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Tremor and Other Hyperkinetic Movements

Review

Review of Prevalence Studies of Tic Disorders: Methodological Caveats

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Abstract

Introduction: Tic disorders are neurodevelopmental disorders of childhood associated with psychiatric comorbidity and academic problems. Estimating the prevalence and understanding the epidemiology of tic disorders is more complex than was once thought. Until fairly recently, tic disorders were thought to be rare, but today tics are believed to be the most common movement disorder, with 0.2–46.3% of schoolchildren experiencing tics during their lifetime. Tentative explanations for differing prevalence estimates include the multidimensional nature of tics with a varied and heterogeneous presentation, and the use of different epidemiological methods and study designs.

Methods: Literature review and analysis of methodological issues pertinent to epidemiological studies of tic disorders.

Results: Epidemiological studies of tic disorders were reviewed, and the main elements of epidemiological studies, including sample selection, case ascertainment strategy, definition of tic disorders, and the degree of coverage of the eligible population (i.e., the response rate) were examined.

Discussion: In order to improve the quality of epidemiological studies of tic disorders, a number of recommendations were made, including but not limited to a review of the diagnostic criteria for tic disorders, and inclusion of new tic disorder categories for those with tics of secondary etiology.

Keywords: Epidemiology, prevalence studies, tics, movement disorders, screening in epidemiology

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Introduction

Tic disorders are hereditary neurodevelopmental disorders of childhood, characterized by being sudden, repetitive, stereotyped motor movements or sounds. The longitudinal outcome is one of gradual improvement in most patients.[1] There are several widely used diagnostic classifications for tic disorders, including the Diagnostic and Statistical Manual of Mental Disorders (DSM),[2] the International Classification of Disease and Related Health Problems 10th revision,[3] and the Classification of Tic Disorders developed by the Tourette Syndrome Classification Study Group.[4] Although clear differences exist between these classification schemes, they are broadly congruent, with each containing well-specified categories; 1) Tourette syndrome (TS), 2) chronic motor or vocal tic disorder (CMT, CVT), 3) transient tic disorder (TTD), and 4) non-specified tic disorders (NSTDs). TS encompasses the combination of chronic (more than 1 year) motor and vocal tics; CMT or CVT, the presence of chronic motor or vocal tics; and TTD, the presence of motor or vocal tics with a duration of at least 4 weeks but less than 1 year. NSTDs include the presence of tics with an adult onset (>18 years) or duration less than 4 weeks. Idiopathic or primary tic disorders typically begin in childhood, mostly at the age of 5–6 years,[5] and boys outnumber girls by a ratio of 4:1 to 6:1.[5,6]

Comorbid conditions are frequently associated with tic disorders. The most frequent conditions are attention deficit disorders (ADHDs) in about 20% of the children, followed by obsessive compulsive disorder (OCD), self-injurious behavior, anxiety, depression, personality disorders, oppositional defiant disorder, and conduct disorders.[7] ADHDs generally occur before tic onset and tend to decrease in 20% of children during adolescence, yet later than tics.[8] OCD symptoms associated with tic disorders usually begin at a somewhat later age than tics (i.e., at around 10 years of age), and tend to remit in only about 40% of patients.[9] Persistence of ADHD and OCD is associated with poorer psychosocial functioning.[10] Family genetic studies strongly indicate a genetic component to tic disorders, with shared genetic

