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

Douglas W. Woods [a,*], Michael R. Walther [a], Christopher C. Bauer [b], Joshua J. Kemp [a], Christine A. Conelea [a]

[a] Department of Psychology, University of Wisconsin-Milwaukee, Garland Hall, P.O. Box 413, Milwaukee, WI 53201-0413, USA
[b] Department of Psychology, North Dakota State University, ND, USA

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ABSTRACT

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.

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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* Corresponding author. Tel.: +1 414 229 4747; fax: +1 414 229 5219.
E-mail address: dwoods@uwm.edu (D.W. Woods).

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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

will be more intense when suppression is reinforced. Likewise, it is predicted that the urge will be more intense in the presence of a stimulus that predicts reinforcement of tic suppression when compared to a stimulus, in the presence of which, suppression has not been attempted.

(2) The second exploratory aim is to replicate the findings of Woods and Himle (2004), which demonstrated that providing reinforcement for tic suppression was more effective in reducing tics than simply instructing participants to suppress their tics. It is hypothesized that the condition in which tic suppression is reinforced will produce greater decreases in tic frequency than the condition in which tic suppression is instructed, but not reinforced.

Method

Subjects

Ten children with a mean age of $M = 10.8$ years (range = 9–15) participated in the study. The sample consisted of 10 boys with TS ($N = 9$) or a chronic tic disorder ($N = 1$). Subjects were recruited through print advertisements placed in newspapers serving southeastern Wisconsin and through self- and physician referrals to the University of Wisconsin-Milwaukee Tic Disorders Specialty Clinic. To be eligible for participation, subjects had to (1) be a healthy male or female between the ages of 9 and 17 years, (2) meet DSM-IV-TR diagnostic criteria for TS or chronic tic disorder, (3) have a minimum Yale-global tic severity score (YGTSS-total score) of $\geq$14 for a TS diagnosis or $\geq$10 for a chronic tic disorder diagnosis, (4) currently be exhibiting tics at a rate of at least 1 per minute averaged across a 10-min videotaped observation, (5) have a Wechsler Abbreviated Scale of Intelligence (WASI; The Psychological Corporation, 1999) of >75, (6) report no history of non-pharmacological treatment for tics (over 3 weeks of treatment) in which suppression strategies were a primary component of the intervention, and (7) report the presence of a noticeable premonitory urge as indicated by a score $\geq$12 on the 9-item Premonitory Urge for Tics Scale (PUTS; Woods et al., 2005).

The sample had a mean YGTSS-Total Tic Severity score of $M = 23.0$ (SD = 6.3), indicating moderately severe TS. The YGTSS Motor Tic Score was $M = 12.5$ (SD = 3.98), and the Vocal Tic Score was $M = 10.5$ (SD = 4.28). The YGTSS Impairment score was $M = 23.2$ (SD = 10.1), indicating a moderate level of impairment caused by the tics. The mean PUTS score was $M = 23.10$ (SD = 6.2). In addition, 70% of the sample had at least one comorbid psychiatric diagnosis, the most common of which were Obsessive-Compulsive Disorder (30%), Social Phobia (30%) and ADHD (any subtype; 30%). Table 1 provides demographic information for the sample. Subjects were paid $250 upon completion of the study.

Measures

Anxiety disorders interview schedule – fourth edition (ADIS-IV; Silverman & Albano, 1996)

The ADIS is a structured diagnostic interview administered by the examiner to the parent and child, which yields a comprehensive listing of all positive DSM-IV diagnoses and diagnostic criteria. The ADIS-IV has demonstrated good to excellent test–retest reliability and good concurrent validity (Silverman, Saavedra, & Pina, 2001; Wood, Piacentini, Bergman, McCracken, & Barrios, 2002).

Premonitory urge for tics scale (PUTS; Woods et al., 2005)

The PUTS is a brief 9-item self-report measure that asks individuals to rate several premonitory urge descriptions on a 1–4 point ordinal scale anchored by "not at all true" and "very true." Initial data show the PUTS to be internally consistent ($\alpha = 0.81$) and temporally stable at 1 ($r = 0.79$) and 2 ($r = 0.86$) weeks (Woods et al.). A mean score of 18.5 with a standard deviation of 6.1 was reported in a clinical sample of children with CTDs (Woods et al.).

WASI (The Psychological Corporation, 1999)

The WASI, based on the Wechsler Intelligence Scales, is a psychometrically acceptable measure of intellectual functioning and was designed for use with individuals aged 6–89 years. The 2-subscale version of the WASI was administered in the current study.

