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CHILD AND DEVELOPMENTAL PSYCHIATRY (M GRADOS, SECTION EDITOR)

The Current Understanding of Premonitory Urge Phenomena in Chronic Tic Disorders

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Abstract
Purpose of Review The goal of this review is to collect the progress made in the area of premonitory urge research in the areas of demographics, psychometrics, neurologic, psychopharmacology, and behavioral psychotherapy.
Recent Findings Individuals with no previous treatment for tics have differing levels of control over their ability to suppress tics. It is unclear if tic suppression reliably causes predictable and uniform changes in urge severity. The literature is unclear regarding the role of comorbid psychological conditions with respect to urge severity. Comprehensive Behavioral Intervention for Tics (CBIT) and several pharmacological treatments have shown decreases in tic severity but it is not clear if there are corresponding decreases in urge severity.
Summary Premonitory urges are prevalent sensations reported by individuals with tic disorders prior to the completion of a tic. Although there has been a recent increase in research on this topic over the past decade, several issues regarding the nature of urges remain ambiguous or in contention. Here, the authors have compiled a brief review of the literature which directly addresses current day understanding of premonitory urges.

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Introduction

Tourette's disorder (TD) and persistent (chronic) motor or vocal tic disorder (PCTD), commonly referred to as chronic tic disorder (CTDs), are child-onset neuropsychiatric disorders, characterized by "tics" or "sudden, rapid, recurrent, vocalizations or motor movements" [1]. Despite tics being the defining features of CTDs, research has suggested a common experience of "premonitory urges" in individuals with tics. Patients have described these urges as tension, urges, impulses, pressure, itches, and tingles [2]. Verbal reports suggest that the anatomical topography of urges is such that they most often occur in the head, neck, face, throat, shoulders, abdomen, and thighs; however, other anatomical regions may also be affected in some individuals. Many individuals report experiencing an unpleasant somatic "build up" of the urge prior to the tic and while attempting to suppress tics [3]. Urges are experienced as unpleasant for those who experience them and are temporarily relieved after ticcing. At this time, the precise relationship between tics and premonitory urges is largely unknown, though indirect evidence suggests that premonitory urges may be related to tic presentation, severity, frequency and develop over time [3]. Despite the theoretical importance of urges, historically they have received little attention in the literature.

Premonitory Urge Measures

To date, no known biological evaluative tool has been developed for assessing premonitory urges. In as much, urges may be best considered a psychological construct and like most psychological

constructs, the lack of direct assessment should not necessarily be considered problematic for phenomenological investigations. Early research on premonitory urges used methods such as informal questionnaires [3], phone interviews [4], and the therapist verbally asking patients if they experience premonitory urges during a therapy session [5]. These studies had high percentages of patients reporting urge experiences (76–93 %). More recently, the Premonitory Urge for Tics Scale (PUTS) was developed in order to obtain an urge-specific severity score for individuals experiencing tics and would suggest that very few TS affected patients (about 2 %) reporting *no* premonitory urge experiences [2]. Total PUTS scores are believed to reflect the degree to which people experience urge phenomena; a subset of items also reflect the urge-tic relation. The PUTS has demonstrated good internal consistency and test-retest reliability. However, the PUTS ratings have not been shown to change as a function of successful pharmacological or behavioral treatment. This suggests that either of the urge phenomena is less modifiable and/or that the PUTS, while specific, is not sensitive to treatment effects.

An "urge thermometer" was developed to evaluate the relationship between urges and tics and is a useful research tool for assessing urge ratings for experimental purposes [6••, 7, 8]. Many of these studies were designed to evaluate changes in urge severity ratings when tics are freely expressed or alternatively suppressed. Participant urge ratings are repeatedly prompted in real time by a computer monitor that displays, at preprogrammed intervals (e.g., 10 seconds), a bar graph depicting increasing urge severity. Study participants verbally report the overall severity of their urges when prompted. This method is useful for obtaining an in-depth perspective on changes in urges in an individual but can be prohibitively time and space intensive and therefore has limited clinical utility. The sensitivity of this evaluative tool to treatment effects has never been assessed to our knowledge.

