Clinical Features of Young Children Referred for Impairing Temper Outbursts

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Abstract

Objective: In light of the current controversy about whether severe temper outbursts are diagnostic of mania in young children, we conducted a study to characterize such children, focusing on mania and other mood disorders, emotion regulation, and parental psychiatric history.

Methods: Study participants included 51 5–9-year-old children with frequent, impairing outbursts (probands) and 24 non-referred controls without outbursts. Parents completed a lifetime clinical interview about their child, and rated their child's current mood and behavior. Teachers completed a behavior rating scale. To assess emotion regulation, children were administered the Balloons Game, which assesses emotion expressivity in response to frustration, under demands of high and low regulation. Parental lifetime diagnoses were ascertained in blind clinical interviews.

Results: No child had bipolar disorder, bipolar disorder not otherwise specified (NOS), or major depression (MDD). The most prevalent disorder was oppositional defiant disorder (88.2%), followed by attention-deficit/hyperactivity disorder (74.5%), anxiety disorders (49.0%), and non-MDD depressive disorders (33.3%). Eleven probands (21.6%) met criteria for severe mood dysregulation. During the Balloons Game, when there were no demands for self-regulation, children with severe outbursts showed reduced positive expressivity, and also showed significant deficits in controlling negative facial expressions when asked to do so. Anxiety disorders were the only diagnoses significantly elevated in probands' mothers.

Conclusions: Overall, young children with severe temper outbursts do not present with bipolar disorder. Rather, disruptive behavior disorders with anxiety and depressive mood are common. In children with severe outbursts, deficits in regulating emotional facial expressions may reflect deficits controlling negative affect. This work represents a first step towards elucidating mechanisms underlying severe outbursts in young children.

Introduction

RELATIVELY LITTLE IS KNOWN about the clinical significance of severe temper outbursts in young children. Outbursts are common in preschoolers (Wakschlag et al. 2012), typically diminishing in severity and frequency by school age (Owens and Shaw 2003). However, when persistent and severe, outbursts may become clinically significant, as suggested by findings that >50% of children (ages 5–12) in an inpatient service had been admitted because of severe outbursts (Carlson et al. 2009). In addition, children whose temper outbursts continued through childhood were reported to have relatively poorer occupational and marital functioning in adulthood (Caspi et al. 1987).

Outbursts are not diagnostically specific. In clinical practice, children with outbursts typically receive disruptive behavior diagnoses such as oppositional defiant disorder (ODD), attention-deficit/hyperactivity disorder (ADHD) (Carlson and Dyson 2012; Margulies et al. 2012), and conduct disorder (Carlson et al. 2009). In recent years, greater focus has been placed on severe tantrums as symptoms of a mood disturbance. For example, studies of children with ODD demonstrate an association between irritability and negative affect, which encompasses temper outbursts, and risk for later depressive or anxiety disorders (Stringaris and Goodman 2009a,b; Burke et al. 2010; Rowe et al. 2010). Conversely, some investigators argue that severe outbursts are the primary manifestation of mania in children, thus justifying the diagnosis of bipolar

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disorder, even in very young children (Wozniak et al. 1995; Wilens et al. 2003). This diagnostic practice has likely contributed to the 40-fold increase in bipolar diagnoses in children since the late 1990s (Moreno et al. 2007; Carlson and Glover 2009). To address this diagnostic controversy, at least in adolescence, Leibenluft et al. compared adolescents with distinct manic episodes who fulfilled American Psychiatric Association, *Diagnostic and Statistical Manual of Mental Disorders*, 4th ed. (DSM-IV) (American Psychiatric Association 1994); criteria for bipolar disorder, to adolescents with severe temper outbursts, chronic irritability, and hyperarousal, a syndrome labeled severe mood dysregulation (SMD) (Leibenluft et al. 2003). Differences between adolescents with SMD and those with bipolar disorder were found in physiological responses to frustration (Rich et al. 2007), neural responses to social stimuli (Brotman et al. 2010), and longitudinal course (Stringaris et al. 2010). These findings have directly informed the development of a new disorder, disruptive mood dysregulation disorder (DMDD) in American Psychiatric Association, *Diagnostic and Statistical Manual of Mental Disorders*, 5th ed. (DSM-V) (American Psychiatric Association 2013). However, a recent study showed that only half of inpatient children (ages 5–12 years) with chronic irritability and explosiveness met criteria for DMDD (Margulies et al. 2012).

By definition, temper tantrums result from a loss of emotional control, but the relationship between childhood temper outbursts and emotional reactivity, and regulation, has not been examined directly. Emotional reactivity predicts internalizing and externalizing problems in disruptive preschoolers (Cole et al. 1996), and adolescents with SMD have been found to exhibit dysfunctional attention mechanisms in response to frustration (Rich et al. 2007). However, these studies did not assess directly the regulation of emotional responses. Data from Gross and Levenson indicates that adults can suppress emotions when instructed to do so (Gross and Levenson 1993; Gross 1998). The experimental paradigm used with adults has been extended to healthy children as young as 5 years of age, through a novel task that assesses changes in emotion expressivity in response to explicit instructions to inhibit emotional expressions (Bar-Haim et al. 2011). This task provided the opportunity to assess emotional responses to frustration (reactivity), as well as the capacity of young children with frequent, impairing outbursts to control their emotional displays.

