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"I swear it is Tourette's!": On functional coprolalia and other tic-like vocalizations

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ABSTRACT

Coprolalia in neuropsychiatry is typically associated with tic disorders, in particular Gilles de la Tourette syndrome. To date, there has been no report of functional coprolalia. Here, we provide the clinical characteristics of 13 adolescent and adult patients with coprolalic and other functional tic-like complex vocalizations who, on the basis of these symptoms, were misdiagnosed with a primary tic disorder, most commonly Gilles de la Tourette syndrome. We describe similarities and highlight the differences from primary tic disorders in order to provide a pragmatic list of clinical clues that will facilitate correct diagnostic labeling and thereby treatment. Finally, we emphasize that the distinction between a primary and a functional tic disorder should rely on a combination of neuropsychiatric symptoms and signs and not on the presence of single, however striking, abnormal behaviors, such as coprolalia.

1. Introduction

Involuntary vocalizations are a well-recognised feature of tic disorders. Indeed, coprolalia, the occurrence of obscene and socially inappropriate vocalizations without intent is seen as almost pathognomonic of Gilles de la Tourette syndrome (GTS). Coprolalia has significant notoriety amongst the public, even though it is a fairly uncommon feature of GTS (lifetime prevalence of less than 20% (Freeman et al., 2009)). Coprolalia in GTS is independently associated with poor quality of life, tic severity, as well as a range of further neuropsychiatric problems, such as increased anxiety, sexually inappropriate and also non-obscene socially inappropriate behaviors (NOSI), obsessive-compulsive and attention-deficit hyperactivity disorder (Eapen et al., 2016; Eddy and Cavanna, 2013a, 2013b; Freeman et al., 2009; Kobierska et al., 2014).

However, repetitive involuntary vocalizations, including coprolalia, are not exclusively encountered in primary tic disorders. Coprolalia has been reported in patients with structural brain lesions and in patients with neurodegenerative and autoimmune disorders (Singer, 1997). Coprolalia, alongside other complex vocal tic behaviors, such as palilalia, echolalia and klazomania (compulsive shouting) has been documented in patients with post-encephalitic neuropsychiatric syndromes (Lees, 1985). Coprolalia has also been described as an ictal or post-ictal phenomenon (Panunzi et al., 2013). Although the functional neuroanatomical basis of coprolalia remains unclear, the limbic circuitry has been suggested to play an important role in the pathophysiology of coprolalic behaviors (Van Lancker and Cummings, 1999).

Here we report 13 patients who presented with prominent vocalizations including coprolalia, palilalia and echolalia, some with additional movement disorders, but where specific features of clinical history and examination are in our view not compatible with the diagnosis of GTS, and where investigations did not reveal a secondary cause. We propose that the diagnosis of these patients is most likely a functional neurological disorder, and we discuss the problems and potential benefits of making this diagnosis in people with tic-like vocalizations and movements.

2. Methods

All patients presented at the GTS referral clinic of one of the authors (KMV; Clinic of Psychiatry, Socialpsychiatry and Psychotherapy, Hannover Medical School) during the period of 1995–2015. Among those, patients with predominant complex vocalizations, such as coprolalia and/or other prominent vocal tic-like behaviors (i.e. palilalia/echolalia/NOSI) were selected and their clinical characteristics were extracted. We selected 13 patients who in our view

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Table 1
Clinical characteristics of patients with functional coprolalia and other functional complex tic-like vocalizations. GTS=Gilles de la Tourette syndrome; NOSI=Non-obscene socially inappropriate behaviors; ADHD = Attention-deficit hyperactivity disorder; OCD = Obsessive-compulsive disorder.

