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Review

The psychology of Tourette disorder: Revisiting the past and moving toward a cognitively-oriented future

Jean-Philippe Gagné*

Department of Psychology, Concordia University, 7141 Sherbrooke St. West, Montreal, QC H4B 1R6, Canada

HIGHLIGHTS

- Chronic tics are influenced by both neurobiological and psychological processes.
- Research supports that emotional states and operant conditioning shape tics.
- Cognitive factors involved in Tourette syndrome are not well understood.
- Negative appraisals of aversive sensations may play a role in Tourette syndrome.
- It is proposed that negative beliefs about discomfort underlie such appraisals.

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ABSTRACT

Tourette syndrome is a neurodevelopmental disorder characterized by chronic tics (i.e., repetitive and stereotyped movements and vocalizations) and premonitory urges (i.e., aversive sensations preceding tics that are alleviated once a tic is performed). Research supports that dysfunctional neurobiological and psychological processes interact and contribute to the development and maintenance of tics. However, psychological theories of Tourette syndrome and accompanying research have mainly focused on the emotional states (e.g., anxiety and frustration) and behavioural principles (i.e., operant conditioning) that play a role in tic exacerbation. This selective review summarizes key discoveries pertaining to the emotional and behavioural aspects of Tourette syndrome but also proposes a more comprehensive, cognitively-oriented conceptualization of the disorder. Specifically, it is proposed that maladaptive beliefs about discomfort and about one's ability to cope with discomfort underlie negative appraisals of unpleasant sensory experiences in individuals with Tourette syndrome. It is further suggested that these beliefs lead individuals to perceive premonitory urges in a catastrophic manner and thereby enhance tic frequency. Concrete research avenues to empirically examine these hypotheses are outlined and clinical implications for the field of cognitive-behaviour therapy are discussed.

1. Introduction

Although it is now well established that both neurobiological and psychological factors contribute to the occurrence and exacerbation of tics (e.g., Woods, Piacentini, & Walkup, 2007), previous explanatory models used to neglect this important interaction. In 1885, Gilles de la Tourette published some of the first reports on tics—in which he described patients struggling with rapid and involuntary motor movements—and hypothesized that those symptoms stemmed from a hereditary and biologically based disorder (Lajonchere, Nortz, & Finger, 1996). Ironically, this relatively accurate theory of Tourette syndrome (TS) was quickly replaced by psychoanalytic explanations, partly due to the popularity of this therapeutic orientation in treating hysteria and to the lack of effective neurological treatments at the time (e.g., Ferenczi, 1921; Kushner, 1999). However, in the 1960s, critical biological discoveries, such as the finding that antipsychotic medication reduces tic frequency, led to the expansion of neurobiological conceptualizations of TS (Kushner, 1999).

Over the last decades, psychology has reintegrated models of the syndrome, with research demonstrating that emotional states (e.g., anxiety) as well as behavioural principles (i.e., operant conditioning) impact tic expression (e.g., Woods & Himle, 2004). In this way, behaviour therapy, either alone or in combination with medication, has been shown to be an effective treatment for TS (e.g., Piacentini et al., 2010). Nevertheless, cognitive-behaviour theory claims that cognitive factors directly influence behavioural outcomes (e.g., Beck, 2011), such that

* Corresponding author.
E-mail address: jean_ga@live.concordia.ca.

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the role of cognition may have been neglected in explanatory models of TS. In support of this idea, O'Connor (2002, 2005) and, more recently, Robinson and Hedderly (2016) suggested that negative *appraisals* of unpleasant body sensations contribute to the exacerbation of tics.

In this selective review, key neurobiological and psychological findings pertaining to TS are first outlined as a way to ultimately propose a more holistic, cognitively-oriented formulation of the syndrome. Specifically, it is suggested that investigating potential maladaptive beliefs underlying negative appraisals of unpleasant sensory phenomena, such as negative beliefs about discomfort, could increase our understanding of the maintaining factors of TS and lead to more comprehensive psychological treatments for this debilitating problem.

2. Tourette syndrome

TS (now "Tourette disorder" in the DSM-5) is categorized as a neurodevelopmental disorder and is defined by motor and vocal tics (American Psychiatric Association, 2013). Tics are sudden, repetitive, and stereotyped movements or vocalizations that are often experienced as being outside voluntary control (Leckman, King, & Cohen, 1999). Simple motor tics involve isolated muscle groups (e.g., head jerking and excessive eye blinking), whereas complex motor tics involve multiple muscle groups and sometimes mimic volitional behaviour (e.g., touching objects and hand gestures). By contrast, simple vocal tics are usually single sounds (e.g., excessive throat clearing), whereas complex vocal tics include multiple syllables, words, and phrases (e.g., repeating one's own words or others' words). A core characteristic of tics is that they are preceded by a *premonitory urge*, an intrusive and localized sensation of inner tension and discomfort (e.g., Woods, Piacentini, Himle, & Chang, 2005). When a tic is performed (and often it has to be performed the 'right' way), the urge fades out or is completely eliminated (e.g., Himle, Woods, Piacentini, & Walkup, 2006).

The onset of TS is typically between 6 and 7 years old (Freeman et al., 2000) and its severity peaks during early adolescence (Leckman et al., 1998). First symptoms often include simple motor tics and, between 8 and 12 years old, complex and vocal tics emerge (Leckman et al., 1998). An important aspect of TS is that tics are unpredictable: they naturally wax and wane in type and intensity (Peterson & Leckman, 1998). Despite this fluctuation, longitudinal studies assert that impairment associated with tics decreases when individuals enter young adulthood (e.g., Coffey et al., 2004). Still, approximately 25% of youths with TS experience moderate to severe tics in adulthood (Leckman et al., 1998).

Together, the prevalence of TS and persistent motor or vocal tic disorder ranges from 1 to 2% in community samples (Scahill, Sukhodolsky, Williams, & Leckman, 2005). Epidemiological studies show that these tic disorders are more common in males than females, with a ratio of 5:1 (e.g., Freeman et al., 2000). Children with TS receive, on average, two other psychiatric diagnoses (Freeman et al., 2000), with attention-deficit/hyperactivity disorder (ADHD) and obsessive-compulsive disorder (OCD) being the most frequent ones (Zohar et al., 1999). Research has shown that half of those diagnosed with TS experience subclinical obsessions and compulsions and that 23% of individuals with TS meet full diagnostic criteria for OCD (e.g., Caine et al., 1988; Zohar et al., 1999). OCD symptoms usually emerge between 11 and 12 years old in youths struggling with TS (Leckman et al., 1998). Although perceived quality of life is lower in children and adolescents with chronic tics (versus healthy controls), youths who also suffer from comorbid disorders (e.g., ADHD and OCD) and/or elevated tic severity report even poorer quality of life across several domains (e.g., social relationships) as well as depressive symptoms (e.g., Cutler, Murphy, Gilmour, & Heyman, 2009; Eddy et al., 2011, 2012). Qualitative research has revealed that impairment and poor quality of life in TS mainly comes from youths' desire to control tics, society's expectations of normal behaviour, and the overall distress associated with tics and premonitory urges (Cutler et al., 2009).

