

ON DIFFERENCES BETWEEN GILLES DE LA TOURETTE SYNDROME AND PSYCHOGENIC/FUNCTIONAL TICS: A NARRATIVE REVIEW

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SUMMARY

A tic is a sudden, rapid, recurrent, nonrhythmic motor movement or vocalization. Motor and phonic tics in a course of over 1 year that first occurred before 18 years are the main features of Gilles de la Tourette syndrome (GTS). Psychogenic/functional (P/F) tics were considered as a diagnosis made per exclusionem in lack of agreement of diagnostic criteria. Recently, emphasis is in rather highlighting positive signs when making diagnosis of P/F tics. Several features in clinical course are important to differentiate GTS from P/F tics. Some of them are acute onset in adulthood, precipitation by physical event, absent family history of tics, variable, complex and inconsistent phenomenology, suggestibility, distractibility. Premonitory urge, feeling of excessive energy and being 'wound up' prior tic, is a usual feature of GTS unlike in P/F tics. If present, such premonitory urge have different qualitative and quantitative marks. Another possible diagnostics tool could be the beireitshaftspotential, an event-related electrical potential associated to initiation of movement which is divided in two phases, early (B1) and late (B2) phase. Early phase, whose occurrence in some papers has been reported prior P/F tics, is absent prior performed tic in GTS. In everyday clinical practice differentiating GTS from P/F tics is often very challenging but taking proper medical history, paying more attention to positive signs and possibly using electroneurophysiology tests could contribute in making the right diagnosis.

Key words: Tourette syndrome - tic - functional tic - psychogenic tic - premonitory urge

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INTRODUCTION

A tic is a sudden, rapid, recurrent, nonrhythmic motor movement or vocalization. According to Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5) there are four diagnostic categories. Gilles de la Tourette Syndrome (GTS) is defined as childhood neurodevelopmental disorder that for valid diagnosis requires both motor and phonic tics during at least period of time of 1 year and with onset before 18 years old. In addition, secondary causes has to be excluded. Second are chronic tic disorder, either motor or phonic. Third category are provisional tics which for valid diagnosis requisite tic lasting less than a year and last category are other specified and unspecified tic disorder. In evaluating certain tic repertoire, Paszek et al. reported importance of assessing tic through another parameter – time. Movement kinematics are hardly distinguishable from normal movements in shorter period of time such as 3 seconds in comparison to 20 seconds). This narrative review tends to compare abovementioned tics and psychogenic tics. The latter is usually defined as hyper- or hypo-kinetic movement disorders, that cannot be directly attributed to a lesion or dysfunction of the nervous system and which are usually thought to derive from psychological stress (Thomas & Jankovic 2004). Throughout last decade, another terminology has been suggested, thus more

recent articles emphasise on using term „functional“ rather than „psychogenic“. Discussion about more appropriate terminology is beyond the scope of this review so the term psychogenic/functional (P/F) will be used this point forward. In DSM-5 functional tics are out of subheading „Tic disorders“ since they have been put under „Somatic Symptom and Related Disorders“. An improvement had been made when a term „Functional Neurological Symptom Disorder“ was introduced in DSM-5 alongside with Conversion disorder. Criteria presented in DSM-4 that required recognized psychological stressor was excluded out of DSM-5, which provided many patients without obvious stressor to be properly diagnosed and treated. Moreover, many patients with psychogenic disorder didn't report any recent psychological stress (Morgante et al. 2013). Since interest has been increasing is discovering pathophysiology of P/F disorders and therefore a biological role in pathogenesis is supported, P/F disorders are being categorized under the neurology section of ICD-11 (Stone et al. 2014). And yet some papers reported phenomena of P/F movement disorders as a 'crisis for neurology' since they can be very challenging in both diagnostic and therapeutic terms for experts (Hallett 2006). The goal of this narrative review is to summarize and concisely present hopefully helpful guidelines for distinguishing GTS from psychogenic tics.

