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Regular Article

Reevaluating the incidence of pervasive developmental disorders: Impact of elevated rates of detection through implementation of an integrated system of screening in Toyota, Japan

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Aim: Although recent epidemiological studies on the pervasive development disorders (PDD) appear to be reporting higher rates of incidence than previously believed, great variation in the reported figures suggests a need for review of the methodology involved. As such, a survey on the incidence of PDD was conducted and compared with data from a previous survey to examine the effects of screening and diagnostic methodology on incidence.

Methods: The incidence of pervasive developmental disorders was surveyed in all children (12 589) born between January 1994 and December 1996 in Toyota, Japan.

Results: Incidence was 1.81% and the ratio of boys to girls was 2.80. Definitive diagnoses were made between 13 months and 7 years 2 months, the average age at diagnosis being 3 years 4 months. Among the cases of PDD, children with normal or borderline intelligence amounted to 66.4%, mild mental retardation (MR) 17.5%, moderate MR 10.3% and severe MR 5.8%.

Conclusion: An approximately 11-fold increase was noted in prevalence of PDD compared to a previous survey two decades ago, and two main factors were believed to account for this apparent sharp increase. First, inclusion of high-functioning subjects detected during infancy, and second, higher rates of diagnosis resulting from an integrated process of screening.

Key words: autism, autistic disorder, epidemiology, incidence, PDD.

SINCE THE FIRST epidemiological study on infantile autism was published in the UK,[1] many surveys have been conducted to date, reporting greatly varying incidence or prevalence. Autistic disorder was generally considered a rare condition with a prevalence of around 2 per 10 000 children in the 1970s. But higher rates have been suggested for some time[2] despite the difficulty of making direct comparisons given the various labeling involved, reflecting transition in the diagnostic standards over time. Of particular note is the English survey published in 2006.[3] In that report, of 56 946 children between 9 and 10 years old residing in a particular region in southern England, 255 children diagnosed with pervasive development disorders (PDD) and 1515 possible cases were subject to further screening. This produced an incidence of 116.1 subjects with PDD/10 000, a higher rate than hitherto believed (Table 1). Whether the apparent rise in incidence starting in the 1990s reflects actual increase or methodological differences in diagnosis and research[11] is as yet unclear. Increasing public awareness of the disorder, including association with high intellectual capacity as well as other types of disorders, and the development of specialist services have been cited as

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Table 1. Prevalence studies

	Year	Prevalence/10000	Diagnosis
Lotter[1]	1966	4.5	Infantile autism (Kanner)
Ishii & Takahashi[4]	1983	16.0	Infantile autism (DSM-III)
Sugiyama & Abe[5]	1989	13.0	Infantile autism (DSM-III)
Honda *et al.*[6]	1996	21.1	Childhood autism (ICD-10)
Kadesjo *et al.*[7]	1999	60.0	Childhood autism (ICD-10)
Baird *et al.*[8]	2000	30.8	Childhood autism (ICD-10)
Chakrabarti & Fombonne[9]	2001	16.8	Autistic disorder (DSM-IV)
Honda *et al.*[10]	2005	27.2	Childhood autism (ICD-10)
Baird *et al.*[3]	2006	116.1	PDD (ICD-10)

PDD, pervasive developmental disorder.

possible reasons for the apparent increase in incidence of PDD, apart from a possible true increase in number. But results indicating that the lower the educational level of the parents, the lower the level of identification, suggest the great significance of screening and diagnostic methodology on determination of incidence rates.

Given this unclear situation, the present study was designed to assess the incidence of PDD in Toyota, Japan, and is distinctive in the following two aspects. First, the 18- and 36-month general health examinations for all children born in the city were utilized as opportunities for the initial screening. A system has been established wherein children thus identified as having the possibility of PDD are advised to consult the Toyota Municipal Child Development Center (TCDC), at which they can be followed from early childhood through early support programs and diagnosed as required. We believe this combined approach has contributed to lowering the possibility of children slipping by undetected. Second, the findings are discussed through comparison with data from an earlier epidemiological study in the same region[4] by one of the authors (Takahashi), investigating the impact of screening systems and research methodology on epidemiological data.

