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Coprolalia and copropraxia in patients with Gilles de la Tourette syndrome

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ABSTRACT

Background and purpose: Involuntary expression of socially unacceptable words (coprolalia) or gestures (copropraxia) is the best-known symptom of Gilles de Tourette syndrome (GTS) that contributes to the social impairment. The aim of the study was to assess the prevalence, age at onset and co-occurring symptoms of coprophenomena.

Materials and methods: One hundred and sixty-eight consecutive subjects with GTS including 94 adults and 74 children and aged between 4 and 54 years (mean: 18.0 ± 8.3) were studied. Demographic and clinical data were obtained from medical history and neurological examination.

Results: Coprolalia or copropraxia appeared in 44 patients. Both coprophenomena were present in 9 patients. Coprolalia occurred in 25.0% ($n = 42$) and copropraxia in 6.5% ($n = 11$) of patients. Mean age at onset was 12.2 ± 5.7 years (range: 4–33) for coprolalia and 12.4 ± 4.9 years (range: 7–24) for copropraxia. Coprolalia started 4.4 ± 3.7 years (range: 0–16) after the onset of disease; copropraxia started 6.1 ± 4.0 years (range: 1–12) after the onset of the disease. Coprolalia began in adulthood in six patients only, and copropraxia in one person. In six patients, coprolalia appeared in the first year of the disease. Copropraxia was never seen in the first year of the disease. Coprophenomena were more frequent in patients with comorbid mental disorders, behavioral problems and severe tics. Three quarters of patients reported significant influence of coprophenomena on daily living.

Conclusions: Coprophenomena affect one quarter of GTS patients, appear in the time when tics are most severe, and are positively associated with comorbidity and more severe form of disease. Coprophenomena may reflect more widespread dysfunction of brain in GTS.

1. Introduction

Gilles de la Tourette syndrome (GTS) is a movement disorder characterized by multiple motor and at least one vocal tic which have been present for more than a year with the age at onset under 18 years [1]. This childhood-onset movement disorder is commonly associated with mental and behavioral comorbidities. GTS is not uncommon, with the prevalence rate about 1% all over the world in children aged 5–18 years [2].

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Coprophenomena encompass coprolalia, complex vocal tic, and copropraxia, complex motor tic. Coprolalia is defined as an involuntary utterance of obscene words or socially inappropriate and derogatory remarks. It is usually expressed out of social or emotional context and may be spoken in a louder tone or different cadence than normal conversation. Copropraxia is involuntarily performing obscene gestures often having a vulgar and insulting content. The prevalence rates vary from 8.5% to 50% for coprolalia [3,4] and from 5.7% to 25% for copropraxia among patients with GTS [5–7]. Coprophenomena are not unique to tic disorders. They may occur after stroke, encephalitis, in other neurological conditions such as choreoacanthocytosis, epilepsy, frontotemporal dementia, obsessive–compulsive disorder without tics, and Lesch–Nyhan syndrome [8–10].

Although coprolalia and copropraxia are two of the most spectacular features of GTS, the research on coprophenomena in Polish patients with GTS is very limited. The present study is an attempt to identify the prevalence of both tics, their typical appearance in relation to tic onset, course into adult life and associations with comorbid mental disorders, behavioral symptoms and tic severity in Polish GTS cohort.

2. Materials and methods

A total of 168 consecutively examined patients with GTS were included into the study. All the subjects were personally interviewed by the author of the study (PJ) using a short questionnaire on demographic and clinical data. Diagnosis of GTS was made according to DSM-IV-TR criteria [1]. Diagnosis of mental disorders was established retrospectively according to earlier psychiatric examinations or at the time of examination according to DSM-IV criteria. Clinical data regarding coprolalia, copropraxia and behavioral problems were collected retrospectively at the time of examination. The study was designed as a one-time registration study and no new clinical data obtained on follow-up visits were included into analysis.

