

Durability, Negative Impact, and Neuropsychological Predictors of Tic Suppression in Children with Chronic Tic Disorder

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Abstract Chronic tic disorders are characterized by involuntary motor and vocal tics, which are influenced by contextual factors. Recent research has shown that (a) children can suppress tics for brief periods of time, (b) suppression is

enhanced when programmed reinforcement is provided for tic-free intervals, and (c) short periods of suppression do not result in a paradoxical “rebound” in tic frequency when active suppression has ceased. The current study extended existing research in three important ways. First, we examined whether tic suppression ability decreased as suppression duration increased from 5 to 25 to 40 min. Second, we examined post-suppression tic frequency to test whether longer periods of suppression were more likely to be associated with a rebound effect. Finally, we explored neuropsychological predictors of tic suppression. Thirteen children with Tourette syndrome or a chronic tic disorder completed the study. Results showed that (a) tic suppression was sustained for all of the suppression durations, (b) rebound effects were not observed following any of the suppression durations, and (c) ability to suppress was correlated with omission, but not commission errors on a continuous performance task. Implications of these findings are discussed.

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Chronic tic disorders (CTDs), including Tourette syndrome (TS), are characterized by involuntary movements and vocalizations (i.e., tics). TS occurs in 0.5% of children, and prevalence rates are slightly higher for chronic motor or vocal tic disorders (Khalifa and von Knorring 2006). Tics are considered involuntary, but can be partially suppressed using both pharmacological and behavioral therapies (Himle et al. 2006b). Medication traditionally has been the first line treatment, but there has been growing interest and outcome research directed toward behavioral approaches (e.g., Deckersbach et al. 2006; Himle et al. 2006b; Wilhelm et al. 2003; Woods et al. 2003). Behavioral treatments are founded on the premise that tics can be exacerbated or

reduced by systematic changes to the environment. These changes include eliminating contextual stimuli that exacerbate tics and/or teaching individuals tic suppression strategies.

To date, several uncontrolled studies have shown a link between tic frequency and contextual factors. For example, Malatesta (1990) reported a case in which a child's tics were more pronounced in the presence of his father, and O'Connor et al. (2003) found that the most common "high risk" (of tics occurring) situations were sedentary activities that involved waiting (e.g., waiting in line), transitioning (e.g., transitioning between appointments or going to/from work), and social activities. These results are consistent with findings reported by Silva et al. (1995), who showed that 42% of persons with TS surveyed reported tic exacerbations in social gatherings. Although these uncontrolled studies demonstrate that various antecedent stimuli *can* influence tic expression, there has been surprisingly little experimental research done to determine *how* these contextual influences obtain their effect. Such information may be particularly important as it may lead to improved behavioral treatments and a more comprehensive understanding of the mechanism (s) by which behavioral interventions are effective.

Recent research has begun to explore tic suppression/expression in controlled laboratory settings. Meidinger et al. (2005) compared the rate of tics during a no-suppression baseline condition to a condition in which participants were instructed to suppress their tics. Using a reversal design (Barlow and Hersen 1984), five participants (adults and children) were recorded during multiple presentations of the baseline and suppression conditions. Successful suppression was defined as a statistically significant difference between the baseline and verbal instructions (to suppress) conditions (with baseline showing higher tic rates). Results showed that participants were able to successfully suppress their tics during an average of 44% of sessions.

Woods and Himle (2004) proposed that "real world" tic suppression is likely to yield reinforcing consequences (e.g., avoidance of teasing), and subsequently developed an analogue paradigm to study tic suppression, which involved the delivery of reinforcers contingent on successful suppression. In one condition, children with TS were instructed to suppress their tics. In a second condition, the same suppression instructions were provided, and token reinforcers (exchangeable for preferred items) were delivered contingent upon 10 s tic-free intervals. Using a reversal design across four participants, results clearly demonstrated the superiority of the reinforcement procedure compared to the instruction-only condition. During the reinforcement condition, tics were reduced by an average of 76.3% from baseline versus an average of 10.3% reduction in the instruction-only condition. These results suggested that tic suppression was enhanced when environmental consequences were delivered contingent upon effective suppression.

In another study, Himle and Woods (2005) sought to replicate the results of the Woods and Himle (2004) and to evaluate clinical observations suggesting that tic suppression may create a paradoxical *increase* in tics (compared to baseline) after tic suppression has ceased (termed the "rebound effect"; e.g., Burd and Kerbeshian 1987). Indeed, various authors have discussed the rebound phenomenon (e.g., Bruun and Bruun 1994; Burd and Kerbeshian 1987) and surveys have shown that up to 77% of professionals who treat those with TS believe the rebound effect occurs (Marcks et al. 2004), although it has not been tested in any controlled fashion.

