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ORIGINAL CONTRIBUTION

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Obsessive compulsive behaviour and depressive symptoms in young people with Tourette syndrome

A controlled study

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Abstract Tourette syndrome (TS) is characterised by multiple motor and one or more vocal tics. There have been no controlled studies using standardised instruments of depressive symptoms and obsessive compulsive symptomatology (OCS) in young people with TS. We completed a study of phenomenology and psychopathology in children with TS, including a controlled evaluation of the association between depressive symptoms, OCS, and TS. 57 people aged 15 or under with TS were recruited. Phenomenology and psychopathology were assessed using standardised instruments. The association between TS, depressive symptoms and obsessionality was investigated using 75 age- and gender-matched controls. There were high levels of depressive symptomatology and OCS in the TS group. Twenty-three (40 %) had carried out self-injurious behaviours and 34 (60 %) met criteria for Attention Deficit Hyperactivity Disorder (ADHD). Depressive symptoms and obsessionality were higher in the TS cohort compared with the control group; this excess persisted after adjustment for the effects of age, gender and comorbidity between depression and obsessionality. This study demonstrates high levels of psychopathology in children with TS, including ADHD, OCS and depressive symptoms. The findings illustrate the potentially complex, challenging combination of difficulties encountered by children with TS and those who care for them.

Key words Tourette syndrome – psychopathology – phenomenology – depression – obsessive compulsive symptomatology – controlled study – young people – children

Introduction

Tourette Syndrome (TS, also known Tourette Disorder or Gilles de la Tourette's Syndrome) is characterised by multiple motor and one or more vocal tics [1, 35]. The mean age of onset of the motor tics in TS is around 7 years of age (range 2–15), while vocal tics begin at 11, and coprolalia, if present begins at around 14–15 years [19, 20]. Although it is accepted that young people with TS have important disturbances of behaviour [33], there have been few systematic studies of the phenomenology and psychopathology of children with TS [9, 16, 31]. Two studies of depression often quoted [11, 34] are small in scale (10 and 33 children respectively) and gathered no data from the young people themselves. Rosenberg et al. [27] using the Child Behaviour Checklist reported that a third of nearly 200 children with TS had depressive symptomatology. Spencer et al. [29] compared groups of children with TS (n = 32), chronic tics (n = 39) and unaffected schoolchildren (n = 28) and reported a higher prevalence of depression in the TS (29 %) and tic groups

(33%) than in the control group (3%). More recently, Carter et al. [8] used the Children's Depression Inventory (CDI) [14] on a group of children with TS with (n = 33) and without (n = 16) Attention Deficit Hyperactivity Disorder (ADHD) and reported higher CDI scores for both TS groups compared with controls (n = 23). High levels of obsessive compulsive symptomatology (OCS) have also been reported in young people with TS [9, 16]. However, there is a lack of data on the associations between depressive symptoms and OCS in TS. We therefore carried out a systematic study of phenomenology and psychopathology in a cohort of children with TS including a controlled evaluation of the association between depressive symptoms, OCS, and TS. The rationale for the study was to address the gaps in the knowledge base using well-validated instruments and an adequate sample size, with the idea that the nature and extent of psychopathology associated with TS may have treatment implications for those children affected.

Method

Subjects

A consecutive series of 57 newly referred people aged 15 or less fulfilling DSM-III-R [1] criteria for TS were enrolled into the study from the Tourette Clinic at the National Hospital for Neurology and Neurosurgery Queen Square, London. This clinic is a national resource receiving both secondary (from GPs) and tertiary referrals (from other hospital specialists). To elicit and record the diagnosis of TS and associated phenomenology systematically, subjects were assessed using the National Hospital Interview Schedule for Gilles de la Tourette Syndrome (NHIS), which incorporates a detailed history taken from parents or other carers as well as a direct examination of the children [24]. The NHIS is a semi-structured interview schedule for the evaluation of TS and related behaviours; the psychometric properties of this instrument have been established with good inter-rater reliability and concurrent validity [24]. In addition, subjects completed the Birleson Depression Self-Rating Scale for Children (DSRS) [5, 6] and the childhood version of the Leyton Obsessional Inventory (LOI) [3, 4]. ADHD was assessed using DSM-IIIR criteria which are incorporated into the NHIS. Data on OCS were obtained by proxy from the parent interview in the NHIS and by self-report.