Parental psychopathology can also contribute to our understanding of childhood psychopathology. For example, parents of adolescents with SMD and parents of adolescents with bipolar disorder were found to have comparable rates of depressive, anxiety, and substance use disorders, but not of bipolar disorder, which was more prevalent in family histories of adolescents with bipolar disorder, thus highlighting the distinction between the two clinical phenotypes (Brotman et al. 2007). Thus far, there has been no examination of parental psychopathology in young children for whom severe temper outbursts are a primary clinical concern.

We present a preliminary study of young children with frequent, impairing outbursts, with the aim of investigating their functioning across multiple domains, including current diagnosis, tantrum characteristics, behavioral and emotional symptoms at home and at school, their ability to inhibit emotional expressivity in response to frustration, and parental psychiatric history. We focused on 5–9-year-old children, as this young age group has not been studied systematically. Children with frequent, severe temper tantrums and a comparison group of children without outbursts, matched for age, sex, race, socioeconomic status, and intelligence quotient (IQ), were recruited to address the following

clinical questions: 1) Do children with severe outbursts present with DSM-IV bipolar disorder, mania, or other mood disorders? 2) Compared with children without outbursts, do children with frequent, impairing outbursts exhibit deficits in regulation of emotional expressivity in response to frustration? Also, we explored whether such deficits, if found, are specific to children with mood or anxiety disorders. 3) Do parents of young children with severe outbursts have greater lifetime prevalence of bipolar and mood disorders than parents of controls? 4) Finally, we assessed SMD symptoms, to examine whether they characterized young children with frequent, severe outbursts, consistent with reports in adolescents. We could not examine DMDD, as the disorder did not exist at the time of the study.

Methods

The study was approved by the New York University (NYU) School of Medicine Institutional Review Board. Parents provided signed informed consent and children provided oral or written (age 9 only) assent. A subset of parents signed an additional informed consent to participate in the parent diagnostic interview. Families received compensation for their time; families of children with outbursts were offered four sessions of behavior therapy, at no cost.

Participants

Children (probands and controls) were recruited over a period of 2 years. They had to be 5.0 through 9.9 years old, medically healthy, with an estimated IQ ≥ 80 , as assessed by the two subtest version of the Wechsler Abbreviated Scale of Intelligence (WASI; Wechsler 1999) (≥ 6 years of age) and the Kaufman Brief Intelligence test (K-BIT; Kaufman and Kaufman 2004) (5-year-olds). Across groups, neurological and psychiatric exclusions were: History of head injury or neurological disorder, pervasive developmental disorder, posttraumatic stress disorder, and use of current antipsychotic medication.

Children with frequent, impairing outbursts (probands). Children whose parents were seeking professional help for their child's frequent, impairing temper outbursts were recruited through the intake service at the NYU Child Study Center ($n = 8$), clinical referrals from NYU and community clinicians ($n = 20$), and advertisements and web postings ($n = 22$). Recruitment information was missing for one child. Outbursts had to 1) be out of proportion to the situation or precipitant and for the child's developmental level, 2) last at least 15 minutes, during which the child was inconsolable, 3) occur at least twice a week for at least 3 months prior to the evaluation, and 4) not occur exclusively during anxiety-provoking situations (e.g., in response to separation from parents). Fifty-one children with severe temper outbursts met these criteria. Five additional children were excluded (three had symptoms of autism; two had IQ's < 80).

Children without frequent, impairing outbursts (controls). A comparison group of 26 children without impairing temper outbursts, ADHD, or ODD were recruited from the community. Two children were excluded because they had ADHD and IQ's < 80 , resulting in a final sample of 24 controls.

Assessments

Child diagnosis. Parents were interviewed about their child with the Schedule for Affective Disorders and Schizophrenia for

School-Aged Children–Present and Lifetime version (K-SADS-PL; Kaufman et al. 1997) by either a trained clinical child psychologist or a trained child psychiatry fellow, blind to group membership. Because children were too young for a full diagnostic interview, only a brief clinical interview was conducted with the children, to further assess specific symptoms endorsed by the parent(s). Children's diagnoses reflected best estimates that relied on all available information, through consensus among the study clinicians blind to group membership. A diagnosis of ADHD-not otherwise specified (NOS) was assigned when teacher ratings were not available, but parents reported ADHD symptoms at home; therefore, the presence of impairment related to these symptoms in more than one setting could not be confirmed. A diagnosis of depressive disorder, NOS was given in circumstances in which the child did not meet criteria for MDD or dysthymia, but was described by parents as moody, irritable, or having impairing sad mood, and as having at least one associated depressive symptom such as low self-esteem. Irritability alone, without any other symptoms of depression, did not qualify for a diagnosis of depressive disorder NOS.

