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Family functioning Deficits in bipolar disorder and ADHD in youth

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ABSTRACT

Background: Rates of diagnosis and treatment for bipolar disorder (BD) in youth continue to rise. Researchers and clinicians experience difficulty differentiating between BD in youth and other conditions that are commonly comorbid or share similar clinical features with BD, especially attention-deficit/hyperactivity disorder (ADHD). Comparative studies of the phenomenology and psychosocial correlates of these conditions help to address this. Family functioning is an important topic for both BD and ADHD since both are associated with numerous family-related deficits. One previous study suggested that manic/hypomanic youths' family functioning differed from ADHD and typically developing control (TDC) groups. However, many family functioning studies with BD and ADHD youth have methodological limitations or fail to use comprehensive, validated measures. **Methods:** This investigation used an adolescent report on the Family Assessment Device (FAD), based on the McMaster Model of family functioning. Youth were recruited in BD ($n=30$), ADHD ($n=36$), and TDC ($n=41$) groups. **Results:** Groups were similar on most demographic variables, but the TDC group scored somewhat higher than the others on IQ and socioeconomic status. FAD results indicated that BD and ADHD groups scored worse than TDC on the General Functioning and Roles scales of the FAD. In addition, the BD group showed impairment on the Problem Solving scale relative to TDC. **Limitations:** sample size, lack of parent report, ADHD comorbidity in BD group. **Conclusions:** Family functioning deficits distinguish both clinical groups from TDC, and problem-solving dysfunction may be specific to BD. These findings may apply to treatment models for both conditions.

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1. Introduction

Pediatric bipolar disorder (BD) represents a significant public health problem, with rates of children and adolescents discharged from psychiatric hospitals with a diagnosis of BD in the United States increasing from 5% to 20% within the last 10–15 years, and similar patterns emerging in other nations (Blader and Carlson, 2007; Holtmann et al., 2010). Rising rates of pediatric BD are also evident in outpatient mental health settings, with a 40-fold increase in office visits by youth with BD to all mental health providers over the same time period (Moreno et al., 2007). In fact, recent evidence from a large community sample suggests that the prevalence of BD in adolescents approaches that of adults (Merikangas et al., 2012).

Among the factors potentially implicated in this rise is the overlap between Diagnostic and Statistical Manual Fourth Edition (DSM-IV) symptoms of mania and symptoms of other psychiatric diagnoses, including attention-deficit/hyperactivity disorder (ADHD). Specifically, the DSM "A" criteria for a manic episode requires either elevated/

expansive (aka "euphoric") or irritable mood. While euphoric mood is specific to mania and BD, irritable mood is not. Moreover, there are similarities between some ADHD symptoms and some DSM "B" criteria for mania—i.e., distractibility (chronically in ADHD and present during manic episodes), blurring out/interrupting (ADHD) vs. pressured speech (BD), and hyperactivity (ADHD) vs. psychomotor agitation (BD). These symptoms have lower discriminant validity between BD and ADHD samples (Geller et al., 2002). Thus, there is a great need for comparative studies given that symptom overlap and similarities in presentation can result in diagnostic disagreement (i.e., ADHD vs. BD vs. comorbid ADHD and BD) between providers.

Towards that end, family functioning is a particularly salient domain to examine in children and adolescents with psychopathology, particularly those with BD and ADHD. Studies have shown that children with BD have impairments in several family-related dimensions, including general family functioning, maternal warmth, expressed emotion, conflict, and family stress (Algorta et al., 2011; Belardinelli et al., 2008; Esposito-Smythers et al., 2006; Keenan et al., 2011; Kim et al., 2007; Miklowitz and Johnson, 2009; Sullivan and Miklowitz, 2010; Townsend et al., 2007). However, it is difficult to determine if family dysfunction is a trait characteristic of pediatric BD, or if it is state-dependent, corresponding to mood status (e.g., mania, depression, or euthymia) or

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overall impairment from psychopathology. This relationship remains unclear in the pediatric BD family functioning literature due to methodological limitations in currently published studies including: the use of mixed diagnostic groups (combining BD youths with those having other mood disorders; e.g., [Fristad and Clayton, 1991](#); [Lange et al., 2005](#)), aggregating BD youths with considerable mood state heterogeneity, or failing to assess current mood state, and limited use of comparison groups such as typically-developing controls (TDC) or psychiatric controls (e.g., those with ADHD).

