

Behavioral and Emotional Problems in Adolescents with Tourette Syndrome

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Background: Patients with Tourette syndrome (TS) are at risk of an array of behavioral and emotional problems, resulting in social, academic and vocational function impairments. This study intended to examine the nature and severity of behavioral and emotional problems in Taiwanese TS adolescents.

Methods: Forty TS adolescents with normal IQ and thirty age- and gender-matched normal controls were evaluated using the Yale Global Tic Severity Scale (YGTSS) and the Child Behavioral Checklist (CBCL) to understand the severity of tic symptoms, and behavioral and emotional problems. The main caretakers of these adolescents were interviewed using the Chinese version of the Schedule for Affective Disorders and Schizophrenia (CK-SADS) to confirm their comorbid psychiatric diagnoses.

Results: Most TS adolescents in this study had mild tic severity. TS adolescents showed significantly higher scores than normal controls in all CBCL subscales. The 'total most severe tics' YGTSS score was positively correlated with internalization behavior problems, externalization behavior problems and aggressive behavior subscales of the CBCL. As TS adolescents got older, their CBCL scores decreased significantly in internalization behavior problems, externalization behavior problems, and obsessive-compulsive and aggressive behavior subscales.

Conclusion: Taiwanese TS adolescents with mild to moderate tic severity still demonstrated prominent behavior and emotional problems. Although the severity of behavior and emotional problems decreased with increasing age, we still suggest systematic inquiry regarding the psychological well-being and psychiatric comorbidities of young TS patients.

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Key words: tourette syndrome, behavior, adolescent

Tourette syndrome (TS) is a complex, neuropsychiatric disorder characterized by repetitive, recurring, involuntary motor and vocal tics. It is now noted that TS patients have a 5- to 20-fold increased

risk of various behavioral and emotional problems compared to their normal peers, and that these problems frequently result in impairments in their social, academic and occupational functioning.⁽¹⁻³⁾ The

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recent study by Zhu et al. showed that the severity of tic symptoms was positively correlated with overall impairments in school and social competence in Chinese TS children and adolescents.⁽⁴⁾ However, an association between tic symptom severity and behavioral disturbance was not established in other studies.^(5,6)

In clinical practice, it is known that the treatment of tic symptoms alone will not improve the overall outcome of TS patients. Although tic related phenomenon might dissipate with age, some studies have shown that the behavioral problems in TS patients persist or become more severe.^(7,8) Research focusing on understanding the course and severity of these comorbid problems in TS patients will improve clinicians' treatment strategies.

The prevalence rate of TS in Taiwan has been estimated at around 0.56%, with 36% having Attention Deficit Hyperactivity Disorder (ADHD), 27% exhibiting minor self-injury behavior and 18% showing obsessive-compulsive symptom (OCS) comorbidity.⁽⁹⁾ Although previous studies have shown that TS patients of all ages present with different behavioral and emotional problems comorbid with different psychiatric disorders, there have not been related studies in Taiwan.^(10,11) It is both interesting and important to understand the relationship between the severity of tic symptoms and the coexisting behavioral and emotional problems, and also the changes of behavioral and emotional problems with increasing age in Taiwanese TS patients.

This study, approved by the Ethics Committee of the Chang Gung Memorial Hospital, was designed to explore the relationship between tic symptoms, age and the behavioral and emotional problems of Taiwanese TS adolescents.

METHODS

Forty adolescents with TS were voluntarily recruited from the Pediatric Neurology and Child Psychiatry clinics in a medical center in northern Taiwan. Written informed consent was obtained from all subjects and their parents. All subjects were paid to participate.

Subjects and their primary caregivers were interviewed. Diagnosis of TS was conducted and confirmed by an experienced child neurologist (HS Wang) and child psychiatrist (HL Chang) based on

the Diagnostic and Statistical Manual of Mental Disorders (DSM-IV) criteria.

A control group was also recruited, made up of thirty subjects with comparable gender and age with the TS subjects. After having obtained written informed consent from the subjects and their parents, we conducted an interview with the parents before the study to ascertain that none presented a positive history of tics or tic-like symptoms.