The Beliefs About Tics Scale (BATS) is a self-report inventory developed to assess various beliefs of children and adolescents regarding the expression and suppression of tics [9]. Total BATS scores were intended to represent the degree to which an individual feels relief after ticcing and the individual's perceptions regarding the relationship between urges and tic suppression. Despite the independent judges excluding items from the final BATS which referred to urges rather than beliefs about tics, the BATS total scores were found to be strongly correlated with the PUTS total scores and less strongly associated with tic severity as assessed via the Yale Global Tic Severity Scale (YGTSS) [10].

Age of Urge Onset

It has long been assumed that urges develop later in the course of CTDs and may develop as a consequence of ticcing. A study questioning patients' experiences via retrospective verbal reports [3] suggested that, on average, individuals first became aware of premonitory urges at age ten. Reports that urges first appeared about 3 years after tic onset was thought to imply lagging development in somatosensory information processing [11, 12]. One study designed to evaluate the validity of this presumption dichotomized children (i.e., less than 10 years and 10 years and older) and found that both age groups reported the presence of premonitory urges, yet only for the older group were YGTSS tic severity and PUTS urge ratings correlated [2]. A similar study found that youths older than 10 years had higher internal consistency in PUTS ratings than youths 10 years and younger [13]. Another study found no differences between patients of different age groups, but only examined patients aged 16 and older [14]. A recent to-be-published study that included participants age 6–21 years found no significant differences in urge presentation in patients 10 years and younger compared to patients older than 10 years. In this study, patients as young as 6 years of age reported the experience of premonitory urges [15]. Despite the longstanding notion that patients under the age of 10 years do not experience urges, recent literature has suggested that patients of all ages who are affected by tics likely experience urges and that urges are an important clinical consideration regardless of age. However, while varied results may arise from different study methodologies, it is also possible that the development of urges and their relationship to tics is complex, evolving over the course of the illness, and perhaps increasingly bidirectional.

Gender and Urge Ratings

It is commonly accepted that CTDs are more common amongst males compared to females at a ratio of approximately 5:1. Most studies regarding urges particularly those with large samples are predominantly male and therefore have not adequately addressed gender differences. The original PUTS article studied a sample of predominantly male participants (93 %), in which no significant differences between genders was reported [2]. Another study of 254 participants observed no gender differences in urge presentation, though 83 % of participants were male [11]. Recently, a study with 65 % male participants suggested that there is no difference between male and female patients in urge presentation [14]. In a master's thesis, it was found that amongst 84 patients, approximately 33 % of which were female; there were no significant differences in urge presentation based on the gender of the patients [15]. At present, there appears to be remarkable consistency across studies suggesting similar urges experiences in TS affected youth irrespective of gender.

Urges and Comorbid Conditions

Patients with CTDs typically have comorbid psychological diagnoses, with attention deficit/hyperactivity disorder

(ADHD) and obsessive compulsive disorder (OCD) being the most common. At this time, the exact causal relation between CTDs and comorbid conditions is unknown, though a review of common theories regarding the etiology of these comorbid disorders has been conducted [16]. In clinical samples, ADHD comorbidity rates have ranged from 26 % [17] to as high as 60 % [18]. It has been suggested that OCD is the most common comorbid disorder which is genetically linked to CTD, afflicting 30–40 % of individuals with CTDs. Additionally, obsessive-compulsive symptoms (OCS) found in CTDs may be part of the CTD symptomatology, which is clinically different from symptoms found in "pure" OCD [19]. An analysis of clinical characteristics of children with tics found that those with comorbid psychological diagnoses did not differ from those with no comorbid conditions in terms of tic severity and tic-related impairment [17]. Similarly, diagnoses of ADHD and OCD were *not* predictive of PUTS total scores in two studies [15, 20]. In another study, ADHD and anxiety were also *not* statistically correlated with urges [13]. However, other studies have found links between comorbid conditions and premonitory urges. Studies have found significant correlations between urge severity and OCD severity [13, 21•, 22]. While other studies have also demonstrated positive correlations between urge severity and ADHD [22] as well as depression [13]. Additionally, urge severity has been significantly correlated with clinical symptoms, but the relation was stronger in individuals with a "pure" CTD than with patients who also have comorbid diagnoses [23]. Taken together, these findings suggest that urge ratings may be influenced by the presence of comorbid psychiatric conditions and that future investigations of urge phenomena should include comorbid symptom severity in addition to age as important covariates.