Over the last decade, preliminary findings have suggested that individuals with TS display slight impairments in social cognition, the cognitive processes used in social interactions that allow adequate functioning (e.g., empathy, facial recognition, emotion perception; Beauchamp & Anderson, 2010). For instance, youths with TS seem to have poorer ability to engage in emotionally appropriate reciprocal social interactions, as compared to those in the general population (Darrow et al., 2017; Güler, Berkem, Yazgan, & Kalaça, 2015). However, these deficits in social reciprocity and responsiveness may be attributable to comorbid disorders (Morand-Beaulieu et al., 2017), as they are most strongly associated with comorbid TS and OCD (Darrow et al., 2017). In an adult sample of patients with TS, Eddy and Cavanna (2013) identified small deficits in interpretations of facial expressions, but a previous study had found similar results between adults with and without TS on a very similar task (Baron-Cohen, Jolliffe, Mortimore, & Robertson, 1997). Likewise, Eddy, Macerollo, Martino, and Cavanna (2015) demonstrated that adults with TS (who did not have any other disorders) had a lower tendency than healthy controls to take others' perspective in everyday situations, but previous research by Channon, Sinclair, Waller, Healey, and Robertson (2004) had shown similar cognitive and emotional empathy levels between adults with TS and healthy controls. Contradictory results are likely due to small sample sizes and the lack of research with younger populations (Morand-Beaulieu, Leclerc, et al., 2017). Despite these inconsistencies, a slight deficit in social cognition—especially for those with a comorbid disorder—appears to emerge from this recent literature (Morand-Beaulieu, Leclerc, et al., 2017).

As mentioned above, a number of theoretical models has been put forward to explain the development and maintenance of tics. Although Gilles de la Tourette first mentioned in his reports that biological processes and genetics were at the root of the problem (Lajonchere et al., 1996), psychoanalytical theorists posited that unconscious aggressive and sexual impulses were causing its emergence (Ferenczi, 1921; Kushner, 1999, 2000). Later, basic research on neurotransmitters (e.g., dopamine) and brain structures (e.g., basal ganglia) improved our understanding of motor control and movement processes (Kushner, 1999). These fundamental discoveries, combined with advancements in genetics and pharmacology (e.g., dopamine antagonists), allowed the refinement of neurobiological models of TS (Kushner, 1999).

3. Neurobiological model

Family and twin studies have confirmed that TS is hereditary (O'Rourke, Scharf, Yu, & Pauls, 2009). For instance, first-degree relatives of individuals with TS have a 5- to 15-fold increased risk of having the disorder, as compared to rates in the general population (e.g., Scharf & Pauls, 2007). Concordance rates of tic disorders are also consistently higher in monozygotic (77%) as opposed to dizygotic (23%) twins (Price, Kidd, Cohen, Pauls, & Leckman, 1985). Similarly, in 25 to 41% of families with TS, both the parents and children experience tics frequently (Hanna, Janjua, Contant, & Jankovic, 1999; Lichter, Dmochowski, Jackson, & Trinidad, 1999). Despite such a clear hereditary component, identifying candidate genes has been particularly challenging for researchers (Scharf et al., 2013) and results are rarely replicated by independent cohorts (Pagliaroli, Vető, Arányi, & Barta, 2016). Structural and copy-number variations in susceptibility genes (e.g., *SLITRK1*, *CNTNAP2*, *HDC*, *NRXN1*, *CNTN6*) have only been identified in very few individuals or single families and may thus be rare instances of causative genetic factors (e.g., Huang et al., 2017; Scharf et al., 2013). In the first genome-wide association study of TS with individuals of European ancestry and Latin American populations, no genetic markers reached a genome-wide significance threshold (although chromosome 9q32 in the *COL27A1* gene emerged as the top signal for both the European and Latin American samples; Scharf et al., 2013). Further, O'Rourke and colleagues explained that the genetic underpinnings of TS overlap with those of frequently comorbid disorders. For example, Pauls, Towbin, Leckman, Zahner, and Cohen (1986) found increased rates of OCD without tics in relatives of individuals with TS without OCD, revealing the complexity of the inheritance of TS and related problems. Moreover, because there is some discordance between monozygotic twins, prenatal (e.g., alcohol abuse, smoking) and perinatal (e.g., streptococcal infection, complicated birth) environmental factors are proposed to play a role in the development of TS via epigenetic mechanisms, such as DNA methylation (e.g., Pagliaroli et al., 2016).

Given these genetic risk factors, it is not surprising that neurobiological abnormalities have been identified in patients with TS. For example, Peterson et al. (2003) found that individuals with TS have a mild (but consistent) volume reduction of the basal ganglia, a region of the brain dedicated to selecting and inhibiting competing motor patterns (i.e., 'gating' functions). Of note, Peterson and colleagues suggested that this reduction may also arise from behavioural (i.e., non-genetic) factors, such as repeated tic suppression. Other groups have also identified decreased volume and/or asymmetry of the caudate nucleus and putamen—structures of the basal ganglia—in both children and adults with the disorder (e.g., Hyde et al., 1995; Moriarty et al., 1997). Using a longitudinal design, Bloch, Leckman, Zhu, and Peterson (2005) demonstrated that a smaller volume of the caudate nucleus in childhood predicted higher tic severity in adulthood. Dopamine—a neurotransmitter involved in the activation of the striatum, the primary input to the basal ganglia—appears to play a key role in TS. Dopamine antagonists have been shown to decrease tic severity (e.g., Roessner et al., 2013), whereas dopamine agonists cause the opposite effect (e.g., Shale, Fahn, & Mayeux, 1986). Further, Steeves et al. (2010) found that amphetamine, a dopamine releaser, led to a significantly greater widespread activation in several brain areas related to motor output, including the striatum, in individuals with TS (versus healthy controls). Therefore, it has been hypothesized that tics may be characterized by an excessive dopamine release in the basal ganglia (e.g., Buse, Schoenefeld, Munchau, & Roessner, 2013).

