

Original article

Psychiatric disorders and behavioral problems in children and adolescents with Tourette syndrome

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Abstract

Objective: Many previous studies have surveyed associations between Tourette syndrome (TS) and co-morbid psychiatric disorders, but they usually did not include oppositional defiant disorder (ODD), conduct disorder (CD), separation anxiety disorder (SAD), and post-traumatic stress disorder (PTSD). **Method:** The subjects were children and adolescents with TS who visited a child and adolescent psychiatric clinic, and who were interviewed using DSM-IV diagnostic criteria. Characteristics of their tics were examined by the Yale-Global Tic Severity Scale (Y-GTSS). Behavioral problems were surveyed by the Child Behavior Checklist (CBCL) filled in by the parents. **Results:** About 87.9% of the subjects were boys. The mean age of the subjects was 11.8 years. The most common psychiatric disorders were attention deficit hyperactivity disorder (ADHD), ODD, nail biting, and obsessive compulsive disorder (OCD). Only one subject was affected by TS without co-morbidities. Among TS patients with co-morbidities, those with disruptive behavioral disorders (DBD) have significantly higher mean scores than patients without DBD on the Externalizing scale, Social problems, Attention problems, Delinquent and Aggression scales. Co-morbidity of anxiety disorders was not related to the CBCL scores. **Conclusion:** Many of our results were similar to those reported in studies conducted in other parts of the world. TS is more common in boys and nearly all of them had at least one co-morbid disorder. The most common co-morbidity was ADHD. Behavioral problems in TS are related to the co-morbidity with the DBD, and possibly not to the anxiety disorders.

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1. Introduction

Tourette syndrome (TS) is characterized by the expression of involuntary motor tics such as sudden gestures or facial movements, and vocal tics such as throat-clearing or utterances [1]. There is no diagnostic test for TS according to DSM-IV-Text Revised [1]. The diagnosis of TS must be accompanied with vocal tics. Vocal tics

are also helpful in ruling out other diagnoses because they are rare in other neurological conditions, except Huntington's disease and Sydenham's chorea [2].

TS is found in all cultures, ethnic groups and social classes. The typical age at onset for TS is 3–5 years of age and the highest prevalence of tics in the general population is at 3–5 and 9–12 years of age [3]. Although, simple tics are quite common, TS is a relatively rare syndrome and its rate is reported up to 0.56% in Taiwanese primary school children and 0.6% in the Swedish school population [4,5]. All of the studies thus far have reported that the rate of TS in boys is more than girls and the ratio is about 4.3:1 [6].

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The co-morbidity rates of attention deficit hyperactivity disorder (ADHD) and obsessive compulsive disorder (OCD) are 36% and 18%, respectively [4]. Among the clinical samples of children and adolescents with TS, 50–90% have co-morbid ADHD, conduct disorder, or oppositional defiant disorder [7]. About one-third of the TS patients have co-morbidity with OCD [8]. A recently published study on 17 children and adolescents with TS reported that the rate of TS without any co-morbidity was about 23.5% [9]. The prevalence of TS plus ADHD, TS plus OCD, and TS plus ADHD and OCD was 11.8%, 41.2%, and 23.5%, respectively [9]. A review study based on multiple sites and with an international database of 3500 individuals diagnosed with TS reported that 2–36% of the individuals with TS had no co-morbidity and the most common reported co-morbidity is ADHD [6]. The ADHD rate was 60% [6]. The rates of OCD and oppositional defiant disorder/conduct disorder (ODD/CD) were 27% and 15%, respectively [6].

The combined TS with ADHD group have significantly higher scores on the CBCL scale in comparison to the TS without comorbidity [10]. Aggressive behavior in TS may be associated with co-morbid ADHD or OCD [11]. However, the relationship of aggressive and explosive behavior (“rage attacks”) with TS is unclear and controversial [12]. TS causes the poor peer relationships in children [13]. Many of the recently published studies have only focused on TS and ADHD and usually have not included other co-morbid psychiatric disorders such as separation anxiety disorder [9], ODD, and CD [10]. There is no association between frequencies of OCD and severity of TS [14]. However, separation anxiety disorder most robustly predicts tic severity, irrespective of the presence of OCD or other anxiety disorders [14].

The majority of previous studies have not dealt with the co-morbid psychiatric disorders [15] or they have just included ADHD and OCD [10] or ADHD and ODD [9]. Therefore, it is essential to study co-morbidities which include more disorders such as SAD, ODD, and CD. There have been no published studies to date in Iran which have surveyed the clinical symptoms and psychopathology of children and adolescent with TS. The aim of this study was to examine the prevalence of co-morbid psychiatric disorders in children and adolescents with TS. In addition, it aimed to compare behavioral problems of different diagnostic categories of TS with other disorders.

