The influence of contextual factors on tic expression in Tourette's syndrome: A review

Christine A. Conelea, Douglas W. Woods*

University of Wisconsin-Milwaukee, Milwaukee, WI, USA

Abstract

Symptoms of Tourette's syndrome vary in frequency and intensity. Although such variability may be the result of deficits in the underlying neurological system, tic expression can also be systematically impacted by contextual factors. This article reviews research on the impact of several contextual factors on tic expression and discusses implications for future research and treatment development.

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Introduction

Chronic motor or vocal tic disorders (CTDs) and Tourette's syndrome (TS) occur in 0.04% to 3.8% of the population [1–5] and are more common in males than females. The presence of single or multiple motor or vocal tics, but not both, for at least 1 year characterizes CTDs, and TS is characterized by multiple motor tics and at least one vocal tic that persist for at least 1 year. Tic frequency and severity often fluctuate in a waxing and waning pattern, and a person's repertoire of tics commonly changes over time. Premonitory urges, which are uncomfortable or aversive bodily sensations (such an itch, tingle, or tension), often occur prior to tics and are temporarily relieved or reduced following tics. Some have suggested that the urge is important for tic control and that its reduction can reinforce tic expression [6–8].

Much TS research has focused on isolating genetic determinants [9–12], neuroanatomic underpinnings [13,14], and associated neurotransmission [15]. Thus, it is not surprising that variation in tic expression has been attributed to these factors [16,17]. However, there is increasing evidence that tic expression may not be the sole product of internal processes but is also influenced by contextual factors [18,19] that impact these biological processes. Understanding how contextual factors themselves impact tic expression is important because it may lead to (a) models of TS etiology and maintenance that better integrate both neurological and contextual factors and (b) the development and refinement of treatment strategies for TS.

In this article, we review what is currently known about the effects of contextual variables on tic expression. By "contextual factors," we refer to environmental events (e.g., presence or absence of specific stimuli, emotional reactions to life events, different settings or activities) that occur in a person's life that can have an immediate and direct impact on tic occurrence. This review does not include static historical variables more closely linked to biological etiologies, such as streptococcal infections [20], events occurring pre- or perinatally [21–24], or the impact of drugs upon tic expression.

We have organized our coverage into two general categories: antecedent and consequent factors. Antecedents are those events that occur prior to a tic, which, when present, alter the likelihood that the behavior will occur. In contrast, consequence factors are events that occur after a tic and serve to reinforce (increase) or punish (decrease) the

* Corresponding author. Department of Psychology, University of Wisconsin-Milwaukee, Box 413, Milwaukee, WI 53201, USA. Tel.: +1 414 229 5335; fax: +1 414 229 5219.

E-mail address: dwoods@uwm.edu (D.W. Woods).

behavior. Under each category, we will review evidence examining the impact of such factors on tic expression, noting particular limitations, implications, and potential future directions.

Articles were identified using the database PsycINFO. Several search terms were used, including the word *tic* plus the terms *environmental factor*, *context*, *functional analysis*, *reinforcement*, *suppression*, and *stress*. The reference lists of relevant articles identified through these searches were examined to identify additional articles of interest.

Antecedent factors

Descriptive studies examining the impact of antecedents

Most research examining the effects of antecedent contextual factors on tic expression has examined the impact of broad events (a.k.a., setting events) using self-report methodology. A summary of the studies reviewed in this section is provided in Table 1.

Combined, these various studies [25–30] offer preliminary evidence that contextual antecedent events impact tic frequency. Some events appear to be more commonly associated with tic exacerbations, such as fatigue [25–28]

and social activities [28,30], and others with tic attenuation, such as relaxation [26,27], concentration or study [26,27,30], and passive states [29–30]. Nevertheless, the studies are limited in several ways. First, the data are typically reported in aggregate form [25–28,30]. Although the reporting of data in this form is common and useful for making general conclusions, it may be of limited value in understanding how tics for any one individual are effected. Given that contextual factors may impact tics differently among individuals [28,30], the assessment of individual response patterns seems essential.