Yale-global tic severity scale (YGTSS; Leckman et al., 1989)

The YGTSS is a clinician-administered semi-structured interview and produces separate severity ratings for motor and vocal tics, impairment produced by the tics, and an overall score of tic severity. Motor and vocal tics are rated separately across five areas (number of tics, frequency, intensity, complexity, and interference) on a scale of 0–5. Scores for each of these five areas are summed to create a motor tic score and a vocal tic score. Total tic severity is calculated by summing the motor tic and vocal tic score, yielding a possible range from 0 to 50. The clinician also provides a separate rating on a 0–50 point scale that is designed to assess impairment produced by tics (50 is most impaired). The YGTSS has demonstrated acceptable psychometric properties. The scale has good internal consistency, good interrater reliability, and acceptable convergent and divergent validity (Leckman et al., 1989).

Procedures

The study occurred over six separate sessions across 1–3 weeks. Session 1 took approximately 3 h and included an initial assessment and preliminary data collection. Sessions 2–5 involved discrimination training, in which the participants were reinforced for effective tic suppression in the presence of one stimulus (a purple light), were instructed to suppress tics but were not reinforced for doing so in the presence of another stimulus (an orange light), and were instructed not to suppress tics at all in a third condition during which no lights were illuminated. In Session 6, a test for the acquisition of stimulus control was conducted. Study procedures are described in detail below.

Assessment/preliminary data collection

Participants and their parents were asked to attend an initial assessment session, during which eligibility was determined and informed consent obtained. The participant and/or parent were asked to complete the demographics form, the ADIS, PUTS, WASI, and YGTSS. After all assessments were completed, the child was overtly recorded (i.e., the camera was placed in plain view of the child) in a room by himself for 10 min. During these 10 min, the child was instructed to (a) sit in the seat while facing the camera without covering his face, (b) remain in the seat the entire time, and (c) feel free to tic as much or as little as he needed to. After the observation was completed, the investigators watched the videotape and derived operational definitions of all tics displayed.

Stimulus discrimination training

All training sessions took place in a 10′ x 15′ observation room equipped with a one-way observation mirror that allowed for covert observation and recording. During each of the four training sessions (Sessions 2–5), the child was consecutively exposed to three different conditions, three times each, in a random order. The three conditions included a baseline (BL) condition, a condition in which the child was instructed to suppress his tics and was reinforced for doing so in the presence of a purple light (SUP-Purple)

Table 1
Participant comorbid diagnoses, tic severity, and current tics

Participant number	Sex	Age	IQ	DSM-IV-TR diagnoses	YGTSS score	Tic medications-current	Tics
1	M	15	98	Tourette disorder Social phobia GAD OCD	37	Haloperidol	*Motor*: Eye blinking, facial grimace, head jerk, abdominal tensing, shoulder shrug, hand tic, toe/foot tic, mouth tic, complex facial movement (mouth), arm into side tic, bending tic, rotating tic, stutter step, tapping/touching, hitting crotch, biting tongue, scooting out of chair. *Vocal*: Screeching up to certain pitch, loud screeching coming to a stop on a certain pitch, barking, coprolalia, words, sniffing.
2	M	9	117	Tourette disorder	20	None	*Motor*: Eye blinking, eye rolling, brushing hair away, nose movements, head jerk/tilt, eye squint, mouth tic, shoulder shrug, finger stretch, hand scratch. *Vocal*: Sniffing, grunt/throat clearing.
3	M	11	103	Tourette disorder ADHD (combined) GAD	18	None	*Motor*: Nose movements, mouth movements, facial grimace/smiling. *Vocal*: Sniffing, throat clearing, grunting.
4	M	9	129	Tourette disorder	22	None	*Motor*: Eye blinking, head jerk, jaw movement, stomach tightening, shoulder shrug, foot flex, eye rolling. *Vocal*: Grunting, snorting, lip smacking.
5	M	15	122	Tourette disorder Social phobia	25	None	*Motor*: Eye blinking, eye rolling/darting, head to side, head to shoulder, head shake, shoulder shrug, abdominal tensing, bicep tensing, arm movement. *Vocal*: Throat clearing, coughing, "m" syllable.
6	M	9	136	Tourette disorder Social phobia	23	None	*Motor*: Eye blinking/squinting, eye rolling/darting, head rolling, shoulder shrugs, heel buttocks kick, dragging toe on ground. *Vocal*: Dolphin sound.
7	M	10	115	Tourette disorder ADHD (inattentive) Enuresis	22	None	*Motor*: Head jerk, facial grimace, leg/feet movements, hand wave, copropraxia. *Vocal*: Words, "f" syllable, coprolalia.
8	M	12	93	Chronic motor tic disorder OCD	16	Risperidal	*Motor*: Eye blinking, eye rolling, head rolling, facial grimace, mouth/tongue movements, shoulder shrugs, leg/feet movements, finger tensing, mouth movements plus eye blink combination.
9	M	10	103	Tourette disorder	28	Guanfacine	*Motor*: Facial grimace, dropping jaw, foot up and down, kick out leg, bicep tensing, hand flick. *Vocal*: Sniffing in and out, fast breath, teeth tic.
10	M	9	118	Tourette disorder OCD ADHD (combined) ODD	17	Guanfacine	*Motor*: Eye rolling, knuckle cracking. *Vocal*: Grunt, sniffing, coughing, repeating commercial songs.