Stage 1. All subjects were assessed using the Wechsler Intelligence Scale for Children (WISC-III)⁽¹²⁾ administered by an experienced clinical psychologist (NC Ko). Only subjects with normal IQ (IQ > 79) were recruited for the second stage of the study.

Stage 2. Subjects completed the Yale Global Tic Severity Scale (YGTSS), a semi-structured interview designed to elicit information concerning the specific character and anatomical distribution of tics. The interview begins with systemic assessment of current tic symptoms (both motor and phonic tic symptoms) that the clinician rates as present or absent over the past week. This is followed by assessing the most severe tic symptoms as perceived by the patient. Motor and phonic tic symptoms are rated according to number, frequency, intensity, complexity and interference on a 6-point ordinal scale. Patients' Current Total Tic score and Total Most Severe Tic score ranged from mild to severe (0 = absent, 1-5 for severity). YGTSS is a widely used scale with excellent psychometric properties.⁽¹³⁾

Stage 3. Main caregivers of all subjects were examined using the Chinese version of the Kiddie-Schedule for Affective Disorders and Schizophrenia - Epidemiology version (CK-SADS-E). The CK-SADS-E is a semi-structured diagnostic interview and was administered by a child psychiatrist (HL Chang). CK-SADS-E is designed to assess psychiatric disorders in children from 6 to 16 years of age.⁽¹⁴⁾ Development of the Chinese version of the K-SADS-E was carried out by the Child Psychiatry Research Group in Taiwan, which included a two-stage translation and modification of several items with psycholinguistic equivalents relevant to Taiwanese culture.⁽¹⁵⁾ The inter-rater reliability of the CK-SADS-E ranged from 0.73 to 0.96 for all mental disorders included in the CK-SADS-E. The overall sensitivity and specificity of the screening interview against any CK-SADS-E diagnostic category were

calculated to be 78% and 98%, respectively.

Caregivers for both TS subjects and controls also completed the Child Behavior Checklist (CBCL). The CBCL is a widely-used questionnaire designed to assess the behavioral problems and social competence of children 4 to 18 years of age. In the current study, the CBCL revised by Achenbach was used after being translated into Chinese.^(16,17) The CBCL-Chinese (CBCL-C) consists of 20 items that assess social competence, and 118 items concerning behavioral and emotional problems. The questionnaires were scored using eight measures: withdrawal, somatic complaints, anxiety and depression, social problems, cognitive problems, attention problems, delinquent behavior and aggressive behavior. They were grouped into two broad band scores: internalization behavior problem score, consisting of withdrawal, somatic complaints, anxiety and depression, and externalization behavior problem score, consisting of delinquent and aggressive behaviors. Standardized T scores with a mean of 50 [standard deviation (SD) = 10] were provided for each subscale. A T score greater than 60 indicated a 'borderline clinical range'. The higher the score, the more severe the child's behavioral and psychological problems.⁽¹⁸⁾

All statistical analyses were conducted using SPSS for Windows software (version 8.0).⁽¹⁹⁾ Data analyses focused on examining the relationship among the various subscales of CBCL and tic severity. TS subjects and controls were compared using the *t*-test for independent variables. Pearson-moment correlations were used where appropriate to examine the relationship between psychiatric comorbidity and clinical data. A *p* value of less than 0.05 was considered statistically significant.

RESULTS

The characteristics of TS subjects

Forty adolescents with TS (28 males and 12 females, 12.0 ± 1.4 years) were recruited. Twenty-one of them were treated with clonidine (35.5-75.0 mcg) and 15 with pergolide (0.05-0.25 mg) at the time of the study. There was a significant difference between male and female patients (*t* = 17.72, *p* < 0.000). The gender differences in our TS group reflected general epidemiological findings. The mean average score of total most severe tic assessed by YGTSS was 25.7 ± 10.5 (range 6-49). Most patients had mild tic severity (YGTSS current total tics < 20) at the time of interview (Table 1).