Numerous facets of family functioning have been associated with a diagnosis of ADHD. For example, there is an extensive literature regarding general psychosocial impairment (e.g., [Altepeter and Breen, 1992](#); [Scahill et al., 1999](#)) and dysfunctional parent-child interactions (e.g., [Johnston, 1996](#)) in samples diagnosed with ADHD. Studies have also shown that families of children with ADHD have impairments in other aspects of family environment compared to TDC, such as greater conflict and decreased organization, more use of negative parenting practices, impaired marital relationship in parents, and increased parental divorce rate ([Johnston and Mash, 2001](#); [Pressman et al., 2006](#); [Schroeder and Kelley, 2008](#); [Wymbs et al., 2008](#)). Thus, there is a large body of literature on family dysfunction in ADHD youths' families which could be compared to other diagnostic groups and linked to general theories of family functioning.

To our knowledge, only one study to date has directly compared family functioning in youth with BD (current mania or hypomania), ADHD, and TDC participants ([Geller et al., 2000](#)). Using the semi-structured Psychosocial Schedule for School Age Children-Revised (PSS-R) interview, this study found that BD and ADHD youths' families had worse functioning in several aspects of parent-child relationships, including consistent limit-setting, parental hostility, mutual warmth, marital problem solving, and parental agreement on child rearing. BD youths' families also reported greater impairments than ADHD and TDC participants in maternal warmth, and maternal/paternal tension and hostility ([Geller et al., 2000](#)). However, this study highlighted the need to evaluate family functioning in non-manic/hypomanic BD participants to clarify the relationship between current mood and family functioning.

Therefore, we sought to expand on the work of [Geller et al. \(2000\)](#) by evaluating family functioning of children in BD, ADHD, and TDC groups using the Family Assessment Device (FAD; [Epstein et al., 1983](#); [Miller et al., 1985](#)). In contrast to the PSS-R, the FAD is solely focused on family functioning, is grounded in extensive literature on the McMaster Model of Family Functioning (MMFF; [Epstein et al., 1978](#)), and has been used to study children and adults ([Garoff et al., 2012](#); [Pritchett et al., 2011](#); [Youngstrom et al., 2011](#)). The FAD measures overall family functioning as well as the six structural elements of the MMFF including: Problem Solving, Communication, Behavior Control, Roles, Affective Responsiveness, and Affective Involvement ([Epstein et al., 1978](#)). We hypothesized that both BD and ADHD groups would have significantly worse family functioning across all FAD scales, compared to the TDC group, but made no a priori hypotheses as to whether or not the BD and ADHD groups would differ significantly from one another on the FAD. We included BD participants in any mood state to allow examination of the relationship between current mood symptoms and family functioning

2. Methods

2.1. Participants

Participants were enrolled in an institutional review board-approved study conducted at an academic-affiliated child psychiatric hospital after informed consent and assent were obtained. Participants were recruited through advertisements distributed to

local physicians' offices as well as placed on local and national websites. Inclusion criteria for all groups were: age between 7–17 years, English fluency, and a consenting parent/guardian. Exclusion criteria for all groups were: IQ \leq 70; autism or an autism spectrum disorder; current psychosis interfering with the child's capacity to comply with study procedures; medical illness that was unstable or could contribute to the symptoms of BD or ADHD; or substance abuse within two months of participation.

The BD group ($N=30$) inclusion criterion was: meeting DSM-IV-TR criteria for BD, including a history of at least 1 episode meeting full duration criteria for hypomania (≥ 4 days) or mania (≥ 7 days) wherein the child exhibited abnormally elevated or expansive mood accompanied by at least 3 other DSM-IV criterion "B" mania symptoms. Children with only irritability, without elevated or expansive mood, were excluded from this group as were children with BD "not otherwise specified." Children with only a hypomanic episode also were required to have at least one lifetime major depressive episode. Thus, all BD participants met [Leibenluft \(2003\)](#) criteria for narrow-phenotype BD.

ADHD group ($N=36$) inclusion criterion was: meeting DSM-IV-TR diagnostic criteria for ADHD, (any subtype) with impairment in at least two settings (i.e., home, school, or peers). An additional exclusion criterion for the ADHD group was: lifetime history of any mood or anxiety disorder.

TDC group ($N=41$) inclusion criteria were: absence of current and lifetime psychiatric illness, absence of substance abuse/dependence, and absence of psychiatric history in first-degree relatives.