Case Number	Age/ Sex	Age at onset of functional tic-like symptoms	Modality of onset/ Precipitants	Symptoms at onset	Simple vocalizations on presentation	Complex vocalizations on presentation		Blocking phenomena	Other movements on presentation	Urge	Suggestibility/ Distractibility/ Suppressibility	Stereotyped nature/ Fluctuations	Other functional symptoms	Previous Treatments/ Improvement	Previous Diagnosis
						Coprolalia	Echolalia/ Palilalia/NOSI								
1	42/ M	33	Abrupt/Mild accident	Episodic hand tremor	“Eh”	“Ficken” “Kacken” “Kackeficken”	Palilalia: Nonsense words (“tiff”, “taff”, “piff”)	No	Blinking, Head banging, Hitting head with hand, Episodic hand tremor	Yes	No/Yes/Yes	Yes/Yes	Urge to hit himself against objects, functional hyposmia, episodic whole-body shivering	None	GTS
2	10/ M	5	Abrupt/ School mobbing	Stuttering	Snuffling, coughing, engine noise, moaning	“Arsch”, “Hurensohn” “Vollidiot”, “fick dich”	Palilalia: “Hilfe” (up to 15 fold)	Speech blocks	Multifocal jerky movements	No	Yes/Yes/No	Yes/Yes	Episodic double vision	None	GTS
3	35/ M	26	Abrupt/ Change of work	Snuffing, blinking	None	"ich will dich in den Arsch ficken", "du bist hässlich", "ich kann dich nicht leiden"	Echolalia while watching TV	Speech blocks	Rapid multifocal jerking, urge to exert pressure on arteries and genitals	Yes	No/Yes/Yes	Yes/Yes	Urge to look into sun, dizzy spells, functional hypaesthesia	Botulinum toxin in vocal cords/ Improvement due to hoarseness; Aripiprazole, Quetiapine, Fluoxetine/No improvement	GTS
4	17/F	14	Abrupt/ Familial conflict	Repetition of a single word (“Korb”)	Screaming, syllables ("ja", "he", "paha", "lalalalala", "eh", "äh", "ehm")	"fick dich", “du Hurentochter", "du abgefickte Schlampe", "Hure", "Fotze", Dreckschwein", "Hurensohn", "fick dich", "Scheiße", "Schlange", Nutte", "ich liebe dich", "ich will ein Kind von dir”	Immediate ambient echolalia; Palilalia: "meine", "ok", "man", "Kevin", "ich bin fertig damit", "leider nein" ("Bolle" (nonsense word))	Slurring	Jerky limb movements, pulling on the curtain (only when listening to songs), pinching others, knocking against the wall, peace sign, showing the middle finger (only in the presence of others)	Yes	Yes/Yes/Yes	Yes/Yes	None	Risperidone, Fluvoxamine/ None	GTS plus ADHD
5	19/ M	13	Abrupt, while watching TV	Sticking out his tongue Noises similar to gagging	Gagging, groaning	Schwein, Sau, Arschloch, Hure, Ficker, Fotze, Pisskopf, Schwuchtel, Hurensohn, Ficklippe,	No	No	Bizarre and complex whole-body movements with twisting and backward arching of trunk	Yes	Yes/Yes/Yes	Yes/ Yes	None	Sulpiride, Lorazepam, Clonazepam, Chlorprothixene, Methylphenidate, Aripiprazole – all without effect, but adverse effects (e. g. not able to speak under treatment with Aripiprazole) Symptom-free when smoking	GTS plus ADHD

