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The development of stimulus control over tics: A potential explanation for contextually-based variability in the symptoms of Tourette syndrome☆

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A B S T R A C T

Research has demonstrated that providing reinforcement for tic-free intervals can decrease tic frequency in controlled analogue settings. The aim of the current study was to determine whether reinforcement could be used to create stimulus control over tic expression. Ten children with chronic tic disorders (including Tourette syndrome) completed four discrimination training sessions. Each session consisted of three exposures to each of three, 5 min conditions presented in a random order. In one condition, participants were reinforced for tic absence on a 10-s fixed interval schedule in the presence of a purple light. In a second condition, participants were instructed to suppress their tics, but were not reinforced for doing so in the presence of an orange light. In a third condition, participants were instructed not to suppress their tics in the presence of two non-illuminated lights. Confirming findings from other studies, results showed that reinforcing tic suppression reduced tic frequency to a greater extent than only providing instructions to suppress. To test for stimulus control, a fifth session was conducted following the aforementioned discrimination training sessions. The fifth session consisted of three exposures to each of three 5 min conditions presented in a random order. 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 all three conditions, instructions to suppress (or not suppress) tics were not provided, and reinforcers for successful suppression were not delivered. Results indicated that in the presence of the purple light, tics were significantly lower than when neither light was illuminated. These findings provide preliminary support for the idea that a history of differential reinforcement in various contexts may play a role explaining variability in tic symptom expression.

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Chronic tic disorders (CTD) include Tourette Syndrome (TS) and chronic motor/vocal tic disorder (CMVT). Tourette syndrome is characterized by the presence of at least two motor tics and one vocal tic over the course of at least 1 year, and CMVT is characterized by the presence of motor or vocal tic(s), but not both, over the course of at least 1 year (American Psychiatric Association, 2000). CTDs occur in approximately 0.6% of the population (Khalifa & von Knorring, 2003) and are more common in boys by a ratio of about 3–4:1 (Robertson & Stern, 2000).

Tics follow a waxing and waning pattern. Onset of CTD is usually between the ages of 4 and 6 years. Peak severity occurs between the ages of 10 and 12 years (Peterson, 1996). In many cases, tic severity diminishes into adulthood, but in some cases, tics remain or even increase in severity as the child develops (Coffey et al., 2000). Tics wax and wane throughout the course of the disorder, and tic expression is heavily influenced by the patient's surroundings (Piacentini et al., 2006; Silva, Munoz, Barickman, & Friedhoff, 1995; Woods, Watson, Wolfe, Twohig, & Friman, 2001).

In addition to tics, many persons with CTDs report a "premonitory urge" (Woods, Piacentini, Himle, & Chang, 2005), which is described as an aversive tension, tickle, or pressure that precedes tics and is relieved after engaging in the tic. Some researchers have suggested the urges emerge around the age of 10 years (Leckman, Walker, & Cohen, 1993), while others have suggested that the urges are present prior to the age of 10 years but not related functionally to the tics until later in childhood (Woods et al., 2005). Research has suggested that premonitory urges are also impacted by the patient's surroundings (O'Connor, Brisebois, Brault, Robillard, & Loiselle, 2003).

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Although it is widely recognized that CTD symptom expression is the result of interacting environmental and biological factors (Chappell et al., 1996; Conelea & Woods, 2008a; Lichter & Jackson, 1996; Woods, Himle, & Conelea, 2006), the vast majority of research on CTDs has focused on biological factors. CTDs are understood to have a polygenetic genetic basis that manifests primarily as dysfunctional cortico–striatal–thalamocortical (CSTC) circuitry (Leary, Reimschisel, & Singer, 2007; Swain, Scahill, Lombroso, King, & Leckman, 2007).

Comparatively little research has examined the influences of environmental factors on tic expression. Although surveys of persons with TS have demonstrated relationships between the occurrence of various environmental variables and changes in tic frequency (i.e., O'Connor et al., 2003; Silva et al., 1995), there has been little controlled work studying the systematic influence of these factors. Most of the existing work has focused on the impact of reinforcement for tic suppression.

Woods and Himle (2004) noted that persons with CTDs do not suppress their tics simply because they are asked to do so. Rather, they often receive some consequence for suppression (e.g., they are able to go to movies or avoid being teased about tics). To explore the hypothesis that reinforcement for suppression could have a powerful impact on tics, Woods and Himle (2004) compared verbal instructions to suppress (i.e., simply instructing someone to suppress their tics) to verbal instructions plus reinforcement for brief tic-free periods of time (i.e., 10-s tic-free periods were reinforced using tokens with a small monetary value). Across four children with TS, results clearly demonstrated the superiority of the reinforced suppression procedure in producing tic reduction. The verbal instruction condition produced a 10.3% reduction in tics from baseline levels, whereas the reinforced suppression condition produced a 76.3% reduction. Later studies replicated the findings, showing that reinforced tic suppression produced reliable suppression with similarly large magnitudes (i.e., Himle & Woods, 2005; Himle, Woods, & Bunaciu, 2008; Himle, Woods, Conelea, Bauer, & Rice, 2007; Woods, Himle et al., 2008).