Using a within-subjects design, Himle and Woods (2005) examined the rebound effect in seven children with TS. Children were observed during a 10-min baseline phase followed by a 5-min phase in which they were instructed to suppress their tics and received reinforcers for brief tic-free intervals. The 5-min suppression phase was immediately followed by a 5-min "rebound evaluation" phase in which the children were instructed to no longer suppress their tics and to "feel free to tic" if they needed. The suppression and rebound phases were replicated within each participant. Results showed that the tic suppression phases produced a mean 70% reduction in tics from baseline and tics returned to baseline levels during the rebound phase. No evidence was found to support the notion of a rebound in tic frequency following 5 min of suppression.

The studies by Woods and Himle (2004) and Himle and Woods (2005) demonstrated that children were able to suppress their tics for brief periods of time when suppression was reinforced and that brief suppression was not necessarily followed by a significant increase in tic frequency. Nevertheless, a number of questions remain. First, it is unclear whether children were able to suppress their tics for longer periods of time, even when reinforced for doing so. Second, given the strong clinical belief in the rebound phenomenon, it is plausible that rebound effects may occur only following longer periods of attempted suppression. Finally, there is considerable variability in the degree to which individual children are able to suppress their tics, and the individual differences that may predict successful suppression are unclear.

Individual differences in attentional ability and impulsive responding/response inhibition have been implicated in tic suppression ability. Evidence implicating the role of attentional processes comes from two sources. Peterson et al. (1998) found that during tic suppression tasks in 22 adults with TS, neural activity (as measured via fMRI) decreased in the basal ganglia and increased in the somatosensory and attention-related cortical regions (Peterson et al. 1998). Likewise, Himle and Woods (2005) showed that increasing attentional problems on the CBCL correlated with poorer suppressibility. Fewer studies have investigated the

role of impulsive responding or response inhibition as a factor in tic suppression, but Deckersbach et al. (2006) demonstrated that poor performance on a response inhibition measure predicted poor response to behavior therapy for tics in adults. Such results imply that both attentional processes and response inhibition deficits may be related to poor tic suppression.

Despite the existing research demonstrating the impact of environmental variables on tic suppression and the possibility that suppression may involve attentional and response inhibition processes, a number of questions remain. First, it is unclear whether tic suppression ability under periods of reinforced suppression can be sustained. As primary hypothesis 1, we predict that reinforced tic suppression durations of 5, 25, and 40 min will produce lower tic rates than a no suppression baseline condition. Second, it is unclear if tic suppression ability degrades with longer periods of suppression. As primary hypothesis 2, we predict that because tic suppression may require sustained effort, the rate of tics in 5-, 25-, and 40-min periods of suppression will linearly increase. Third, it may be the case that while brief periods of suppression do not create subsequent rebounds in tic frequency, the likelihood of rebound increases with duration of suppression. As primary hypothesis 3, we predict that tic rates in the 25- and 40-min rebound conditions will be significantly higher than the baseline condition. Finally, because prior research suggests that attentional and response inhibition deficits may be related to tic suppression, we test the exploratory hypothesis that more impaired performance on a continuous performance test (commission and omission errors) will be inversely related to tic suppression ability.

Method

Participants

All data were collected by researchers at North Dakota State University (NDSU) and Western Michigan University (WMU). To obtain broad sample representation, recruitment took place through print advertisements placed in newspapers in Fargo, ND and Kalamazoo, MI. Children were eligible to participate if they met the following criteria: (1) generally healthy males or females between the ages of 10 and 17; (2) a diagnosis of TS; (3) intellectual functioning in the low-average range or above as indicated by a score of greater than 75 on the Wechsler Abbreviated Scale of Intelligence; and (4) no history of behavioral treatment for tics or other treatment in which suppression strategies were a primary component of the intervention. Children were allowed to be on psychotropic medication, though they were required to have been on a stable dose for

4 weeks prior to participation. Fourteen children, ages 10–17, with TS or chronic motor or vocal tic disorder were recruited to participate in the study. Data for one participant were unusable due to recording error. CPT data were unavailable for 2 of the remaining 13 due to a computer malfunction.

Of the 13 remaining children, 7 were collected at NDSU and 6 were collected at WMU. Demographic characteristics are described in Table 1. Of note, all were Caucasian and 69% had at least one comorbid psychiatric disorder, the most common of which were attention-deficit/hyperactivity disorder (46.2%) and generalized anxiety disorder (38.5%). Forty-six percent were on psychotropic medications. The mean Yale Global Tic Severity Scale total tic score was 28.2 (SD=5.3), indicating moderate severity.