and a condition in which the child was instructed to suppress his tics, but received no reinforcement for doing so (SUP-No Reinforce-Orange). Each condition was 5 min in duration. In summary, each child was exposed to 12, 5-min BL conditions; 12, 5-min SUP-Purple conditions; and 12, 5-min SUP-No Reinforce-Orange conditions over the course of Sessions 2–5. The conditions are described in greater detail below.

Baseline (BL)

During all BL conditions, the child was seated in the observation room by himself with a token dispenser (a $12' \times 12' \times 24'$ box with an attached, non-functioning internet camera and a clear plastic receptacle attached to the front) placed in front of him. Two colored lights (orange and purple) were on the device, but neither light was illuminated. Following the protocol established by Woods and Himle (2004), the child was told the machine was a "tic detector" that had the ability to monitor and count the child's tics (note: the device was manually operated by the investigator from behind a one-way mirror). The child was asked to sit in front of the machine alone for 5 min. Prior to leaving the room, a male research assistant instructed the participant to freely tic as much or as little as needed and to ignore the "tic detector" as much as possible while remaining in his seat. No tokens were delivered during the BL condition. At the end of each BL session, the child was asked to provide an overall estimate of premonitory urge intensity experienced in the previous 5 min.

Reinforced suppression in presence of a purple light (SUP-Purple)

During the SUP-Purple condition, the child was seated in a room with the aforementioned token dispenser placed directly in front of him. Two colored lights (orange and purple) were on the token dispenser, but only the purple light was illuminated. The child was told that the device was a "tic detector," that would monitor and count his tics. To produce suppression, the child was told that he could earn a token for each 10-s interval during which he had no tics (note: the child was not told how tics could be suppressed). The child was told that each token was exchangeable for a small monetary reward upon completion of the study (*note: for ethical reasons, the child always received the same amount of money (i.e., a $20 gift certificate in addition to the participation payment) at the end of the study regardless of the number of tokens earned*). The investigator then provided a detailed overview of the tics that the child was to suppress (including each tic described on the YGTSS and each tic observed during the 10 min preliminary data collection observation during Session 1). The child was asked to repeat the instructions to ensure that he understood the task. During the suppression condition, the "tic detector," was controlled by a research assistant observing from behind a one-way mirror. For each consecutive 10-s interval during which the child did not have a tic, a token was delivered into a clear receptacle on the front of the token dispenser. In the event that a tic occurred in a given interval, the 10-s interval was reset without token delivery. If tics occurred that were not targeted for suppression (i.e., "tic substitution"), such tics were noted so that they could be later coded, but they did not prevent the delivery of tokens. At the end of each 5 min SUP-Purple condition, the child was asked to provide an overall estimate of premonitory urge intensity experienced in the previous 5 min.

Suppression with no reinforcement in presence of an orange light (SUP-No Reinforce-Orange)

During the SUP-No Reinforce-Orange condition, the child was seated in a room with the aforementioned token dispenser placed directly in front of him. Two colored lights (orange and purple) were on the token dispenser, but only the orange light was illuminated. The child was told that the device was a "tic detector," which would monitor and count his tics. The child was asked to attempt tic suppression during the SUP-No Reinforce-Orange condition, but informed that no tokens would be delivered for his efforts. At the end of each 5-min SUP-No Reinforce-Orange condition, the child was asked to provide an overall estimate of premonitory urge intensity experienced in the previous 5 min.

