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How Are Child Restricted and Repetitive Behaviors Associated with Caregiver Stress Over Time? A Parallel Process Multilevel Growth Model

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Abstract The impact of raising a child with autism spectrum disorder (ASD) is frequently accompanied by elevated caregiver stress. Examining the variables that predict these elevated rates will help us understand how caregiver stress is impacted by and impacts child behaviors. This study explored how restricted and repetitive behaviors (RRBs) contributed concurrently and longitudinally to caregiver stress in a large sample of preschoolers with ASD using parallel process multilevel growth models. Results indicated that initial rates of and change in RRBs predicted fluctuations in caregiver stress over time. When caregivers reported increased child RRBs, this was mirrored by increases in caregiver stress. Our data support the importance of targeted treatments for RRBs as change in this domain may lead to improvements in caregiver wellbeing.

Keywords Autism · Caregiver stress · Restricted and repetitive behaviors

Introduction

The impact of raising a child with autism spectrum disorder (ASD) is frequently accompanied by elevated rates of caregiver stress and decreased wellbeing (Baker-Ericzn et al. 2005; Kasari and Sigman 1997; Osborne and Reed 2009; Schieve et al. 2007). Caregivers assume a number of roles when caring for their child with ASD, such as advocating for effective services and frequently participating in time-consuming treatment plans. In fact, caregiver variables, such as stress, are known to affect treatment efficacy (Osborne et al. 2008). Examining variables that contribute to caregiver stress will help us further understand how stress is impacted by and impacts child behavior. The aim of this study was to explore how the core deficit of restricted and repetitive behaviors in preschoolers with ASD contributed concurrently and longitudinally to caregiver stress using parallel process multilevel growth models.

Restricted and Repetitive Behaviors in ASD

Restricted and repetitive behaviors (RRBs) occur within a number of neurodevelopmental disorders; however, are considered a hallmark feature of ASD. RRBs characterize a wide range of idiosyncratic behaviors including stereotyped movements, repetitive self-injury, compulsive/ritualistic behaviors, insistence on sameness, repetitive language, and unusual and/or intense preoccupations (Turner, 1999).

These behaviors often dominate the daily activities of children with ASD. Furthermore, RRBs have been found to interfere with opportunities for the child to engage socially (Lee et al. 2007; Loftin et al. 2008), develop adaptive skills (Cuccaro et al. 2003; Dunlap et al. 1983) as well as appropriately learn and explore (Dunlap et al. 1983; Lewis et al. 2007; Pierce and Courchesne 2001; Tanimura et al. 2008). Caregivers frequently rate these behaviors as more difficult to manage than social-communication deficits (Bishop et al. 2007, Lecavalier et al. 2006). Further, RRBs

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are perceived as stigmatizing for both children and families (Gabriels et al. 2005; Nadig et al. 2010), and can lead to negative caregiving styles (Durand and Carr 1987; Greenberg et al. 2004). Children with ASD often become agitated, irritable, disruptive and even aggressive or self-injurious when RRBs are disrupted (Mercier et al. 2000). Thus it is unsurprising that RRBs have been hypothesized to influence family quality of life and serve as a significant driver of caregiver stress (Gabriels et al. 2005; Lecavalier et al. 2006; Lounds et al. 2007; Mercier et al. 2000).

Parenting Stress in ASD

While parenting any child can at times be stressful and challenging (Crnic and Greenberg 1990), researchers have consistently found elevated rates of stress in caregivers of children with ASD when compared to both caregivers of typically developing children and children with other developmental disorders (see Hayes and Watson 2015, for a recent review and meta-analysis). Research appears to indicate that there is something unique about the ASD phenotype, which contributes to the higher rates of caregiver stress. Researchers have begun to unpack the behavioral profile of ASD in order to identify specific child characteristics that may explain higher rates of parent stress.

To date, a wide range of child behaviors have been found to correlate with caregiver rated stress, including ASD severity (Hastings and Johnson 2001), social communication deficits (Davis and Carter 2008), executive functioning (Epstein et al. 2008), and sensory behaviors (Ben-Sasson et al. 2013; Epstein et al. 2008). A recent meta-analysis (Hayes and Watson 2015) found that core deficits of ASD (i.e., social-communication impairments and RRBs) contributed to caregiver stress above other variables (such as developmental functioning); however, very few studies have explored the longitudinal contributions of child characteristics to caregiver stress.

