

FOCUS REVIEW

Prevalence and Interpretation of Recent Trends in Rates of Pervasive Developmental Disorders

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ABSTRACT: The aims of this article are to provide an up-to-date review of the methodological features and substantive results of published epidemiological surveys of the prevalence of pervasive developmental disorders (PDD). This article updates previous reviews (1, 2) with the inclusion of new studies made available since then. The specific questions addressed in this article are: a) how are cases of PDD defined and identified in epidemiological surveys?; b) what are the best estimates for the prevalence of autism and related pervasive developmental disorders considering the methodological implications of the surveys, and c) what interpretation can be given to time trends observed in prevalence rates of PDDs given the hypothesized secular increase in PDDs?

INTRODUCTION

Selection of Studies

The studies were identified through systematic searches from the major scientific literature databases and from prior reviews (1-3). Only studies published in the English language were included. Overall, 57 studies published between 1966 and 2009 were selected which surveyed PDDs in 17 countries; half of the studies have been published since 2001. The age range of the population included in the surveys is spread from birth to early adult life, but most studies have relied on school-aged samples. There was huge variation in the size of the population surveyed (mean: 279,000; median: 44,900).

Study Designs

In designing a prevalence study, two major features are critical for the planning and logistics of the study, as well as for the interpretation of its results: case definition, and case ascertainment (or case identification methods) (4).

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Case Definition

Over time, the definitions of autism have changed as illustrated by the numerous diagnostic criteria that were used in both epidemiological and clinical settings. The first diagnostic criteria reflected the more qualitatively severe forms of autism and it is only in the 1980s that less severe forms of autism were recognized, either as a qualifier for autism occurring without mental retardation or as separate diagnostic categories within a broader class of autism spectrum disorders (ASD) denominated 'Pervasive developmental disorders' (PDD, an equivalent to ASD) in current nosographies. Current nosographies include the Diagnostic and Statistical Manual of Mental Disorders, Fourth edition, Text Revision (DSMIV-TR) (5) and the International Classification of Diseases, Tenth edition (ICD-10) (6). Whilst there is generally high agreement among experts on the diagnosis of PDD's, some differences persist between nomenclatures about the terminology and precise criteria of PDD's. In addition, in recent years, the definitions of syndromes falling on the autism spectrum have been expanded further with reference to the broader autism phenotype, which is a pattern of mild autistic developmental symptoms seen in relatives of individuals affected with a diagnosed PDD. As no diagnostic criteria are available for these mild forms of autism, the resulting boundaries with the spectrum of PDDs are left uncertain. Whether or not this plays a role in more recent epidemiological studies is difficult to know, but it is a possibility that should be considered in assessing results for the new generation of surveys.

Case Identification

When an area or population have been identified for a survey,

some studies have solely relied on existing service providers databases (7); on special educational databases (8,9,10); or, on national registers (11) for case identification. These studies have the common limitations due to relying on local access to existing services for case ascertainment. As a result, subjects with the disorder who are not in contact with these services are not included as cases, leading to an underestimation of the prevalence proportion.

Other investigations have relied on a multi-stage approach to identify cases. The aim of the first screening stage of these studies is to cast a wide net in order to identify subjects possibly affected with a PDD, with the final diagnostic status being determined at a next phase. Then, subjects identified as positive screens go through a more in-depth evaluation to confirm their case status. The source of information used to determine whether an individual has a PDD usually involved a combination of data coming from different sources (medical records, educational sources, other health professionals, etc.), with a direct assessment of the person with autism being offered in some but not all studies. Obviously, surveys of very large populations did not include a direct diagnostic assessment of subjects, as in the studies conducted in the US by the Center for Disease Control (CDC) (12,13) or in national registers (11). However, investigators could generally confirm the accuracy of the diagnosis of their cases by undertaking, on randomly selected subsamples, a more complete diagnostic workup.

When subjects were directly examined, assessments were conducted with various diagnostic instruments, ranging from a typical unstructured examination by a clinical expert (without demonstrated psychometric properties), to the use of batteries of standardized measures by trained research staff. The Autism Diagnostic Interview (14) and/or the Autism Diagnostic Observational Schedule (15) have been increasingly used in the most recent surveys.