Table 1. Epidemiological Studies on the Prevalence of Tic Disorders in Children

First Author, Year	Country	Age (Years)	Screening	Sample Size	Point Prevalence (%)	Period Prevalence (%)
Boncour, 1910[59]	France	2–13	Teacher observation	1759	23.7	–
MacFarlane, 1954[26]	USA	1–14	Parent interview	116	–	25
Lapouse, 1964[27]	USA	6–12	Parent interview	482	12	–
Pringle, 1967[28]	UK	7	Parent questionnaire, medical history and examination	7949	7.7	–
				7958		
				7965		
Rutter, 1970[60]	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[61]	UK	5–15	Parent questionnaire	6290	–	20
Achenbach, 1978–9[30,31]	USA	6–16	Parent questionnaire	2200		
			Population sample	1100	7.7	
			Referral sample	1100	28.8	
Kurlan, 2001[44]	USA	8.5–17.5	Parent questionnaire			
			Technician exam			
			Special education	341	29	
			Regular education	1255	17.9	
Khalifa, 2003[50]	Sweden	7–15	Parent questionnaire			
			Medical exam			
			Regular education	4438	6.3	
			Special education	41	46.3	
Lanzi, 2004[22]	Italy	6–11	Teacher observation	2347		2.9
Ooki, 2005[33]	Japan	3–15	Questionnaire	1986	6.8 (males) 4.1 (females)	
Linazasoro, 2006[62]	Spain	4–16	Parent and teacher questionnaire Observation	867	6.5	
Stefanoff, 2007[24]	Poland	12–15	Parent and teacher questionnaire Exam	1579	6.7	9.9
Schlander, 2009[36]	Germany	0–50+	Database code	2.238.460	0.2	

Table 1. Continued

First Author, Year	Country	Age (Years)	Screening	Sample Size	Point Prevalence (%)	Period Prevalence (%)
Cubo, 2011[48]	Spain	6–16	Parent, teacher questionnaire			
			Observation			
			Neurologist interview			
			Regular education	741	16.8	
			Special education	54	20.3	

Modified from Shapiro et al,[22] and Lanzi et al.[25]

influences between tics and OCD, but a less clear set of relationships with ADHD.[11] A large Genome Wide Association Study within the Tourette Syndrome Association genetic consortium is underway.[12]

The epidemiology of tic disorders is more complex than was once thought. Until fairly recently, tic disorders were thought to be rare and, according to some, a psychogenically mediated disorder,[13] but today tics are believed to be the most common movement disorder diagnosed in children, with 0.2–46.3% of schoolchildren experiencing tics during their lifetime (Table 1).[6,14–32] Tentative explanations for differing prevalence estimates include the multidimensional nature of tics with a varied and heterogeneous presentation, and the use of different epidemiological methods.[7,13] The diagnosis of tic disorders is challenging, given the fact that tics classically wax and wane, can be suppressed, and may not be present at the time of interview. Furthermore, if tics are diagnosed by history, they can be difficult to distinguish from other repetitive behaviors (compulsions, habits), and the proxy (teachers or parents) cannot provide detailed information about tic suppression or urgency. Owing to the important association of tic disorders with psychiatric conditions and academic difficulties,[34] knowledge of the prevalence of different tic disorders is critical, and reasonable estimates are necessary for planning and providing early support in education and health care.

In this review, a review and discussion of epidemiological studies will be presented. The main elements of epidemiological studies, including sample selection, case ascertainment strategy, definition of tic disorders, and the degree of coverage of the eligible population (i.e., response rate) will be examined and discussed. Given the nature of the literature, the primary focus is on TS.

Methods

For the review, a literature search was performed using Medline, Psychlit, and Index Medicus, up to August 1, 2011.

Results

Sample selection

Earlier estimates of the epidemiology of tic disorders were based on single or multicenter clinic samples. However, with the use of such referral samples, the detection and diagnosis of tic disorders, especially among mild cases and/or children with limited access to specialty health-care services, is not optimal, and prevalence estimates may be low.[6] On the other hand, administrative databases (state-population databases or large national samples) provide wide coverage of the population, and provide an opportunity to include samples with different sociodemographic characteristics. In this regard, the use of administrative databases and/or telephone-based surveys, may provide us with estimates of the public health impact of tic disorders.[6,35,36]