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Table 1 *(continued)*

Case Number	Age/ Sex	Age at onset of functional tic-like symptoms	Modality of onset/ Precipitants	Symptoms at onset	Simple vocalizations on presentation	Complex vocalizations on presentation		Blocking phenomena	Other movements on presentation	Urge	Suggestibility/ Distractibility/ Suppressibility	Stereotyped nature/ Fluctuations	Other functional symptoms	Previous Treatments/ Improvement	Previous Diagnosis
						Coprolalia	Echolalia/ Palilalia/NOSI								
														cannabis.	
6	19/F	16	Abrupt, during inpatient admission in adolescent psychiatry due to depression, panic attacks, and self-injurious behavior .	Head jerking	"hm", ba, ne, miaowing, he, ja, eo, bababa,	Fotze. "Heil Hitler du Fotze", "Ich hab dich nicht beleidigt du Fickfotze", "Bullshit"	Echolalia: "Katzengulasch" (upon meeting patient No 8) "Policracker" "so viel Sand und keine Förmchen" Palilalia: "Interessiert keinen", "Guck mal du Frettchen"	No	Copropraxia: Middle finger sign (she can say exactly that this occurs once a week) Hitting her hand against her head and chest (copied upon seeing other GTS patients performing these actions), head jerking, hand jerking, stamping feet on the ground	Yes	Yes/Yes/Yes	Yes/Yes	None	Nabiximols improves noises by 30–40% No effect with Atomoxetin, Aripiprazole, Tiaprid, Risperidone	GTS plus ADHD;
7	41/ M	33	Abrupt, during withdrawal treatment from opiate addiction	Whole body jerks	None	"Schwule Sau", "fette Sau", "Arschfick", "Arschficken mit Kindern", "Pädophil", "Sex mit Kindern", "Scheißtürken"	NOSI: "man bist du hässlich", "ich bin pädophil"	No	Rarely grimacing, Sticking of tongue	Yes, but not before co-prola-lia or voca-liza-tions	Yes/Yes/Yes	No/Yes	None	Aripiprazole/tics deteriorated; Amisulpride/no effect; Quietiapine/ improvement of jerks, but deterioration of coprolalia; Improvement of symptoms when smoking marijuana	GTS plus OCD
8	18/F	14	Insidious/ Episodes of school mobbing	Left leg tremor	None	–	Echolalia: only sounds from people with GTS; Palilalia: "Sägemehl", "Katzengulasch", "aber hey", "ha hey", "ahu", "hallelujah"; NOSI: "Handy weg", "Kettenraucher", "Feuer", "Katzenfeuer"	No	Blinking, head jerks, tongue protrusion, grimacing, hitting head with hands	Yes	Yes/Yes/Yes	Yes/Yes	None	Tiapride, Aripiprazole, Pimozide, Risperidone, Fluoxetine, Quetiapine/None	GTS
9	41/ M	29	Unknown/No	Repetition of a single word ("Wüstchen")	None	–	Palilalia: "Wüstchen"	No	None	No	No/No/No	Yes/No	None	None	Tic disorder
10	41/ M	39	Abrupt/No	Episodic whole-body shaking	"Ah", "Nah", "Hm"	–	Palilalia: "lalalala" in bouts of about 20 s	Speech blocks	Episodic whole-body shaking, maintaining left	Yes	No/Yes/Yes	Yes/No	None	Sulpiride, Haloperidol, Clonidine/None	GTS plus ADHD

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Table 1 *(continued)*

Case Number	Age/ Sex	Age at onset of functional tic-like symptoms	Modality of onset/ Precipitants	Symptoms at onset	Simple vocalizations on presentation	Complex vocalizations on presentation		Blocking phenomena	Other movements on presentation	Urge	Suggestibility/ Distractibility/ Suppressibility	Stereotyped nature/ Fluctuations	Other functional symptoms	Previous Treatments/ Improvement	Previous Diagnosis
						Coprolalia	Echolalia/ Palilalia/NOSI								
									leg in the air when walking, pressing buttocks in mattress when lying						
11	13/F	11	Abrupt/No	Noises (hissing, sniffing, coughing)	None	–	Echopalilalia: completing and repeating technical terms -	No	None	No	No/Yes/No	Yes/Yes	Aversion towards certain noises (smacking, sniffing)	Aripiprazole/ Transient improvement	Chronic vocal tic disorder
12	51/ M	50	Abrupt/ Divorce	Noises, syllables, words, episodic leg tremor	Very loud shouts	–	Palilalia: repetition of own words and sentences up to 100 fold	Speech blocks	Multifocal jerky movements	Yes (but only during the past few months)	No/Yes/Yes	Yes/Yes	None	Lorazepam/ Improvement of movements but not sounds; Doxepin, Promethazin/ None	GTS
13	56/ M	46	Insidious/ Post minor surgical procedure on the left shoulder	Left arm tremor	Noises (“brr”, “tsch”)	–	Palilalia: “Ach-so ja ja” always 3 fold	No	Complex movements of left arm and hand	Yes	Yes/Yes/Yes	Yes/Yes	Functional gait disorder	Aripiprazole, Tetrabenazine/ None	GTS

had specific atypical features that led us to question the diagnosis of GTS. These features included: age at onset, abrupt symptom onset with physical and/or psychological precipitators, presence of additional functional neurological symptoms, including other functional movement disorders, such as functional tremor, as well as atypical contextual factors, including lack of response to typical anti-tic medication, such as neuroleptics. Relevant clinical and paraclinical examinations in these patients, including laboratory tests, electroencephalography and brain imaging had been performed prior to referral, and were unrevealing. Patients with functional movement disorders and additional simple and less prominent vocalizations mimicking "simple vocal tics" (such as sniffling and coughing) were not included.

