

ORIGINAL ARTICLE

Prevalence of tic disorders among primary school students in the city of Pavia, Italy

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Arch Dis Child 2004;**89**:45–47

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Accepted 2 March 2003

Background: The prevalence of tic disorders in children varies from 1% to 29% depending on the characteristics of the study population, the diagnostic criteria, and the study design and methods.

Aims: To calculate the prevalence of tic disorders among primary school children in Italy.

Methods: The study population comprised 2347 primary school children from the city of Pavia (pop. 80 073), Northern Italy. Using trained school teachers as the source of cases, all children with motor or vocal tics occurring intermittently and unpredictably out of a background of normal motor activity were accepted. The type, frequency, and circumstances of tic disorders were noted. School performance was correlated to the presence of tics.

Results: A total of 68 children (56 boys, 12 girls) aged 6–11 years were identified with tic disorders. The period prevalence was 2.9% (95% CI 2.3 to 3.7). The prevalence was 4.4% in boys and 1.1% in girls, with no detectable trends at age 6–11. Motor tics were present in 46 cases, vocal tics in 6, and motor and vocal tics in 16. Situation related tics were noted in 37 cases. A significant correlation was found between the presence of tic disorders and impaired school performance.

Discussions: Tic disorders are a fairly uncommon but disabling clinical disorder among primary school children from an urban community. The fairly low prevalence of this clinical condition, as compared to other reports, can be explained by the choice of stringent diagnostic criteria and the exclusion of patients with other movement disorders.

Estimates of the prevalence of tic disorders in children vary from 1% to 29% depending on the characteristics of the study populations, the diagnostic criteria, and the methods of case ascertainment.^{1–12} First, information can be obtained through parent questionnaires or interviews, school surveys, or medical examinations. The lowest rates have been found in populations subjected to mail interview and the highest in selected populations (for example, children with psychiatric disorders with or without epilepsy, parents with tics, or patients attending special classes). Second, although most studies assessed the prevalence of Tourette's syndrome (TS) in regular or special school populations, transient tics, chronic motor tics, and other adventitious movements are not always differentiated. Third, epidemiological indices varied across studies, which assessed incidence, point or period prevalence, or cumulative incidence. Finally, the varied descriptions of tics preclude a definite, differential diagnosis for many tic conditions.

The aim of the present study was to obtain the period prevalence of tic disorders among primary school students in Pavia, a small town (pop. 80 073) in Northern Italy.

METHODS

This is an observational population based survey. The study population included 2717 children aged 5–12 years from 15 primary schools. Of these, three were excluded (for a total of 370 children) as they declined participation. The purposes of the study and the definition and characteristics of the tic disorders were illustrated by one of the authors (CT) to all the school teachers, with the support of videotaped interviews. The cases illustrated in the videotapes included examples of vocal and motor tics as well as the main disorders considered in the differential diagnosis (stereotypies, dystonias, etc). Diagnostic criteria for a tic disorder were those of the Tourette Syndrome Classification Study Group.¹³ According to this classification, tics are brief movements (motor tics) or

sounds (vocal tics) that occur intermittently and unpredictably out of a background of normal motor activity. In the present study we also attempted to identify patients with probable TS—that is, with both motor and vocal tics.¹³ The school teachers used semistructured questionnaires to list each student with reference to the main demographic characteristics, the school performance (assessed by a panel of teachers on the basis of cognitive and behavioural functions and coded arbitrarily as normal, mildly impaired, and severely impaired), and the presence of tics. For children with tic disorders, the clinical features (type and frequency) and the most commonly associated circumstances were noted. The screening procedure started in December 1995 and ended in June 1996.

A pilot study was conducted on 232 children from one school, evenly distributed across school years (two classes for each school year). This sample served to test the validity and reliability of the school teachers as a source for the ascertainment of patients with tics. In each class, one of us with experience in the field of movement disorders (CT) spent four morning hours observing the students and noted any symptoms resembling tics. To prevent any emotional impact on the children, the expert was introduced as a teacher. The school teachers' ability to confirm or exclude tic disorders was matched against that of our expert (chosen as the standard) and used as a measure of the validity of the screening technique. Inter-rater agreement was tested by asking two teachers to examine the same classes. Based on the results of the pilot study, the sensitivity of the school teachers (obtained after matching with the expert) was 80% and the specificity was 97%. The measure of the teachers' inter-rater agreement was satisfactory (kappa 0.74).