The Relation between Tics and Urges

While the exact relation between tics and premonitory urges is unclear, it has long been assumed that the two phenomena are intimately related. For some time now, it has been suggested that the primary functional impact of performing a tic is to alleviate the unpleasant premonitory urge. While tics may function to alleviate premonitory urges in some cases, it is also true that tics are performed in the absence of reported urges. It has also been suggested that tics with an associated urge were more bothersome to patients than tics not accompanied by a preceding urge, thus furthering the evidence that urges should be a consideration when administering treatment for tics [12, 14]. Studies with varied methodologies for assessing urge severity have reported that the expression of a tic served to eliminate urges in as few as 68 % [24] and as many as 92 % [3] of patients. In a study of two patients, tics were described as "not a pure 'unaware' phenomenon but an 'intentional' reaction to an unpleasant sensation" and also stated that patients with urges are not surprised by tics but are able to anticipate them [25]. The limited information about the relationship between urges and tics has lead researchers to champion a negative reinforcement or "urge reduction" model to explain the functional relationship between tics. A negative reinforcement model is also convenient for explaining that habituation to the premonitory urge produces symptom reduction.

Another model suggests that urge ratings are merely a reflection of an affected individuals' *belief* about the urge/tic relation as opposed to the actual severity of their urges. The BATS was found to be statistically correlated with the PUTS urge ratings despite that the authors of the BATS constructed the measure specifically to exclude information regarding urges. Interestingly, BATS total scores were moderately correlated with the YGTSS impairment subscale, but *not* the YGTSS tic severity score. Moreover, the PUTS was not correlated with the YGTSS total or impairment score in the same study. These results could imply that much like depression and anxiety [26], the more an individual embraces catastrophic beliefs about their urges, the more impairing their tic disorder will become, perhaps irrespective of tic severity. In fact, BATS scores were strongly related to depression in older children but not in younger children. This finding suggests that negative beliefs about tics could have a bidirectional relationship with comorbid depression. Additionally, one study observed that most patients believed their urges would increase if they tried to suppress their tics, but in reality their urge ratings remained unchanged during tic suppression periods [27••]. At present, the precise relationship between urges and tics is unknown; however, emerging literature suggests that the relationship may be complex, change over time, and be influenced by an individuals' interpretations and or cognitive sets associated with psychiatric comorbidity.

Premonitory Urges and Tic Suppression

Premonitory urges have been of central importance in developing behavioral models for CTDs. Urge reduction models suggest that negative reinforcement is central to the maintenance and exacerbation of tics; however, results have been inconsistent. Most affected individuals report differing ability to refrain from ticcing for variable periods of time. Several studies have shown evidence that urge severity is not necessarily related to one's ability to suppress their tics and that tic suppression does not necessarily increase urge severity as would be predicted by the negative reinforcement model. In a study of 15 adult patients, all patients reported urges and were capable of suppressing their tics [28] and urge ratings and tic inhibition were *not* correlated. Another study showed that treatment-naïve youth were more likely to tic following medium and high urge ratings during tic freely conditions; but interestingly, while showing individual differences in capacity to suppress (65–90 %) tics under contingently

reinforced tic suppression, they were equally capable of suppressing following low, medium, and high urge ratings [29••]. In a study of four children with CTDs, patients were exposed to conditions in which suppressing their tics was reinforced and failure to suppress tics was punished via token economy [30]. Both the reinforcement and punishment conditions achieved significant decreases in tics from baseline levels and neither condition showed significant differences from one another in urge presentation. In addition, for three out of the four patients, urge ratings were not consistently higher during the experimental conditions compared to the tic freely condition again lending to the notion that tic suppression does not necessarily increase the severity of urges. Conversely, one study of 13 youths found that urge strength was higher during reinforced tic suppression periods [31]; however, the tic suppression condition with an escape option resulted in higher tic rates during the subsequent escape portions. Also, urges were higher during the onset of break periods than at the offset of these periods, which would suggest that ticcing may influence premonitory urge ratings.

