Brief Report

Obsessive-Compulsive Symptoms Among Patients with Sydenham Chorea

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Background: *Among patients with tic disorders, a distinctive clinical profile of obsessive-compulsive symptomatology has been described. The present investigation was designed to document the phenomenology of obsessive-compulsive symptoms (OCS) among patients with Sydenham chorea (SC), the neurologic variant of rheumatic fever. We hypothesized that OCS occurring in association with SC would be similar to those among patients with tic disorders.*

Methods: *The authors studied the presence of OCS in 73 patients with SC by using the Yale-Brown Obsessive-Compulsive Scale at the Pediatric Clinics of the University of São Paulo Medical Center in São Paulo, Brazil (*n *= 45) and at the National Institute of Mental Health in Bethesda, Maryland (*n *= 28).*

Results: *The most frequent symptoms observed among subjects with comorbid SC and OCS were aggressive, contamination, and somatic obsessions and checking, cleaning, and repeating compulsions. A principal component factor analysis yielded a five-factor solution (accounting for 64.5% of the total variance), with contamination and symmetry obsessions and cleaning compulsions loading highly.*

Conclusions: *The symptoms observed among the SC patients were different from those reported by patients with tic disorders but were similar to those previously noted among samples of pediatric patients with primary obsessive-compulsive disorder.*

Key Words: Sydenham chorea, rheumatic fever, obsessive-compulsive symptoms, obsessive-compulsive disorder, tic disorders, factor analysis

Obsessive-compulsive disorder (OCD) is a neurobiological disorder with heterogeneous symptomatology. Although the symptoms of OCD are diverse, they fall into several general categories that are consistent across culturally and geographically diverse populations (Honjo et al 1989; Thomsen and Mikkelsen 1991). Patients today present with symptoms similar to those first described by Janet in 1903 (Janet 1903). The similarity of the symptoms over time and across different cultures implies a neurobiological basis for OCD, and a growing body of evidence suggests that obsessive-compulsive symptoms (OCS) result from dysfunction of the orbito–frontal–striatothalamocortical circuitry. Neuroimaging studies have demonstrated structural and functional abnormalities of the caudate nucleus (Giedd et al 1996, 2000; Insel 1992; Luxenberg et al 1988; Rauch 2000).

Obsessive-compulsive disorder symptoms can also occur in disorders of the basal ganglia that are associated with abnormal movements. For example, OCD has been observed in patients with Sydenham chorea (SC), a neurologic variant of rheumatic fever (Swedo et al 1989a). In prospective studies of SC, up to 70% of patients acutely develop obsessions and compulsions associated with the choreic movements (Asbahr et al 1998; Swedo 1993). Recently, Swedo et al (1993, 1994) have suggested that SC might be a medical model for OCD, although, for this to be the case, the OCD symptoms in patients with SC should be similar to those found in patients who do not have SC. To date, no one has examined the OCD symptom profile in patients with SC. It is possible that OCD associated with movement disorders is distinct from the basic disorder (OCD without movement disorders). This has been suggested by studies of tic-related OCD symptoms. There is a particularly close association between tic disorders, including Tourette's syndrome, and OCD. Findings from studies of OCS among patients with tic disorders suggest that these patients have a unique symptom profile. Unlike symptoms in patients who have OCD with no abnormal movements, the predominant symptoms in tic-related OCD are aggressive or violent thoughts and images, symmetry concerns, and compulsive rituals involving blinking, touching, tapping, or rubbing (George et al 1993; Holzer et al 1994; Leckman et al 1994, 1995). The results of a recent study by Leckman et al (2003) suggested that familial factors contributed to the symptom dimension phenotypes among these patients and their first-degree relatives. The distinctive symptom profile, in combination with unique reports in gender ratio, age at onset, family history, and diverging responses to treatment, suggests that tic-related OCD might be a separate phenotype of the disorder.

The purpose of the present investigation was to describe the OCS experienced by patients with SC and to compare them with those seen in children with tic disorder. A comparison of these disorders is important because it might provide insights into whether SC is a medical model of OCD. We thus hypothesized that the obsessive-compulsive symptomatology observed in the SC group would be similar to that previously reported for patients with tic disorders.