Child temper outbursts. A clinical interview version of the Outburst Monitoring Scale (OMS) was administered to parents as a measure of outburst severity (Kronenberger et al. 2007). This scale contains 20 items rated on a five point scale of how frequently they occurred over the past week (never, rarely [1–2 times], sometimes [3–6 times], often [1–2 times/day], or very often [≥ 3 times/day]). Items are classified into four subscales of aggressive behavior (verbal, property, physical, and self-injury). The subscales have shown adequate to excellent internal consistency, as well as moderate to strong relations with measures of disruptive behavior and aggression (Kronenberger et al. 2007).

SMD. SMD criteria include: non-episodic irritability, overreaction to negative emotional stimuli ≥ 3 times per week, and hyperarousal (i.e., at least three of the following symptoms: insomnia, distractibility, psychomotor agitation, racing thoughts/flight of ideas, pressured speech, intrusiveness) (Leibenluft et al. 2003). Irritability symptoms and their duration were assessed systematically using the ODD and Depression sections of the K-SADS-PL. Additionally, severity of irritability and other symptoms of SMD were evaluated using the Mood Symptoms Questionnaire (MSQ-7-Child Version; E. Leibenluft, personal communication, 2007). This clinician-completed measure generates three factor scores: Irritability (four questions related to severity, frequency, and duration of irritability), Outward Arousal (three questions related to distractibility, psychomotor agitation, intrusiveness), and Inward Arousal (three questions related to insomnia, racing thoughts, pressured speech) (Argyris et al., article in preparation). In this study sample, internal consistency estimates were excellent for the Irritability ($\alpha = 0.95$), good for Outward Arousal ($\alpha = 0.74$), and poor for Inward Arousal ($\alpha = 0.39$), likely because of the low base rate of these behaviors. Therefore, only the Irritability and Outward Arousal scores were examined.

Parent ratings of child behavior. Parents completed the Preschool version (for 5-year-olds; 134 items) or Child version (for children ≥ 6 years of age; 160 items) of the Parent Rating Scale (PRS) of the Behavior Assessment Scale for Children, 2nd edition (BASC-2; Reynolds and Kamphaus 2004), a norm-referenced measure of mood and behavioral symptoms. The PRS includes 2 broad scales of internalizing and externalizing problems, 12 clinical and adaptive scales, and 7 content scales. Of interest in this

study were the Aggression, Anxiety, Depression, Anger Control, Emotional Self-Control and Negative Emotionality scales.

Teacher ratings of child behavior. Teachers were asked to complete the Preschool version (for 5-year-olds, 100 items) or Child version (≥ 6 years of age; 139 items) of the BASC-2 Teacher Rating Scale (TRS) (Reynolds and Kamphaus 2004). The Clinical, Adaptive, and Content scales were the same as for the PRS, as were the subscales of interest. Teacher ratings were obtained for 43/51 (84.3%) probands and 20/24 (83.3%) controls.

Child self-report. Self-reports of behavior and mood symptoms were obtained from 6–9-year-olds (38 probands, 17 controls) using the BASC-2 Self-Report of Personality (SRP; Reynolds and Kamphaus 2004) administered orally by a trained research assistant to insure comprehension of scale items. Two children were unable to complete this measure. The Anxiety, Depression, Interpersonal Relations, and Social Stress scales were examined.

Regulation of emotional expressivity. Children completed the Balloons Game, a computerized task that assesses regulation of emotional expression in young children (Bar-Haim et al. 2011). Children see red, blue, and green balloons floating to the top of the screen. They are instructed to “pop” all of the green balloons by clicking on them with the mouse. Using practice trials, the speed of the balloons is calibrated individually so that each child is able to pop all the green balloons. At the end of each “success trial,” when the child has popped all the green balloons, the computer shows a happy picture and plays rewarding music. Unbeknownst to the child, the task is designed so that, during half of the trials, the mouse fails, and the child cannot pop all the balloons, inducing mild frustration. Following each “frustration trial,” the computer plays a failure sound and shows a sad picture.

The game is played under two conditions, low and high emotion regulation, while a video camera records the child's facial expressions. In the “low regulation” condition, the child plays the Balloons Game without additional instructions. In the “high regulation” condition, the child is asked to suppress any display of emotion so that “my friend who will watch the tape will not be able to know, by looking at your face, whether you are winning or losing the game.” The low regulation condition always preceded the high regulation condition to avoid carryover effects from the latter.

Each condition includes eight 40-second trials. The first two trials of each condition are success trials. The remaining six trials (three success and three failure) are presented in pseudo-random order. The last 12 seconds of each of these trials (6 seconds before and 6 seconds after the display of the picture and sound indicating success or failure) are coded for emotional expressivity using the Facial Action Coding System (FACS; Ekman et al. 2002). Following the procedures of Bar-Haim et al. (2011), facial expressions were coded using a selected set of 10 action units indexing negative emotional displays and 4 action units indexing positive emotion displays. Facial expressions were coded five times per second of the 12 second coding period resulting in 60 coding frames for each trial. This yielded 180 coding units for success trials and 180 coding units for failure trials for each of the two emotion regulation conditions. Facial expressions were coded by an undergraduate psychology student, blind to study goals, group membership, and condition, who was trained by a senior staff member who had obtained reliability with Dr. Bar-Haim's staff.