2.2. Procedure

Following a telephone interview to screen for relevant symptoms, potential participants were invited for an on-site assessment which included a diagnostic interview, the Schedule for Affective Disorders and Schizophrenia for School-Aged Children, Present and Lifetime Version (K-SADS-PL; [Kaufman et al., 1997](#)). The K-SADS-PL was administered by doctoral-level clinicians (M.D. or Ph.D.) with established inter-rater reliability ($\kappa \geq 0.85$). The principal investigator, a board-certified child and adolescent psychiatrist with more than a decade of expertise with pediatric BD, led a consensus conference to generate diagnoses. Comorbid diagnoses for the BD group were assessed by asking about symptoms and associated impairment during periods of generally euthymic mood, to avoid counting manic or depressive BD symptoms toward another diagnosis. Current mood symptom severity was assessed for the BD participants with the Young Mania Rating Scale (YMRS; [Young, Biggs, Ziegler, & Meyer, 1978](#)) and Children's Depression Rating Scale—Revised (CDRS-R; [Poznanski et al., 1984](#)). For the BD and ADHD groups, current global functioning was measured with the Children's Global Assessment Scale (C-GAS; [Shaffer et al., 1983](#)). The Wechsler Abbreviated Scale of Intelligence ([Wechsler, 1999](#)) was administered to all participants as a brief estimate of cognitive abilities. Demographic information was collected from parents, and socioeconomic status (SES) was categorized according to the Hollingshead Index ([Hollingshead, 1975](#)).

2.3. Family functioning assessment

Current family functioning was assessed through child report on the Family Assessment Device (FAD), a self-report questionnaire based on the MMFF ([Epstein et al., 1983](#)). The FAD consists of 60 items across seven scales: one for each of the six dimensions of the MMFF, and one which measures overall family health/pathology. The six MMFF dimensions include (1) **Problem Solving**: the family's ability to address problems in an adaptive way; (2) **Communication**: the style and clarity of verbal information sharing among individuals; (3) **Roles**: established behavior patterns to fulfill family functions; (4) **Affective Responsiveness**:

family members' ability to experience appropriate affect across situations; **(5) Affective Involvement**: the interest and value placed upon other family members' behaviors and concerns, and **(6) Behavior Control**: the way in which family members maintain expectations for each other's actions (Epstein et al., 1983). The seventh FAD scale, **General Functioning**, is an overall summary of family processes and its items are independent of the other six scales.

Items for each FAD scale are averaged to produce a summary score for that domain ranging from one to four, with higher scores indicating poorer current functioning. The FAD and its subscales have demonstrated reliability and validity across a number of age ranges and patient populations, and clinical cutoffs are available for classifying healthy versus unhealthy functioning on each subscale (Miller et al., 1985, 1986). The FAD and its underlying scales performed similarly to more in-depth, interview-based family functioning assessments (e.g., Barney and Max, 2005; Kabacoff et al., 1990). The FAD was chosen due to its wider age range than some similar measures (e.g., Family Environment Scale; Moos, 1990) and broader coverage of family function dimensions than others (e.g., Family Adaptability and Cohesion Evaluation Scales; Edman et al., 1990).

2.4. Statistical analyses

Analyses were conducted using IBM SPSS Statistics (version 19.0, IBM, Inc.). Group demographic differences were investigated using multivariate analysis of variance (MANOVA), with post-hoc contrasts for any significant differences identified. Lastly, the relationship between FAD scores and other clinical variables (i.e., CDRS-R, YMRS, and C-GAS) was calculated via Spearman rank correlation coefficients (rather than Pearson correlation coefficients) because these variables did not meet assumptions about normality and interval-level data.

A multivariate analysis of covariance (MANCOVA) was conducted using diagnosis (BD, ADHD, TDC) as the fixed factor and the seven FAD scales' total scores as dependent variables. SES was included in the model as a covariate *a priori*, due to evidence of its association with family functioning (e.g., Bradley and Corwyn, 2002). Given a significant main effect in the MANCOVA model, individual tests of between-subject effects were examined to identify which FAD scales significantly differed by group. Significant results for each individual FAD scale were followed by post-hoc between-group contrasts, with Bonferroni adjustment for multiple comparisons, to identify which groups were significantly different and generate estimates of effect size (Cohen's *d*).

In addition, we used established clinical cutoffs to categorize participants as "healthy" or "unhealthy" for each FAD scale (Miller et al., 1985, 1986). Chi-square analyses were conducted to test for between-group differences in the proportion of participants classified as "unhealthy" for each FAD scale. Where significant between-group differences emerged, individual chi-square contrasts were conducted to determine which specific groups scored differently. Again, a Bonferroni correction was applied to correct for multiple comparisons, resulting in a significance threshold set at $p \leq 0.01$. For each significant group contrast, phi (ϕ) coefficients were calculated as an estimate of effect size (Rosenthal, 1994).

