Eur Child Adolesc Psychiatry (2007)
DOI 10.1007/s00787-007-0651-y

ORIGINAL CONTRIBUTION

Prevalence of tic disorders among schoolchildren in Warsaw, Poland

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Accepted: 8 August 2007
Published online: 14 September 2007

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Abstract *Objective* To estimate the prevalence and to describe clinical characteristics of tic disorders in 12–15 year old Warsaw schoolchildren. *Method* Children attending 24 randomly selected schools were screened by inquiring their parents and teachers. Children indicated as tic-positive by the screening procedure were investigated using semi-structured questionnaires and the Polish version of YGTSS scale. A validity study involved random selection and investigation of 130 non-indicated subjects. *Results* Out of 1,579 screened children, 104 met criteria for tic disorders, giving a lifetime prevalence of 9.9% (95% CI 7.1–12.6%) and a point prevalence of 6.7% (4.3–9.1%). Lifetime prevalence of ICD-10 tic disorders was 2.6% (95% CI 1.2–4.1%) for transient tic disorder (TTD); 3.7% (1.9–5.4%) for chronic tic disorder (CTD); 0.6% (0.2–0.9%) for Tourette disorder (TD); and 2.9% (1.2–4.6%) for non-specific tic disorder. Screening procedure had high sensitivity (92%) and low positive predictive value (18%). *Conclusion* Tic disorders are common among Warsaw schoolchildren, have mild severity and form a continuum. The present study has confirmed numerous problems with studying neurobehavioral disorders in general population not referred to physicians, and stressed out the need to improve education on tic disorders in the general public.

Key words childhood tic disorders – neurobehavioral manifestations – prevalence – Poland

Introduction

Tic disorders are the most common movement disorders in children. Their etiology involves complex interactions between genetic and environmental factors that influence neurotransmitters homeostasis [13]. According to present classification, four syndromes are distinguished: transient tic disorder (TTD), chronic tic disorder (CTD), Tourette disorder (TD) and non-specific tic disorder (NSTD) [1, 20]. According to current disease classifications, tic disorders are a continuum of disturbances with different expression of tics and related symptoms [17]. Also, several psychiatric syndromes exhibit co-morbidity with tic disorders, including attention-deficit hyperactivity disorder (ADHD), obsessive-compulsive disorder (OCD) and other anxiety and mood disorders [6, 8, 15].

The epidemiology of TD was extensively studied since the early 80s. Historically, most of studies of TD prevalence were based on review of medical documentation from hospitals and ambulatories. They

typically involved patients with most severe symptoms and used diverse case classifications (from DSM III to DSM-IV). The TD prevalence established in these studies ranged between 0.5–0.7 per 10,000 [3, 15] and 15 per 10,000 [8]. According to recent studies utilising widely accepted methodology of screening large population samples and administering comparable protocols to confirm and classify cases, Tourette syndrome appeared to be a more prevalent and milder disease, than was previously thought [2, 7, 9]. Tourette disorder prevalence established in population-based studies ranged from 4.3 per 10,000 [2] to 76 per 10,000 [7]. Only few population-based studies estimated the prevalence of specific tic disorders using modern syndrome classification. In a Japanese screening of 1,321 schoolchildren, the established overall point prevalence of tic disorders was 8.3%, and the prevalence of particular syndromes was: TTD—5.1%; CTD—2.2%, TD—0.5% [12]. A recent Swedish study investigated a population sample of 4,479 children aged 7–15 years and established an overall point prevalence of 6.7%, TTD prevalence of 4.8%, CTD prevalence of 0.8% and TD prevalence of 0.6% [9].

There are multiple problems related to studying tic disorders epidemiology, including subjective diagnosis with no confirmatory test, different perception of tic disorders in different cultures and societies [10], and uneven knowledge and attitudes towards behavioural diseases among physicians. In commonly used DSM classifications the difference between TD and CTD relied on coincidence of motor and vocal tics in TD, which makes population-based studies even more difficult. Additionally, tic disorders have complicated interrelations with other behavioural syndromes including ADHD and OCD, which often influence the outcome of tic disorders [6].

