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ORIGINAL PAPER

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Clinical Correlates of Parenting Stress in Children with Autism Spectrum Disorder and Serious Behavioral Problems

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Abstract

Objectives We examined associations between parent-reported stress on the Parenting Stress Index (PSI) and clinical characteristics in children with autism spectrum disorder (ASD) and serious behavioral problems.

Methods The 298 children (259 males, 39 females; mean age 5.8 ± 2.2 years) were participants in one of two multisite randomized trials. The pre-treatment evaluation included standardized assessments of cognitive and adaptive functioning (Vineland Adaptive Behavior Scales) and parent ratings such as the Aberrant Behavior Checklist (ABC).

Results Parents of children above the median on disruptive behavior (ABC Irritability) and social disability (ABC Social Withdrawal) reported higher levels on PSI Parent–Child Interaction than children below the median (Irritability 33.0 ± 7.7 vs 28.4 ± 7.3; Social Withdrawal 33.4 ± 7.5 vs 27.9 ± 7.2, $p < .05$). Similar findings were observed for the PSI Difficult Child subscale. Bivariate logistic regression identified that these measures as well as greater adaptive functioning deficits (≤median on Vineland Daily Living) predicted parental membership in the upper quartile on the PSI. Stepwise logistic regression models showed that greater severity on ABC Social Withdrawal and greater deficits on Vineland Daily Living uniquely predicted parental membership in the highest quartile on the Parent–Child Interaction PSI subscale (ABC Social Withdrawal odds ratio = 3.4 (95% CI 1.82–6.32); $p < .001$; Vineland Daily Living odds ratio = 2.6 (95% CI = 1.34–4.87; $p < .001$).

Conclusions In addition to disruptive behavior, higher levels of social disability and lower levels of adaptive functioning are associated with parental stress on the PSI.

Keywords Autism Spectrum Disorder · Serious behavioral problems · Parenting stress · Parenting Stress Index-Short Form

The multiple and daily demands of parenting can be a source of stress for many parents (Abidin and Wilfong 1989; Deater-Deckard 2004). Rearing children with serious medical conditions or chronic disabilities predictably

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increases parental stress (Bouma and Schweitzer 1990; Deater-Deckard 2004). Autism spectrum disorder (ASD) is a chronic condition beginning in early childhood that is often accompanied by behavioral problems including tantrums, noncompliance, aggression, and self-injury. Results from several studies attest that parents of children with ASD report higher levels of stress than parents of typically developing children, children with other developmental disabilities or children with medical conditions (Estes et al. 2013; Hayes and Watson 2013; McStay et al. 2013; Schieve et al. 2011).

Children with ASD and behavioral problems pose additional challenges that may amplify parental stress (Giovagnoli et al. 2015; Hall and Graff 2012; Huang et al. 2013; Lecavalier et al. 2006; Zaidman-Zait et al. 2014; Zaidman-Zait et al. 2017). Behavioral problems in young children with ASD may interfere with parental efforts to promote daily living skills (Scahill et al. 2016). High levels of parenting stress in the context of behavioral problems may hinder the child's ability to make use of early intervention programs and undercut parental sense of competence (Iadarola et al. 2018; Osborne et al. 2008). Although findings are not consistent, some studies have shown that higher scores on measures of ASD severity in the child can also contribute to greater parenting stress (Huang et al. 2013; Shepherd et al. 2018; Zaidman-Zait et al. 2017). These differences across studies may be due to differences in the sample size, source of sample and measures applied.

The 36-item Parenting Stress Index Short Form (PSI) has been used to measure parenting stress in several samples of children with ASD (Hall and Graff 2012; Huang et al. 2013; Lecavalier et al. 2006; Zaidman-Zait et al. 2011; Zaidman-Zait et al. 2014; Zaidman-Zait et al. 2017). Although most of these studies accepted the original PSI factor structure, a few studies questioned the validity of the original PSI factor structure and did not use the original factors structure of the PSI (Dardas and Ahmad 2014; McStay et al. 2013; Zaidman-Zait et al. 2011). For example, in a sample of 411 young children with ASD, Zaidman-Zait et al. (2011) conducted a confirmatory factor analysis, rejected the original PSI factors and identified 5 factors in an exploratory factor analysis.