3. Results

3.1. Participant characteristics

BD ($N=30$), ADHD ($N=36$) and TDC ($N=41$) groups did not differ significantly in age, sex, race, or ethnicity (Table 1). However, we did find a significant between-group difference in full-scale IQ

Table 1
Demographic characteristics of participants.

	BD ($N=30$)	ADHD ($N=36$)	TDC ($N=41$)
Age	13.4 (2.8)	12.4 (2.7)	12.6 (3.4)
Sex (% male)	63.3	52.8	39.0
Race (%)			
Caucasian	83.3	69.4	78.0
African American	3.3	8.3	2.4
Asian	–	5.6	4.9
Native American	–	5.6	–
> 1 race	13.3	8.3	7.3
Unknown/withheld	–	2.8	7.3
Ethnicity (% Hispanic)	3.3	8.3	7.3
Full-scale IQ	105.4 (11.0)	108.2 (12.8)	116.6 (11.2) ^a
SES	41.9 (14.4)	39.2 (16.4)	50.2 (12.9) ^b
Comorbidity (%)			
ADHD	70.0	–	–
ODD	70.0 ^c	19.4	–
Conduct disorder	3.3	0.0	–
Depression (CDRS–R)	29.17 ^c	20.31	–
Mania (YMRS)	8.33 ^c	4.51	–

Note. CDRS–R=Child Depression Rating Scale–Revised; YMRS=Young Mania Rating Scale.

^a TDC significantly higher than BD and ADHD groups ($p \leq 0.05$).

^b TDC significantly higher than ADHD group ($p \leq 0.05$).

^c BD group significantly higher than ADHD group.

(FSIQ) [$F(2, 104)=9.15, p \leq 0.01$] and SES [$F(2, 100)=5.63, p < 0.01$]. Pair-wise post-hoc analyses showed that this was driven by TDC participants having higher mean FSIQ than the BD ($p < 0.01$) and ADHD ($p < 0.01$) groups. Post-hoc analyses also indicated that the TDC group had significantly higher SES than the ADHD ($p < 0.01$) group.

A similar proportion of participants in the BD (86.2%) and ADHD (91.7%) groups were taking one or more psychiatric medication, (chi-square=0.50, *ns*), while no TDC participants took psychiatric medications, by design. A greater proportion of the ADHD group was currently prescribed stimulant medication, compared to the BD group (88.9% vs. 30.0% respectively, chi-square=23.81, $p < 0.01$). The rate of non-stimulant ADHD medication in the ADHD (13.9%) and BD (16.7%) groups did not significantly differ (chi-square =0.14, *ns*). No other medications were prescribed to participants in the ADHD group. Current medications for the BD group also included atypical neuroleptics (56.7%), lithium (36.7%), anti-epileptics (16.7%), anti-depressants (16.7%), and benzodiazepine sedatives (6.7%).

The BD group was largely euthymic, based on YMRS and CDRS mood ratings. Specifically, 22 participants (73.3%) were euthymic (defined as CDRS–R score < 40 and YMRS score ≤ 12), 4 participants (13.3%) were hypomanic (CDRS–R < 40 and YMRS > 12 and < 25), 2 participants (6.7%) were depressed (CDRS–R ≥ 40 and YMRS ≤ 12), and 2 participants (6.7%) were in a mixed state (CDRS–R ≥ 40 and YMRS > 12). No participants were manic (CDRS–R < 40 and YMRS ≥ 25) at the time of study participation. Importantly, all participants in this group were BD Type I, although Type II was not exclusionary. As expected, the mean CDRS–R score was higher in the BD group (29.17 ± 11.66) than the ADHD group (19.91 ± 5.78 ; $t=3.95, p < 0.01$). Similarly, the BD group (8.33 ± 7.44) scored significantly higher on the YMRS than the ADHD group (4.51 ± 4.36 ; $t=2.45, p=0.02$). Within the ADHD group, diagnostic sub-types were as follows: Combined Type=17 participants (81.0%), Hyperactive/Impulsive Type=one participant (4.8%), Inattentive Type=three participants (14.3%).

In the BD group, 21 participants (70.0%) had a current comorbid diagnosis of ADHD (85.7% Combined, $n=18$; 4.8% Hyperactive/Impulsive $n=1$; 9.5% Inattentive, $n=2$). In addition, participants in

the BD group had a significantly higher rate (70.0%) of current comorbid Oppositional Defiant Disorder (ODD) than the ADHD group (19.4%; chi-square=17.12, $p < 0.05$). The rate of comorbid Conduct Disorder did not differ significantly between the BD (0.3%, $n=1$) and ADHD (0%; chi-square=1.22, ns) groups.