METHODS

Region demographics

The study was conducted in Toyota, Japan, a city in central Japan with a population of 355 711 as of January 2002, at the time of the survey.

Identification of children with PDD

Public health examination system for children in Toyota

The 18-month checkup was used as the opportunity for initial screening, to identify children with disorders in psychological development including PDD. The 18-month checkup consists of parent interviews, direct observation, and examination by public health nurses. Items such as whether one's gaze is met naturally, enunciation of three or more meaningful words, ability to finger-point, build with blocks etc. are reviewed. In Toyota a checklist has been developed independently to eliminate difference in standards arising from subjective evaluation by caregivers or the attending public health nurse (Table 2).

The screening and support system is depicted in Fig. 1.

Toyota Municipal Child Development Center

Children suspected of some developmental disorder through this 18-month or other health examinations are encouraged to participate in the early parent-child support group *Aozora* established in 1981, then integrated into TCDC founded in April 1996. The aim of Aozora is to support parents with concerns regarding their child's development, difficulties handling problem behavior such as intense tantrums, or children with problems playing with other children, through provision of advice and assistance in disciplining in actual situations of play between parents and children. Among all subjects in this survey born between 1994 and 1996, 4.6% have participated in the group at some point. When disorders such as

Table 2. Questionnaire used in the 18-month checkup (extract)

1.	Does your child react/answer when you call him/her?	Yes/No
2.	Are you worried about your child's eyes or vision?	Yes/No
3.	Can your child pile 2 or 3 little wood blocks (2–3 cm each)?	Yes/No
4.	Does your child scribble with a pencil?	Yes/No
5.	Does your child point out animals or objects when you read a book for him/her?	Yes/No
6.	Does your child understand simple requests?	
	(1) Throw it in the garbage	Yes/No
	(2) Bring this/that. . . (an object)	Yes/No
7.	When did your child start speaking meaningful words?	Yes/No
	How many words can he/she speak now?	Yes/No
8.	Which are his/her favorite toys, games, plays?	
9.	Does your child show interest in playing with other children?	Yes/No
10.	Does your child imitate adults?	Yes/No
11.	Does your child ask you to play with him/her?	Yes/No

PDD are suspected through observing the child or the parent–child relationship, the parents are advised to consult a child psychiatrist with the Nozomi Medical Clinic, the medical division of the TCDC, specializing in the treatment of developmental disorders.

Network with nursery schools and kindergartens

As noted out by Sugiyama and Abe,[5] it is difficult to reliably identify and classify mild to moderate speech disabilities at 18 months. Almost all children with PDD are referred to the Nozomi Medical Clinic via the 18-month checkup and the early support group Aozora, but there still remain some who are false negative and those who select not to participate in the support group. In either case, all children are again screened at the health checkup for 3-year-olds, although a number of children with PDD do end up entering nursery schools or kindergarten before diagnosis, even with these repeated measures. But once there, stepped-up staff awareness cultivated by the TCDC through its support system for nursery schools and kindergartens enables the identification of possible cases by staff members, who then encourage the parents to consult a child psychiatrist.

