Title

Parenting stress in caregiver-mediated interventions for toddlers with autism: An application of quantile regression mixed models

Journal

Autism Research, 15(2)

ISSN

1939-3792

Authors

Schlink, Andrew
Williams, Justin
Pizzano, Maria
et al.

Publication Date

2022-02-01

DOI

10.1002/aur.2637

Peer reviewed

Published in final edited form as:

Autism Res. 2022 February ; 15(2): 353–365. doi:10.1002/aur.2637.

Parenting Stress in Caregiver-Mediated Interventions for Toddlers with Autism: An Application of Quantile Regression Mixed Models

Andrew Schlink[1], **Justin Williams**[2], **Maria Pizzano**[1], **Amanda Gulsrud**[3], **Connie Kasari**[1]

[1]Department of Education, University of California, Los Angeles, Los Angeles, CA, UCLA Semel Institute 68-268, 760 Westwood Plaza, Los Angeles, CA 90095

[2]Department of Biostatistics, University of California, Los Angeles, Los Angeles, CA, UCLA Semel Institute 68-268, 760 Westwood Plaza, Los Angeles, CA 90095

[3]Semel Institute for Neuroscience and Human Behavior, University of California, Los Angeles, Los Angeles, CA, UCLA Semel Institute 68-268, 760 Westwood Plaza, Los Angeles, CA 90095

Abstract

Traditional longitudinal modeling approaches require normally distributed data and do not account for sample heterogeneity. Parenting stress, in particular, can be difficult to model across time without transforming the data as it is usually high for caregivers of children with ASD. This study used novel linear quantile mixed models (LQMMs) to model non-normal parent stress scores across two caregiver-mediated interventions involving toddlers with ASD. The sample included 86 caregiver-child dyads who were randomized to either a parent-only psychoeducational intervention or hands-on parent training in a naturalistic developmental intervention. Child and parent-related domains of the Parenting Stress Index (PSI) were the primary outcomes in this study. The PSI was collected at entry, 10-week exit, 3-month follow-up, and 6-month follow-up periods. Separate LQMMs were used to model five specific quantiles (τ = 0.1, 0.25, 0.5, 0.75, and 0.9) of the two PSI domains across the complete intervention timeline. These five quantiles effectively modeled the entire conditional distribution of parenting stress scores. The LQMMs indicated that child-related parenting stress decreased across all quantiles within both interventions, with no difference in the rate of parenting stress change between the intervention groups. For parent-related parenting stress, the effect of intervention depended on the group's stress level; some parents increased their perceived stress within the hands-on intervention at the 3-month follow-up. Overall, this study demonstrated that the use of LQMMs yielded additional information, beyond traditional longitudinal models, regarding the relationship between parenting stress within two caregiver-mediated intervention protocols. This study also discussed the methodological contributions and potential future applications of LQMMs.

Lay Summary

Corresponding Author: Andrew Schlink, UCLA Semel Institute 68-268, 760 Westwood Plaza, Los Angeles, CA 90095, aschlink@ucla.edu, 818-427-8183.

Clinical trial information:
Optimizing Social and Communication Outcomes for Toddlers with Autism, ClinicalTrials.gov identifier: NCT00999778

This study used a newer longitudinal modeling technique to examine how parenting stress changed across two caregiver-mediated interventions for toddlers with ASD. Results showed that certain parents in the JASPER condition might require additional support as they exit the study and enter into their first follow-up period. It was also determined that this new modeling technique could be a valuable tool to analyze highly variable data often present in ASD intervention studies.

Keywords

autism spectrum disorder; toddlers; parenting stress; caregiver-mediated interventions; JASPER; linear quantile mixed models

Introduction

Young children with autism experience difficulties in social communication, a core characteristic of the disorder (American Psychiatric Association, 2013). Early social communication, including joint attention, can be learned via caregiver interactions (Kasari et al., 2006, 2008, 2010). Co-orientation between child and caregiver serves as a "zone of proximal development" (Vygotsky, 1978), where caregivers introduce language during social interactions (Adamson et al., 2004, 2009; Tomasello, 1995). Caregivers therefore are powerful agents of change and can be trained in evidence-based interventions to optimize child outcomes, improve the implementation of interventions, and maximize caregivers' feelings of efficacy.

Caregiver-Mediated Interventions

In caregiver-mediated approaches to intervention, caregivers learn strategies to implement at home with their child. This mode facilitates natural learning opportunities for the child and encourages the generalization of skills (Oono et al., 2013). One consideration of caregiver-mediated approaches is their complexity; they introduce an additional source of variability to intervention response beyond the child — caregiver characteristics.

These additional and potentially highly variable caregiver characteristics likely contribute to social communication outcomes in caregiver-mediated interventions and require a closer examination. Some parents are highly motivated and ready to actively engage in an intervention targeting their child's social communication outcomes. Others are not, and may be anxious and/or resistant to making changes in their own interaction strategies. Parents are also coping with the stressors that come about from parenting a young child generally, as well as concerns over their child's development.

Parenting Stress

Self-reported parenting stress has been identified as a potentially influential factor to intervention efficacy and therefore warrants examination (Osborne et al., 2008). First, it is widely understood that parenting stress is a pervasive issue in the autism population; caregivers of children with ASD experience levels of parenting stress significantly higher than those experienced by parents of typically developing children and by parents of children with other disabilities (Brobst et al., 2009; Duarte et al., 2005; Dumas et al.,

1991; Eisenhower et al., 2005; Hoffman et al., 2009; Schieve et al., 2007). In particular, toddlers' difficulty with reciprocal social interactions and caregivers' perception of poor social relatedness with their child are both salient predictors of overall parenting stress (Davis & Carter, 2008). When training caregivers, the optimal intervention intensity for parental self-efficacy depends on parenting stress pre-treatment, indicating that parenting stress is a crucial variable to consider in tailoring interventions for optimal parent and child outcomes (Estes et al., 2021). Therefore, understanding how this stress manifests throughout a caregiver training program may be an important step in improving intervention protocols and maximizing child improvement.

Next, parenting stress is a complicated construct that has two distinct domains: child-related and parent-related. Child-related parenting stress refers to the perceived stress from their child's specific behaviors, while the parent domain reflects more general stress related to parenting in general (Loyd & Abidin, 1985). Independently examining each facet of stress may highlight additional ways to support caregivers in caregiver-mediated interventions.

Lastly, evidence suggests that although parenting stress may be especially prevalent in the autism population, it can be alleviated via education programs that teach strategies to manage stress as well as child behaviors (Feinberg et al., 2014; Kasari et al., 2015; Tonge et al., 2006). Reframing parenting stress as a malleable construct further substantiates the argument to understand how it manifests in intervention contexts. Further, applying a new longitudinal modeling approach can help identify the nuances of changes in non-normally distributed parent stress throughout the course of learning intervention strategies for their child with autism.

The Current Study

Previous work from our group found that caregivers reported lower child-related parenting stress throughout active treatment and at the 6-month follow-up after participating in a psychoeducational intervention, but not in a hands-on caregiver-mediated intervention (Kasari et al., 2015). Additionally, there were no differences in parent-related parent stress between the groups. The present study adds to Kasari et al.'s (2015) previously reported findings by using a longitudinal modeling technique to examine how parenting stress fluctuates throughout intervention for parents with heterogeneous stress levels.