3.2. Family functioning

Fig. 1 displays the mean scores for each of the FAD scales for the three groups. The results of our MANCOVA with the FAD scales as dependent variables and SES as the covariate indicated a significant effect of group (Wilks' Lambda=0.74, $F[14, 186]=2.13$, $p=0.01$), but not the covariate SES (Wilks' Lambda=0.93, $F[7, 93]=1.07$, ns). Three TDC participants and one participant in each of the BD and ADHD groups were excluded from the model because they did not report information for the covariate SES. Individual group by FAD scale comparisons were then examined. The effect of group was significant for Problem Solving [$F(2,99)=5.65$, $p < 0.01$], Roles [$F(2,99)=5.22$, $p=0.01$], and General Functioning [$F(2,99)=9.45$, $p < 0.01$].

Pair-wise comparisons for the Problem Solving scale revealed that the BD group had worse functioning than the TDC group ($p < 0.01$) with a large effect size (Cohen's $d=0.80$). There was no significant difference between the BD and ADHD groups, or between the ADHD and TDC groups. To further probe the specificity of this effect to BD vs. TDC, we ran a post-hoc ANCOVA comparison of the three groups on Problem Solving, again including SES as a covariate, but excluding all BD participants with comorbid ADHD ($n=21$). Despite a small BD group size ($n=9$), there was still a significant main effect of diagnostic group on Problem Solving, [$F(2, 78)=8.58$, $p < 0.01$]. Pair-wise comparisons indicated significantly higher scores in the BD group than both the TDC group ($p < 0.01$; Cohen's $d=1.33$) and ADHD groups ($p < 0.01$; Cohen's $d=0.98$).

On the Roles scale, the BD group scored significantly worse than the TDC group ($p=0.01$), with a large effect size (Cohen's $d=0.94$). The ADHD group also scored significantly worse than the TDC group ($p=0.03$), with a medium effect size (Cohen's $d=0.72$). There was no significant difference on this scale between the BD and ADHD groups.

On the General Functioning scale, group contrasts revealed that the BD group scored significantly worse than the TDC group

($\leq p < 0.01$), again with a large effect size (Cohen's $d=1.12$). There was no significant difference between the BD and ADHD or the ADHD and TDC youth on the General Functioning scale scores.

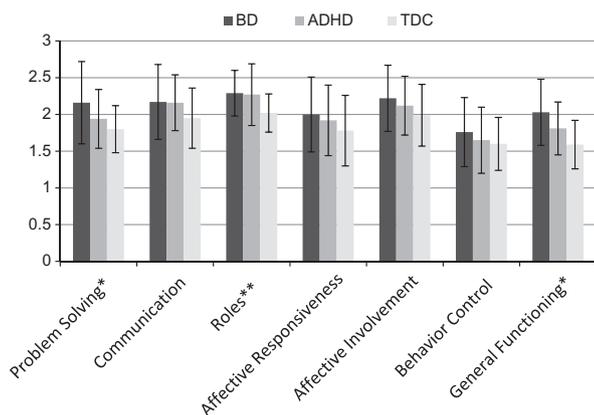
3.3. Influence of BD mood state on group differences in family functioning

To evaluate the influence of BD group participants' mood status on the findings above, we then conducted exploratory analyses by restricting BD group to only euthymic participants ($n=22$) and repeating the main analyses. That is, we sought to investigate whether current mood symptoms in BD participants had accounted for the group differences noted above, by removing those who were not in a euthymic state at assessment. Consistent with our main analyses, the MANCOVA indicated a significant main effect of group (Wilks' Lambda=0.68, $F[14, 166]=2.51$, $p < 0.01$), but not the covariate SES (Wilks' Lambda=0.92, $F[7, 83]=1.05$, ns) on FAD scores. The effect of group was significant for Problem Solving [$F(2,89)=10.15$, $p < 0.01$], Communication [$F(2,89)=4.46$, $p=0.01$], Roles [$F(2,89)=6.28$, $p < 0.01$], and General Functioning [$F(2,89)=12.32$, $p < 0.01$].