Controls

Seventy-five students were randomly selected as controls from five classes attending two mainstream schools (one primary and one secondary) in East London. This was completed after the age and gender distribution of the TS cohort had been ascertained. On the basis that school classes are to a large extent age determined, batch age matching was carried out by drawing random samples of pupils from a random selection of classes, based on the age distribution of the TS cohort. Following this a gender-specific random sampling ratio was applied to yield 80% males and 20% females to correspond with the gender distribution of the TS cases. These subjects all consented to participate, their age and gender were recorded and they then completed the DSRS and the LOI. The results from this group were compared with data from the subjects with TS.

Statistical analysis

Descriptive statistics were calculated to summarise the phenomenology of the TS group. Data were analysed using Mann Whitney U tests for non-normally distributed continuous variables, and χ^2 statistics for categorical variables. In order to investigate differences in DSRS and LOI scores between the TS and the control groups univariate associations were first calculated using simple t tests. Next, multivariate analyses were carried out using the General Linear Model in order to calculate the proportion of the variance of DSRS and LOI scores which was attributable to belonging to the TS group (the η statistic) controlling for the effects of age and gender. Analyses were completed using SPSS 8.0 for Windows and Confidence Interval Analysis.

Results

Demographic data

Fifty-seven subjects were studied in the TS group; 45 (79%) were male. Their mean age was 11.3 years (sd 2.4; range 4 to 15; 95% confidence interval (95% CI) 10.6 to 11.9). The mean age at onset of TS symptoms was 6 years (95% CI 5.4 to 6.6). The mean age at diagnosis was 10 years (95% CI 9.3 to 10.7). The mean duration of TS was 6 years (95% CI 5.2 to 6.8). The mean cumulative number of motor tics each individual had had was 25 (95% CI 21.7 to 28.3) and of vocal tics was 7 (95% CI 5.4 to 8.6).

Phenomenology

Twenty-three (40%) had coprolalia, 14 (25%) copropraxia, and 8 (14%) mental coprolalia. Twenty-two (39%) had echolalia, 18 (32%) echopraxia, 17 (30%) palilalia, and 4 (7%) palipraxia. Tics were improved by relaxation in 15 (26%), concentration in 8 (14%), sport in 4 (7%), and other non-specified situations in 4 (7%). Tics were aggravated by stress in 30 (53%), tiredness in 3

(5 %), boredom in 2 (4 %), and other non-specified situations in 4 (7 %). Eighteen (32 %) reported no aggravating factors. Nineteen (33 %) walked in their sleep, 34 (60 %) talked in their sleep, 15 (26 %) had night terrors, and 25 (44 %) required a night light to sleep. Twenty-three (40 %) had carried out self-injurious behaviours, of which the most common was head banging (n = 12; 21 %).

Fourteen (25 %) had obsessional thoughts, including 7 (12 %) who had obsessional thoughts involving violent scenes. Twenty-three (41 %) had compulsive rituals, and 11 (19 %) reported themselves to be, or were reported to be, excessively tidy. Seventeen (30 %) exhibited "evening-up" behaviours (a concern with symmetry), and 34 (60 %) forced touching of objects of whom 13 (23 %) touched dangerously hot objects, such as hot cookers or irons. Arithmomania (obsessions and compulsions involving excessive and inappropriate counting) was reported in 13 (23 %) cases. In only 5 (9 %) cases was the OCS reported to be bothering socially, and only one (2 %) subject had received specific treatment for OCS prior to assessment.

Thirty-six (63 %) had a history of hyperactivity, 35 (61 %) of impulsivity, and 35 (61 %) of inattention; 34 (60 %) met DSM-III-R criteria [1] for ADHD. Seventeen (30 %) had a history of being aggressive to people, and 9 (16 %) of aggression towards property.

Controlled study of depressive and obsessive compulsive symptomatology

The mean age of the control group was 10.7 (sd 3.0; range 6 to 17; 95 % CI 10.0 to 11.4), the mean age of the TS group was 11.3. There was no statistically significant difference in age when the two groups were compared calculating the 95 % CI for the difference in mean age and using a t test (95 % CI –0.5 to 1.5; $t = 1.08$, $p = 0.282$). Seventy-nine per cent of the TS group were male compared with 85 % of the control group, this was not statistically significant ($\chi^2 = 0.92$, df 1, $p = 0.338$).

