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BRIEF REPORT

Brief Report: Prevalence of Pervasive Developmental Disorder in Brazil: A Pilot Study

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Abstract This pilot study presents preliminary results concerning the prevalence of Pervasive Developmental Disorder (PDD) in South America. It was a three-phase study conducted in a typical town in Southeast Brazil. Case definition was based in a combination of standardized instruments and clinical evaluations by experts. The prevalence of PDD was 27.2/10,000 (95% CI: 17.6–36.8) and some hypotheses were raised to explain this low frequency. Clinical findings of PDD cases were consistent with previous data, such as, male preponderance, more children diagnosed with PDD-NOS than with autistic disorder, and half of them born from older mothers. Moreover, the study raised concerns about treatment of cases, because identification of PDD had been late and access to services has been very limited.

Keywords Prevalence · Pervasive developmental disorders · Autism · Epidemiological studies · South America

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Introduction

Autism is a pervasive developmental disorder (PDD) defined by early social, communication and behaviors impairments. In recent surveys, a best estimate of 70-90/10,000 has been proposed for the prevalence of PDD (Fombonne 2009a; US Center of Disease Control [CDC] 2009). Since the first studies in the 1960's, prevalence has been rising mainly due to changes in diagnostic concepts and criteria**,** age of identification, improved professional, public awareness and changes in services and public policy. Nevertheless, the contribution of other risk factors in this increase cannot be excluded (Center for Disease Control 2009; Fombonne 2009a; Parner et al. 2008; Atladóttir et al. 2007).

The influence of cultural, ethnic, geographic and socioeconomic factors associated to PDD is unclear (Fombonne 2009a; Newschaffer et al. 2007). Most surveys were conducted in North America, Northern Europe and Japan, and cross-national comparisons have been scarce (Elsabbagh et al., in press; Fombonne 2009a). With respect to ethnicity, some studies have reported lower rates of PDD among Latino/Hispanic children (Palmer et al. 2010; Center for Disease Control 2009; Liptak et al. 2008)**,** while some, but not all, surveys from Northern Europe have pointed at higher rates of autism among immigrants (Newschaffer et al. 2007; Fombonne 2006.). Some studies have reported higher PDD rates among white compared to black children (Center for Disease Control 2009), others found a raised risk in the black population (Croen et al. 2002) while others have described similar rates between these two groups (Newschaffer et al. 2007). These inconsistent findings may reflect differential patterns of access to services, but the hypothesis of a true difference in PDD risk by ethnic/racial groups remains plausible.

Only two studies have been carried out in South-America. The first one, a Venezuelan clinic-based data-review survey, found a PDD frequency of 17/10,000 in 3–9 years old children (Montiel-Nava and Peña 2008). The second study was based on a non-representative sample of service users from 3 health care centers in a town close to Buenos Aires, Argentina. It provided a prevalence estimate of 1.3% for PDD among 839 children under age 5 (Lejarraga et al. 2008). This study was focusing on psychomotor developmental problems and had several methodological limitations that call for caution in interpreting its results for PDD rates. The limitations of these studies prompted us to conduct a pilot study in order to estimate the prevalence of PDD in a Brazilian town and to examine the clinical presentation of PDD cases. To the best of our knowledge, this is the first population-based study in this region to rely on direct evaluation of cases, instead of record reviews, to determine caseness.

Methods

Site

Brazil is composed of five regions. The Southeast is the most populous one with 1,668 municipalities, with less than 300,000 inhabitants each. This region is highly urbanized (90.5%), with a literacy rate of 91.5% and a Human Developmental Index (HDI) higher (0.824) than the Brazilian average (0.693) (Instituto Brasileiro de Geografia e Estatística [IBGE], 2000). Brazil has a reliable population registry based on national census according to residence (IBGE, 2000). Since 2001, all children 7–14 years of age are required by law to be registered and to attend school (Lei de Diretrizes e Bases 2001). The combination of these two demographic data sources allows for the precise determination of the number of school age children living in a specific district, in any Brazilian municipality.