The objective of the present study was to screen mainstream school population with enough sensitivity to identify tic-positive children and estimate minimal prevalence of ICD-10 tic disorders. The study design, validated in previous studies, was selected to maximise screening sensitivity and allow assessing disease prevalence in general population not referred to physicians.

Methods

Study population

The sample consisted of schoolchildren attending Warsaw primary schools. A total of 24 schools were chosen randomly and all 6th and 7th grade students from these schools were included in the study. Schools teaching mentally retarded children were excluded from the randomisation process. Other forms of special education, i.e. special integration classes for disabled children or children with learning problems were not excluded. The age group was chosen to meet the age compatibility of used questionnaires and consideration of tics as disorders occurring in young children. All subjects were screened for lifetime occurrence of motor and/or vocal tics using a two-stage process. Warsaw Medical University Ethical Committee reviewed and approved the study project.

Study procedure

The procedure used to estimate the prevalence of tic disorders in this study is presented on Fig. 1. The first stage of the assessment utilised a screening questionnaire. After receiving the approval of directors of selected schools, meetings with parents were arranged. After short 5-min introduction by interviewers on the clinical picture of tic disorders parents were asked to give their consent for the inclusion of their children in the study and to complete a three-item questionnaire developed by Apter et al. [2] (Table 1). Teachers of enrolled children completed the same screening questionnaire for each child.

In the second stage of ascertainment, all children indicated by parents or teachers as tic-positive were examined. Inclusion criteria consisted of informed consent by parent or legal guardian and positive parent or teacher's answer to any of the screening questions. Trained investigators personally interviewed all indicated children. The interview took 10–45 min and involved the observation of subjects and completion of semi-structured interview. Information on past symptoms was obtained from the interviewed children. Altogether 10 trained physicians and medical students participated in the study. Their training included videos from Tourette Syndrome Association and 10-h training sessions in tics and associated disorders clinical picture, diagnostic criteria, performed by certified child psychiatrists. Questionnaires used in the second-stage interviews were designed at the Department of Child and Adolescent Psychiatry of Warsaw Medical University. The set of questionnaires included an 18-questions socio-demographic interview, a semi-structured questionnaire for differentiation of tic disorders based on ICD-10 criteria, and the Polish version of Yale Global Tic Severity Scale (YGTSS). The YGTSS scale was developed by Leckman [14]. It serves for clinical assessment of patients with diagnosed tic disorder and is measuring the severity of motor and vocal disturbances in several dimensions (number, frequency, severity, complexity, influence). The global score ranges from 0 to 100. The

