

Clinical phenomenology of episodic rage in children with Tourette syndrome

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Abstract

Objective: Episodic rage of unknown etiology causes significant morbidity in children with Tourette's syndrome (TS). Using modified Diagnostic and Statistical Manual of Mental Disorders (DSM-IV) criteria for intermittent explosive disorder (IED), we developed a screen and symptom questionnaire to explore rage attack phenomenology and to preliminarily investigate whether symptom clusters can identify clinical subgroups of TS children with rage attacks. **Methods:** 48 children with TS between ages 7 and 17 years consecutively presenting with rage attacks completed the Rage Attacks Screen and Questionnaire.

Data was subjected to factor analysis. Cluster analytic procedures were used to identify clinical subgroups. **Results:** Final cluster solution revealed four homogeneous subgroups of TS children with rage who were differentiated by predominant clinical characteristics: specific urge resolution, environmentally secure reactivity, nonspecific urge resolution or labile nonresolving. **Conclusion:** Episodic rage in TS has stereotypic features, but diverse and complex etiologies. Identifying particular symptom clusters may facilitate improved treatment strategies. © 2003 Elsevier Inc. All rights reserved.

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Introduction

Tourette's syndrome (TS) is a neuropsychiatric disorder of childhood onset characterized by multiple motor and phonic tics, with a prevalence of 10 cases per 10,000 [1]. TS is increasingly identified among children in special educational settings [2]. In adults, TS prevalence appears lower, with a minority suffering debilitating tic symptoms [3].

The natural course of tic symptoms includes fluctuations in type, location and impairment. Tic severity appears to peak between ages 8 and 15 years, often followed by a steady decline in symptoms by late adolescence [4]. Factors influencing the natural history of TS are largely unknown but may involve psychiatric comorbidity, endocrine mech-

anisms or environmental exposures such as infections, stress and medications [5–7].

Clinical studies show TS associated with a variety of behavioral symptoms and psychiatric disorders [8–10]. Referral and selection bias in such populations tends to distort the relationship of psychopathology with tics, yielding relatively high rates of comorbidity compared with community samples [11]. Nonetheless, while the relationship of behavioral symptoms with the underlying tic diathesis has been difficult to untangle, it is clear that psychiatric comorbidity including attention-deficit hyperactivity disorder (ADHD), obsessive compulsive disorder (OCD), non-OCD anxiety disorders and mood disorders are commonly associated with higher levels of TS morbidity and impairment [5,12,13].

Among the most disruptive behaviors in some children with TS are recurrent episodes of explosive anger or aggression (commonly termed *rage attacks*). Approximately 25–70% of patients with TS in clinical settings report experiencing episodic behavioral outbursts and anger control problems [14–19]. When present, episodic

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rage is often identified by families as the most impairing symptom in their children with TS [20]. Similar symptom clusters have been described in other medical and psychiatric conditions and by the DSM-IV diagnosis of intermittent explosive disorder (IED) (when the episodes cannot be better accounted for by another mental disorder, substance or general medical condition) [21–25]. Recent studies demonstrate an association between comorbid psychiatric disorders and episodic rage in TS [14,27,28]. However, specific treatment interventions require a better understanding of rage phenomenology and potential etiologies. For this reason, we developed a screen and questionnaire to explore rage attack phenomenology and to preliminarily investigate whether symptoms clusters can identify clinical subgroups of TS children with episodic rage.

Method

Patients were voluntarily recruited from the Movement Disorders Center, Departments of Psychiatry and Neurology at North Shore University Hospital. Attempt was made to enroll 50 consecutive outpatients referred for assessment of TS and comorbid rage attacks; at the time of presentation, no subject was being medicated specifically for rage symptoms. Parental and child informed consents for participation were obtained by a clinician not involved in their care. Assessments of comorbidity and rage status were made by a research clinician blinded to the patient's previous diagnosis, comorbidity, rage status and concurrent medication usage. Ethics approval was obtained from the Office of the Internal Review Board, North Shore-Long Island Jewish Health System.

TS diagnoses were made by a TS specialist after comprehensive evaluation using the Diagnostic and Statistical Manual of Mental Disorders (DSM-IV) criteria [25]. An independent assessment battery including the Schedule for Affective Disorders and Schizophrenia for School-Age Children—Epidemiologic version (K-SADS-E) [26] was administered by a research clinician not involved in the subjects' care. Children with evidence from medical records or examination of mental retardation, head trauma, seizure disorder, autism or pervasive developmental disorder, psychosis, alcohol or substance abuse, sexual or physical abuse, or known organic causes of rage were excluded. Episodic rage attacks were characterized as unpredictable, uncontrollable and atypical of the child's baseline personality; cases of overtly goal-directed or predatory aggression were excluded. All participants were on stable doses of medication for a minimum of 2 months prior to entry into the study, and no medication changes were permitted until evaluation was completed.

Clinical diagnosis of episodic rage was made using modification of DSM-IV diagnostic criteria for IED with the omission of criterion C (i.e., that aggressive episodes are

not better accounted for by another mental disorder, medication or drug, or general medical condition) as follows:

Several discrete episodes of failure to resist aggressive impulses that result in serious assaultive acts or destruction of property.

Degree of aggressiveness expressed during the episodes is grossly out of proportion to any precipitating psychosocial stressors.

Parents provided examples of a typical rage attack to confirm that the above criteria were satisfied. In addition, the following frequency thresholds were selected to establish symptom severity, based on standard research criteria for IED [29,30], to insure a minimal frequency over time that is critical in order to make the diagnosis of episodic rage reliable across clinicians and to ensure that subjects with only occasional impulsive aggressive outbursts were not assigned with episodic rage:

At least three episodes per week \geq
four episodes during a 1-month period of time.

After rage attacks were documented by the rage screen, the subject and his/her parent or guardian were asked to complete the 22-item parental and self-report questionnaire designed to assess the presence or absence of specific characteristics believed to be associated with episodic rage.

The Rage Attacks Questionnaire was factor analyzed and a six-factor solution was utilized. Factor scores were normalized with *Z* transformations and the profiles were subjected to cluster analysis. The cluster analytically derived subgroups were checked with a K-Means iterative partitioning procedure in which the means for each variable were used to specify the basis for cluster membership. Univariate

Table 1
KSADS-E psychiatric comorbidities

Diagnoses	<i>n</i> (%)
ADHD	37 (77)
Inattentive subtype	11 (23)
Hyperactive subtype	2 (4)
Combined subtype	24 (50)
Bipolar disorder	12 (25)
I	7 (15)
II	1 (2)
Not otherwise specified	4 (8)
Unipolar depression	21 (48)
Major depressive disorder	8 (17)
Dysthymic disorder	7 (15)
Not otherwise specified	6 (13)
OCD	25 (52)
Non-OCD anxiety disorders	26 (54)
Generalized anxiety disorder	11 (23)
Separation anxiety disorder	5 (10)
Panic disorder	2 (4)
Simple phobia	2 (4)
Oppositional defiant disorder	20 (42)

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