methodology employed by a multidisciplinary field research team and pediatric geneticists/dysmorphologists. However, the four small towns and their surrounding rural areas studied here had never been studied for FASD epidemiology before. And they are more rural, remote, and lower SES than the community of previous WCP studies.

2. Methods

2.1. Sampling and recruitment

Active, written consent for children to participate in the study was sought from parents and guardians of all first grade pupils ($n = 1866$) enrolled in all 53 primary schools of the four town region and child assent forms were obtained from all children seven years and older. Consent was received for 1354 (72.6%). As in Fig. 1, a three-tier process of screening, data collection, and diagnosis was instituted for all consented children. In Tier I all children were measured for height, weight, and head circumference. If a child was ≤25th centile on height, weight, and/or occipitalfrontal (head) circumference (OFC), he/she was advanced to Tier II which was an in-person pediatric dysmorphology exam to evaluate potential cases. In addition, 559 child enrollment numbers were picked randomly as potential controls (normal/not FASD comparison children) from all children on the school roles, and 406 had consent to participate. Each of the qualifying children (small and/or randomly-selected) was advanced to Tier II where they received the same dysmorphology exam from a dysmorphologist assisted by a scribe to record exam data. Of the children advanced to Tier III, the racial composition was generally a mirror of that of the four town region: 92.9% colored (mixed race), 5.6% black, and 1.5% white.

2.2. IOM diagnostic categories

The IOM diagnostic criteria of the four FASD diagnoses are presented in Fig. 2 and described in more detail elsewhere (Hoyme et al., 2005, 2015). Significant growth retardation and significant dysmorphia are present in children with FAS, less growth restriction is present in children with PFAS, but at least two of the three cardinal facial features and a constellation of other minor anomalies are present in both FAS and PFAS. The clinical dysmorphic traits of FAS and PFAS have been clearly linked with prenatal alcohol exposure in thousands of cases and in multiple correlation studies (May et al., 2011b, 2013b), and these diagnoses can be made by qualified pediatricians without direct documentation of alcohol exposure, particularly after ruling out other anomalies with similar phenotypes. In previous studies in WCP communities, very seldom is it necessary to diagnose a child with FAS or PFAS without direct or strong collateral evidence of prenatal alcohol use (May et al., 2008, 2013b). Children with ARND do not have a characteristic pattern of facial characteristics, and therefore direct evidence of prenatal alcohol exposure and evidence of substantial cognitive impairment are required.

2.3. Assessment of cognitive and behavioral traits

In Tier III, all randomly-selected control candidates and all children with significant features common to a diagnosis within FASD were advanced to cognitive testing, and their teachers completed Achenbach Teacher Report Forms (TRF; Achenbach and Rescorla, 2001) to evaluate inattention and total behavioral issues. The tests were: Test of Reception of Grammar (TROG; Bishop, 1989) to measure verbal abilities; Raven Coloured Progressive Matrices (Raven, 1981) for non-verbal abilities; and the Digit Span subtest of the Wechsler Intelligence Scales for Children, Third edition (WISC-III; Wechsler, 2003) to measure working memory. The tests were administered in the individual schools by blinded, contract psychometrists. Most tests were administered in Afrikaans, the dominant language of the region, although 0.4% were administered in English, and 5.4% were administered in isiXhosa for the