Methods and Materials

Subjects

One hundred fourteen patients with SC were assessed at the Pediatric Clinics of the University of São Paulo Medical Center (USP) in São Paulo, Brazil, or at the National Institute of Mental Health (NIMH) in Bethesda, Maryland during the study period (USP, $n = 62$, 54.4% of the total sample; NIMH, $n = 52$, 45.6% of the total sample). Seventy-three (64%) of the 114 SC patients were eligible for the study. Subjects were selected according to the development of OCS among all patients with SC. For the screening of patients, the following instruments were used: the

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checklist of the Yale-Brown Obsessive-Compulsive Scale, child version (CYBOCS); the Schedule for Affective Disorders and Schizophrenia for School-Age Children (K-SADS); and the Diagnostic Interview for Children and Adolescents (DICA). Patients with a prior history of OCD or Tourette's syndrome were excluded; children with recent-onset OCS or tics were not excluded. The Ethics Committee for Research Projects of the University of São Paulo Medical School (USP site) and the National Institute of Mental Health (NIMH) Institutional Review Board (NIMH site) approved the study, and all subjects and their parents gave written informed assent and consent, respectively, for participation in this investigation.

Thirty-three of the 73 participants were boys (45.2%) and 40 were girls (54.8%). At the USP, 25 of the 45 subjects were boys (56%), and 20 were girls; whereas only 8 of the 28 subjects at the NIMH were boys (29%), and 20 were girls. This difference is statistically significant [$\chi^2(1) = 5.07$, $p = .02$]. The mean (±SD) age at onset of SC of the total sample was 9.98 ± 2.53 years. The mean ages at onset of SC of the NIMH and USP samples were 9.6 ± 2.2 years and 10.1 ± 2.8 years, respectively. No differences were found in the comparison of the mean age at onset of SC at the two sites [$t = 1.04$; $p = .30$] or in the mean age at onset of SC by gender in the combined sample [$t = 1.14$; $p = .26$]. Contrary to prior reports (Mercandante et al 1997), no motor or vocal tics were observed among patients at either site.

Clinical Measures

The CYBOCS was used to assess symptom content obtained through responses to the CYBOCS checklist. This instrument was chosen owing to its use in several previous reports, and it provides systematic information about more than 60 symptoms, divided into 15 separate categories of obsessions and compulsions (Scahill et al 1997).

The K-SADS (Chambers et al 1985) and the DICA (Welner et al 1987) were administered to ascertain psychiatric diagnoses.

Data Analysis

The frequency of specific obsessions and compulsions was calculated. Differences in frequency by diagnostic status (OCS or OCD), comorbid conditions (e.g., separation anxiety disorder), gender, age at onset, or site (USP or NIMH) were analyzed by χ^2 or Fisher exact tests, Student's t test, or Mann-Whitney rank–order test (U), depending on the type and distribution of the data.

A principal components factor analysis with varimax rotation was used to define the number and structure of symptom dimensions (factors or grouping of OCS). In keeping with studies previously published (Baer 1994; Leckman et al 1997; Mataix-Cols et al 1999), data from three categories—sexual obsessions (none of the patients had such symptoms), miscellaneous obsessions, and miscellaneous compulsions—were excluded from the initial factor analysis. A minimum eigenvalue of 1 was the criterion for the selection of the numbers of factors in the principal components analysis. Each of the factor solutions was rotated through the varimax rotation to maximize the variance explained by the factor, and summary results are presented.

Results

Of the 73 patients, 28 (38.4%) met DSM-IV diagnostic severity criteria for OCD (15 from USP and 13 from NIMH) (American Psychiatric Association 1994). No differences were observed between the two sites in the number of patients meeting criteria for OCD [$\chi^2(1) = 1.252$, $p = .263$], their gender distribution [$\chi^2(1) = 1.652$, $p = .199$], or age at SC onset [$t(68) = 1.728$, $p = .089$].

Table 1 shows the frequency of symptoms reported at the NIMH and USP centers and in the total sample. All 12 of the symptom categories were compared by gender, center, age at onset, and the presence of the diagnosis of OCD. There was a trend toward female patients reporting more hoarding obsessions than male patients ($U = 720$, $p = .053$); no other male/female differences were observed. No correlation was observed between the mean age at onset and reported rates of specific OCS. Symptoms such as touching, blinking, rubbing, and staring compulsions (included under the miscellaneous compulsions category) were reported by three of the 73 SC patients (4.1%).

A comparison between the two centers found that the USP site had more children reporting aggressive obsessions than the NIMH ($U = 451.5$, $p = .021$), whereas the NIMH had more children reporting contamination ($U = 839$, $p = .004$) and religious ($U = 720$, $p = .009$) obsessions and repeating ($U = 811$, $p = .013$) and hoarding ($U = 802$, $p = .010$) compulsions.