Second, all studies relied on self-reports or parental reports and were entirely descriptive in nature [25–30]. Participant bias and distortion can impact responding on self-report measures, as can the format, wording, and order of items [31]. The descriptive nature of the research did not allow for the examination of contextual variables upon tics in a controlled experimental setting, thereby precluding interpretations about causality. Third, only a few of these studies contained information regarding a formal description of how tics [26–30] or comorbid conditions [26,27,29,30] were diagnosed and how medication history was established. Such information is important, as the latter two may moderate the relationship between contextual factors and tic expression. Finally, none of the studies systematically investigated the

Table 1
Summary of descriptive studies examining the impact of antecedent variables on tics

Study	Participants	Design	Primary measure(s)	Factors associated with tic exacerbation	Factors associated with tic attenuation
Bornstein et al. [25]	763 children and adults	Descriptive	Survey	Stress or anxiety (98.2%) Fatigue (74.5%) Holidays, birthdays (67.9%) Return to school (68.4%)	Stress or anxiety (0.3%) Fatigue (5.9%) Holidays, birthdays (2.3%) Return to school (3.1%)
Eapen et al. [26]	91 adults	Descriptive	National Hospital Interview Schedule for Gilles de la Tourette Syndrome	Stress (83.5%) Tiredness (7.7%) Boredom (3.3%)	Relaxation (52.7%) Concentration (23.1%) Sport (8.8%)
O'Connor et al. [29]	13 adults	Descriptive	Daily self-report diary	Working under pressure Overstimulation Waiting Multitasking	Habitual, automatic actions (e.g., showering, typing) Passive states Interactions with familiar people
O'Connor et al. [30]	39 adults	Descriptive	Daily self-report diary	Socialization (50%) Transportation (43%) Waiting (43%) Passive attendance (38%) Intellectual work (28%)	Passive attendance (45%) Study activity (43%) Physical exercises (32%) Socialization (30%) Leisure activity (28%)
Robertson et al. [27]	57 children	Descriptive	National Hospital Interview Schedule for Gilles de la Tourette Syndrome	Stress (53%) Tiredness (5%) Boredom (4%)	Relaxation (26%) Concentration (14%) Sport (7%)
Silva et al. [28] [a]	14 children	Descriptive	Tourette Syndrome Questionnaire, clinical interview	Anxious/Upset (78%) Emotional trauma (50%) Fatigue (50%) Watching TV (46%) Being alone (44%) Social gatherings (42%)	Doctor visits (27%) Social gatherings (25%) Talking to friends (23%) Reading for pleasure (20%)

Values enclosed in parentheses (except those for the Silva et al. [28] study) represent the percentage of participants whose tics were exacerbated/attenuated by stated factor.

[a] For the Silva et al. [28] study, values in parentheses represent the number of participants that endorsed the exacerbating/attenuating factor over the number of participants that reported experiencing that factor.

impact of contextual variables on the urge to tic. Given the purported importance of the urge in tic expression, it seems important that any examination of broad contextual events and their impact on tics should consider the impact of such events on the urge.

Experimental studies examining the impact of antecedents

Studies utilizing experimental designs have also examined the impact of contextual antecedent stimuli on tic expression to address some of the issues raised by descriptive designs, such as self-reporting biases and an inability to experimentally examine causal relations [18,32–35]. Frequently, these studies utilize single-case experimental designs, which are often confused with a case study design. Whereas case study designs lack both internal and external validity, single-case experimental designs include various controls for threats to internal validity and are deemed internally valid designs [36]. A summary of the studies reviewed in this section is provided in Table 2.

Malatesta [32] observed that tic frequencies in a 9-year-old were highest in the presence of the child's father, who was critical and punitive regarding the tics. This clinical observation was followed by an experiment in which tics and frontalis electromyographic (EMG) responses were measured during an alone condition and compared to conditions in which certain observers were absent or present (i.e., father, mother, therapist, custodian, mother and father simultaneously). Results showed an increase in tics and EMG responses when in the presence of others, but especially in the presence of the father.

Watson et al. [34] conducted informal interviews with two male child participants and their teachers. For one child, a teacher noted that tics were likely to occur most often in science class. Direct observations in that class indicated that tics occurred most frequently during seatwork and least

frequently during lab work. Because seatwork usually consisted of reading and writing assignments, the type of assignment was compared, and more tics were observed during reading tasks. Finally, the authors compared difficult and easy reading material and found that tic frequencies were higher when the child was reading easy material. Using a similar procedure, the second child's tics were found to be most frequent when he was reading in a quiet classroom and between assignments.