Pair-wise comparisons revealed the same differences previously found, with the BD group scoring worse than TDC on Problem Solving ($p < 0.01$, $d=0.53$), both BD ($p=0.01$, $d=1.09$) and ADHD ($p=0.01$, $d=0.74$) with significantly worse scores than TDC on Roles, and the BD group with worse functioning than TDC ($p < 0.01$, $d=1.40$) on General Functioning. Three additional group differences emerged as well. The difference between the BD and TDC groups on Communication reached statistical significance ($p=0.01$, $d=0.91$), with the BD group scoring in the direction of worse functioning. In addition, the BD group showed significantly worse functioning than the ADHD group on Problem Solving ($p=0.01$, $d=0.14$) and General Functioning ($p=0.01$, $d=0.74$). Therefore, restricting our sample only to euthymic BD participants maintained or increased the group differences found in the main analyses.

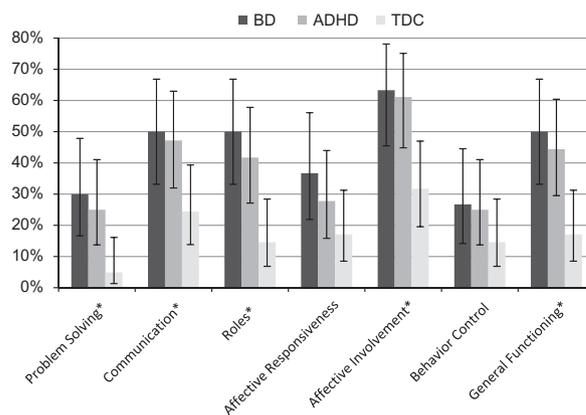
3.4. Post-hoc comparison of rates of clinical dysfunction by group

To evaluate the potential clinical impact of group differences in family functioning, we used published clinical cutoff scores to divide participants into "healthy" and "unhealthy" categories for each FAD dimension (Miller et al., 1986, 1985; see Fig. 2). We then conducted chi-square analyses to evaluate between-group



Note. Higher scores indicate worse functioning; significant differences based on MANCOVA model and post-hoc group contrasts
* TDC < BD
** TDC < both BD and ADHD

Fig. 1. FAD scores by diagnostic group. Note: Higher scores indicate worse functioning; significant differences based on MANCOVA model and post-hoc group contrasts. *TDC < BD, **TDC < both BD and ADHD.



Note. Error bars represent 95% confidence intervals
*At least one significant pairwise difference found between groups. See Table 2.

Fig. 2. Percentage of families in the clinical range of dysfunction by diagnostic group. Note: Error bars represent 95% confidence intervals. *At least one significant pairwise difference found between groups. See Table 2.

Table 2

Post-hoc group comparisons of percentage of families in the clinical range of dysfunction on FAD scales.

FAD subscale	BD-TDC χ^2 (sig.) ^a	ES ^b	ADHD-TDC χ^2 (sig.) ^a	ES ^b	BD-ADHD χ^2 (sig.) ^a	ES ^b
Problem solving	8.35 (< 0.01)	0.34^c	6.34 (0.01)	0.29^c	0.21 (0.65)	0.06
Communication	4.98 (0.02)	0.27	4.39 (0.04)	0.24	0.05 (0.82)	0.03
Roles	10.40 (< 0.01)	0.38^c	7.06 (< 0.01)	0.30^c	0.46 (0.50)	0.08
Affective involvement	7.00 (< 0.01)	0.31^c	6.68 (0.01)	0.30^c	0.03 (0.85)	0.02
General functioning	8.78 (< 0.01)	0.35^c	6.86 (< 0.01)	0.30^c	0.20 (0.65)	0.05

Note. Higher scores indicate worse functioning.

^a Significance threshold set at $p \leq 0.01$ to account for multiple comparisons.

^b Effect size (Phi [ϕ] coefficient).

^c Statistically significant effect size (i.e., $p \leq 0.01$).

differences in the proportion of families displaying “unhealthy” family functioning on each FAD scale. We found between-group differences in the following FAD dimensions: Problem Solving (chi-square=7.81, $p=0.02$), Communication (chi-square=6.62, $p=0.04$), Roles (chi-square=11.86, $p < 0.01$), Affective Involvement (chi-square=10.02, $p < 0.01$), and General Functioning (chi-square=9.46, $p=0.01$).

Post-hoc chi-square pairwise contrasts for these five scales revealed a number of significant group differences. Specifically, we found that the BD and ADHD groups both had a significantly higher proportion of families in the unhealthy range on the Problem Solving, Roles, Affective Involvement, and General Functioning scales, compared to TDC. There were no significant differences between the BD and ADHD groups for the proportion of families falling within the healthy vs. unhealthy range on any FAD scale (Table 2). Each of the significant group differences represented a medium effect size, based on phi coefficients, with one exception: the higher percentage of unhealthy functioning on Problem Solving in the ADHD group than the TDC group represented a small effect size.