Across the many studies that have used the PSI to explore parenting stress in samples of children with ASD, the source of the sample, the age range of the sample and sample size have varied widely. Here we extend previous research by exploring the association of disruptive behavior and parenting stress in a large sample of children with ASD who were selected for disruptive behavior. In this well-characterized sample, we examined the severity of disruptive behavior, repetitive behavior and social disability as well as impaired adaptive behavior in order to identify clinical characteristics in children with ASD that uniquely predict parenting stress. The children were participants in one of two federally-funded multisite trials conducted by the Research Units on Pediatric Psychopharmacology (RUPP) Autism Network and Research Units on Behavioral Intervention (RUBI) (Aman et al. 2009; Bearss et al. 2015).

Methods

Participants

The three-site, RUPP trial tested whether combined treatment with risperidone and parent training ($n = 75$) would be superior to risperidone alone ($n = 49$) in children (aged 4–14 years) with ASD diagnosis and serious behavior problems (Aman et al. 2009; Scahill et al. 2009). In the six-site RUBI trial, children (aged 3–7 years) with ASD and disruptive behavior were randomly assigned to parent training ($n = 89$) or parent education ($n = 91$) (Bearss et al. 2015; Scahill et al. 2016). Of the combined total of 304 children (aged 3–14 years), PSI surveys collected at baseline were available for 298 participants. Each study was approved by the institutional review board at each site and written informed consent was obtained from parents or legal guardian prior to data collection.

Procedure

For each trial, an experienced multidisciplinary team conducted a pretreatment diagnostic evaluation and behavioral assessments as well as medical and developmental histories. Following the clinical assessment, ASD diagnoses of autistic disorder, pervasive developmental disorder–not otherwise specified, or Asperger disorder were based on the Diagnostic and Statistical Manual of Mental Disorders-Fourth Edition-Text Revision (American Psychiatric Association 2000) criteria. These diagnoses were supported by the Autism Diagnostic Interview-Revised (Lord et al. 1994). The RUBI trial also used the Autism Diagnostic Observation Schedule (Lord 2000). Intellectual ability was measured on one of three standardized tests: Abbreviated Stanford-Binet Intelligence Scales, fifth edition, the Leiter International Performance Scale, or the Mullen Scale of Early Learning (Mullen 1995; Roid and Miller 1997; Roid 2003).

From the combined dataset of 304 subjects, complete PSI data were available for 298 children (259 males and 39 females) with a mean of 5.8 ± 2.2 years (range 3.1 to 13.8 years). Table 1 presents demographic and clinical characteristics. The alpha coefficients of the PSI subscales were 0.86, 0.81 and 0.79 for the PSI Parental Distress (PSI-PD), PSI- Child Dysfunctional Interaction (PSI-CDI)

Table 1 Demographic and clinical characteristics of children with ASD and disruptive behavior ($N = 298$)