This secondary data analysis reexamined parent stress data from the original Kasari et al. (2015) study. We were interested in better understanding how the level of reported child and parent-related parenting stress fluctuated across the entire distribution of parenting stress scores (i.e., parents with different parenting stress levels) within intervention and education programs across the complete timeline of study participation (i.e., entry, 10-week exit, 3-month, and 6-month follow-ups). We modeled the full distribution of stress scores with an extension of traditional mixed modeling techniques, a linear quantile mixed model (LQMM).

This study aims to determine whether LQMM results yield additional information, beyond traditional longitudinal models, regarding the relationship between parenting stress and two caregiver-mediated intervention protocols. We first examined individual domains of

parenting stress (child and parent) with LQMMs. Next, we compared results from the LQMMs to results from a traditional modeling technique. Finally, we discussed the methodological contributions and potential future applications of LQMMs.

Methods

This study includes data on toddlers with ASD and their caregivers from a previously published randomized controlled trial (Kasari et al., 2015).

Participants

This sample includes the 86 toddler-caregiver dyads from Kasari and colleagues' (2015) RCT. Toddlers were initially recruited from the same intensive 10-week outpatient early intervention (EI) program, which used a combination of therapy approaches, including behavioral, speech, and occupational. A University Institutional Review Board approved the original study, and parents provided written consent.

Before entrance into the original study, children met the specific inclusion and exclusion criteria, as previously described by Kasari et al. (2015). Children needed to be younger than 36 months at the entry of the study, have a clinical diagnosis of ASD confirmed by both the Autism Diagnostic Interview-Revised (ADI-R; Lord et al., 1993) and the Autism Diagnostic Observation Schedule (ADOS; Lord et al., 2000), and be available for follow-up assessment.

An independent data-coordinating center used a random numbers list to randomize participants to either the JASPER or PEI group. Despite randomization, the average chronological age at baseline of children in the JASPER group (30.7 months) was significantly younger ($p = 0.01$) than the PEI group (32.3 months). The remaining demographic variables were successfully matched between the two groups, as shown in Table 1. Three dyads discontinued treatment, and another ten did not complete follow-up assessments. Figure 1 shows the full participant recruitment diagram. The original study employed an intent-to-treat model, and all participants were included in the statistical models regardless of attrition.

Procedures

After informed consent, families were randomized to receive either JASPER or PEI interventions. Each intervention protocol included one hour of interventionist contact per week.

The goal of the JASPER condition was to teach parents the strategies of JASPER via direct coaching with their child present. JASPER is an evidence-based, manualized treatment for toddlers and preschoolers, whose focus is on increasing children's joint engagement with their caregiver to facilitate the frequency of joint attention gestures and play skills (Kasari et al., 2006, 2008, 2012, 2014).

The purpose of the PEI group was to provide 1:1 individual education and support to parents of young children with autism. PEI intervention goals were to teach parents the principles of managing children's behaviors, methods to manage parental stress, and strategies to teach

new skills to their child. More detail about PEI is provided in the original publication (Kasari et al., 2015; Brereton & Tonge, 2005).

Families continued JASPER or PEI interventions for the duration of the same 10-week EI program. Due to the intensity of the EI program (30 hours per week of therapy), families suspended other early intervention services. During the follow-up periods, families resumed their early intervention services.

Among the original study's various outcome measures, parents completed a parenting stress questionnaire at four time points throughout the study; entry, 10-week exit, 3-month follow-up, and 6-month follow-up. Participants were excluded from the present analyses if the questionnaire was not completed.

Measures

The Parenting Stress Index (PSI; Loyd & Abidin, 1985)

Structure.: The PSI is a 101-item inventory that measures parent-reported stress within the parent/child system. The PSI has been frequently used to measure parenting stress among parents of children with autism as well as children with other developmental disabilities (Abidin, 1995). The PSI structure contains two subdomains, one that measures child characteristics and the other that measures parent characteristics. The PSI was administered at entry, exit, and at 3 and 6-month follow-ups.

Child-Domain.: The child domain assesses parenting stress that derives from child behaviors that may make it difficult for parents to fulfill their parenting duties. The child domain consists of six subscales: Distractibility/Hyperactivity, Adaptability, Reinforces Parent, Demandingness, Mood, and Acceptability. Examples of items include, "Sometimes I feel my child doesn't like me and doesn't want to be close to me"; "My child smiles at me much less than I expected."

Parent-Domain.: The parent domain estimates perceived parental stress that stems from parents' functioning, specifically, the parent's relationship with their child and how the parent feels about his or her life outside the parent-child system. The parent domain consists of seven subscales: Competence, Isolation, Attachment, Health, Role Restriction, Depression, and Spouse/Parenting Partner Relationship. Examples of items include, "I feel that I am successful most of the time when I try to get my child to do or not do something"; "When I think about the kind of parent I am, I often feel guilty or bad about myself."

Scoring.: Parents rate each item on a 5-point Likert scale that describes the degree to which they agree or disagree with statements regarding parental stress. Items are summed to yield child and parent domain scores, where higher scores indicate greater perceived stress. The two domains are summed to obtain a total stress raw score. The normal range for the total stress raw scores is between 175–245, approximately the 10^{th} to 75^{th} percentile rank. Parents who obtain very high scores at or above the 85^{th} percentile (>260 total stress raw score) are considered to be borderline clinically significant based on the norms of the full PSI (Abidin, 2012; Loyd & Abidin, 1985).

Reliability.: The PSI provides scores that demonstrate stability over repeated applications. In a sample of 37 mothers who were re-administered the PSI after a one-year interval, the test-retest reliability coefficients were 0.55 for the child domain and 0.70 for the parent domain, which indicate sufficient stability over time considering typical child development changes in the family system (Hamilton, 1980).

Data Analysis

Linear Mixed Effect Models vs. Linear Quantile Mixed Models—Parenting stress scores that are observed in clinical studies offer unique challenges when it comes to analysis. They are often non-normally distributed and do not lend themselves naturally to traditional longitudinal modeling techniques. In the original study, researchers first inverted the parenting stress scale to determine if the data were zero-inflated. There was a significant overrepresentation of very highly stressed parents, requiring the use of a generalized linear mixed model (GLMM) with zero-inflated Poisson distribution to model the average stress scores over time (entry, exit, and 6-month follow-up) by treatment group (JASPER vs. PEI). The GLMM model indicated an *on average* reduction for child-related parenting stress over time for parents in the parent education intervention group compared to the hands-on intervention group. There was no difference between the two treatment groups in the change of average parent-related stress over time (Kasari et al., 2015).

This study proposes using a linear quantile mixed model (LQMM) (Geraci & Bottai, 2007). The premise of a LQMM is that factors may induce differential rates of change to distinct quantiles of the outcome variable; for instance, infants with low birthweight may be more adversely affected by parental smoking than average weight infants (Koenker & Bassett 1978; Geraci, 2014). The objectives of using a LQMM to model parenting stress were to understand how particular intervention protocols may differentially affect parents with varying levels of parenting stress. LQMMs, which directly model *discrete quantiles* of the outcome variable, differ from commonly used longitudinal models like linear mixed effect regression (LMER) models, which model the *mean* response. While LQMMs are estimated with respect to discrete quantiles, the entire dataset is used to estimate each quantile assuring that there is no data loss, and the consistency of estimation is a function of the sample size of the data, not the sample size within quantiles. By modeling quantiles, LQMMs also offer protection against strong outliers that influence the mean response more strongly.