Because of technical issues, complete data from all six trials in each condition were not available for every child. Therefore, we

used mean facial expression counts, calculated by dividing the total number of coded expressions by the number of trials for that condition, rather than total counts (as in Bar-Haim et al. 2011). Means were derived for three variables per regulation condition: Positive expressivity during success (positive expressions during success trials/number of success trials coded), negative expressivity during failure (negative expressions during failure trials/number of failure trials coded), and total expressivity (positive and negative expressivity across all trials /total number of trials coded). Emotional reactivity was measured by these three indices during the low regulation condition, which serves as a baseline because the child is given no explicit instructions regarding regulation. Emotion regulation indices were calculated by subtracting expressivity during the low regulation condition from expressivity during the high regulation condition.

Parental psychiatric history. The Structured Clinical Interview for DSM Disorders (SCID; First et al. 1996) was administered to biological mothers and fathers by a trained clinical psychologist blind to the child's status. For the 51 probands, 43 biological mothers and 34 biological fathers were available. Of these, 34/43 mothers (79.1%) and 19/34 fathers (55.9%) consented to be interviewed. The 26 controls had 23 biological mothers and 16 biological fathers; 17/23 mothers (73.9%) and 10/16 fathers (62.5%) who consented to be interviewed. The low number of participating fathers precluded meaningful contrasts; therefore, findings in fathers were omitted. To assess inter-rater diagnostic reliability, an independent clinical psychologist with extensive SCID training reviewed 25% of the audiotaped interviews. Kappas ranged from 0.61 for past anxiety disorder to 1.0 for current mood disorder, with an overall mean kappa = 0.82.

Statistical analyses

Independent samples *t* tests, and χ^2 tests were used to test group differences on continuous variables and categorical measures, respectively. For the Balloons Game, three univariate analyses of covariance (ANCOVAs) were used to test for group differences in mean total expressivity, mean positive expressivity during success trials, and mean negative expressivity during failure trials for the low regulation (baseline) condition. Group contrasts for regulation of emotion expressivity were subjected to the same analyses, using total, positive, and negative emotion regulation indices as the dependent variables. Medication status was covaried to control for possible stimulant effects (three probands had taken stimulant medication the day of the evaluation). Significance was set at $p < 0.05$. As this study is the first of its kind, we also reported trends ($0.05 > p > 0.10$). For variables showing significant group differences, additional ANCOVAs were conducted to explore differences in emotion expressivity and regulation scores between probands with and without comorbid mood or anxiety disorders.

Results

Demographics

Probands. Of the 51 children with frequent, impairing outbursts (mean age = 6.59 ± 1.3 years), 66.7% were boys, 70% were Caucasian, 65% had parents who were married, and 48% were from families who reported an annual household income $> \$100,000$ (Table 1). Thirty-two probands (62.3%) had received treatment: 12 (23.5%) psychopharmacological, and 20 psychological. At study entry, 9 probands (17.6%) were on medication (1 fluoxetine, 8

TABLE 1. PARTICIPANT CHARACTERISTICS

Characteristic	Probands (n = 51)	Controls (n = 24)
Age in years (mean [SD]) ^a	6.59 (1.3)	6.71 (1.3)
Sex (n [%]) ^a	34 boys (66.7%)	16 boys (66.7%)
Race (n [%]) ^a		
Caucasian	36 (70.6%)	17 (70.8%)
African American	7 (13.7%)	4 (16.7%)
Asian	2 (3.9%)	1 (4.2%)
Other/Mixed	6 (11.8%)	2 (8.3%)
Parent marital status (n [%]) ^a		
Married	33 (64.7%)	16 (66.7%)
Not married, living together	3 (5.9%)	1 (4.2%)
Separated or divorced	2 (3.9%)	4 (16.7%)
Never married, not living together	13 (25.5%)	3 (12.5%)
Adoptive parents (n [%])	7 (13.7%)	0
Family income (n [%]) ^a		
< \$50,000	7 (14%)	6 (25%)
\$50,000- \$100,000	19 (38%)	6 (25%)
> \$100,000	24 (48%)	12 (50%)
IQ estimate (mean [SD]) ^a	108.4 (16.9) ^b	109.6 (13.0)
School-based services (n [%]) ^c		
Classroom interventions ^d	15 (29.4%)	1 (4.2%)
Specialized services ^e	29 (56.8%)	5 (20.8%)
Counseling	17 (33.3%)	1 (4.2%)

^aNo significant group differences for age, sex, race, parental marital status, family income, or IQ.

^bIQ missing for one proband.

^cMany children were receiving more than one service.