Table 1. Comparison of IQ, YGTSS and CBCL Scores of TS Group and Controls in IQ

	TS n = 40 Mean ±SD	Control n = 28 Mean ±SD	T-test	<i>p</i> value
Full IQ	106 ± 7	110 ± 11	-1.64	0.11
Verbal IQ	104 ± 7	106 ± 10	-1.30	0.19
Performance IQ	108 ± 12	113 ± 13	-1.42	0.16
YGTSS total current	19.4 ± 11.7	N/A		
YGTSS total most severe	25.7 ± 10.5	N/A		
CBCL Internalization	63.4 ± 14.2	48.3 ± 10.9	4.54	0.000
CBCL Externalization	58.8 ± 13.3	48.8 ± 10.8	3.15	0.003
CBCL Obsessive-compulsive	60.4 ± 12.3	50.3 ± 8.9	3.57	0.001
CBCL Somatic complaints	60.8 ± 11.9	49.9 ± 8.4	4.14	0.000
CBCL Withdrawn	60.3 ± 10.9	52.6 ± 8.4	2.98	0.004
CBCL Hyperactivity	66.0 ± 13.1	50.8 ± 9.6	3.0	0.003
CBCL Aggressive behavior	60.6 ± 13.5	51.4 ± 8.9	3.07	0.003
CBCL Uncommunicative	61.9 ± 14.6	49.4 ± 11.4	3.088	0.003
CBCL Depression	74.4 ± 16.9	54.5 ± 8.4	3.81	0.001
CBCL Anxiety	62.6 ± 15.2	50.7 ± 8.5	3.63	0.001

Abbreviations: YGTSS: Yale Global Tic Severity Scale; CBCL: Child Behavior Checklist; TS: Tourette Syndrome; SD: standard deviation; N/A: not assessed.

Comparison between TS patients and controls

Thirty age-, gender- and IQ-matched healthy adolescents were also recruited to participate in the study. Two subjects from the control group were excluded from the study because of comorbid with ADHD. The average age for control subjects was 12.1 ± 1.5 years. There was no significant difference in age and IQ between the TS patients and controls.

TS patients had significantly higher (more severe) scores than controls in all CBCL subscales (Table 1): 46.7% of TS patients had a CBCL Hyperactivity subscale score exceeding the clinical cut off point, 56.6% had Anxiety subscale scores exceeding the clinical cut off point and 30% had Depression subscale scores exceeding the cut off point. From the CK-SADS-E clinical interviews, 22.5% of TS patients met DSM-IV criteria for ADHD, 7.5% met DSM-IV criteria for anxiety disorder and 2.5% met DSM-IV criteria for depressive disorders. Of the controls, 14.2%, 21.4% and 7.8% exceeded the cut off points in Hyperactivity, Anxiety and Depression subscales, respectively but none met DSM-IV criteria for ADHD or anxiety/depressive disorders.

Correlations between CBCL subscales and YGTSS total most severe tic score

Pearson correlation was used to examine the relationships between CBCL subscales and YGTSS total most severe tic score. Although all CBCL subscales were positively correlated with YGTSS total most severe tic scores, only Internalization behavior problems ($r = 0.418, p < 0.05$), Externalization behavior problems ($r = 0.452, p < 0.05$) and Aggressive behavior ($r = 0.419, p < 0.05$) subscales reached significant levels. This positive correlation suggested that the patients with more severe tic symptoms showed more behavioral and emotional problems, and more aggression (Table 2).

To examine whether behavioral and emotional problems in TS patients are related to tic severity, CBCL scores that were significantly related to YGTSS total most severe tic scores were analyzed by multiple linear regression. By R^2 assessment, YGTSS most severe tic scores accounted for 17.4%, 20.4% and 17.5% of the Internalization behavior problems, Externalization behavior problems and Aggressive behavior, respectively.

Correlations between CBCL subscales and age of TS patients

Pearson correlation was used to examine the relationships between CBCL subscales and ages of TS patients. All CBCL subscales were negatively correlated with age but only Internalization behavior problems ($r = -0.441, p < 0.05$), Externalization behavior problems ($r = -0.418, p < 0.05$), Aggressive Behavior ($r = -0.465, p < 0.01$) and Obsessive-Compulsive ($r = -0.408, p < 0.05$) subscales reached significant levels (Table 3). These negative correlations indicated TS adolescents outgrew these problems as they got older (Figs. 1-4).

