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Patterned changes in urge ratings with tic suppression in youth with chronic tic disorders



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ABSTRACT

Background: Premonitory urges are central to emerging behavioral models of chronic tic disorders (CTD). Urge reduction has been proposed as a behavioral explanation for tic maintenance and exacerbation as well as the efficacy of behavioral treatments. Prior investigations have produced inconsistent findings despite common methodologies. The current study evaluated the possibility that data aggregation obscures distinct and meaningful patterns of change in urge ratings when tics are freely expressed versus suppressed.

Method: Participants (n=12) included children with moderate-to-marked tic severity and noticeable premonitory urges. Tic frequencies and urge ratings were obtained at 15 s and 10-s intervals, respectively, across an alternating sequence of 10-min tic freely and 40-min tic suppression conditions. Patterns were established using a two step approach.

Results: Five distinct patterns of urge rating change emerged, suggesting data aggregation may obscure meaningful patterns in the urge—tic relationship when tics are completed versus suppressed.

Limitations: Eligibility criteria may have unintentionally excluded younger affected children and included older participants with more severe tic disorders than commonly seen. Additional research with less stringent eligibility criteria and a larger sample size will help validate the results.

Conclusions: The relationship between urges and tics is much more complex than previously theorized. Investigations that rely on global assessments of urge and tic severity and/or assume uniformity when aggregating participant data may obscure meaningful differences in the urge—tic relationship. Future investigations should examine the possibility that individual differences and/or developmental considerations modulate the functional urge—tic relationship.

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Chronic tic disorders (CTDs) are neurodevelopmental disorders that begin in early childhood, are characterized by motor and/or vocal tics (i.e., brief, repetitive movements or vocalizations), and often result in functional impairment (Conelea, Woods, & Brandt, 2011; Conelea, Woods, Zinner, et al., 2011). By the age of ten, most affected individuals report premonitory urges, or unpleasant

sensory experiences that precede tics (Himle, Woods, Piacentini, & Walkup, 2006; Leckman, Walker, & Cohen, 1993). An emerging behavioral model suggests that tics result from basal ganglia dysfunction and may be exacerbated as tics serve to alleviate unpleasant premonitory urges (Capriotti, Espil, Conelea, & Woods, 2013; Evers & Van de Wetering, 1994; Himle, Wood, Piacentini, & Walkup, 2006). This view is referred to as the "urge-reduction" or "negative reinforcement" model and has been investigated in several recent studies (Himle, Woods, Conelea, Bauer, & Rice, 2007; Specht et al., 2013, 2014; Verdellen et al., 2008). Habituation to

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premonitory urges through disruption of the negative reinforcement cycle has been proposed as a behavioral mechanism underlying the efficacy of tic suppression-based therapies (Capriotti & Woods, 2013; Himle, Woods, et al., 2006); however, findings regarding the urge-reduction model are inconsistent and have called into question the exact nature of the urge—tic relationship.

Research regarding the urge phenomenon was initially grounded in case studies and ad hoc self-report data (e.g., Banaschewski, Woerner, & Rothenberger, 2003; Kwak, Dat Vuong, & Jankovic, 2003; Leckman et al., 1993). Self-reports of tic completion alleviating the aversive premonitory urge were believed to accurately reflect the relationship between urges and tics (Bliss, Cohen, & Freedman, 1980; Kane, 1994). Current conceptualizations of the urge reduction model have built on these self-reports by hypothesizing that individuals would rate urges as less intense during periods of free ticcing and more intense early in periods of tic suppression, with gradual reduction as habituation to the urge occurs over prolonged periods of tic suppression (Evers & Van de Wetering, 1994; Himle, Woods, et al., 2006). Recent investigations have compared urge ratings during periods of free ticcing and reinforced tic suppression to address this hypothesis.

Two studies have provided support for the urge reduction model using slightly different methodologies (Capriotti, Brandt, Turkel, Lee, & Woods, 2014; Himle et al., 2007). Three of five participants in a within-subjects reversal design demonstrated relatively low urge ratings during tic-freely periods and higher urge ratings during tic suppression periods (Himle et al., 2007). A more recent study also found that urge ratings increased during periods of reinforced tic suppression, and that urge severity was higher immediately before participants self-initiated a brief period of escape from reinforced tic suppression (Capriotti et al., 2014). The volitional nature of these breaks from the tic suppression contingency suggests that gaining access to urge reduction was a reinforcing event. The findings of both of these studies support the urge reduction model and the hypothesis that tics serve to relieve the unpleasant urge.