During the pilot study, the school medical records of each child in the target population were reviewed in a search for cases with tic disorders who were not otherwise identified, but none were traced. The methods used to train school

teachers in the pilot study were then applied in the other schools. Each teacher received the same information and was taught how to identify patients with tic disorders, excluding other movement disorders, and how to complete the questionnaires. Fifteen days after completion of the pilot study, on receiving the completed forms, the investigators discussed any controversial case with the teachers and any problem arising with data collection.

Ninety five per cent confidence intervals (95% CI) were calculated for the prevalence estimates, based on the Poisson distribution.¹⁴

The parents of each school child enrolled in the pilot study signed an informed consent form delivered to their homes through the local school authorities. In each school, the parents' committees, elected to represent each family's child, and their teachers signed the study protocol. The study was approved by the parent-staff committees, which act as ethical committees in each school. The anonymity of each child was rigorously preserved, in line with national privacy legislation.

RESULTS

Only one school head (representing 6% of the total primary school children of Pavia) decided not to participate in the study. The target population included 1274 boys (54%) and 1073 girls (46%) aged 5–12 years. A total of 68 children with tic disorders were identified by the school teachers through our screening procedure, giving a prevalence rate of 2.9% (95% CI 2.3 to 3.7). The prevalence was 4.4% (95% CI 3.3 to 5.7) in boys and 1.1% (95% CI 0.6 to 1.9) in girls. The children with tic disorders were evenly distributed in each school and class.

The sample comprised 56 boys (82%) and 12 girls (18%) aged 6–11 years. Twenty two children (33%) were 6–8 years old and 46 (67%) were 8–11 years. The prevalence rate at age 6 was 2.5% (10/395). The corresponding rates at age 7, 8, 9,

10, and 11 were 1.9% (9/460), 3.1% (14/451), 4.9% (24/489), 2.7% (9/331), and 1.5% (2/128). Forty six cases (67%) presented motor tics, 6 cases (9%) had vocal tics, and 16 (24%) had motor and vocal tics. Situation related tics (reading, writing, etc) were present in 37 cases (55%). Eleven patients (16%) complained of rare tics, 31 (46%) had tics sometimes during the day, and 26 (38%) several times a day. School performance was normal in 40 children (59%), mildly impaired in 16 (23%), and severely impaired in 12 (18%). The corresponding values in children without tic disorders were 1937 (85%), 250 (11%), and 92 (4%) (Mantel-Haenszel χ^2 for trend 41.8; $p < 0.00001$).

DISCUSSION

This study aimed to calculate the prevalence of tic disorders in a fairly large sample of primary school children living in a urban community. The prevalence (2.9%) was lower than that reported by others,^{1–8 11 12} the only studies with a rate lower than ours being those of Caine and colleagues⁹ and Comings colleagues.¹⁰ The difference may be largely explained by the use of different diagnostic criteria and data sources (table 1). In contrast with others,^{1–8} we used more stringent criteria and excluded patients with other movement disorders, such as stereotypic behaviours. In the published reports, different sources have been used to identify subjects with tic disorders, including direct observation at school,^{1 12} parents' interview^{2 3 5 9} or questionnaire,^{4 6–8 11} teachers' interview⁵ or questionnaire,¹² and clinical examination.^{4 9 11} Persons in close contact with the children (parents and teachers) may be the most reliable observers. However, the sensitivity and specificity of parents require validation, and training parents is time consuming. Interestingly, according to MacFarlane and colleagues² and Pringle and colleagues,⁴ mothers tend to overestimate tics compared to doctors in a medical interview. By contrast, physicians tend to be more