In another study, Woods et al. [35] examined the impact of tic-related conversation on tics in two males with TS using a withdrawal design [36]. Tics were repeatedly compared across two different 5-min conditions. In the *control* condition, the participant and the clinician had a conversation that did not involve discussion of tic-related topics. During the *tic-talk* condition, the participant and the clinician discussed tic-related topics (e.g., described the topography of tics and the social consequences of the tics). Results showed that vocal (but not motor) tic frequencies increased considerably during tic-talk conditions and decreased during the control conditions. It is unclear why motor and vocal tics were impacted differently, but the findings are consistent with Silva et al. [28], who suggested that vocal tics may be more reactive to contextual variables, and O'Connor et al. [30], who suggested that the physical location of tics seemed, in some cases, to be functionally related to the activity in which the person was engaged.

Another antecedent condition that has been evaluated involves the giving of an explicit instruction to suppress tics. Some have suggested that exposure to this particular variable can lead to a rebound in frequency after it is removed [37]. As such, Meidinger et al. [18] examined the effects of suppression instructions and the tic rebound phenomenon by comparing three conditions, baseline, suppression (suppression when alone or suppression when in the presence of another person), and post-suppression, in seven adults and

Table 2
Summary of experimental studies examining the impact of antecedent variables on tics

Study	Participants	Design	Primary measure(s)	Factors associated with tic exacerbation	Factors associated with tic attenuation
Malatesta [32]	1 child	Experimental, single-subject (withdrawal design)	Direct observation Frontalis EMG responses	Presence of others Presence of father	
Meidinger et al. [18]	7 adults and children	Experimental, single-subject (withdrawal design)	Direct observation		Verbal instructions to suppress tics
Piacentini et al. [33]	43 children	Experimental, single-subject (alternating treatments)	Direct observation	Overt observation (80%) Overt observation with person in room (26%) Being in clinic (15%)	Overt observation with person in room (46%) Being in clinic (30%)
Watson et al. [34]	2 children	Experimental, single-subject (alternating treatments)	Direct observation	Reading tasks Easy reading tasks Reading in quiet classroom Between assignments	
Woods et al. [35]	2 children	Experimental, single-subject (withdrawal design)	Direct observation	Tic-related conversation (vocal tics only)	

Values enclosed in parentheses represent the percentage of participants whose tics were exacerbated/attenuated by stated factor.

children with TS. During the alone suppression condition, participants were instructed to suppress their tics while sitting alone in a room for 30 min. During the social suppression condition, the experimenter and the participant engaged in a non-tic-related conversation for 30 min and no instructions to suppress were given. The participant was instructed to stop suppression efforts during the post-suppression condition. Reductions in tic frequencies were observed in almost one half of all suppression sessions, indicating that verbal instructions to suppress can exert a moderate degree of control over tics. No consistent differences were observed between suppression when alone and suppression when with another person. Adults in the study demonstrated suppression more frequently than the children. Rebounds in tic frequencies were not found in the post-suppression condition. Meidinger et al. also asked participants to rate their urge to engage in tics on a Likert-type scale following each experimental session. A moderate relationship between tic frequency and urge ratings was found (r=.235, P=.052). Unfortunately, the authors did not report whether urge ratings changed during or after suppression, making the impact of suppression instructions on the urge unclear.

In a final study, Piacentini et al. [33] examined the reactivity of tics to observation and the stability and variability of tics across home and clinic observation settings in children and adolescents with TS or CTD. Participants' medication dosages had to be stable for study inclusion. Comorbid diagnoses were formally assessed using a structured clinical interview, and the most common comorbid conditions were ADHD (51%) and ODD/CD (23%).

Participants were exposed to three experimentally manipulated situations to investigate whether tic expression was reactive to observation and/or the presence of others: alone-overt, in which the child was taped with the video camera visible; present-overt, in which the child engaged in a non-tic-related conversation with the experimenter while a camera was visible; and alone-covert, in which the child sat alone while being taped by a hidden video camera. Each condition lasted for 15 min and was randomly ordered across three weekly laboratory visits. Results showed that 80% of participants experienced a ≥20% increase in tics during the overt observation conditions compared to the covert conditions, suggesting that being observed generally increases tics. However, the addition of a live person to an overt observation condition increased tics notably for 26% of participants but produced a notable decrease in tics for 46%.

The study also examined the stability of tics across home and clinic settings. Participants were videotaped three times at home and three times in the clinic. Each observation lasted for 30 min. For the majority (55%), tic frequencies did not differ between the two settings. However, 15% of participants had more tics (i.e., ≥20% difference) in the clinic and 30% had fewer (i.e., ≥20% difference), suggesting that environment-specific variables may play a role in explaining differences in tic frequencies.