Subjects

Children born between January 1994 and December 1996 were selected as the subjects. According to a preliminary survey, we found that the number of children with PDD consulting child psychiatrists for the first time after age 5 was minimal. Thus, it was believed that examination of the patients brought

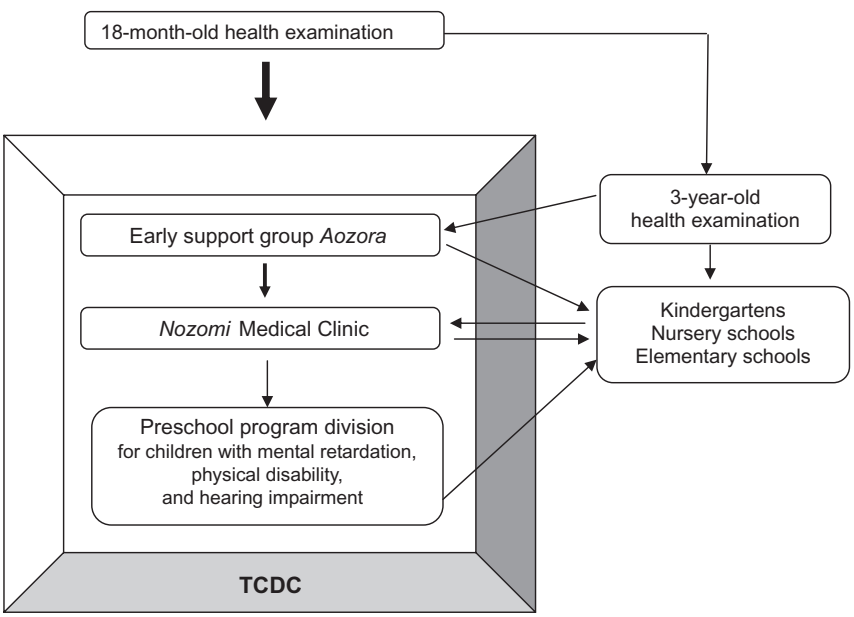

Figure 1. Screening and support system in Toyota, Japan.

Table 3. Incidence of PDD

Year of births	Subjects			Cases			Incidence	Ratio of boys to girls
	Boys	Girls	Total	Boys	Girls	Total		
1994	2196	2111	4 307	56	15	71	1.65%	3.73
1995	2063	2002	4 065	50	12	62	1.52%	4.17
1996	2128	2089	4 217	62	33	95	2.25%	1.88
Total	6387	6202	12 589	161	56	228	1.81%	2.80

to medical attention before age 5 should allow for analysis of incidence, because diagnosis of most patients should have been finalized by that time. Consequently, the initial consideration was to select children older than 5 years of age as of April 2002 when the survey was conducted. Next, because the TCDC is also attended by children who have transferred into the city, the present subjects were limited to the 12 589 children (6387 boys, 6202 girls) born in Toyota in the 3-year period between 1994 and 1996 for calculation of incidence rate, circumventing the effect of population influx and outflow. Naturally there are children born in Toyota who fall through this screening due to transfers outside the city. Although the number of such subjects is believed to be minimal because screening starts at 18 months of age, it is possible that the actual incidence may be somewhat higher than the current result.

Diagnosis and testing for IQ

Given the screening system described in the previous section, most children suspected of having PDD are believed to have visited the TCDC. There, a child psychiatrist – one of the authors – interviewed parents, observed each child directly for 90–120 min, and rendered diagnoses according to DSM-IV criteria.[12] The examinations were carried out in a playroom where the child was allowed to play freely.

As a rule the Tanaka–Binet test (Japanese adaptation of the Stanford–Binet) has been used to assess intelligence. But the Enjoji Infantile Developmental Test (similar to the Denver Developmental Screening Test) was used for children with mental retardation because the Tanaka–Binet is not suitable for children with marked retardation, given absence of items on the 0-year level. When the tests had been given on several occasions, the latest result was used in the survey.

Ethical considerations

The survey was reviewed and deemed ethically appropriate by the ethics committee of the TCDC in accordance with the guideline for epidemiological research set forth by the Ministry of Health, Labor and Welfare, and the provisions regarding use of personal information decreed by Toyota City.

RESULTS

Incidence of PDD

The total number of children with PDD was 228 (168 boys, 60 girls). Incidence was 1.81% and the ratio of boys to girls was 2.8. The age at which definitive diagnoses were made was between 13 months and 86 months, the average being 40 months of age (Table 3). Diagnoses that had been made at an early age were subject to reconfirmation according to DSM-IV standards through review of past interview records.