PREVALENCE ESTIMATIONS

Autistic Disorder

Prevalence estimates for autistic disorder are summarized in Table 1. There were 47 studies, half of them published since 1999, and the sample size varied from 826 to 4.95 millions, with a median of 38,000 (mean: 217,000) subjects in the surveyed populations. The age ranged from 3 to 15 years, with a median age of 8.5 years. The male/female ratio ranged from 1.33 to 16.0 in 39 studies, leading to an average male/female ratio of 4.3:1. Prevalence rates varied from 0.7/10,000 to 72.6/10,000 with a median value of 12.7/10,000. Small-scale studies reported higher prevalence rates. A

significant positive correlation between prevalence rate and year of publication was found. Therefore, a current estimate for the prevalence of autistic disorder must be derived from more recent surveys with an adequate sample size. After exclusion of 4 studies with the smallest and largest sample sizes, the best current estimate for autistic disorder is 22/10,000. In 25 studies where the proportion of subjects with IQ within the normal range was reported, the median value was 20% (interquartile range: 17.5%-50%). In these surveys, there was a significant correlation between a higher proportion of normal IQ subjects and a higher male/female ratio (Spearman's r : 0.53; $p=0.007$), a result consistent with the association between gender and IQ in autism. Over time, there were minor associations between the year of publication of the survey and the sample male/female ratio (Spearman's r : 0.33; $p=.039$) and the proportion of subjects without mental retardation (Spearman's r : 0.34; $p=.094$). Taken in conjunction with the much stronger increase over time in prevalence rates, these results suggest that the increase in prevalence rates is not entirely accounted for by the inclusion of milder forms (i.e. less cognitively impaired) of autistic disorder, albeit this might have contributed to it to some degree.

Asperger Syndrome

Epidemiological studies of Asperger Syndrome (AS) are sparse, due to the fact that it was acknowledged as a separate diagnostic category only recently. Twelve studies (already listed in Table 1) published since 1998, have examined samples with respect to the presence of both autistic disorder and Asperger Syndrome. The median population size was 200,000 and the median age 8.25 years. The number of children with AS varied from 6 to 427, with a median sample size of 38. There was a 160-fold variation in estimated rates of AS (range: 0.3 to 48.4/10,000) that highlights the lack of reliability of these estimates. The median value was 11.0/10,000. The prevalence ratio (autistic disorder/ Asperger Syndrome) had a median value of 2.05, indicating that the rate of AS was consistently lower than that for autism. The epidemiological data on AS are of dubious quality, reflecting difficult nosological issues as well as lack of proper measurement strategies that ensure a reliable difference between AS and autistic disorder.

Childhood Disintegrative Disorder

Eleven surveys have provided data on childhood disintegrative disorder (CDD). In 5 of these, only 1 case was reported. Prevalence estimates ranged from 0 to 9.2/100,000, with a median rate of 2.0/100,000. The pooled estimate based on 11 identified cases and a surveyed population of about 604,000 children, was 1.8/100,000.