Combined, the experimental studies demonstrate that specific antecedent factors can influence tics. These include the presence of others [32,33], academic tasks [34], tic-related conversation [35], verbal instructions to suppress tics [18], and overt observation [33]. In addition, the methodology of these studies improves upon the limitations inherent in the earlier self-report studies [25,28] by including experimentally manipulated variables and direct observation of tics. Individual patterns of responding were also reported, allowing for inspection of individual variability. Nevertheless, these studies also have a number of limitations. First, in all but the Piacentini et al. [33] study, the effects were examined in very few participants; thus, generalizability of results remains unclear. Second, the studies were conducted within relatively brief time periods, and given the fluctuating nature of tics, longer periods of observation may yield different results. Third, the number of variables explored in these studies remains small. Future research should consider expanding the methodology to test additional variables noted in the descriptive studies as possibly influencing tics (i.e., fatigue, excitement). Fourth, as mentioned previously, the premonitory urge may be an important antecedent event involved in the maintenance of tics, and only Meidinger et al. [18] assessed the phenomenon, albeit in a limited fashion.

Finally, both the experimental and descriptive studies examining the effects of antecedents on tics suffer from a similar problem. Both stop before a level of analysis that is satisfying because the question remains as to *why* the antecedent factors exert control on tics. A number of possibilities exist. For example, the presence of certain antecedents may create an emotional experience (i.e., anxiety) that either increases tics directly or decreases one's ability to inhibit tics when contextually appropriate. Likewise, antecedent events may signal the presence or absence of reinforcement or punishment for tic expression. In the next two sections, the role of emotional variables on tic expression and the impact of consequences on tic expression will be explored.

Emotions and tic expression

Preliminary evidence suggests that emotional variables impact tics [25,28–30,38]. For example, nearly all participants (98.2%) in the Bornstein et al. [25] study reported that stress or anxiety worsened their tic symptoms, and similar results were reported by Silva et al. [28] and by Eapen et al. [26]. A summary of the studies reviewed in this section is provided in Table 3.

Findley et al. [38] assessed the global impact of stressful life events on tic and OCD symptoms in 32 children and their parents. Results indicated that the children with TS and OCD experienced a greater number of stressful events than matched normal controls, and a significant correlation between tic severity and daily live stressors was also found. After controlling for the effect of age on tic severity improvement, Lin et al. [39] found that current levels of

Table 3
Summary of studies examining the impact of emotions on tics

Study	Participants	Design	Primary measure(s)	Factors associated with tic exacerbation	Factors associated with tic attenuation
Findley et al. [38]	32 children	Correlational	Daily Life Stressors Scale, Yale Global Tic Severity Scale	Daily life stressors	
Lees et al. [42]	53 adults and children	Descriptive	Direct observation	Timed mental arithmetic task (11%)	Timed mental arithmetic task (13%)
Lin et al. [39]	37 children	Multigroup, prospective, longitudinal	Parent Perceived Stress Scale, Daily Life Stressors Scale, Yale Children's Global Stress Index, Yale Global Tic Severity Scale	Psychosocial stress	
Hoekstra et al. [40]	32 adults and 25 children	Single-group, prospective, longitudinal	Inventory of Small Life Events (adults), Small Events Inventory-Child Reports (children), Yale Global Tic Severity Scale	Undesirable small life events (found for adult group only)	
O'Connor et al. [29]	13 adults	Descriptive	Daily self-report diary	Worry Frustration	
O'Connor et al. [30]	39 adults	Descriptive	Daily self-report diary	Active/tense appraisal	
Wood et al. [41]	4 children	Experimental, single-subject (alternating treatments)	Direct observation	Neutral movie scenes	Emotionally charged movie scenes (particularly anger and happy scenes)

Values enclosed in parentheses represent the percentage of participants that endorsed the exacerbating/attenuating factor.

psychosocial stress and depression were significant, although modest, predictors of future tic severity.

Hoekstra et al. [40] conducted a prospective longitudinal study of adults (*n*=32) and children (*n*=25) to examine possible associations between small life events and self-reported tic severity. A questionnaire, completed weekly by participants for 12 consecutive weeks, required them to rate the perceived tic severity and frequencies of small life events across several domains (e.g., family, leisure, education, and work). A significant correlation (*r*=.268, *P*<.001) between negative small life events and tic severity was found for the adult group but not for the pediatric group. The authors noted a number of explanations for the relatively weak relationship, including (a) that anxiety-provoking contextual events may have a more temporally proximal impact on tics than was measured in the study and (b) that the *perceived stress* associated with the small life events endorsed may be more important than the simple occurrence of the event.