A study designed to examine the efficacy of Exposure and Response Prevention, in which tic suppression was established via "coaching" from a therapist with no systematic reinforcement, did not find initial increases in average urge ratings during the first 5 minutes of tic suppression, but did note within- and between-session reductions in urge ratings (Verdellen et al., 2008). These findings lend partial support to the urge reduction model, as the within- and between-session reductions in urge ratings are indicative of habituation to the urge despite no evidence of immediate increases in urge ratings during periods of tic suppression.

Two studies using similar methodologies found no change in average urge ratings between tic-freely and reinforced tic suppression periods, inconsistent with the urge reduction model (Specht et al., 2013; Woods, Walther, Bauer, Kemp, & Conelea, 2009). Subsequent analyses of the Specht et al. (2013) data suggested that when tics were allowed, there was a trend toward tic completion being least likely following low-intensity urge ratings and most likely following high-intensity urge ratings (Specht et al., 2014). This finding partially supports the negative reinforcement hypothesis, as more intense urges may have prompted escape through tic completion (negative reinforcement). Taken together, these findings suggest that the negative reinforcement model is viable but fails to explain the entire spectrum of results observed.

Disparities across studies cannot simply be dismissed as anomalous, as there were many close methodological parallels. One possible explanation for inconsistent findings is that the urge—tic relationship is moderated by environmental variables, individual differences, and/or developmental considerations (Banaschewski et al., 2003; Capriotti et al., 2013; Leckman et al., 1993; Woods,

Piacentini, Himle, & Chang, 2005). Consistent with this notion, we re-examined data from a previously reported study (Specht et al., 2013), which found no statistically significant changes in urge ratings across an alternating sequence of tic-freely and reinforced tic suppression conditions. We began by examining the possibility that data aggregation (i.e., comparing mean urge ratings) obscured meaningful patterns of change in urge ratings across tic freely and tic suppression periods. This possibility was based on a review of previous studies in which some participants evidenced patterns of changing urge ratings that appeared consistent with the urge reduction model, with more ambiguous patterns for others (Capriotti, Brandt, Ricketts, Espil, & Woods, 2012; Himle et al., 2007).

The primary aim of the current study was to examine individual participant urge ratings. Although this study was exploratory in nature, we hypothesized that participants could be grouped based on common patterns of change in the urge phenomena across ticfreely and tic suppression conditions. We speculated that these patterns may occur in such a way that they would essentially nullify each other when averaged, thus accounting for the previous null findings (Conelea, Woods, & Brandt, 2011; Conelea, Woods, Zinner, et al., 2011; Specht et al., 2013; Woods et al., 2009). Our secondary aim was to further describe characteristics (i.e., changes in tic frequency and urge ratings across conditions, global tic and urge severity, and age) of each group for pattern validation and prospective investigation.

1. Method

The current study involved follow-up analyses from a previously reported study (Specht et al., 2013). With the exception of the data analysis section, the methods described below are identical to those previously reported and have been substantially edited for the sake of brevity. For more details regarding methodology, please refer to Specht et al. (2013).

1.1. Participants

Eligible participants included healthy males and females (ages 10-17) who met the Diagnostic and Statistical Manual of Mental Disorders-Fourth Edition-Text Revision (DSM-IV-TR; American Psychiatric Association, 2000) diagnostic criteria for Tourette's syndrome or a chronic motor or vocal tic disorder. Additional inclusion criteria included (a) no history of >3 weeks of behavioral or suppression-based treatment for tics, (b) moderate-to-severe tic severity determined by a minimum total score of >14 for both motor and vocal tics or > 10 if motor or vocal tics only on the Yale Global Tic Severity Scale (YGTSS, Leckman et al., 1989), (c) lowaverage range or better intellectual functioning defined by a two scale score of >75 on the Weschler Abbreviated Scale of Intelligence (WASI, Psychological Corporation, 1999), (d) the presence of a noticeable premonitory urge as indicated by a score \geq 12 on the Premonitory Urge for Tic Scale (PUTS, Woods et al., 2005), (e) current presentation of one or more motor and/or vocal tics at a rate of at least 1 per minute. Children with significant oppositional defiant disorder or conduct disorder symptoms, as determined by the Anxiety Disorders Interview Schedule-Research Lifetime Version (ADIS, Silverman & Albano, 2002), were excluded from the study. Children with other co-occurring conditions (e.g., obsessivecompulsive disorder, attention-deficit/hyperactivity disorder) were not necessarily excluded provided they met all other eligibility requirements. Pharmacological tic and/or urge suppression would unnecessarily confound results; therefore, potential participants were excluded if they reported a current medication

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