Measures

The Anxiety Disorders Interview Schedule for DSM-IV, Research and Lifetime Version for Children and Parents (ADIS-RLV) The ADIS-RLV is an expanded version of the Child Version of the ADIS (Silverman and Albano 1996). The ADIS-RLV is a semi-structured diagnostic interview that assesses the major *DSM-IV* anxiety, mood, and externalizing disorders along with a range of psychiatric disorders affecting youth. In the present study, only current disorders were assessed. The ADIS-IV, on which the ADIS-RLV is based, possesses favorable psychometric properties including acceptable test–retest reliability ($\kappa=0.76$) and high interrater reliability ($r=0.98$ and 0.93 for parent and child interviews, respectively; Silverman and Eisen 1992;

Table 1 Means and SDs for the sample characteristic data across comorbidity status

	CTD-alone N=4	CTD+co- occurring N=9	Overall N=13
Gender			
Males	4	8	12
Females	0	1	1
Mean age	10.0 (2.0)a	12.1 (2.8)a	11.5 (2.7)
IQ	103 (11.3)a	107 (14.6)a	106 (13.2)
CY-BOCS	6.8 (8.6)a	14.6 (7.7)a	12 (8.5)
YGTTSS			
Overall total	19.2 (4.1)a	29.33 (5.6)a	28.2 (5.3)
CBCL			
Internalizing	52.0 (13.3)	59.9 (9.3)	57.46 (10.8)
Externalizing	56.3 (14.7)	59.8 (11.1)	53.38 (11.9)
Total problems	56.0 (15.5)	59.8 (11.1)	58.62 (12.0)
CPT			
Omissions	69.8 (10.9)a	49.4 (5.5)b	54.99 (12.0)
Commissions	52.2 (6.6)	49.2 (6.1)	50.00 (6.04)

Reading from left to right for each row, different letters indicate significant mean differences at $p<.05$.

Silverman and Nelles 1988; Silverman and Albano, *Anxiety Disorders Interview Schedule for DSM-IV, Research and Lifetime Version for Children and Parents*, unpublished).

Child Behavior Checklist (CBCL) The CBCL, a 118-item parent-report measure of child psychopathology, is one of the most extensively tested and normed rating scales available and possesses excellent psychometrics (Achenbach 1991). *T* scores allow for normative comparisons across three broadband factors (social competence, and internalizing and externalizing behavior problems) and eight subscales measuring various domains of psychiatric symptoms (Achenbach 1991).

Wechsler Abbreviated Scale of Intelligence (WASI) This measure is used to assess intellectual functioning in persons aged 6–89 years (The Psychological Corporation 1999). The two-subtest version of the WASI was used and has a mean score of 100 with a standard deviation of 15. The two-subtest version has shown adequate split-half reliability ($r=0.95$), temporal stability ($r=0.85$), and concurrent, content, and construct validity (The Psychological Corporation 1999).

Children's Yale–Brown Obsessive–Compulsive Scale (CYBOCS) The CYBOCS is a semi-structured clinical interview assessing obsessive-compulsive disorder (OCD) severity and change over time in youth ages 6–17 (Scahill et al. 1997). The child version was adapted from the adult Yale–Brown Obsessive–Compulsive Scale, which also has good psychometric properties (Goodman et al. 1989). The CYBOCS consists of a checklist and a rating scale, each of which is divided into obsessions and compulsions. The checklists contain commonly endorsed obsessions and compulsions, and the rating scales include ratings of frequency/duration, interference, distress, resistance, and control related to OCD symptoms. Separate scores, ranging from 0 to 20, are obtained for obsessions and compulsions, with a combined total of 0 to 40.

Yale Global Tic Severity Scale (YGTSS) The YGTSS is a clinician-completed measure that provides 0–5 point ratings on five different dimensions: tic number, frequency, duration, intensity, and complexity (Leckman et al. 1989). Each of the dimensions is scored separately for motor and vocal tics to produce total motor tic and total vocal tic severity scores. The motor and vocal tic severity scores are combined to produce a total tic severity score ranging from 0 to 50, with higher numbers indicating more severe tics. The YGTSS has demonstrated acceptable internal consistency and acceptable convergent and divergent validity (Leckman et al. 1989). Psychometric analyses have shown high correlations between the motor and vocal tic subscales

and the global score, high interrater reliability (intraclass correlation coefficient for the total tic score=0.84), and factor analyses have revealed a two-factor structure (motor tics and vocal tics; Leckman et al. 1989).

Conners' Continuous Performance Test (CPT II) The CPT is a commonly used, normed cognitive task in which subjects respond on most trials, with response inhibition required on rare target trials (Conners and Multi-Health Systems Staff 2002). Two particular CPT variables were examined in the current study; omission errors and commission errors. Omission errors suggest poor orientation toward incoming stimuli, hence reflect poor attentional functioning. Commission errors are believed to be a specific measure of response inhibition, and high scores suggest more impulsive responding. Adequate reliability and validity information on the CPT scores has been reported (Halperin et al. 1991). Scores are reported as *T* scores with higher scores indicating more impaired performance.

Procedures

The procedures for this study occurred across two sessions, which were conducted on consecutive days. The first session included the assessment and preliminary data collection. The second session involved the experimental procedures. The study procedures are described in detail below.

Assessment and preliminary data collection All interviews and assessment instruments were administered by advanced graduate students with training in *DSM-IV* diagnostic criteria and clinical interviewing. The examiners were trained in the specific assessment instruments by doctoral-level psychologists (D.W.W., R.G.M., J.E.C.) during a cross-site meeting prior to beginning the study.