The role of perceived emotion and tic exacerbation was examined in studies by O'Connor et al. [29,30]. O'Connor et al. [29] asked participants to identify the emotions associated with high-risk situations. Frustration was the emotion most commonly associated with high-risk situations. Using a similar procedure, O'Connor et al. [30] found that activities associated with a high risk for tic expression were most likely to be appraised as active or tense. Interestingly, the authors noted that high-risk activities were not associated with appraisals of anxiety or nervousness.

In another study, Wood et al. [41] experimentally examined the effects of emotional stimuli on tic severity. Four children watched eight movie scenes associated with different emotional states while being videotaped. Tapes were coded by blinded independent raters using the Goetz Video

Rating Scale. Measures of autonomic arousal (respiratory rate and heart beat) were taken while the children watched the scenes and were not found to be associated with changes in tic severity. Tic severity was found to generally be the highest during baseline periods than during the emotionally charged scenes. Among the emotionally charged scenes, tic severity was lowest during scenes of happiness and anger and most severe during scenes of anticipation and emotional resolution, suggesting that tic severity may be differentially impacted by certain emotional stimuli.

Finally, as part of a larger study, Lees et al. [42] observed 53 adults and children with TS during a typically stressful situation, completing mental arithmetic under time pressure, and found that tic severity increased in 6 participants and decreased in 7. The impact of the mental arithmetic task on the tics of the remaining participants was not reported.

Combined, these studies suggest that stress, anxiety, frustration, and tension are often associated with an increase in tics. Nevertheless, a number of methodological concerns limit our ability to make specific conclusions about the role of emotional variables on tics. First, these studies typically used nonexperimental designs and primarily relied on retrospective self-reports or parental reports of both the emotional experience and tic occurrence, thus subjecting the data to various biases [35]. The Wood et al. [41] study expanded upon this research by using an experimental manipulation and direct observation measure, but the generalizability of the results remains unclear due to the small sample size.

Second, the emotions examined are limited primarily to some derivative of anxiety, and even this has not been reliably defined, with some studies calling it stress, some calling it tension, and some calling it anxiety. Similarly, the

distinction between proximal and distal anxiety was not clearly examined. It may be the case that anxiety related to a current or soon-to-occur event impacts tics differently than anticipatory anxiety related to a more distal event. Third, it is unclear which aspect of the emotional experience is specifically related to tic expression. Is it sympathetic nervous system arousal, cognitive activity, the behavioral tendency to avoid or escape such situations, or a combination of all factors? Fourth, as with previous studies, the effect of emotional variables on the premonitory urge has not been examined. Does anxiety increase the salience of the urge, making it more noticeable or aversive? Finally, the studies do not tell us *why* tics are impacted by these emotional variables. Do particular emotions disrupt inhibitory processes, making tic exacerbation more likely? Do particular emotions function to exacerbate the underlying neurological deficits responsible for tics? Or do particular emotions, as discussed previously, increase the salience of, or aversion to, the premonitory urge, which is reduced with more frequent tics? Future research must consider these possibilities.

Consequence factors

In addition to the research on antecedent contextual factors associated with tic expression, numerous studies have focused on the effect of contextual consequences on tics. A summary of the studies reviewed in this section is provided in Table 4.

Packer [43] asked 69 parents of children with tic disorders to identify specific consequences that had been implemented to modify their children's tics at school. Negative consequences, such as sending the child from the room or publicly commenting on tics, and neutral consequences, such as signaling the child discreetly, were reported to have negative or neutral outcomes. Positive consequences, such as reward for modifying a tic or using a competing response, reportedly yielded positive outcomes. However, as the author noted, it was unclear if "outcomes" referred to tic frequencies or some other variable, such as emotional functioning. Therefore, although this study suggests that contextual consequences may have an impact on tics, the results should be considered tentative.