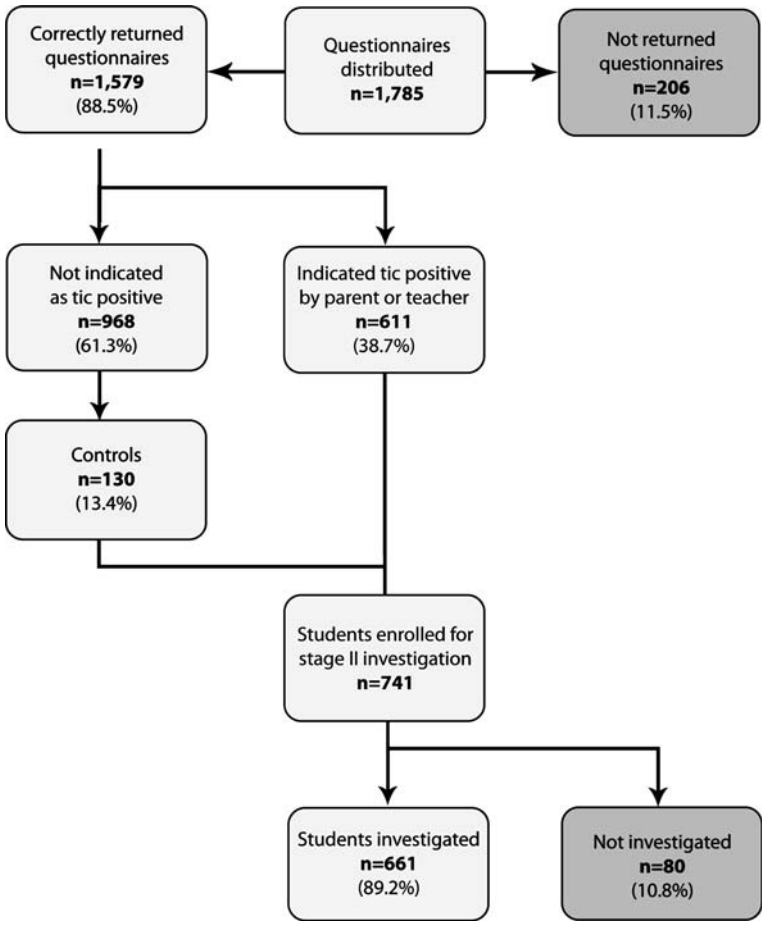

Fig. 1 Schematic presentation of the study procedure

psychometric properties of the Polish version of the YGTSS scale are published elsewhere [18].

To determine the sensitivity of the screening procedure, 130 randomly selected subjects not pointed out by screening were assessed. The inclusion criteria were informed consent of the parent or legal guardian, and presence in the school during the interviews. All subjects were interviewed using the Stage II study protocol.

Statistical analysis

The description of data was expressed using mean values (±SD). Comparison of groups was performed using chi-square test for categorical variables and one-way ANOVA test for continuous variables. In case of inequality of variance or lack of normal distribution, Kruskall–Wallis test was used. When comparing more then two groups, Mann–Whitney *U*-test was used. The significance level of 5% was used. When multiple tests were performed, the Dunn–Sidak alpha adjustment was applied. The lifetime prevalence and point prevalence estimates of tic disorders were computed using the formula for stratified sampling to account for separate sampling of children indicated by parents and those randomly chosen for the control group. The denominators for the estimates were the studied populations. The distribution of demographic and social factors was compared between schoolchildren diagnosed with tics and schoolchildren free of tics. Tic disorders were compared by tic symptom distribution and severity of symptoms. The sensitivity, specificity and positive predictive value of the screening procedure were computed. To assess the correlation of positive answers to screening questions with the diagnosis of tics, τ-Kendall coefficients were used. Stepwise logistic regression analysis was performed to assess the independent effect of parental and teacher answers on the final diagnosis of tic disorders. The analysis was performed using SPSS ver. 14.0 software.

Results

A total of 1,579 children were screened. Out of 1,785 questionnaires distributed during school meetings,

Table 1 Screening questionnaire items list [2]

Many children have habits like blinking their eyes, moving their nose, clearing their throat, and other involuntary tics.
1. Has the child ever had (or currently has) facial tics, jerks of other parts of his or her body, or any other unusual movements or habits?
2. Did you ever have noticed (or currently find) the child involuntary making noises other than normal talking, like grunts, throat clearing, or saying words?
3. Did you ever have noticed (or currently find) the child involuntary touching objects, other people, or parts of his or her body repeatedly?

Table 2 Selected demographic factors in the studied group, compared to official statistics for Poland in 1999.