Age (years), mean (SD)	5.8 (2.2)
Gender, *N* (%)	
Male	259 (86.9%)
Female	39 (13.1%)
Race	
Caucasian	252 (84.6%)
African American	33 (11.1%)
Asian	9 (3%)
Other	4 (1.3%)
Hispanic ethnicity	
Yes	37 (12.4%)
No	261 (87.6%)
Educational placement	
Regular class	124 (41.6%)
Special education class	75 (25.2%)
Special education school	71 (23.8%)
Home instruction	14 (4.7%)
No school	14 (4.7%)
IQ	
≥70	199 (67.7%)
<70	95 (32.3%)
Mother age (years) ($N = 297$), mean (SD)	35.6 (6.9)
Mother education	
Eighth grade or less	5 (1.7%)
Some high school	8 (2.7%)
High school graduate or GED	46 (15.4%)
Some college or post-high school or 2 year degree	98 (32.9%)
College graduate	79 (26.5%)
Advanced graduate or professional degree	62 (20.8%)
Father age (years) ($N = 266$), mean (SD)	38.6 (8.0)
Two-parent household	249 (83.6%)
Vineland standard scores, Mean (SD)	
Communication	71.7 (21.3)
Daily living	65.3 (22.7)
Socialization	66 (14.8)
Aberrant behavior checklist	
Irritability	26.1 (7)
Social withdrawal	14.1 (8.6)
Stereotypic behavior	7.3 (5.3)
Hyperactivity/noncompliance	32.6 (9.3)
Inappropriate speech	5.8 (3.4)
ECI/CASI ($N = 296$)	
ODD	12.2 (5.6)
ASD	21.3 (7.1)
ADHD	34.9 (9.3)
CYBOCS-ASD ($N = 297$)	14.0 (3.3)
CGI – Severity	
Moderate (4)	101 (33.9%)
Marked (5)	139 (46.6%)
Severe (6)	57 (19.1%)
Extreme (7)	1 (0.3%)

SD Standard Deviation, *IQ* Intelligence Quotient, *ECI/CASI* Early Childhood Inventory/Child and Adolescent Symptom Inventory, *ODD* Oppositional Defiant Disorder subscale, *ASD* Autism Spectrum Disorder subscale, *ADHD* Attentional Deficit Hyperactivity Disorder subscale, *CYBOCS-ASD* Children's Yale Brown Obsessive-Compulsive Scale-Autism Spectrum Disorder, *CGI* Clinical Global Impression

and Difficult Child (PSI-DC) subscales, respectively. Pearson correlations across the three subscales ranged from 0.40 to 0.47. The total score of the PSI was not used in the analysis.

Inclusion criteria of these trials were as follows: moderate or greater behavioral problems, as measured by the Aberrant Behavior Checklist-Irritability (ABC-I) subscale (RUPP: ABC-I > 18; RUBI: ABC-I > 15); Clinical Global Impression-Severity (CGI-S) score ≥ 4; Intelligence Quotient (IQ) ≥ 35 (or mental age ≥ 18 months). The RUPP trial required subjects to be healthy and medication-free (7–28 days depending on the prior medication). In the RUBI trial, children on stable medication were eligible if there were no planned medication changes during the trial. Both studies allowed entry of children on anticonvulsant medications for seizure disorder if the dose was stable and if the participant was seizure-free for at least 6 months.

Measures

Parenting Stress Index-Short Form (PSI)

The PSI is a 36-item parent self-report designed to measure parental impressions and difficulties about the parenting role (Abidin 1995). The validity and reliability of the PSI have been demonstrated in the general population and a wide range of pediatric clinical populations (Abidin 1995). The PSI consists of three, 12-item subscales: Parental Distress (PD) (burdens and limitations in the parenting role); Parent–Child Dysfunctional Interaction (P-CDI) (level of dissatisfaction in the parent–child relationship); and Difficult Child (DC) (child behaviors and characteristics that are challenging for the parent). The total PSI score is an overall index of parenting stress. For 33 of 36 items, parents are asked to circle *strongly agree*, *agree*, *not sure*, *disagree*, or *strongly disagree*. Three items ask parents to select the most appropriate phrase of five choices. Parental responses are simultaneously transferred to the score sheet rendering a score from 1 to 5 for each item with higher scores reflecting greater stress.

Vineland Adaptive Behavior Scales

The RUPP study used the Vineland Adaptive Behavior Scales by interview (Vineland; Sparrow et al. 1984). The RUBI trial used the parent-rated Vineland Adaptive Behavior Scales-second edition (Vineland-II) (Sparrow et al. 2005). Both versions of the Vineland measure the child's actual performance of adaptive skills in three domains: Communication, Socialization and Daily Living Skills. Items are scored 0 (child does not perform the skill); 1 (child performs the skill sometimes) or 2 (child regularly

performs the skill). Raw score are converted to standard scores (mean 100; SD 15) with higher scores indicative of greater adaptive functioning.