Additionally, LQMMs make no distributional assumptions about the outcome variable, while LMER assumes that the errors and random effects are normally distributed. This is a key difference as when the assumptions of LMER are violated, the resulting estimates are known to be biased (Geraci & Bottai, 2007). If the data are in fact normally distributed and LQMMs are used, there is no bias in the results, but the efficiency and precision of the estimation are reduced (Geraci & Bottai, 2007). This means that quantile regression often needs more data than linear regression to achieve the same accuracy level. By relaxing the assumption of normality, LQMM models can be used to make inferences in the presence of significantly skewed or inflated data without requiring transformations. This can lead to a more natural interpretation of the coefficients on the response variable.

For practitioners, one of the key decisions when running a LQMM analysis is the choice of quantiles to estimate. The choice of quantiles should be driven by the research question of interest, data availability as quantiles require more data to be estimated accurately as they approach 0 or 1, and the underlying distribution of the response. Concerning parenting stress, an LQMM may illuminate the differential effect of treatment in parents. It is also particularly useful as the PSI scores within treatment groups exhibit some evidence of non-normality. In particular, we were interested in the effect on very low, low, middle, high, or very high levels of parenting stress, which were functionally defined as the 10th, 25th, 50th, 75th, and 90th quantiles. We would suggest using similar quantiles to research in order to determine if any differential effects exist along a reasonable range of quantile values. Inclusion of the 50th quantile, i.e., the median, can often serve as a useful comparison to the LMER mean response model.

Details of the Linear Quantile Mixed Models—Two separate linear quantile mixed models (LQMM) were constructed to model the longitudinal trajectory of both parent and child raw domain scores to assess potential treatment effects. We examined stress subdomain raw scores, rather than percentiles or total stress scores, with the assumption that this would give a more nuanced description of the nature of stress in this sample. Each LQMM examined changes in stress scores across four timepoints: entry, 10-week exit, 3 and 6-month follow-ups.

Structure.: In each model, five specific quantiles ($\tau = 0.1, 0.25, 0.5, 0.75,$ and 0.9) of the outcome variable were identified a priori and investigated for differential treatment effects. These quantiles were chosen to accurately model the full distribution of stress scores in each PSI domain. Operationally, we can think of these groups as very-low, low, middle, high, and very-high parenting stress.

Each model contained the fixed effects of the treatment assignment (JASPER vs. PEI) and time (entry, exit, 3-month follow-up, and 6-month follow-up) along with an interaction of treatment by time. Treatment effects were defined as the interaction between treatment and time, resulting in 3 potential significant periods: exit vs. entry, 3-month follow-up vs. entry, and 6-month follow-up vs. entry. Time by treatment interactions were examined within each quantile. Additionally, random intercepts were incorporated to account for the within-subject variability due to the repeated measurement design. Chronological age was controlled for in each model due to evidence of significant differences between groups.

Novelty.: While typical linear mixed-effect regression (LMER) relies on normality assumptions on the residuals and random effects, LQMM invokes an asymmetric Laplace density to estimate parameter coefficients numerically. Under settings where data are skewed or have few observations, the normality assumptions for LMER may no longer be valid. As a comparison, each stress domain was also fit using an LMER with the same fixed effects, treatment interaction, and random intercept as those used for LQMM. In this way, the fitted values for the average outcome could identify particular quantiles where differential treatment effects were occurring. We would expect that with normally distributed data, the results from the LMER and the median LQMM should be identical with slightly larger error

bars for the latter. The standard errors for LQMM were calculated using a block bootstrap method with $R = 1000$ replications.

LQMMs are a relatively new technique that have not yet been applied to autism intervention research. The variability in autism symptomatology presentation combined with differential treatment responses offer an opportunity for LQMMs to track responses to treatment over time more accurately.

Software.: Descriptives were conducted in jamovi version 1.2 (The jamovi project, 2020), and statistical analysis was conducted in R version 3.6 (R Core Team, 2019) using the lqmm package (Geraci, 2014; Geraci & Bottai, 2014).

Results

Descriptives

At entry, 57 (67%) of the parents had extreme total stress raw scores, and 47 (55%) of those parents scored above 260, effectively classifying more than half the sample as having clinically significant stress. Table 2 outlines total stress scores by timepoint and treatment group. Violin and scatter plots in Figure 2 depict the distributions of PSI raw stress scores in each domain by treatment group and time point.

Child Domain Model

LQMM Effects—There were no significant treatment effects within the child domain across any of the quantiles, as shown in Table 3. Figure 3 shows the estimated trajectory of parental stress and corresponding 95% confidence intervals by quantile. The model indicates that all parents decreased their child domain stress scores across all quantile levels, after controlling for chronological age. This uniform decrease in stress indicates that both interventions mitigated child-related stress for parents across the full distribution of raw child domain stress scores.

Visual inspection of Figure 3 indicates that this decrease in stress scores was especially pronounced for the entry to exit period. Parents in the PEI group with very low, low, middle, high, and very high stress entered the study with child domain stress scores of 119.43, 124.08, 136.52, 146.82, and 154.20, respectively. At the exit of the study, these scores decreased to 106.61 (−12.82), 109.64 (−14.44), 124.49 (−12.03), 136.20 (−10.62), and 143.85 (−10.35). At the 6-month follow-up, these scores were maintained at 106.87, 110.08, 124.67, 136.40, and 143.38.

Similarly, parents in the JASPER group with very low, low, middle, high, and very high stress entered the study with child domain stress scores of 109.80, 115.22, 132.08, 145.77, and 152.59, respectively. At exit of the study, these JASPER parents decreased their child-related stress to 101.91 (−7.89), 103.82 (−11.40), 123.54 (−8.54), 138.82 (−6.95), and 143.91 (−8.68). Additionally, these parents slightly decreased their stress scores at the 6-month follow-up with scores of 96.70, 100.55, 120.49, 136.33, and 140.67. However, it is important to note that the stress reduction rate across quantiles from entry to exit and entry to the 6-month follow-up did not statistically differ between treatment groups.

Table 3 outlines the estimates of the child domain model. Each estimate corresponds to the difference in slope of parental stress raw score for JASPER vs. PEI group from entry to either exit, 3-month follow-up, or 6-month follow-up. Positive values indicate an increase in parental stress.

LMER vs. LQMM—Figure 3 shows that the linear mixed-effect regression model estimates for the child domain of the PSI returned similar values as the median quantile in LQMM. In this model, we also observe no significant treatment effects, but both groups decreased their child-related stress through the course of the study. The similarity between the median quantile and the LME model is evidence that the data are not strongly skewed for the child domain raw scores.

Parent Domain Model

LQMM Effects—A nuanced secondary analysis of Kasari et al.'s (2015) original study found significant effects in the parent domain stress scores across time and treatment assignment. Significant treatment by time interactions occurred from entry to 3-month follow-up in the very-low ($\tau = 0.1$), low ($\tau = 0.25$), middle ($\tau = 0.5$), and high ($\tau = 0.75$) stress groups (Table 4). Among parents in these four stress quantiles, the change in parent-related stress scores from entry to the 3-month follow-up depended on treatment assignment.

Parents in the JASPER group with very low, low, middle, and high stress entered the study with stress scores of 97.64, 114.62, 133.23, 136.66, respectively. At the 3-month follow-up, these stress scores had risen to 103.57 (+5.93), 118.40 (+3.78), 137.44 (+4.21), and 141.48 (+4.82).