CLINICAL COURSE

Psychogenic/functional tics are usually described as rare phenomena. According to few papers P/F tics represented 4-15% of the all P/F movement disorder (Baizabal-Carvalho & Jankovic 2014, Ertan et al. 2009), while P/F disorders encountered in neurologic clinics represents 15% of the admitted patients (Maccotta et al. 2009). However other researches suggests otherwise somewhere between 1.5 and 3% (Factor et al. 1995, Edwards & Bhatia 2012, Hallett 2006). It is clear that consensus about prevalence of P/F disorders in general, and within it P/F movement disorders has not been brought out, and exact number is yet to be determined. On the other hand, Gilles de la Tourette is more consistently reported to prevail around 1% in general population, with higher prevalence in males than females opposite to distribution of patients with FMD (Cath et al. 2011). Even though there has been papers reporting rare cases where patients had both GTS and psychogenic tic (Dooley et al. 1994), usually that is not a case, rather a diagnostic challenge when determining origin of certain tic symptomatology. As noted above, there are clear criteria when making diagnosis of Gilles de la Tourette syndrome, while there are still not clear and consistent consensus, therefore criteria for declaring a certain movement disorder psychogenic/functional. In recent years it has been consistently suggested that positive signs should be considered, rather than solely deciding on diagnosis per exclusionem (Demartini et al. 2015, Morgante & Edwards 2013). Regarding that, nowadays the diagnosis is mostly based on neurologic examination focusing on specific positive clinical sign along with taking proper history of disorder. In support of a P/F tics are: acute onset in adulthood, precipitation by physical event, absent family history of tics, variable, complex and inconsistent phenomenology, suggestibility, distractibility, entrainment and a *Bereitshaftspotential* preceding the movement (Dreissen et al. 2016, Ganos et al. 2014). Demartini et al. also reported in their research that lack of the typical rostrocaudal tic distribution, usually related to GTS, presence of blocking tics, absence of echophenomena and coexistence of other functional movement disorder were common among patients. According to Fahn and Williams criteria, response to placebo gives one the highest level of diagnostic certainty. In addition, waxing and waning course is usually related to GTS (American Psychiatrist Association 2013), which is not the case in patients with P/F movement disorder. Although generally thought, coprolalia is not exclusive symptom of GTS. Coprolalia was recognized pathognomonic for GTS in less than 20% of cases with somewhat higher frequency in males (Freeman et al. 2008), but Ganos et al. (2016) reported on functional coprolalia. They also suggested that qualitative view should be taken while thinking about diagnosis. They showed rather quite broad spectrum of

used word and unusual length of coprolalic vocalization, in contrast to GTS where coprolalia is comprised usually of short words.

In terms of diagnosing Gilles de la Tourette syndrome, there are few features from patient's history commonly related to the condition. In contrast to late onset of P/F tics, GTS symptomatology starts around mean age of 5 years, with motor tics appearing first followed by phonic tics in later years. Longitudinal studies pinpointed favourable course of disorder in up to 80% of patients. Decreased intensity and frequency of tics occurs mostly during adolescence (Pappert et al. 2003). Moreover, poorer quality of life in adolescents and adults with GTS, if present, is related to persistence of comorbidities, especially OCD (Cath et al. 2011). Psychiatric comorbidities are consistently reported to be present in up to 90% of patients, whereas only 10% of patients have so-called „pure GTS“ (Grados & Mathews 2009). In addition, Cravedi et al. investigated GTS phenotypes in 174 children and adolescents in French university clinic. Three clusters were identified. One of them corresponded to a abovementioned „pure GTS“ whereas another cluster included learning and intellectual disabilities, ASD and ADHD. The third cluster corresponded to an ADHD profile with rather high intelligence. Interestingly, two of the most often comorbidities, OCD and ADHD, typically occur at different stages. ADHD usually occur before onset of tics (Stewart et al. 2006) and OCD after onset (Palermo et al. 2011). On the other side, profile of comorbidities associated with P/F tic are somewhat different. Higher frequency of major depressive episodes, anxiety disorders and personality disorders have been associated with psychogenic movement disorders (Demartini et al. 2014).