For the TS patients, we attempted to predict significant CBCL subscales by age using simple linear regression analysis. For the Internalization subscale, slope = -4.653 ; 95% CI = -8.31 to -0.991 ; $t = -2.603$; $p < 0.05$; $Y = -4.653X + 119.4$; $r^2 = 0.19$. For the Externalization subscale, slope = -4.131 ; 95% CI = -7.60 to -0.66 ; $t = -2.44$; $p < 0.05$; $Y = -4.131X + 108.51$; $r^2 = 0.175$. For the Aggressive behavior subscale, slope = -4.636 ; 95% CI = -8.01 to -1.22 ; $t = -2.77$; $p > 0.1$; $r^2 = 0.22$. For the Obsessive-Compulsive subscale, slope = -3.707 ; 95% CI = -6.919 to -0.496 ; $t = -2.465$; $p < 0.05$; $r^2 = 0.166$. The slope of the regression line was signifi-

Table 2. Association of YGTSS Total Most Severe Tic Score and Significant CBCL Subscales

CBCL subscales	Internal-ization	External-ization	Aggressive behavior
r	0.418	0.452	0.419
p value	0.022	0.012	0.020
95% CI	0.09 – 1.05	0.14 – 1.02	0.09 – 0.99

Abbreviations: YGTSS: Yale Global Tic Severity Scale; CBCL: Child Behavior Checklist; r: Pearson coefficient; sig.: significant (2 tailed); CI: confidence interval.

Table 3. Association of Age and Significant CBCL Subscales

CBCL subscales	Internal-ization	External-ization	Aggressive behavior	Obsessive-compulsive
r	-0.441	-0.418	-0.465	-0.408
p value	0.015	0.021	0.010	0.025
95% CI	-0.832 – -0.99	-7.60 – -0.66	-8.06 – -1.22	-6.92 – -0.49

Abbreviations: CBCL: Child Behavior Checklist; r: Pearson coefficient; sig.: significant (2 tailed); CI: confidence interval.

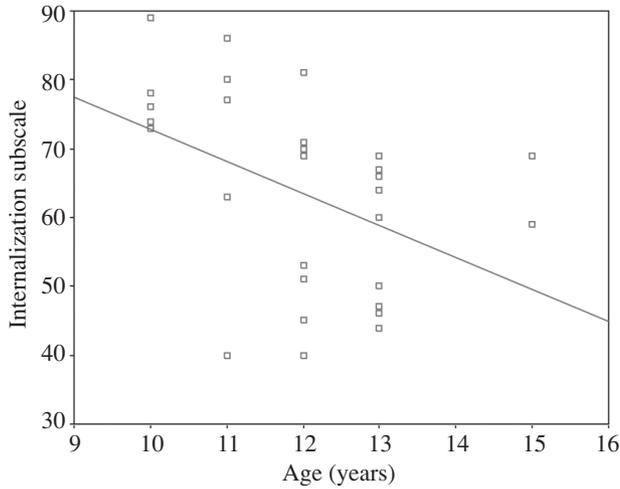


Fig. 1 Association between age and Child Behavior Checklist Internalization behavior.

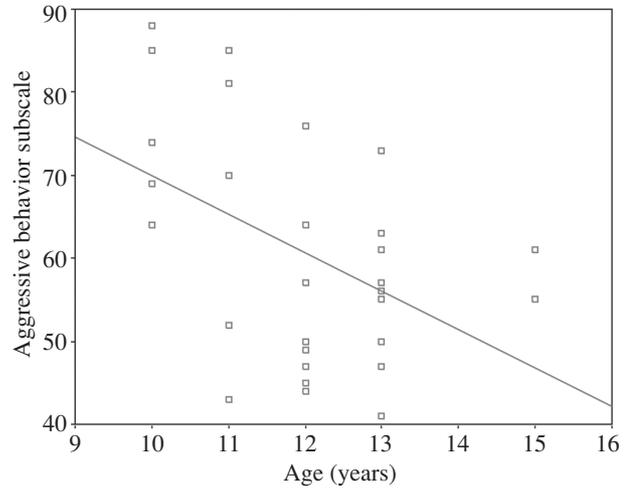


Fig. 3 Association between age and Child Behavior Checklist Aggressive behavior.