3.5. Post-hoc evaluation of mood and functional status:

To examine the relationship between family functioning and clinical symptoms, we calculated Spearman rank correlation coefficients between the FAD scales and our measures of mood state and general psychosocial impairment in the BD and ADHD groups. In the BD group, we did not find a significant correlation between any FAD scale scores and current C-GAS, mania (YMRS), or depression (CDRS). In the ADHD group, C-GAS score was significantly correlated with Problem Solving (Spearman's rho=0.35, $p=0.04$) and General Functioning (Spearman's rho=0.38, $p=0.02$).

4. Discussion

There are three main findings from the present study, which compared family functioning in children with BD or ADHD to TDC. Most broadly, we found that both BD and ADHD families had worse General Functioning scores and were more likely to score in the clinical range on this domain than TDC families. Second, BD families had a specific deficit on the domain of Problem Solving. Third, BD and ADHD families were significantly impaired vs. TDC on Roles. In addition, the family impairments observed in the BD group were not accounted for, or even significantly associated with, current mood symptoms. Taken as a whole, our study evidences the resultant stress and impairment in families struggling with children with pediatric psychopathology, as well as highlighting potential new targets for family-based interventions for ADHD and BD youths.

This study extends the findings of Geller et al. (2000) by replicating their finding of family functioning impairments in BD

and ADHD groups using narrower TDC (i.e., no psychopathology in first degree-relatives) and broader BD (i.e., any current mood state) inclusion criteria. By using the FAD, the current study links these data to the broader literature on the MMFF, demonstrates cross-method convergence by utilizing a youth questionnaire as opposed to a semi-structured clinical interview, and allowed for classification of significant impairment through the use of clinical cutoff scores. By including BD participants regardless of current mood status, we were able to explore the relationship between mood symptoms and family functioning. When restricting our BD sample to only euthymic participants, group differences on FAD scores remained or increased, suggesting that these are trait, not state, phenomena. However, further work is necessary to confirm this using the FAD in samples of manic or depressed BD youths.

Our finding of worse functioning in BD families relative to TDC families on the Problem Solving scale is consistent with the literature regarding the importance of modifying maladaptive problem solving skills in the treatment of BD in youth (e.g., Simoneau et al., 1999; Townsend et al., 2007; Young and Fristad, 2007). That is, the psychosocial treatments currently available for BD in youth target problem solving skills by increasing ability to identify family conflicts/stressors and generate adaptive solutions (Fristad et al., 2009; Miklowitz et al., 2004; Pavuluri et al., 2004). Our post-hoc analyses evaluating the specificity of problem solving deficits in BD youths, although preliminary due to small group size, showed that BD youths without comorbid ADHD scored significantly worse than TDC and ADHD families. These differences, relative to the ADHD and TDC groups, were also observed when only euthymic BD participants were included. While these results are promising in suggesting that problem solving impairments may be specific to BD, we recognize that further study in larger samples of BD with and without comorbid ADHD are needed to confirm this possibility.

Impaired problem solving in BD families is also interesting, given the growing body of research demonstrating that BD youth show deficits in the brain/behavior interactions underlying these skills. For example, BD youths show impaired cognitive flexibility, or adaptation to changing rewards and punishments, as indexed by reversal learning tasks that require participants to learn by trial-and-error which of two stimuli is rewarded (vs. punished) and then to adapt when the previously rewarded stimulus is now punished (Dickstein et al., 2010). This has been demonstrated using two different behavioral tasks - the probabilistic response reversal task and the Cambridge Neuropsychological Test Automated Battery (CANTAB) intra-dimensional/extra-dimensional shift task—and is evident in BD youths compared to TDC, youths with depression or anxiety, and youth meeting Leibenluft et al. (2003) criteria for severe mood dysregulation (Dickstein et al., 2010, 2007, 2004; Gorrindo et al., 2005). Furthermore, functional MRI studies have shown that BD youths have the opposite pattern of brain activation in the prefrontal cortex and striatum than would be expected when performing a reversal learning task

(Dickstein et al., 2010), suggesting a possible neural mechanism involved in poor Problem Solving for BD youth. In context of our present study, further study is warranted to determine the role of aberrant brain/behavior interactions underlying cognitive flexibility in mediating family-based problem solving deficits in BD youths, as well as to determine if such reversal learning deficits are present in family members of BD youths.