The number of mental disorders was calculated for each patient by assigning the value 1 to each existing disorder and adding the values together. Mean value was counted by dividing the sum of all comorbidities by the number of patients. Obsessive–compulsive behavior (OCB) was diagnosed if obsessive thoughts and activities did not significantly influence daily activity and lasted less than an hour per day. In contrast to mental disorders, behavioral problems did not have strictly established criteria of diagnosis according to available psychiatric classifications. The illness was identified on the grounds of patient's behavior and author's experience. Behavioral problem was diagnosed if the condition had a significant impact on patient's daily life activities. Behavioral problems included: anger control problems, sleep problems, sexually inappropriate behavior, self-injurious behavior, significant social skills problems. Anger control problems were defined as uncontrolled bursts of anger and aggression which appeared suddenly of minimal provocation, and manifested as physical or verbal aggression against other people. These symptoms result from the inability to tolerate minor frustration and delay of gratification. Sleep problems and sexually inappropriate behavior were diagnosed when the symptoms did not meet the criteria of severe disorders according to DSM-IV. Examples of sexually inappropriate behavior are: touching other people's genitals (parents, family members, strangers), frequent and overt masturbation, constant talking about matters concerning sex and forcing other people to listen. Social skills problems are difficulties in establishing and maintaining contacts with other people, lack of friends, wrong interpretation of others' cues and intensions, excessive suspiciousness toward other people and tendency to blame other people for one's failures. Self-injurious behavior included self-cutting, head banging, self-hitting, biting (cheeks, nails, lips), scar-scratching, etc. The presence of one self-injurious behavior enabled the diagnosis. The total and mean value of behavioral symptoms was counted the same way as in case of mental disorders.

The intensity of tics was defined as the maximal intensity ever experienced by the patient (not necessarily at the moment of examination). Thus, tics were determined retrospectively on the basis of a medical history. This definition prevented the disease from, e.g. being classified as mild in adults with mild tics at the moment of evaluation but with a history of severe tics during childhood. The intensity of tics was defined descriptively. Tics were divided into mild, moderate and severe. Mild tics were defined if they were not related to physical or mental discomfort, problems in relations with peers, less than expected academic achievements and the need to treat. Moderate tics generated only small and temporary restrictions in patients' daily life (e.g. few-day absence from school, difficulties with homework). Severe tics caused the inability to continue normal daily activities (e.g. repeating grades, losing work), a physical discomfort (e.g. neck pain caused by cervical tics), a significant deterioration of quality of life and a necessity of neuroleptic therapy.

The negative impact of coprophenomena on daily living was marked if the patient reported subjective and significant social impairment with regard to the family, himself/herself and school or work. No quantitative scales were used.

For the purpose of the study, certain groups had been distinguished. Those patients with coprophenomena were termed K+, whereas those without coprolalia and copropraxia as K−. Likewise, those patients having tics without mental disorders as the clinical manifestation of the disease were termed GTS− and those with comorbid mental disorders as GTS+.

Although GTS is a developmental disorder of childhood onset, adult persons prevailed among subjects were included into the study. Demographic data of children, adults and all GTS patients were presented in Table 1. Children were defined as age $\leq$17 years.

3. Statistical analysis

Obtained data were statistically analyzed with Excel and Statistica 8.0 software. Quantitative data were shown as the mean $\pm$ standard deviation and evaluated using the Mann–Whitney *U*-test. Qualitative data were compared using the χ^2 test. Significance level was set at $p = 0.05$.

Table 1 – Demographic data of children, adults and all Gilles de la Tourette syndrome (GTS) patients included into the study.

Characteristics	Children ($n = 74$)	Adults ($n = 94$)	All GTS patients ($n = 168$)
Age at evaluation [years]			
Mean ± SD (range)	12.1 ± 3.3 (3.8–17.9)	25.4 ± 6.6 (18.1–53.7)	18.0 ± 8.3 (3.8–53.7)
Disease duration [years]			
Mean ± SD (range)	5.4 ± 3.1 (1–13.2)	16.6 ± 6.8 (1–43.7)	10.2 ± 7.4 (1–43.7)
Gender			
Males/females (*n*)	86/8	59/15	145/23
Age at tic onset [years]			
Mean ± SD (range)	6.7 ± 3.1 (2–16)	8.2 ± 3.6 (3–18)	7.3 ± 3.4 (2–18)
Intensity of tics			
Severe	13.5% ($n = 10$)	22.3% ($n = 21$)	18.5% ($n = 31$)
Moderate	56.8% ($n = 42$)	61.7% ($n = 58$)	59.5% ($n = 100$)
Mild	29.7% ($n = 22$)	16.0% ($n = 15$)	22.0% ($n = 37$)

SD – standard deviation.