Conversely, parents in the PEI group within these same stress groups ($\tau = 0.1$, $\tau = 0.25$, $\tau = 0.5$, $\tau = 0.75$) entered the study with stress scores of 99.49, 118.35, 130.22, 137.08, respectively. The PEI groups' stress scores decreased at their 3-month follow-up to 94.12 (−5.37), 112.75 (−5.60), 124.84 (−5.38), and 131.52 (−5.56), respectively. Figure 4 shows how parent domain stress scores changed over time by treatment group and quantile.

LMER vs. LQMM—Linear mixed-effect regression for the parent domain scores shows that parents entered the study with average stress scores of 126.06 and 127.46 for the PEI and JASPER groups, respectively. Similar to the LQMM results, there was a significant interaction at the 3-month follow-up time point. This indicates that, on average, these stress scores significantly differed over time, depending on the treatment assignment. At the 3-month follow-up, parents in the PEI group decreased their stress scores to 124.83 (−1.23) on average, while parents in the JASPER group increased their stress scores to 135.97 (+8.51) on average. These score changes from entry to the 3-month follow-up corresponded to a significant treatment effect ($\beta = 9.75$, $SE = 4.92$, $t(219.3) = 1.98$, $p = 0.049$). Comparing these to the estimated quantile treatment effects (τ; 0.1 = 11.30, 0.25 = 9.78, 0.5 = 9.59, 0.75 = 10.31, 0.9 = 6.23), we see that the parents with very-low stress were most adversely affected (largest increase in stress) while those with very high stress did not see a significant treatment effect.

Discussion

Several notable findings emerged from this study's nuanced examination of parenting stress within caregiver-mediated early interventions involving toddlers with ASD. This examination suggests that quantile models offer potential opportunities to improve intervention implementation. Overall, trends in LQMM results suggest that although both treatment groups in this study may alleviate child-related parenting stress, the quantile regression indicates that caregiver-related stress develops differently across quantiles for the two caregiver-mediated interventions from entry to the 3-month follow-up. Applying a relatively novel modeling approach and utilizing all timepoints is essential to better understand the complexity of parenting stress in caregiver-mediated interventions.

Differential Parenting Stress Patterns for Child and Caregiver Domains

Child-Related Stress—The original study previously reported a reduction in average child-related parenting stress over time for parents in the PEI group compared to the JASPER group (Kasari et al., 2015). Our model, including 3-month follow-up data, indicates that both treatment groups alleviate child-related stress from entry to all other time points, but with no statistical difference in rates of decrease between treatment groups across any of the quantiles. As a different statistical method was used, it is unsurprising that results differed slightly between this study and the original study. However, it is encouraging to see that parents across all quantiles and both treatment groups drastically reduced their child-related parenting stress from entry to all other time points. Quantile regression results imply that both interventions effectively taught strategies to parents that helped them manage and understand their children's behaviors.

Suggestions to Mitigate Child-Related Parenting Stress.: Reducing child-related parenting stress is a crucial next step. Mothers' negative perceptions about their children's temperament negatively affect the time spent engaged with their children (Kasari & Sigman, 1997). Minimizing child-related stress can foster an environment where a caregiver-mediated social communication intervention can be most effective. Caregiver support groups are one way to incorporate the opportunity to interact with other similar parents and may improve parents' well-being (Catalano et al., 2018; Stuart & McGrew 2009).

Caregiver-Related Stress—We previously reported no statistical difference between the mean parent-related stress scores of the two treatment groups (JASPER vs. PEI) across any timepoints (entry, exit, and 6-month follow-up) while using a zero-inflated GLMM (Kasari et al., 2015). Our findings from the current study revealed that caregiver-related parenting stress does change over time but depending on individual stress levels in addition to treatment assignment.

Parent-related stress increased for the JASPER group and slightly decreased for the PEI group across all quantiles from the entry to the 3-month follow-up period. In this time period, the difference in the rate of change between treatment groups was statistically significant for all quantiles except for those with very high stress levels ($\tau = 0.9$). One group, in particular, the very low stress JASPER group, had a significantly steeper increase rate of change in parent-related parenting stress compared to the very low stress PEI group.

The increase in parent-related stress for the JASPER group across quantiles into the 3-month follow-up period may indicate that parents lost confidence in using specific JASPER skills and strategies once active support from the research team stopped. It is possible that parents in the low stress JASPER group may be less concerned during active treatment due to the potential over-reliance on the research team's support. For these low stress parents, the rigors of using new intervention strategies may only be realized once support from the research team has stopped after the exit timepoint.

Suggestions to Mitigate Parent-Related Parenting Stress.: Reducing parent-related parenting stress may be one way to improve caregiver-mediated intervention efficacy. Implementing occasional booster sessions after a study's termination can increase the maintenance of strategies learned throughout the study. Continuing use of effective strategies may then improve parents' confidence to use a hands-on intervention like JASPER, decrease stress, and potentially improve intervention efficacy.

Methodological Contribution of LQMMs

LQMMs are a useful method to analyze longitudinal autism intervention data. In autism intervention studies, there are typically strict inclusion and exclusion criteria for child characteristics, thus creating a homogeneous sample of children. A sample of children that is similar on observed variables helps ensure that significant effects can be attributed to the intervention rather than confounding variables. In caregiver-mediated interventions, parents play a crucial role by delivering the intervention to their children. However, there are typically no inclusion and exclusion criteria for parents. This results in a sample of parents with highly variable characteristics, which may obfuscate treatment effects. Parenting stress is one characteristic that could influence both child and parent outcomes. Instead of statistically controlling for parenting stress or modeling how average stress scores change over time, we aimed to further explore the variability in parenting stress development within caregiver-mediated interventions with novel linear quantile mixed models.

Advantages of LQMMs

Relaxed Statistical Assumptions.: LQMMs offer a clear advantage over LMERs as it does not assume common parametric distributions, meaning it can be used with many different types of data (Koenker & Bassett, 1978). A major drawback of LMER is its strict normality assumption criteria. If these assumptions are not met, LMER may yield inaccurate inferences since the mean may not represent the skewed or inflated distribution. LQMMs, on the other hand, can model the median and any number of quantiles associated with the dependent outcome, which may more accurately depict non-parametric data. In autism intervention research, the heterogeneous nature of the disorder paired with potentially highly variable responses to treatment may benefit from quantile regression that is appropriate for these types of data.

Explores Unexplained Variability.: In the context of intervention studies, it may be of substantive interest to determine if an intervention affects individuals differently. For caregiver-mediated studies, parents could have different stress responses, which may affect the implementation of an intervention. To understand how parents may have different stress

reactions to intervention, our study modeled five quantiles (τ = 0.1, 0.25, 0.5, 0.75, *and* 0.9) of stress scores over time by treatment group. In intervention data, a thorough investigation of a dependent outcome's full conditional distribution may illuminate trends among certain participants and highlight those who may need additional support.

We also included traditional linear mixed effect analyses in our study to highlight the differences between the models. The LMER analysis plotted the *average* stress score by timepoint and treatment group. Although plotting the average scores across time may show general trends, we may lose information on caregivers with greater variability in their stress scores. Therefore, by analyzing changes in parenting stress by quantile, we can tap into additional variability that may help tailor interventions to benefit all parents.