Aberrant Behavior Checklist (ABC)

The ABC is a 58-item, parent-rated scale comprising five subscales: Irritability (I) (includes tantrums, aggression, and self-injurious behaviors, 15 items); Social Withdrawal (SW) (includes response to others, initiation of interaction, 16 items); Stereotypic Behavior (S) (includes mannerisms and repetitive movements, 7 items); Hyperactivity/Noncompliance (H) (includes hyperactivity and noncompliance, 16 items); and Inappropriate Speech (IS) (repetitive vocalizations, 4 items) (Aman et al. 1985; Aman and Singh 2017). Each item is rated 0 to 3 with higher scores indicating greater severity. Normative data in children with autism spectrum disorder offer guidance on interpretation of scores on the ABC (Kaat et al. 2014).

Clinical Global Impression-Severity (CGI-S)

The CGI-S is a clinician rating of current illness severity based on all available information including parent reports, clinician interviews, and clinical observation (Guy 1976). The seven-point scale ranges from 1 (normal, not at all ill) to 7 (among the most extremely ill patients).

Early Childhood Inventory (ECI)/ Child and Adolescent Symptom Inventory (CASI)

The Early Childhood Inventory (ECI) and the Child and Adolescent Symptom Inventory (CASI) are DSM-IV-referenced parent-rated scales (American Psychiatric Association 2000; Gadow and Sprafkin 2000; Gadow and Sprafkin 2005). The ECI and CASI cover a full range of psychiatric disorders with each disorder treated as a separate subscale. Here we selected the Attention Deficit/Hyperactivity Disorder (ADHD; 18 items), Oppositional Defiant Disorder (ODD; 8 items) subscales as measures of disruptive behavior, and the ASD subscale (12 items) as a measure of ASD severity. Items are rated from 0 (never) to 3 (very often). Each subscale is the sum of the items in that subscale with higher scores reflecting greater symptom severity.

Children's Yale Brown Obsessive-Compulsive Scale-Modified for Autism Spectrum Disorder (CYBOCS-ASD)

The Children's Yale-Brown Obsessive-Compulsive Scale-Modified for Autism Spectrum Disorder (CYBOCS-ASD) is a clinician-rated interview designed to evaluate the severity of repetitive behavior in children with ASD (Scahill et al. 2006). This modified version was derived from the CYBOCS, which was developed to assess the symptom severity in children with obsessive-compulsive disorder (Scahill et al. 1997). Because of commonly observed language limitations in children with ASD, the CYBOCS-ASD only includes the five compulsion items: time spent, interference, distress, resistance to repetitive behavior, and control of repetitive behavior. Each item is rated from 0 (none) through 4 (extreme), and scores can range from 0 to 20.

Data Analyses

Demographic and clinical measures were calculated as means with standard deviations, medians with interquartile ranges (IQR), or percentages as appropriate. Findings presented by Zaidman-Zait et al. (2011) raised questions about the fit of the existing three- factor structure of the PSI in children with ASD. Thus, we conducted a confirmatory factor analysis (CFA) using MPlus v7.2 (Los Angeles, CA) to evaluate the fit of three PSI factors (PD, P-CDI, DC).

We used weighted root mean square residual (WRMR) given the ordinal format of the PSI. To assess model fit, root mean square error of approximation (RMSEA), comparative fit index (CFI) and Tucker-Lewis index (TLI). Guidelines to evaluate whether a given model provided a good approximation to the data included: RMSEA <0.1; CFI and TLI greater than 0.9 (Browne and Cudeck, 1992; Hu and Bentler, 1999). Results of the CFA for the original three-factor solution were: (RMSEA = 0.078; 90% CI: 0.074–0.082; TLI = 0.815; CFI = 0.827) (Browne and Cudeck 1992; Hu and Bentler 1999). Although not all fit indices met the convetional benchmark, we proceeded with the original PSI factors (PD, P-CDI, DC) in all subsequent analyses to facilitate comparisons with other studies.