4. Results

Coprolalia occurred in 25.0% (42/168) and copropraxia in 6.5% (11/168) of patients. Both coprophenomena were present in nine patients. At least one of two coprophenomena (coprolalia or copropraxia) was present in 44 patients.

Age at onset of coprolalia and copropraxia was not known in two and one patient, respectively. Mean age at onset was 12.2 ± 5.7 years (range: 4–33) for coprolalia and 12.4 ± 4.9 years (range: 7–24) for copropraxia. The age at the onset of coprophenomena is presented in Fig. 1. Coprolalia started 4.4 ± 3.7 years (range: 0–16) and copropraxia appeared 6.1 ± 4.0 years (range: 1–12) after the tic onset. Only in 15% (6/40) of patients, coprolalia was the early symptom of GTS which means that it appeared in the first year of the disease. Copropraxia was never seen at that stage of GTS. Majority of patients developed coprolalia (85%, 34/40) and copropraxia (90%, 9/10) before the age of 18.

Comorbid mental disorders occurred in 75% (126/168) of patients (GTS+ group); GTS− group consisted of 25% (42/168) of cases. Coprophenomena occurred considerably more often in GTS+ group compared to GTS− group and were associated with more frequent appearance of mental disorders and behavioral problems (Table 2). Tics rated as severe were reported more frequently in K+ group but the difference reached only borderline statistical significance ($p = 0.068$). Characteristics that differed K+ and K− groups were shown in Table 2. K+ and K− groups did not differ with regard to sex: 79.5% (35/44) vs. 88.7% (110/124) of males, respectively ($p = 0.1287$). We analyzed also data reported by children and adults separately. These findings are shown in Table 3. Positive family history did not differentiate K+ and K− groups ($p = 0.42$).

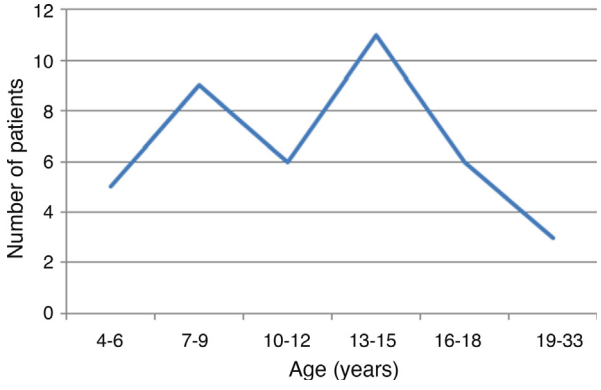

Fig. 1 – The age at the onset of coprophenomena in patients with Gilles de la Tourette syndrome.

Coprolalia was continuing in 61.9% (26/42) of patients at the time of evaluation, and copropraxia in 81.8% (9/11). In the remaining patients, coprolalia ($n = 16$) and copropraxia ($n = 2$) disappeared entirely before the clinical evaluation. Duration of discontinuous and persistent coprolalia did not differ (3.4 ± 4.8, range: 0–18 vs. 4.2 ± 5.0, range: 0–21 years; $p = 0.4332$). Duration of discontinuous and persistent copropraxia was similar as well (1.25 ± 1.1, range: 0.5–2.0 vs. 1.9 ± 3.2, range: 0–8 years; $p = 0.4862$).

The course of coprolalia and copropraxia did not change over time in more than half of the patients (57.1%, 24/42 and 63.6%, 7/11, respectively). Minority of patients developed intermittent (come-go-come again) or self-limited (come and go) course of coprophenomena.