The Roles dimension within the MMFF assesses “the extent to which families have established patterns of behavior for handling family tasks” (Miller et al., 1985). Our finding of impaired family roles in BD and ADHD youths aligns with previous studies which found similar deficits in youth and adults with ADHD, as well as in children with mood and anxiety disorders compared to TDC (Eakin et al., 2004; Lange et al., 2005). This difference was not accounted for by the present mood state of the BD participants, nor was it associated with global functioning in either clinical group. The high rate of comorbid ADHD in our BD group prevents us from evaluating whether the impairment on roles in the two groups represents a shared deficit present in both disorders, or is characteristic of ADHD overlap in the BD group. However, results from a large sample of BD youth suggests that comorbidity has an additive detrimental effect on family functioning (Esposito-Smythers et al., 2006). Future studies could investigate whether poor functioning on roles is present in BD in the absence of ADHD, and in other diagnostic groups. Our findings on this dimension, taken in context with the others mentioned above, may suggest novel targets for clinical assessment and psychotherapy intervention, considering that this impairment has been found across multiple clinical populations.

Our finding of overall family dysfunction (i.e., General Functioning scores) in the BD families is consistent with a several past studies demonstrating family-related dysfunction in these youth, such as family cohesion and conflict, maternal warmth, expressed emotion, parenting strategies, and family stress. However, some studies have found no difference in family functioning between BD youth and controls (Fristad and Clayton, 1991; Robertson et al., 2001). These inconsistent findings might be explained by methodological differences across studies such as mood state at assessment, different definitions or measures of family functioning, and grouping youth by broad diagnostic categories (e.g., mood or internalizing disorders). These differences limit our understanding of the specificity of family dysfunction dimensions to BD and the extent to which they are associated with mood state. We sought to address this last limitation by investigating the relationship between current mood symptoms and family functioning in our sample, and found no relationship between current global functioning, manic symptoms, or depression and family functioning in the BD group. However, as noted above, our BD sample was mostly euthymic at the time of assessment.

The implications for our finding of impaired affective involvement for both the BD and ADHD groups are less clear. This dimension of the MMFF measures “the extent to which family members are interested in and place value on each other’s activities and concerns” (Miller et al., 1986). One other study has also found impairment in affective involvement on the FAD in groups of youth with ADHD or “emotional disorder” (i.e., depression or anxiety; Lange et al., 2005). Replication with larger samples and clearly defined groups would aid in our understanding of these relationships.

4.1. Limitations

Our study has several potential limitations, including possible confounding effects of informant bias or parental psychopathology, SES and IQ differences by group, ADHD comorbidity in the BD group, and potential risk for type II error due to limited sample size.

Regarding demographic differences by group, SES was chosen as a covariate in the main analyses *a priori* due to potential confounding effects on the BD group. For example, higher income has been associated with more frequent euthymia and less time in manic/hypomanic states (Bauer et al., 2011). In the present sample, we observed that the TDC group scored higher than the others on IQ and SES; this may have contributed to the current results. Due to concerns about using a covariate in MANCOVA when it is related to the independent variable (see Miller and Chapman, 2001), the main analyses were repeated without any covariate, and the same results were observed.

With respect to potential informant bias, this investigation specifically targeted children’s evaluation of family functioning, so only child report on the FAD was obtained. Incorporating parent (s) report could have produced different findings, as parent–child disagreement on family functioning is not uncommon. Also, parental psychopathology was not measured in the BD and ADHD groups (though it was exclusionary in the TDC group), so its influence on our sample’s family functioning is unknown. Parental psychopathology may impact aspects of family environment (e.g., Oyserman et al., 2000; Pilowsky et al., 2006). However, one study found no association between parental diagnoses and FAD scores specifically in a child sample (Fristad and Clayton, 1991).

The high rate of comorbid ADHD in the BD group is consistent with similar studies in this population (e.g., Geller et al., 2000). Although we attempted to address this with exploratory post-hoc analyses, these rates reflect comorbidity found in clinical samples with BD (Pavuluri et al., 2005). Finally, this investigation could be limited by low power due to sample size. We observed medium effect size results on the Affective Involvement and General Functioning scales that were not statistically significant. It is possible that larger samples in each group may have produced a different pattern of group differences. This investigation is an important first step in understanding family functioning in children and adolescents with BD vs. ADHD through the use of a validated, systematic assessment like the FAD.

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Conflict of interest

All authors state that they have no conflicts of interest.

All authors have contributed to and have approved the final manuscript.

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