Test of stimulus control (session 6)

Session 6 occurred an average of 1 day after Session 5 (range = 0–2 days after Session 5). In this final session, the children were exposed to each of the following 5-min conditions three times in a random order. In the *Baseline-Test (BL-Test)* condition, the child was seated in the observation room by himself with the "tic detector" in front of him, with neither stimulus light illuminated. The child was told that the tic detector would be counting tics but was given no instructions about suppression. In the *Test-Purple*, condition the child was seated in the observation room by himself with the "tic detector" in front of him and the purple stimulus light illuminated. The child was told that the tic detector would be counting tics, but the child was given no instructions about suppression and did not receive tokens during the condition. Finally, in the *Test-Orange* condition, the child was seated in the observation room by himself with the "tic detector" in front of him and the orange stimulus light illuminated. The child was told that the tic detector would be counting tics, but the child was given no instructions about suppression and did not receive tokens during the condition.

Data recording and scoring procedures

Recording procedures

During all conditions, the subject was video-recorded from behind a one-way observation mirror. Parents were informed of the recording, but the children were uninformed until they were debriefed at the end of the study. At that point, children could allow the researchers to use their data or to have it destroyed immediately. All children allowed for the use of their data.

Training of coders

Coders were trained in the operational definitions and observational coding procedures using procedures established by the first author and outlined elsewhere (Himle, Chang et al., 2006; Piacentini et al., 2006).

Defining tics for scoring

Operational definitions of each tic displayed by each child were created from data gathered during administration of the YGTSS and from the 10-min videotaped sample obtained during the preliminary data collection session. Coders were trained in the operational definitions using procedures established by the first author and outlined elsewhere (Himle, Chang et al., 2006; Piacentini et al., 2006). During the course of the experimental portion, two participants (#5 and #7) displayed tics that had not been endorsed on the YGTSS or detected during the initial 10 min assessment in the first session. Participant #5 demonstrated an additional vocal tic (i.e., sniffing), and #7 demonstrated three motor tics (i.e., shoulder shrugging, finger snapping, and evening up behavior). Because the participants had not been instructed to suppress these tics, the tics were coded, but not included in the final data analyses. Nevertheless, for both participants, data on the distribution of these tics across conditions paralleled the results found with the targeted tics (i.e., lower in the SUP-Purple conditions than in either the BL or SUP-Orange conditions).

Frequency counts

Each videotape was scored for event frequency, which was subsequently transformed into a tic rate (i.e., tics per minute (TPM) by dividing the total number of tics for that condition by the number of minutes in that condition during which the child was visible to the observers). Preliminary research from a multi-site observational study has shown event frequency counts to be reliable measures of tic expression (Himle, Chang et al., 2006).

Interrater reliability

Interrater reliability was calculated using procedures outlined in earlier work by the first author (e.g., Himle, Chang et al., 2006; Piacentini et al., 2006). Twenty-four percent of the conditions were randomly sampled across all participants and scored by an independent rater. Interobserver agreement was calculated at 87% (range = 76–95%), indicating acceptable reliablity.

Measuring the premonitory urge

Using a method employed in earlier research (Himle et al., 2007), the premonitory urge was measured by asking participants to provide an overall rating of the urge experience for each 5-min condition. Children were given an "urge thermometer" printed on a sheet of paper, and asked to rate the intensity of the urge on a 0 (no urge) to 8 (intense urge) point scale.

Independent variable integrity

To assure procedural consistency and independent variable integrity, 18% of the conditions were randomly sampled and scored for correct implementation of the independent variable. Independent raters noted whether or not the researcher correctly delivered the condition instructions to the participant for all conditions. Results demonstrated that instructions had been correctly delivered in 100% of the conditions. In addition, the delivery of the reinforcer contingent on tic-free periods during all SUP-Purple conditions was recorded by calculating the time between token delivery and the previous tic. Accurate token delivery included any token delivered 10 ± 1 s after the previous tic. Results showed that tokens were delivered accurately 84% of the time, consistent with findings from other studies using the same experimental paradigm (e.g., Woods, Himle et al., 2008). In addition, during the training phases, after each 5-min condition, participants were asked to state what the instructions had been for that condition. Participants were able to accurately recite the experimental instructions in all conditions, with the exception of Participant 3, who failed to accurately restate the instructions during all three baseline conditions of Session 2. Because removal of these data points did not alter study findings, and inclusion of these data represented a more conservative analytic approach, these data points were retained in the reported analyses.