Limitations

Findings from this study should be interpreted with caution due to the small sample size. 86 participants were included in this analysis, with a total of 306 data points for parent-related and child-related stress across the four time points. The multilevel structure of the data, especially with the numerous quantiles, may have limited the power of the analysis. The longitudinal nature of these caregiver-mediated intervention studies indicates the need for larger sample sizes so that quantile trends can be more robust. A larger sample size would allow for an even more detailed analysis of how parenting stress is affected by caregiver-mediated interventions. Future research should look into the dyadic variability to determine the relationship between parenting stress and autism severity.

Conclusions

This study aligns with previous work highlighting the need to address parenting stress among caregivers of children with autism, especially in the context of caregiver-mediated interventions (Keen et al., 2010; Osborne et al., 2008). In longitudinal autism intervention studies, it is of substantive interest to model the complete conditional distribution of a dependent variable, not just the mean, to understand the full effects of intervention more thoroughly. A thorough statistical analysis like LQMMs may help unpack the heterogeneity of autism symptoms and associated behaviors to understand treatment responses better. Applying LQMMs was beneficial in further explaining the variability in parenting stress scores within caregiver-mediated interventions.

Acknowledgments

This study was supported by NICHD, Autism Center of Excellence P50-HD-055784, Determinants of Social, Communicative, and Other Core Deficits in Autism (Bookheimer, PI) project 4, Optimizing Communication in Toddlers with Autism (Kasari, PI), Clinical trials.gov # NCT00999778, and 655054CL (SPROUT) from the Simons Foundation Autism Research Initiative. AS, JW, MP, AG, and CK have no potential conflicts of interest to declare.

References

Abidin RR (1995). Parenting stress index: Professional manual (3rd ed.). Odessa, FL Psychological Assessment Resources.

Abidin RR (2012). Parenting Stress Index, Fourth Edition (PSI-4). Lutz, FL: Psychological Assessment Resources.

Adamson LB, Bakeman R, & Deckner DF (2004). The Development of Symbol-Infused Joint Engagement. Child Development, 75(4), 1171–1187. 10.1111/j.1467-8624.2004.00732.x [PubMed: 15260871]

Adamson LB, Bakeman R, Deckner DF, & Romski M (2009). Joint Engagement and the Emergence of Language in Children with Autism and Down Syndrome. Journal of Autism and Developmental Disorders, 39(1), 84–96. 10.1007/s10803-008-0601-7 [PubMed: 18581223]

American Psychiatric Association. (2013). Diagnostic and statistical manual of mental disorders (5th ed.). Washington: American Psychiatric Association.

Brereton A, & Tonge B (2005). Preschoolers with autism: An education and skills training programme for parents: Manual for clinicians. London: Jessica Kingsley Publishers.

Brobst JB, Clopton JR, & Hendrick SS (2009). Parenting Children With Autism Spectrum Disorders: The Couple's Relationship. Focus on Autism and Other Developmental Disabilities, 24(1), 38–49. 10.1177/1088357608323699

Catalano D, Holloway L, & Mpofu E (2018). Mental Health Interventions for Parent Carers of Children with Autistic Spectrum Disorder: Practice Guidelines from a Critical Interpretive Synthesis (CIS) Systematic Review. International Journal of Environmental Research and Public Health, 15(2). 10.3390/ijerph15020341

Davis NO, & Carter AS (2008). Parenting Stress in Mothers and Fathers of Toddlers with Autism Spectrum Disorders: Associations with Child Characteristics. Journal of Autism and Developmental Disorders, 38(7), 1278–1291. 10.1007/s10803-007-0512-z [PubMed: 18240012]

Duarte CS, Bordin IA, Yazigi L, & Mooney J (2005). Factors associated with stress in mothers of children with autism. Autism, 9(4), 416–427. 10.1177/1362361305056081 [PubMed: 16155057]

Dumas JE, Wolf LC, Fisman SN, & Culligan A (1991). Parenting stress, child behavior problems, and dysphoria in parents of children with autism, down syndrome, behavior disorders, and normal development. Exceptionality, 2(2), 97–110. 10.1080/09362839109524770

Eisenhower AS, Baker BL, & Blacher J (2005). Preschool children with intellectual disability: Syndrome specificity, behaviour problems, and maternal well-being. Journal of Intellectual Disability Research, 49(9), 657–671. 10.1111/j.1365-2788.2005.00699.x [PubMed: 16108983]

Estes A, Yoder P, McEachin J, Hellemann G, Munson J, Greenson J, Rocha M, Gardner E, & Rogers SJ (2021). The effect of early autism intervention on parental sense of efficacy in a randomized trial depends on the initial level of parent stress. Autism, 25(7), 1924–1934. 10.1177/13623613211005613 [PubMed: 33858234]

Feinberg E, Augustyn M, Fitzgerald E, Sandler J, Ferreira-Cesar Suarez Z, Chen N, Cabral H, Beardslee W, & Silverstein M (2014). Improving Maternal Mental Health After a Child's Diagnosis of Autism Spectrum Disorder: Results From a Randomized Clinical Trial. JAMA Pediatrics, 168(1), 40. 10.1001/jamapediatrics.2013.3445 [PubMed: 24217336]

Geraci M (2014). Linear Quantile Mixed Models: The lqmm Package for Laplace Quantile Regression. Journal of Statistical Software, 57(13). 10.18637/jss.v057.i13

Geraci M, & Bottai M (2007). Quantile regression for longitudinal data using the asymmetric Laplace distribution. Biostatistics, 8(1), 140–154. 10.1093/biostatistics/kxj039 [PubMed: 16636139]

Geraci M, & Bottai M (2014). Linear quantile mixed models. Statistics and Computing, 24(3), 461–479. 10.1007/s11222-013-9381-9

Hamilton EB (1980). The relationship of maternal patterns of stress, coping, and support to quality of early infant-mother attachment. Unpublished doctoral dissertation, University of Michigan.

Hayes SA, & Watson SL (2013). The Impact of Parenting Stress: A Meta-analysis of Studies Comparing the Experience of Parenting Stress in Parents of Children With and Without Autism Spectrum Disorder. Journal of Autism and Developmental Disorders, 43(3), 629–642. 10.1007/s10803-012-1604-y [PubMed: 22790429]

Hoffman CD, Sweeney DP, Hodge D, Lopez-Wagner MC, & Looney L (2009). Parenting Stress and Closeness: Mothers of Typically Developing Children and Mothers of Children With sAutism. Focus on Autism and Other Developmental Disabilities, 24(3), 178–187. 10.1177/1088357609338715

Kasari C, Freeman S, & Paparella T (2006). Joint attention and symbolic play in young children with autism: A randomized controlled intervention study. Journal of Child Psychology and Psychiatry, 47(6), 611–620. 10.1111/j.1469-7610.2005.01567.x [PubMed: 16712638]

Kasari C, Gulsrud A, Freeman S, Paparella T, & Hellemann G (2012). Longitudinal Follow Up of Children with Autism Receiving Targeted Interventions on Joint Attention and Play RH = Targeted Interventions on Joint Attention and Play. Journal of the American Academy of Child and Adolescent Psychiatry, 51(5), 487–495. 10.1016/j.jaac.2012.02.019 [PubMed: 22525955]

Kasari C, Gulsrud A, Paparella T, Hellemann G, & Berry K (2015). Randomized comparative efficacy study of parent-mediated interventions for toddlers with autism. Journal of Consulting and Clinical Psychology, 83(3), 554–563. 10.1037/a0039080 [PubMed: 25822242]