Population	Parents' Education		Parents' Employment		Families			
	Secondary (%)	Higher (%)	Employed (%)	Unemployed (%)	Complete (%)	Incomplete (%)	≤2 children (%)	>2 children (%)
Studied group								
Screened positive (n = 531)	46.5	29.6	81.6	2.0	84.4	15.6	83.0	17.0
Controls (n = 130)	47.0	26.2	83.8	2.0	85.4	14.6	86.1	13.9
Official statistics								
Warsaw	48.1	16.9	81.4	2.3	67.9	32.1	93.5	6.5
Urban population	58.8	9.8	47.4	15.9	76.0	24.0	78.0	22.0
Rural population	43.5	1.9	49.0	14.5	81.3	18.7	71.2	28.8
Poland	53.1	6.8	48.0	15.3	78.0	22.0	80.2	19.8

1,579 (88.5%) forms from both parents and teachers were obtained. The mean age in the screened group was 13 years and 4 months (SD 8 months), 848 children (53.7%) were boys and 731 (46.3%) were girls.

Out of 1,579 screened children, 611 (38.7%) were indicated as tic-positive. Both parents and teachers indicated 124 (20.3%) children, parents only indicated 234 (38.3%) children and teachers only indicated 253 (41.4%) children. Only in 31 cases all answers of teachers and parents were matching, and no correlation of parent's and teacher's answers was revealed (lowest $\tau = 0.007$ for the correlation between the parental question No. 3 and teacher question No. 2, highest $\tau = 0.153$ for the correlation between parental question No. 2 and teacher question No. 1). A logistic regression model confirmed a lack of association between teacher and parental answers to screening questions. The highest association ($r^2 = 0.02$) was established between the answer to the teacher question No. 3 (complex motor or vocal tics) and parental question No. 2 (vocal tics).

A total of 611 eligible children were enrolled in the second stage of the study, of which 531 (86.9%) were interviewed. There were 353 (66.5%) boys and 178 (33.5%) girls interviewed. The mean age in this group was 13 years and 4 months (SD 9 months). Of the 80 remaining children, 30 (37.5%) were not present in the schools during the study period, 20 children (25.0%) refused to participate, 19 (23.8%) moved to other schools, and 11 (13.8%) were not interviewed due to the lack of parent informed consent.

About 130 children were randomly recruited into the reference group, including 54 boys (41.5%) and 76 girls (58.5%). The mean age in the control group was 13 years and 4 months (SD 8 months). The control group was different from the screened schoolchildren population in terms of gender distribution ($\chi^2 = 8.434$, $P = 0.002$) and did not differ in terms of age distribution. The representativeness of the studied groups was assessed by comparing to officially published statistical data on socio-demographic factors (Table 2).

A total of 104 children were found to meet criteria for tic disorders, 96 of them were indicated by screening and 8 were diagnosed in the control group. There were 77 boys (74%) and 27 girls (26%). The mean age in the group was 13 years and 3 months (SD 9 months). Children with tics differed from the population free of tics only in terms of gender distribution ($\chi^2 = 8.11$, $P = 0.004$). Overall lifetime prevalence of tics was 11.6% (95% CI 7.9–15.4%) for boys, and 7.7% (95% CI: 3.5–11.9%) for girls. Thirty-seven children (35.6%) had tics in the past, and 67 children (64.4%) had tics at the moment of interview. The overall lifetime prevalence of tics established in this study was 9.9% (95% CI 7.1–12,7%), and the point prevalence was 6.7% (95% CI 4.3–9.1%). Out of 104 children with tics, 87 (83.7%) had motor tics, and 26 (25.0%) had vocal tics. The most common symptoms were simple tics – eye blinking (38 children; 36.5%), and simple movements involving facial muscles (32 children; 30.8%). The most common vocal tics were simple sounds (throat clearing, coughing; 25 children; 24.0%). Nine children with tics have been receiving medications at the moment of the study: anti-allergics (7 children), carbamazepine (1) and haloperidol (1).