One of the most consistent predictors of caregiver stress to date has been child behavior problems—a variable while frequently reported at elevated rates in ASD (Beck et al. 2004; Lecavalier et al. 2006; McStay et al. 2014a, b; Orsmond et al. 2003) is not a core deficit of ASD or syndrome specific. Concurrent and predictive relationships have been found between child behavior problems and stress in a number of studies across a wide range of ages (Beck et al. 2004; Estes et al. 2013b; Falk et al. 2014; Hastings 2003; Orsmond et al. 2003), and these associations often are stronger than those found for the core ASD deficits of social-communication and RRBs (Davis and Carter 2008). While some researchers also have found a longitudinal relationship between child behavior problems and caregiver stress (Lecavalier et al. 2006), this has not been a consistent finding (Totsika et al. 2013). One issue is overlap with child RRBs—behaviors that fall under the umbrella of RRBs are frequently included on measures of problem behavior. For example, the Aberrant Behavior Checklist (Aman and Singh 1986) includes items such as motor stereotypy, self-injury, rituals and over-sensitivity. In this context it becomes important to understand the potentially unique contributions of the RRBs of children with ASD to caregiver stress.

Both Gabriels et al. (2005) and Bishop et al. (2007) found that caregiver reported RRBs were strongly associated with caregiver stress. In a large sample ($n = 110$) of caregivers of a child with ASD (mean age 9 years), Bishop and colleagues found that child RRBs accounted for unique variance in caregiver perceived negative impact. In this study, social-communication impairments did not uniquely account for any of the variance in caregiver perceived negative impact. In a smaller sample of children separated by high and low IQ, Gabriels et al. (2005) reported strong correlations between child RRBs and ratings of caregiver stress. Together these studies suggest that the core deficit of RRBs may directly affect caregiver stress; yet, we do not know if this relationship persists over time.

Aims and Hypotheses

Given that RRBs are frequently reported as difficult to manage by caregivers (Bishop et al. 2007; Lecavalier et al. 2007), interfere with other areas of child development (Cuccaro et al. 2003; Lee et al. 2007; Tanimura et al. 2008) and continue to persist overtime (Harrop et al. 2014; Honey et al. 2008; Joseph et al. 2013; Moore and Goodson 2003; Richler et al. 2010), the aim of this study was to examine how this core deficit contributed to caregiver stress over time in preschoolers with ASD. To date, studies examining the relationship between child RRBs and caregiver stress have been cross-sectional, and in general, few longitudinal examinations of the relationship between parent stress and child ASD symptoms have been conducted (Lecavalier et al. 2006; Osborne and Reed 2009). Additionally, the handful of longitudinal studies on RRBs during the toddler and preschool period have included relatively small samples, but indicated slight increases in these behaviors over time (Harrop et al. 2014; Honey et al. 2008). Therefore, a further aim was to examine developmental trajectories of RRBs in a large sample of preschoolers with ASD. Examining the extent to which change in children's RRBs are associated with and predicts fluctuations in caregiver stress will allow us to better understand the relationship between core features of ASD and caregiver wellbeing. Based on previous research, we predicted the following:

1. Caregiver report of child RRBs will not change significantly over the course of a short-term longitudinal study (i.e., 1½ years).
2. Caregiver reported stress will associate concurrently with child RRBs, with higher rates of RRBs correlating with higher rates of caregiver stress.
3. Change in caregiver stress over time will be predicted by change in child RRBs, after controlling for parent socioeconomic status, ethnicity, and sex as well as child sex, overall ASD severity and developmental functioning.

Methods

Data for this study were drawn from a larger study comparing the efficacy of school-based comprehensive treatment models (CTMs) for preschool-aged (3–5 years) children with ASD (Boyd et al. 2014). Preschool classrooms across four states (North Carolina, Colorado, Florida and Minnesota) were recruited into the larger study. Caregivers in this study completed various rating scales and questionnaires about themselves and their children across three time points over the course of 12–18 months (referred to hereafter as T0, T1 and T2 for data analysis purposes—see *time points* for further information).