Kasari C, Gulsrud AC, Wong C, Kwon S, & Locke J (2010). Randomized Controlled Caregiver Mediated Joint Engagement Intervention for Toddlers with Autism. Journal of Autism and Developmental Disorders, 40(9), 1045–1056. 10.1007/s10803-010-0955-5 [PubMed: 20145986]

Kasari C, Kaiser A, Goods K, Nietfeld J, Mathy P, Landa R, Murphy S, & Almirall D (2014). Communication Interventions for Minimally Verbal Children With Autism: Sequential Multiple Assignment Randomized Trial. Journal of the American Academy of Child and Adolescent Psychiatry, 53(6), 635–646. 10.1016/j.jaac.2014.01.019 [PubMed: 24839882]

Kasari C, Paparella T, Freeman S, & Jahromi LB (2008). Language outcome in autism: Randomized comparison of joint attention and play interventions. Journal of Consulting and Clinical Psychology, 76(1), 125–137. 10.1037/0022-006X.76.1.125 [PubMed: 18229990]

Kasari C, & Sigman M (1997). Linking parental perceptions to interactions in young children with autism. Journal of Autism and Developmental Disorders, 27(1), 39–57. [PubMed: 9018581]

Keen D, Couzens D, Muspratt S, & Rodger S (2010). The effects of a parent-focused intervention for children with a recent diagnosis of autism spectrum disorder on parenting stress and competence. Research in Autism Spectrum Disorders, 4(2), 229–241. 10.1016/j.rasd.2009.09.009

Koenker R, & Bassett G (1978). Regression Quantiles. Econometrica, 46(1), 33–50. JSTOR. 10.2307/1913643

Lord C, Risi S, Lambrecht L, Cook EH Jr, Leventhal BL, DiLavore PC, Pickles A, & Rutter M (2000). The autism diagnostic observation schedule-generic: a standard measure of social and communication deficits associated with the spectrum of autism. Journal of autism and developmental disorders, 30(3), 205–223. 10.1023/A:1005592401947 [PubMed: 11055457]

Lord C, Storoschuk S, Rutter M, & Pickles A (1993). Using the ADI-R to diagnose autism in preschool children. Infant Mental Health Journal, 14(3), 234–252. 10.1002/1097-0355(199323)14:3<234::AID-IMHJ2280140308>3.0.CO;2-F

Loyd BH, & Abidin RR (1985). Revision of the Parenting Stress Index. Journal of Pediatric Psychology, 10(2), 169–177. 10.1093/jpepsy/10.2.169 [PubMed: 4020601]

Oono IP, Honey EJ, & McConachie H (2013). Parent-mediated early intervention for young children with autism spectrum disorders (ASD). Evidence-Based Child Health: A Cochrane Review Journal, 8(6), 2380–2479. 10.1002/ebch.1952

Osborne LA, McHugh L, Saunders J, & Reed P (2008). Parenting Stress Reduces the Effectiveness of Early Teaching Interventions for Autistic Spectrum Disorders. Journal of Autism and Developmental Disorders, 38(6), 1092–1103. 10.1007/s10803-007-0497-7 [PubMed: 18027079]

R Core Team (2019). R: A Language and environment for statistical computing. (Version 3.6) [Computer software]. Retrieved from https://cran.r-project.org/.

Schieve LA, Blumberg SJ, Rice C, Visser SN, & Boyle C (2007). The Relationship Between Autism and Parenting Stress. Pediatrics, 119(Supplement 1), S114–S121. 10.1542/peds.2006-2089Q [PubMed: 17272578]

Stuart M, & McGrew JH (2009). Caregiver burden after receiving a diagnosis of an autism spectrum disorder. Research in Autism Spectrum Disorders, 3(1), 86–97. 10.1016/j.rasd.2008.04.006

The jamovi project (2020). jamovi. (Version 1.2) [Computer Software]. Retrieved from https://www.jamovi.org.

Tomasello M (1995). Joint attention as social cognition. In Moore C & Dunham PJ (Eds.), Joint attention: Its origins and role in development (pp. 103–130). Hillsdale, NJ: Erlbaum

Tonge B, Brereton A, Kiomall M, Mackinnon A, King N, & Rinehart N (2006). Effects on Parental Mental Health of an Education and Skills Training Program for Parents of Young Children With Autism: A Randomized Controlled Trial. Journal of the American Academy of Child & Adolescent Psychiatry, 45(5), 561–569. 10.1097/01.chi.0000205701.48324.26 [PubMed: 16670650]

Vygotsky L (1978). Mind in Society: Development of Higher Psychological Processes (Cole M, Jolm-Steiner V, Scribner S, & Souberman E, Eds.). Cambridge, Massachusetts; London, England: Harvard University Press.