The specific neurobiological mechanisms underlying tic expression are also currently unknown. However, contemporary theories attribute TS to a lack of inhibitory tone in the striatum or, in order words, to "deficits in gating functions" (Godar & Bortolato, 2017, p. 124). Specifically, neuroimaging studies support that tics and premonitory urges stem from a dysfunctional activation of the cortico-striatal-thalamic-cortical pathway, a brain circuitry that integrates motor and sensory functions (e.g., Ganos, Roessner, & Münchau, 2013; Mink, 2001; see Albin & Mink, 2006 and Godar & Bortolato, 2017 for a full discussion on the neurobiology of TS). First, premonitory urges are proposed to emerge from a hyperactivation of *cortical* areas, including the insular cortex, supplementary motor area, anterior cingulate cortex, and parietal operculum (e.g., Bohlhalter et al., 2006; Neuner et al., 2014; Wang et al., 2011; Worbe et al., 2014), possibly due to a lack of inhibitory (i.e., GABAergic) interneurons (Vaccarino, Kataoka-Sasaki, & Lennington, 2013). Indeed, research has shown that individuals with TS have reduced GABAergic interneurons in the insular cortex and that low GABA content in the somatosensory cortex predicts elevated tic severity (Puts et al., 2015; Tinaz et al., 2014). Interestingly, premonitory urges are also associated with a high activation of the amygdala—a brain region dedicated to emotional responses—likely because of the aversive nature of such urges (Wang et al., 2011). Second, it is posited that this hyperactivation of cortical areas and of the amygdala (i.e., neurobiological correlates of premonitory urges) leads to an excessive dopamine release to the *striatum* (Neuner et al., 2014), which causes a discrete set of striatal neurons to become inappropriately overactive—an "aberrant focus" (Albin & Mink, 2006; Mink, 2001). Third, this aberrant focus in the dorsal striatum inappropriately inhibits basal ganglia output neurons in the globus pallidus pars interna and substantia nigra pars reticulata, which in turn disinhibits output neurons in the *thalamus* and *primary motor cortex* (Albin & Mink, 2006; Mink, 2001; Neuner et al., 2014). Essentially, a dysfunctional activation of the striatum prevents the basal ganglia from inhibiting or 'braking' involuntary movements. Fourth, it is hypothesized that excessive dopamine release in several areas of the cortico-striatal-thalamic-cortical loop contributes to the reinforcement of tics (e.g., Albin & Mink, 2006; Godar & Bortolato, 2017). This theory is consistent with research showing that habit formation, a normal learning process involving dopamine, is hyperactive in those with TS (Delorme et al., 2015).

Although inhibitory deficits are at the core of these neurobiological theories, there are important inconsistencies in the literature regarding whether individuals with TS actually display inhibitory control impairments on several executive functioning tasks (Morand-Beaulieu et al., 2017). As an illustration, some authors found impaired performance (i.e., pressing a key to a "No-Go" stimulus) on the Go/No-Go task for participants with TS (Goudriaan, Oosterlaan, de Beurs, & van den Brink, 2005), whereas many lines of research demonstrated no significant differences with healthy controls (Delorme et al., 2015; Draper, Jude, Jackson, & Jackson, 2015; Serrien, Orth, Evans, Lees, & Brown, 2005). Contradictory findings have been found using a number of other neuropsychological tests, such as the Stroop test, with either impaired (e.g., Chang, McCracken, & Piacentini, 2007), unaffected (e.g., Lavoie, Thibault, Stip, & O'Connor, 2007), or even enhanced (Thibault, O'Connor, Stip, & Lavoie, 2009) performance for individuals with TS. Such inconsistencies are often attributed to comorbidity and psychiatric medication (Morand-Beaulieu, Grot, et al., 2017), although assessment methods, age, gender, and history of therapy and deep-brain stimulation are other potential reasons (Morand-Beaulieu, Leclerc, et al., 2017). In a recent meta-analysis, Morand-Beaulieu and colleagues (2017a) attempted to shed light on these inconsistencies and found a small-to-medium effect toward inhibitory control impairments in individuals with TS (versus healthy controls). However, those with TS *and* ADHD (i.e., comorbidity) displayed significantly greater inhibitory deficits than those with 'pure' TS. The authors further showed that inhibitory impairments tended to be more important in studies including medicated samples, which is likely due to higher symptom severity. Still, specific medications appear to impact one's performance on a task in different ways, with pimozide being associated with lower errors on the Go/No-Go task and haloperidol being associated with more errors, for example (Sallee, Sethuraman, & Rock 1994).

Despite some empirical evidence supporting inhibitory control deficits in TS, proposed neurobiological models cannot adequately explain why tics are quite variable and unpredictable. To account for this symptom fluctuation, Mink (2001) proposed that, *under various circumstances and influences*, different clusters of striatal neurons become inappropriately overactive, such that different involuntary movements (i.e., tics) are produced. In this way, researchers hypothesized that environmental and psychological factors interact with neurobiological processes to shape tic expression.

4. Psychological Factors

To study psychological and environmental factors influencing tic occurrence, researchers have focused on two types of variables: antecedents and consequences (Himle et al., 2014). On the one hand, antecedents are internal and/or external cues, events, and feelings that immediately precede a tic, altering the likelihood that the tic occurs (Conelea & Woods, 2008). On the other hand, consequences are internal and/or external outcomes that occur immediately after a tic, making that specific tic more or less likely to reoccur (Conelea & Woods, 2008).

Initially, researchers used self-report methodology to investigate antecedent factors, which mainly allowed them to gather descriptive information (Conelea & Woods, 2008). For instance, in a study by Bornstein, Stefl, and Hammond (1990), 98.2% of participants reported that anxiety and stress exacerbate the frequency of their tics, and this finding has been replicated in several studies (e.g., Eapen, Fox-Hiley, Banerjee, & Robertson, 2004; Silva, Munoz, Barickman, & Friedhoff, 1995). Conversely, participants reported that relaxation and other passive states helped with tic reduction (e.g., Eapen et al., 2004; Robertson, Banerjee, Eapen, & Fox-Hiley, 2002). Other frequently reported *internal* antecedents included fatigue, boredom, frustration, and excitement (e.g., Eapen et al., 2004; Silva et al., 1995). Individuals with TS and their parents were also asked to elaborate on *external* tic-exacerbating antecedents. Common responses included returning to school, holidays and birthdays, social gatherings, watching television, and being alone (e.g., Bornstein et al., 1990; Silva et al., 1995). Therefore, internal antecedents seem to primarily relate to different *emotional states*, such as anxiety and excitement. Likewise, external antecedents appear to be the situations in which these emotions are experienced (e.g., anxiety when returning to school and excitement during holidays). Of note, these lines of descriptive research also highlight the idiosyncratic nature of situations as some common tic-exacerbating events (e.g., social gatherings and doctor's office visits) were reportedly tic-alleviating for other youths (e.g., Himle et al., 2014).

Self-reports and parental reports were further utilized to collect qualitative data on consequences (Conelea & Woods, 2008). The most frequently reported *internal* consequence of tic expression is the removal of the unpleasant premonitory urge (Kwak, Vuong, & Jankovic, 2003). Typically, *external* consequences include negative attention (e.g., being teased by peers), positive attention (e.g., being comforted by parents), gaining access to situations (e.g., television time to relax) and rewards (e.g., ice cream for excessive throat clearing), and leaving stressful situations (e.g., Packer, 2005). Accordingly, *behavioural principles* stemming from operant conditioning appear to play a critical role in tic reoccurrence. Specifically, external consequences such as attention may be positively reinforcing tics, whereas the internal consequence of removing the premonitory urge may be negatively reinforcing tics. These contingencies are thus proposed to maintain tics and shape them in form and frequency (e.g., Himle et al., 2006).