Year of publication	Authors	Country	Size of target population	Age	Diagnostic criteria	Prevalence Rate/10,000	95% CI
1966	Lotter (41)	UK	78,000	10-Aug	Rating scale	4.1	2.7 ; 5.5
1970	Brask (42)	Denmark	46,500	14-Feb	Clinical	4.3	2.4 ; 6.2
1970	Treffert (43)	USA	899,750	12-Mar	Kanner	0.7	0.6 ; 0.9
1976	Wing et al. (18)	UK	25,000	14-May	24 items rating scale of Lotter	4.8	2.1 ; 7.5
1982	Hoshino et al. (17)	Japan	609,848	0-18	Kanner's criteria	2.33	1.9 ; 2.7
1983	Bohman et al. (44)	Sweden	69,000	0-20	Rutter criteria	5.6	3.9 ; 7.4
1984	McCarthy et al. (45)	Ireland	65,000	10-Aug	Kanner	4.3	2.7 ; 5.9
1986	Steinhausen et al. (46)	Germany	279,616	0-14	Rutter	1.9	1.4 ; 2.4
1987	Burd et al. (47)	USA	180,986	18-Feb	DSM-III	3.26	2.4 ; 4.1
1987	Matsuishi et al. (48)	Japan	32,834	12-Apr	DSM-III	15.5	11.3 ; 19.8
1988	Tanoue et al. (49)	Japan	95,394	7	DSM-III	13.8	11.5 ; 16.2
1988	Bryson et al. (50)	Canada	20,800	14-Jun	New RDC	10.1	5.8 ; 14.4
1989	Suivama & Abe (51)	Japan	12,263	3	DSM-III	13	6.7 ; 19.4
1989	Cialdella & Mamelle (52)	France	135,180	9-Mar	DSM-III like	4.5	3.4 ; 5.6
1989	Ritvo et al. (53)	USA	769,620	27-Mar	DSM-III	2.47	2.1 ; 2.8
1991	Gillberg et al. (36)	Sweden	78,106	13-Apr	DSM-III-R	9.5	7.3 ; 11.6
1992	Fombonne & du Mazaubrun (39)	France	274,816	9 & 13	Clinical-ICD-10 like	4.9	4.1 ; 5.7
1992	Winyosumarto et al.	Indonesia	5,120	7-Apr	CARS	11.7	2.3 ; 21.1
1996	Honda et al. (55)	Japan	8,537	5	ICD-10	21.08	11.4 ; 30.8
1997	Fombonne et al. (40)	France	325,347	16-Aug	Clinical ICD-10-like	5.35	4.6 ; 6.1
1997	Webb et al. (33)	UK	73,301	15-Mar	DSM-III-R	7.2	5.3 ; 9.3
1997	Arvidsson et al. (56)	Sweden (West Coast)	1,941	6-Mar	ICD-10	46.4	16.1 ; 76.6
1998	Sponheim & Skjeldal (57)	Norway	65,688	14-Mar	ICD-10	5.2	3.4 ; 6.9
1999	Taylor et al. (22)	UK	490,000	0-16	ICD-10	8.7	7.9 ; 9.5
1999	Kadesjö et al. (70)	Sweden (Central)	826	6.7-7.7	DSM-III-R/ICD-10 Gillberg's criteria (Asperger's)	72.6	14.7 ; 130.6
2000	Baird et al. (58)	UK	16,235	7	ICD-10	30.8	22.9 ; 40.6
2000	Powell et al. (59)	UK	25,377	5-Jan	Clinical/ICD10/DSM-IV	7.8	5.8 ; 10.5
2000	Kielinen et al. (31)	Finland	27,572	7-May	DSM-IV	20.7	15.3 ; 26.0
2001	Bertrand et al. (60)	USA	8,896	10-Mar	DSM-IV	40.5	28.0 ; 56.0
2001	Fombonne et al. (24)	UK	10,438	15-May	DSM-IV/ ICD-10	26.1	16.2 ; 36.0
2001	Magnússon and Saemundsen (32)	Iceland	43,153	14-May	Mostly ICD-10	13.2	9.8 ; 16.6
2001	Chakrabarti & Fombonne (23)	UK (Midlands)	15,500	2.5-6.5	ICD10/	16.8	10.3 ; 23.2
2001	Davidovitch et al. (61)	Israel	26,160	11-Jul	DSM-III-R/	10	6.6;14.4
2002	Croen et al. (7)	USA	4,950,333	12-May	CDER « Full syndrome »	11	10.7;11.3
2002	Madsen et al. (11)	Denmark	63,859	8	ICD-10	7.2	5.0 – 10.0
2004	Tebreeqee et al. (62)	UK	2,536	9-Aug	ICD-10	23.7	9.6 ; 49.1
2005	Chakrabarti & Fombonne (23)	UK (Midlands)	10,903	7-Apr	ICD-10/DSM-IV	22	14.4 ; 32.2
2005	Barbaresi et al (63)	USA, Minnesota	37,726	0-21	DSM-IV	29.7	24.0; 36.0
2005	Honda et al (64)	Japan	32,791	5	ICD-10	37.5	31.0; 45.0
2006	Fombonne et al. (9)	Canada (Quebec)	27,749	17-May	DSM-IV	21.6	16.5 ; 27.8
2006	Gillberg et al. (65)	Sweden	32,568	12-Jul	Gillberg's criteria	35.3	29.2 ; 42.2
2006	Baird et al. (20)	UK	56,946	10-Sep	ICD-10	38.9	29.9; 47.8
2007	Ellefsen et al. (66)	Denmark	7,689	17-Aug	ICD-10	16	7.0; 25.0
2007	Oliveira et al. (67)	Portugal	67,795	9-Jun	DSM-IV	16.7	14.0; 20.0
2007	Latif and Williams (68)	UK	39,220	0-17	Kanner	12.7	9.0;17.0
2008	Williams et al. (69)	UK	14,062	11	ICD-10	21.6	13.9 ; 29.3
2009	Lazoff et al. (10)	Canada	23,662	5 – 17	DSM-IV	26.2	19.7 ; 32.7