Second, children were classified above or below the median (50^{th} percentile) on measures of disruptive behaviors, repetitive behaviors and social disability. On the Vineland children ≤50th percentile (lower adaptive functioning) were compared to children >50th percentile. *T*-tests, adjusted for multiple comparisons with the Benjamini-Hochberg method, were used to examine differences on PSI subscale scores across these dichotomized child groups.

Third, to identify associations between the more impaired group of children (i.e., ≥50th percentile on behavioral problems and ≤50th percentile on the Vineland) and parents with the highest stress levels, we performed a series of bivariate and multivariate logistic regression models. For these categorical analyses, the highest quartile (≥75th percentile) on PSI subscales was used to classify parents in the "high stress" group (parents <75th percentile on each PSI subscale was used as the reference group). All logistic

regression analyses were adjusted for age of child (≥6 years versus <6 years). Children with ≥median scores on behavioral measures and ≤median on the Vineland were regressed to evaluate the association with parents in the "high stress" group (≥75th percentile) on each PSI subscale. Model results are presented as odds ratios (OR) with 95% confidence intervals (CI). Multivariate logistic models were constructed using forward selection guided by the strength of the statistical associations in the bivariate calculations. To test the significance of each added variable, we used likelihood ratio statistics (see Supplemental Table 1 for details). Inferential analyses and statistical modeling were performed using SAS v9.4 (Cary, NC).

Results

Parental Stress in Children with Higher versus Lower Behavioral Problems

Table 2 shows the mean scores on PSI subscales after dividing the child sample at the median on parent-rated ABC, pre-selected ECI/CASI subscales, and the CYBOCS-ASD (p-values were adjusted for multiple comparisons). Parents reported slightly higher scores on the PSI-PD subscale for children ≥50th percentile on the ABC-SW subscale ($p = .020$). In this sample of children with disruptive behavior, there were no differences in PSI-PD subscale scores on measures of disruptive behavior. On the PSI-CDI and PSI-DC subscales, however, several differences emerged. Although children enrolled in these studies were selected for disruptive behavior, the observed trend in Table 2 is clear. Parents who rated children ≥50th percentile on measures of disruptive behavior had significantly higher scores on the self-reported PSI-CDI and PSI-DC subscales. In addition, parents who rated children ≥50th percentile on measures reflecting social disability (e.g., ABC-SW and ECI/CASI ASD subscales) also had significantly higher scores on the self-reported PSI-CDI and PSI-DC subscales.

Associations between Behavioral Measures and Greater Parental Stress

PSI subscale scores ≥75th percentile were used to classify parents in the "high stress" group. In the bivariate logistic regression analyses, predictor variables for parents in the "high stress" group included children ≥50th percentile on behavioral problems and ≤50th percentile on Vineland domain scores (see Table 3). For example, children ≥50th percentile on the ABC-SW subscale, a measure of social disability, were twice as likely to have parents in the upper quartile on the PSI-PD subscale as children below the median. High scores on the ABC SW subscale were also

Table 2 Mean PSI subscale scores for children classified above or below the median on ratings of child behavior