Participants

Eligibility criteria for the study included being a caregiver of a child with a clinical diagnosis or educational label of ASD, aged between 3 and 5 years. Child community diagnosis was verified through administration of the Autism Diagnostic Observation Schedule (ADOS-G; Lord et al. 2000) by a research-reliable administrator. Caregivers had to be proficient in English in order to complete caregiver rating scales and questionnaires. A total of 198 caregivers were recruited into the study at T0 (i.e., pretest) with 184 providing complete data on both the Repetitive Behavior Scales-Revised (Bodfish et al. 1999) and the Parenting Stress Index-Short Form (Abidin 1995). A total of 181 caregivers completed T1 (i.e., posttest) assessments. Fewer caregivers (n = 91) participated at T2 (i.e., follow-up) because the duration of the project (4 years) precluded the collection of follow up data on all study participants. Thus as planned, child and caregiver data were only collected from families in the North Carolina and Miami sites at T2. We did examine if there were differences on parent or child demographic variables between families who participated at T2 versus those who did not. We found no significant differences for caregiver socioeconomic status between those that did ($M = 9.64$) and did not ($M = 9.20$) contribute T2 data [$t(189) = -1.17$, $p = 0.22$]; in addition, there were no differences for children's Mullen standard scores between those that did ($M = 61.69$) and did not ($M = 66.92$) contribute T2 data [Welch's $t(177.22) = 1.88$, $p = 0.061$]. A series of Fisher's exact tests also indicated that the T2 sample did not significantly differ from the participants not assessed at T2 on race ($p = 0.118$), sex ($p = 0.174$), or ethnicity ($p = 0.357$).

Female caregivers made up the vast majority of the sample (86 %). Most primary caregivers also self-identified as White, non-Hispanic (47 %) in comparison to Hispanic (36 %), Black (12 %) or Asian (5 %), and the vast majority of caregivers were the child's biological parents (94.95 %). Socioeconomic status (SES) was defined as a composite of the highest educational level reached across caregivers and their combined family income since these two variables were moderately correlated ($r = 0.53$, $p < 0.0001$). Income was measured on a six-point scale representing a range of income levels and education was measured on a five-point scale. Thus, these values were combined to yield an SES indicator that could range from two (lowest income and education level) to eleven (highest income and education level). The mean SES composite score for the sample was 9.40 (SD = 2.62), indicating that this sample was relatively high in SES. The mean age of the child participants at study enrollment was 48.32 months (SD = 7.64) and their Mullen standard score at T0 averaged 64.39 (SD = 19.30). Caregivers were paid a total of $250 for study participation, and this amount was equally distributed across the three time points. Participant characteristics are reported in Table 1.

Measures

Child RRB data was gathered using the Repetitive Behavior Scale-Revised (RBS-R; Bodfish et al. 1999). Caregiver stress data was obtained through the Parent Stress Index-Short Form (PSI-SF; Abidin 1995).

Repetitive Behavior Scales-Revised (RBS-R)

The RBS-R (Bodfish et al. 1999) is an informant-based rating scale of restricted and repetitive behavior that rates the occurrence of a behavior on a 4 point-likert scale from (0) does not occur to (3) occurs frequently and/or is severe. The RBS-R is composed of 43 items from six subscales that measure a variety of RRBs in ASD. Items from the Stereotypical Behavior subscale measure body movements and actions that are repeated in the same manner and serve no apparent purpose. The Self-Injurious Behavior subscale captures movements or actions that have the potential to cause injury to the child and are repetitious in nature. Items from the Compulsive Behavior subscale measure behaviors

Table 1 Participant characteristics at T0

Child sex (boys:girls)	165:33
T0 Chronological age (months)	48.32 (7.64)
T0 Mullen standard score	64.39 (19.30)
T0 ADOS severity score (range 1–10)	7.22 (1.64)
Child race	
White	155
Black	24
Asian	10
Multi-Racial	8
Other	1
Child ethnicity	
Hispanic	69
Non-Hispanic	128
Missing	1
Caregiver sex (female:male)	171:23 (4 missing)
SES composite (education and income)	9.40 (2.62)
Caregiver race	
White	150
Black	25
Asian	10
Multi-Racial	6
Missing	7
Caregiver type	
Biological	188
Adoptive	3
Maternal grandparent	2
Missing	5

that the child repeats or performs in a "just so" way. The Ritualistic Behavior subscale asks informants to rate whether their child performs activities of daily living in a similar manner, such as a preference for particular routines. The Sameness Behavior subscale includes items that capture a resistance to change or insistence that things stay the same. The final subscale, Restricted Behaviors, includes items relating to a child's limited range of focus, interests or activities. The RBS-R generates six subscale scores, which were summed to create a total sum score for analysis. The RBS-R has a test–retest reliability ranging from 0.52 to 0.96. The same caregiver was asked to rate the child's RRBs at all three time points.

