



## Reward enhances tic suppression in children within months of tic disorder onset



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### ABSTRACT

Tic disorders are childhood onset neuropsychiatric disorders characterized by motor and/or vocal tics. Research has demonstrated that children with chronic tics (including Tourette syndrome and Chronic Tic Disorder: TS/CTD) can suppress tics, particularly when an immediate, contingent reward is given for successful tic suppression. As a diagnosis of TS/CTD requires tics to be present for at least one year, children in these tic suppression studies had been living with tics for quite some time. Thus, it is unclear whether the ability to inhibit tics is learned over time or present at tic onset. Resolving that issue would inform theories of how tics develop and how behavior therapy for tics works. We investigated tic suppression in school-age children as close to the time of tic onset as possible, and no later than six months after onset. Children were asked to suppress their tics both in the presence and absence of a contingent reward. Results demonstrated that these children, like children with TS/CTD, have some capacity to suppress tics, and that immediate reward enhances that capacity. These findings demonstrate that the modulating effect of reward on inhibitory control of tics is present within months of tic onset, before tics have become chronic.

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### 1. Introduction

Tic disorders (including Tourette syndrome and Chronic Tic Disorder: TS/CTD) are complex childhood-onset neuropsychiatric disorders of the central nervous system characterized by the presence of motor and/or vocal tics.

Tics are movements or noises, often brief and repeated many times a day in a stereotyped fashion, that may look intentional but serve no useful purpose (Black, 2010). Common tics include forceful eye blinking, nose twitching, head jerking, sniffing, and throat clearing. The average age of tic onset in TS/CTD is ~6 years old (Leckman et al., 1998, 2006), and tics must be present for at least a year to diagnose TS/CTD. Though historically thought to be rare disorders, careful epidemiologic studies show that TS and CTD affect at least 2–6% of all children (Hornsey et al., 2001; Robertson, 2008; Cubo et al., 2011). Many more children have tics for less than a year, with point prevalence estimates of about 20% of school-aged children (Kurlan et al.,

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2001; Snider et al., 2002; Khalifa and von Knorring, 2003; Cubo et al., 2011). Therefore, within the first year after tic onset, some children experience marked improvement or become asymptomatic, while others develop a chronic disorder that can substantially impair their quality of life (Cavanna et al., 2008; Eddy et al., 2011a, 2011b).

Tics are distinguished from other abnormal movements in several ways, one of which is that there is usually some degree of voluntary control (Robertson et al., 1999; Black, 2010; Leckman et al., *in press*). Specifically, many adults and children with TS/CTD can suppress tics, at least briefly, and often attempt to suppress tics, especially in certain environmental contexts such as social gatherings or school. Because of this partial voluntary control, TS/CTD has been thought to involve faulty inhibitory control processes (Mink, 2001). In addition, tics are often described as being preceded by a “premonitory urge,” defined as a feeling of discomfort (e.g., a sensation like itch or pressure, or a sense that one must tic). This premonitory urge is temporarily relieved by the performance of a tic (Leckman et al., 2006). Thus, tics may reflect deficient inhibitory control over the motor response to this urge.

The issue of inhibitory control in TS/CTD is actually quite complex, as there is debate over whether certain domains of inhibitory control are in fact affected. For example, some electrophysiological studies (using transcranial magnetic stimulation) have found reduced cortical inhibition in the primary motor cortex in TS/CTD (Ziemann et al., 1997; Gilbert et al., 2004), suggesting altered motor inhibition. However, other studies have shown that differences in cortical excitability may be more related to comorbid attention deficit hyperactivity disorder (ADHD) symptoms than to tics (Gilbert et al., 2005; Orth and Rothwell, 2009). Similarly, although behavioral studies in children and adults with TS/CTD have demonstrated impairments in response inhibition, selective attention, and cognitive flexibility (Bornstein et al., 1991; Channon et al., 2003, 2009; Watkins et al., 2005), some have argued that these impairments are driven by comorbid conditions, including ADHD and obsessive-compulsive disorder (OCD) (Ozonoff et al., 1998; Denckla, 2006). Further, some studies have even shown evidence for enhanced executive function in TS/CTD (Mueller et al., 2006; Jackson et al., 2007). Neuroimaging data are also somewhat inconsistent (for a review, see Greene et al., 2013). There is some evidence for atypical and immature task control systems in the brain in TS/CTD (Church et al., 2009a, 2009b; Wang et al., 2011) as well as atypical activation of frontostriatal regions posited to support inhibitory control (Aron et al., 2014) in TS/CTD (Hershey et al., 2004a; Marsh et al., 2007; Baym et al., 2008; Raz et al., 2009). However, the directions of specific effects were inconsistent among the fMRI studies, and others have not been able to replicate differences between TS/CTD and controls with similar study designs (Hershey et al., 2004b; Debes et al., 2011). EEG and fMRI studies specifically investigating tic suppression have shown increased activation in frontostriatal regions that support inhibitory control during active tic suppression in children and adults with TS/CTD (Peterson et al., 1998; Hong et al., 2013). Thus, while studies of inhibitory control per se in TS/CTD may

be inconsistent, a relationship between tic suppression and inhibitory control mechanisms likely exists.

The ability to suppress tics has been measured using a standardized tic suppression paradigm (Woods and Himle, 2004). In this task, children are seated in front of a “tic detector” (described below) and asked to suppress or not to suppress their tics under varying conditions. Studies using this task have demonstrated that children with TS/CTD can suppress tics in response to a simple verbal request, though suppression is inconsistent and varies among individuals (Meidinger et al., 2005; Conelea and Woods, 2008). By contrast, when immediate rewards are given for brief periods of successful tic suppression, children with TS/CTD can robustly and reliably reduce tic rate (Woods and Himle, 2004; Himle and Woods, 2005; Himle et al., 2007, 2008; Woods et al., 2008; Specht et al., 2013). Further, rewards delivered specifically when tics were suppressed led to better tic suppression than rewards given without a temporal link to tic behavior (Himle et al., 2008). In other words, contextual variables that were not immediately linked to tic behavior (i.e., a verbal request to suppress tics, or non-contingent reward) had less impact on tics than contingent rewards. Thus, the presence of a reward that is specifically contingent upon tic behavior may create an environmental context for more consistent tic suppression in TS/CTD.

Since a diagnosis of TS/CTD requires tics to be present for a minimum of one year, the children in the cited tic suppression studies had been living with tics for at least a year and usually much longer. Years of experience with tics usually brings years of experience attempting to suppress tics, and some argue that this experience enhances inhibitory control in TS/CTD (Jackson et al., 2011). Thus, it is unclear whether inhibitory control over tics is present at the onset of the tic disorder or develops with experience. In the present study, we investigated children whose tics began within the previous six months. Surprisingly little is known from controlled studies about children with recent-onset tics. Anecdotally, many of these children display little awareness of their tics, and few of them have experienced the social pressure to inhibit their tics that children with TS/CTD have experienced. Children with recent-onset tics have, at most, a few months' experience suppressing tics, and the extent to which they can suppress tics has not been reported.

The present study tested whether children with recent-onset tics are capable of suppressing their tics and if so, whether an environmental contingency (namely, reward) modifies this ability. Given evidence that reward can enhance inhibitory control in children without tics (Padmanabhan et al., 2011; Geier and Luna, 2012), we hypothesized that children with recent-onset tics would be able to suppress tics successfully when rewarded for doing so, even though they have little to no experience suppressing tics.

## 2. Methods

### 2.1. Participants

Children with recent-onset tics (DSM-IV-TR Transient Tic Disorder) were recruited for this study via clinicians in

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