Child measure	<50th percentile on child rating Mean (SD)	≥50th percentile on child rating Mean (SD)	P-value[a]
Parental Distress			
ABC-I	32.0 (8.6)	33.8 (9.5)	.330
ABC-SW	31.2 (8.3)	34.5 (9.5)	.020
ABC-S	32.1 (8.3)	33.4 (9.7)	.330
ABC-H	31.6 (9.0)	34.0 (9.1)	.161
ABC-IS	32.3 (9.0)	33.4 (9.2)	.330
ECI/CASI ODD	31.8 (9.1)	33.8 (9.1)	.330
ECI/CASI ASD	32.3 (8.9)	33.4 (9.3)	.330
ECI/CASI ADHD	31.5 (8.6)	34.2 (9.4)	.108
CYBOCS-ASD	32.1 (8.2)	33.3 (9.6)	.330
Parent–Child Dysfunctional Interaction			
ABC-I	28.4 (7.3)	33.0 (7.7)	<.001
ABC-SW	27.9 (7.2)	33.4 (7.5)	<.001
ABC-S	28.9 (8.4)	32.1 (7.2)	<.001
ABC-H	29.0 (7.9)	32.3 (7.5)	<.001
ABC-IS	29.7 (7.6)	31.7 (8.0)	.054
ECI/CASI ODD	29.0 (7.3)	32.3 (8.0)	<.001
ECI/CASI ASD	29.0 (7.6)	32.5 (7.7)	<.001
ECI/CASI ADHD	29.3 (8.1)	32.2 (7.4)	.006
CYBOCS-ASD	29.6 (7.5)	31.4 (8.0)	.060
Difficult Child			
ABC-I	41.5 (6.9)	46.2 (5.9)	<.001
ABC-SW	42.9 (7.2)	44.7 (6.5)	.046
ABC-S	43.3 (7.4)	44.3 (6.4)	.221
ABC-H	42.5 (7.1)	45.1 (6.4)	.006
ABC-IS	42.7 (7.7)	44.8 (6.0)	.033
ECI/CASI ODD	41.6 (7.2)	45.8 (5.9)	<.001
ECI/CASI ASD	42.7 (6.8)	44.9 (6.7)	.024
ECI/CASI ADHD	42.2 (6.9)	45.4 (6.5)	<.001
CYBOCS-ASD	42.4 (6.3)	44.7 (7.0)	.024

SD standard deviation, *ABC-I* Aberrant Behavior Checklist-Irritability subscale, *ABC-SW* Aberrant Behavior Checklist-Social Withdrawal subscale, *ABC-S* Aberrant Behavior Checklist-Stereotypy subscale, *ABC-H* Aberrant Behavior Checklist-Hyperactivity subscale, *ABC-IS* Aberrant Behavior Checklist-Inappropriate speech subscale, *ECI/CASI ODD* Early Childhood Inventory/Child and Adolescent Symptom Oppositional defiant disorder subscale, *ECI/CASI ASD* Early Childhood Inventory/Child and Adolescent Symptom Autism Spectrum Disorder subscale, *ECI/CASI ADHD* Early Childhood Inventory/Child and Adolescent Symptom Attention Deficit Hyperactivity Disorder subscale, *CYBOCS-ASD* Children's Yale Brown Obsessive-Compulsive Scale-modified for Autism Spectrum Disorder

[a]*P*-values presented are adjusted using the Benjamini-Hochberg method

Table 3 Odds ratios for child variables predicting parental membership in the highest quartile of PSI subscale scores