The aforementioned studies demonstrated that providing a token reinforcer for effective tic suppression resulted in significant tic reduction. Nevertheless, the effect could have been produced by the direct effects of the reinforcer delivery or by the demand for increased attention toward tics suppression. To clarify the role of contingent reward, Himle et al. (2008) compared the effects of contingent versus non-contingent reinforcement for suppression in four children with TS. Using an alternating treatments design, results showed that 3 of the 4 children demonstrated reliable suppression. Of these three, all showed similar patterns of responding. Notably, when children were contingently reinforced for successful suppression, they showed dramatic decreases in tic frequency from baseline. In contrast, during the non-contingent reinforcement condition, children were instructed to suppress their tics, but received frequent token reinforcers in a non-contingent fashion (i.e., participants received rewards that were unrelated to tic occurrence/nonoccurrence, but that participants were told were "reminders to suppress"). During this latter condition, tic frequency failed to reliably differentiate from baseline levels. Combined, the study suggests that reinforcement needs to be delivered in a contingent fashion to produce maximal tic reduction.

The aforementioned studies examining the effects of reinforcement on tic suppression focused only on tic frequency as a dependent variable. However, it is also important to consider the impact that reinforcing tic suppression may have on the premonitory urge. The emerging neurobehavioral model of tic disorders posits that tics may be partially maintained via a negative reinforcement process in which tics serve to reduce an aversive premonitory urge (Evers & van de Wetering, 1994; Leckman et al., 1993). If this is true, then it may be predicted that tic suppression would result in an increase in the perceived intensity of the urge. To test this hypothesis, Himle et al. (2007) exposed five children to baseline (i.e., no instructions to suppress), suppression (reinforced for suppression), and free-tic (instructions to avoid suppression) conditions in a single-subject experimental reversal design. In addition to monitoring tic frequency, the researchers also asked the children to provide an intensity rating of their premonitory urges throughout the study using a 0 (low) to 8 (high) scale. Four of the five children in the study showed a successful suppression effect (the one who did not was the youngest at 8 years of age). Of these four, three showed the predicted pattern, with urges being higher during the suppression conditions when compared to the free-tic periods. The fourth child showed no apparent relationship between the tics and premonitory urges. Combined, these results suggest that premonitory urges may also be influenced by contingently reinforcing the suppression of tics.

Summarizing the aforementioned work, it is clear that reinforcement can have a significant and reliable impact on tics, reducing them if suppression behaviors are reinforced or potentially increasing them if the tic produces a reduction in the premonitory urge. Given these findings, it is worth considering whether other related behavioral processes may be active and useful in forming a more complete understanding of TS symptom expression. As stated earlier, it is well-understood that tic expression can be heavily influenced by environmental events. Often the "reactive" nature of the tics to such events is described as, or implied to be a poorly understood characteristic of the disorder (Hoekstra, Steenhuis, Kallenberg, & Minderaa, 2004). However, one possible explanation for such environmentally induced variability is that tics come under the control of events that predict the availability or absence of reinforcement for tic suppression. As the acquisition of stimulus control is a core behavioral phenomenon that can emerge during the process of reinforcement (Dinsmoor, 1995a, 1995b), and given that reinforcement can play a clear role in creating tic reduction, this explanation seems quite plausible. Likewise, given the evidence that urges are increased during suppression, then stimuli that predict reinforcement for suppression should also yield increases in urges. The current study attempts to determine whether reinforcing tic suppression in particular settings, but not others, can create stimulus control over both the tics and premonitory urges through the testing of two primary and two exploratory aims/hypotheses.

Primary aims & hypotheses

(1) The first primary aim of the study is to replicate earlier research showing that reinforcing children for suppressing tics can result in a significant tic reduction. Consistent with prior findings (e.g., Himle & Woods, 2005; Woods & Himle, 2004), it is predicted that tic frequency will be lower in a condition in which tic suppression is reinforced in comparison to a condition in which suppression is not attempted.

(2) The second primary aim is to determine whether stimulus control develops after reinforcing the suppression of tics. It is hypothesized that following a period of stimulus discrimination training, children will tic less frequently in the presence of a stimulus light predicting the availability of reinforcement for tic suppression when compared to a stimulus condition, in the presence of which, suppression has not been attempted.

Exploratory aims

(1) The first exploratory aim is to determine the impact of stimulus control training on the premonitory urge. Consistent with the findings of Himle et al. (2007), it is predicted that urge ratings

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