This cross-sectional study was conducted in Atibaia, a typical town from São Paulo state. Atibaia is similar to other municipalities from Southeast Brazil in terms of urbanization (91.0%), literacy (93.7%) and HDI (0.819). The population is 126,851 inhabitants, 10,503 7–12 years-old children living in urban areas and 1,902 living in rural areas (IBGE, 2000). One Atibaia urban district with 1,470 children aged 7–12 was selected as study site. This particular district was chosen because it benefits from an extensive Family Health Program (Portuguese acronym, PSF). PSF teams are each responsible to promote health and prevention for 1,000 families through regular home visits, being well suited to ascertain cases in an epidemiological survey. This district is representative of the town with respect to literacy (93.5%), proportion of the population under age 15 years (26.7 vs. 26.9% for the entire town), waste collection in the neighborhood (99.0 vs. 95.4%) and other similar indices (Sistema de Informação da Atenção Básica [SIAB], 2007; Fundação Sistema Estadual de Análise de Dados [SEADE], 2010).

Study Design

The design was a three-phase study, including 2 screening phases followed by a diagnostic confirmation phase. In phase 1, Atibaia's health and educational professionals were asked to screen and refer all children, aged 7–12, suspected to have a PDD. In phase 2, Autism Screening Questionnaires (ASQ) (Berument et al. 1999) were obtained from referred children and subjects with a score at or higher than 15 were considered to form the positive screening group. The cut-off of 15 for the Brazilian version of the ASQ was previously validated, achieving a sensitivity of 92.5% and a specificity of 95.5% (Sato et al. 2009). In phase 3, psychologists and a senior child psychiatrist assessed all screen positive children identified in phase 2 in order to determine the final diagnostic status of each child.

Case Identification

All district public services were identified and visited. We additionally searched for PDD cases in specialized schools, health services, one specialized clinic, and one inpatient unit. At the beginning of the study, local educational, health and social authorities were contacted. The research team coordinated a training program on PDD for professionals of the district. A short 10 items symptoms-list, based on the DSM-IV (American Psychiatric Association 1994), and the CDC guidelines were also provided in order to help them identify suspected PDD cases. In addition, bi-monthly follow-up meetings with the research team were organized to further train professionals in referral procedures. To evaluate co-occurring behavioral problems, the Strength and Difficulties Questionnaire—SDQ (Goodman 1997) was filled out by parents in phase 2 with a 92% response rate. In phase 3, diagnostic confirmation by professionals was facilitated by administration of the K-SADS (Ambrosini 2000) and of the ADI-R (Lord et al. 1994). Final diagnostic status was established by reviewing all relevant clinical and developmental data.

Case Definition

In this study, children with any PDD as defined in the DSM-IV (APA 1994) were considered as cases. Thus, we included children with Autistic Disorder, PDD-NOS, Childhood Disintegrative Disorder and Asperger Disorder.

Results

Of the 1,470 screened children, 94 subjects (6.4%) were referred as suspected cases of PDD. Eleven who did not meet inclusion criteria (such as age) were excluded. Of the remaining 83, 12 children had scores at or higher than 15 on the ASQ. This translated into a prevalence of 0.82% (95% CI: 0.47–1.42) for the screen positive children. Based on SDQ scores, 72.4% of those 83 suspected cases of PDD had significant behavioral problems, such as ADHD or conduct problems, including all 12 children with high ASQ scores.

In phase 3, the 12 screen positive children with high ASQ scores and 12 controls (randomly selected from the 71 cases initially referred but with low ASQ scores) were blindly assessed by psychologists and the senior child psychiatrist. The mean ages were not statistically different, respectively 8.25 years (SD = 1.76) for PDD suspected children, and 8.81 years (SD = 1.63) for the controls.

One child of the control group and 3 of the 12 PDD-suspected children met PDD criteria (all boys). This translated in a PDD prevalence of 27.2/10,000 (95% CI: 17.6–36.8). Only one child attending a special school had been diagnosed prior to the study (at age 6). The other three children did not receive any type of treatment (Table 1).

Discussion

We found a prevalence of almost 0.3% of PDD in a Brazilian town, which is lower than figures reported in most other surveys (Center for Disease Control 2009; Fombonne 2009a). Only one other reliable prevalence study on PDD was published in South America. In Venezuela, Montiel-Nava and Pena (2008) also found a low PDD prevalence of 17 per 10.000 (11 per 10.000 when considering autistic disorder only). As mentioned earlier, there are several hypotheses to explain the variability in rates of PDD between epidemiological studies worldwide. Lower frequencies of PDD in low/middle-income countries may reflect methodological issues, such as small samples, lack of awareness of local population/professionals, poor record keeping, misdiagnosis, and later age at diagnosis (Center for Disease Control 2009). In this study, we tried to overcome some of these pitfalls, especially with the implementation of an intensive population awareness and training program for front line health/educational and a multistage ascertainment procedure. We capitalized on primary care/preventative services (the PSF Program), since the same health professionals perform regular home visits and are well acquainted with children of their family's caseload. Even though some children with PDD may have been missed, it is noteworthy that children referred for phase 2 screening had high levels of behavioral problems. This suggests that local professionals were willing to refer children to the study, probably since alternative referral pathways to diagnostic and treatment centers are almost nonexistent.