Characteristic	Bivariate		Multivariate	
	≥75th Percentile OR (95% CI)	*P*-value	≥75th Percentile OR (95% CI)	*P*-value
Parental Distress				
ABC Social Withdrawal ≥50th percentile	2.15 (1.26–3.66)	.005	2.0 (1.16–3.41)	.013
ABC Stereotypy ≥50th percentile	1.72 (1.01–2.93)	.048		
ECI/CASI ADHD ≥50th percentile	1.88 (1.11–3.17)	.019	1.7 (1.02–2.95)	.043
Parent–Child Dysfunctional Interaction				
Vineland Communications ≤50th percentile	2.33 (1.32–4.13)	.004		
Vineland Daily Living ≤50th percentile	3.25 (1.79–5.88)	<.001	2.6 (1.34–4.87)	.004
Vineland Socialization ≤50th percentile	3.00 (1.68–5.37)	<.001		
ABC Irritability ≥50th percentile	3.17 (1.81–5.57)	<.001		
ABC Social Withdrawal ≥50th percentile	3.73 (2.09–6.66)	<.001	3.4 (1.82–6.32)	<.001
ECI/CASI ODD ≥50th percentile	2.90 (1.64–5.11)	<.001	3.4 (1.85–6.20)	<.001
ECI/CASI ASD ≥50th percentile	1.89 (1.10–3.22)	.020		
ECI/CASI ADHD ≥50th percentile	1.89 (1.11–3.23)	.020		
Difficult Child				
Vineland Communications ≤50th percentile	1.90 (1.08–3.33)	.026		
Vineland Daily Living ≤50th percentile	2.25 (1.27–4.00)	.006		
ABC Irritability ≥50th percentile	2.96 (1.69–5.19)	<.001	2.1 (1.15–3.91)	.016
ECI/CASI ODD ≥50th percentile	2.18 (1.26–3.80)	.006		
ECI/CASI ADHD ≥50th percentile	2.35 (1.36–4.07)	.002	1.8 (1.02–3.24)	.043
CYBOCS-ASD ≥50th percentile	2.69 (1.46–4.97)	.002	2.1 (1.09–4.12)	.027

All estimates adjusted for patient age (≥ 6 years versus <6 years)

ABC Aberrant Behavior Checklist, *HSQ* Home Situation Questionnaire, *ECI/CASI* Early Childhood Inventory/Child and Adolescent Symptom Inventory, *CYBOCS-ASD* Children's Yale Brown Obsessive-Compulsive Scale-ASD

associated with parents in the high stress group on the PSI-CDI subscale. The level of child disruptive behavior was associated with higher parental stress on the PSI-CDI and PSI-DC subscales. Children ≥50th percentile on disruptive behavior measures such as the ABC-I and ECI/CASI ODD conferred a two to three- fold increase in the likelihood of parents classified in the high stress group. By contrast, child disruptive behavior was not strongly associated with high scores on the PSI-PD subscale. Children ≤the 50th percentile on all Vineland standard scores were also significantly associated with parental membership in the highest quartile on the P-CDI subscale (odds ratios across Vineland domains ranged from 2.3 to 3.25). The highest quartile on the PSI-DC subscale was significantly associated with children ≤the median on Vineland Communication and Daily Living domain.

Because the measures of child behavior may be intercorrelated, we used the same dichotomized groups on child measures and PSI subscales (highest quartile) in multivariate logistic models. Table 3 also includes the results of forward selection logistic regression (see Table 1 in Supplemental material for details) used to identify the child measures that uniquely contributed to the association with parental membership in the highest quartile on the PSI subscales. After adjusting for other significant bivariate associations, children with greater severity of social disability (ABC-SW), and ECI/CASI ADHD symptoms significantly predicted parents being in the high stress group on the PSI-PD (OR 1.7–1.9). Higher severity of social disability (ABC-SW) and noncompliance (ECI/CASI-ODD), and lower Vineland Daily Living Skills, were significantly associated with parents being in the highest quartile on the PSI-CDI subscale (OR 2.5–3.4). Finally, a model that included children ≥ 50th percentile on the ABC-I, ECI/CASI-ADHD, and CYBOCS-ASD scores was predictive of parental classification in the upper quartile on the PSI-DC subscale (OR 1.8 to 2.1).

Discussion

This study examined pre-treatment data in a sample of 298 children (age 3–14 years) with ASD accompanied by moderate to severe behavioral problems. To our knowledge, this is the largest sample of children with ASD selected for serious behavioral problems to explore the associations of child clinical characteristics and parenting stress as measured on the PSI. As a prerequisite, we conducted a

confirmatory factor analysis that generally supported the original three-factor structure of the PSI. This observation contrasts with the report by Zaidman-Zait et al. (2011) - perhaps due to differences in sample selection. The Zaidman-Zait et al. (2011) sample was younger (between 20 and 72 months of age) and participants were not selected for disruptive behavioral problems.