Parenting Stress Index: Short Form (PSI-SF)

The PSI-SF is a caregiver report measure derived from the Parenting Stress Index (Abidin 1995). The PSI includes 101 items across 13 subscales. The PSI-SF consists of 36 items derived from the PSI. The PSI-SF has been widely used in ASD research (Baker-Ericzn et al. 2005; Beck et al. 2004; Lecavalier et al. 2006). The PSI-SF consists of three subscales each containing 12 items; Parenting Distress (PD), Parent–Child Dysfunctional Interactions (PCDI), and Difficult Child (DC). Caregivers rate each item on a 5-point scale ranging from (1) strongly disagree to (5) strongly agree. Items on the PD subscale are intended to assess distress related to personal factors that influence parenting, such as lack of social support. Examples include "problems with spouse" and "cannot do things I like since having this child." The PCDI subscale is intended to examine how the caregiver perceives interactions with their child and whether these interactions are positive or negative. An example of the PCDI scale is "my child rarely does things for me that make me feel good." The DC subscale measures child characteristics that make him/her easy or difficult to manage with a focus on compliance, defiance, demanding behaviors and temperament. However, the subscale does not include items that would be considered RRBs. An example from the DC scale is "getting child to do something is hard." The PSI-SF total score reflects the caregiver's overall experience of parenting stress. A score in the 75th percentile is considered a "clinically significant" level of parenting stress. The PSI-SF reports internal reliability coefficients of 0.80-0.87 for the three subscales and test–retest reliability of 0.76 (Abidin 1995).

Time Points

Data were collected across three time points—T0, T1 and T2 (see Boyd et al. 2014 for further details). T0 occurred within the first three months of the child's school year with all children and their caregivers enrolled into the study at T0 (n = 198). T1 occurred at least six months and no more than nine months after T0 (M = 191 days, SD = 31.8). T2—the final time point—occurred a minimum of six months but no more than nine months after T1 (M = 231 days, SD = 32.6). As previously stated, the design of the study precluded the collection of follow up data on all study participants, as such, only 91 participants completed T2 assessments.

Data Analysis

Descriptive statistics were calculated for child RRBs and caregiver stress at each time point (T0, T1 and T2). Data were not separated by CTM due to the lack of group differences in the main study (Boyd et al. 2014). Cross sectional correlations were run between RBS-R total scores and PSI-SF total scores and subscale scores. Data were then analyzed via a two-stage parallel-process multilevel growth modeling approach.

The general purpose of a parallel-process growth model is to understand how change over time on one variable is related to change over time on another. In this case, we sought to understand how severity and rate of change of RRBs for children in treatment is related to the severity and rate of change of caregiver stress. While the analysis is complex, the underlying idea is relatively simple. In the first step, we fit a model to the RBS-R data that allowed us to estimate a unique initial severity and rate of change for each child. Next, we fit a model to the PSI-SF data, allowing us to use RBS-R initial severity and rate of change from the first model as predictors of initial parent stress as well as the rate of change for caregiver stress over time.

In the first step, an unconditional linear growth model for the RBS-R total score was fit to the data to explore change in RRBs over the course of the study. This model, and all subsequent models, included random effects for the intercept and slope parameters, with the intercept-slope covariance term freely estimated. The Empirical Bayes (EB) residuals from this model were saved for analysis in the following step. These residuals represent precision-weighted estimates of each individual's initial score on the RBS-R total score (the intercept) as well as rate of change over time (the slope). These estimates are grand-mean centered. Therefore a participant with a positive intercept EB estimate has a higher than average mean score on the outcome at time zero. A participant with a positive slope EB estimate has a rate of change on the outcome variable that is higher than the sample mean. Conversely, a negative EB slope indicates a rate of change less than the sample mean.

The EB estimates saved in step one were used as predictors in step two. In step two, growth models were fit to the caregiver-reported PSI data (total score, difficult child, parent–child dysfunctional interaction and parent distress subscales). The focus of these models was to understand the extent to which initial RBS-R scores and trajectories are related to baseline (T0) caregiver stress and change over time. Growth models allow T0 caregiver stress scores to be influenced by T0 child RBS-R scores as well as change over time in this variable.

