Repetitive behaviors in Tourette Syndrome and Obsessive-Compulsive Disorder

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Abstract

Tourette Syndrome (TS) is a neuropsychiatric disorder characterized by tics that are frequently associated with obsessive-compulsive symptoms (OCS), estimated to be present in 28-63% of patients, though not always sufficient for the diagnosis of an Obsessive-Compulsive Disorder (OCD). Epidemiological, phenomenological and family-genetic studies support an interrelationship between tic disorders and OCD. The repetitive behaviors present in either TS and OCD—tics and compulsions—have been associated with dysfunctional cortico-striato-thalamo-cortical circuits, which is consistent with the hypothesis that OCS and tics might be part of the same spectrum. According to that, OCD with tics assumes an intermediate position between OCD without tics and TS, sharing clinical features with both entities. On the other hand, many studies have reported that tic-related OCD presents specific clinical, neurobiological, genetic and treatment response patterns and TS can be differentiated from OCD subgroups according to obsessive-compulsive symptomatology, presence of sensory phenomena and treatment response. Thus, the phenomenological distinction of repetitive behaviors in patients with TS is important to consider, since the treatment of tics and OCS differs.

Keywords: Tourette Syndrome, Obsessive-Compulsive Disorder, Repetitive Behaviors, Tic, Compulsion, Treatment.
Introduction

Tourette Syndrome (TS) is a neuropsychiatric disorder characterized by the presence of multiple motor tics and one or more vocal tics, not necessarily concurrently, for a period of more than one year and onset before the age of 18 years. TS is relatively common, affecting around 1% of school-aged children [1–3]. Males suffer from TS more frequently than females (3/4:1) [4] and a multifactorial etiology has been proposed, in which immunological, environmental and genetic factors interact to establish vulnerability. TS has a polygenic inheritance, with the SLITRK1 gene in the chromosome 13q31 being proposed as one of the possible genes involved in TS [5].

Tics, the clinical hallmark of TS, are sudden, rapid, recurrent, nonrhythmic motor movements (motor tics) or sounds (vocal tics). Motor tics can be simple, such as blinking, or complex: coordinated and sequenced movements resembling normal motor acts or gestures that are repetitive and inappropriately intense and timed. Examples of complex motor tics are repetitive touching and jumping. Vocal tics may also be simple, like coughing or grunting, or complex, when they include linguistically meaningful verbalizations, such as shouting obscenities or profanities (coprolalia), repeating someone else’s words or phrases (echolalia), and repeating one’s own utterances, particularly the last syllable or word in a sentence (palilalia).

By early adulthood, roughly three-quarters of children with TS will have greatly diminished tic symptoms and more than one-third will be virtually tic free [6].

Both motor and vocal tics are often preceded by a sense of urgency and imperiousness. This premonitory phenomenon consists of either localizable sensation or discomfort in the region of the tic that compels it for its execution and is relieved by the movement [7].

Obsessive-Compulsive Disorder (OCD) is characterized by obsessive thinking and/or compulsive behavior and has a lifetime prevalence rate in the general population between 1.9 and 3.2% [8]. Although its diagnostic criteria remain the same in DSM-5, this disorder is now grouped in a new chapter entitled “Obsessive-Compulsive and Related Disorders”, along with Hoarding Disorder, Skin-Picking Disorder, Body Dysmorphic Disorder and Trichotillomania, among others. This new categorization reflects the recent evidence of these disorders’ similarities, such as an obsessive preoccupation and repetitive behaviors, and probable relatedness to each other, albeit several contradicting family and twin studies proving an association between OCD and anxiety disorders, namely in paediatric populations [9,10].

Although children may present obsessive ideas and compulsive behaviors as elements of normal development, a very similar phenomenology of adult OCD may also be present in children. So, as in adults, child OCD is an often chronic and in many cases disabling condition affecting 1-3% of the pediatric population [11]. This early onset subtype seems to be related to tic disorders, namely TS, being frequently comorbid conditions. Roughly one-third to one-half of children with TS will experience comorbid OCD throughout their lifetime. Symptoms of OCD that are subclinical in severity are even more common [6].

Repetitive behaviors (RB) in TS have been the subject of several analyses, mainly due to its high prevalence and controversy in phenomenological categorization: are they complex motor tics or compulsions? Are they part of TS or indicate OCD comorbidity [12]?

These questions prompt another one: to what extent are RB in TS and OCD mediated by overlapping versus distinct substrates [13]?