Intelligence testing

Table 4 shows the IQ/DQ distribution of 223 patients (excepting five patients not subjected to testing for various reasons). The age at which the tests were conducted ranged from 33 to 93 months, the average being 67 months old.

The Tanaka–Binet test was used in 85% of cases, and the Enjoji Infantile Developmental Test in 15%.

Among the 223 patients, children exhibiting average intelligence of IQ/DQ >85 amounted to 52.9%; inclusion of subjects with borderline intellectual functioning (IQ/DQ >70) raises this figure to 66.4%. Mild mental retardation (IQ/DQ 50–69) was found in 17.5%, moderate mental retardation (IQ/DQ 35–49) in 10.3%, and severe mental retar-

Table 4. Distribution of IQ and DQ

IQ/DQ		<34	35–49	50–69	70–84	>85
IQ	Male	0	8	22	24	85
	Female	0	1	9	4	33
	Total	0	9	31	28	118
DQ	Male	10	11	6	0	0
	Female	3	3	2	2	0
	Total	13	14	8	2	0
Total	Male	10	19	28	24	85
	Female	3	4	11	6	33
	Total	13	23	39	30	118
		5.8%	10.3%	17.5%	13.5%	52.9%

dation (IQ/DQ <35) was noted in 5.8%. No difference was noted in gender regarding distribution of intelligence (Table 4).

DISCUSSION

Comparison with a previous study in Toyota

A survey on the prevalence of infantile autism was conducted in Toyota in 1981.[4] That study differs from the current survey in terms of subject age and standards for diagnosis, and does not allow for direct comparison regarding incidence of PDD. Nevertheless, the studies, conducted in the same area, reflecting changes in the system of screening, are is crucial in considering support for children with PDD, who comprise increasingly larger proportions of the population, meriting the comparison despite the limitations involved. In the 1981 study a questionnaire was used as the initial screening reference. In that survey the staff involved were neither specialists in mass screening, nor did they have specialized knowledge of the developmental disorders. This time the results were obtained from mass screening by screening specialists at 18 months, with the additional opportunity of following up on most children identified with autistic features.

Comparing these two methods allows for insight into the apparent drastic increase of children with PDD. In the previous survey the subjects were elementary school children living in Toyota in May 1981, that is, children born between April 1969 and March 1975. The initial screening was in the form of teacher questionnaires. Two child psychiatrists then interviewed parents and observed all children directly, and determined whether they met criteria for infantile autism listed in Table 5. DSM-III (1980)[13] did not yet contain a listing of concrete symptoms, and both availability and recognition of the standards were still low in the years before publication of the Japanese translation in 1982. For this reason a group of Japanese specialists had convened to compile a clinical picture of autism[14] (of which one of the authors was a member), which formed the basis of the diagnostic standards used at the time.

In that study 56 children fulfilled criteria for infantile autism. Prevalence was 0.16% and the male : female ratio was 6:1. Mean age at diagnosis was 8 years. Of the 56 children, severe mental retardation was seen in 31, moderate to mild retardation in 16, and only nine exhibited above-borderline levels of intelligence.

The following differences are seen in the results of the two surveys. First, while the previous survey identified 0.16% of the children as having infantile autism, the current survey was able to identify and support 1.81% with PDD. Second, while most of the children identified in the previous survey were those with moderate to severe mental retardation, a very large increase is seen in the proportion of children with above-borderline intelligence.