Table 3 Classification of children diagnosed with tics by tic disorder, according to ICD-10 criteria

Diagnosis	*N*	%[a]	M/F ratio	Prevalence[b]	95% CI
Transient tic disorder (F95.0)	29	1.8	19/10	265.3	120.6–410.0
Chronic tic disorder (F95.1)	39	2.5	30/9	369.5	195.0–544.0
Tourette syndrome (F95.2)	9	0.6	6/3	57.0	20.3–93.7
Non-specific tic disorder (F95.8)	27	1.7	22/5	293.5	123.8–463.1
Overall prevalence	104	6.6	77/27	985.2	707.5–1262.9

[a]Percent of studied group (N = 1,579)
[b]Lifetime prevalence per 10,000 children aged 13–14 years

Table 4 Clinical characteristic of children with tic disorders

Characteristic		Non-specific tic disorder (n = 27)	Transient tic disorder (n = 29)	Chronic tic disorder (n = 39)	Tourette disorder (n = 9)
Gender—number (%)	Males	22 (81.5%)	19 (65.5%)	30 (76.9%)	6 (66.7%)
	Females	5 (18.5%)	10 (34.5%)	9 (23.1%)	3 (33.3%)
Tic symptoms—number (%)	Eye blinking	6 (22.2%)	7 (24.1%)	19 (48.7%)	6 (66.7%)
	Other simple tics of face	4 (14.8%)	9 (31.0%)	15 (38.5%)	4 (44.4%)
	Head movements	5 (18.5%)	3 (10.3%)	4 (5.5%)	3 (33.3%)
	Other simple motor tics	9 (33.3%)	9 (31.0%)	13 (33.3%)	9 (100.0%)
	Complex motor tics	3 (11.1%)	0 (0.0%)	10 (25.6%)	3 (33.3%)
	Simple vocal tics	8 (29.6%)	5 (17.2%)	4 (10.3%)	8 (88.9%)
	Complex vocal tics	0 (0.0%)	0 (0.0%)	2 (5.1%)	1 (11.1%)
YGTSS results—mean (95% CI)	Number	1.53 (1.25–1.82)	–	1.64 (1.33–1.97)	3.00 (2.37–3.63)
	Frequency	1.73 (1.12–2.34)	–	2.57 (2.16–2.98)	6.62 (5.22–8.03)
	Intensity	2.00 (1.45–2.55)	–	2.62 (2.31–2.93)	5.37 (3.70–6.78)
	Complexity	0.33 (0–0.73)	–	0.57 (0.21–0.92)	0.87 (0.05–1.70)
	Interference	1.64 (0.98–2.31)	–	1.95 (1.59–2.30)	4.00 (2.82–5.18)
	Overall impairment	5.33 (1.22–9.45)	–	9.73 (6.26–13.20)	18.75 (11.77–25.73)
	Global scale	11.64 (6.38–16.91)	–	19.08 (14.63–23.54)	38.62 (28.85–48.40)

The established lifetime prevalence of ICD-10 tic disorders is summarised in Table 2, and descriptive data for children diagnosed with tic disorders are reported in Table 3. TTD was diagnosed in 29 schoolchildren with symptoms reported in the past, yielding a lifetime prevalence of 2.6% (95% CI 1.2–4.1%). Sixteen children (55.2%) had tics of eye and face muscles. Simple vocal tics were reported by 5 children (17.2%). CTD was diagnosed in 39 children (38 at present and 1 in the past), giving a lifetime prevalence of 3.7% and a point prevalence of 3.6% (95% CI 1.9–5.4%). In 32 children (82.1%), the movements involved muscles of eye and face. More than one simultaneous simple motor tics was reported by 14 children (35.9%). Simple vocal tics (throat clearing) were reported by 4 children with CTD (10.3%). TD was diagnosed in 9 schoolchildren. In all cases the symptoms were present at the moment of study, yielding a lifetime and point prevalence of 0.6% (95% CI 0.2–0.9%). All cases had simultaneous motor and vocal tics. Complex motor or vocal tics were reported by 3 children. Only two children with TD were referred to a physician because of neurological syndromes, and one have been treated for tics. Non-specific tic disorder was diagnosed in 27 children, mostly having current symptoms and not yet meeting criteria for specific tic disorders. Lifetime prevalence of non-specific tics was 2.9% (95% CI 1.2–4.2%) and their point prevalence was 2.5% (95% CI 0.8–4.2%). The most prevalent tic symptoms in this subgroup were movements involving muscles of eye and face (9 cases, 33.3%) and simple vocal tics (8 children, 29.6%) (Table 4).