^dThese include special education classroom, having a classroom aide, and receiving resources room services.

^eThese include speech/language therapy, occupational therapy, and physical therapy.

IQ, intelligence quotient.

methylphenidate) and 10 (19.6%) were receiving outpatient psychological treatment. As shown in Table 1, many were receiving school-based services.

Controls. The 24 controls (mean age = 6.71 ± 1.3 years; 16 boys) did not differ significantly from the probands in age, sex, race, socioeconomic status, IQ, or parent marital status (Table 1). None was receiving outpatient treatment; four (16.7%) were receiving school-based support services (Table 1).

DSM-IV disorders

As shown in Table 2, no child met DSM-IV criteria for bipolar disorder types I, II, or NOS, or for mania, and none had a diagnosis of major depression. Of 51 probands, 17 (33.3%) were diagnosed with a mood disorder other than major depression: 2 (3.9%) were diagnosed with dysthymia, and 15 (29.4%) with a depressive disorder, NOS. The children with depressive disorder, NOS exhibited significant symptoms of negative mood or irritability, but did not meet criteria for a specific disorder. For example, most of these children exhibited irritability along with frequent brief periods of sad or negative mood that interfered with their functioning, and/or other associated symptoms (i.e., low self-esteem, heightened rejection sensitivity). None of the comparison children met criteria for a depressive disorder.

TABLE 2. DSM-IV DISORDERS

Diagnosis ^a	Probands n = 51 n (%)	Controls n = 24 n (%)
ADHD	38 (74.5%)	0
Combined type	20 (39.2%)	
Predominantly inattentive type	10 (19.6%)	
Predominantly hyperactive-impulsive type	1 (2.0%)	
ADHD NOS	7 (13.7%)	
Oppositional defiant disorder	45 (88.2%)	0
Any mood disorder	17 (33.3%)	0
Bipolar disorder (I, II, NOS)	0	
Mania	0	
Major depressive disorder	0	
Dysthymia	2 (3.9%)	
Depressive disorder, NOS	15 (29.4%)	
Any anxiety disorder	25 (49.0%)	4 (16.7%)
Generalized anxiety disorder	6 (11.8%)	1 (4.2%)
Separation anxiety disorder	8 (15.7%)	1 (4.2%)
Social phobia	6 (11.8%)	0
Specific phobia	5 (9.8%)	2 (8.3%)
Obsessive-compulsive disorder	2 (3.9%)	0
Other disorders		
Severe mood dysregulation (SMD)	11 (21.6%)	0
Disruptive behavior disorder NOS	2 (3.9%)	0
Tic disorder	4 (7.8%)	0
Enuresis/ Encopresis	1 (2.0%)	1 (4.2%)
Gender identity disorder	1 (2.0%)	0
Meets diagnostic criteria for 1 disorder	5 (9.8%)	5 (20.8%)
Meets diagnostic criteria for 2 disorders	20 (39%)	0
Meets diagnostic criteria for ≥ 3 disorders	26 (51%)	0

^aBased on best estimate by clinicians.

DSMIV, American Psychiatric Association, *Diagnostic and Statistical Manual of Mental Disorders*, 4th ed.; ADHD, attention-deficit/hyperactivity disorder; NOS, not otherwise specified.

ODD and ADHD were highly prevalent among probands, with 45/51 (88.2%) meeting criteria for ODD, 38 (74.5%) for ADHD, and 34 (66.7%) for both. Twenty-five probands (49%) were diagnosed with at least one anxiety disorder. More than 90% of probands met DSM-IV criteria for two or more disorders (Table 2). Twenty (39%) met criteria for two, and 26 (51%) met criteria for three or more disorders. Four (16.7%) of the comparison children met criteria for an anxiety disorder (generalized anxiety disorder [GAD], social anxiety disorder [SAD], specific phobia), and one met criteria for enuresis.

Parent reports of outbursts

On average, probands exhibited three or more developmentally inappropriate outbursts per week. Parents reported that temper outbursts were most often triggered by frustration, or the child not getting his or her way. Verbal aggression was the most frequent behavior, occurring 3–6 times a week (mean OMS score = 2.0 ± 0.76), followed by property aggression (mean OMS score = 1.37 ± 0.68), physical aggression (mean OMS score = 0.65 ± 0.50), and self-injury (mean OMS score = 0.43 ± 0.50).

Parent reports of SMD symptoms

On the MSQ-7, parents of probands reported more severe irritability (mean = 11.9 ± 2.6) than did parents of controls (mean =

1.6 ± 2.5) ($t[72] = 16.0, p < 0.001$). Thirteen children with outbursts (25.5%) met the SMD criterion of having irritable mood >50% of the day. The Outward Arousal factor score was significantly elevated in the children with severe outbursts (mean = 4.6 ± 1.8) relative to controls (mean = 1.3 ± 1.3; $t[73] = 7.68, p < 0.001$). This group difference may be largely accounted for by probands who had ADHD ($n = 38/51, 75\%$), as they had significantly higher outward arousal scores (mean = 5.2 ± 1.4) than did probands without ADHD ($n = 13$; mean = 2.8 ± 1.7) ($t[49] = 4.95, p < 0.001$). However, hyperarousal scores in the 13 probands without ADHD were still significantly higher than those of controls ($t[34] = 3.38, p = .002$). Overall, based on the MSQ-7 and parent clinical interviews with the K-SADS-PL, 11 children (21.6%) with frequent, impairing outbursts met criteria for SMD. This group included the two children diagnosed with dysthymia, five with depressive disorder, NOS, as well as four children with no DSM-IV mood diagnosis.