Table 1: Prevalence surveys of autistic disorder

PREVALENCE FOR COMBINED PDDs

Unspecified Autism Spectrum Disorders in Earlier Surveys

Several studies performed in the 1960's and 1970's have provided useful information on rates of syndromes similar to autism but not meeting of the strict diagnostic criteria for autistic disorder then in use (1,2). At the time, different labels were used by authors to characterize these clinical pictures, such as the triad of impairments involving deficits in reciprocal social

interaction, communication, and imagination (16), and among others, autistic mental retardation (17). These syndromes would be falling within our currently defined autistic spectrum, probably with diagnostic labels such as atypical autism and/or pervasive developmental disorder – not otherwise specified (PDD-NOS). In 8 of 12 surveys providing separate estimates of the prevalence of these developmental disorders, higher rates for the atypical forms were found compared to those for more narrowly defined autistic disorder (see Fombonne, 2003, Table 3, p.172 (1)). This

group received little attention in previous epidemiological studies and these subjects were not counted in the numerators of prevalence calculations, thereby underestimating systematically the prevalence of what would be defined today as the spectrum of autistic disorders. For example, in Wing et al.'s study (1976) (18), the prevalence was 4.9/10,000 for autistic disorder, but, adding the figure of 16.3/10,000 (16) corresponding to the triad of impairments, the prevalence for the whole PDD spectrum was in fact 21.1/10,000. For historical purposes, it is important to be attentive to this earlier figure, bearing in mind that the study was conducted in the early 1970s and that autism occurring in subjects with an IQ within the normal range was not yet being investigated. Progressive recognition of the importance and relevance to autism of these less typical clinical presentations has led to changes in the design of more recent epidemiological surveys (see below), that are now using case definitions that incorporate upfront these milder phenotypes.

Newer Surveys of PDDs

The results of surveys that estimated the prevalence of

the whole spectrum of PDDs are summarized in Table 2. Of the 23 studies listed, 13 also provided separate estimates for autistic disorder (see Table 1) and other types of PDD; 10 studies provided only an estimated rate for all the PDDs combined. Sample sizes ranged from 2,536 to 4,247,206 (median: 32,568; mean: 270,026) and the median age of samples ranged from 5.0 to 12.5. The diagnostic criteria used in the studies listed in Table 2 reflect reliance on modern diagnostic schemes, such as the DSM-IV (19), the DSM-IV-TR (5) and the ICD-10 (6). In 14 studies where IQ data were reported, the proportion of subjects within the normal IQ range varied from 30% to 85.3% (median: 54.4%; mean: 55.7%), a proportion that is higher than that for autistic disorder and reflects the lesser degree of association, or lack thereof, between intellectual impairment and milder forms of PDD's. The male/female ratio ranged from 2.7 to 15.7 (mean: 5.5). There was a 6-fold variation in prevalence proportions among studies. The median rate was 61.9/10,000 and the mean rate was 66.6/10,000. This mean rate coincides with the rate reported recently for PDDs in 14 sites, in a large sample of 8-year-old US children born in 1994 (13). The CDC value represents, however, an