Specific covariates were added to all models based on associations with caregiver stress in previous cross-sectional studies. Both family SES and caregiver sex have been found to be associated with caregiver stress in parents of a child with ASD (Baker-Ericzn et al. 2005; Dabrowska and Pisula 2010; Jones et al. 2013; McStay et al. 2014a, b). We also wanted to explore how child RRBs contributed to caregiver stress over and above the contribution of child developmental ability as this variable is related to both caregiver stress (Bebko et al. 1987) and child RRBs (Gabriels et al. 2005; Harrop et al. 2014). ASD severity (as indexed through ADOS-G Calibrated Severity Scores) was included as a covariate to examine if RRBs contributed above and beyond overall symptom severity. Finally, we added caregiver ethnicity to the analytic models as this demographic factor has been associated with family stress in one study (Bishop et al. 2007).

Due to the study's time constraints, the research plan purposefully involved follow-up data collection from only half the participants. Therefore, missingness was determined by the research team rather than by the participant and was considered "ignorable", meaning that the probability of missingness was not driven by the unobserved missing values themselves. Under ignorable missingness (MCAR or MAR), the multilevel model for change over time produces unbiased estimates of effects by allowing each subject's influence on the model estimates to vary in proportion to the amount of data they contribute (Singer and Willett 2003). So subjects measured at all three time points contribute more information to the analysis than those measured only twice, but all the data are used. Layered over the missingness by design was infrequent missingness on specific predictor variables which caused cases to be listwise deleted from the dataset prior to analysis. For instance, the T0 RBS-R sum scores could be computed for 193 of 198 participants. The small proportion of missingness due to missing predictors was approximately 10 % (178 of 198 with complete PSI-SF data at T0) and did not justify the use of multiple imputation or other techniques, which could have at maximum only addressed 75 % of the missing data due to the two-step process for fitting the parallel process model.

Results

Restricted and Repetitive Behaviors

RBS-R scores decreased over the duration of the study (Fig. 1). Using a growth model, time was a significant predictor of change in RBS-R scores ($B = -1.24$,

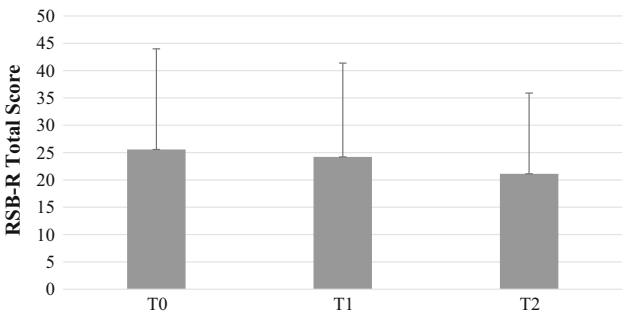

Fig. 1 RBS-R total sum score across timepoints

$p = 0.04$) with an average reduction of about 2.5 points over the course of the study.

Caregiver Stress

Caregiver stress also showed some reduction over the course of the study (Fig. 2; Table 3). PSI total scores decreased significantly over the course of the study ($B = -2.64$, $p = 0.002$), with PSI total scores reducing by around 5.3 points from T0 to T2. All PSI subscales showed some decrease by T2, with reductions ranging between 1.4 and 2.2 points (Fig. 2; Table 3).

Associations Between Child RRBs and Caregiver Stress: Cross-Sectional and Longitudinal

RBS-R mean scores were significantly correlated with PSI Total and Subscale scores at T0 (all $p < 0.05$; Table 2). The strongest association was between the RBS-R total sum score and the PSI DC subscale (Table 2).

Results for the caregiver PSI (total score) outcome as a function of RBS-R total score at T0 (i.e., pretest) and change in RBS-R scores are reported in Table 3. RBS-R scores at T0 were positively associated with T0 PSI caregiver stress scores ($B = 0.64$, $p < 0.001$). Results indicated that for each additional *unit* on the T0 RBS-R score, caregiver stress was about 0.64 points higher at this same time point. Similar cross-sectional associations were found between T0 RBS-R scores and T0 PSI Subscale Scores (Table 3). The PCDI subscale also was associated with Mullen Standardized Scores at T0 ($B = -0.06$, $p = 0.015$) indicating that for each additional *unit* on the T0 Mullen Score (indicating higher cognitive abilities), caregiver stress on the PCDI subscale was about 0.06 points lower at T0.