Patterns of urge ratings during tic freely and tic suppression conditions have been evaluated to determine the validity of the negative reinforcement hypothesis of tic maintenance. In a study of five patients using the urge thermometer, four participants showed a clear and reliable suppression of tics during a reinforced suppression condition when compared to a condition in which patients were asked to tic freely [6••]. Of these participants, three demonstrated a pattern in which urge ratings were relatively higher during the reinforced suppression condition when compared with the tic freely condition. The other two patients were able to suppress their tics but did not report higher urge ratings during the reinforced suppression condition when compared to the tic freely condition. Similar results were found by a more recent and larger study [7]. This study examined the possibility of distinct patterns of changes in urge ratings when patients are instructed to tic freely compared to when they are instructed to suppress their tics. In an examination of 12 patients, three participants had lower urge ratings during tic freely periods and high urge ratings during tic suppression periods (i.e., an "M" pattern) suggestive of a negative reinforcement model; however, four additional participants had higher urge ratings during tic freely periods and lower urge ratings during tic suppression periods (i.e., a "W" pattern), the inverse of the expected pattern. An additional five participants did not demonstrate reliable change patterns in urge ratings with or without tic suppression. Three of the participants who did not have significant differences between the two conditions did have significant differences during the first shift between ticcing freely and tic suppression, leading to two participants being labeled as having a "partial M" pattern and one participants labeled as having a "partial W" pattern. Taken together, the expected negative reinforcement "M" pattern was not observed in the majority of the cases examined in this study. These results strongly suggest that the generally accepted model of tic maintenance via a negative reinforcement or urge reduction model may be overly simplistic and is insufficient to capture the experience of all TS affect youth and suggests possible developmental considerations.

Neurological Models

Tic suppression followed by habituation to urges is a key in behavioral explanations for the effectiveness of behavioral treatments of CTDs. Although the presence of urges in patients with tics is thoroughly documented throughout the literature, the neurological correlations of this phenomenon are still relatively unknown, as most studies assessing neurological models have low sample sizes. In a study of 33 adolescents with CTDs, it was found that individuals with CTDs had a different functional connectivity when compared to a healthy sample, particularly within the fronto-parietal network which is thought to be important for online adaptive control [32]. Adolescents with CTDs have less than typical functional communication between distant regions of cortex, and over-communication between regions in close proximity, which is similar to connection patterns found in younger healthy control children. The notion of neurodevelopmental immaturity could explain the remitting course and why CTDs are much more common in children than adults, with approximately 1 % of school aged children exhibiting symptoms [33] compared to 0.1 % of adults with CTDs [34].

One study investigated bereitschafts potentials (BPs) using electromyography (EMG) and electroencephalography (EEG) recordings as a measure of premonitory urges [35]. With a sample size of three patients, the results suggested that BPs can be used as a marker for premonitory urges. Somatosensory evoked potentials (SEP) were examined using median nerve stimulation in 18 children with CTDs, 18 children with ADHD, and 10 controls [36]. Of these patients, 7 with ADHD and 9 with CTDs, and none of the controls had SEP abnormalities including latency or amplitude deviations. These findings suggest hyperactive sensorimotor fronto-subcortical circuit loops may give rise to premonitory urge symptoms. It was also suggested that high SEP amplitudes along with shortened cortical silent periods may be suggestive of a motor inhibitor dysfunction in CTDs. An examination of 40 adults with CTDs in comparison with 40 controls showed a reduction in gray matter volume in orbitofrontal, anterior cingulate, and ventrolateral prefrontal cortices bilaterally in patients with CTDs [37]. Tracts corresponding to cortico-cortical and cortico-subcortical connections seemed to be related to both premonitory urges and motor tics.

