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Functional tic-like behaviors in patients with Gilles de la Tourette syndrome

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Background:

It is widely believed that **functional tics** are a rare presentation of functional movement disorders (FMD). Hence, possible co-occurrence of both **tics** – due to Gilles de la Tourette syndrome (GTS) - and **functional tic-like behaviors** (FTB) – due to FMD - in one and the same patient is a heavily understudied clinical topic. This is the more surprising, since it is well known that patients may suffer from both a neurological disease and a functional neurological disorder (FND), for example **psychogenic nonepileptic seizures (PNES)** in patients with **epilepsy**. In GTS, up to now, only very few patients have been described suffering from **both "primary" tics and comorbid FTB**. With this study we want to raise awareness of comorbid FTB in patients with GTS, to present clinical characteristics of a large sample of patients suffering from both tics and FTB, and to give some advice, how to differentiate one from the other.

Methods:

Between 2002 and 2021, **in our specialized Tourette outpatient clinic at Hannover Medical School,** 71 patients were diagnosed with both GTS and comorbid FTB. We analyzed data of these patients retrospectively and compared the results with data obtained from a large GTS sample (n=1032) from the same center. Of note, in this sample we included no cases with recently described rapid onset FTB related use of social media (mass social media induced illness, MSMI).

Tab. 1: Comparison of groups GTS-FTB and GTS+FTB.

	TS	TS+FTB	p
Female n (%)	236 (23%)	27 (38%)	**0.003**
Age (mean, SD)	18.98 (12.97)	21.5 (12.7)	0.1
Tic onset (mean, SD)	7.17 (3.72)	6.3 (2.3)	0.05
OCD n (%)	103 (10%)	16 (23%)	**0.0009**
OCS n (%)	637 (67%)	41 (58%)	0.5
Anxiety n (%)	323 (31%)	19 (27%)	0.4

GTS – Gilles de la Tourette syndrome, FTB – functional tic-like behaviors, SD – standard deviation, OCD – obsessive-compulsive disorder, OCS – obsessive-compulsive symptoms

Results:

1. The GTS+FTB group comprises **significantly more females** compared to the GTS sample (Table 1).
2. Age at onset of comorbid FBT was found to be **on average 14 years** (mean age: 20.8 years, SD 11.8, range 9 and 47 years) after tic onset (Table 1).
3. Most typically onset of FTB was **abrupt** (n=38, 54%) enabling all these patients to precisely indicate the exact date of onset of FTB.
4. Based on patients' reports and clinical judgement, more than half of patients reported **a concrete triggering factor** (n=38, 54%).
5. Different from motor tics, in most cases, motor FTB presented with **complex movements** not following the typical rostro-caudal distribution of tics.
6. Vocal FTB were also mainly complex. Accordingly**, coprophenomena** were much more common in GTS+FTB compared to GTS (44% vs 28%).
7. Different from tics, FTB **did not show the typical waxing and waning course**.
8. While most patients reported stress being the strongest influencing factor for their tics, FBT was mainly influenced **by situations that usually have no major impact on tic severity** such as long period of sitting or unhealthy food. Remarkably, presence and severity of FTB strongly depended on contact with particular people.
9. **Self-injurious behaviors** such as self-hitting were more common in GTS+FTB than GTS (55% vs. 30%).
10. Due to misdiagnosis of FTB as tics, a remarkably high number of patients with diagnosed **"treatment-resistant"** (47%). In addition, a large number of patients with GTS+FTB reported unusual or even unknown **side effects** after use of typical anti-tic medications.

Table 2. Distinguishing features between severe GTS and GTS+FTB

Category	Severe TS*	TS+FTB**
Age of onset	<18 years	Before and after 18 years
Onset	1. Gradual onset 2. Simple tics mainly localized in the face	1. Abrupt onset, patients can often identify exact day of onset 2. Triggering factor/situation preceeding onset 3. Complex tics present at onset
Course	1. Waxing and waning 2. Rostro-caudal distribution	1. After abrupt onset the symptoms are progressing gradually 2. Not rostro-caudal distribution (n=57; 80%)
Sex distribution	Males predominate	Significantly more females than in TS group
Period of worst symptom severity	Between 10-12 years	24 +/- 13 years
Symptom awareness	Patients can often describe both past and present tics	Patients often have problems listing all their symptoms
Suppressibilty	Usually could be suppressed for a short period of time	Could be suppressed in 72% of patients
Factors influencing symptom remission/exacerbation	1. Stress usually provokes symptoms exacerbation 2. Common symptoms' reduction upon relaxation and concentration	Symptom remission and exacerbation provoked by diverse and sometimes unusual factors (ex. presence of a particular person)
Main impairing factors	Complex tics and OCD	FTB
Comorbidities	OCS, OCD, ADHD are the most frequent ones	1. Distribution of comorbidities similar to TS 2. OCD less frequent than in TS
SIB	1. Found in around 39% of patients 2. Secondary to tics and/or comorbidities	1. Found in 56% of patients 2. Seems to occur independent of FTB and comorbidities
Coprophenomena	Found in around 30% of patients Reported by the patients and caregivers, but are not seen during clinical examination	1. Found in 44% of patients 2. Often seen during clinical examination
Secondary gain	Rare	Common
Reaction to anti-tic medication	Treatment-resistance in limited number of patients	1. 28% of patients experienced atypical side effects 2. 15% of patients experienced atypical reaction to medication 3. In 48% of patients the treatment was not effective or suddenly no longer effective

GTS – Gilles de la Tourette syndrome, FTB – functional tic-like behaviors, OCD – obsessive-compulsive disorder, OCS – obsessive-compulsive symptoms, ADHD – attention deficit hyperactivity disorder, SIB – self-injurious behaviors; * based on Sambrani et al. (25), ** based on sample of 71 patients presented in this study

Conclusions:

1. **The additional diagnosis of FTB in patients with GTS** should be taken into consideration, when mainly **complex motor** (including self-hitting) and **vocal tic-like behaviors** occur, which so far were not present in the course of the disease, with **abrupt onset** several years after tic onset characterized by completely different influential factors and response to treatment compared to preexisting tics (Table 2).
2. Based on our sample, it can be assumed **that FTB is common in patients with GTS**.
3. The diagnosis of FTB should not be missed to avoid inappropriate treatments.