Participants and their parents were asked to attend the initial assessment session to determine eligibility for the study. At the first session, examiners obtained (a) consent/assent (all consented/assented to being videotaped), (b) background information about the participant, (c) a history of the tic disorder and its treatment, and (d) general demographic information. In addition, participants were asked to complete the WASI and the CPT. Parents completed the CBCL and the CPRS. The evaluator administered the ADIS, CYBOCS and the YGTSS with the participants and parents. After all interviews and rating scales had been completed, children were videotaped alone in a room for 10 min to obtain a videotaped sample of their tics. After the observation was completed, the evaluators and site supervisors watched the videotape and derived operational definitions of each tic displayed during the observation.

Experimental Conditions

All experimental conditions took place the day after the initial assessment. Each child was exposed to each of three general conditions: baseline, reinforced suppression, and rebound evaluation. The procedures were conducted identically at the NDSU and WMU sites and are described in more detail below.

Baseline condition (BL) A baseline-recording phase was conducted at the beginning of the experimental session. During this observation, the child was seated alone in the observation room in front of a token dispenser (a 12"×18"×12" metal box with a non-functioning Internet camera on top and a clear plastic receptacle attached to the front). Using the protocol outlined in Woods and Himle (2004), the child was told that the machine was a "tic detector" and that it was able to monitor and count tics (the device was manually operated by the investigator from another room). The child was asked to sit in front of the machine for 10 min. In addition, he/she was told to tic as much or as little as needed and to ignore the machine as much as possible while remaining seated. No instructions to suppress were delivered, and no tokens were delivered during BL.

Suppression conditions Utilizing a within-subject design, tic suppression and rebound effects were examined in two phases: a *suppression* phase and 5 min *rebound evaluation* phase. Three durations of suppression: 5, 25, and 40 min were evaluated. Each suppression duration was immediately followed by a 5-min rebound evaluation phase. Each participant received each of the three suppression durations, with the suppression durations delivered randomly to control for practice effects. Suppression was created using the preparation described by Woods and Himle (2004). In this procedure, the child was seated in a room in front of the token dispenser. The child was reminded that the device was a "tic detector" that would monitor and count his/her tics. To produce suppression, the child was told that he/she could earn a token for each 10 s interval during which he/she refrained from having tics and that the tokens were exchangeable for money (a few cents each) upon completion of the study. The investigator then provided the child with a detailed overview of each of his/her tics that were to be targeted for suppression (including each tic described on the YGTSS and each tic observed during the preliminary data collection observation). The child was asked to repeat the instructions to ensure understanding. A research assistant, who was observing from behind a one-way mirror or via close-circuit camera/TV, controlled the token dispenser. For each 10 s interval during which the child did not tic, the investigator delivered a token into a visible receptacle on the front of the token dispenser.

During the *rebound evaluation* phase, the child was told that the token dispenser was turned off and that although the machine would monitor and count his/her tics, it would no longer deliver tokens. In addition, the child was told to tic as much or as little as needed. Upon completion of the study, all children received a pre-determined amount of compensation, regardless of the number of tokens earned.

Data Coding

All videotape scoring was conducted at the University of Wisconsin-Milwaukee (UWM) by research assistants trained in direct observation procedures by the first author (DWW). Operational definitions for each tic exhibited by each child were generated based on information obtained (1) during the interview, (2) upon review of the observation from the preliminary data collection, and (3) by noting tics during the experimental procedure.

All videotaped observations were scored using the event (frequency) recording method. Based on information obtained during the initial assessment and review of the initial 10-min videotapes by the site-specific primary investigators, a list of operationally defined tics was created for each participant. Each condition was then randomly ordered for each participant and scored for tic occurrence by the UWM research assistants, who were blind to segment number. Each segment was scored separately for motor tics, vocal tics, and total tics. To score the videotapes, a research assistant watched each segment on a standard television. Each tic was recorded using a simple computer program capable of recording and time-stamping computer keystrokes. As the research assistant watched the videotape, he/she indicated the presence of each tic by pressing a designated key on a laptop computer. After the segment had been scored, the computer provided an output listing each keystroke (corresponding to each tic) and the precise timing of each keystroke, sensitive to a tenth of a second. A separate key was also used to designate the onset and offset of any moments during which the child was not in full camera view (e.g., turned away from the camera, left the chair). This procedure yielded a direct frequency measure, which was used to calculate the rate of tics per min. Preliminary research has shown frequency counts to be a reliable measure of tic expression (Himle et al. 2006a).

Interobserver reliability A second observer at UWM independently scored 23% of the tapes using the same scoring methods. A raw agreement score (lowest frequency/highest frequency×100%) was calculated for motor tics, vocal tics, and total tics, yielding a percent of total agreement. Mean agreement on the occurrence of motor tics was 81.1%, for vocal tics was 83.6%, and for total tics

was 81% (range=0–100%¹). These data suggest that the direct observation was reliable across observers.