In order to investigate the factors behind this large increase in PDD children with above-borderline intelligence, raw data from the 1981 survey were re-examined to examine why the nine children comprising the high-functioning group had been identified in the 1981 survey. This showed that six of the nine had exhibited clear symptoms such as hyperactivity or inability to join groups of other children that were obvious to any teacher, in addition to frequent appearance of symptoms such as echolalia, or oblique gaze. The other three children had been capable of taking part in group activity to the extent of participating in regular classes, but two had already been diagnosed in other institutions during infancy. In other words, initial screening in the form of teacher questionnaires in the elementary school years used in the previous survey was capable of identifying only those children with clear mental retardation or problems in group behavior such as hyperactivity that would have been obvious to anyone in the group situation.

Therefore, although our analysis does not rule out a true increase in high-functioning subjects, it appears more reasonable to assume that close observation of behavior by trained professionals is requisite for the accurate identification of high-functioning cases in early childhood.

Table 5. Diagnostic standards used in the 1981 survey

Criteria were regarded as fulfilled by the appearance of more than one item in each of A, B, C and D before age 3.
A. Disturbance in interpersonal relationships
(1) Delay in response to being called by name.
(2) Avoidance of eye-to-eye gaze.
(3) Does not point to things, but guides people by the arm to the object of their wishes.
(4) Dislikes intervention by others.
(5) Can relate to adults, but shies from interaction with peers.
(6) Hyperkinesis (escapes from the classroom in kindergarten).
(7) Cannot apply the rules of conduct on one's own, capable of understanding the situation only after watching others acting accordingly.
(8) Addresses people, even strangers, with undue familiarity.
(9) Cannot tackle tasks they are incapable of doing, turning away or voicing dissent.
B. Disturbance in speech and language
(1) Severe delay in understanding and speech
(2) Echolalia
(3) Talks to oneself voicing phrases entirely irrelevant to the situation (e.g. TV commercials and such)
(4) Pronominal reversal
(5) Can only answer to set questions; peculiar/idiosyncratic answers to questions
(6) Monotonous upward inflection in pronunciation and sentence delivery.
(7) Pronounced lack in reading skills and solving problems in applied mathematics.
(8) Limited ability with sentence construction and writing compositions
C. Insistence on the preservation of sameness or resistance to change
(1) Dexterity in manipulation of objects, string, paper, water etc.
(2) Arranges things in a line, and displays anger when it is moved around.
(3) Obsessive questions related to child's particular preoccupation.
(4) Cannot stand having succession in time or arrangement in space changed.
(5) Becomes unstable or panics unable to adapt to new situations.
(6) Limited or deviated interest in objects (trade-marks, calendars, train timetables, and certain picture books, etc.)
D. Sensory/motor disturbances
(1) Head tilting and looking askance at objects or persons.
(2) Fluttering or holding hands aloft in front of face.
(3) Fondness for high places.
(4) Only glances at objects.
(5) Will not look directly at what they are doing when working with their hands.
(6) Severely unbalanced diet.
(7) Dull response to pain and cold.
(8) Likes to whirl around (and does not become dizzy doing so.)
(9) Walks on toes.
(10) Has difficulty copying other people's movements.

The screening and support system currently available in Toyota has undergone great improvement in the two decades between this and the previous survey. The checkup for 18-month-olds incorporated in 1977 did not exist when the subjects of the previous survey were infants. Moreover, establishment of the early intervention system is considered an integral factor. As stated previously, the early support group Aozora began operation in 1981. The number of children participating in the program was 39 in the first year. In 1996 it was integrated into the TCDC, and in 2002, at the time of the survey, more than 350 children aged 1–3 were participating in the program.

The institution of the Nozomi Medical Clinic has been another essential factor. Before establishment, patients had to visit hospitals outside the city to be diagnosed. Following establishment of the TCDC in 1996, child psychiatrists have been posted at the Clinic within the TCDC. The early support group Aozora and the Nozomi Medical Clinic are located in the same building, allowing for ready coordination between the two.

Comparison with other regions

As another angle for examining the Toyota approach, our findings are first compared with that from a study conducted in Yokohama, Japan, using a comparable system of screening.