Results

Data analysis

For each subject, the mean TPM was calculated across the 12 SUP-Purple, 12 SUP-No Reinforce-Orange, and 12 BL conditions obtained during the training phase (i.e., Sessions 2–5). In addition, for each subject, the mean TPM was calculated across the 3 Test-Purple, 3 Test-Orange, and 3 BL-Test conditions obtained during Session 6. As a result, each subject ended up with six scores; SUP-Purple, SUP-No Reinforce-Orange, BL, Test-Purple, Test-Orange, and BL-Test.

Test of primary hypothesis 1

To test the first primary hypothesis, a paired-samples *t*-test was used to compare the scores of the BL and SUP-Purple conditions from the training phase. Results confirmed the hypothesis, showing that TPM in the BL condition ($M = 4.29$, $SD = 3.04$) was significantly higher than when tic suppression was reinforced in the SUP-Purple condition ($M = 1.57$, $SD = 1.71$), $t(9) = 4.30$, $p = 0.002$, $d = 1.10$.

Test of primary hypothesis 2

To test the second primary hypothesis, a paired-samples *t*-test was used to compare the scores of the BL-Test and Test-Purple conditions from Session 6. Results confirmed the hypothesis, showing that TPM in the BL-Test condition ($M = 3.98$, $SD = 2.99$) was significantly higher than in the Test-Purple condition ($M = 2.18$, $SD = 2.46$), $t(9) = 4.07$, $p = 0.003$, $d = 0.66$.

Test of exploratory hypothesis 1

To test the first exploratory hypothesis, paired-samples *t*-tests were used to compare the mean urge ratings in the BL vs. SUP-Purple and BL-Test vs. Test-Purple conditions. Results failed to support the hypothesis. During the training phase, the mean urge rating across BL conditions ($M = 2.34$, $SD = 1.69$) was not significantly different from the mean urge rating across the SUP-Purple condition ($M = 2.90$, $SD = 2.37$), $t(9) = 0.76$, $p = 0.47$, $d = 0.27$. In addition, the mean urge rating in the BL-Test condition ($M = 1.9$, $SD = 1.69$) was not significantly different from the urge rating in the SUP-Purple condition ($M = 2.40$, $SD = 2.13$), $t(9) = 0.64$, $p = 0.54$, $d = 0.26$ during the test phase.

Test of exploratory hypothesis 2

To test the second exploratory hypothesis, separate difference scores were calculated between the SUP-Purple and SUP-No Suppression-Orange conditions and the BL scores during the training phase. The result was two separate change scores, one for the SUP-Purple condition and one for the SUP-No Reinforcement-Orange condition. A paired-samples *t*-test supported the hypothesis, showing that the SUP-Purple condition produced greater decreases in TPM ($M = -2.71$, $SD = 1.99$) than the SUP-No Reinforcement-Orange condition ($M = -1.86$, $SD = 1.59$), $t(9) = 2.54$, $p = 0.03$, $d = 0.47$.

Discussion

Previous research has demonstrated that reinforcing tic suppression can effectively reduce tics and increase premonitory urges (i.e., Himle & Woods, 2005; Himle et al., 2007; Woods & Himle, 2004; Woods, Himle et al., 2008). The current study sought to replicate the findings and extend research to determine if tic and urge expression could come under the control of discriminative stimuli.

Consistent with previous research examining the impact of reinforcement on tic suppression (e.g., Himle & Woods, 2005, Woods & Himle, 2004; Woods, Himle et al., 2008), results from the current study demonstrated that when tic suppression is reinforced (i.e., SUP-Purple), tics occur at much lower rate than when tics are allowed to occur freely (i.e., BL). Similar to magnitudes found in other studies (e.g., Conelea & Woods, 2008b; Himle & Woods, 2005; Woods & Himle, 2004; Woods, Himle et al., 2008), a 63.6% reduction in TPM was found from baseline to the SUP-Purple conditions during the training phase. Results also replicated prior findings (Woods & Himle, 2004) showing that verbal instructions to suppress tics produced a lower magnitude of suppression than providing instructions to suppress plus reinforcement for successful suppression. However, in contrast with the findings of Himle et al. (2007), during tic suppression conditions, the urge was not significantly different from the BL condition.

Extending prior research, the current study also tested whether tic frequency and premonitory urges could come under the control of stimuli that predicted the availability of reinforcement for successful suppression. In terms of the impact on tic frequency, results showed that a stimulus (i.e., purple light) that had predicted the availability of reinforcement for tic suppression (even though the reinforcement was no longer delivered) evoked lower rates of tics than a stimulus (i.e., no lights) in the presence of which tic suppression was not required. In contrast, stimulus discrimination training was ineffective with respect to the premonitory urge. Results suggested that during the testing phase, urge intensity experienced in the presence of a stimulus that had predicted the availability of reinforcement for suppression was not significantly different from urge intensity during the baseline condition.