Independent Variable Integrity

To determine the integrity of reinforcer delivery, all suppression conditions were scored for the correct delivery of tokens during suppression conditions. Accurate token delivery was defined as any token delivered within 10±2 s of the previous token (if there were no intermediate tics) or the previous tic. Data indicated that across all suppression conditions (5, 25, and 40 min), tokens were delivered accurately 84% of the time. Analyses were conducted to determine whether longer periods of suppression yielded poorer independent variable integrity. Results showed that the 5, 25, and 40 min suppression conditions did not differ with respect to IV integrity, $F(2, 22)=0.18, p=ns$.

Results

Tests of Suppression Effects

To test whether suppression occurred across all durations and to assess whether comorbid status impacted tic suppression ability, a 2 (CTDs-alone vs CTDs+comorbid psychiatric condition(s))×4 (baseline, 5, 25, and 40 min suppression conditions) mixed ANOVA was run using tics per minute (TPM) as the dependent variable. Results showed no main effect of comorbid status, $F(1,11)=0.184, p=0.68, \eta^2=0.016$. The tic rate for the CTDs-alone group across the four conditions ($M=4.68, SE=1.43$) was similar to the tic rate for the CTDs+comorbid conditions group ($M=3.95, SE=0.95$).

Results did demonstrate a main effect of condition, $F(3,33)=8.73, p=0.000, \eta^2=0.44$. Follow-up comparisons demonstrated that the 5 min ($p=0.004, d=1.24$), 25 min ($p=0.027, d=0.97$), and 40 min ($p=0.001, d=1.22$) suppression conditions yielded significantly fewer TPM than BL. Comparisons between the three different suppression conditions yielded no significant differences. The interaction between comorbid status and condition was not significant, $F(3, 33)=1.06, p=0.38, \eta^2=0.09$. Table 2 contains the means and SDs across the BL and suppression conditions.

As predicted, results showed that tic rates were significantly lower than BL in all three suppression durations (5,

¹ The reported 0% inter-observer reliability coefficient represents a case in which one rater observed and recorded a single tic during a suppression condition whereas the second rater observed zero tics during that same segment.

Table 2 Mean tics per minute across site and condition

	CTD-alone N=4	CTD+co-occurring N=9	Overall N=13
Baseline	7.3 (6.3)	9.3 (5.2)	8.7 (5.4)
5 min suppression	3.8 (4.8)	1.4 (1.5)	2.2 (2.9)
25 min suppression	4.1 (4.1)	3.2 (4.3)	3.5 (4.1)
40 min suppression	3.5 (2.6)	1.9 (1.6)	2.4 (2.0)
5 min rebound	12.9 (8.8)	8.4 (8.3)	9.7 (8.4)
25 min rebound	10.0 (11.6)	7.5 (5.5)	8.3 (7.4)
40 min rebound	7.9 (5.7)	8.0 (8.2)	8.0 (7.3)

25, and 40 min). However, contrary to our hypotheses, there were no significant differences between the three different durations of suppression. Participants were able to suppress as well in the 40- and 25-min conditions as in the 5-min condition. In sum, these results suggest that reinforced tic suppression is a durable phenomenon, which can last for at least 40 min. To test for site effects, a separate 2 (WMU vs NDSU)×4 (BL, 5, 25, and 40 min) ANOVA was conducted and yielded similar findings.² The effects of co-occurring psychotropic medication status were also explored in a 2 (medicated vs nonmedicated)×4 (BL, 5, 25, and 40 min) ANOVA and produced similar results.³ Finally, to explore the relationship between tic severity and tic suppression ability, a correlation was calculated between YGTSS total tic severity scores and the mean percent reduction (from baseline) of tics across the suppression conditions. The resulting correlation was small and nonsignificant ($r(13)=-0.20, p>0.05$).

Tests of Rebound Effects

To test whether rebound occurred following all durations of suppression, a 2 (CTDs-alone vs CTDs+comorbid psychiatric conditions)×4 (Baseline, 5, 25, and 40 min rebound conditions) mixed ANOVA was calculated using TPM as the dependent variable. Results showed no main effect of comorbid status, $F(1,11)=0.11, p=0.75, \eta^2=0.01$. Results also demonstrated no main effect of condition, $F(3,33)=0.66, p=0.58, \eta^2=0.06$, nor a significant interaction

² We ran a 2 (WMU vs NDSU)×4 (BL, 5, 25, 40 min) ANOVA for the suppression data. Results showed a main effect of site, $F(1,10)=6.22, p=0.03$, with the tic rate across the four conditions at NDSU being significantly higher than the tic rate from WMU participants. The interaction between site and condition was not significant, $F(3, 30)=0.13, p=0.94$. The results of the main effect of suppression were similar to those reported in the body of the paper.

³ We ran a 2 (medicated vs nonmedicated)×4 (BL, 5, 25, 40 min) ANOVA for the suppression data. The suppression data showed no main effect of med status, $F(1, 11)=0.13, p>0.05$, nor a med status×condition interaction, $F(3,33)=0.39, p>0.05$. The results of the main effect of suppression were similar to those reported in the body of the paper.

between comorbidity and condition, $F(3, 33)=0.96$, $p=0.42$, $\eta^2=0.08$. Table 2 contains the means and SDs across the BL and rebound conditions.