References	Country	Size	Age	Prevalence/10,000	95% CI
Baird et al, 2000 (58)	UK	16,235	7	57.9	46.8 – 70.9
Bertrand et al, 2001 (60)	USA	8,896	10-Mar	67.4	51.5 – 86.7 *
Chakrabarti & Fombonne, 2001 (37)	UK	15,500	7-Apr	61.9	50.2 – 75.6
Madsen et al, 2002 (11)	Denmark	---	8	30	---
Scott et al, 2002 (70)	UK	33,598	11-May	58.3 †	50 – 67 †
Yeargin-Allsopp et al, 2003 (72)	USA	289,456	10-Mar	34	32 – 36
Gurney et al, 2003 (8)	USA	---	8 – 10	52.0 † 66	---
Icasiano et al, 2004 (73)	Australia	≈ 54,000	17-Feb	39.2	---
Tebruegge et al., 2004 (62)	UK	2,536	9-Aug	82.8	51.3 – 126.3
Chakrabarti & Fombonne, 2005 (38)	UK	10,903	6-Apr	58.7	45.2 – 74.9
Baird et al, 2006 (20)	UK	56,946	10-Sep	116.1	90.4 – 141.8
Fombonne et al, 2006 (9)	Canada	27,749	5–17	64.9	55.8 – 75.0
Harrison et al, 2006 (34)	UK	134,661	0-15	44.2 ‡	39.5 – 48.9
Gillberg et al, 2006 (69)	Sweden	32,568	7-12	80.4	71.3 – 90.3
CDC, 2007a (12)	USA	187,761	8	67	—§
CDC, 2007b (13)	USA	407,578	8	66	63 – 68
Ellefsen et al, 2007 (66)	Denmark	7,689	17-Aug	53.3	36 – 70
Latif & Williams, 2007 (68)	UK	39,220	0-17	61.2	54 – 69 †
Wong et al, 2008 (74)	China	4,247,206	0-14	16.1 (1986-2005) 30.0 (2005)	---
Nicholas et al, 2008 (75)	USA	47,726	8	62	56 – 70
Kawamura et al, 2008 (21)	Japan	12,589	8-May	181.1	158.5 – 205.9 *
Williams et al, 2008 (69)	UK	14,062	11	61.9	48.8 – 74.9
Lazoff et al, 2009 (9)	Canada	23,662	17-May	80.3	68.9-91.7

Table 2: Newer epidemiological surveys of PDDs

average, and that study conducted at 14 different sites utilizing the same methodology found a three-fold variation of rate by state (13). Alabama had the lowest rate of 3.3/1,000 whereas New-Jersey had the highest value with 10.6/1,000 (13). As surveillance efforts continue, it is likely that awareness and services will develop in states that were lagging behind, resulting in a predictable increase in the average rate for the US as time elapses. These CDC findings apply to other countries as well, and prevalence estimates from any study should always be regarded in the context of the imperfect sensitivity in case ascertainment that results in downward biases in prevalence proportions.

In conclusion, the convergence of estimates around 60 to 70 per 10,000 for all PDDs combined, which translates into 1 child out of 150 suffering from a PDD, is striking especially when derived from studies with improved methodology. This is the best estimate for the prevalence of PDDs currently available; however, this represents an average figure and there is substantial variability across studies, and within studies, across sites or areas. However, some studies have reported rates that are even two to three times higher (20, 21).