Patients who met diagnostic criteria for OCD had significantly more of the following symptoms than patients with OCS: contamination [$\chi^2 = 7.533$, $p = .006$], religious ($p = .019$), aggressive [$\chi^2 = 5.505$, $p = .019$], and somatic [$\chi^2 = 4.401$, $p = .036$] obsessions; and cleaning [$\chi^2 = 8.851$, $p = .003$] and repeating compulsions [$\chi^2 = 6.386$, $p = .012$]. Regarding checking compulsions, a tendency to statistical difference was observed [$\chi^2 = 3.802$, $p = .051$].

In reference to comorbid conditions, none of the subjects had motor or vocal tics at the time of their SC diagnosis. It is noteworthy, though, that 28 of the 73 patients (38.35%; USP, $n = 20$; NIMH, $n = 8$) developed symptoms of separation anxiety along with OCS or OCD. Of these 28 patients, 15 (54% [USP, $n = 11$; NIMH, $n = 4$]) met diagnostic criteria for separation anxiety disorder (SAD). No gender-related differences were observed between subjects with or without separation anxiety symptoms [$\chi^2(1) = .422$,$p = .516$] or diagnosis of SAD [gender: $\chi^2(1) = 1.668$,$p = .196$], nor were there significant between-group differences in the age at SC onset [$t(68) = .871$, $p = .387$]. Of the 28 individuals with symptoms of SAD, 21 (75% [USP, $n = 15$; NIMH, $n = 6$], $p = .673$) concomitantly presented aggressive obsessions (including fear of harm coming to parents). Patients who developed symptoms of SAD had significantly more aggressive obses-

Table 1. Frequencies of the Major Symptom Categories of the CYBOCS Checklist Reported for Children with Sydenham Chorea

Symptom Category	USP ($n = 45$)	NIMH ($n = 28$)	USP + NIMH ($n = 73$)
Obsessions			
Aggressive	33 (73)	13 (46)	46 (63)
Contamination	9 (20)	16 (57)	25 (34)
Hoarding	3 (6.7)	0	3 (4.1)
Religious	0	4 (14)	4 (5.5)
Symmetry	3 (6.7)	1 (3.6)	4 (5.5)
Somatic	15 (33)	6 (21)	21 (29)
Compulsions			
Cleaning	17 (38)	14 (50)	31 (42)
Checking	22 (49)	17 (61)	39 (53)
Repeating	11 (24)	15 (54)	26 (36)
Counting	4 (8.9)	5 (18)	9 (12)
Ordering	10 (22)	8 (29)	18 (25)
Hoarding	7 (16)	12 (43)	19 (26)

Children were evaluated at the University of São Paulo (USP) or the National Institute of Mental Health (NIMH). Data are presented as n (%).
CYBOCS, Yale-Brown Obsessive-Compulsive Scale, child version.

sions [$\chi^2(1) = 4.717$, $p = .030$] and were more likely to meet diagnostic criteria for OCD [$\chi^2(1) = 4.447$, $p = .035$] than were those who did not develop this symptomatology.

The principal component factor analysis of the 12 symptom categories yielded a five-factor solution that accounted for 64.46% of the total variance. Contamination and symmetry obsessions and cleaning compulsions loaded highly (greater than .50) in the first factor, which accounted for 19.26% of the total variance of the sample. Factor 2, accounting for 13.25% of the total variance, reflected a symptom dimension that included hoarding obsessions and compulsions and ordering compulsions (loadings greater than .50). Factor 3 accounted for 12.38% of the total variance and included only compulsions (checking and repeating). Both categories had a loading greater than .50 in this factor. Accounting for 10.38% of the total variance, factor 4 included religious obsessions and counting compulsions, also with loadings greater than .50 in this factor. Finally, factor 5, which accounted for 9.19% of the total variance, included only obsessions, aggressive and somatic, both with robust loadings (greater than .65). The results, after a varimax rotation of the factors, are shown in Table 2.

Discussion

To our knowledge, this is the first study to describe the obsessive-compulsive symptom profile of patients with Sydenham chorea. Our findings are similar to those previously reported among children and adolescents with OCD, in which the two most commonly reported obsessions are aggressive thoughts and contamination fears (Hanna et al 2002; Swedo et al 1989b, 1998). Geller et al (2001) reported that 29 of the 46 children in their sample (63%) had aggressive/catastrophic obsessions and 24 (52%) had contamination concerns. Rettew et al (1992) found similar proportions of aggressive and contamination obsessions (56% and 60%, respectively) at baseline assessments of a sample of 79 children and adolescents followed longitudinally, but noted that the primary obsessions shifted over time. Two subsequent studies (Hanna 1995; Thomsen and Mikkelsen 1995) also noted changes in obsessional content as children matured. A recent report from Scahill et al (2003) found that the most common obsessions among children aged 11 years or younger were contamination fears and worries about harm, whereas adolescents were more likely to have religious concerns (Hodgson and Rachman 1977). Thus, the prevalence of aggressive and contamination obsessions among the SC patients might reflect the children's young age at onset and developmental immaturity, rather than represent a salient difference in phenotypes.