3. Results

Thirteen patients who presented with complex atypical tic-like vocalizations were identified (complete clinical characteristics provided in Table 1). All patients had previously received a diagnosis of a primary tic disorder and were referred for further specialist treatment.

Seven patients had onset of symptoms in adulthood and the remaining six during childhood or adolescence (range: 5–50 years). Ten patients had abrupt onset of symptoms. At symptom onset, patients presented with abnormal vocalizations, but also movement disorders such as functional tremor or functional jerks. Coprolalic behaviors were the predominant symptom in seven cases and in four of those this was already during early childhood or adolescence (cases 2, 4–6). Echolalia was present in five patients and palilalia in 10. Interestingly, two patients with echolalia would echo sounds or vocalizations from other people with GTS (case 6, 8), but also from each other. Two patients had NOSI (case 7, 8). Eleven patients also had jerky tic-like movements, which were, however, in most cases milder compared to their repetitive vocalizations.

Nine patients reported a general sensory premonition prior to the occurrence of their vocalizations. Tic-like symptoms were suggestible in seven cases, and could be temporarily suppressed by 10 patients. Fluctuations of symptom severity were present in 11 patients and were atypical in most cases (e.g. full remission during holidays with sudden symptom onset at the moment the patient crossed the doorstep of his house upon his return; case 11). Ten patients had been treated – in most cases with several different - classical anti-tic medications, for example antipsychotics, without any symptom improvement or with atypical adverse effects (e.g. inability to speak after treatment with low dose aripiprazole, case 5). Remarkably, in three, otherwise pharmacological treatment-resistant patients (cases 5–7), usage of cannabinoids led to marked symptom improvement.

Five patients had additional functional symptoms at the time of last follow-up, including functional movement disorders (see Table 1). Four patients had received a diagnosis of attention-deficit hyperactivity disorder (cases 4–6, 10) and one of obsessive-compulsive behavior (case 7). There was no family history of tic disorders in any of the patients.

4. Discussion

Functional tic disorders are a rare presentation of functional movement disorders (Demartini et al., 2015). Their distinction from organic tics is challenging, as the very nature of the latter sits at the phenomenological edge between volition and involuntariness (Ganos et al., 2015). Functional tic disorders were acknowledged in earlier literature, but their clinical characteristics, perhaps due to the difficulties in ascertaining a definite diagnosis, have only recently been summarized in two case series (Baizabal-Carvallo and Jankovic, 2013; Demartini et al., 2015). However, emphasis was given at motor manifestations, whereas complex vocalizations, such as coprolalia were not reported.

The patients presented here were atypical, in our view, from patients with vocalizations due to GTS, and had no features of a secondary cause for their vocalizations. In most cases, additional tic-like movements were present, but these usually rather accompanied the loud utterances, were milder in severity and did not present an equal source of distress. In fact, the majority of the presented cases had sought medical attention due to vocalizations and not due to their other symptoms and had all, therefore, been first diagnosed as primary tic disorder.

Some of the clinical features of these cases overlapped with previous suggested clues to distinguish functional tic-like movements from their organic counterparts (Baizabal-Carvallo and Jankovic, 2013; Demartini et al., 2015). For example, as in previous reports (Baizabal-Carvallo and Jankovic, 2013; Demartini et al., 2015) also in our cases onset of symptoms in adulthood, psychological or physical precipitants and tic-related blocks were common. In addition, response to anti-tic medications was disappointing. Further functional symptoms, including functional movement disorders, were present in five of 13 cases and only five patients had psychiatric comorbidity profiles comparable to those observed in primary tic disorders. Interestingly, obsessive-compulsive behavior was very rare in our group of patients (only in one case).