The mean DSRS score for the 57 subjects in the TS group was 8.2 (95 % CI 7.0 to 9.4; range 0–19), and for the 75 controls was 5.8 (95 % CI 5.2 to 6.4; range 0–14). The control group mean was statistically significantly lower than the TS group ($t = 3.79$; $p < 0.001$). The mean LOI score for the 56 subjects in the TS group who completed the instrument was 7.2 (95 % CI 5.9 to 8.6; range 0–20) and the mean interference score was 8.3 (95 % CI 5.9 to 10.7; range 0–38). For the control group the mean LOI score was 4.3 (95 % CI 3.6 to 5.1; range 0–15) and the mean interference score was 4.1 (95 % CI 3.2 to 5.0; range 0–13). The mean LOI and interference scores were significantly lower in the control group than the TS group (LOI: $t = 4.0$; $p < 0.001$; interference score: $t = 3.6$; $p < 0.001$).

The results of the multivariate modelling to assess the association of having TS with DSRS score controlling for the effects of age and gender are presented in Table 1. The strong association between TS and ratings of depression and OCS observed using univariate statistics remained, with being a case of TS accounting for: 8.7 % of the variance in DSRS scores ($p = 0.001$); 10.3 % of the variance of total child LOI score ($p < 0.001$); and 8.5 % of the child LOI interference score ($p = 0.001$). Given the potential for correlation of depressive symptoms and obsessionality a statistical model was constructed to investigate whether there was still an association between having TS and depression score controlling for the contribution of obsessionality. In this, the association remained but was diminished in strength with being a case of TS accounting for 3.8 % of the variance in DSRS score ($p = 0.027$).

Discussion

This study provides the first detailed systematic description of TS and its associated psychopathology and phenomenology in a cohort of young people. It also demonstrates statistically significant associations between depression, obsessionality and TS.

Limitations

The main limitation of this study is that it was carried out in a specialist clinic so our findings may be subject

Table 1 Multivariate analyses of associations between TS and depression and obsessionality controlling for the effects of age and gender

Dependent variable	Variables	% age of variance accounted for (η^2)	p value
Model One – Total Leyton obsessional score			
	TS	10.3 %	< 0.001
	Female gender	0.2 %	0.579
	Increasing age	0.2 %	0.643
Model Two – Leyton obsessional interference score			
	TS	8.5 %	0.001
	Female gender	0.4 %	0.472
	Increasing age	0.1 %	0.727
Model Three – Child depression score (without Leyton)			
	TS	8.7 %	0.001
	Female gender	2.4 %	0.079
	Increasing age	3.3 %	0.037
Model Four – Child depression score (with Leyton)			
	TS	3.8 %	0.027
	Female gender	0.2 %	0.115
	Increasing age	0.2 %	0.114
	Total Leyton score	17.2 %	< 0.001

to ascertainment bias, in that children may be referred to the clinic because of complicating psychopathology such as depression or obsessionality. Our patients may be more severely affected than children with TS in the community undiagnosed and also the specialist nature of the clinic may mean that more severely affected individuals are referred. Secondly the study group is relatively small which will limit the precision of the parameter estimates. These characteristics of the study may limit the generalisability of the data generated. However given the rarity of TS (its prevalence being between 0.03 % [7] and 1.15 % [13]), the case control methodology used is more practicable than attempting to carry out a population-based community study. It is also the case that the TS clinic used to generate cases serves the country as a whole. No attempt was made to control for socio-economic class; this is unlikely to have influenced the data generated but is a potential limitation to the methodology used. These limitations notwithstanding, the gaps in the current evidence base mean that the data reported here may be a useful contribution to elucidating the nature and extent of psychopathology in children with TS and the care they need.

Phenomenology

These data suggest that there are similarities between the phenomenology of TS in childhood and that seen in adults with TS [20, 33]. The male to female ratio and the length of time between age at onset and age at diagnosis (4 years) in our study is similar to that previously reported [10, 15]. Just over 60 % of our group of children with TS had ADHD, twice the prevalence reported in some [10, 17] though not all [30] series in the USA. The differences in proportion may reflect a higher threshold for recognition and referral of TS by UK health services compared with those in the USA. The rates in this study fall within the range reported in an international multisite study which found the mean comorbidity rate for all sites to be 60 % (range 33 % to 91 %) [12]. Self-injurious behaviours occurred in 40 % of our cases, which is similar to the 30 % found in adult cohorts [26]. A high rate of self-injurious behaviour has now been documented in a wide variety of TS individuals including members of a large pedigree which included children and individuals who had not been referred to a clinic and who had mild TS [25].