The use of functional magnetic resonance imaging (fMRI) to examine possible neurological models of urges has been documented in three studies. In one study, brain activity patterns were observed in 16 patients before and during tic expression using fMRI scanning [38]. Before tic onset, the supplementary motor

area (SMA) showed the most activity, and the anterior cingulate cortex (ACC), insular region, posterior putamen, parietal operculum, and ventrolateral thalamus also showed activity. However, only the SMA, ACC, insular, and thalamus appeared to be associated with urge generation. Another study compared the fMRI imaging data of 13 patients with CTDs and 21 control subjects during spontaneous and simulated tics [39]. Activity in the somatosensory and posterior parietal cortices, putamen, and amygdala/hippocampus complex was stronger in the CTDs group compared to the control group and suggests that premonitory urges may originate in these brain areas. Another study of 14 adult patients with CTDs, but no comorbid diagnoses, examined neural correlates of tic functioning using a resting-state fMRI (RS-fMRI) in free ticcing, as well as voluntary tic inhibition states [40•]. Results suggested that although the left inferior frontal gyrus is a key-area in tic inhibition, premonitory urges were not related to this neuronal signature.

Behavioral Treatment Methods

According to Martino and Leckman, "premonitory urges are very important in the tic-management strategies taught in cognitive behavioral therapies (CBT) for CTDs, and changing patients' responses to them may be a key mechanism of change", yet most research focuses on treatment for the tics themselves with less emphasis on premonitory urges [41]. Despite this, front-line treatment methods for tics, such as habit response therapy [42] exposure with response prevention [43], and comprehensive behavioral intervention for tics [44] have not had extensive research conducted on their efficacy with respect to premonitory urges themselves. In a study of 240 child and adult participants, cognitive behavioral intervention therapy (CBIT) was shown to be a more effective treatment than psychoeducation and supportive therapy (PST) with respect to both tics and urges. Interestingly, while there is no evidence to suggest that the treatment reduced urges it was suggested that tics associated with premonitory urges at baseline were unexpectedly less amendable to treatment [45]. Another study reported a decrease in urge ratings within and between exposure and response prevention sessions for tic treatment in 19 patients [46]. These limited findings are inconclusive regarding the effectiveness of behavioral treatment with respect to urges and the degree to which urges should be thought of as the target of therapeutic efforts.

Pharmacological Treatment

Although tics have been successfully treated by antipsychotics, alpha-2 agonists [47], guanfacine [48], atomoxetine, clonidine, and risperidone [49], the same treatment methods have not been thoroughly researched for the treatment of premonitory urges. One study found that tic impairment was reduced after 8 weeks of treatment with a D1 receptor antagonist, ecopipam, though no significant decreases in premonitory urges were observed [50]. A study on the effects of botulinum toxin (BTX) type A in 15 patients with 18 tics total observed short term efficacy in 16 tics and long term efficacy in 12 tics with urge reduction reported by eight patients [51]. Topiramate, a dopamine-receptor-blocking drug, has shown improvement in tic severity and urge severity [52]. In a placebo-controlled study of 24 patients with CTDs, Tetrahydrocannabinol (THC) was shown to reduce tic and premonitory urge severity for patients in the experimental condition [53]. At present, the degree to which medications function to reduce urge severity in addition to tics remains largely unclear.

Conclusions

Premonitory urges have now been the subject of several research papers over the past two decades, yet it is still difficult for researchers to pinpoint their nature. Much of what is known about premonitory urge phenomena is gleaned from studies that incorporate urge ratings assessed via questionnaire or an urge thermometer each with inherent strengths and weaknesses. Given the lack of a clear biological index of urge experiences and considering our current methods for assessment, we must consider urges as largely psychological phenomena. Our growing knowledge suggests that a significant majority of CTD affected individuals experience premonitory urges regardless of gender. One unresolved issue pertains to whether tics predate urges, urges predate tics, or if they develop concurrently. Contrary to historical reports, it would appear that urges are present as early as 5–7 years of age, when tics characteristically onset, and remain prominent throughout the typical remitting course. The degree to which urges impact the severity of CTD illness and related impairment as well as the degree to which urges are impacted by or impact comorbid psychiatric symptoms is largely unknown at present. There is little information regarding the precise relationship between urges and tics. Little is known about the strength of the actual association between urge and tic severity. It would appear that CTD affected individuals come to believe the two phenomena to be intimately linked; however, current empirical evidence suggests a modest relationship between urges and ticcing. The functional relationship between urges and tics is also unclear and recent research suggests that the relationship is likely complicated, may be related to developmental phenomena, and cannot be characterized solely by a negative reinforcement or "urge reduction" model of tics. It has also been suggested that treatments that encourage tic suppression safely and effectively allow for habituation to the premonitory urge and lead to lasting reduction in tic severity; however, some