In contrast to our predictions, results showed no significant differences between the 5, 25, and 40 min rebound conditions and BL. Such a pattern suggests that a rebound in tic frequency did not occur, nor did it become more likely following periods of longer suppression.

To test for site effects, a separate 2 (WMU vs NDSU) \times 4 (BL, 5, 25, and 40 min) ANOVA was conducted and yielded similar findings.⁴ The effects of co-occurring psychotropic medication status were also explored in a 2 (medicated vs nonmedicated) \times 4 (BL, 5, 25, and 40 min) ANOVA and produced similar results.⁵ Finally, to explore the relationship between tic severity and tic rebound ability, a correlation was calculated between YGTSS total tic severity scores and the mean percent reduction (from baseline) of tics across the rebound conditions. The resulting correlation was small and nonsignificant ($r(13)=0.19$, $p>0.05$)

Neuropsychological Predictors of Tic Suppression

To explore the relationships between tic suppression, attentional problems, and response inhibition, we calculated Spearman correlations between scores on the CPT (omissions and commissions) and the percent of tic reduction (from Baseline) seen in the 40 min suppression condition. Higher numbers indicated greater reduction of tics from baseline. The 40 min condition was chosen as we felt it would provide the most reliable estimate of tic suppression ability.

Results showed a significant correlation between the CPT omissions score and the percentage of tic reduction, $r(11)=-0.63$, $p<0.05$, suggesting that poor orientation to stimuli were related to poorer suppression. In contrast, the correlation between CPT commissions score and tic suppression was not significant, $r(11)=-0.06$, $p=0.87$. Combined, these results suggest that orientation to a task, but not impulsive responding was related to tic suppression ability.

⁴ We ran a 2 (WMU vs NDSU) \times 4 (BL, 5, 25, 40 min) ANOVA for the rebound data. Results showed no main effect of site, $F(1,11)=3.99$, $p=0.07$. Results showed no significant interaction between site and condition, $F(3, 33)=1.0$, $p=0.40$. The results of the main effect of rebound were similar to those reported in the body of the paper.

⁵ We ran a 2 (medicated vs nonmedicated) \times 4 (BL, 5, 25, 40 min) ANOVA for the rebound data. There was no main effect of med status, $F(1, 11)=0.05$, $p>0.05$, nor a med status \times condition interaction, $F(3,33)=1.65$, $p>0.05$. The results of the main effect of rebound were similar to those reported in the body of the paper (Table 1).

Discussion

There has been a growing interest in the use and efficacy of nonpharmacological interventions for tic disorders (Himle et al. 2006b). Two approaches with the strongest empirical basis are teaching and reinforcing tic suppression strategies (e.g., Habit Reversal Training) and modifying environmental variables believed to worsen tics (e.g., function-based treatments). Unfortunately, little work has been done to understand how environmental factors (e.g., reinforcement) influence tic expression. The current study replicates earlier findings showing that instructions to suppress plus reinforcement for tic-free periods can produce substantial decreases in tic frequency (Himle and Woods 2005; Woods and Himle 2004) and extends this earlier work by showing that the effects can be maintained for at least 40 consecutive minutes. In addition, consistent with previous studies (e.g., Himle and Woods 2005), the current study failed to support the notion that attempts to suppress tics necessarily results in a post-suppression worsening of tics (i.e., a rebound), even when suppression is maintained for up to 40 min.

The implications of, and future directions stemming from these findings are important for a number of reasons. First, the current study replicates and extends previous findings showing that environmental variables can have a substantial effect on tic expression. Second, these findings suggest that an important area of future investigation may involve understanding the interactions between reward circuitry and neural circuitry involved in tics and the inhibition of movements. Third, it is well understood that contextual stimuli, which predict the delivery of reinforcers, can acquire the ability to control the expression of a reinforced behavior. Determining the extent to which tic suppression can come under the control of specific contextual features (e.g., discriminative stimuli) may begin to explain the wide situational variation exhibited by many individuals with TS. Fourth, future studies should determine which features of reinforcement are responsible for the decreases in tic frequency. It is possible that the stimuli delivered as reinforcers (i.e., the tokens) serve an attention-enhancing function such that increased attention to tic suppression is present when receiving reinforcement. It is also possible that the contingency simply strengthens competing behaviors, while doing little to the tics themselves. Clearly, more research is needed to better understand the mechanism by which contextual stimuli influence tics.