RBS-R scores at T0 and change in RBS-R scores over time predicted change in caregiver PSI scores (Table 3). The positive effects indicated that caregivers of children with higher T0 RBS-R scores have a lower rate of decrease in stress over time ($B = 0.51$, $p < 0.001$). Similarly change in child RBS-R scores over time is mirrored by change in caregiver PSI scores ($B = 4.81$, $p < 0.001$). In other words, as the child's RBS-R scores decrease over time, so does caregiver stress. Therefore, change in caregiver stress is related to both initial levels and change in child RRBs.

Similar associations were found with the individual PSI subscales (Table 3). Caregiver reported stress decreased on all subscales when child RBS-R scores decreased over time with the strongest association observed between RBS-R change and change in caregiver stress for the DC subscale ($B = 2.12$, $p < 0.001$).

Demographic variables (SES, ethnicity, child or caregiver sex) did not predict change in caregiver reported stress over time. The child's overall ASD severity also did not contribute to change in caregiver stress.

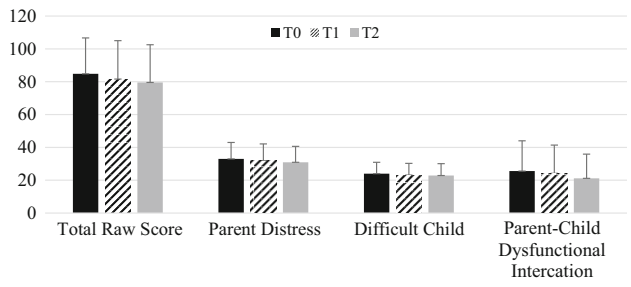

Fig. 2 PSI total and subscale scores across timepoints

Discussion

In general, child RRBs and caregiver stress decreased over time (~1½ years). This is likely attributed to all children being enrolled in high quality preschool classrooms and receiving targeted ASD interventions (see Boyd et al. 2014). However, child RRBs (both initial rates and change over time) predicted changes in caregiver stress over time. Thus, when caregivers reported increasing rates of their child's RRBs over the course of the study, this was mirrored by increases in caregiver reported stress over and above overall child ASD severity, developmental functioning and parent and child demographic variables. Our data support the importance of targeted treatments for RRBs as meaningful change in this core deficit may lead to related improvements in caregiver wellbeing, particularly in a subgroup of children who enter intervention with elevated RRBs.

While we found that increasing RRBs predicted elevated and increasing rates of caregiver reported stress overall and on all three PSI-SF subscales, this relationship was strongest for the Difficult Child subscale. Associations were stronger between RBS-R totals and this subscale than associations with overall PSI-SF scores and the remaining two subscales. This subscale measures specific child characteristics that make him/her difficult or easy to manage. While the PSI-SF DC subscale does not measure ASD specific characteristics or RRBs, caregivers frequently report RRBs as difficult to manage and stigmatizing (Mercier et al. 2000) therefore the association between RRBs and the DC subscale further supports this.

One potential explanation for the strong relationship between RRBs and scores on this subscale is the way in which caregivers of a child with ASD may interpret the items relative to caregivers of a typically developing child. For example, for the item "getting child to do something is hard" caregivers may attribute this to their child's RRBs,

Table 2 Correlations between RBS-R total sum score and PSI total score and subscale scores at T0 ($n = 188$)

	RBS-R	PSI: total	PSI: PCD	PSI: DC	PSI: PD
RBS-R total	–				
PSI total	0.427*	–			
PSI parent–child Dysfunction	0.306*	0.820*	–		
PSI difficult child	0.518*	0.874*	0.600*	–	
PSI parental distress	0.227*	0.834*	0.558*	0.550*	–

* $p < 0.05$

Table 3 Fixed effects for parallel-process growth model: parent stress index (PSI) as a function of child T0 RBS-R total sum score and rate of change