Tourette Syndrome and Obsessive-Compulsive Disorder

Epidemiological, phenomenological and family-genetic studies support an interrelationship between tic disorders and OCD. This association appears to be bidirectional, with 20-60% of tic disorder patients meeting criteria for OCD, and 20-38% of children with OCD reporting comorbid tics [14].

Obsessive-compulsive symptoms (OCS) are present in 28-63% of patients with TS and genetic studies report an increased prevalence of OCD in first-degree relatives of TS patients, independently of concurrent OCD in these patients, and an increased prevalence of tics or TS in first-degree relatives of OCD patients [15]. However, OCD seems to be an etiologically heterogeneous entity, with only a tic-related form of OCD being related to TS, which is characterized by an earlier age of onset, greater prevalence in males and higher frequency of premonitory sensory phenomena preceding RB [12,16]. The average age at symptom onset is even lower in a subgroup of children, the PANDAS (pediatric autoimmune neuropsychiatric disorders associated with streptococcal infections) subgroup, in which an abrupt onset of tic disorders, obsessions and compulsions and a temporal association with a group-A-beta-hemolytic streptococcal (GAS) infection were identified. In this PANDAS subgroup, the average onset age is nearly 3 years lower than that reported for childhood-onset OCD and up to 2 years lower than the average age of onset for tic disorders. These neuropsychiatric symptoms are postulated to arise from an interaction of GAS antibodies with neurons of the basal ganglia [17].

TS and OCD share several characteristics: both disorders have a chronic waxing and waning course throughout the lifetime; they are characterized by the presence of RB, and have a childhood or juvenile onset. And although OCD may manifest later in life, an association is apparent, such that a large proportion of children diagnosed with OCD also present with tics. But despite the many similarities between TS and OCD, there are of course clear differences, as tics appear overall more likely to improve with age, whereas OCD symptoms may worsen over time [18]. Research has also shown that while OCD phenom-
ena are accompanied by more complex thought processes and autonomic anxiety, tics are usually preceded by short-lived sensory symptoms and feelings of incompleteness. This difference in how RB are experienced by patients has led to their subdivision into two categories: “OCD-like”, or OCD-related compulsions and “Tic-like”, or TS-related impulsions [12,15]. Other differentiations include the ego-syntonic/ego-dystonic nature of the RB, perceived voluntariness and duration of the RB. Thus, RB in TS are considered to be not anxiety-related, to have an egosyntonic character, to be performed in a stereotypical way, to be stimulus-bound and to be more frequently accompanied by premonitory sensory phenomena, feelings of incompleteness and “not just right” perceptions. Although these “not just right” experiences are also frequently described in OCD [19], these overall features near RB to the concept of impulsion, but it is important to note that many of these behaviors may actually be complex motor tics. Also, the use of terminology impulsion in TS, although is related mainly with RB's purpose and relation with anxiety, may associate it with Impulse Control Disorders and to date there are no indications that these disorders are specifically related to TS. For this reason, a more neutral terminology has been suggested, such as “non-anxiety related” or “stimulus bound” repetitions [15].

TS and OCD-related RB, tics and compulsions, have been associated with a dysfunction of cortico-striato-thalamo-cortical circuits, consistent with the hypothesis that obsessive-compulsive symptoms and tics might be part of the same spectrum [12,20]. The corticostriatial circuits are multiple parallel, segregated feedback circuits with outputs from striatum targeting primary motor areas, and specific pre-motor and prefrontal cortical areas. They are typically grouped in the sensorimotor circuit (sensorimotor cortex), the associative or cognitive circuit (dorsolateral prefrontal cortex) and the limbic circuit (anterior cingulated and orbitofrontal cortex). The primary function of the corticostrialat circuits is to control and select goal-directed motor, cognitive and motivational behavior. Although anatomical and functional disturbances in corticostral circuits are thought to be centrally involved in TS and OCD, recent neuroimaging data suggest that motor and vocal tics in TS correspond to changes in sensorimotor loop, whereas obsession and compulsion in OCD are associated with the limbic loop. In TS with comorbid OCS, not only the sensorimotor cortex is involved in motor stereotypies, but also the orbitofrontal circuit is implicated in comorbid obsessions and compulsions. The involvement of the associative loop has been associated with impulsive and rigid behaviors [13]. Differences in the neurobiological mechanisms underlying RB in TS and OCD may help to explain differences in clinical presentations and disparities in how RB are experienced by OCD and TS patients. Therefore, the driving forces behind RB in OCD are likely to include greater focus on mental events while in TS patients there may be hyperawareness of sensory and physical experiences. In other words, repetitive thinking in OCD is more associated with anxiety and negative emotions, involving greater high-order conscious experience of the RB [8] and consequently being a more prefrontal-limbic phenomenon than repetitive behavior in TS.