Of course, self-report methodology does not allow researchers to draw conclusions regarding causality and mechanisms of action (Conelea & Woods, 2008), and biases can often influence responding in interviews and surveys (e.g., Kazdin, 2003). These descriptive data were used to inform the creation of more controlled designs (e.g., experiments, daily diaries, animal models) and better explore the emotional and behavioural aspects of TS.

4.1. Emotional aspect

Anxiety and stress are currently the emotional states that have been the most empirically examined in relation to TS. For example, in a sample of children with TS and OCD, Findley et al. (2003) found a positive correlation between daily life stressors and tic severity. Hoekstra, Steenhuis, Kallenberg, and Minderaa (2004) replicated this result in a sample of adults with TS who were asked to record, on a weekly basis, the number of stressful life events they had experienced and their tic severity. Lin et al. (2007) investigated this same relationship longitudinally and demonstrated that *current* stress level predicted *future* tic severity. Still, the mechanism through which anxiety and stress exert their effect on tics is unknown. Current neurobiological models suggest that such negative feelings further stimulate the already overactive amygdala (i.e., the brain region associated with both fear and premonitory urges), triggering aberrant foci in the striatum (e.g., Godar & Bortolato, 2017; Wang et al., 2011). In line with this theory, research has shown that elevated anxiety symptoms are associated with higher scores on a measure of premonitory urges (Rozenman et al., 2015). Specifically, panic and somatic symptoms (e.g., feelings of dizziness, shakiness, and nausea) were tightly linked with premonitory urge severity, indicating that youths with TS may pay selective attention to unpleasant sensory phenomena in general.

O'Connor, Gareau, and Blowers (1994) also investigated the role of emotion in TS. They asked participants to complete a daily diary in which they had to identify the emotions they were experiencing in high-risk situations (i.e., elevated tic frequency) and frustration emerged as the main predictor. The role of frustration in the occurrence of tics was also examined using animal models. Waitt and Buchanan-Smith (2001) modified monkeys' food delivery schedule, a paradigm known to induce frustration (e.g., Bassett & Buchanan-Smith, 2007), and observed an increase in spontaneous repetitive behaviour. Godar and Bortolato (2017) used a similar paradigm in mice (that were already engaging in tic-like behaviour due to a genetic intervention) and observed exacerbation of tic-like responses. Neuroimaging studies have shown that frustration is associated with similar brain activity patterns as anxiety and acute stress (e.g., Yu, Mobbs, Seymour, Rowe, & Calder, 2014), such that similar neurobiological mechanisms might be at play with these several emotions.

Other states have been given less attention in the tic literature but are still relevant to examine. Cohrs et al. (2001) investigated sleep quality in patients with TS by using polysomnography and video monitoring and demonstrated a positive association between number of awakenings at night and tic severity during the day, as well as a negative association between sleep efficiency and tic severity. Although causal relationships have not yet been established, it is posited that fatigue makes individuals more susceptible to experiencing frustration, anxiety, stress, and irritability (e.g., Aniţei, Chraif, & Liliana, 2013), which then enhances tic severity as explained above (e.g., Cohrs et al., 2001; Godar & Bortolato, 2017). Still, coping with tics during the day may lead to a state of hyperarousal, thereby maintaining sleep disturbance and the negative effects of fatigue on tics (Cohrs et al., 2001).

Additionally, situations resulting in overstimulation (e.g., excitement) and understimulation (e.g., boredom) are proposed to enhance tic occurrence (e.g., Barnea et al., 2016; Silva et al., 1995). Regarding overstimulation, it is hypothesized that information overload might exacerbate 'gating' deficits, which are already impaired in individuals with TS (Godar & Bortolato, 2017). Indeed, there is some evidence showing that sensory integration may lower filtering functions in humans (e.g., Marsh, Hoffman, & Stitt, 1976). Finally, boredom may be problematic for those struggling with chronic tics: low stimulation may encourage one to shift their attention inward and focus on interoceptive and other sensory phenomena, such as premonitory urges (e.g., Bench & Lench, 2013).

Although these lines of research provide support for a link between key emotional states and tic exacerbation, descriptive research reminds us that these several emotions are experienced in idiosyncratic situations (e.g., Himle et al., 2014). As such, one's positive or negative *evaluation* of a given situation likely drives the relationship between emotions and tic manifestation (e.g., O'Connor et al., 2009). In other words, emotional states may trigger neurobiological mechanisms involved in tic expression but one's idiosyncratic cognitions may underlie emotional and physiological responses in the first place. For instance, Wood et al. (2003) had children with TS watch sequences from movies and experimentally manipulated the type of scenes they were watching based on the emotional state primarily associated with the scenes. They found that tic severity was higher during "anticipation" as opposed to "happiness" scenes, but that overall tic severity was actually at its highest before the movie started. Although it is unclear which scenes/situations elicited which emotional states in participants, results suggest that tic frequency can be shifted in the laboratory based on various emotions that are triggered by individuals' perception of a situation.

4.2. Behavioural aspect

Self-reports, parental reports, and even direct observation led researchers to believe that behavioural principles are a core component of psychological conceptualizations of TS. For example, in 1973, Rosen and Wesner (1973) conducted a single-case experiment with a 12-year-old male. The authors illuminated a light in the therapy room after every 30-s interval in which the youth had not performed a tic. At the end of therapy, he was given a candy for each time the light had been turned on. The intervention was then brought to his classroom: all students were given a piece of candy for every 30-s tic-free interval. Tics decreased to near-zero levels and the authors concluded that tics could be shifted via operant conditioning. Similarly, Watson and Sterling (1998) focused on the case of a 4-year-old female who had a vocal tic—excessive coughing—that mainly occurred at the dinner table. A functional analysis indicated that parental attention during meal time was maintaining the tic. The parents were asked to stop providing attention when the child was coughing and to instead provide attention when the child was *not* coughing. This intervention led to a clear decrease in coughing and the tic eventually faded out.

However, more controlled designs, especially laboratory experiments, are essential to further support the reinforcement hypothesis. In 2004, Woods and Himle told four children with TS that a 'tic detector' was standing in front of them. During the baseline period, participants were instructed to behave normally and to freely engage in tics. During the verbal instructions period, participants were asked to suppress their tics in any way possible. Finally, during the reinforcement period, participants were instructed to suppress their tics and were told that the 'tic detector' would give them a token (to be later exchanged for money) for every 10-s tic-free interval. The authors observed higher tic suppression during the reinforcement period (76.3% reduction rate as compared to baseline) than during the verbal instructions period (10.3% reduction rate as compared to baseline), indicating that tic occurrence could be modified by using positive reinforcement. Himle, Woods, and Bunaciu (2008) expanded these findings by showing that reinforcers had to be delivered contingent upon tic suppression to cause maximal tic reduction, such that tokens delivered based on a fixed time schedule (regardless of tic suppression performance) did not lower tic frequency to the same extent. Recently, it was demonstrated that even mild punishment (i.e., removing tokens) could enhance tic suppression (Capriotti, Brandt, Ricketts, Espil, & Woods, 2012). Of note, experiments have confirmed that reinforcing tic suppression with these strategies does not lead to a rebound effect post-suppression (e.g., Capriotti et al., 2012; Himle & Woods, 2005; Woods et al., 2008).