Depressive symptoms

In accordance with other studies, our data suggest that children with TS have more depressive symptoms and obsessionality than other school age children. The multivariate statistical modelling suggests that the association between TS and depression scores is independent of the important association between TS and OCS. The mean DSRS scores of our TS cases (8.2) were higher than those reported for "normal" school children (4.3) [5]. this is a statistically significant difference calculating the difference between the means (3.9; 95 % CI 1.6 to 6.1). The scores of our TS cohort are similar to those reported for "maladjusted" school children and non-depressed clinic attenders, but lower than those for children with formal depressive disorder attending clinics [5, 6]. Our data therefore suggest that children with TS may have depression scores that are higher than those in other school age children but lower than those found in children with depressive disorder.

This excess of depressive symptoms is of interest. If the association between TS and depressive symptoms is not mediated genetically [18], then social and environmental factors may be of aetiological importance. While our findings may be complicated by the ascertainment bias implicit in clinic referrals, with children referred because of complicating psychopathology, the observed association has plausibility. Children with TS have vocal and motor tics and these involuntary movements and noises in a school setting might amount to a chronic stigmatising disorder, which for example could isolate the individuals or make them a subject for bullying and which could lead to depression [28].

Obsessional symptoms

A high proportion of the cohort had obsessional symptoms as assessed by the NHIS, which is similar to the findings of studies of adults [22, 23] and young people [9, 17] with TS. In the controlled element of the study, total LOI scores and interference scores were higher for the TS group than the control group. However, the mean scores in the TS group were lower than those reported in children with obsessive compulsive disorder [32]. This is consistent with the suggestion that the OCS seen in TS are different from, and often not as severe as those seen in formal obsessive compulsive disorder [21].

Conclusions

The results from this study confirm high levels of psychopathology in children with TS, including ADHD, OCS and depressive symptoms. These data indicate the potentially complex and challenging combination of difficulties encountered by young people with TS and those that care for them.