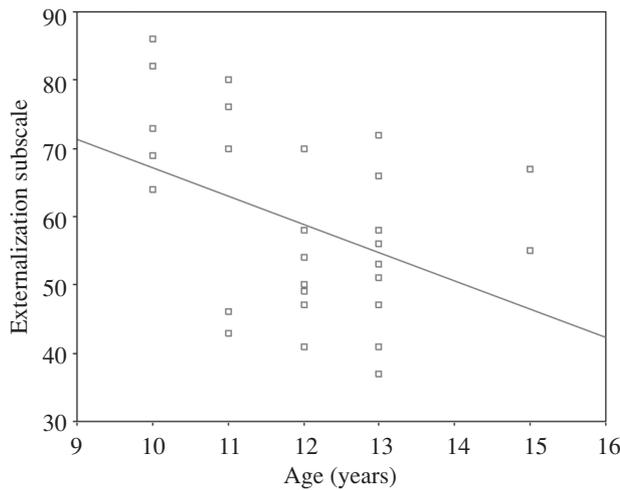


Fig. 2 Association between age and Child Behavior Checklist Externalization behavior.

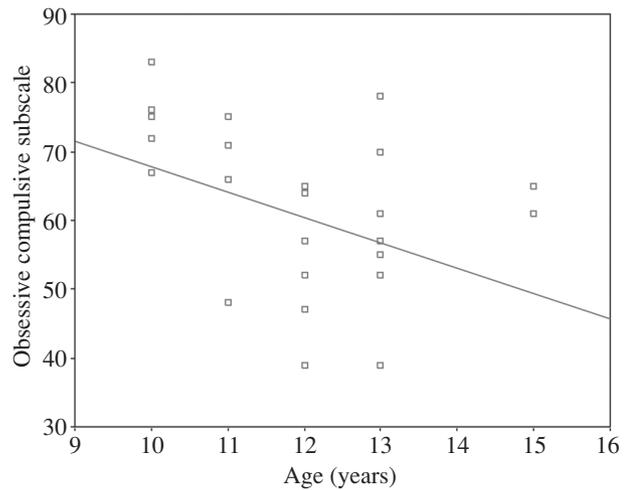


Fig. 4 Association between age and Child Behavior Checklist Obsessive-Compulsive behavior.

cantly smaller than zero, indicating that these CBCL subscales tended to decrease as age increased.

DISCUSSION

It has been a common clinical experience that the treatment of tic symptoms alone in TS patients does not improve such patients' overall problems.^(20,21) This is because, although vocal and motor tics are the most prominent symptoms that define TS, but for many children with tics, the associated

emotional and behavioral problems are more disruptive among the tic symptoms.^(22,23)

This is the first report in which Taiwanese TS adolescents with normal intelligence were well-characterized with regard to diagnosis, tic severity and comorbidity, and were matched to controls. In this study, TS adolescents had significantly higher CBCL scores on internalizing problems, externalizing problems, obsessive-compulsive behavior, withdrawal, hyperactivity, anxiety/depression and aggressive behavior than normal controls.

This study also assessed the relationship between tic severity, and behavioral and emotional problems in TS adolescents. We found that their tic severity positively correlated with internalization behavior problems, externalization behavior problems and aggressive behavior. In summary, TS adolescents experienced a variety of internalization and externalization behavior problems.