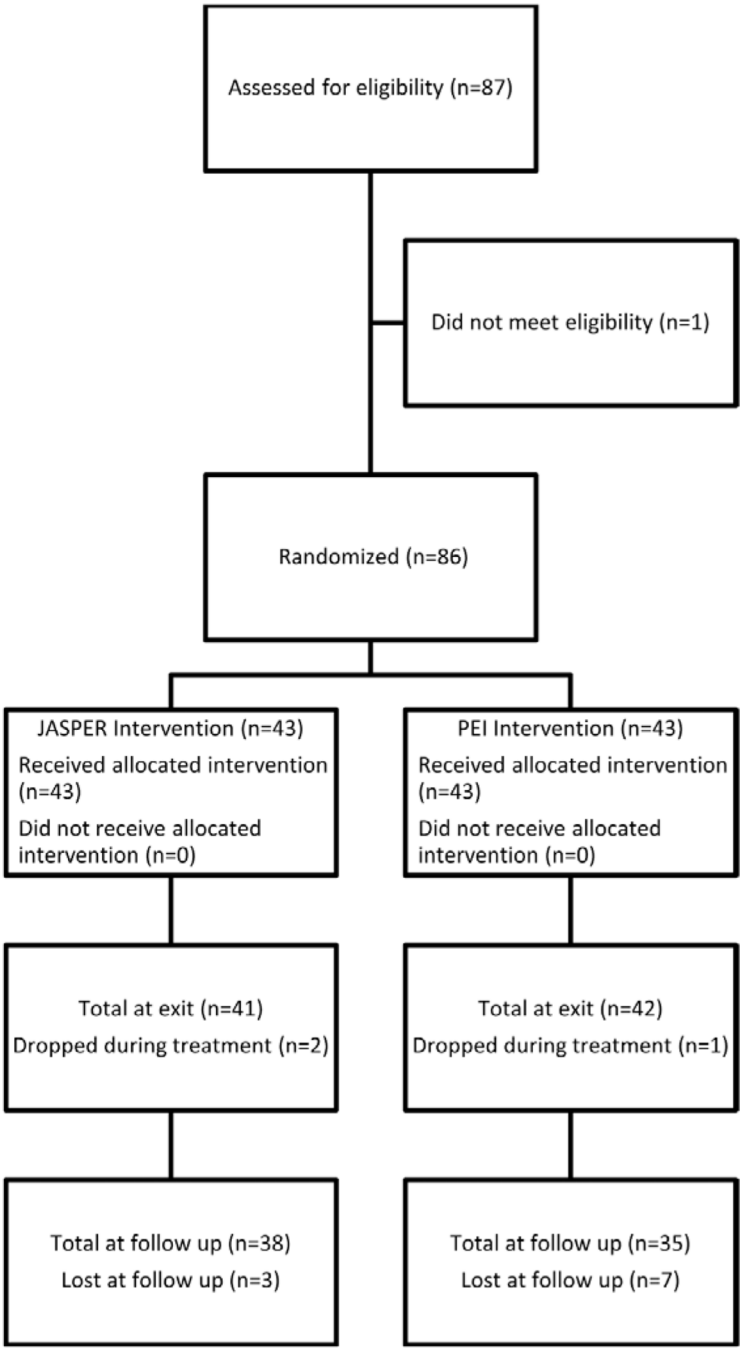

Figure 1.
Recruitment Flow Diagram

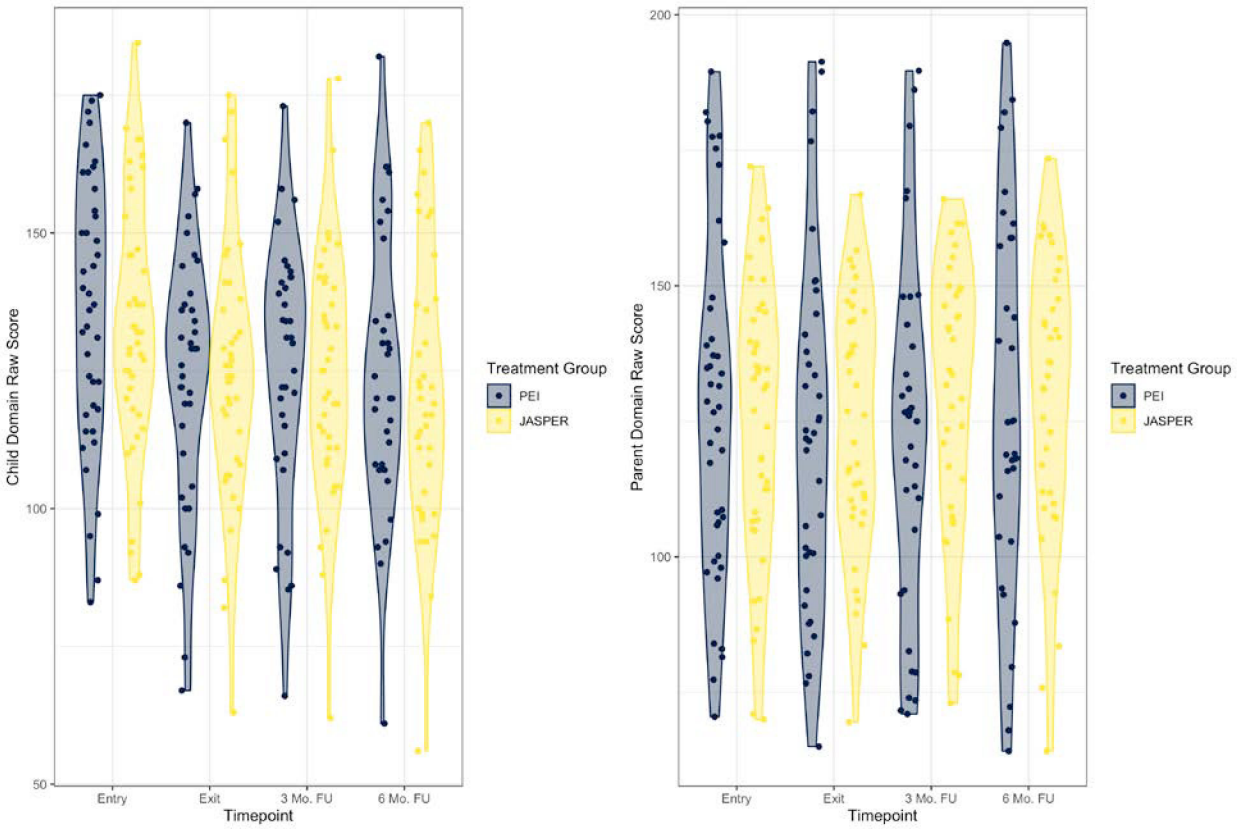

Figure 2.

Distribution of Child and Parent Domain Raw Stress Scores by Timepoint and Treatment Group