Parent and teacher BASC-2 ratings

As shown in Table 3, on the BASC-2 Parent Rating Scale, mean T scores of children with outbursts were >1.5 standard deviations

TABLE 3. PARENT, TEACHER, AND CHILD RATINGS ON BASC-2 SCALES

	Probands Mean (SD) n = 51	Controls Mean (SD) n = 23	Group difference $t(72) =$	p <
<i>Parent scales</i>				
Hyperactivity	66.4 (12.0)	48.2 (9.0)	6.54	0.001
Attention problems	63.7 (9.0)	47.0 (8.8)	7.44	0.001
Conduct problems ^a	65.8 (14.4)	47.6 (7.8)	5.02	0.001
Aggression	67.7 (10.3)	45.6 (6.1)	9.55	0.001
Depression	65.9 (11.6)	46.3 (5.7)	7.64	0.001
Anxiety	55.0 (11.9)	47.8 (10.0)	2.54	0.014
Anger control	67.0 (11.2)	46.7 (8.8)	7.65	0.001
Emotional self-control	72.4 (9.9)	46.4 (6.7)	11.38	0.001
Negative emotionality	70.0 (8.0)	47.0 (9.3)	10.99	0.001
<i>Teacher scales</i>				
	n = 43	n = 20	$t(61) =$	p <
Hyperactivity	65.9 (13.5)	46.4 (6.9)	6.05	0.001
Attention problems	59.9 (9.4)	47.3 (8.5)	5.10	0.001
Conduct problems ^a	58.8 (11.6)	47.3 (6.9)	3.73	0.001
Aggression	65.6 (17.9)	47.8 (7.4)	4.26	0.001
Depression	64.0 (18.2)	47.9 (5.6)	3.86	0.001
Anxiety	50.1 (14.2)	48.7 (11.5)	2.86	0.01
Anger control	63.6 (12.9)	46.8 (7.0)	5.44	0.001
Emotional self-control	67.9 (16.0)	49.1 (6.5)	5.03	0.001
Negative emotionality	64.9 (16.5)	47.3 (8.2)	4.15	0.001
<i>Child scales</i>				
	n = 38	n = 17	$t(53) =$	p =
Depression	53.3 (12.2)	46.0 (6.3)	2.93 ^b	0.005
Anxiety	53.0 (13.0)	48.1 (8.6)	1.41	0.16
Interpersonal Relations	51.5 (11.4)	53.0 (8.2)	0.49	0.63
Social Stress	52.9 (10.8)	47.5 (8.6)	1.82	0.08

^aConduct problems subscales were only available for children ≥ 6 years of age (39 probands, 18 controls for parent ratings and 32 probands, 17 controls for teacher ratings).

^bDegrees of freedom = 51.9, adjusted for unequal variances (Levene's test: $F = 10.25; p = 0.002$).

BASC-2, Behavior Assessment Scale for Children, 2nd ed.

(>65) above the population mean (50) for each scale of interest, except Anxiety. Similarly, on the BASC-2 Teacher Rating Scale, mean T scores of children with outbursts were at least one standard deviation above the mean (>60) for all scales except Anxiety. For both parent and teacher BASC-2 scales, mean T scores of comparisons were all <50, resulting in significant group differences, as shown in Table 3.

Child self-report

Among children who were able to complete the BASC-2 self-report (38 probands, 17 controls), probands had higher scores on the Depression subscale than did comparison children ($t[51.9]=2.32$, $p=0.005$, Table 3). No group differences were observed on the Anxiety, Interpersonal Relations, and Social Stress scales.

Emotion regulation

Data were obtained for 30 probands and 16 controls. The task was not available for the first 5 participants, and 24 children were excluded (12 because of technical problems and 12 because they failed to complete the task). There were no significant differences in group membership, age, sex, or BASC-2 scores between children who completed the task and those who did not.

During the low regulation (baseline) condition, a significant group difference was found in positive expressivity in response to success ($F[1, 46]=4.44$, $p=0.04$); children with outbursts exhibited fewer positive expressions (mean = 10.0 ± 6.97) than did controls (mean = 14.73 ± 9.6) (Fig. 1A). No group differences were found in total expressivity ($F[1, 46]=2.30$, $p=ns$), or in negative expressivity during failure (frustration) trials ($F[1, 46]=0.04$, $p=ns$). A significant group difference was found for regulation of negative expressivity in response to frustration ($F[1, 46]=3.95$, $p=0.05$). As shown in Figure 1B, compared with controls (mean = -0.78 ± 4.6), probands exhibited less regulation of negative expressivity (mean = 3.5 ± 6.8). There were no group differences in regulation of total expressivity ($F[1, 46]=1.05$, ns) or in positive expressivity during success trials ($F[1, 46]=0.61$, $p=ns$).