2. Materials and methods

Subjects in this study were 35 children and adolescents with the diagnosis of TS who had referred for the first time to the child and adolescent psychiatry clinic. They had either not received any pharmaceutical

treatment or were drug-free for at least 4 weeks before the study.

The subjects were enrolled in the study if they met the DSM-IV-Text Revised diagnostic criteria for TS. They also needed to give history of multiple motor tics and one or more phonic tics, had to be younger than 19 years of age; and the children and their parents had to give their consent for participation in the study. Subjects with transient tics as well as cases with mental retardation, gross neurological or other organic disorders were not included. Physical examination was conducted for all patients.

All of the subjects and their parents were interviewed using the Farsi version of Schedule for Affective Disorders and Schizophrenia for School-age Children—Present and Lifetime version (K-SADS-PL) by a child and adolescent psychiatrist [16,17]. Diagnosis was made using clinical observation and history provided by the parents and the patients themselves. The KSADS-PL Farsi-Version has sufficient reliability and validity. It has been shown that consensual validity of all the psychiatric disorders is good to excellent. There is sufficient validity and test-retest and inter-rater reliability and good to excellent sensitivity and specificity and positive and negative predictive validity for nearly all of the disorders. Test-retest reliability of attention deficit hyperactivity disorder (ADHD) is 0.81. Inter-rater reliability of ADHD is 0.69. Sensitivity and specificity for tic disorders is 81.8% and 100% [17].

The semi-structured interview using the Yale-Global Tic Severity Scale (Y-GTSS) elicits the characters of tics. It provides information about number, frequency, intensity, complexity, and interference of motor and phonic symptoms with behaviors [18].

The parents of the children and adolescents filled in the Child Behavior Checklist (CBCL) [19]. The CBCL is a standardized questionnaire in which parents report about the frequency and intensity of behavioral and emotional problems of their child aged 4–18 years in the past 6 months. A response format is ‘0–2’ (0, not true; 1, sometimes true; 2, true). The CBCL has 11 scales including delinquency, aggression, withdrawal, somatic complaints, anxiety/depression, social problems, thought problems, attention problems, externalizing problems (includes delinquent and aggressive behavior), internalizing problems (including withdrawal, somatic complaints, and anxiety/depression), and total problems (including externalizing, internalizing, social, thought, and attention problems). The total behavioral problem scale score is the sum of all the responses to all the items [19]. The CBCL is one of the most widely used instruments in research and clinical work in children. It has been used in many studies in Iran and has enough validity and reliability [20]. The parents of the patients completed the CBCL-parent form while they were in the outpatient clinic room. Most of the parents completed

the questionnaire without difficulty. However, assistance including clear explanations was provided if needed.

Demographic data questionnaire included the patient's age, gender, and educational level.

The collected data were analyzed statistically with SPSS. Chi-squared analysis was used for categorical data and continuous data were analyzed by analysis of variance. Statistical significance was defined at 5% level and all tests were two-tailed.

3. Results

From the 35 subjects in this study; 31 of them (88.6%) were boys and four subjects (11.4%) were girls. The mean age of the subjects was 11.8 (SD = 3.1) years, with an age range of 6–18 years. The mean age at onset of tic was 9.1 (SD = 2.9) years. Delay in diagnosis of TS was about 2.26 (SD = 1.7) years.

About 80% of the children had at least one motor tic at the time of study. Sixty percent had at least one vocal tic. About 80% of the patients had more than one type of motor tic. The most common motor tics were blinking (54.3%), mouth movement (31.4%), and shoulder motor tic (31.4%). Vocalizations included throat clearing and grunting which were the most common vocal tics. Coprolalia was seen in 9.1%.

The mean age of subjects with TS with AD and TS–AD was 11.6 (SD = 3.8) and 12.0 (SD = 2.4) years old, respectively ($t = 0.4$, $df = 31$, $p = 0.6$). The mean age of subjects with TS with DBD and TS without DBD was 11.7 (SD = 3.1) and 12.1 (SD = 3.2) years, respectively ($t = 0.3$, $df = 31$, $p = 0.7$).

Gender was not related to co-morbidity of disruptive behavior disorders (DBD) in the subjects with TS ($\chi^2 = 1.6$, $df = 1$, $P = 0.2$). Anxiety disorders of diagnostic group was not related to gender ($\chi^2 = 0.03$, $df = 1$, $P = 0.8$).