That being said, *the* most cited internal reward following tic occurrence is the removal of the aversive and unpleasant premonitory urge (Kwak et al., 2003). According to self-reports, the urge fades out immediately after a tic is performed (e.g., Himle et al., 2006), such that negative reinforcement likely plays an important role in the maintenance of tics (e.g., Woods et al., 2005). Himle, Woods, Conelea, Bauer, and Rice (2007) examined this possibility in the laboratory. During baseline periods, five children and adolescents were instructed to freely engage in tics; during reinforcement periods, they were all told that they would receive a token for every 10-second tic-free interval. Every 30 s, the severity of participants' premonitory urges was measured. The authors found that subjective urge ratings were higher during reinforcement periods (i.e., when actively suppressing tics), as opposed to during baseline periods (except for one participant who reported similar urge ratings across conditions). Although this study is largely limited in terms of sample size, the authors provided preliminary experimental support for the negative reinforcement theory, such that results are, for most participants, in line with the assertion that freely ticcing relieves aversive sensory phenomena.

Lastly, Woods, Walther, Bauer, Kemp, and Conelea (2009) designed a laboratory experiment to shed light on a phenomenon commonly observed in TS: tic variability across contexts. The authors hypothesized that a history of reinforcers in specific contexts could influence tic expression/suppression at a later time. In this study, ten children with chronic tics underwent four training sessions. All training sessions contained the same three counterbalanced conditions. In the reinforcement condition, participants were instructed to suppress their tics and were told that, for every 10-s tic-free interval, they would get a token; a purple light was illuminated as the instructions were provided. In the no reinforcement condition, participants were asked to suppress their tics, but no reinforcement was provided; an orange light was associated with this condition. In the control condition, participants were instructed to freely engage in tics; both lights were turned off. Following the four training sessions, three experimental tests took place randomly for all participants: in one condition, a purple light was illuminated; in a second condition, an orange light was illuminated; in a third condition, neither light was illuminated. Across the three experimental tests, instructions to suppress and reinforcers were *not* provided at any point. The authors found that tic expression was significantly lower in the purple light condition (but not in the orange light condition), as compared to the no lights condition. It appears that antecedent stimuli, and perhaps specific contexts, can alter tic expression/suppression through their association with reinforcers, even when these reinforcers are not provided anymore.

The experiments described above have a number of limitations, including extremely small sample sizes and low ecological validity. For example, reinforcers provided in the laboratory (e.g., tokens and money) certainly differ from the ones provided under real-world conditions (e.g., a back rub from a parent). Nonetheless, current evidence points to the importance of reinforcement and emotional states in conceptualizations of TS. Accordingly, a contemporary model of the disorder has been put forward and emphasizes the *interactions* between neurobiological and psychological factors to explain how symptoms are developed, maintained, and exacerbated in a comprehensive manner.

5. Comprehensive integrated model and related interventions

Both neurobiological and psychological research findings show that tics can be shifted in several ways. In line with these results, the comprehensive integrated model of TS acknowledges that two interacting forces underlie tic occurrence: one's biological processes and environment (Woods et al., 2007). According to this model, tics and premonitory urges exist in the first place because of genetic predispositions and dysfunctional neurobiological mechanisms. However, tics are then shaped in form, frequency, and intensity because of internal and external environmental factors (i.e., emotional states and positive/negative reinforcers) that directly impact underlying neurobiological and dopaminergic processes.

Although this interaction is proposed to be at the root of the problem, medication remains the most commonly prescribed intervention by physicians (e.g., Sallee, Nesbitt, Jackson, Sine, & Sethuraman, 1997; Scahill, Leckman, Schultz, Katsovich, & Peterson, 2003; Shapiro et al., 1989). These include alpha agonists (e.g., clonidine and guanfacine) and antipsychotics (e.g., risperidone), among others (Shprecher & Kurlan, 2009). Unfortunately, pharmacotherapy is associated with a number of side effects and randomized controlled trials have shown that medications are only moderately effective in suppressing tics (e.g., Sallee et al., 1997; Shapiro et al., 1989), possibly because other (psychological) factors are not addressed.

Comprehensive Behavioural Intervention for Tics (CBIT) is currently the gold standard psychological intervention for TS (e.g., Espil, Elkin, & Young, 2017). The main component of CBIT is *habit reversal training*, a form of therapy that stems from behavioural principles (e.g., Woods et al., 2007) and that originates from Azrin and Nunn's (1973) work on nervous habits and tics. Specifically, Azrin and Nunn posited that such habits (e.g., nail biting, head and shoulder jerking, eyelash picking) start either as a behavioural reaction from a psychological trauma/physical injury or as a normal but infrequent behaviour. Eventually, the behaviour blends into other daily movements and is performed outside of one's and others' awareness. The behaviour then becomes a habit. It also strengthens the muscles required to perform it and inhibits antagonistic muscles. Azrin and Nunn further claimed that social reinforcement plays a role in the behaviour's maintenance. As a result, the authors' initial treatment protocol aimed to increase awareness that one is engaging in such behaviour, interrupt the nervous habit, replace it with a movement that strengthens antagonistic muscles, and decrease social reinforcement.

Today, the overall goal of CBIT is to identify and then modify factors that exacerbate tics (e.g., Himle et al., 2006). Similar to Azrin and Nunn's (1973) protocol, individuals first undergo awareness training, during which they learn to detect cues indicating that a tic is imminent: this involves becoming aware of premonitory urges and of initial movements of complex tics. Second, individuals undergo habit reversal training, during which they practice engaging in a physical response that competes with the tic. A competing response is a movement that is incompatible with the tic—preventing its occurrence—and that is performed for 1 to 3 min or until the urge fades out naturally. In other words, patients are aware that they are experiencing an aversive premonitory urge but break the cycle of negative reinforcement by engaging in a different behaviour. Still, in some cases, competing responses may not necessarily include muscles that are antagonistic to the tic (as in Azrin and Nunn's protocol) and may instead emphasize strategies such as relaxation and/or diaphragmatic breathing (to target excessive throat clearing for example). Third, encouragement is provided when patients use their competing responses and, if necessary, parents of youths with TS learn to reinforce treatment progress instead of paying attention to tics.