The information about the treatment was not available from one patient with coprolalia, and one person with both coprophenomena and six cases without coprophenomena. Eighty-one percent of patients (34/42) in K+ group, and 68.6% (81/118) cases in K− group were ever treated with tic reducing medications ($p = 0.13$). The mean number of all tic-reducing medications ($p = 0.22$), dopamine receptor blocking drugs ($p = 0.82$), benzodiazepines ($p = 0.06$) and clonidine ($p = 0.47$) used in K+ patients was similar compared to K− group. All tic-reducing medications were analyzed retrospectively, including drugs used before the onset of coprophenomena and after coprophenomena had ceased. All used tic-reducing medications in GTS patients are listed in Table 4.

Seventy-five percent of all patients with coprophenomena ($n = 44$) reported significant influence of coprolalia and copropraxia on daily living. These tics affected the family (25/44), the patient himself/herself (17/44), school or work (15/44). Only a quarter of patients reported coprophenomena as being not troublesome in their lives.

Table 2 – Characteristics of Gilles de la Tourette syndrome (GTS) patients with and without coprophenomena.[a]

	Patients with GTS (n = 168)		
	Patients with coprophenomena (n = 44)	Patients without coprophenomena (n = 124)	P-value
GTS without mental disorders (n = 42)	13.6% (6)	29.0% (36)	
GTS with mental disorders (n = 126)	86.4% (38)	70.1% (88)	0.0434
Mental disorders			
ADHD	54.5% (24)	35.5% (44)	0.0269
OCD/OCB	61.4% (27)	36.3% (45)	0.0039
Learning disorders	40.9% (18)	25.8% (32)	0.0598
Anxiety disorders	38.6% (17)	13.7% (17)	0.0004
Mood disorders	20.4% (9)	9.7% (12)	0.0641
Conduct disorder	20.4% (9)	4.0% (5)	0.0007
Enuresis	15.9% (7)	9.7% (12)	0.2636
Stuttering	9.1% (4)	10.5% (13)	0.9780
Trichotillomania	6.8% (3)	0% (0)	0.0231
Behavioral problems			
Anger control problems	52.3% (23)	16.1% (20)	0
Sleep problems	50.0% (22)	21.8% (27)	0.0004
Self-injurious behavior	38.6% (17)	8.9% (11)	0.00001
Sexually inappropriate behavior	18.2% (8)	4.8% (6)	0.0061
Social skills problems	29.5% (13)	4.0% (5)	0
Mean number ± SD of			
Mental disorders	2.7 ± 1.9	1.5 ± 1.3	0.0001
Behavioral problems	1.9 ± 1.4	0.6 ± 0.9	0
Intensity of tics			
Severe	31.8% (14)	18.6% (23)	0.0680
Moderate	56.8% (25)	60.5% (75)	0.6704
Mild	11.4% (5)	21.0% (26)	0.1595

ADHD – attention deficit with hyperactivity disorder; OCD/OCB – obsessive-compulsive disorder/obsessive-compulsive behavior; SD – standard deviation.

[a] Data shown as percentages (numbers) unless otherwise stated.

5. Discussion

The frequency of coprophenomena in Polish population with GTS was 25% for coprolalia and 6.5% for copropraxia. Our data are consistent with other studies, finding that most people with GTS never display these tics. Frequencies reported by other authors are quite variable: 8–50% for coprolalia [11,4] and 3–25% for copropraxia [6,7,12]. It is considered that their prevalence is much lower in non-selected samples. The rate of coprophenomena is higher in selected samples which consist of the patients who are referred to psychiatrist due to their mental disorders, neurologist due to predominant tics or pediatrician because they are children. Our findings are in the middle of the range for coprolalia and on the low side of range for copropraxia. Coprolalia in Polish cases is nearly four times as common as copropraxia that is the highest ratio compared to literature (range 1.12–3.23) [5,6].

Copropraxia is highly socially unacceptable symptom and some adult patients may not admit its presence. Coprolalia was much more likely than copropraxia to occur as the exclusive expression of coprophenomena. 'Isolated' copropraxia appeared very rarely.