Children who present at the onset with pure TS generally have a quite good long-term clinical course, whereas those who present a comorbid condition at the onset present a more severe prognosis. Rizzo et al. (2012) reported that OCD can have a significant negative impact on the course of TS because of its deleterious effects on quality of life [4].

**Obsessive-Compulsive Spectrum**

Several authors agree with the existence of an obsessive-compulsive spectrum incorporating diverse clinical entities that share characteristics with OCD, including symptom overlap, family history, neurobiologic features, clinical course and treatment response. Thus, obsessive-compulsive spectrum disorders (OCSD) can be viewed along a continuum with a compulsive risk-averse pole at one end and an impulsive risk-seeking pole at the opposite end [15]. In this scheme, tic-free OCD with predominant obsessions and compulsions is positioned at the compulsive end, whereas Impulse Control Disorders are positioned at the impulsive pole. However, as Eric Hollander underlines, it has now become evident that many of the OCSD have both compulsive and impulsive features, so rather than polar opposites, compulsivity and impulsivity represent key factors that contribute in varying degrees to each one of the OCSD that will in this sense adopt different positions along the spectrum. Hollander et al. also point that the one unifying feature of OCSDs seems to be that they all are linked by a RB domain [21].

Even though OCD is classified as an unitary nosologic entity, recent research has indicated that OCD seems in fact to be an heterogenous disorder, with a variety of putative clinical phenotypes and probably different etiologies. So far, one of the best described OCD subgroup is composed of OCD patients with an associated tic disorder. Many studies have reported that the tic-related OCD subgroup presents specific clinical, comorbidity, neurobiologic, genetic and treatment response patterns. The existence of this tic-related OCD subgroup with an overlap of clinical features of TS and OCD explains why it may be so difficult to differentiate these two entities [16].

By integrating RB into the referred obsessive-compulsive spectrum, TS with TS-related impulsions, compulsions and obsessions will assume a position slightly deviated towards the impulsive pole, while tic-related OCD will be positioned closer the compulsive pole, between tic-free OCD and TS with or without OCS. This intermediate position of tic-related OCD is justified by its shared clinical features with both tic-free OCD and TS. But what are the exact relative positions of these categories?
The precise delineation of TS and OCD subgroups is important because the different subtypes might be related to different treatment responses and genetic transmission. One way to delineate TS from OCD subgroups is to compare their (a) obsessive-compulsive symptomatology, evaluate the presence of (b) sensory phenomena and (c) treatment response.

**Obsessive-Compulsive Symptomatology**

It has been described that intrusive aggressive or inappropriate sexual thoughts and images are more common in TS than OCD, whereas obsessions and compulsions in OCD alone are more often related to fears about contamination or harm coming to another person [8].

According to symptomatology, patients with tic-related OCD have more frequently religious obsessions, contamination worries and washing compulsions, whereas in TS with comorbid OCD (TS+OCD) patients have more intrusive violent and sexual images/thoughts, hoarding behaviors, counting and symmetry searching rituals, touching behaviors, “just right” feelings, echophenomena and self-injurious behaviors. There may also be more mental rituals in TS+OCD than in pure OCD [11,18]. Thus, according to phenomenology, although tic-related OCD presents intermediate symptoms, it seems that it is more closely related to tic-free OCD than to TS, whereas TS+OCD is more similar to TS than OCD [12,15,22].

The presence of touching behaviors and echophenomena seems to better discriminate between TS and OCD [8,15].

Although there are phenomenological differences between OCD with and without tics, they do not seem to influence the severity of OCS [14,16], which is also not related to the presence of sensory phenomena [23].

When symptoms are measured based on dimensions, two OCD symptom dimensions have been more frequently associated with tic-related OCD: the aggressive/sexual/religious and the symmetry/ordering/arranging dimensions [18].

It has also been suggested in the literature that OCS in TS changes in character with age, with younger patients exhibiting more difficulties related to impulse control and older patients being more concerned with checking, arranging and fear of contamination [24,25], closer to the symptoms of non tic-related OCD. Rizzo et al (2012) reported that almost half of the children with pure TS at onset changed their phenotype to TS+OCD after 10 years of follow-up [4]. Together these findings raise the intriguing question of whether OCD could actually reflect a more neurodevelopmentally mature manifestation of the same condition diagnosed as TS in some (often younger) patients [8].