The efficacy of CBIT in reducing tic frequency has been supported by case reports, single-subject and group experimental designs, open clinical trials, and randomized controlled trials. As an illustration, Piacentini et al. (2010) conducted a randomized controlled trial comparing CBIT to support therapy with 126 youths (9 to 17 years old). They found that those who underwent CBIT (versus support therapy) had significantly greater tic reduction, with a 30.8% decrease on the Yale Global Tic Severity Scale (YGTSS) for the CBIT condition compared with a 14.2% decrease for the support therapy condition at the end of treatment (i.e., 10 weeks). Treatment gains were maintained at 6-month follow up with 87% of available responders to CBIT still showing therapy benefits. Later, Wilhelm et al. (2012) conducted a randomized controlled trial also comparing CBIT to support therapy with 122 adults (16 to 69 years old). As with the youth sample, participants who underwent CBIT (versus support therapy) displayed significantly greater tic reduction, with a 25.8% decrease on the YGTSS for the CBIT condition compared with a 11.5% decrease for the support therapy condition at the end of treatment (i.e., 10 weeks). It was thus concluded that CBIT is a viable alternative to pharmacotherapy, transcranial magnetic stimulation, or more extreme forms of treatment (e.g., deep brain stimulation) for children and adults with TS.

Because competing responses often prevent tic occurrence, one of the proposed mechanisms of action in CBIT is habituation to the aversive nature of premonitory urges (e.g., Hoogduin, Verdellen, & Cath, 1997; Woods, Hook, Spellman, & Friman, 2000). This hypothesis primarily comes from a randomized controlled trial examining the efficacy of exposure and response prevention (ERP) in reducing tic frequency (Verdellen, Keijsers, Cath, & Hoogduin, 2004). ERP is the gold standard treatment for OCD: individuals are exposed to feared stimuli (e.g., contaminants) and are instructed to refrain from engaging in compulsions (e.g., washing behaviour; Foa et al., 2005). In the context of TS, ERP consists in triggering premonitory urges and instructing patients to refrain from ticcing. Verdellen and colleagues found that ERP produced significant reductions in both tic occurrence and ratings of *premonitory urges* as compared to baseline. In CBIT, habit reversal training also requires patients to tolerate unpleasant sensory phenomena—as in ERP—while engaging in competing responses. That being said, research is needed to empirically examine habituation as a potential mechanism.

Cognitive-behaviour therapy (CBT) can also be used to treat TS and appears to be effective (e.g., O'Connor et al., 2001, 2009). In CBT, habit reversal training is supplemented by cognitive restructuring. Specifically, individuals' appraisals of high-risk tic situations are also a treatment target. By challenging one's idiosyncratic cognitions regarding a given situation, this component of CBT simultaneously targets the specific emotional states that are known to exacerbate tics (e.g., anxiety and stress). For example, challenging catastrophic thoughts and beliefs related to ticcing in a classroom can consequently alleviate one's negative emotions while in that situation (thereby reducing tic frequency). Of note, the cognitive-psychophysiological approach to treating TS also targets a number of other cognitive processes related to tics, such as appraisals of the appearance of tics and beliefs about organizational standards and action planning (e.g., Leclerc, O'Connor, J-Nolin, Valois, & Lavoie, 2016; Leclerc, Valois, et al., 2016; O'Connor, Lavoie, Blanchet, & St-Pierre-Delorme, 2016; O'Connor, Lavoie, & Schoendorff, 2017). However, these perhaps important processes are not included in the comprehensive integrated model of TS, indicating that cognitive factors may have been neglected in formulations of the disorder and that current psychological treatments may be limited in scope.

6. Cognitive factors

As with other mental disorders characterized by negative behavioural and emotional aspects (e.g., anxiety and depressive disorders), there are reasons to believe that cognitive factors are important to consider in conceptualizations of TS. For instance, research has shown that individuals with chronic tics have elevated scores on the personal organization and personal standards subscales of the Frost Multidimensional Perfectionist Scale (Frost, Marten, Lahart, & Rosenblate, 1990), a comprehensive measure of perfectionism beliefs (O'Connor, Gareau, & Borgeat, 1997). In other words, those struggling with chronic tics *believe* that they should be extremely efficient and do as much as possible (e.g., at work, in school, or in everyday life) so they do not waste their time ("Either I do everything at once or I'm lazy"; O'Connor, 2002, p.1129). These positive beliefs about perfectionism are also accompanied by a need to put extensive effort in a given task, a desire to always over-prepare and be 'in advance' of oneself, and an unwillingness to relax (O'Connor, 2002). This finding led O'Connor to propose the cognitive-behavioural/psychophysiological model of tics. Specifically, he posited that these maladaptive beliefs cause a number of negative psychophysiological consequences, such as chronic heightened motor activation, elevated muscle tension, and feelings of frustration and anxiety, which are maintaining factors of tics. A more elaborated version of this model indicates that metacognition—thinking that a tic will occur, that one is not allowed to perform a tic, that ticcing will prevent one from completing activities, and that ticcing will impact one's appearance—also contribute to the maintenance of tics by promoting muscle contraction/tension and negative emotions (Lavoie, Leclerc, & O'Connor, 2013). In fact, a recent psychometric investigation provided support for the role of metacognition in influencing tic severity (O'Connor, St-Pierre-Delorme, Leclerc, Lavoie, & Blais, 2014).

Further, O'Connor (2002, 2005) suggested that individuals with tics are hypersensitive to various sensory phenomena (e.g., discomfort, tingling, itches) and that this hypersensitivity is exacerbated by another dysfunctional cognitive process: selective attention to unpleasant sensory experiences. O'Connor (2002, 2005) explained that premonitory urges may thus be the result of a pre-existing hypersensitivity (as supported by neurobiological data), combined with this increased attentional focus. O'Connor (2002, 2005) claimed that this selective attention to unpleasant sensory phenomena may stem from negative *appraisals* of these various body sensations, such that people with chronic tics may view aversive sensations very negatively. This hypothesis resonates with finding presented above from a recent study by Rozenman et al. (2015), which demonstrated a positive association between reported panic symptoms characterized by unpleasant somatic sensations (e.g., feelings of dizziness) and the severity of premonitory urges. This perhaps indicates that the more individuals with chronic tics are bothered by unpleasant sensory phenomena in general, the more they experience premonitory urges (and consequently tics). By integrating these theoretical propositions and lines of empirical evidence, it appears that those struggling with TS may be motivated to perform a tic to quickly remove an unpleasant premonitory urge, which they *perceive* negatively and *believe* is extremely aversive. Hence, as predicted by cognitive-behaviour theory (e.g., Beck, 2011) and alluded by O'Connor (2002, 2005), individuals with chronic tics may hold negative beliefs that make premonitory urges and other unpleasant body sensations seem intolerable.