Fifth, the current study failed to support the widely held clinical belief that teaching tic suppression results in a rebound in tic frequency (Marcks et al. 2004). The question remains, however, of how to interpret the current findings without ignoring the numerous clinical reports of rebound effects. In our clinic, many parents report that their children

tic frequently when they return home after school, whereas teachers observe relatively few tics throughout the day. Often, parents attribute this scenario to the ill effects of suppression and the emergence of a subsequent rebound. Although the results of the current study run contrary to such observations, a number of possible explanations for the discrepancy could be offered. First, it may be the case that 40 min of suppression was not adequate to produce a rebound effect. Perhaps hours of suppression are needed to produce a detectable rebound in tics. As a second explanation, it is possible that the children in the current study were still inhibiting their tics to some degree during the rebound conditions because of implicit social pressures to suppress. If this is the case, the removal of such social pressures (i.e., when the child is at home in a familiar and supportive environment) may lead to a rebound following suppression. A third explanation is that suppression may not lead to a rebound in tic frequency, but rather changing environments (i.e., transitioning from school to home) has a tic exacerbating effect. A final explanation is that the child's tic frequency at home is truly his/her "baseline" level of tics, and the child has fewer tics at school because such suppression avoids teasing from peers. Thorough observations conducted across multiple contexts might help to clarify the discrepancy in the rebound phenomenon between clinical observations and findings in the current study.

In addition to extending previous work on reinforcement's role in tic suppression/expression, the current study also explored the relationship between attentional functioning, impulsive responding, and tic suppression ability. Although, the sample size limits the reliability of the findings, decreased tic suppression ability was associated with poorer attentional functioning. Specifically, the ability to orient to a task. In contrast, impulsive responding was not related to tic suppression abilities. These results are clearly preliminary, but if replicated in future research, they could have a number of implications. For example, the findings may suggest that behavioral interventions for tics should include modules for increasing focus on tic suppression tasks. Likewise, the results may imply something about the process by which tics are suppressed. If successful suppression depends on early awareness of a premonitory urge, as some have suggested (e.g., Leckman et al. 1993), then those who orient poorly to stimuli (even internal stimuli) may ultimately be poor at tic suppression. Clearly these implications are speculative and await further clarification through future research. In such research, efforts should be made to look more carefully at the attentional and response inhibition constructs as they relate to tic suppression ability. Larger sample sizes and the use of multiple measures of both the attentional and response inhibition constructs would be helpful in providing a more complete understanding of the relationship.

The current study also had a number of limitations that warrant mention. First, the sample size was small. Although the magnitude of the suppression effects made it possible to detect significant differences, the power to detect more subtle effects (e.g., rebound effects) was likely much lower. Nevertheless, this is not of great concern as the effects in the rebound comparisons were small and in the direction opposite of that which would cause clinical concern. A second limitation was the use of an indirect method of assessing tic suppression. We can only speculate that children were engaging in a competing, effortful, tic incompatible behavior during the suppression task. Because tic frequency was the dependent measure used in the study, suppression was defined simply as a reduction in tics, without regard to how the reduction was achieved. Future research should consider alternative measures (e.g., electromyographic recording or more sensitive observational recording) that may help generate hypotheses about particular suppression behaviors. As a third limitation, we relied only on tic rate as the dependent variable, effects on other relevant dimensions of tic responding including duration, intensity, or complexity were not considered. Fourth, the current study did not assess the impact of reinforced suppression on the premonitory urge experience. Individuals often report unpleasant somatic sensations prior to the onset of tics that are relieved by performance of the tic and are worsened by tic suppression. Future research examining other dimensions of tics and premonitory urges will be useful in forming a complete picture on the effects of reinforcement on tic expression. Finally, the current study only examined the impact of one environmental variable (differential reinforcement for the absence of tics) on tic expression. The impact of a host of behavioral variables remains to be systematically explored. It is our hope that researchers continue to conduct the basic and preclinical work designed to enhance our understanding of TS and nonpharmacological treatment options.

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References

- Achenbach, T. M. (1991). *Manual for the child behavior checklist/4-18 and 1991 profile*. Burlington: University of Vermont Press.
- Barlow, D. H., & Hersen, M. (1984). *Single case experimental designs: Strategies for studying behavior change* (2nd ed.). Boston: Allyn and Bacon.