**Sensory Phenomena**

Sensory phenomena consisting of mental or bodily sensations preceding the compulsions in the absence of any thought, fear or worry were first described in TS patients. Different terms have been used in the literature to describe these subjective experiences, including energy release, pressure, “just-right” perceptions, premonitory urges and other mental sensations [26]. It has been shown that up to 93% of patients with TS can report premonitory urges, whether local or generalized with regard to body location, with a trend towards increased premonitory urges’ awareness with increased age through adolescence [26]. However, several OCD patients have described that their compulsions are preceded not by obsessions but by subjective experiences that are compatible with the descriptions of sensory phenomena. These phenomena, which have been initially described in TS, have now been also associated with OCD [23], especially tic-related OCD [16,18], although some authors consider that these urges have a cognitive origin in OCD whereas they are originated peripherally in TS [11].

When OCD presents sensory phenomena, often there is higher frequency of symmetry/ordering/arranging and contamination/washing OCS dimensions, higher frequency of comorbid TS, more common family history of tic disorders, and lower levels of insight [23]. Thus, according to the presence of sensory phenomena, tic-related OCD is closer to TS than to tic-free OCD.

**Treatment response**

The phenomenological distinction of repetitive behaviors in patients with TS is important to consider, since the treatment of tics and OCS differs [12].

According to treatment response, TS+OCD and tic-related OCD respond differently to Selective Serotonin Reuptake Inhibitors (SSRIs) when compared to non tic-related OCD. While “Tic-like” repetitive behaviors respond to dopamine antagonists and α2-receptors agonists (clonidine), “OCD-like” repetitive behaviors improve under serotoninergic agents. As the use of antipsychotics in monotherapy does not control obsessive symptoms and the single use of SSRIs may worsen tics, the addition of antipsychotics to SSRIs is particularly useful in patients with TS+OCD and tic-related OCD [12,27,28]. Studies evaluating the augmentation of SSRIs with antipsychotics for treatment of refractory OCD suggest that OCD patients with comorbid tics on an SSRI may have better response to this intervention [6,18]. These data may support, from a pharmacological perspective, the existence of a continuum between OCD and TS. They also show the importance of knowing whether tics, or obsessions and compulsions, are the primary target of treatment, as this will affect the selection of pharmacological agents.

Both TS and OCD might also improve with Cognitive-Behavioral Therapy, but with slight differences in the behavioral strategies and approaches: while exposure-based cognitive behavioral therapy is useful in OCS, Habit Reversal Training (HRT) is directed to tics [29]. Thus, regarding treatment response, tic-related OCD is closer to TS than to tic-free OCD.
To date, pharmacotherapy has been considered the treatment of choice for TS with several randomized trials documenting the efficacy of haloperidol, pimozide, risperidone and clonidine. However, many patients refuse or discontinue medication because of intolerable side effects, while others are unresponsive to medication or suffer from residual tics despite pharmacotherapy. As a result, a behavioral treatment, HRT, has become increasingly popular, mainly as a non-pharmacologic adjunct to pharmacological treatments. Briefly, HRT involves training patients to respond to uncomfortable sensations preceding tics with voluntary behavior that is physically incompatible with tics. This means that patients learn to apply antagonistic, competing movements to inhibit the occurrence of tics. Competing response training involves implementation of contingent movements/behaviors on early sensations/urges preceding a tic that are incompatible with its execution, thereby preventing their occurrence. For this reason, the sensory component of tics is an essential element of HRT, as gaining awareness and describing it is a key procedure in this behavioral treatment. The trained responses, that aim to interrupt the tic execution cycle, function as negative reinforcements and thereby reduce the probability of future recurrence of the tic [6,30,31].

Brain stimulation techniques, including transcranial and deep brain stimulation, constitute new approaches. Although the efficacy of repetitive transcranial magnetic stimulation (rTMS) has not been proven yet, some studies have reached promising results with this non-invasive brain stimulation, as low-frequency (1Hz) rTMS to the supplemental motor area may correct abnormal cortical excitability in TS patients and improve symptoms [32]. Further studies using rTMS are needed in order to recognize it as an useful therapeutic tool in the future.

**Chronic Motor/Vocal Tics**

Although comorbid OCD is more frequent in TS than in other tic disorders [6], phenomenological differences in OCD phenotype have been detected according to the presence of either TS or Chronic Motor-Vocal Tics (CMVT) [16].

CMVT is characterized by the presence of either motor or vocal tics, but not both, for a period of more than one year [3], having a prevalence of approximately 5% in children [6]. Patients with OCD with comorbid CMVT present clinical characteristics that are intermediate between those of tic-related OCD and TS+OCD according to age at onset of OCS, frequency of sensory phenomena, bodily sensations and "just-right" perceptions and clinical characteristics. From a phenomenological point of view, these results provide further evidence of a possible continuum between OCD and TS, where patients with OCD plus tics have intermediate phenotypic features [16,18].