The use of community-based samples has resulted in higher estimates of the prevalence of tic disorders. Epidemiological studies of tic disorders have been conducted in different populations, sampling different races and geographical areas, cohorts from movement disorder clinics, community-based samples, and school-based samples (including mainstream schools and special education centers [SECs]). Overall, TS, the most studied tic disorder, has been reported all over the world, and from all sociocultural, socioeconomic, religious, linguistic, and ethnic groups.[37] However, while TS has been researched extensively in Western populations, the clinical characteristics and associated features are less well known in non-Western cultures.[13] Lower prevalences of TS have been reported in China and sub-Saharan and other regions of Africa.[7] According to Robertson,[13] the following reasons may account for the apparent rarity in these regions: other medical priorities, lack of medical training, ethnic and epigenetic differences, and genetic and allelic differences in different races.

The age of the population may also influence estimates of the prevalence of tic disorders. Inclusion of persons in younger age groups in population-based studies (e.g., as young as 4 years of age) may result in lower estimates of prevalence, since the symptoms may not yet have manifested at such young ages. For example, in one study conducted in primary schools, TS was found in 0.15–1.1%.[37,38] In contrast, the prevalence among older people, especially in adult psychiatric units, may also be relatively low (e.g., around 9.5%) when compared to the prevalence in younger people, as studies have shown that tics diminish with age.[39]

Prevalence estimates of tic disorders in populations with mental disabilities also seem to be higher, especially in children with autistic spectrum disorders, learning difficulties and behavioral problems; these estimates range from 6% to 23.4% for TS, and up to 46.3% for other tic disorders.[22,31,40–44] Interestingly, in our study conducted in SECs, tic disorders were very frequent in children diagnosed with Down syndrome, and there was no gender preponderance.[32]

Screening and case ascertainment

There is no doubt that community-based surveys are costly and therefore difficult to perform. Yet they offer an ideal strategy for obtaining an adequate and representative sample size, especially for the study of low prevalent conditions or populations at high risk of dropout (i.e., populations of low income, of those with low education background, etc.). The logistic difficulties increase when epidemiological studies are carried out in populations diagnosed with diseases such as tic disorders that have fluctuating symptoms and signs. Validated screening tools and methodologies are needed to define each case. Ideally, screening should be a multistage procedure, which in a prior pilot study specific for that population and setting, has been validated against a gold standard (i.e., a diagnosis established by medical personnel).

Few epidemiological studies have previously reported the sensitivity of their screening procedures for tic disorders.[21,22,44] Prior studies have used different methods to identify individuals with tic disorders, including direct observations at school, parent, or teacher interviews or questionnaires, student questionnaires, and clinical examination.[19,37,44–47] The use of information from trained teachers is reported to have a sensitivity of up to 80% in mainstream schools.[22] A study designed to validate screening procedures in a random selection of parents and teachers for 130 individuals in mainstream schools, demonstrated that this approach had a high sensitivity (92%) but low positive predictive value (18%).[45] We conducted a pilot study designed to validate our screening procedures for tic disorders in different settings.[48] We developed a two-stage screening tool. First, a Proxy-Report Questionnaire completed by parents and teachers, and, second, classroom observation of up to 3 hours by three trained lay observers. The training of the three observers was also validated by the combined rating of 11 individuals (six with tics and five without tics), which resulted in a high sensitivity (100%) and good specificity (60%) to identify tics when compared with the neurologist, good inter-observer agreement (kappa coefficient [k]=0.62), and good neurologist–observer agreement (k=0.62). We found that 2 hours' observation of the classroom offered the best strategy. However, this length of direct observation can be considered suboptimal as it seems to have lower single screening accuracy when compared with the information provided by the teachers and parents. The decision to use a screening test on a given group of patients is made on the basis of a cost–benefit analysis. Hence, if one is only able to use a single screening source, parents would be the best source in mainstream schools, given that their modest sensitivity (58%) is compensated by a high specificity (92%) and the highest likelihood ratio (7.38%) for a positive result prior to testing (Table 2). On the other hand, teachers are the best single screening source of information in SECs, with moderate sensitivity