Effect	PSI total score (N = 414, n = 178)		Difficult child (N = 414, n = 178)		Parent–child dysfunctional interaction (N = 414, n = 178)		Parent distress (N = 414, n = 178)	
	Estimate (SE)	*p*	Estimate (SE)	*p*	Estimate (SE)	*p*	Estimate (SE)	*p*
Intercept	81.890 (1.829)	<.001*	31.907 (0.742)	<.001*	23.133 (0.555)	<.001*	26.846 (0.79)	<.001*
Time	−2.672 (0.835)	0.002*	−0.923 (0.382)	0.017*	−0.718 (0.279)	0.011*	−1.083 (0.372)	0.004*
RBSR initial	0.646 (0.104)	<.001*	0.377 (0.042)	<.001*	0.136 (0.032)	<.001*	0.134 (0.045)	<.001*
Time × RBSR initial	0.510 (0.101)	<.001*	0.220 (0.047)	<.001*	0.141 (0.034)	<.001*	0.154 (0.045)	<.001*
Time × RBSR change	4.811 (0.88)	<.001*	2.111 (0.405)	<.001*	1.231 (0.292)	<.001*	1.504 (0.394)	<.001*
Caregiver = male	0.850 (4.432)	0.848	−1.19 (1.809)	0.513	0.289 (1.355)	0.832	1.392 (1.911)	0.468
Female	7.285 (4.016)	0.073	3.499 (1.643)	0.036	1.767 (1.231)	0.155	1.753 (1.732)	0.314
Caregiver race = Hispanic	−15.616 (13.912)	0.265	−4.797 (5.725)	0.405	−2.541 (4.318)	0.558	−7.762 (5.995)	0.199
Caregiver race = Black	−2.998 (4.314)	0.489	−1.492 (1.754)	0.398	−0.832 (1.311)	0.528	0.007 (1.861)	0.997
Caregiver race = Asian	1.402 (6.402)	0.827	−0.477 (2.61)	0.855	1.500 (1.95)	0.444	−0.272 (2.762)	0.922
SES composite	0.107 (0.587)	0.856	0.228 (0.239)	0.342	−0.038 (0.178)	0.832	−0.071 (0.253)	0.780
Mullen std score	−0.036 (0.083)	0.669	0.048 (0.034)	0.163	−0.063 (0.026)	0.015*	−0.019 (0.036)	0.596
ADOS severity score	0.428 (0.954)	0.655	0.454 (0.387)	0.245	0.325 (0.289)	0.265	−0.303 (0.412)	0.465

* $p < 0.05$. *n* to the number of subjects, *N* to the number of repeated measures. All continuous predictors except for time were grand-mean centered

particularly if the child has elevated and/or increasing rates of these behaviors. This could mean that even though this scale measures child characteristics such as compliance and defiance, these may be interpreted differently by caregivers of a child with ASD compared to other caregivers. Further investigation of this relationship is required to understand the association between these variables.

We did not find any predictive associations between demographic variables and caregiver stress. Stress has been found to manifest differentially in mothers and fathers of a child with ASD, with greater rates consistently reported in mothers (Jones et al. 2013; McStay et al. 2014a, b) and different child factors predict stress in mothers versus fathers (Baker-Ericzn et al. 2005; Falk et al. 2014; McStay et al. 2014a, b). Caregiver sex was not associated with caregiver stress in our sample, perhaps in part due to the small number of fathers included in the study. Additionally, ethnicity did not predict caregiver stress. As with previous research, we also did not find any association between SES and caregiver stress (Totsika et al. 2013); however, this could be due to the relatively high socioeconomic status of our sample.

Overall ASD severity (as indexed through the ADOS-G Calibrated Severity Scores) did not predict caregiver stress despite previous research indicating an association between severity and caregiver stress (Hastings and Johnson 2001; Hayes and Watson 2015). Within our sample, the findings indicate that RRBs uniquely contributed to change in caregiver-reported stress over and above the contribution of overall ASD severity. One potential factor could be the relative lack of variance in the ADOS-G Severity Scores in this sample, with the majority falling in the more severe end of the scale. With more variance in functioning and severity levels, associations between severity and caregiver stress might emerge.

We did find a significant association between child developmental abilities and caregiver stress on the PCDI subscale, indicating that caregivers of children with lower developmental functioning reported greater levels of stress on this subscale. The PCDI subscale measures how the

caregiver perceives their interactions with their child and whether these are viewed as positive or negative—caregivers of children with lower developmental abilities perceived their interactions to be more negative, suggesting that child functioning exerts an independent effect on caregiver stress related to specific aspects of the caregiver-child dyad. Child developmental abilities have not been consistently found to independently predict caregiver stress (Totsika et al. 2011), therefore this finding also merits further investigation.