Our results suggest that coprophenomena appear usually around the age of 12. This finding is consistent with other studies. The mean age at onset of coprolalia in our GTS sample was within the range of 8–15 years reported by other authors [5,11,13–16]. However, one-third of Polish patients developed coprophenomena in early childhood or late adulthood. We found that coprolalia could start as late as 33, and copropraxia – 24 years of age. Fifteen percent of patients developed coprolalia after the age of 18, and in another 15% coprolalia was present at time of tic onset. Copropraxia started in adulthood in 10% of patients and was never present at the beginning of the disease. Freeman et al. [5] in large multicenter study found that 1/10 of patients may developed coprolalia at the time of tic onset and in adult age as well, which is similar to our findings. However, they also found that copropraxia may be the initial sign of the disease that we did not see in Polish patients. The reason for that is probably small number of copropraxia cases in our series. In Polish patients, the delay between tic and coprophenomena onset was 4 years and 5 months for coprolalia and 6 years and 1 month for copropraxia. This finding is within the range of other studies (2.2–8.1 years) [5,11,13,15].

We confirm the results of other studies that coprophenomena appear in time when tics are at their peak severity. Tics tend to be most severe during the preadolescent period, between 10 and 12 years of age, and in most cases diminish during adolescence and adulthood [17,18]. We believe that tic severity is related to the presence of coprophenomena that force the patients to seek medical counseling. Four-fifths of

Table 3 – Characteristics of children and adult Gilles de la Tourette syndrome (GTS) patients with and without coprophenomena.

	GTS patients group					
	Children (n = 71)			Adults (n = 97)		
	Patients with coprophenomena (n = 21)	Patients without coprophenomena (n = 50)	P-value	Patients with coprophenomena (n = 23)	Patients without coprophenomena (n = 74)	P-value
GTS without mental disorders	14.3% (3)	34.0% (18)	0.0910	13.0% (3)	25.7% (20)	0.3278
GTS with mental disorders	85.7% (18)	66.0% (33)		87.0% (20)	74.3% (55)	
Mental disorders						
ADHD	52.4% (11)	40.0% (20)	0.3371	56.5% (13)	32.4% (24)	0.0378
OCD/OCB	57.1% (12)	24.0% (12)	0.0071	65.2% (15)	44.6% (33)	0.0840
Learning disorders	38.1% (8)	28.0% (14)	0.4045	43.5% (10)	24.3% (18)	0.0766
Anxiety disorders	38.1% (8)	6.0% (3)	0.0023	39.1% (9)	18.9% (14)	0.0477
Mood disorders	9.5% (2)	2.0% (1)	0.4284	30.4% (7)	14.9% (11)	0.0951
Conduct disorder	14.3% (3)	2.0% (1)	0.1375	26.1% (6)	5.4% (4)	0.0140
Enuresis	14.3% (3)	12.0% (6)	0.8993	17.4% (4)	8.11% (6)	0.3755
Stuttering	9.5% (2)	6.0% (3)	0.9829	8.7% (2)	13.5% (10)	0.8023
Trichotillomania	0% (0)	0% (0)		13.0% (3)	0% (0)	0.0136
Behavioral problems						
Anger control problems	47.6% (10)	8.0% (4)	0.0005	56.5% (13)	21.6% (16)	0.0014
Sleep problems	42.9% (9)	18.0% (9)	0.0291	56.5% (13)	24.3% (18)	0.0038
Self-injurious behavior	71.4% (6)	94.0% (3)	0.0265	47.8% (11)	10.8% (8)	0.0003
Sexually inappropriate behavior	28.8% (6)	2.0% (1)	0.0028	8.7% (2)	6.8% (5)	0.8828
Social skills problems	33.3% (7)	4.0% (2)	0.0027	26.1% (6)	4.1% (3)	0.0056
Mean number ± SD of						
Mental disorders	2.3 ± 1.8	1.2 ± 1.1	0.0080	3.0 ± 2.0	1.6 ± 1.4	0.0033
Behavioral problems	1.8 ± 1.5	0.4 ± 0.8	0.0002	2.0 ± 1.22	0.7 ± 0.9	0.0000
Intensity of tics						
Severe	23.8% (8)	10.0% (14)	0.24896	39.1% (9)	24.3% (18)	0.1664
Moderate	61.9% (12)	60.0% (12)	0.8809	52.2% (12)	60.8% (45)	0.4623
Mild	14.3% (11)	30.0% (20)	0.2756	8.7% (2)	14.9% (11)	0.6831