The comparison of cases by tic disorder diagnosis revealed no significant differences in gender distribution (χ^2 = 2.30; P = 0.51). In all tic disorders, male gender was predominant. Since the analysis of YGTSS scores involved 7 comparisons, the adjusted alpha level of 0.0073 was used. Significant differences of scores between particular tic disorders were found for all items, except the tic number and complexity scores (Fig. 2). The significant differences were based mainly on the difference between scores for TD and scores for other tic disorders.

The validity study of the screening questionnaire revealed its high sensitivity (92%), in contrast to very low specificity (22%) and positive predictive value (18%). When restricting this comparison to parents answers to the tic screener, the sensitivity of this tool would be 51.9%, specificity 53.5% and positive pre-

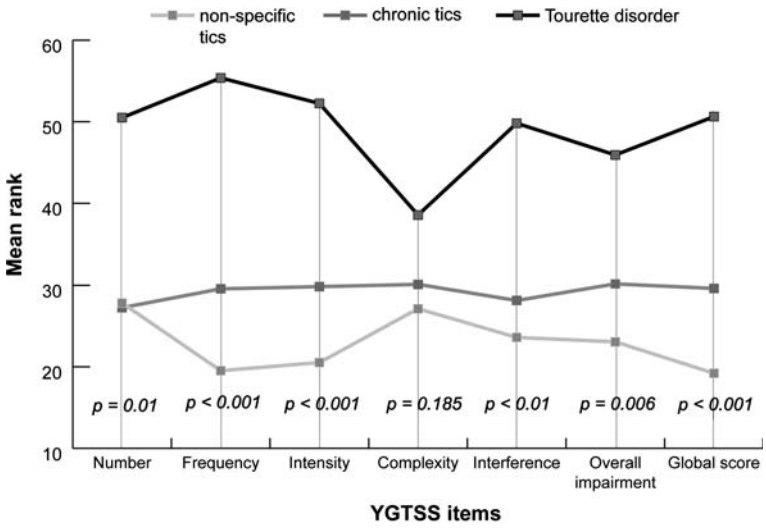

Fig. 2 Comparison of YGTSS scale results by tic disorder, Kruskall–Wallis test, $n = 60$

dictive value 17.3%. In contrast, the properties of the tic screener for teachers answers was: sensitivity 74.0%, specificity 54.8% and positive predictive value 23.4%. The analysis of predictive usefulness of screening questions revealed only a weak correlation between a positive answer to the teacher's question No. 1 (motor tics) and the final diagnosis of tics ($r = 0.339$; $P < 0.001$), and a lack of correlation for answers to other questions. In the stepwise logistic regression analysis of independent effects of each screening question on the final diagnosis of tics, only the positive answers to teacher's and parent's questions on motor tics (No. 1 in both questionnaires) were left in the model.

Discussion

The study explored the minimum prevalence of ICD-10 tic disorders in a population of schoolchildren aged 13–14 years, living in Warsaw. Compared to official socio-demographic data for Poland, the studied population was similar to the population of Warsaw in terms of percent of parents employed and having higher education, but not in terms of family structure, and differed importantly from the urban, rural and general populations. Thus, the results of this study can be considered representative only for the population of 13–14 year old schoolchildren living in a large Polish agglomeration. To avoid selection bias, the schools were chosen randomly and should not differ systematically from other mainstream schools. Additionally, selection bias could arise from attending of the school meetings selectively by parents, who give more attention to their children's education, or who have problems with their children. Non-participants were not assessed because of logistical constraints.