TIME TRENDS IN PREVALENCE AND THEIR INTERPRETATION

The debate on the hypothesis of a secular increase in rates of autism has been obscured by a lack of clarity in the measures used by investigators to determine the occurrence of disease, or rather in their interpretation. Methodological requirements must be borne in mind whilst reviewing the evidence for a secular increase in rates of PDDs, or testing for the epidemic hypothesis. Several approaches to assess the question concerning a secular increase have been used in the literature that fall into 5 broad categories.

a. Use of Inappropriate Referral Statistics

Increasing numbers of children referred to specialist services or known to special education registers have been taken as evidence for an increased incidence of autism spectrum disorders. Upward trends in national registries, medical and educational databases have been seen in many different countries (8,11, 22, 23). However, trends over time in referred samples are confounded by many factors such as referral patterns, availability of services, heightened public awareness, decreasing age at diagnosis and changes over time in diagnostic concepts and practices, to name only a few. Failure to control for these confounding factors was obvious in some recent reports (24), such as the widely quoted reports from California Developmental Database Services (25, 26). Firstly, these reports applied to numbers rather than rates, and failure to relate these numbers to meaningful denominators left the

interpretation of an upward trend vulnerable to changes in the composition of the underlying population. Second, the focus on the year-to-year changes in absolute numbers of subjects known to California state-funded services detracts from more meaningful comparisons. Third, with one exception (see below), no attempt was made to adjust the trends for changes in diagnostic concepts and definitions. Fourth, age characteristics of the subjects recorded in official statistics were portrayed in a confusing manner where the preponderance of young subjects was presented as evidence of increasing rates in successive birth cohorts (24). Fifth, the decreasing age at diagnosis leads to increasing numbers of young children being identified in official statistics (27) or referred to already busy specialist services. Please see Fombonne, Quirke and Hagen (in press) (28) for a more elaborate analysis of these factors.

b. The Role of Diagnostic Substitution

One possible explanation for increased numbers of a diagnostic category is that children presenting with the same developmental disability may receive at one time one diagnosis, and later another diagnosis. Such substitution may occur when diagnostic categories are becoming increasingly known and recognized by health professionals and/or when access to better services is insured by using a new diagnostic category. The strongest evidence of “diagnostic switching” was produced in all US states in a complex analysis of Department of Education Data in 50 US states (23), indicating that a relatively high proportion of children previously diagnosed as having mental retardation were now identified as having a PDD diagnosis. Shattuck (2006) (23) showed that the odds of being classified in autism category increased by 1.21 during 1994-2003. In the meantime, the odds decreased significantly of being classified in the learning disability (LD) (odds ratio: OR=0.98) and the mental retardation (MR) categories (OR=0.97). However, this investigation has largely relied on ecological, aggregated data that have known limitations. Using individual level data, a new study has re-examined the hypothesis of diagnostic substitution in the California DDS dataset (29) and has shown that 24% of the increase in caseload was attributable to diagnostic substitution from mental retardation to autism. Similarly, a recent study in the UK (30) has shown that up to 66% of adults previously diagnosed as children with developmental language disorders would meet diagnostic criteria for a broad definition of PDD.

c. Comparison of Cross-Sectional Epidemiological Surveys

As shown earlier, epidemiological surveys of autism each possess unique design features which could

account almost entirely for variations in rates among studies, and time trends in rates of autism are therefore difficult to gauge from published prevalence rates. The significant correlation previously mentioned between prevalence rate and year of publication for autistic disorder could merely reflect increased efficiency over time in case identification methods used in surveys as well as changes in diagnostic concepts and practices (23, 30, 31, 32, 33). In studies using capture-recapture methods, a statistical method for indirectly estimating prevalence, it is apparent that up to a third of prevalent cases may be missed by an ascertainment source, even in recently conducted studies (34). Evidence that factors due to study methodology could account for most of the variability in published prevalence estimates comes from a direct comparison of 8 recent surveys conducted in the UK and the USA (2). In each country, 4 surveys were conducted around the same year and with similar age groups. As there is no reason to expect huge differences in rates across areas, prevalence estimates should therefore be comparable within each country. However, there was a six-fold variation in rates for UK surveys, and a fourteen-fold variation in US rates. In each set of studies, high rates derived from surveys where intensive population-based screening techniques were employed whereas lower rates were obtained from studies relying on passive administrative methods for case finding. Since no passage of time was involved, the magnitude of these gradients in rates can only be attributed to differences in case identification methods across surveys. Even more convincing evidence comes from the large survey by the CDC (13) where there was more than a three-fold variation in state specific rates (see above). However, the substantial differences reflected ascertainment variability across sites in a study that was otherwise performed with the same methods and at the same time. Thus, no inference on trends in the incidence of PDDs can be derived from a simple comparison of prevalence rates over time, since studies conducted at different periods are likely to differ even more with respect to their methodology.