Research suggests that early targeted intervention (Estes et al. 2013b; Feinberg et al. 2014; Kasari et al. 2015; Tonge et al. 2006) and CTMs (D'Elia et al. 2014) are beneficial for both child behaviors and caregiver stress levels; yet, most early intervention currently targets social-communication behaviors in ASD (Boyd et al. 2012; Harrop 2015). Thus it remains unclear whether caregivers and their children would benefit from more targeted interventions for RRBs. The overall reduction in caregiver reported stress across the whole cohort supports the importance of comprehensive intervention not only for child progress but also for caregiver wellbeing. Still greater gains may be observed if additional support in the management of RRBs was available, in particular for children with elevated rates of RRBs at entry into intervention.

A recent pilot study found increased levels of caregiver self-efficacy following brief caregiver training specifically targeting RRBs in 3–7 years olds (Grahame et al. 2015). Higher levels of maternal self-efficacy have been associated with increased maternal well-being and decreased feelings of guilt in mothers of children with ASD (Kuhn and Carter 2006). Therefore improvements in self-efficacy could potentially mediate the relationship between child behaviors and stress.

While caregiver stress is reported to remain relatively consistent over childhood and adolescence, there are undoubtedly key developmental transitions, such as entering preschool, that may result in fluctuations in both child behavioral characteristics and caregivers stress levels (Estes et al. 2013a). Indeed research has indicated that caregiver depression is elevated during the preschool years due to the transition into preschool services (Carter et al. 2009). Thus caregiver stress may rise upon entry into preschool but reduce over time, as such it is important to study caregiver stress over the course of the lifespan.

We did not study the potential transactional and bidirectional relationships between child RRBs and caregiver stress, although these have been previously reported (Lecavalier et al. 2006; Totsika et al. 2013). It is important to recognize factors that may influence caregiver stress (such as increasing RRBs) but also identify the factors that may affect child success within a given intervention (Osborne et al. 2008). As caregivers frequently act as advocates, educators and interventionists in addition to caring for their child with ASD, it is important to study how caregiver variables (such as stress) influence treatment implementation and progress. It is important to note that caregiver report was used to measure both child RRBs and caregiver stress which may have led to informant bias—as such caregivers who were more stressed may have rated their children as having greater RRBs or vice versa. While this is a limitation, it also represents a key feature of the study as it is important to understand how caregivers' perceptions of their child's behaviors impact their own wellbeing.

Further, RRBs in ASD are associated with other clinical features, such as sleep problems (Gabriels et al. 2005), therefore the relationship found between child RRBs and caregiver stress could be transmitted through a third factor. Additionally, recent mediation analyses suggest other maternal variables, such as fatigue and self-efficacy, may mediate the relationship between child problem behaviors and caregiver stress (Kuhn and Carter 2006; Seymour et al. 2013). Thus it will be important in future research to explore potential mediating factors in the relationship between caregiver stress and the behavioral phenotype of children with ASD.

Limitations

It is worth acknowledging that while the PSI-SF has been used extensively to explore parenting stress in ASD, it was not developed for families with a child with developmental disabilities. The suitability of the three subscales of the PSI-SF have been questioned for families with a child with ASD (Zaidman-Zait et al. 2011) due to the qualitative differences in raising a child with ASD relative to typically developing children. Researchers have proposed a more suitable six factor solution for caregivers of children with ASD based on the 36 PSI-SF items, which—while has not been extensively validated—should be considered in future research. Finally, due to the design of the larger comparative study (Boyd et al. 2014), not all families were seen at all three time points. While the caregivers who completed T2 questionnaires did not differ from the T0 and T1 samples in terms of key demographic variables, data on the full sample would have been advantageous.

Conclusions and Recommendations

Despite overall reductions in both child RRBs and caregiver stress, when increased rates of RRBs were reported this was mirrored by increased caregiver stress ratings. As RRBs have been shown to change minimally following intensive intervention (Dawson et al. 2010; Wetherby et al. 2014), are often difficult to redirect (Harrop et al. 2015; Mercier et al. 2000) and impact how caregivers interact

with their child (Woolfson and Grant 2006), it is important to identify effective intervention strategies for these behaviors as these may have beneficial effects on caregiver wellbeing. This study further supports the importance of involving caregivers within research and treatment and identifying what predicts caregiver stress in order to help guide intervention efforts.

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