Exploratory analyses found that within the proband group, those with an anxiety disorder exhibited less positive expressivity than did those without anxiety disorders ($F[1, 30]=5.30$, $p=0.03$; Fig. 2A); no difference in positive expressivity was found between those with and without a depressive disorder ($F[1, 30]=0.80$, $p=ns$). Conversely, those with depressive disorders exhibited poorer regulation of negative expressivity (higher scores) than did those without depressive disorders ($F[1, 30]=4.70$, $p=0.04$; Fig.

2B); no difference was found for those with and without an anxiety disorder ($F[1, 30]=1.03$, $p=ns$).

Maternal lifetime psychiatric diagnoses

Of the 51 probands, 43 had biological mothers who were invited to complete the parent psychiatric interview (6 had adoptive mothers, 2 were living exclusively with their fathers). Diagnostic interviews were completed with 34/43 biological mothers of probands, and 17/24 mothers of controls. No group differences were found for lifetime diagnoses of bipolar disorder (probands, $n=2$; controls, $n=0$). There was a trend for a higher prevalence of lifetime depressive disorders in mothers of probands ($n=21$; 61.8%) than in those of controls ($n=6$; 35.2%) ($\chi^2[1]=3.19$; $p=0.07$). Among the 21 mothers of probands with a lifetime history of depression, the onset of the depressive disorder preceded the probands' birth in 57.1% of cases (12/21). Mothers of probands had a higher rate of lifetime anxiety disorders ($n=26$; 76.5%) than did mothers of controls ($n=6$; 35.3%) ($\chi^2[1]=8.22$; $p=0.004$). Onset of anxiety disorders preceded the birth of probands for 13 mothers (50%). No group differences were found for lifetime diagnoses of substance disorders (probands $n=6$; controls $n=3$) or ADHD (probands $n=2$; comparisons $n=2$).

Discussion

This study represents an initial effort to characterize young children with impairing temper outbursts and their parents, and to experimentally examine emotion regulation deficits. The significant impairment suffered by young children with frequent, impairing, outbursts is demonstrated by the high rates of psychiatric comorbidity, with >50% meeting criteria for three or more DSM-IV disorders. A key finding is the lack of clinical evidence of mania in these children. Rather, disruptive behavior disorders were common: ODD was virtually ubiquitous (88%), followed by ADHD (75%).

No child qualified for a diagnosis of current or past major depression, but one third (17/51) exhibited impairing depressive mood symptoms. In most of these children, irritability was the predominant mood symptom, accompanied by either brief periods of sad mood or associated depressive features such as low self-esteem and rejection sensitivity. Persistent sad mood without prominent irritability was a salient feature for only three children. Therefore, most young children with severe outbursts (78.4%) did not exhibit persistent negative mood (i.e., "irritable or sad mood more than half of the time"), and therefore did not meet criteria for SMD, nor would they meet criteria

FIG. 1. (A) Mean number of positive facial expressions (expressivity) during the low regulation condition in probands and comparisons and (B) mean negative regulation scores in probands and comparisons.

FIG. 2. Mean number of positive facial expressions (expressivity) during the low regulation condition (**A**) and mean negative regulation scores (**B**) in probands with (ANX) and without an anxiety disorder (NO ANX) and probands with (DEP) and without a depressive disorder (NO DEP). * $p < .005$.

for DMDD according to the upcoming revision of the DSM. However, as our subjects were considerably younger than those in studies of SMD (current sample: 6.6 ± 1.3 years; SMD sample 11.7 ± 2.5 years [Leibenluft 2011]), they may represent an early developmental manifestation of SMD, with chronic irritability possibly emerging later in childhood.

Young children with severe temper outbursts showed altered emotional reactivity as well as deficits in emotional regulation. When the children were not instructed to restrict their emotional expressions while playing the Balloons Game (low regulation condition), probands exhibited less positive expressivity in response to success than their peers, but groups did not differ in negative expressivity. Probands also showed a significantly lower ability to regulate negative emotional expressivity when frustrated. In fact, probands showed an *increase* in negative emotional expressivity during high regulation trials, relative to controls. Within probands, this increase during the high regulation condition was even more pronounced in those with a depressive disorder than in those without. Overall, these findings suggest that young children with severe outbursts may not have increased rate of negative emotions in response to frustration, but rather have deficits in the regulation of these reactions once they occur. Integration of this behavioral task with neuroimaging techniques will inform whether these deficits are related to disruptions in neural circuits mediating emotion regulation.