References

1. American Psychiatric Association (1987) Diagnostic and Statistical Manual of Mental Disorders (3rd edn revised), (DSM III-R). Washington DC: American Psychiatric Association
2. American Psychiatric Association (1994) Diagnostic and Statistical Manual of Mental Disorders (4th edn), (DSM IV). Washington DC: American Psychiatric Association
3. Berg CJ, Rapoport JL, Flament M (1986) The Leyton Obsessional Inventory – child version. Journal of the American Academy of Child Psychiatry 25:84–91
4. Berg CZ, Whitaker A, Davies M, Flament M, Rapoport JL (1988) The survey form of the Leyton Obsessional Inventory – child version: norms from an epidemiological study. Journal of the American Academy of Child and Adolescent Psychiatry 27:759–763
5. Birleson P (1981) The validity of depressive disorder in childhood and the development of a rating scale: a research report. Journal of Child Psychology and Psychiatry 22:73–88
6. Birleson P, Hudson I, Buchanan DG, Wolff S (1987) Clinical evaluation of a self-rating scale for depressive disorder in childhood (Depression Self-Rating Scale). Journal of Child Psychology and Psychiatry 28:43–60
7. Caine ED, McBride MC, Chiverton P, Bamford KA, Rediess S, Shiao J (1988) Tourette's syndrome in Monroe County school children. Neurology 38:472–475
8. Carter AS, O'Donnell DA, Schultz RT, Scahill L, Leckman JF, Pauls DL (2000) Social and emotional adjustment in children affected with Gilles de la Tourette's Syndrome. Journal of Child Psychology and Psychiatry 41:215–223
9. De Groot CM, Janus MD, Bornstein RA (1995) Clinical predictors of psychopathology in children and adolescents with Tourette Syndrome. Journal of Psychiatric Research 29:59–70
10. Erenberg G, Cruse RP, Rothner AD (1986) Tourette syndrome: an analysis of 200 pediatric and adolescent cases. Cleveland Clinic Quarterly 53:127–131
11. Ferrari M, Matthews WS, Barabas G (1984) Children with Tourette syndrome; results of psychological tests given prior to drug treatment. Developmental and Behavioral Pediatrics 5: 116–119
12. Freeman RD, Fast DK, Burd L, Kerbeshian J, Robertson MM, Sandor P (2000) An international perspective on Tourette syndrome: selected findings from 3,500 individuals in 22 countries. Developmental Medicine and Child Neurology 42:436–447
13. Kadesjo B, Gillberg C (2000) Tourette's Disorder: epidemiology and comorbidity in primary school children. Journal of the American Academy of Child and Adolescent Psychiatry 39:548–555
14. Kovacs M (1985) The Children's Depression Inventory (CDI). Psychopharmacology 21:995–1000
15. Leckman JF, Zhang H, Vitale A, Lahnin F, Lynch K, Bondi C, Young-Shin K, Peterson BS (1998) Course of tic severity in Tourette syndrome: the first two decades. Pediatrics 102:14–19
16. Leonard HL, Lenane MC, Swedo SE, Rettew DC, Gershon ES, Rapoport JL (1992) Tics and Tourette's disorder: a 2–7 year follow-up of 54 obsessive-compulsive children. American Journal of Psychiatry 149:1244–1251
17. Park S, Como PG, Cui L, Kurlan R (1993) The early course of the Tourette's syndrome clinical spectrum. Neurology 43: 1712–1715
18. Pauls DL, Leckman JF, Cohen DJ (1994) Evidence against a genetic relationship between Tourette's syndrome and anxiety, depression, panic and phobic disorders. British Journal of Psychiatry 164: 215–221
19. Robertson MM (1989) The Gilles de la Tourette syndrome: the current status. British Journal of Psychiatry 154: 147–169
20. Robertson MM (1994) Annotation: Gilles de la Tourette syndrome – an update. Journal of Child Psychology and Psychiatry 35:597–611
21. Robertson MM (1995) The relationship between Gilles de la Tourette syndrome and obsessive compulsive disorder. Journal of Serotonin Research 1(Suppl 1):49–62
22. Robertson MM, Channon S, Baker J, Flynn D (1993) The psychopathology of Gilles de la Tourette Syndrome: a controlled study. British Journal of Psychiatry 162:114–117
23. Robertson MM, Trimble MR, Lees AJ (1988) The psychopathology of the Gilles de la Tourette syndrome: a phenomenological analysis. British Journal of Psychiatry 152:383–390
24. Robertson MM, Eapen V (1996) The National Hospital Interview Schedule for the assessment of Gilles de la Tourette syndrome. International Journal of Methods in Psychiatric Research 6:203–226
25. Robertson MM, Gourdie A (1990) Familial Tourette's syndrome in a large British pedigree: associated psychopathology, severity of Tourette syndrome and potential for linkage analysis. British Journal of Psychiatry 156:515–521
26. Robertson MM, Trimble MR, Lees AJ (1989) Self-injurious behaviour and the Gilles de la Tourette syndrome: a clinical study and review of the literature. Psychological Medicine 19:611–625
27. Rosenberg LA, Brown J, Singer HS (1995) Behavioral problems and severity of tics. Journal of Clinical Psychology 51:760–767
28. Salmon G, James A, Smith DM (1998) Bullying in schools: self reported anxiety, depression, and self esteem in secondary school children. British Medical Journal 317:924–925
29. Spencer T, Biederman J, Harding M, Wilens T, Faraone S (1995) The relationship between tic disorders and Tourette's syndrome Revisited. Journal of the American Academy of Child and Adolescent Psychiatry 34:1133–1139
30. Spencer T, Biederman J, Harding M, O'-Donnell D, Wilens T, Faraone S, Coffey B, Geller D (1998) Disentangling the overlap between Tourette's disorder and ADHD. Journal of Child Psychology and Psychiatry 39:1037–1044
31. Wand RR, Matazow GS, Shady GA, Furer P, Staley D (1993) Tourette syndrome: associated symptoms and most disabling features. Neuroscience and Biobehavioral Reviews 17:271–275
32. Whitaker A, Johnson J, Shaffer D, Rapoport JL, Kalikow K, Walsh BT, Davies M, Braiman S, Dolinsky A (1990) Uncommon troubles in young people: prevalence estimates of selected psychiatric disorders in a nonreferred adolescent population. Archives of General Psychiatry 47:487–496
33. Wilson RS, Garron DC, Tanner CM, Klawans HL (1982) Behavior disturbance in children with Tourette syndrome. In: Friedhoff AJ, Chase TN (eds) Gilles de la Tourette syndrome. New York: Raven Press 35:329–333
34. Wodrich DL, Benjamin E, Lachar D (1997) Tourette's syndrome and psychopathology in a child psychiatry setting. Journal of the American Academy of Child and Adolescent Psychiatry 36:1618–1624
35. World Health Organisation (1992) International Classification of Diseases and Health Related Problems – Tenth Revision. Geneva: WHO