Presence of tics during sleep and therefore decreased mean slow wave sleep period (Młodzikowska-Albrecht et al. 2007) could possibly also contribute to decreased quality of life, whereas tic presence has not been reported in patients with P/F tics. According to European Clinical Guidelines for GTS and other tic disorder another feature that is distinguishing between GTS and other movement disorder and therefore P/F tics is feeling of active involvement in performing a tic, especially in adults. It is explained as being a sense of intentional movement performed to relieve premonitory urge. Although P/F tics and GTS appear separately, psychogenic/functional component is reported to deteriorate intensity and frequency of tics resulting in bouts of tics, phenomena described as 'tic attacks'. Robinson & Hedderly (2016) proposed that such tic attacks may resemble a combination of tic and psychogenic/functional neurological movement. In their paper, a strong correlation between level of anxiety and increased internal focus of attention on tic was found. Maladaptive coping mechanism regarding increased sensory urge prior tics and cognitive misattribution are hypothesized to contribute maintenance of such phenomenon.

PREMONITORY URGE

Feature that is often mentioned in literature preceding executed tick is premonitory urge. According to assessment of tics using the Premonitory Urge to Tic scale, it is the most commonly described either as excessive energy needed to be released or feeling of being tense or „wound up“ (Reese et al. 2014, Dallochio et al. 2010). After tics is done, urge resolves along with present discomfort prior tic. Recently it has been reported that premonitory urge is present in 73% individuals with GTS, with higher prevalence in those with complex tics (78.6%) over those with simple tics (68.9%) (Jakubovski et al. 2018). Also, in same paper it is noted that urges tends to localize in same body area where tic is going to be performed. Interestingly, it is believed that premonitory urge is not present in children younger than 10 years (Raines et al. 2017, Leckman et al. 1993). Using PUTS, a correlatation is found between degree of premonitory urges and both estimated IQ and tic severity. In contrast, age, gender and severity of comorbid obsessive-compulsive disorder or attention-deficit hyperactivity disorder didn't have any correlation (Reese et al. 2014). There are researches (Soler et al. 2019) reporting that other elevated sensory phenomena dysregulation exists in GTS spectrum, especially linked to complex tics and GTS phenotype with comorbidities. Premonitory urge could be used as potential diagnostic tool as it is well-known sensory phenomena related to GTS, but appears not to have such relations to psychogenic tic.

Premonitory sensations prior tic are also thought to play a central role in the maintenance of tics. Current behavioral models annotate that tics are negatively reinforced every time they rid the patient of the discomfort associated with the premonitory urge (Crossley et al. 2014). Therefore, it has become a target for treatment in Habit Reversal Training (HRT), part of Comprehensive Behaviour Intervention for Tics (CBIT) which according to meta-analysis shows promising results (Mcguire et al. 2014). Habit Reversal features set of techniques that help the patient become aware of occurrence of tic, followed by a so-called competing response training to interrupt or inhibit the tic. Moreover, for less severe forms of GTS, behavioural therapy is considered as a first choice therapy (Verdellen et al. 2011, Pringsheim et al. 2019). In addition, it has been reported that psychiatric comorbidities such as attention-deficit/hyperactivity disorder or compulsive-obsession disorder did not moderate on improvement after CBIT. Improvement was even greater for those patients who prior CBIT weren't on tic-suppression medications (Sukhodolsky et al. 2017).

On the other hand, treatment of P/F tics starts with very first explanation of diagnosis, since few studies emphasized importance of patient's acceptance of diagnosis which can be challenge for itself. Although treatment has not been standardized yet, physiotherapy has been recently proven beneficial for functional movement disorders other than tics (Dallochio et al. 2010,

Demartini et al. 2019). Although physiotherapy is suggested as a valid form of therapy for patients with functional motor symptoms, therapeutic value should be validated further. More traditionally, common approach is still psychotherapy either with psychodynamic or cognitive-behavioural approach, both resulting in modest improvement of condition. According to some studies, long-term prognosis is still unfavourable and requires more effective strategies in treating P/F movement disorders (Gelauff et al. 2014). It is worth mentioning that patients with GTS have ability to suppress tic for a while, but followed by increased urge until it has to be done, providing therefore a relief. To compare it with P/F tic, no such urge or ability to suppress tic has been recognized to occur, which is valuable in differentiating GTS from P/F tics (Dreissen et al. 2016). However, it has been reported that patients with P/F tic-like complex vocalization had premonitory sensation, however qualitative differently described. Sensation were atypical compared to patients with GTS (e.g. „a sudden energy pulse“, „generalized whole body pressure“). Since there has been reports on few cases where patients with GTS were unable to suppress tic, inability to halt tic or urge prior tic is highly supportive but not sufficient for definite diagnosis (Espay & Lang 2015).

