



Subjective versus objective measures of tic severity in Tourette syndrome – The influence of environment



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ABSTRACT

The objective of this study was to examine the influence of environmental challenges on tic expression by subjective and objective measures. The study group consisted of 41 children aged 6–18 years ($M=10.15$, $SD=2.73$) with a primary diagnosis of Tourette syndrome. Subjective measures included the Functional Assessment Interview developed for this study and three standard validated instruments. The objective measure was a video-recording of the patients in five daily-life situations: watching television, doing homework, being alone, receiving attention when ticcing, and talking to a stranger. In addition, the effect of premonitory urges on assessment of tic expression was evaluated. The associations between the subjective and objective measures of tic expression were moderate to low. A significantly higher number of tics were observed in the television situation, and a significantly lower number in the alone situation, compared to the other situations. Higher levels of premonitory urge were associated with greater awareness of objectively measured tic expression. In conclusion, tic expression is significantly influenced by the environment. Subjective measures of tic expression may be misleading. These results have implications for refining the clinical assessment of tics, improving research methodology, and developing new therapeutic strategies.

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1. Introduction

Tourette syndrome, a relatively common neuropsychiatric tic disorder, serves as a model for understanding brain-environment interactions (Murphy and Eddy, 2013). Although tics are an observable motor phenomenon driven by disturbances in underlying brain circuitry, they are accompanied by subjective sensory and psychological manifestations and are highly sensitive to environmental influences (Conelea and Woods, 2008). Apart from their theoretical implications, these factors are important considerations in the clinical assessment. In addition to history, comorbidities, and observation, tic intensity, frequency, and impact must be precisely calibrated for clinicians to fully understand and appropriately treat the disorder (Woods et al., 2007). However, most of the currently available instruments for quantifying tic

characteristics consist of subjective self-report questionnaires, parent- and teacher-rated scales, and semi-structured clinician-administered interviews (Cavanna and Piedad, 2013).

Several groups have attempted to develop more objective tools, usually based on video-recordings (Himle et al., 2006; Pappert et al., 2003), but these are limited to measurements of tic expression, without accounting for sensory or psychological phenomena. In addition, technical and logistic issues prevent their widespread use, and video-recording itself have been found to influence tic expression (Piacentini et al., 2006). It is also noteworthy that the role of premonitory urges in tic expression has hardly been addressed. Theoretically, because premonitory urges predict tic occurrence, patients with a greater awareness of urges would be expected to display greater awareness of tics, making their subjective reporting more accurate.

The sensitivity of tic expression to environmental influences may be related to both internal and external factors and to various types and levels of activity. To identify and analyze the conditions under which tic expression increases, and thereby achieve better

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behavioral management of the disorder, clinicians use functional assessment procedures (Gresham et al., 2001; Piacentini et al., 2010). However, function-based assessments harbor methodological problems. Most of the research so far has been done on small samples or in artificial environments unrelated to the patient's daily life. In addition, few efforts have been made to understand the mechanisms underlying the effect of the environment on tic expression, apart from those related to learning theory and broad emotional effects (Conelea and Woods, 2008). One plausible theory is that an environment with highly stimulating properties can interfere with motor inhibition and thereby exacerbate tic expression (Belluscio et al., 2011).

The aim of the present study was fourfold: 1. To test the ability of common subjective measures of tics to predict objective tic expression; 2a. To test the ability of the Functional Assessment Interview (FAI), a semi-structured instrument designed by our group, to systematically assess the influence of environmental situations on tic levels 2.b To test the FAI validity, by examining its ability to predict tic expression across research situations; 3. To examine the effect of premonitory urges on the associations between subjective and objective measures of tic expression; and 4. To compare the effect of different daily-life situations with different levels of stimulation on tic expression.

The following hypotheses were tested: 1. Subjective measures of tic expression accurately predict objective recordings of tic activity; 2a. Children and parents report different levels of tic expression in different environmental situations; 2b. The FAI is an accurate measure of expected tic expression in different environmental situations; 4. The intensity of premonitory urges affects the strength of the association between patient self-reports and objective assessments of tics; 5. Tic expression differs by type and degree of environmental stimulation.

2. Methods

2.1. Sample

Forty-one children and adolescents with a primary diagnosis of Tourette syndrome participated in the study. All participants were patients in a child and adolescent Tourette syndrome clinic at a university-affiliated children's hospital. Consecutive admissions to the clinic were recruited to the study, and when both parent and child gave permission, they were admitted to the study. Children with IQ less than 80 were excluded ($M=102.48$, $SD=18.87$).