Table 4
Summary of studies examining the impact of consequence variables on tics

Study	Participants	Design	Primary measure(s)	Factors associated with tic exacerbation	Factors associated with tic attenuation
Carr et al. [45]	1 child	Experimental, single-subject (alternating treatments)	Direct observation	Contingent attention Contingent escape	
Himle & Woods [50]	7 children	Experimental, single-subject (reversal design)	Direct observation		Reinforcement for tic-free periods (70% tic reduction from baseline)
Himle et al. [49]	4 children	Experimental, single-subject (alternating treatments)	Direct observation		Reinforcement for tic-free periods
Himle et al. [53]	5 children	Experimental, single-subject (reversal design)	Direct observation Premonitory Urge Severity Ratings	Tic suppression [a]	Reinforcement for tic-free periods
Packer [43]	69 parents of 71 children	Descriptive	Survey	Sending child from room Publicly commenting on tics Signaling child discreetly	Reward for modifying a tic or using a competing response
Roane et al. [7]	1 adult	Experimental, single-subject (alternating treatments)	Direct observation	Being alone Sitting upright	Contingent attention Contingent escape from demand Contingent access to tangibles or toys Lying down
Rosen & Wesner [47]	1 child	Experimental, single-subject (withdrawal design)	Direct observation		Social and tangible reinforcement for tic-free periods
Scotti et al. [44]	1 adult	Experimental, single-subject (alternating treatments)	Direct observation	Contingent escape/ avoidance from demand	
Watson et al. [46]	1 child	Experimental, single-subject (alternating treatments)	Parental direct observation	Contingent parental attention	Parental attention contingent upon tic-free periods
Woods & Himle [19]	4 children	Experimental, single-subject (withdrawal design)	Direct observation		Verbal instructions to suppress tics (10.3% tic reduction from baseline) Reinforcement for tic-free periods (76.3% tic reduction from baseline)
Woods et al. [51]	13 children	Experimental, single-subject (withdrawal design)	Direct observation		Reinforcement for tic-free periods

[a] Factor associated with premonitory urge exacerbation.

Several studies have examined the impact of specific consequences on tic frequencies using functional behavior analysis (FBA) methodology, also known as alternating treatments, single-case experimental designs [7,44–46]; other experimental single-case designs [47–50,52]; or group designs [19].

Scotti et al. [44] conducted a functional analysis on the tics of a 32-year-old male with TS and profound mental retardation. Tics were assessed during five situations to determine possible behavioral functions. In the escape/avoidance situation, the participant was asked to do various tasks. Contingent upon the first tic occurring after a request, the task was withdrawn for a brief period of time. In the positive social interaction and tangible items condition, the researcher gave the participant food and made an approving statement contingent upon a tic. In the disapproving social attention condition, socially disapproving statements were delivered contingent upon tics. In the "alone" condition, the participant sat alone in the assessment room to assess whether the tic served a sensory stimulation function. The final situation consisted of the participant sitting alone in a rocking chair and served as the control condition, due to reports that tics were lowest in that situation. Each condition lasted for 15 min, followed by a 5-min break, and each condition was presented 10 times in random order. Tic frequencies were highest in the escape/avoidance situation and lowest during the social disapproval situation. Results indicated that tics for the subject predominately functioned to produce escape from demand.

Carr et al. [45] used a similar procedure as a diagnostic tool to assess an 11-year-old male with multiple vocal tics. Tic frequency was observed in several analogue classroom conditions: attention, escape, alone, free play, and high sensory stimulation. Although tics occurred frequently across all conditions, they were particularly elevated in the attention and escape conditions, suggesting that the child's tics were not solely operant but may have been partially controlled by positive and negative reinforcement contingencies. Roane et al. [7] observed a similar data pattern in a 22-year-old man with autism and TS.

Another study clearly demonstrated the influence of social attention in the expression of a transient vocal tic in a 4-year-old girl [46]. An interview with the child's parents suggested that the coughing tic occurred solely during meal times. A functional analysis was conducted during meal times to assess the impact of social attention and tangible reinforcers. Results indicated that parental attention was maintaining the cough. The rate of coughing decreased to zero following an intervention in which parental attention was withheld following occurrences of the cough and was instead provided contingent upon no coughing.

Overall, studies using function-based methodologies have shown that tics can be impacted by contextual consequence factors. However, such studies may be limited in several ways. First, there are no standard procedures to guide researchers when they select the scenarios to include in analogue situations [44]; hence, it is possible that researchers overlooked consequence factors that impact tics. Second, the analogue settings used in the studies may be too contrived and thereby limit the applicability of findings to natural settings. Third, these studies failed to assess the impact of consequences on the premonitory urge. Function-based methodologies could be a useful means by which to assess premonitory urge changes in a systematic fashion and could provide additional information about the urge–tic relationship. Finally, the current studies did not consider the possibility that emotional variables may have mediated the relationship between consequences and tic expression.

The aforementioned studies identified maintaining contextual consequences by using FBA procedures to establish the impact of controlling variables. In the next group of studies, single-subject and group experimental designs were employed to study the unique impact of reinforcement on tic expression/suppression.