None of the SC patients in this sample had tics, nor did their OCS profile resemble those previously reported for patients with tic disorders. Simple compulsions, such as touching and tapping, and aggressive obsessions have been reported more frequently among patients with tics than among those without a tic disorder (George et al 1993; Holzer et al 1994; Leckman et al 1994, 1995; Zohar et al 1997). Touching, blinking, rubbing, and staring compulsions were reported by only three of the 73 SC patients (4.1%). Thus, it appears that the OC symptoms of SC are more similar to those of childhood-onset OCD, than are the OC symptoms seen in children with tic disorders. This might reflect a shared neuropathology between childhood-onset OCD and SC that differs from that of tic disorders; future studies can address this hypothesis through structural and functional methodologies.

Aggressive obsessions were reported by 46 SC patients (63%). Interestingly, children with symptoms of SAD were noted to have aggressive obsessions more frequently than children without separation anxiety. This association is not surprising, because the majority of aggressive obsessions involved harm coming to the child's parents and contributed to the diagnosis of both OCD and SAD. Furthermore, there was no observed association between the child's age or developmental stage and the frequency of SAD symptoms, as would have been expected if the SAD were primary; however, the separation anxiety symptoms began during the SC illness and seem to be an additional manifestation of the basal ganglia dysfunction.

To further investigate the symptomatology of the 73 children with SC in our sample, we used factor analysis, a methodology that has been used successfully in a number of investigations of OCS in adults (Alsobrook et al 1999; Baer 1994; Leckman et al 1997, 2001; Mataix-Cols et al 1999). Instead of categorizing individuals into mutually exclusive subgroups (obsessions vs. compulsions, impulsive vs. non-impulsive) as has been done in earlier phenomenologic investigations (Hodgson and Rachman 1977; Hoehn-Saric and Barksdale 1983; Khanna et al 1990), the choice of factor analysis aims to explore a structural definition of

Table 2. Varimax Rotated Factor Structure for the CYBOCS Checklist Category Scores

	Factor Loading[a]				
Symptom Category	Factor 1	Factor 2	Factor 3	Factor 4	Factor 5
Contamination Obsessions	.792[b]	0	0	0	0
Cleaning Compulsions	.707[b]	0	0	0	0
Symmetry Obsessions	.530[b]	0	0	−.400	−.299
Hoarding Obsessions	0	.760[b]	0	0	0
Hoarding Compulsions	0	.747[b]	0	0	0
Ordering Compulsions	.278	.548[b]	0	.302	−.299
Checking Compulsions	0	0	.854[b]	0	0
Repeating Compulsions	0	.296	.757[b]	0	0
Counting Compulsions	0	0	0	.827[b]	0
Religious Obsessions	.367	0	0	.565[b]	.393
Aggressive Obsessions	0	0	0	0	.756[b]
Somatic Obsessions	0	0	0	0	.688[b]
Percentage of Variance Explained	19.26	13.25	12.38	10.38	9.19

CYBOCS, Yale-Brown Obsessive-Compulsive Scale, child version.
[a]Loadings less than .250 and greater than zero were substituted by zero.
[b]Robust loadings (greater than .50).

the symptom dimensions within a sample of SC patients, identifying symptoms that appear frequently together in the same individual. In our study, this analysis was primarily exploratory and was performed to identify potentially meaningful symptoms relationship. Thus, any conclusion concerning associations of OCD symptoms is clearly limited. Although the replication of factor scores in different samples might differ, future investigations (with other samples of SC patients) are necessary to determine the nature of symptoms associations (and better establish the consistency of our results) or address causality. An additional limitation of the data analysis was the use of the CYBOCS symptom checklist. The responses to its screening questions are dichotomous (yes/no), which limits the descriptive utility of the instrument.

In summary, the primary OCS among patients with SC were different from those reported by patients with tic disorders but similar to those reported by patients with childhood-onset OCD. The descriptive nature of the data does not address the question of etiopathogenesis, but the results do suggest that the use of SC as a medical model for OCD (Swedo 1994) might have face validity.

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