It is nevertheless tempting to speculate that the prevalence of PDD might indeed be lower due to specific characteristics of developing countries. Recent studies have showed high levels of stigma among adults with mental health problems from developing countries (Alonso et al. 2008), including Brazilian studies (Peluso and Blay 2010). Stigma is associated to social isolation, poorer quality of life, lower income and non-adherence to treatment (Brohan et al. 2010); Alonso et al. 2009). In addition, PDD subjects living in developing countries may have poorer prognosis due to inadequacies of health care systems, including poorer recognition of mental health problems, lower budget spent on mental health and the absence of skilled mental health practitioners (Knapp et al. 2006; Johnson et al. 2007). Another possibility is that PDD mortality in low and middle-income countries may be higher due limited capacities of the health system to prevent and treat medical correlates of PDDs, such as low birth weight, genetic syndromes, mental retardation, epilepsy, and the like (Gillberg et al. 2010; Mouridsen et al. 2008). In turn, the cumulative effect of these factors might result in a higher reproductive disadvantage for subjects with PDD that, in view of the high heritability of PDDs, may account for our

Table 1 Clinical presentation of PDD cases

Age (years)	Sex	Age at diagnosis (years)	Diagnosis	Type of school	Current treatment	Maternal age at child's born
10	M	6	Autistic Disorder	Special	Regular treatment with child psychiatrist from a known university plus multidisciplinary assistance at the Atibaia's inpatient clinic	36
7	M	–	PDD-NOS	Regular	None; waiting list for speech therapy	35
8	M	–	PDD-NOS	Regular	None; waiting list for psychological and speech therapy	23
7	M	–	PDD-NOS	Regular	None	23

lower prevalence findings. However, data are required to test further these hypotheses.

Clinical characteristics of the four PDD cases identified in this pilot study shed some light on how the Brazilian health system currently deals with children with PDDs. First, only one of them had a previous diagnosis of PDD, the most severe case. Yet, he was only identified late at age 6, long after parents usually report the first developmental concerns in their children (De Giacomo and Fombonne, 1998; Fombonne 2009b). Late identification of PDDs is a universal problem. Even in developed countries, children, especially those with PDD-NOS, are identified in primary school age years (Atladóttir et al. 2007; Shattuck et al. 2009), as most children in the current study. National data on age at diagnosis for Brazilian children diagnosed with PDD are not available (Teixeira et al. 2010), and, if confirmed, these preliminary findings point to the need to establish a PDD early diagnosis initiative in Brazil.

Second, only one child was educated in a special school. This is in keeping with the recent law mandating that all Brazilian children aged 7–14 years be educated in regular schools. This law is controversial, and research is required to evaluate the efficacy of this inclusion policy (Teixeira et al. 2010).

Improved outcomes have been associated with early enrollment in appropriate intervention programs (Jonhson et al. 2007). Therefore, it was concerning that only one PDD child was receiving regular intervention. As São Paulo is the most developed Brazilian State, these preliminary data should alert coordinators of the public health system nationwide.

Third, clinical features of this small case series were consistent with established correlates of PDD: (1) more children had a diagnosis of PDD-NOS compared to that of autistic disorder (Fombonne et al. press); (2) two children out of four were born from older mothers—a finding consistent with recent studies documenting parental age as a risk factor for autism (Kolevzon et al. 2007); and (3) a typical male preponderance—the absence of girls being simply consistent, in such a small sample, with the 4:1 male/female ratio reported for PDD (Fombonne et al. in press).

This pilot study has several limitations. The small sample size reduced the precision of estimates and increased the likelihood of sampling biases. Although the sample was selected from a typical town of the Southeast, Atibaia has a HDI (0.819) higher than the Brazilian (0.693) and the Latin American indices (0.701) (Human Development Report 2009). Furthermore, the sample was selected from a single district of the town, suggesting that results should be conservatively interpreted.

To summarize, this first PDD population-based prevalence study in South America found a lower frequency than previous international PDD studies. Methodological limitations likely explain this result, especially the small sample size. However, other explanations should also be considered. Further epidemiological surveys of PDD in this region are required to replicate these preliminary results and to improve service planning for children with PDD in Brazil.

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