Table 2. Screening Instruments vs. Gold Standard in Mainstream Schools

N=63	Sensitivity % 95% CI	Specificity % 95% CI	PPV% 95% CI	NPV% 95% CI	+LR ratio 95% CI
Teachers	40 (9.64–70.36)	74 (59.81–88.77)	30 (5.68–55.86)	81 (67.73–94.77)	1.64 (0.55–4.87)
Parents	58 (30.44–86.92)	92 (86.35–99.61)	63 (35.21–92.07)	91 (68.08–87.80)	**7.38 (2.85–19.87)**
Observers					
1 hour	33 (6.66–60.00)	80 (68.30–90.88)	28 (4.91–52.23)	83 (72.24–93.72)	
2 hours	58 (30.44–86.22)	80 (68.30–90.88)	41 (17.78–64.58)	89 (79.26–98.02)	2.9 (1.37–5.93)
3 hours	58 (30.44–86.22)	78 (65.87–89.23)	39 (16.37–61.45)	88 (78.79–97.95)	
Multiple sources					
Teachers+ parents +2-hour ob.	**92 (76.03–100)**	65 (51.59–77.83)	38 (20.27–55.59)	**97 (91.38–100)**	2.59 (1.72–3.90)
Teachers+parents	83 (62.24–100)	**74 (62.55–86.47)**	43 (23.22–63.74)	95 (88.25–100)	3.26 (1.91–5–57)
Teachers+2-hour ob.	67 (40.00–93.34)	70 (57.30–82.70)	35 (15.32–54.24)	90 (80.22–99.26)	2.22 (1.24–3.97)
Parents + 2-hour ob.	**92 (76.03–100)**	72 (60.30–84.80)	**44 (24.54–63.46)**	**97 (92.28–100)**	3.33 (2.07–5.38)

Data obtained from Cubo et al, study.[48]

Abbreviations: +LR ratio, likelihood ratio for a positive result; NPV, negative predictive value; ob., observation; PPV, positive predictive value; CI, confidence interval.

(73%) and specificity (55%), but with the highest likelihood ratio (1.63%) for a positive result prior to testing (Table 3). If multiple sources can be used, the most efficient screening source in mainstream schools is the combination of parents and a 2-hour classroom observation. By contrast, the information from teachers plus 2 hours of observation provides the best combination for screening in SECs. Overall, this screening procedure seems to be more accurate in mainstream schools than in SECs, most likely due to the presence of repetitive motor behaviors, such as stereotypes, in children with mental disability.

A selection bias is commonly observed in studies for which the cases are ascertained based on a positive screen.[16] Using such methodology, false-negative cases may be underestimated and, likewise, the estimated prevalence of tic disorders may be low. Similarly, there is a failure to ascertain cases in some studies, or assessments that are only performed by trained technicians.[44] Preferably, case ascertainment should be performed by medical personnel.[21,48,49]

In order to decrease the burden of conventional face-to-face interviews and to avoid dropouts, telephone interviews can be an efficient alternative. In our study, we analyzed the intra-rater (neurologist) reliability of telephone interviews compared to in-person interviews to achieve a correct diagnosis of tic disorders.[48] A high k of 0.83 was found, suggesting that telephone interviews may make it easier to perform clinical interviews, and to increase participation. However, in our study, the use of telephone interviews decreased the level of diagnostic certainty by increasing the frequency of possible tics up to 19.4%.[32] Unfortunately, similar reliability of data from telephone interviews has not been published in other studies, so comparisons are not possible.[16,50]