THE BEIREITSHAFTSPOTENTIAL

The BP is an event-related electrical potential reflecting activity of SMA prior to voluntary movement associated to initiation of movement (Colebatch 2007, Obeso et al. 1981). Although first mentioned in 1980es (Libet et al. 1982) and went under series of experiments, until nowadays criteria, classification and underlying neurophysiology remain unclear. At the beginnings origin of BP was placed in supplemental motor area (Obeso et al. 1981), but later on, as SMA itself was better understood, it became divided into rostral and caudal SMA. SMA is usually associated as a cause to longer latency of BP (Cunnington et al. 2003). Recently, for the lack of established criteria for presence of the BP, some studies (Van Der Salm et al. 2012, Colebatch 2007) suggested two phases of BP. Van et al. proposed BP to be classified in the early and late BP. Early potential is measured from 0.2 to 0.1 seconds prior to the onset of the burst on EMG ($t=0$), whereas late potential is defined between 1 second and a half of a second. This classification has been shown as beneficial in distinguishing GTS tics apart from myoclonus and P/F movement disorders, although less decisive than patient's medical history, neurological examination and first impression, respectively (Van Der Salm et al. 2013, 2017). In contrary to believed event-related nature of a BP, a novel finding shows absence of BP prior to volitional movement in patients with P/F tics (Van der Salm 2014). BP phase that certain type of tick movement features is also valuable as differentiating tool since there is shown for GTS to lack early phase (BP1), opposite to

P/F tics which majority were preceded by early phase. As mentioned before, GTS in majority cases encompasses comorbidities such as OCD, ADHD. These condition could be disruptive to interpretation of a BP since it is shown that ADHD for itself has reduced BP amplitudes compared to controls (Jarczok et al. 2019, Jarczok & Haase 2019). Some papers suggest EMG to be first choice in differentiating functional from nonfunctional origin of tic, since burst time during jerk that are less than 75 ms are considered hardly to be functional. However, low specificity in distinguishing GTS from P/F jerks decreases diagnostic value of mentioned examination (Dreissen et al. 2016). Specificity for the diagnosis of P/F jerks and tics are 0.68 and 0.26, respectively, which is still notably low, but further standardized measurements and better understanding of electrophysiology in the background is needed. In addition, The Gupta Lang criteria suggested laboratory supported approach for distinguishing primary from psychogenic movement disorders, including also frequency analysis and EMG-EEG back averaging with Bereitschaftspotential.

CONCLUSION

Differentiating GTS from P/F tics are usually challenging in everyday clinical practice. In spite of lacking more thoroughly understanding of biological background, there are enough papers and knowledge about P/F movement disorders to consider it as valid diagnosis very disruptive for patients and not just diagnosis of exclusion when nothing fits. Approach to positive diagnosis has been consistently encouraged. However, in terms of differentiating GTS from psychogenic tics, we pinpointed few possibly helpful features of each disorder. Critical clinical assessment which can provide brighter course to correct diagnosis is crucial along with properly taken medical history. Inconsistency, unusual distribution and non-rhythmic tics with certain distractibility are highly suggestive of P/F diagnosis. Electrophysiological measurements could be helpful but clarification on diagnostic value is needed. Since functional movement disorders are primarily diagnosed by neurologists with expertise in movement disorders and part of the treatment of possible underlying intrapsychic conflicts is up to psychiatrist, further close liaison between two specialties is encouraged.

Contribution of individual authors:

Ante Periša made substantial contributions to conception and design of this review, extracting and interpretation of data, as well as in drafting the article.

Srđana Telarović was involved in interpretation of data and has made crucial suggestions about the scope of this review along with revising the manuscript and giving the final approval of the version to be submitted.

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