Two sample *t-tests*, with adjustment for multiple comparisons, compared children rated at or above the median on measures of disruptive behavior, repetitive behavior and social disability to children below the median on these measures. On the PSI-CDI and the PSI-DC subscales (but not the PSI-PD subscale), parent-reported stress was significantly higher in the more impaired child group on nearly all measures (Table 2). Differences were especially prominent on measures of disruptive behavior. These findings are consistent with prior reports showing that disruptive behavior in children with ASD may contribute to higher levels of parental stress (Hall and Graff 2012; Huang et al. 2013; Lecavalier et al. 2006; Zaidman-Zait et al. 2014; Zaidman-Zait et al. 2017). Our results extend the findings of prior studies by showing that parenting stress is greater in children with more severe disruptive behavior.

As noted, on the PSI-PD subscale (a measure of parental distress) in our sample, there were no group differences on ratings of disruptive behavior. Indeed, the only PSI measure that was significantly higher in the children ≥50th percentile across all PSI subscales was the ABC-SW subscale. This finding on the impact of social disability as measured on the ABC-SW subscale on parenting stress awaits replication.

To examine associations between children with greater impairment and parents classified in the high stress group on PSI subscales (≥75th percentile), we conducted a series of logistic regression analyses. Bivariate models showed that children in the more impaired group on several available measures were significantly associated with parental membership in the high stress group (Table 3). When the variables showing a significant association were included in a forward selection multivariate logistic regression model, a parsimonious model emerged for each PSI subscale. Children with more severe social disability and ADHD symptoms each contributed to parental classification in the highest quartile on the PSI-PD subscale. On the PSI-CDI subscale, model fitting showed that higher social disability and lower scores on Vineland Daily Living domain contributed to parental classification in the highest quartile. The group of children at or above the median on measures of disruptive behavior and the CYBOCS-ASD (which includes child's over-reaction to interruption of repetitive behavior) predicted classification of parents in the high stress group on the PSI-DC subscale.

Collectively, these findings suggest that, in addition to disruptive behavior, greater impairment in social disability and adaptive functioning also contribute significantly to parenting stress in children with ASD. The strong association between the PSI-CDI and children ≥50th percentile on the ABC-SW subscale, the ECI/CASI ODD subscale and <50th percentile on the Vineland Daily Living domain may be partially explained by the items on this PSI subscale: "My child smiles at me much less than I expected;" "My child is not able to do as much as I expected;" "When playing, my child doesn't often giggle or laugh;" "I get the feeling that my efforts are not appreciated very much." Endorsement of these items may reflect parental a disparity between expected and actual quality of parent-child interaction in children with ASD and serious behavioral problems.

Limitations

First, all child and parent measures were completed by a single informant (i.e., the child's parent). High levels of parenting stress may affect parent ratings of child behavior. Second, this was a sample of convenience that included children with ASD and serious behavioral problems. Therefore, our results may not generalize to the wider population of children with ASD. Third, the age range of the children in this sample (3–14 years) was somewhat broad. We note, however, that statistical analyses were adjusted for age. Finally, this was a cross-sectional analysis, the observed associations do not prove causation. As observed in several previous reports, we showed that the level of disruptive behavior in children with ASD is associated with high levels of parenting stress. Less well established is the observed association of social disability in the child and parenting stress. Future studies in a larger sample of children with a wider sampling frame could confirm this association.

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Compliance with Ethical Standards

Conflict of Interest Over the past two years, Dr. Scahill has served as a consultant to Roche, Shire, Suupernus, Neurocrine, Janssen, Yamo, and the Tourette Association of America; royalties from Guilford, Oxford, American Psychological Association. Dr. Aman has received research contracts, consulted with, served on advisory boards, or done investigator training for Aevi Genomic Medicine; Bracket Global; Hoffman-La Roche; MedAvante, Inc.; MedAvante-Prophase; Ovid Therapeutics; and Zynerba Pharmaceuticals. He receives royalties

from Slosson Educational Publications received research. The remaining authors declare that they have no conflict of interest.

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