Table 1 shows the co-morbid psychiatric disorders in the children and adolescents with TS. The common psychiatric disorders were ADHD, ODD, nail biting, and OCD. Only one out of the 35 subjects was affected by pure TS, without any other co-morbid psychiatric disorder.

A comparison of CBCL T scores of children and adolescents with TS between those with DBD and AD are shown in Table 2. TS subjects were divided on the basis of the presence/absence of the main co-morbid psychiatric diagnostic group, i.e. DBD(+) versus DBD(–), and AD(+) versus AD(–) (Table 1).

Comparison of the CBCL T scores revealed that TS patients co-morbid with DBD (25/35) have no significantly higher mean scores than patients without DBD on the total scale (52.8, SD = 10.2 versus 44.2, SD = 5.9). Meanwhile, the T scores of Externalizing scale, Social problems, Attention problems, Delinquent and Aggression scales in TS cases co-morbid with

Table 1
Frequency on co-morbid psychiatric disorders and diagnostic classifications in children and adolescents with Tourette disorder

Disorder	Number	Percent
<i>Mood disorders</i>		
Major depressive disorder	1	2.9
Bipolar mood disorder	4	11.4
<i>Anxiety disorders</i>		
Obsessive–compulsive disorder	9	25.7
Separation anxiety disorder	5	14.3
Post-traumatic stress disorder	2	5.7
<i>Disruptive behavior disorders</i>		
Attention deficit hyperactivity disorder	24	68.6
Oppositional defiant disorder	10	28.6
Conduct disorder	2	5.7
<i>Others</i>		
Schizophrenia	1	2.9
Autistic disorder	1	2.9
Enuresis	3	8.6
Lip biting	1	2.9
Bruxism	7	20.0
Nail biting	10	28.6
Sleep talking	7	20.0
At least one of the above-mentioned disorders	34	97.1
<i>Diagnostic classification</i>		
TS without co-morbidity	1	2.9
TS + disruptive behavior disorders	25	71.4
TS + anxiety disorders	15	42.9
TS + mood disorders	5	14.3
TS + disruptive behavior disorders + anxiety disorders + mood disorders	30	85.7

TS, Tourette disorder.

DBD were significantly more than in TS cases without DBD (Table 2).

On the other hand, no statistically significant difference emerged on the CBCL T scores between TS patients co-morbid with anxiety disorders and TS patients without co-morbidity of anxiety disorders.

Comparison of CBCL T scores of the three diagnostic subgroups of TS patients including TS with DBD, TS with AD, TS with DBD with AD revealed no significant difference between the groups for any subscales of CBCL.

4. Discussion

‘The prevalence of TS patients without co-morbidities was only 2.9% which is much lower than previously reported [9]. The prevalence of coprolalia was less than the rate of 14% which was reported in a review study [6]. The higher frequency of TS in boys is in accordance with all of the previously published studies [6]. Notably, delay in referring for diagnosis was 2.2 years which is much shorter than the rate of 6.4 years in other studies [6]. It might indicate that our subjects had referred much sooner than in other studies or another possible expla-

Table 2

Comparison of the means of the scores obtained on the CBCL (T-scores) in the simple diagnostic subgroups into which the TS patients could be divided on the basis of the presence/absence of the main co-morbidities investigated: DBD(+) vs DBD(–) and AD(+) vs AD(–)

CBCL scores	Disruptive behavior disorders (DBD)				<i>P</i> ^a	Anxiety disorders (AD)				<i>P</i> ^b
	With DBD		Without DBD			With AD		Without AD		
	Mean	SD	Mean	SD		Mean	SD	Mean	SD	
Total	52.8	10.2	44.2	5.9	0.058	49.3	10.7	50.4	9.9	0.8
Externalizing	52.9	9.9	43.4	6.6	0.02	48.5	9.	50.8	10.4	0.5
Internalizing	51.1	10.9	44.8	5.4	0.1	50.0	11.6	48.8	8.9	0.7
Withdrawal	49.9	10.9	50.2	7.7	0.9	50.5	9.6	49.6	10.6	0.8
Somatic complaints	51.6	11.2	45.8	3.6	0.1	49.8	0.4	11.9	8.5	0.8
Anxiety/depression	51.8	10.6	45.1	6.2	0.1	50.7	12.2	49.4	8.4	0.7
Social problems	52.6	9.4	44.3	9.4	0.04	48.8	11.0	51.4	8.9	0.5
Thought problems	50.3	10.8	48.9	7.7	0.7	50.5	8.07	49.4	11.7	0.7
Attention problems	53.2	8.8	41.1	7.5	0.002	49.0	12.4	50.7	7.9	0.6
Delinquent behavior	52.6	10.2	43.7	4.9	0.03	50.1	10.7	49.9	9.9	0.9
Aggressive behavior	52.5	9.7	43.3	7.5	0.02	49.4	10.3	50.4	10.0	0.8
Other problems	51.6	10.6	45.8	7.1	0.2	51.6	12.2	48.8	8.3	0.5