- Burd, L., & Kerbeshian, J. (1987). Treatment-generated problems associated with behavior modification in tourette disorder. *Developmental Medicine and Child Neurology*, *29*, 831–832.
- Bruun, R. D., & Bruun, B. (1994). *A mind of its own: Tourette's syndrome: a story and a guide*. New York: Oxford University Press.
- Conners, K. J., & Multi-Health Systems Staff (2002). *Conners' continuous performance test (CPT II)*. North Tonowanda, NY: Multi-Health Systems.
- Deckersbach, T., Rauch, S., Buhlmann, U., & Wilhelm, S. (2006). Habit reversal versus supportive psychotherapy in Tourette's disorder: A randomized controlled trial and predictors of treatment response. *Behaviour Research and Therapy*, *44*, 1079–1090.
- Goodman, W., Price, L., & Rasmussen, R. A., Mazure, C., Fleischmann, R. L., Hill, C. L., et al. (1989). The Yale–Brown obsessive–compulsive scale: Part I. Development, use and reliability. *Archives of General Psychiatry*, *46*, 1006–1011.
- Halperin, J. M., Sharma, V., Greenblatt, E., & Schwartz, S. T. (1991). Assessment of the Continuous Performance Test: Reliability and validity in a nonreferred sample. *Psychological Assessment*, *3*, 603–608.
- Himle, M. B., Chang, S., Woods, D. W., Pearlman, A., Buzzella, B., Bunaci, L., et al. (2006a). Direct observation of tics in children with chronic tic disorders: Reliability, validity, and feasibility. *Journal of Applied Behavior Analysis*, *39*, 429–440.
- Himle, M. B., & Woods, D. W. (2005). An experimental evaluation of tic suppression and the tic rebound effect. *Behaviour Research and Therapy*, *43*, 1443–1451.
- Himle, M. B., Woods, D. W., Piacentini, J. C., & Walkup, J. (2006b). Brief review of habit reversal training for Tourette's syndrome. *Journal of Child Neurology*, *21*, 719–725.
- Khalifa, N., & Von Knorring, A. L. (2006). Psychopathology in a Swedish population of school children with tic disorders. *Journal of the American Academy of Child and Adolescent Psychiatry*, *45*, 1346–1353.
- Leckman, J. F., Riddle, M. A., Hardin, M., Ort, S. I., Swartz, K. L., Stevenson, J., et al. (1989). The Yale Global Tic Severity Scale: Initial testing of a clinician-rated scale of tic severity. *American Academy of Child and Adolescent Psychiatry*, *28*, 566–573.
- Leckman, J. F., Walker, D. E., & Cohen, D. J. (1993). Premonitory urges in Tourette's syndrome. *American Journal of Psychiatry*, *150*, 98–102.
- Malatesta, V. J. (1990). Behavioral case formulation: An experimental assessment study of transient tic disorder. *Journal of Psychopathology and Behavioral Assessment*, *12*, 219–232.
- Marcks, B. A., Woods, D. W., Teng, E. J., & Twohig, M. P. (2004). What do those who know, know? Investigating providers' knowledge about Tourette syndrome and its treatment. *Cognitive and Behavioral Practice*, *11*, 298–305.
- Meidinger, A. L., Miltenberger, R. G., Himle, M. B., Omvig, M., Trainor, C., & Crosby, R. (2005). An investigation of tic suppression and the rebound effect in Tourette's disorder. *Behavior Modification*, *29*, 716–745.
- O'Connor, K., Brisebois, H., Brault, M., Robillard, S., & Loiselle, J. (2003). Behavioral activity associated with onset in chronic tic and habit disorder. *Behaviour Research and Therapy*, *41*, 241–249.
- Peterson, B. S., Skudlarski, P., Anderson, A. W., Zhang, H., Gatnby, J. C., Lacadie, C. M., et al. (1998). A functional magnetic resonance imaging study of tic suppression in Tourette syndrome. *Archives of General Psychiatry*, *55*, 326–333.
- Scahill, L., Riddle, M. A., McSwiggin-Hardin, M., Ort, S. I., King, R. A., Goodman, W. K., et al. (1997). Children's Yale–Brown obsessive–compulsive scale: Reliability and validity. *Journal of the American Academy of Child and Adolescent Psychiatry*, *36*, 844–852.
- Silva, R. R., Munoz, D. M., Barickman, J., & Friedhoff, A. J. (1995). Environmental factors and related fluctuation of symptoms in children and adolescents with Tourette's disorder. *Journal of Child Psychology and Psychiatry*, *36*, 305–312.
- Silverman, W., & Albano, A. M. (1996). *Anxiety disorders interview schedule for DSM-IV—child and parent versions*. San Antonio, TX: Psychological Corporation.
- Silverman, W. K., & Eisen, A. R. (1992). Age differences in the reliability of parent and child reports of child anxious symptomatology using a structured interview. *Journal of the American Academy of Child and Adolescent Psychiatry*, *31*, 117–124.
- Silverman, W. K., & Nelles, W. B. (1988). The Anxiety Disorders Inventory Schedule for Children. *Journal of the American Academy of Child and Adolescent Psychiatry*, *27*, 779–784.
- The Psychological Corporation (1999). *Wechsler abbreviated scale of intelligence*. San Antonio, TX: The Psychological Corporation.
- Wilhelm, S., Deckersbach, T., Coffey, B. J., Bohne, A., Peterson, A. L., & Baer, L. (2003). Habit reversal versus supportive psychotherapy for Tourette's disorder: A randomized controlled trial. *American Journal of Psychiatry*, *160*, 1175–1177.
- Woods, D. W., & Himle, M. B. (2004). Creating tic suppression: Comparing the effects of verbal instruction to differential reinforcement. *Journal of Applied Behavior Analysis*, *37*, 417–420.
- Woods, D. W., Twohig, M. P., Flessner, C. A., & Roloff, T. J. (2003). Treatment of vocal tics in children with Tourette syndrome: Investigating the efficacy of habit reversal. *Journal of Applied Behavior Analysis*, *36*, 109–112.