The analysis of the screening process showed that teachers indicated tic-positive children more often. The possible reasons for that could be the presence of tics at school, and more accurate perception of involuntary movements by teachers, compared to parents. This is further supported by the higher sensitivity and positive predictive value of teacher's answers in the tic screener. Because the reference group differed significantly from the total study population in terms of gender distribution, the number of subjects with tics found in this group could be even higher, leading to lower positive predictive value. The school system at the time of the study comprised 8-year primary school and 4-year secondary school. Teachers interviewed in the study protocol knew their students for 3–4 years and were coping with them on a daily basis as their formal tutors. The original version of the questionnaire appeared to be highly sensitive (91.1%), and to have very low positive predictive value (16.0%) [2]. A British study analysed the validity of its screening procedure [11] and its results showed, similarly, a high sensitivity (76.9%) and low predictive positive value (33.3%). The false positive results were related in this setting to the high proportion of subjects which refused to participate in the II stage of the study. The choice of a simple and sensitive screening method, even allowing an important increase in workload related to the need of investigation of a large proportion of false-positive subjects, seems justified in a country where knowledge of tic disorders in general population is low. Tics are misinterpreted by teachers and parents as stereotypic behaviours or hyperactivity. A relatively high proportion of false negative subjects in the reference group is the result of poor knowledge of involuntary movements in the polish population and indicates a possible underestimation of prevalence estimates. In

summary, the screening procedure, with all its limitations, allowed screening a representative sample of Warsaw schoolchildren and sensitive estimation of the minimal prevalence of tic disorders in the studied population.

Potential misclassification could occur, related to the application of diagnostic protocol and of the YGTSS scale to subjects by particular interviewers. To minimise differential misclassification, a uniform training and semi-structured interview methodology were applied. Also, classification of tic disorders based on the ICD-10 criteria could yield misdiagnosing of tic disorders. The ICD classification reflects the current concepts of tic disorders as a behavioural continuum, and two most severe syndromes—CTD and TD—differ only in terms of duration of tic symptoms. The use of ICD criteria should not constitute a problem in terms of comparability with previous epidemiological studies, especially that most of them utilised DSM-III-R criteria, very similar to ICD-10 classification. Several authors have argued that impairment and emotional distress (included in DSM-IV criteria) are not necessary for definite diagnosis of tic disorders [4, 19]. Our decision to use ICD criteria was based on its universal use in medical documentation and diagnostic protocols in Poland, as well as other European countries.

Despite these limitations, the present study has important strengths. It used sensitive two-stage screening procedure and precise measurement tools. It was the first study of tic disorders epidemiology in Poland. Also, it was one of the first comprehensive attempts to establish a population-based prevalence of tic syndromes. The prevalence of TTD was in the lower limit of the previously established figures [9, 12]. However, TTD is most common among preschool children and is a mild a self-resolving disorder [16]. The diagnosis of TTD is possible only for past symptoms. Therefore all children meeting ICD-10 criteria for TTD and having current symptoms were classified as non-specific tic disorder. This makes precise estimation of TTD prevalence possible only during longitudinal studies, preferably using multiple informants. The prevalence of CTD was higher than established in previous studies. The prevalence of TD established in the present study was comparable to results of published population-based studies, involving similar age groups [5, 7–9].

The presently studied sample of the general population had less severe clinical picture of tic disorders than suggested by published data on patients referred to hospitals. They had mild symptoms, and were not previously diagnosed with a tic disorder. As stated in previous publications, children and their parents have often problems with interpretation of their symptoms and referral to appropriate doctors [8, 15]. The diagnosis of tics could be further complicated by the co-occurrence of other behavioural disorders (ADHD, OCD), which influence the clinical presentation of tics.

Acknowledgements The authors wish to thank the study participants, their caregivers and teachers for their participation in the study. This study was supported by a research grant from the Warsaw Medical University # AEN-2-262b-98.

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