^a The difference of comparison of the means in the TS patients on the basis of the presence/absence of DBD.

^b The difference of comparison of the means in the TS patients on the basis of the presence/absence of AD.

nation is that these subjects were suffering from more severe forms of TS or co-morbid psychiatric disorders than in the other studies. The rate of mood disorder was relatively low. Depression in TS is related to the duration of TS [21].

Nearly all of the subjects with TS were co-morbid with at least one of the psychiatric disorders. This is in accordance with reports that TS alone is the exception rather than the rule in different clinical samples [22]. The most common co-morbid disorder was ADHD. This is also in accordance with prior studies that the rate of ADHD in TS is as frequent as 60–90% [14,23,24]. Furthermore, the rate of OCD was about 25% which is much lower than the rates of 32% and 50% reported by some other studies [6,3].

Meanwhile, our co-morbidity rates were completely different from a study on a clinical sample which reported that the prevalence of TS plus ADHD, TS plus OCD and TS plus ADHD and OCD was 11.8%, 41.2%, and 23.5%, respectively [9]. The most common co-morbid psychiatric disorder in their study was OCD and then ADHD [9]. This is in contrast with our study's results but it is also not in accordance with other mentioned studies. In addition, their study reported that the subscales of CBCL were not different between the two groups of TS with ADHD and TS without ADHD, except for aggressive behavior. The lack of difference between subscales of CBCL between TS with AD group and TS without AD is not in accordance with the study that reported that internalizing score and anxiety/depression score of TS with OCD are more than TS without OCD [9]. It is notable to state that their study was only conducted on 17 patients and the number of children with TS with ADHD was just two patients and the number of TS with OCD patients was only 7. Furthermore, they had not examined many of the other

psychiatric disorders such as ODD, and SAD [9]. It is possible that some of the subjects, who were not suffering from OCD, were suffering from another anxiety disorder and this might have affected their study results. Finally, there is a study that co-morbidities of TS change with age [25] but the mean age of our study sample was very similar to their's (11.8 versus 11.4 years) [9].

Lack of significant association of groups of TS with DBD and TS without DBD, and groups of TS with AD and TS without AD might be due to the low number of female patients. The lack of a meaningful difference between mean age of the TS with AD and TS without AD might be related to the low sample size.

The association of CBCL *T* scores of TS with DBD group and externalizing, aggression, attention, delinquent problem subscales is in accordance with results that the co-occurrence of TS and ADHD can be associated with disruptive behaviors such as aggression, explosive behavior, and low frustration tolerance [12,26,27]. It is interesting to note that many of the previous studies have not included other disruptive behaviors (ODD and CD) [9,10,15].

The CBCL *T* scores of TS patients with AD and TS patients without AD were not significantly different while some of the subscales were different between the two groups of TS with DBD and TS without DBD. This might support the idea that DBD is an integral part of the clinical spectrum of TS and shares a common etio-pathogenetic basis. At the same time, it is not supportive of AD being an integral part of the clinical spectrum of TS and sharing a common etio-pathogenetic basis.

Although the CBCL was filled out by the parents, the higher scores of externalizing items in the TS with DBD group in comparison to the TS without DBD group are congruent with the clinical interview results.

The different pattern shown by these data in comparison to some of the previous studies might reflect different referral patterns. It is possible that many of the children and adolescents with TS have not had time to develop co-morbid disorders and there is a probability that some of them will show it in the future.

There were some limitations to this study that should be considered in the interpretation and generalization of the results. This study was based upon cases at a single site and OCD intensity was not considered. Also, more importantly CBCL is a questionnaire filled in by the parents.

Despite these limitations, this study was based on the clinical interview conducted by child and adolescent psychiatrist, using standard measures, and included all the psychiatric disorders in a considerable number of children and adolescents with TS. Further prospective studies with larger samples sizes from different sites are recommended.

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