ADHD – attention deficit with hyperactivity disorder; OCD/OCB – obsessive–compulsive disorder/obsessive–compulsive behavior; SD – standard deviation

patients with coprophenomena had to be on symptomatic treatment, three quarters of them found these tics severely affecting their lives and two-thirds of cases had at least one of coprophenomena at the time of evaluation.

What is of special interest in our study is that course of coprophenomena may be unusual and different from typical course of tics. In most patients, severity of tics usually changes over time with waxing and waning pattern. In more than half of our patients, coprophenomena, that are kind of complex tics, were persistent and invariable over time, with the mean duration time of 4 years for coprolalia and 2 years for copropraxia which is rather unusual for tics. This may result from excessive proportion of adult persons in our series (56%) in whom tics tend to stabilize. However, our finding is very similar to that reported by Freeman et al. [5] who included into the study mainly children (85%). It cannot be excluded that persistent coprophenomena may eventually cease but long duration, 21 years for coprolalia and 8 years for copropraxia in two extreme cases, suggest that in substantial proportion of patients these tics will have invariable course. On the other hand, nearly all patients with coprophenomena were treated with tic-reducing medications and the agents may have suppressed these tics to decrease the number of persistent coprophenomena. In the remaining patients, nearly less than half of the total population, coprophenomena had a typical course for tics that changed over time. In these cases coprophenomena remitted, resumed, ceased or had intermittent course.

Tics rated as severe were found more often in patients with coprophenomena at borderline significance. However, tic-reducing medications in patients without coprophenomena were used as frequently as in K+ group during all disorder. Haloperidol, thioridazine and risperidone were three most popular medications in both K+ and K− groups (Table 4). Sulpiride and clonazepam were even more frequently used in patients without coprophenomena. These results confirm our previous findings that there is a tendency to treat the patients even with mild, not troublesome tics in our country [19].

We have found a very clear association of coprophenomena with comorbidity and, to a lesser degree, tic severity. The prevalence of mental disorders, behavioral symptoms and severe intensity of tics in patients with GTS was higher if coprophenomena occurred. The incidence of coprophenomena among GTS− group was very low. Although it is not known whether coprophenomena are the risk factor for some psychiatric disorders or rather inversely, the presence of coprophenomena contributes to more complex and more severe clinical phenotype of GTS. Additionally, there is some evidence from functional neuroanatomy studies that complex

Table 4 – Tic-reducing medications used in patients with and without coprophenomena (*n* – number of patients treated with particular medication; *N* – number of all treated patients in patients with or without coprophenomena; % – calculated as *n*:*N* × 100%).

	Patients with coprophenomena (*N* = 34)	Patients without coprophenomena (*N* = 79)
Neuroleptics		
Haloperidol	70.6% (24)	65.8% (52)
Thioridazine	20.6% (7)	26.6% (21)
Risperidone	32.4% (11)	27.9% (22)
Sulpiride	5.9% (2)	12.7% (10)
Pimozide	5.9% (2)	3.8% (3)
Tiapride	5.9% (2)	5.1% (4)
Chlorprothixene	0% (0)	1.3% (1)
Olanzapine	5.9% (2)	2.5% (2)
Perazine	2.9% (1)	0% (0)
Zuclopenthixol	0% (0)	2.5% (2)
Quetiapine	2.9% (1)	2.5% (2)
Chlorpromazine	0% (0)	1.3% (1)
Perfenazine	0% (0)	2.5% (2)
Trifluoperazine	0% (0)	1.3% (1)
Benzodiazepines		
Clonazepam	2.9% (1)	15.2% (12)
Clorazepate	2.9% (1)	2.5% (2)
Diazepam	2.9% (1)	3.8% (3)
Lorazepam	0% (0)	1.3% (1)
Bromazepam	0% (0)	2.5% (2)
Other medications		
Clonidine	14.7% (5)	22.8% (18)
Diltiazem	2.9% (1)	1.3% (1)
Botulinum toxin	0% (0)	1.3% (1)
Baclofen	0% (0)	1.3% (1)
Pergolide	0% (0)	1.3% (1)
Ropinirole	0% (0)	1.3% (1)
Topiramate	0% (0)	2.5% (2)