Definition of tic disorders

Tics can be motor, vocal, sensory or cognitive, simple or complex, isolated or multiple, and brief or more dystonic and tonic. Classically, tics are described as sudden, brief, and rapid, repetitive sounds or movements that can be controlled voluntarily during short periods of time, they may change and affect other body parts periodically, and improve or worsen from time to time. Since tics are a fluctuating condition, the neurologist may not observe the tics during the exam, but can establish the diagnosis based on patient/relative reports or videotapes.[51,52] Likewise, many clinicians erroneously believe that coprolalia must be present for the diagnosis of TS to be made, and some habits, such as foot tapping or finger drumming that occur when subjects are bored or anxious, are incorrectly diagnosed as tics. Although it seems to be obvious, the definition of tic phenomenology should be clear for standardization of epidemiological research efforts. In Appendices 1 and 2, we provide an example of the screening and interview schedule for tic disorders followed in our study.[48]

The DSM criteria for tic disorders have also changed over time.[2] Specifically, the upper age limit, which is somewhat arbitrary, has changed, and the inclusion of a tic period of at least three consecutive months for TS, CVT, or CMT has been added, although there seems to be no evidence base for this.[13] By contrast, in the World Health Organization (WHO) criteria,[3] there has never been an age of onset requirement, resulting in the inclusion of adult-onset tic disorders. Whereas there is a clear definition of tic disorders based on the DSM, in the International Classification of Disease and Related Health Problems 10th revision,[2,3] and the Classification of Tic Disorders developed by the Tourette Syndrome Classification Study Group,[4] tics

Table 3. Screening Instruments vs. Gold Standard in Special Education Settings

N=57	Sensitivity % 95% CI	Specificity % 95% CI	PPV% 95% CI	NPV% 95% CI	+LR ratio 95% CI
Teachers	73 (46.41–99.05)	55 (41.04–70.08)	28 (11.84–45.30)	76 (61.14–90.38)	**1.63(1.00–2.66)**
Parents	36 (7.93–64.79)	42 (27.79–56.65)	27 (9.87–43.97)	87 (74.51–98.83)	0.62 (0.27–1.42)
Observers					
1 hour	27(0.95–53.59)	55 (41.04–70.08)	13(–0.72–26.80)	76 (61.14–90.38)	
2 hours	36 (7.93–64.79)	44 (29.92–58.96)	14 (1.24–26.34)	74 (57.54–90.60)	0.65(0.28–1.49)
Multiple sources					
Teachers+ parents +2-hour ob.	**73 (39.02–93.97)**	**58 (36.77–78.60)**	**42 (20.25.66.50)**	83 (58.58–96.42)	1.02 (0.74–1.40)
Teachers+ parents	**73 (39.02–93.97)**	29 (16.07–43.03)	22 (9.56–35.44)	87 (69.47–100)	1.15 (0.82–1.62)
Teachers+ 2-hour ob.	**73 (39.02–93.97)**	35 (21.56–49.54)	23 (10.16–37.20)	**89 (74.37–100)**	**1.26 (0.89–1.80)**
Parents + 2-hour ob.	45 (30.00–60.36)	22 (10.07–34.36)	12 (2.25–22.74)	62 (38.77–86.22)	0.58 (0.30–1.13)

Data obtained from Cubo et al, study.[48]

Abbreviations: +LR ratio, likelihood ratio for a positive result; NPV, negative predictive value; ob., observation; PPV, positive predictive value; CI, confidence interval.

represent a symptom, and their definition is not included in these diagnostic revisions. Adult-onset tics have been reported, but in contrast to child-onset tics, they are likely to be secondary to trauma, drugs, stroke, or infection.[53]

In addition, there also several ambiguities about the definition of tic disorders, with regards to the inclusion or not of impairment criteria, and comorbid conditions. The current version of the DSM-IV TR,[2] and in the future DSM-V,[54] the distress item, which had been obligatory in order to establish a tic diagnosis in previous classification schemes, has been omitted. Clearly, more restrictive criteria yield much lower prevalence estimates, and, thus, misleading results.[32,44,55,56] The use of impairment criteria as a means of distinguishing cases from non-cases of tic disorders is controversial, given the fact that in most cases, the source of impairment (tics or comorbidity conditions) cannot be clearly established.[57]