Rosen and Wesner [47] demonstrated the impact of reinforcement on tics by manipulating contingencies within therapeutic and classroom environments of a 12-year-old male with TS. Within the therapy setting, tics were reduced using a differential reinforcement of zero-behavior (DRO) procedure, wherein a light was illuminated for every 30-s interval in which no tics occurred. At the conclusion of the therapy sessions, the participant was given a piece of candy for each time the light had been illuminated and was simultaneously praised for engaging in a tic-incompatible behavior. Following reductions in tic frequencies using this particular DRO procedure, the authors began to implement the procedure in the child's classroom. The child's entire class was given candy for each time the child exhibited a 30-s tic-free interval in order to establish the peers as reinforcers for symptom control. The reinforcement schedule was thinned and eventually faded out, and treatment gains were maintained by training the child's teacher to praise symptom control and to avoid reinforcing tics. In both the therapy and classroom settings, tics were reduced to near-zero levels when the DRO contingency was in place. Tics increased during baseline conditions, in which the contingency was not in effect. The authors concluded that TS symptoms could be operantly controlled. Similar procedures yielded similar results in a study of a 9-year-old male with chronic vocal tics [48].

Woods and Himle [19] investigated the specific role of contingent reward in the suppression of tics by comparing the effects of verbal instruction to suppress to verbal instruction plus DRO. During all conditions, a token dispenser was placed in front of the participants, and they were told that the machine was a "tic detector" capable of detecting tics via motion sensors. Participants were told to tic freely during the baseline condition. During the verbal instruction condition (VI), participants were told to suppress their tics in any way possible. During the DRO-enhanced instruction condition (DRO), participants were told to suppress their tics and were also told that the

machine would dispense a token for every 10-s tic-free period, which could later be exchanged for money. Each condition lasted for 5 min, and the experimental conditions were each repeated twice. Averaged across the four participants, the VI condition produced a 10.3% reduction in tic rates from baseline levels, whereas the contingency-enhanced condition (DRO) produced a 76.3% reduction from baseline, thereby supporting the belief that tics can be influenced by operant contingencies.

Himle et al. [49] further examined whether tic reductions observed from a DRO condition were attributable specifically to the reinforcement contingency. Tic frequencies were measured during conditions of baseline, DRO, and non-contingent reinforcement (NCR), in which tokens were delivered on a fixed time schedule regardless of successful suppression. Overall, tic frequencies in the DRO condition were lower than in the NCR condition, leading the authors to conclude that the contingent delivery of reinforcers for successful suppression was necessary for producing maximal tic reduction.

Himle and Woods [50] examined the potential relationship between tic suppression and a rebound effect using a similar manipulation as the Woods and Himle [19] study. Each of the seven participants was exposed to conditions of baseline, DRO-enhanced suppression, and post-suppression. Tic rates in the DRO-enhanced suppression condition, but not the post-suppression condition, were significantly lower than in baseline. Across participants, there was no evidence of a rebound effect. These results have been replicated using larger sample sizes and longer durations of suppression [51]. Combined, these findings demonstrate that reinforcing tic suppression for up to 40 consecutive minutes reduces tics and does not lead to later exacerbation. Other research has shown that rebounds in tic frequency do not occur following tic suppression for 2-h-long sessions of exposure and response prevention therapy, further contradicting the purported rebound effect [52].

In a study designed to examine empirically the relationship between tic suppression and the premonitory urge, Himle et al. [53] used a reversal design to obtain subjective ratings of urge severity during conditions of baseline and DRO. Three of the four children who demonstrated a suppression effect reported higher urge ratings during suppression compared to baseline. These findings demonstrated some support for the negative automatic reinforcement hypothesis, which contends that urge removal or reduction maintains or strengthens some tics [8]. Although this study provides preliminary insight into the relationship between contextual factors, tics, and the premonitory urge, further research should be conducted to better understand how age, tic location, and particular urge experience impact this relationship. Combined, these studies [19,47–51] provide experimental evidence that tic frequencies can be impacted by consequent stimuli. However, it is unclear if the tics themselves were modified. It may be the case that the consequences only influenced inhibitory behavior. It is possible that situations thought to be associated with tic exacerbations may be more accurately described as situations in which tic frequencies are at a baseline level or where reinforcement contingencies for tic suppression are absent. Additional research is needed to test this hypothesis. The generalizability of these results is also unclear. Although experimental, single-subject withdrawal designs are internally valid, the extent to which results are externally valid is uncertain. Future research should address whether these findings generalize to non-laboratory settings and to other children with TS, including those with comorbid psychiatric conditions.