On the other hand, there were some other features that differed from previously published case series of functional tics. Indeed, six patients developed symptoms under the age of 18, and one of them at the age of 5 years. This is consistent with some reports on functional movement disorders in children and adolescents showing that functional tics can occur in this age group (Ahmed et al., 2008; Isaacs et al., 2011), but may also point to the common situation of co-occurrence of functional and organic neurological disorders. Although establishing the exact prevalence of functional tic disorders is beyond the scope of this report, our data emphasize that, albeit rare, the diagnosis of a functional tic disorder should be considered even in children who present with complex tic-like vocalizations, including coprolalia. Further, different than previous reports (Baizabal-Carvallo and Jankovic, 2013; Demartini et al., 2015), nine of our 13 patients did in fact report the presence of a sensory premonition prior to tic-like vocalizations. Even though qualitative descriptions of premonitory sensations were atypical compared to patients with primary tic disorders (e.g. "a sudden energy pulse", "generalized whole body pressure"), this highlights that the mere presence or absence of sensory experiences related to tics and tic-like phenomena cannot be a single diagnostic criterion. Finally, the absence of symptom fluctuation has been suggested as a further helpful hint to discern functional from primary tics (Demartini et al., 2015). However, 11 of our patients did in fact report (unusual) changes in their tic frequency and severity over the course of time. This again highlights that clinicians should not rely on a single clinical characteristic in order to reach diagnosis.

Another interesting point to raise relates to the content of coprolalic behaviors. Coprolalic words in GTS are usually uttered loudly during sentence pauses, often with imprecise pronunciation of phonemes and in a different pitch and tone than that of the ongoing conversation (Singer, 1997). Common coprolalic utterances comprise short words, typically, in English language, four-letter words (Nuwer, 1982; Singer, 1997). On the other hand, in most patients reported here the selection of words was rather different. Patients with functional coprolalia uttered not only longer or compound words or even short sentences of obscene content, but also an atypically high number of different swear words (up to thirteen different words) or unusual coprolalic utterances the authors never encountered before in patients with GTS (examples presented in clinical table). Although our patient sample is quite small, we suggest this to be a further hint to guide diagnosis.

We appreciate that the clinical categorization of a functional disorder for the patients presented here is difficult, for reasons, which have been highlighted in previous papers on functional (motor) tics (Baizabal-Carvallo and Jankovic, 2013; Demartini et al., 2015). We, also, cannot exclude that some patients may have had primary

(organic) tics at some point. Indeed the co-occurrence of organic and functional disorders is common in movement and also other neurological (particularly paroxysmal) disorders, such as seizures (Benbadis et al., 2001; Erro et al., 2016; Erro and Tinazzi, 2014; Ganos et al., 2014). However, the predominant clinical signs of the cases presented here, are, we argue, functional for the reasons outlined above. Though we accept that our classification may be open to error, we believe it to be important to try to separate out these patients from those with GTS, as treatment is likely to be different, as evidenced by the poor response of most of our patients to anti-tic medication. In addition, this is a retrospective study and hence there are limitations as to the clinical information that has been retrieved. However, thorough evaluation at the time of presentation allowed for a precise characterization of the main clinical features of these patients. Further, our study sample is relatively small. On the other hand, this is the first report to highlight functional coprolalia and other complex tic-like vocalizations, which are particularly rare, as we identified 13 of a large sample of about 1.500 patients seen in a large tic disorders psychiatric clinic in a period of 20 years. However, the clinical impression was that the prevalence of functional coprolalia increased within this period, particularly over the last decade, owing, we believe, to raised awareness concerning GTS, as a result of media coverage. Finally, we do not provide follow-up details and information on treatment outcome. Although the diagnosis was explained in detail to all patients, and in fact accepted by their majority, they were, subsequently, referred back to their attending physicians for treatment of functional neurological symptoms. However, the nature of such treatment for functional symptoms in general is still much debated, with limited evidence available to guide decision making, and therefore may be a case for continuing to look after such patients within a specialist tic clinic.

To conclude, we here present the clinical characteristics of patients with functional coprolalia and other complex tic-like vocalizations, who on the basis of their symptoms had been misdiagnosed as having a primary tic disorder, most commonly GTS. We wish to highlight that the diagnosis of a primary tic disorder should rely on a combination of neuropsychiatric symptoms and signs and not on single clinical features, even in the presence of striking behaviors such as coprolalia.

Conflict of interest

All authors report no conflict of interest.
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