6.1. Negative beliefs about discomfort

In order to properly inform explanatory models of TS and increase our understanding of cognitive factors exacerbating tics, it is essential to know more about the content of these negative beliefs. To accomplish this, one can focus on the automatic thoughts that individuals experience when they are triggered by premonitory urges. For instance, in a series of case studies, patients reported that urges are "impossible to bear" and that they can "drive me crazy all day long" (Mansueto & Keuler, 2005, p. 791). Similarly, individuals with chronic tics indicate having difficulty tolerating uncomfortable sensations caused by confining clothes, clothing tags, and scratchy fabrics (e.g., Mansueto & Keuler, 2005), as well as somatic symptoms such as dizziness and headaches (e.g., Rozenman et al., 2015). As a result, the beliefs underlying negative appraisals of unpleasant body sensations clearly extent beyond premonitory urges and appear to broadly target *discomfort*. It is thus proposed that individuals struggling with TS erroneously believe that experiencing discomfort is catastrophic, intolerable, and long-lasting and that coping with discomfort is very difficult.

Focusing on mental illnesses related to and frequently co-occurring with TS, such as OCD, can further corroborate this hypothesis. Although the phenomenology of both disorders differs in important ways (with concerns revolving around threats and potential negative consequences in OCD and around the relief of physical tension in TS), 'not just right' experiences—a subtype of OCD—seem to share key characteristics with tics. 'Not just right' experiences are conceptualized as feelings of incompleteness and *discomfort* when objects in one's surroundings are not organized or positioned a certain way (Miguel et al., 2000). For instance, an individual with OCD could be highly anxious and uncomfortable when exposed to one tilted frame placed among many other symmetrical frames. 'Not just right' experiences contrast with other obsessions (e.g., contamination and doubting) as they are rarely associated with a concrete threat but, instead, with tension, discomfort, and an urge to fix one's environment (e.g., Coles, Heimberg, Frost, & Steketee, 2005). Interestingly, some of the most commonly reported obsessions in individuals diagnosed with both OCD and TS are 'not just right' experiences. Psychometric investigations have shown that beliefs about perfectionism predict elevated levels of 'not just right' symptoms (e.g., Wheaton, Abramowitz, Berman, Riemann, & Hale, 2010). As indicated previously, individuals with TS also score highly on measures of perfectionism (Anholt et al., 2006; O'Connor et al., 1997, 2001, supporting a cognitive, belief-oriented link between tics and 'not just right' experiences. Given this phenomenological similarity regarding a need to alleviate discomfort, it might be that perfectionism *and* negative beliefs about discomfort are involved in both sets of symptoms. For example, individuals with complex motor tics often report that, in order to eliminate feelings of discomfort, they need to perform a tic until they attain the 'right' or 'perfect' sensation (e.g., Castellanos, 1998; Towbin, 1988). Several variations and levels of these dysfunctional beliefs likely interact with other psychological and neurobiological factors (involved in the TS-OCD spectrum) and ultimately emerge as either tics, obsessions, compulsions, and/or other related behavioural manifestations (e.g., body-focused repetitive behaviours and nervous habits).

Importantly, these negative beliefs about discomfort are proposed to be *maladaptive*, as they likely exacerbate tic occurrence. Although neurobiological abnormalities might be driving ticcing behaviour in the first place, individuals with TS appear to learn very quickly that a tic allowed them to put an end to feelings of discomfort. In this way, they rarely experience prolonged exposure to premonitory urges, which could possibly lead to and/or reinforce (pre-existing) negative beliefs about discomfort (e.g., "I could have never tolerated that urge any longer"). These maladaptive beliefs then encourage individuals with chronic tics to perceive premonitory urges in a catastrophic manner and to selectively pay attention to unpleasant body sensations, thereby exacerbating ticcing behaviour.

Likewise, negative beliefs about discomfort appear to be *irrational*. In both CBIT and ERP, premonitory urges—or feelings of discomfort—naturally fade out when patients are instructed to engage in a competing response (e.g., Woods et al., 2007) or to refrain from performing a tic (e.g., Verdellen et al., 2004). This further supports that patients falsely believe that discomfort will "drive [them] crazy all day long" (Mansueto & Keuler, 2005, p. 791). Therefore, another mechanism of action (other than habituation) that could potentially be at work in CBIT and ERP for TS is belief disconfirmation: patients learn through these behavioural exercises that they can tolerate and cope with discomfort and that premonitory urges do not last as long as previously expected. Actually, changes in dysfunctional beliefs have been shown to precede and predict symptom reduction in trials of CBT for OCD (e.g., Wilhelm, Berman, Keshaviah, Schwartz, & Steketee, 2015), providing evidence for a cognitive mechanism of change in a disorder tightly related to TS. Nonetheless, empirical investigations are necessary to support the hypothesis that beliefs about discomfort are involved in the exacerbation of tics.

6.2. Research avenues

A first step would be to interview children, adolescents, and adults with TS to collect qualitative data on beliefs about discomfort. Researchers could ask them questions about their perceived ability to cope with discomfort, their perceived consequences of experiencing discomfort, and about the different body sensations, somatic symptoms, and clothing materials that trigger feelings of discomfort. Then, this phenomenon could be examined psychometrically. Based on interview responses, an inventory could be developed to quantitatively assess one's negative beliefs about discomfort. Individuals could be asked to rate the extent to which they agree with specific statements, such as "unpleasant body sensations will bother me all day long", "discomfort is unbearable", "annoying body sensations like head pressure will prevent me from doing any activities", "wearing confining clothes will drive me crazy", etc. One could expect a positive correlation between negative beliefs about discomfort and severity of tics/premonitory urges to emerge.

However, experiments allow to directly test for causality and could provide more compelling evidence in favour of the hypothesis that negative beliefs about discomfort *lead* to tic exacerbation. For instance, researchers could ask individuals with TS to complete a tic-inducing task in order to assess their baseline tic frequency. Then, beliefs about discomfort could be experimentally manipulated by using the cold pressor test, a common laboratory method to induce acute pain during which participants submerge their hand in cold water (e.g., Dodo & Hashimoto, 2017). Following the cold pressor test, participants could be provided with false feedback about their performance: some could be told, depending on condition assignment, that they were better (or worse) than a normative sample at tolerating discomfort based on the amount of time they submerged their hand and based on bogus physiological measures. Researchers could then ask participants to complete the same tic-inducing task as before the manipulation and evaluate whether those in the high (versus low) negative beliefs about discomfort condition performed more tics as compared to baseline.

Such psychometric and experimental evidence would then encourage the creation of novel *cognitive* interventions to target these beliefs in the clinic and alleviate tic severity. As mentioned above, previous work by O'Connor and colleagues (1997, 2001, 2009) has shown that adding cognitive restructuring to treatment is beneficial for individuals with TS; unfortunately, beliefs about discomfort were not targeted in these protocols. Behavioural experiments are usually an optimal intervention strategy in cognitive-behaviour therapy for a number of mental disorders including OCD, as they allow patients to collect new and credible evidence disconfirming their maladaptive beliefs (e.g., Radomsky, Shafran, Coughtrey, & Rachman, 2010). In the case of TS, patients could be asked to estimate how long they believe they can tolerate a premonitory urge (or any uncomfortable body sensation like wearing a tight turtleneck) and then test out if they can tolerate the feeling for five extra minutes. Of course, behavioural experiments can be tailored to patients' idiosyncratic beliefs (e.g., test out if tolerating discomfort for 30 s versus 5 min will make them lose control or go crazy). Intervention studies could be utilized to assess the efficacy of these clinical techniques.