In Yokohama the incidence of autistic disorder has been reported at 0.211–0.272%,[6,10] which is low compared to that in the present study. They also use the 18-month health examination for the initial screening. The screening system is similar to that in Toyota, that is, it is also combined with an early support group for PDD, although participation in the group is after the children have been seen by a child psychiatrist, and the program itself is not regarded as an opportunity for auxiliary screening. The scale of the group is smaller and the term is short, and thus does not fulfill the secondary screening role allocated to the support program in Toyota.

In Toyota many children suspected of having PDD take part in the early support group. Nursery school teachers in charge of the group observe the children and parents in the long term, enabling them to discern the features of PDD in children who may otherwise escape detection, recognized only as being 'difficult'. In other words, while the Yokohama group provides post-diagnosis intervention in specialized facilities, the objective of the Toyota group is different in that it aims to provide support with day-to-day childcare.

It is believed these differences in the system of identifying PDD, which may appear minor at first glance, may actually be playing a major role in contributing to lowering the number of children slipping by unidentified in Toyota, compared to the Yokohama approach.

Next, in comparison with a recent study by Baird *et al.* citing high figures in a cohort study conducted in the UK,[3] their prevalence rate of 1.161% is considerably higher than previous reports, although lower than our estimation of 1.81%. A possible reason for this discrepancy, as in the case of the Toyota study in 1981, is difference in the age of subjects and method of initial screening. In the Baird *et al.* study, among 56 946 children between 9 and 10 years old in a particular region, 255 already diagnosed as having PDD, and 1515 identified as having special educational needs without a diagnosed disorder, were subjected to further analysis. Because diagnosis in the present study started with screening of all subjects early on, it is to be expected that such subjects identified at 18 months will include children who will not have special educational needs upon entering school, thus raising our initial figure.

Furthermore, a survey in Atlanta, Georgia on children between 3 and 10 years old reported overall prevalence at 0.34%,[15] considerably lower than the present survey. Exclusion of subjects with above-average intelligence (IQ > 70) is believed to factor largely in this difference. According to age group, prevalence among 3–4 years olds is low, rising to 0.41–0.45 between ages 5–8, and becoming lower again from age 9–10. This disparity is believed to be associated with autistic disorders not yet being identified at age 3–4; and the low rate among those between 9 and 10 years old is thought to be a reflection of the services provided to children with autistic disorder in the 1990s. That such systematic approaches through identification to support should affect outcome in epidemiological surveys is a phenomenon in common with our current survey.

Limitations

Within this renovated framework of screening a number of situations can be envisioned leading to children falling through the screening process: transfers out of the city, parents or guardians not wishing for diagnoses, or symptoms being so light as to escape detection even by the specialist.

Next, there is the issue surrounding the diagnostic standards themselves. According to diagnosis at first presentation, 217 children (161 boys, 56 girls) were diagnosed with autistic disorder, seven children (four boys and three girls) with Asperger's disorder, and four children (three boys and one girl) with PDD not otherwise specified (PDD-NOS). Incidentally, no cases of Rett's disorder or childhood disintegrative disorder, also listed under PDD according to DSM-IV, were found in the present sample. There have been a number of studies on the stability of diagnosis of PDD. Both Chawarska *et al.* and Stone *et al.* report a relative stability of autistic disorders and instability in diagnosis of PDD-NOS,[16,17] indicating that diagnoses may not necessarily be stable regarding the subclassifications of PDD. Because the present study was based on symptoms at first presentation, and because age at diagnosis was not controlled, the findings are reported as overall prevalence rate of PDD.

And last, although the elevation in detection rate is clear, it does not rule out the possibility of a true rise in the incidence of PDD.

CONCLUSION

The principal factor behind the high incidence of PDD found in this survey is believed to be the presence of the improved system for screening and support encompassing both infants and the high functioning subjects that are more valid and functional compared to the previous format in Toyota city, and the systems in other regions.

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