tics cause abnormal activity in many regions of brain. Two studies using positron emission tomography and functional magnetic resonance showed that complex tics, including coprolalia, involve widespread changes of activity in basal ganglia and cerebral cortex [20,21]. Thus, if coprophenomena are associated with more severe clinical phenotype and widespread changes of neural activity, their presence is probably related to more pronounced dysfunction of brain in GTS.

We are aware of important limitations of our study. There was substantial amount of adults included into the study. Data obtained from these patients were retrospective with the possibility of recall bias. This may be due to patients' forgetfulness or natural course of some diseases, e.g. tic severity and attention deficit with hyperactivity disorder (ADHD) symptoms decrease with age in contrast to OCD symptoms that increase with age [17,18]. However, in our cohort adult patients with coprophenomena reported ADHD more frequently and OCB/OCD less frequently compared to children with coprophenomena (Table 3). It suggests that recall bias has been unlikely to affect the results of our study significantly.

Methodology of collecting data was different in children and adults. Most clinical information regarding children are provided by their parents whereas adults report themselves. Despite it, the findings in both groups were very similar compared to the all GTS patients with regard to mean number of mental disorders and behavioral problems. There was also no difference between children and adults regarding the reporting of mental disorders except ADHD, OCB/OCD and conduct disorder (Table 3). In the latter, there were very small groups of patients that limited to make the conclusions. Moreover, all but one behavioral symptoms, sexually inappropriate behavior that is probably troublesome for adults to admit, were reported by children and adult patients in similar rate. In contrast to comorbidity, tic severity did not differ between children and adults (Table 3). However, tics rated as severe reached borderline statistical significance and were reported more often by all GTS patients with coprophenomena compared to those without coprolalia and copropraxia (Table 2). The number of patients and descriptive way of tic severity may have affected these findings. In conclusion, (1) the data regarding the particular mental disorder and behavioral symptom occurring in K+ and K− groups should be interpreted with caution due to small number of cases in most of these disorders and (2) different methodology of collecting data in children and adults may have influenced our results.

There is also possible referral bias because the patients were evaluated by neurologist and the cases with more severe psychopathology and behavioral problems (and probably high proportion of coprophenomena) were referred to psychiatric clinics. Most researchers found that only 8–12% of GTS patients had no other psychopathology [22,23]. In our series, 25% of patients did not have comorbid mental disorders (GTS− group). It might have been the reason for lower incidence of copropraxia in Polish patients with GTS. The data regarding the course of coprophenomena should be interpreted with caution due to retrospective and one-time registration study design. The assessment of natural course of coprophenomena would require prospective study.

6. Conclusions

1. Coprophenomena appear only in minority of GTS patients, typically around the time that tics are at their peak severity.
2. Coprolalia is nearly four times as common as copropraxia.
3. The course of coprophenomena may be unusual for tics in some patients because of their persistence and invariability.
4. Coprophenomena are more frequent in patients with comorbid mental disorders, behavioral problems and severe tics.
5. Coprophenomena may reflect more widespread dysfunction of brain in those patients with GTS.

Conflict of interest

None declared.

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None declared.

Ethics

The work described in this article has been carried out in accordance with The Code of Ethics of the World Medical Association (Declaration of Helsinki) for experiments involving humans; Uniform Requirements for manuscripts submitted to Biomedical journals.

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