Findings from the current study contribute to the literature on tic disorders in a number of ways. First, the data provide further evidence for the potentially important role of operant variables in the expression of tics. Second, the finding that repeatedly reinforcing tic suppression in the presence of a particular antecedent stimulus results in that antecedent stimulus acquiring some control over tic occurrence provides one empirically plausible explanation for environmentally-based variability in tic expression. Extrapolating from this analogue study, it is possible that one reason tics vary from situation to situation is that each situation has come to predict the presence or absence of contingencies that make tics more or less likely to happen. As opposed to simply being a poorly understood characteristic of the disorder itself, the current study suggests that environmental variability in tic expression may be a predictable, controllable, and potentially modifiable phenomenon. Even more intriguing is that stimulus control over tics can emerge after a relatively brief exposure to contingencies, as the children in this condition received only 1 h of training in the purple condition.

The study hypotheses with respect to tics were largely supported, but the results failed to support hypotheses involving the premonitory urge. Given the findings of Himle et al. (2007), who demonstrated that reinforcing tic reduction increased the premonitory urge, it was expected that during the training phase, ratings of the urge in the SUP-Purple condition would be higher than those obtained during the BL condition. However, results showed that this was not the case and in the absence of urge differentiation between the SUP-Purple and BL conditions, the acquisition of stimulus control over the urge could not be expected.

Reasons for the failure to replicate Himle et al. (2007) are unclear, but because the means were in the predicted direction, and the study was not adequately powered to detect small differences, it is possible that with additional power the differences found in this study would be detected as significant. In addition, because the correlation between the PUTS scores and premonitory urge ratings taken during the BL training conditions were weakly correlated ($r = 0.21$, $p = 0.41$), it is possible that the measurement strategy used to assess the urge was inadequate. Nevertheless, because the premonitory urge, and the reduction of the urge following tic expression, is believed to play a functional role in behavioral model of TS (e.g., Himle, Woods, Piacentini, & Walkup, 2006), the incongruent findings from the current study should prompt researchers to conduct additional well-powered studies to examine the effects of tic suppression on the premonitory urge experience, using multiple methods of assessing the urge. Such research may provide additional clues into the behavioral mechanisms underlying tic maintenance.

The current study also had a number of limitations that should be addressed in future research. First, the small sample sizes did not allow us to examine potential moderators. For example, it would have been interesting to determine whether or not a particular comorbid diagnosis (i.e., ADHD or OCD) predicted greater or poorer acquisition of stimulus control. Second, it would have been interesting to explore the maintenance of these results over time. Although a repeated measures ANOVA across the three purple conditions during the testing phase did not show a statistical difference, $F(2, 18) = 1.22$, $p = 0.32$, it is possible that over the course of many days, the stimulus control effect would have diminished. Future research should be conducted to establish the durability of stimulus control and to estimate the decay of effect as a function of time spent in training. Third, the current study was conducted in an analogue, highly controlled setting. The generalizability of the current findings to "real-world" settings remains in question and should be addressed in future research. Should future studies in "real-world" settings replicate the current findings, it would have a number of clinical implications. For example, behavior therapy for TS is typically done using only one or two specific support people to facilitate the acquisition of tic suppression skills, and this is usually in the home context (Woods, Piacentini et al., 2008). Should the current studies be replicated, it would suggest that behavior therapy structure multiple environments (i.e., home, school, athletic and social settings) to yield contingencies for successful tic suppression. Such strategies would likely yield greater overall tic reduction.

As the biological nature of tic etiology is clear (Leary et al., 2007; Swain et al., 2007), the emerging behavioral model of tics does not offer environmental variables as an etiological cause of the disorder (Woods et al., 2006). Rather, behavioral models have been developed to explain (a) how tics can be influenced by fluid environmental factors (Conelea & Woods, 2008a), (b) how the influence of such factors can be effectively understood, predicted and controlled (Woods, Piacentini, & Walkup, 2007), and (c) how these factors can be utilized to bring about effective tic management in the form of behavior therapy (Woods, Piacentini et al., 2008). Although the current study represents another step in the development of a plausible behavioral model for tic disorders, many questions remain, and it is our hope that they will be answered through rigorous experimental study.

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