In this small sample, there were no group differences in the rates of maternal lifetime depressive or bipolar disorder. However, lifetime anxiety disorders were more than twice as prevalent in mothers of children with outbursts than in mothers of typically developing children. We explored whether these disorders arose in mothers after the birth of a child who was difficult to manage, but found no clear evidence of a relationship between onset of mothers' mental disorders and birth of probands. This suggests a possible nonspecific relationship between maternal anxious psychopathology and offspring mood dysregulation (Leckman-Westin et al. 2009). Maternal psychopathology has been shown to impact mothers' reporting of their child's difficulties (Briggs-Gowan et al. 1996); such a bias cannot be ruled out in the current study.

Our finding of relatively elevated prevalence of anxiety disorders among children with severe outbursts (nearly 50%) is consistent with reports of adolescents with SMD (Dickstein et al. 2005) and of outpatient children with rages (Carlson and Dyson 2012). In our sample, as indicated, we also found that anxiety disorders were associated with significantly less positive expressivity during the low regulation condition of the Balloons Game. Further, children with pervasive emotion dysregulation across contexts as indicated

by elevated teacher and parent ratings on the BASC-2 Emotional Self-Control scale were more likely to receive an anxiety disorder diagnosis. Together, these findings suggest a relationship between anxiety and emotional expression in children with severe temper outbursts. A recent study of child inpatients found that those with severe outbursts who also had anxiety disorders exhibited greater distress during tantrums than nonanxious children with outbursts (Potegal et al. 2009). At this time, the nature of the relationship between anxiety and temper outbursts is unclear and, therefore, we cannot discern whether the outbursts are a complication of anxiety disorders and whether treatment of the anxiety may reduce tantrums for this group of children.

Limitations

A number of limitations are noted. First, we present a largely Caucasian, middle class sample, and findings may not apply to other groups. Second, the number of mothers evaluated is limited, as is the number of children with mood and anxiety disorders, and power may have been insufficient to detect true associations between parent and child psychopathology. Third, this initial study relied on comparisons with a group of children without significant behavioral concerns, as we did not know which pathological group would have been an appropriate comparison (i.e., depressed children or those with other disorders) at the outset. As we found that $\sim 75\%$ of probands had ADHD, it would seem appropriate for future studies to include children with ADHD, free of severe outbursts, as comparisons, to identify deviance specific to severe outbursts. Fourth, we did not include any teacher-reported measures of tantrum activity. Recent evidence suggests diagnostic differences between children who exhibit outbursts across settings and those who tantrum in only one setting (either school or home) (Carlson and Dyson 2012). We took an initial look at this using the Emotional Self-Control scale of the BASC-2 for which we had parent and teacher reports, and found trends (p 's < 0.06) for increased frequency of anxiety and mood disorders in those children who had elevated ratings on both parent and teacher reports versus those with only parent-reported difficulties (elevated teacher ratings without parent ratings were only found for five children, precluding any statistical analyses in this group). This contrasts previous findings of increased mood and anxiety symptoms in children with only parent-reported rages (Carlson and Dyson 2012). Finally, we relied on the K-SADS-PL and MSQ to assess SMD symptoms, rather than the diagnostic module for SMD (Leibenluft et al. 2003), perhaps limiting detection of SMD.

Conclusions

The National Institute of Mental Health (NIMH) recently initiated the Research Domain Criteria project (RDoC) to improve classification of psychopathology based on observable behavior. One of the key elements of this initiative is to focus investigations on behaviors that cut across diagnostic categories, with the ultimate goal of understanding their biological bases, and using this information to define new classifications. In children, severe temper outbursts are a source of significant clinical impairment across a range of diagnoses including ADHD, ODD, and mood and anxiety disorders, as was clearly the case in the present sample. As such, this study represents a preliminary step toward this broader research initiative. In particular, the introduction of a novel behavioral task to directly assess affect regulation supports a key facet of the RDoC initiative, which is to use behavioral measures to improve the objective characterization of individual differences.

Overall, study findings confirm that young children with frequent, severe outbursts compose a heterogeneous clinical group characterized primarily by oppositional symptoms, behavioral disinhibition, and anxiety. No child met DSM-IV criteria for bipolar, or major depressive, disorder. Only a minority presented with significant, impairing mood symptoms besides tantrums, suggesting that chronic irritability and mood disturbances are not typical, as has been suggested for adolescents with SMD. Whether the co-occurrence of severe outbursts, periodic sad mood, anxiety, and poor emotion regulation, presages future development of full-fledged mood disorders in adolescence or adulthood awaits longitudinal study.

Clinical Significance

Over the past few years, there has been significant discussion regarding the appropriate diagnosis and treatment of children with severe temper outbursts. The present findings do not support that severe temper outbursts are indicative of bipolar disorder; rather, most children with these tantrums have multiple comorbidities including ADHD, oppositional defiant disorder, and anxiety and depressive disorders. These results have significant implications for the pharmacological treatment of these children. Further, these young children demonstrate deficits in the expression of positive emotions and the regulation of negative facial expressions in response to frustration. This provides preliminary experimental evidence of specific emotional deficits that might benefit from targeted interventions

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Disclosures

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