7. Revisiting the comprehensive integrated model

Given past and more recent efforts to conceptualize tics from a cognitive framework (e.g., O'Connor, 2002; Robinson & Hedderly, 2016), it is surprising that the comprehensive integrated model of TS focuses solely on neurobiological and environmental factors (Woods et al., 2007). This is perhaps due to the lack of experimental findings providing evidence for a direct link between dysfunctional beliefs and chronic tics. Still, it is important to predict how these various biological, behavioural, emotional, and cognitive factors interact to later investigate them. Hence, a revised version of the comprehensive integrated model is proposed (see Fig. 1).

Initially, genetic vulnerabilities likely underlie dysfunctional neurobiological processes (i.e., cortico-striatal-thalamic-cortical pathway) that lead to premonitory urges and tics. Prenatal and perinatal environmental risk factors may also contribute to the development of tics via epigenetic mechanisms, such as DNA methylation (e.g., Pagliaroli et al., 2016). Various emotional states—elicited by one's idiosyncratic appraisal of a given situation—are proposed to exacerbate tic occurrence, supposedly by stimulating an already hyperactive amygdala and thereby triggering these abnormal neurobiological/dopaminergic mechanisms (e.g., Godar & Bortolato, 2017; Neuner et al., 2014; Wang et al., 2011). Moreover, it is suggested that other cognitive processes (i.e., high personal/organizational standards and metacognition) intensify anxiety, frustration, motor activation, and muscle tension—together enhancing premonitory urges and tics (Lavoie et al., 2013; O'Connor, 2002). Tics are then shaped in form and frequency based on reinforcement contingencies. Actually, internal and external reinforcers or 'rewards' (e.g., elimination of premonitory urges) possibly strengthen neurobiological mechanisms associated with tics through excessive dopamine release, a process involved in habit formation (e.g., Albin & Mink, 2006; Delorme et al., 2015). Finally, it is suggested here that negative beliefs about discomfort and about one's ability to cope with discomfort lead individuals to negatively perceive and pay selective attention to premonitory urges, which in turn increases one's motivation to engage in ticcing behaviour. These beliefs about discomfort likely stem from interacting biological and developmental sources, but are most certainly maintained via negative reinforcement when a tic is performed (e.g., "I would not have been able to tolerate that urge").

Researchers in the fields of neurobiology, cognitive-behavioural psychology, and related domains should join forces to shed light on these proposed mechanisms, ideally by conducting experimental investigations. Empirical evidence supporting contemporary theories of TS, including the importance of cognitive factors in exacerbating symptoms, would then lead to more comprehensive treatment plans for clients and patients living with this debilitating condition and would help practitioners target idiosyncratic maintaining factors more effectively.

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Conflict of interest

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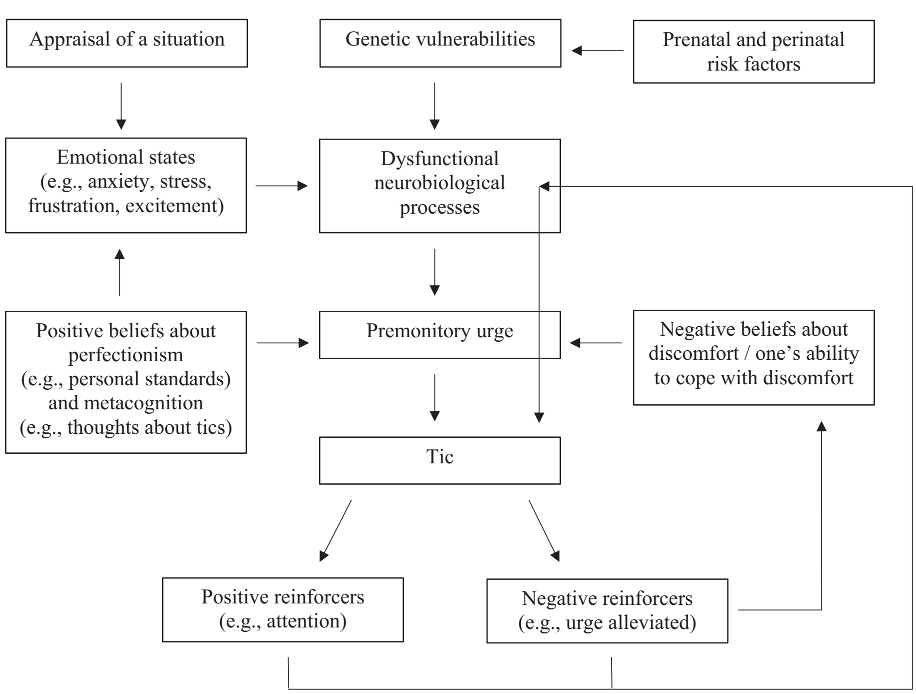

Fig. 1. Revised comprehensive integrated model of Tourette syndrome. Interacting genetic vulnerabilities are proposed to cause dysfunctional neurobiological and dopaminergic processes (i.e., cortico-striatal-thalamic-cortical pathway), leading to aversive premonitory urges and tics. Prenatal and perinatal risk factors may also contribute to the development of tics via epigenetic mechanisms. Emotional states such as anxiety and frustration (elicited by one's appraisal of a given situation) are proposed to exacerbate tics by overstimulating the amygdala and activating the same abnormal neurobiological processes. These emotional states could also be triggered by positive beliefs about perfectionism (e.g., high personal and organizational standards) and metacognition (e.g., thoughts about tics and their consequences). Perfectionism beliefs and metacognition may also enhance premonitory urges by intensifying muscle tension and motor activation. Research supports that tics are shaped in form and frequency via positive and negative reinforcement; actually, internal and external reinforcers/rewards likely contribute to excessive dopamine release, thereby fostering habit formation. Maladaptive beliefs about discomfort are hypothesized to underlie negative appraisals of premonitory urges, which in turn enhances their salience and motivates ticcing behaviour. Elimination of the premonitory urge (due to ticcing behaviour) is thought to negatively reinforce irrational beliefs about discomfort.

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Jean-Philippe Gagné is a PhD student in Clinical Psychology at Concordia University under the supervision of Dr. Adam Radomsky and a Vanier Scholar (Social Sciences and Humanities Research Council of Canada). Previously, he completed his BSc in Honours Psychology at McGill University (2015) and his MA in Clinical Psychology at Concordia University (2017), which included an exchange internship at McLean Hospital/Harvard Medical School. His research interests centre upon the role of cognitive-behavioural mechanisms in the development and maintenance of anxiety, obsessive-compulsive, and tic disorders.