studies suggest that urges may not predictably change as a function of effective behavioral or pharmacological intervention for tics.

Given the current knowledge regarding premonitory urges, we suggest it may be useful to consider the prospect that urges and tics have a similar age of onset. That early in the development of CTDs urges and tics may be separable, co-occurring phenotypic expressions of an underlying neurodevelopmental immaturity in the down-stream and up-stream cortico-striato-thalamo-cortical (CSTC) loops. In this proposed model, urges initially represent observable consequences of sensorimotor abnormalities in the CSTC circuitry and tics the observable consequences of basal ganglia dysfunction. We propose that much of the urge-tic relationship develops overtime and that their co-occurrence gives rise to the perception that urges and tics are functionally related in a meaningful way, which in turn binds them functionally. To draw a parallel, OCD affected individuals with good insight regarding the irrational nature of their fears often report that they do not actually believe that their compulsive behaviors protect them from the perceived risks associated with their obsessions but feel compelled to perform them anyway. We propose that, early in the course, primary or "biological" tics are spontaneous, involuntary motor behaviors that occur largely without warning; however, these involuntary motor behaviors will at times coincide with primary or "biological" sensorimotor urge phenomena. It is also important to consider that both motor and vocal tics involve repetitive movements and that repeated use of muscles would conceivably increase awareness of these anatomical regions. In this way, performing tics may serve to both fatigue and/or aggravate musculature involved in ticcing and subsequently increase one's awareness of the associated anatomical region, the later may in turn prompt subsequent voluntary secondary or "environmentally" determined tics. For instance, thrusting one's head back will aggravate the neck muscles and increase awareness of this anatomical region. Neck pain and/or discomfort may in turn prompt additional head/neck movements, which will temporarily interrupt the pain but further aggravate the associated musculature. In the proposed model, while some tics would continue to occur involuntarily, tics would over time be increasingly performed as a voluntarily responses to unpleasant sensory experiences in the anatomical region where tics previously occurred due to both bottom-up and top-down processes. This theory may help to explain the tendency for tics to worsen in adolescence.

Considering that presently, we must consider premonitory urges to be psychological rather than biological phenomena, we propose that urges may best be considered to be akin to somatosensory obsessions. That many, but not all, tics are performed voluntarily due to the belief that ticcing will alleviate an associated symptom that in actuality will dissipate on its own even if a tic is not performed. Moreover, much in the same way that compulsive behaviors often serve to make obsessions worse over time, it may be the case that voluntarily performing tics increasingly in response to urges may serve to "link" parallel phenomena that in actuality are modestly correlated. With this in mind, behavioral treatments may serve to demonstrate to affected individuals that urges and tics are not intimately linked in the way they have come to believe them to be, thereby "shaving off" the maladaptive, learned behavior, and reducing CTDs to their core biological underpinnings. Much like with exposure and response prevention, OCD affected individuals learn via repetition that compulsions, in reality, do little to address obsessional fears and/or associated autonomic hyperarousal and OCD becomes reduced to "false alarms".

In sum, the current literature regarding premonitory urge phenomena has laid a good foundation for the future of premonitory urge research, but there are many topics which deserve further empirical scrutiny. Particularly, it would be interesting to see future studies further examine the somatic obsession hypothesis presented here.

Compliance with Ethical Standards

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Human and Animal Rights and Informed Consent All reported studies/experiments with human or animal subjects performed by the authors have been previously published and complied with all applicable ethical standards (including the Helsinki Declaration and its amendments, institutional/national research committee standards, and international/national/institutional guidelines).

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