At the time of assessment, most of our patients suffered from mild tic severity. All our TS patients were receiving medication; it is possible that their tic problems were more severe in the past. However, even with the decrease of tic severity after treatment, TS patients' caregivers still reported significantly more emotional and behavioral problems. From this study, we believe that even in TS patients with a reduction of tic symptoms to a mild degree after medical treatment, behavioral and emotional problems are still more prominent than in normal controls. The behavioral and emotional problems in TS may result from previous severe tic symptoms that caused frustration in peer interaction and built negative self concept.^(5,6) Alternatively, tic symptoms and psychiatric symptoms may originate from a different neurobiological mechanism, thus the elimination of tic symptoms will not improve the psychiatric problems at the same time.^(24,25)

Although our study showed that internalization behavior problems, externalization behavior problems and aggressive behavior are positively correlated to tic symptoms, this only explained about 20% of the problems by R^2 analysis. As a cross-sectional study, our study could not address the influence on behavioral and emotional problems that may be due to the effect of a chronic illness or the specific effect of tic symptoms alone. Further research should explore the interaction of chronic tic symptoms with behavioral and emotional status of TS children and adolescents.

In this study, TS adolescents showed a reduction in behavioral and emotional problems with increasing age, suggesting improved functioning in TS adolescents with mild tic severity as they grow older. Previous studies have indicated that the various comorbid psychiatric illnesses in TS had different peak ages, thus the behavioral and emotional problems in TS may occur independently of tic severity.⁽²⁶⁾ However, so far there are no precise clinical prognostic indicators to assist in determining tic

severity and its relation or outcome with behavioral/emotional problems.

The long-term influence of behavioral and emotional problems on the global outcome of TS is controversial. Some studies have shown that children with mild to moderate tics can also be withdrawn, aggressive and less popular, and tic severity is not a linear predictor of behavioral disturbance.⁽²⁷⁾ The most significant clinical implication of this study is that TS adolescents with mild to moderate tic severity still show prominent behavioral and emotional problems. We, thus, suggest that systematic inquiry regarding the behavioral and emotional problems of TS adolescents should be a general clinical procedure. The identification of these problems in TS adolescents could lead to appropriate intervention and treatment strategies.

Limitations

There are some limitations in this study that require careful consideration in the interpretation of these findings. As our study was a cross-sectional study, we could not fully assess the relationship between tic symptoms, age, and behavioral and emotional problems. Thus, our findings should be interpreted as correlational and not causal. In addition, our studied population was a clinically referred sample; our findings may not generalize to non-referral TS populations. It is possible that TS adolescents with more behavioral and emotional problems are treated at medical centers more often than TS adolescent patients without these problems.

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青少年妥瑞氏症患者的行爲及情緒問題

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背景：青少年妥瑞氏症患者合併行爲及情緒問題的危險性高，常導致患者在社會及職業功能、學業成就上的損害。本研究的目的在了解台灣地區青少年妥瑞氏症患者行爲及情緒問題之嚴重度與病徵。

方法：研究對象爲 40 名患有妥瑞氏症之智能正常青少年，及 30 名性別、年齡、智商吻合之正常對照組。全部受試者均以耶魯整體抽搐嚴重度量表 (YGTSS) 及兒童行爲量表 (CBCL) 來評估抽搐嚴重度及其行爲與情緒之問題。研究中同時以中文版 K-SADS 針對主要照顧者，評估受試者之精神科共病症。

結果：本研究共有 28 名男性及 12 名女性妥瑞氏症青少年，男女比例達有意義的差別。妥瑞氏症青少年的平均年齡爲 12.0 ± 1.4 歲。研究中之妥瑞氏症青少年大多爲抽搐症狀較輕微之個案。妥瑞氏症青少年在 CBCL 各項得分均有意義的高於對照組。YGTSS 中最嚴重之抽搐症狀得分與 CBCL 中內化、外化行爲問題及攻擊行爲成正相關。年齡越長的妥瑞氏症青少年，其 CBCL 中內化、外化行爲問題、強迫症狀及攻擊行爲得分越低。

結論：本研究顯示，即使是抽搐症狀較爲輕微的妥瑞氏症青少年，亦合併多項行爲及情緒問題。妥瑞氏症青少年的行爲及情緒問題雖會隨著年齡的增長而減緩，但臨床醫師在診治妥瑞氏症青少年時，仍應固定篩選其行爲及情緒問題，及精神科共病診斷的可能性。

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