Summary, limitations, and future directions

Research examining the impact of contextual factors on tic expression has increasingly generated support for the hypothesis that variability in tic expression may, in part, be explained by contextual factors.

Overall, findings in this area indicate that several contextual factors are more commonly associated with tic exacerbations. Stressful, frustrating, or anxiety-provoking events were reported to exacerbate tics in several studies [25,26,28,29,38–40,42], but this finding has not been universal [30]. Although these types of events are frequently reported to exacerbate tics, it is important to interpret this finding with caution. Stress, frustration, and anxiety are emotional states that are evoked by a set of circumstances, and the circumstances themselves, rather than the emotional states, may ultimately be the factors of interest. To better translate these findings into treatment applications, it will be important to identify and examine the contextual factors evoking these emotional responses. For example, research in the animal literature has demonstrated that manipulation of an animal's environment (in this case, the introduction of new elephants into an existing elephant group) may be associated with increases in stereotypies as well as cortisol levels [54].

It will also be helpful for future research to identify the neurochemical pathways by which stress-inducing events impact tic expression. Some researchers have hypothesized that neurobiological mechanisms related to stress, such as increased levels of stress-related hormones in the hypothalamic–pituitary–adrenal axis and of corticotropin-releasing factor in CSF, may impact tic expression [55]. Stress-inducing events may also increase motor cortex excitability and thereby hinder motor control [56], which may make tic suppression more difficult.

Other contextual events frequently associated with tic exacerbation were fatigue [25–28], social events [25,28,30,33], and starting school in the fall [25]. Events frequently reported to coincide with tic reductions included social interactions with familiar people [28,29], situations in which the individual is a passive participant [29,30], and leisure activities [28,30]. Once again, it would be beneficial for

future research to examine each reported event more closely to identify the specific contingencies involved in tic exacerbation or reduction.

One of the limitations of self-report and survey studies is that individual data are obscured. Given that several of the events reported above have been associated with both tic exacerbations and reductions, it is important to examine how these factors impact tics at the level of the individual. Functional analysis and single-case experimental design studies have begun to address this limitation. Findings from these studies have demonstrated that tic frequencies can increase when tics are positively reinforced, as in the case of attention following a tic [32,45,46], or negatively reinforced, as in the case of escape from demand situations [44,45]. Furthermore, reinforcement for tic-free periods has consistently been shown to reduce the occurrence of tics [19,47–51,53].

Although the current literature offers evidence that contextual factors can be involved in tic expression, there is still much work to be done. Few studies have examined the relationship between tics and contextual factors within a controlled experimental setting [18,19,35,49–51,53]. Experimentally isolating variables associated with increases in tic expression would greatly enhance treatment development and application, as such variables could be targeted specifically.

Likewise, there is limited understanding of the role of the premonitory urge in the development and maintenance of tic disorders. Preliminary evidence supports the notion of an urge–tic relationship [18,53]. There are data to suggest that urge severity increases during periods of reinforced suppression, supporting the notion that urge reduction may reinforce tics [53]. However, various aspects of the urge–tic relationship, as well as the impact that contextual factors have upon the urge, remain unclear. A comprehensive behavioral account of tic disorders should ultimately account for how the urge emerges and how it is impacted by contextual factors.

Finally, an interesting question for future research to address is whether it is tics themselves that are impacted by contextual factors or if it is suppression or inhibitory behaviors that are modifiable. What past researchers have labeled "tic exacerbations" may instead be situations in which there is little or no reinforcement for tic inhibition. Thus, "tic exacerbations" may represent naturally occurring frequencies of tics. If this is the case, future research may need to focus on isolating the variables associated with situations in which tic suppression is evident, rather than focus on isolating the variables associated with "increased" tic frequencies. It may also be the case that what is currently thought of as "voluntary" tic suppression is instead suppression under the control of certain contextual variables that have yet to be precisely identified. It is possible that awareness of contingencies, such as knowledge of a reinforcement contingency [19,50] or of being observed [33], improves suppression. However, it is likely that suppression behaviors are controlled by contextual factors whether or not an individual is aware of those factors. Therefore, research focusing on identifying the contingencies controlling suppression would likely enhance our understanding of how suppression occurs and how it may be enhanced.

In summary, research has demonstrated that tic expression is, at least in part, impacted by contextual factors. However, more research must be done to understand the role of these factors. Such work will allow for a better understanding of the disorder's etiology, maintenance, and treatment.

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