Coverage of the eligible population

Overall, there is a lack of information about attrition (i.e., the loss of participants) in neuroepidemiological studies of tic disorders. Attrition can result in selection bias and reduce the internal or external validity of the research findings. Likewise, the response rate, defined as the number of respondents who complete a questionnaire compared to the number assigned, may also impact on the quality of the data. In our study,[32] we observed different response rates for mainstream schools (70%) vs. SECs (55%). Caucasians and younger students without academic problems were more likely to participate in a survey of tic disorders. In other studies, conducted in youth cohorts, the main demographic variables associated with participation were gender (girls were more likely to participate) and a high education background of parents.[57] Other variables that may contribute to dropout include the frequency of contacts, format of the survey (face-to-face interview vs. telephone interview), distances, contact reminders for initial contact failure, and gender of the parent. A detailed description and analysis of attrition variables is then of major importance for future epidemiological studies, to improve the enrollment of participants at higher risk of dropout.

Discussion

Defining the prevalence of tic disorders is an important step in determining the public health impact of these disorders. Although tics might disappear in some cases by early adulthood, symptoms of co-occurring conditions may persist. Specific recommendations to improve the quality of epidemiological studies include the following. First, to provide an adequate definition of tic disorders with or without associated comorbidities, and to provide optimal sample selection (preferably by enrolling individuals from 6 to 18 years of age, and by sampling communities or performing school-based studies). Second, to use validated screening procedures, and preferably ones that are specific to that population and setting, and to use medical personnel with experience in tic disorders to ascertain cases. Third, to provide adequate coverage of the target population, including efforts to improve participation (e.g., phone calls, multiple mailings and postcard reminders, use of telemedicine, etc.). Fourth, to provide an adequate analysis of attrition variables in order to reduce sample selection bias.

Additional general recommendations would be as follows: to review the diagnostic criteria for tic phenomenology, to standardize the inclusion and exclusion criteria for participants' selection, to simplify the duration criterion, to revise the term "transient tic disorder", and to establish new tic disorder categories for those tics of secondary etiology.

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Appendix 1

Proxy Report Questionnaire for Parents and Teachers

Tics are abnormal movements with the following characteristics:

a. They are sudden, brief and rapid
b. They are repetitive
c. They can be controlled voluntarily during short periods of time
d. They can change and affect other body parts periodically
e. They improve and worsen from time to time
f. Boys are more frequently affected than girls

The most common tics are eye blinking; elevating the eyebrows; twitching the nose and the mouth, and shoulder shrugging; shaking the head; twitching the neck; touching objects, other people, or body parts (hair, nose, etc.); kicking the legs; throat clearing; sniffing, barking, and verbalizations.

According to these characteristics,

- Do you believe that your son/daughter has had tics? (yes or no)
- Do you believe that your son/daughter (or pupil) has tics? (yes or no)

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Appendix 2

Essential criteria of tics

1 Does your child make repeated and short movements or sounds? To give an example: constant blinking, screwing up of the nose, continuously clearing of the throat….
Yes ☐No ☐
2 Do these movements/sounds, despite being repetitive, change? To give an example: one period it may be blinking and in another period head shaking movements….
Yes ☐No ☐
3 Do these movements/sounds fluctuate over time? To give an example: are there days/periods of time with many movements/sounds and other days/periods of time with none.
Yes ☐No ☐
4 Is there a sense of "urgency" before making the sound/movement? To give an example: feeling of discomfort/itching in the area of the tic, sensory symptoms….
Yes ☐No ☐
5 Can he/she suppress them for a short period of time? To give an example: it is normal for children to have few tics in school and have more at home. Young people can even notice this suppression
Yes ☐No ☐
6 Is there a feeling of relief when the tic movements are made? To give an example: those affected can often make themselves nervous when they suppress tics for a long time and afterwards, they notice a decrease in this "tension".
Yes ☐No ☐
Obtained from Cubo et al, study.[48]