Since several genetic studies have been unsuccessful in identifying the most likely mode of transmission of OCD, the hypothesis that gene are added to produce a cumulative effect seems plausible. Therefore, from a quantitative point of view, some authors speculate that the degree of genetic loading is higher for the disorders at the poles of this spectrum: OCD and TS+OCD, whereas patients with OCD+CMVT fall in between. Supporting this hypothesis there are recent data pointing towards earlier age at onset of OCS in the TS pole, and the earlier the age at onset, the higher the genetic load in OCD [11,16]. But not all studies support these data: Lewin et al. (2010) found no significant differences in the onset age of OCS between the OCD and OCD with comorbid tic disorder groups [14].

**Conclusion**

Several authors have pointed out the similarities and differences between OCD and TS, which are frequently comorbid, supporting the growing evidence that these two entities are neurobiologically and genetically related.

There are clinical differences in OCD patients according to the presence of tics, CMVT or TS, presenting the phenotype OCD with comorbid CMVT intermediate characteristics compared to the other disorders, probably reflecting the different number and type of genes involved in the several phenotypes of tic disorders [15]. The frequent comorbidity between OCD and tic disorders, mainly in the pediatric population, has opened the way to the discussion about a possible inclusion of a new tic-related OCD subgroup in the future diagnostic classifications.

The term RB refers to broad and often disparate classes of behavior linked by repetition, rigidity, invariance and inappropriateness. RB are characteristic of many psychiatric disorders, as well as of stages of typical development.

Two types of RB can be found in TS: "Tic-like", which are very likely an integral part of TS and "OCD-like", which can be considered as a comorbid condition of TS. When TS patients present "OCD-like" RB, generally they present more complex and severe tics, better response to SSRIs and antipsychotic drugs and less successful socio-professional adaptation [12]. Thus, a meticulous semiological analysis of RB in TS patients will help adjusting treatment and predict prognosis.

Attempts to distinguish between OCD and TS based on the specific types of RB presented are not without limitations, as categorization will depend, among other factors, on the awareness of the compulsive experience which will vary according to the individual's own subjective nature of interpretation. This in turn could be affected by other factors that are rarely considered in RB research studies, such as personality, intelligence and locus of control, and that can potentially influence results when administering self-assessment measures. Also, evaluation of the literature pertaining to the phenomenological experience of OCD and TS has its own limitations as different studies categorize patients differently: for example, some included TS+OCD and tic-related OCD in the same group, while
others did not differentiate TS+OCD from TS with comorbid subclinical OCS. These diverse methodologies reflect the difficulties in RB categorization. Overall, however, it seems that contamination worries are probably indicative of OCD, and RB linked to echophenomena, touching, symmetry and counting may be most common in TS. It may be of use also to note that while patients with OCD seem to be focused on avoiding harm as much as possible, patients with TS often display RB which are ironically self-defeating, dangerous (e.g. touching very hot or sharp objects) and socially inappropriate (e.g. coprolalia) [8].

A further difficulty with assessing RB includes the dynamic of the phenomenological experience. That is, these symptoms can change in nature over time, perhaps as the RB become more automated, or as continued reflection allows the subject to become more aware of any preceding cognitions or sensory cues. This could help explain why the report of RB can change with age and the finding that changes in diagnosis may occur over time, being tics more often diagnosed in children and OCD more often seen in adults [4,8]. Furthermore, corticostriatal circuits have a protracted development trajectory and neurobiological changes associated with RB may change over development.

Finally, TS is characterized by different clinical phenotypes at the onset that are not always stable over the years and that mean different severity of symptoms, response to the pharmacological treatment and impairment in quality of life. Further research focusing the response of different RB to SSRIs, anti-dopaminergic medications and other interventions will be of great value in tailoring interventions to the needs of individual patients within the obsessive-compulsive spectrum [4,7].

Abreviations

CMVT: Chronic motor-vocal tics; HRT: Habit reversal training; OCD: Obsessive-compulsive disorder; OCS: Obsessive-compulsive symptoms; OCSD: Obsessive-compulsive spectrum disorders; rTMS: Repetitive transcranial magnetic stimulation; RB: Repetitive behaviors; SSRIs: Selective Serotonin Reuptake Inhibitors; TS: Tourette Syndrome

Competing interests

The authors declare no conflict of interest.

References