2.2. Instruments

2.2.1. Effect of environment on tic expression

2.2.1.1. Functional Assessment Interview (FAI). The FAI is a semi-structured clinical interview constructed specifically for the present study. It was designed to identify the effect of five common, daily-life environmental challenges on tic expression: being alone, watching television, doing homework, talking to a stranger, receiving attention when ticcing. All these situations have a precedent in the literature (Conelea and Woods, 2008) and are relatively easy to simulate in the laboratory. Children and parents (separately) rate the frequency of ticcing experienced when each situation is encountered in everyday life on a scale of 1 (low frequency) to 4 (high frequency).

2.2.2. Presence and characteristics of tics

2.2.2.1. Yale Global Tic Severity Scale (YGTSS) (Leckman et al., 1989). The YGTSS is a clinician-rated instrument designed to assess various tic characteristics (severity, number, frequency, intensity, interference, and complexity), each scored on a 5- point

scale. Separate scores are obtained for vocal tics and motor tics, and these are summed to yield a total severity score (range 0–50). The scale also includes an impairment rating, scored on a 5-point scale (range 0–50). The YGTSS has demonstrated excellent psychometric properties with solid internal consistency, inter-rater reliability, and convergent and divergent validity (Leckman et al., 1989; Storch et al., 2005). The internal reliability in this study (Cronbach's α) was 0.86 for the motor scale and 0.90 for the vocal scale.

2.2.2.2. Parent Tic Questionnaire (PTQ) (Chang et al., 2009; Woods et al., 2007). The PTQ is a parent-report questionnaire designed to assess the presence of 14 common motor and vocal tics, in addition to their frequency (1=weekly, 2=daily, 3=hourly, 4=constantly) and intensity (1=lowest intensity, 4=highest intensity). The intensity and frequency scores for each item are summed, yielding a total score of 0 (no tics) to 8 (constant and intense tics). Motor and vocal tics may be evaluated separately, and the scores summed for a total score. The PTQ was found to have excellent validity against other tic-severity measures (Cavanna and Piedad, 2013).

2.2.2.3. Tourette Syndrome Clinical Global Impression (TS-CGI) (Leckman et al., 1988). The TS-CGI is a clinician-rated 5- item scale designed to assess the severity of Tourette syndrome, obsessive-compulsive disorder (OCD), anxiety disorder, depression, and attention deficit-hyperactivity disorder (ADHD). Each item is scored on an ordinal scale from 1 (normal) to 7 (very severe); a score of 4 or more indicates a significant clinical disorder. The TS-CGI shown excellent reliability and validity (Walkup et al., 1992) and has been widely used (Kwon et al., 2011; Pringsheim and Steeves, 2011).

2.2.2.4. Premonitory Urge for Tics Scale (PUTS) (Woods et al., 2005). The PUTS is a 9- item self-report questionnaire designed to assess the presence and frequency of premonitory sensory urges. Each item is rated on an ordinal scale from 1 (not at all true) to 4 (very much true); the total score ranges from 9 to 36. The PUTS has demonstrated high internal consistency and convergent validity (Woods et al., 2005). The Hebrew version was found to have good psychometric properties (Steinberg et al., 2010). The internal reliability in this study (Cronbach's α) was 0.83.

2.2.3. Co-morbidities

2.2.3.1. The Children's Yale Brown Obsessive Compulsive Scale (CYBOCS) (Scahill et al., 1997). The CYBOCS is a clinician-administered semi-structured interview for the assessment of the severity of obsessive-compulsive symptoms in children. The contents of the obsessions and compulsions are divided into 5 sub-domains, each rated on a scale from 0 (none) to 4 (severe): duration of the symptoms throughout the day, symptom-induced interference, level of stress, level of resistance, and control. Separate scores are obtained for obsessions and compulsions; their sum yields a total severity score (range 0–40); 16 is the cutoff total score for a diagnosis of OCD. The CYBOCS has demonstrated acceptable internal consistency and convergent validity (Gallant et al., 2008; Storch et al., 2006). The internal reliability in this study (Cronbach's α) was 0.89 for the obsession scale, 0.93 for the compulsion scale, and 0.93 for the total score.

2.2.3.2. Screen for Child Anxiety Related Emotional Disorders (SCARED) (Birmaher et al., 1997). The SCARED is a self-report scale for children with anxiety disorders. It contains 38 items describing different emotions and behaviors. Children rate the frequency of each on a 3-point scale from 0 (never) to 2 (often). The sum of all items yields a total anxiety score; 25 is the cutoff total score for a

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