d. Repeat Surveys in Defined Geographical Areas

Repeated surveys, using the same methodology and conducted in the same geographical area at different points in time, can potentially yield useful information on time trends provided that methods are kept relatively constant. The Göteborg studies (35,36) provided three prevalence estimates which increased over a short period of time from 4.0 (1980) to 6.6 (1984) and 9.5/10,000 (1988) (36). However, comparison of these rates is not straightforward as different age groups were included in each survey. Secondly, the increased prevalence in the second survey was explained by

improved detection among the mentally retarded, and that of the third survey by cases born to immigrant parents. That the majority of the latter group was born abroad suggests that differential migration into the area could be a key explanation. Taken in conjunction with a change in local services and a progressive broadening of the definition of autism over time acknowledged by the authors (36), these findings do not provide evidence for an increased incidence in the rate of autism. Two separate surveys of children born 1992-1995 and 1996-1998 in Staffordshire in the UK (37, 38) were performed with rigorously identical methods for case definition and case identification. The prevalence for combined PDDs was comparable and not statistically different in the 2 surveys (38), suggesting no upward trend in overall rates of PDDs, at least during the short time interval between studies.

e. Successive Birth Cohorts

In large surveys encompassing a wide age range, increasing prevalence rates among most recent birth cohorts could be interpreted as indicating a secular increase in the incidence of the disorder, provided that alternative explanations can confidently be eliminated. This approach was used in two large French surveys (39, 40). The surveys included birth cohorts from 1972 to 1985 (735,000 children, 389 of whom had autism), and, pooling the data of both surveys, age-specific rates showed no upward trend (40). An additional example is provided in the analysis of Gurney and colleagues' (2003) (8) study in which a sixteen-fold increase in the number of children identified with a PDD from 1991-1992 to 2001-2002 was shown to not be specific to autism since, during the same period, an increase of 50% was observed for all disability categories identified. In addition, the analysis also showed a marked period effect that identified the early 1990s as the period where rates started to increase in all ages and birth cohorts.

CONCLUSION

The recent upward trend in rates of prevalence cannot be directly attributed to an increase in the incidence of the disorder, or to an 'epidemic' of autism. There is good evidence that changes in diagnostic criteria, diagnostic substitution, changes in the policies for special education, and the increasing availability of services are responsible for the higher prevalence figures. The rise in number of children diagnosed occurred at the same time in many countries, when radical shifts occurred in the ideas, diagnostic approaches and services for children with PDDs. Alternatively, this might, of course, reflect the effect of environmental influences operating simultaneously in

different parts of the world. However, there has been no proposed and legitimate risk mechanism to account for this world-wide effect. Most of the existing epidemiological data are inadequate to properly test hypotheses on changes in the incidence of autism in human populations. Moreover, due to the relatively low frequency of autism and PDDs, variations of small magnitude in the incidence of the disorder are very likely to go undetected. Equally, the possibility that a true increase in the incidence of PDDs has also partially contributed to the upward trend in prevalence rates cannot, and should not, be eliminated based on available data.

From recent studies, a best estimate of 60 to 70/10,000 (equivalences = 6 to 7/1,000; or 0.6 to 0.7%; or 1 child in about 150 children) can be confidently derived for the prevalence of autism spectrum disorders. Current evidence does not strongly support the hypothesis of a secular increase in the incidence of autism, but statistical power to detect time trends is seriously limited in existing datasets. To assess whether or not the incidence has increased, method factors which account for an important proportion of the variability in rates must be tightly controlled. New survey methods have been developed to be used in multinational comparisons, and ongoing surveillance programs will inform soon this hypothesis. Meanwhile, the available prevalence figures carry straightforward implications for current and future needs in services and early educational intervention programs.

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