Table 1 Epidemiological studies on the prevalence of tic disorders in children

First author, year (ref)	Country	Age (y)	Screening procedure	Sample size	Mannerisms and stereotypies	Point prevalence (%)	Period prevalence (%)
Boncour, 1910 ¹	France	2–13	Teacher observation	1759	+	23.7	–
MacFarlane, 1954 ²	USA	1–14	Parent interview	116	+	–	25
Lapouse, 1964 ³	USA	6–12	Parent interview	482	+	12	–
Pringle, 1967 ⁴	UK	7	Parent questionnaire	7949	+	7.7	–
			Medical history and examination	7958		5.2	–
				7965		4.1	–
Rutter, 1970 ⁵	UK	9–12	Parent and teacher interview	2057	+	–	–
			Non-psychiatric sample	1919		4	–
			Psychiatric sample	95		18	–
			Non-psychiatric epileptic sample	26		8	–
			Psychiatric epileptic sample	17		24	–
Shepherd, 1971 ⁶	UK	5–15	Parent questionnaire	6290	+	–	20
Achenbach, 1978–9 ^{7 8}	USA	6–16	Parent questionnaire	2200	–	–	–
			Population sample	1100		7.7	–
			Referral sample	1100		28.8	–
Caine, 1988 ⁹	USA	5–18	Parent interview				
			Physicians				
			Other health professionals	41	+	–	0.03
			School personnel				
			Local news media				
			Medical history and examination				
Comings, 1990 ¹⁰	USA	7–14	Classroom observation; teacher and/or parent interview	3034	–	1.4	–
Kurlan, 1994 ¹¹	USA	9–15	Parent questionnaire and medical examination	70	–	–	–
			Normal classes	35		4.9	–
			Special classes	35		21.9	–
Mason, 1998 ¹²	UK	13–14	Parent questionnaire	111	–	18	–
			Pupil questionnaire	152		–	–
			Teacher questionnaire	147		–	–
			Classroom psychologist observation	166		–	–
Present study	Italy	6–11	Teacher observation (after training)	2347	–	–	2.9

Modified from Shapiro *et al.*¹

specific but less sensitive than parents or teachers because they spend less time with the children. This assumption is confirmed by others⁴ and by the medical records of our school children, in none of whom were any movement disorders reported.

In our study, teachers were instructed how to identify tics as the accuracy of teachers may be poor in the absence of proper training. Our method of case ascertainment can be considered the most serious flaw of the study, as there was no attempt to verify the teachers' observations through direct examination of all the affected children. However, based on the results of the pilot study, the method proved fairly accurate and reliable. In addition, we do not know whether teachers not participating in the pilot study were matched to their colleagues in terms of motivation and skills. However, participants in the pilot study were not volunteers, supporting the assumption that teachers involved in this part of the study were comparable to the other study participants. Further comments arise from comparison of our findings with other recent epidemiological surveys, in which the prevalence rate of tic disorders was 1.4%,¹⁰ 4.9%,¹¹ and 18%¹² (see table 1). In the only studies giving a prevalence of tics lower than ours,^{9, 10} patients were included if they satisfied the DSM-III diagnostic criteria after a detailed medical interview and medical examination,⁹ or were referred for psycho-educational assessment.¹⁰ Conversely, the two studies giving higher rates^{11, 12} were conducted in small samples of children, for which a chance overestimate may be likely. Interestingly, in our study the prevalence of motor and vocal tics (probable TS) was 0.7% (16/2347). This is in keeping with the 0.1–1.1% rates of TS reported in the United States by Comings and colleagues¹⁰ and in Sweden by Kadesjo and Gillberg,¹⁵ which tend to confirm the accuracy of our screening procedure. Several studies reported tics and TS were more common among males.^{4, 9, 10} The association between tic disorders and impaired school performance is also an expected finding. TS has been associated with poor school performance¹⁶ which may be related to a combination of factors like tic severity, use of medications, executive dysfunction, stigmatisation, and coexisting attention deficit disorders, hyperactivity, obsessive-compulsive behaviour, or other psychopathological conditions.¹⁷ In addition, susceptibility genes may be implicated in the liability to many neurodevelopmental disorders, including TS.¹⁸ These correlations could not be assessed here because the collection of data regarding the clinical characteristics of tic disorders (including co-morbidities and related treatments) was beyond the scope of the study. In conclusion, the even distribution of our cases in each school and class, the choice of stringent diagnostic criteria, the use of fairly accurate and reliable data sources, and the size of the target population, all tend to support the validity of our study design and show that tic disorders are a fairly uncommon clinical but disabling

disorder among primary school children in an urban community.

ACKNOWLEDGEMENTS

The study was supported by grant ICS 57.2/RF 94.126 from the Italian Ministry of Health. The authors wish to thank the personnel of the entire school district for their generous assistance, Ms Judy Baggott for editorial help, and Miss Susanna Franceschi for typing the manuscript.

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