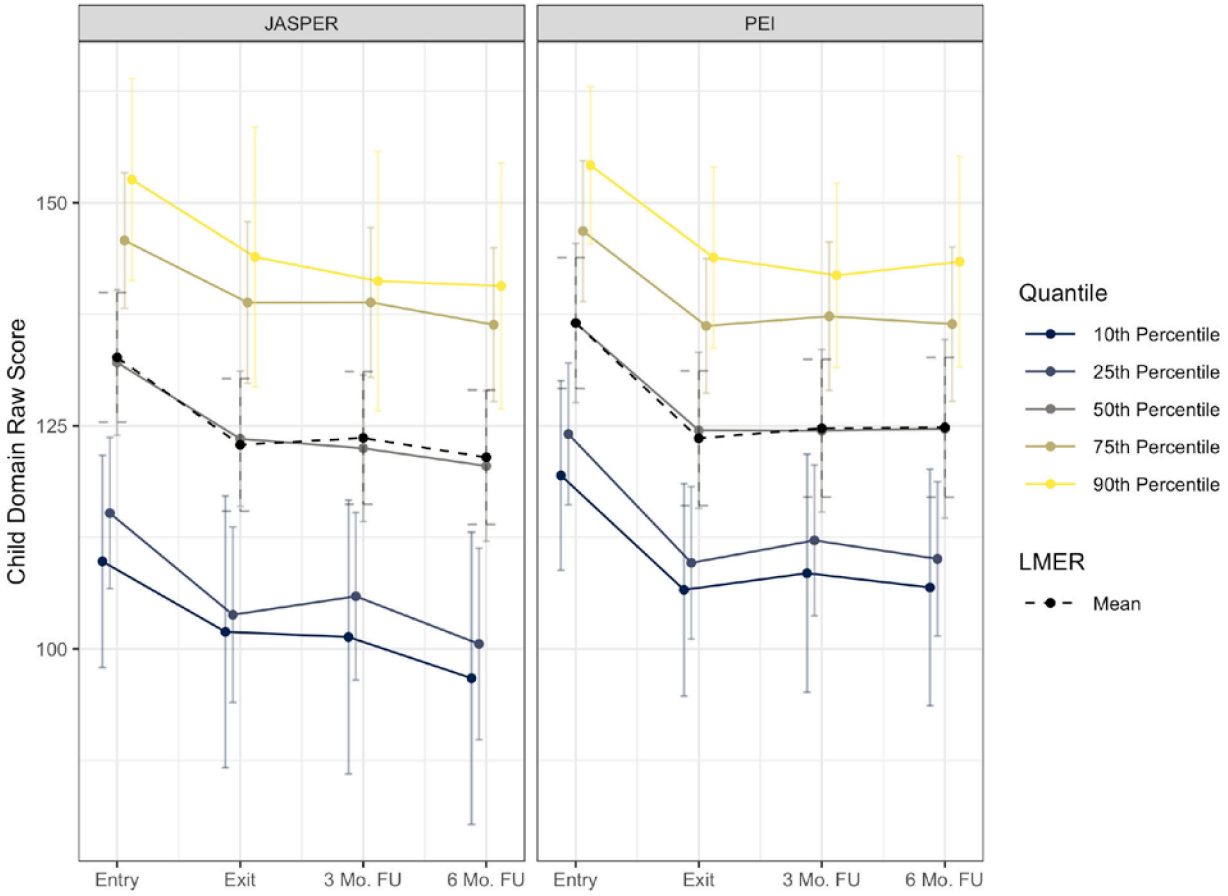

Figure 3.
Child Domain Stress Scores by Treatment Group Over Time

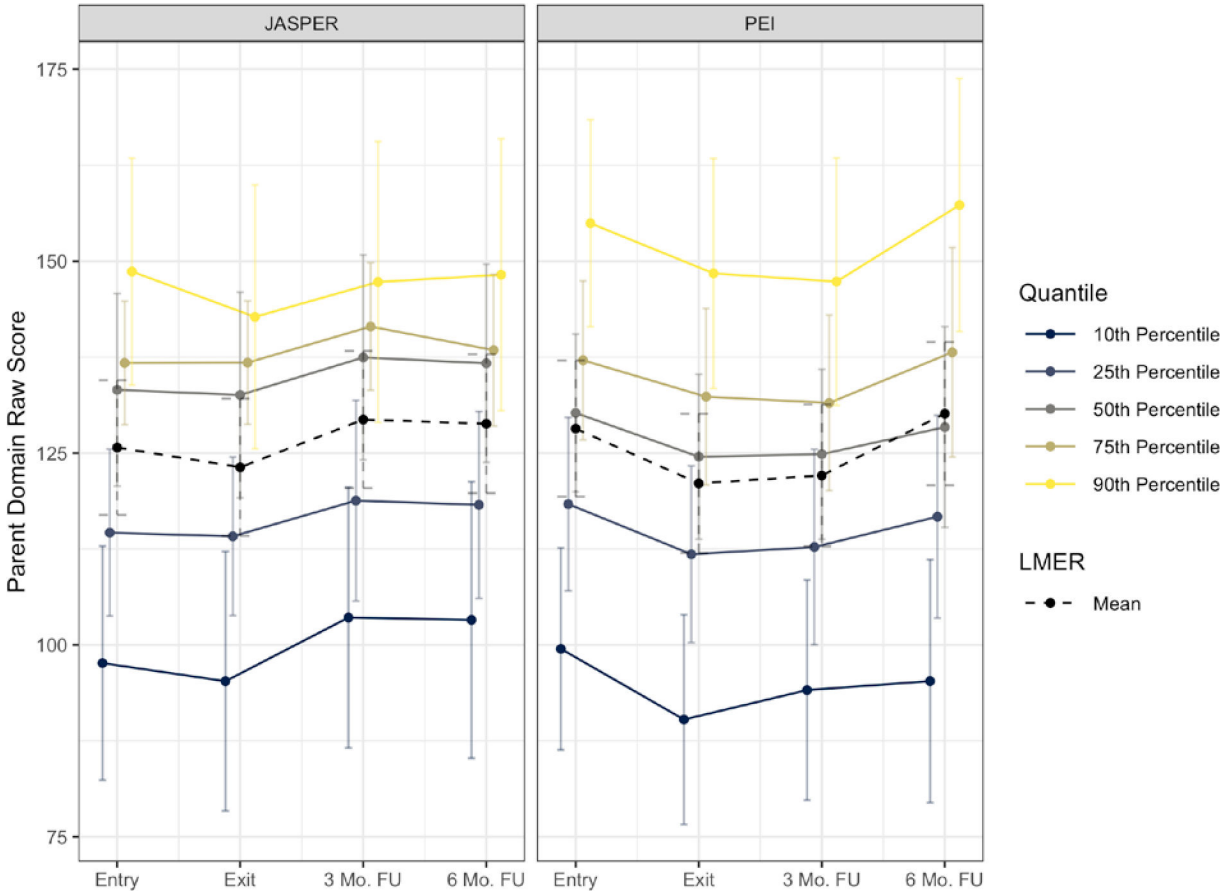

Figure 4.
Parent Domain Stress Scores by Treatment Group Over Time

Table 1

Child and Parent Demographics (N = 86)

Child Characte ristics N (%)	JASPER N=43	PEI N=43	Total	Test	*p*
Chronological Age (Months): Mean (SD)	30.7 (3.5)	32.3 (2.7)	31.5 (3.2)	F(1,84)=6.3	.01*
Gender					
Female	8 (19%)	8 (19%)	16 (19%)		
Race/Ethnicity					
African American	0 (0%)	2 (5%)	2 (2%)		
Caucasian	27 (63%)	26 (60%)	53 (61%)		
Hispanic	3 (7%)	4 (9%)	7 (8%)		
Asian/Pacific Islander	4 (9%)	6 (14%)	10 (12%)		
Other	9 (21%)	5 (12%)	14 (17%)		
Mullen Age Equivalency (Months)					
Developmental Quotient: Mean (SD)	68.0 (20.3)	68.1 (20.6)	68.0 (20.3)	F(1,84)=0.0	.98
Age of Mother	36.9 (4.4)	34.9 (4.7)	35.9 (4.6)	F(1,83)=3.9	.05
Maternal Education					
Years of Education	17.2 (2.3)	16.4 (2.6)	16.8 (2.4)	F (1,84)=2.6	.11

Note

* $p < 0.05$.

Table 2

Total Parenting Stress Raw Scores by Timepoint and Treatment Group

Timepoint	Treatment Group	Mean	Median	Min	Max	SD
Entry	JASPER	259	266	158	347	42.7
	PEI	264	266	154	350	50.1
Exit	JASPER	249	249	155	329	41.8
	PEI	246	247	144	349	47.6
3-month follow-up	JASPER	254	261	141	340	43.0
	PEI	248	251	140	346	50.6
6-month follow-up	JASPER	251	245	132	335	45.7
	PEI	253	257	129	351	55.8

Table 3

Estimates of Effect for Child Domain Stress Using Linear Quantile Mixed Effect Models

Time	τ	Estimate	SE	95% LB	95% UB	p-value
Entry-Exit						
	0.10	4.93	4.89	−4.67	14.53	0.31
	0.25	3.04	4.78	−6.34	12.43	0.52
	0.50	3.49	4.56	−5.47	12.44	0.45
	0.75	3.67	4.70	−5.55	12.89	0.43
	0.90	1.67	4.73	−7.61	10.94	0.72
Entry-3 Mo.						
	0.10	2.49	5.42	−8.15	13.12	0.65
	0.25	2.59	5.18	−7.57	12.75	0.62
	0.50	2.47	4.96	−7.26	12.19	0.62
	0.75	2.60	4.98	−7.16	12.37	0.60
	0.90	0.96	5.12	−9.08	11.00	0.85
Entry-6 Mo.						
	0.10	−0.53	6.13	−12.56	11.50	0.93
	0.25	−0.67	6.00	−12.44	11.11	0.91
	0.50	0.26	5.70	−10.92	11.45	0.96
	0.75	0.98	5.81	−10.41	12.38	0.87
	0.90	−1.10	6.07	−13.00	10.81	0.86

Note

$^{*}p \leq 0.05$.

Table 4

Estimates of Treatment Effect for Parental Stress Using Linear Quantile Mixed Effect Models

Time	τ	Estimate	SE	95% LB	95% UB	p-value
Entry-Exit						
	0.10	6.84	6.16	−5.24	18.92	0.27
	0.25	6.06	5.67	−5.07	17.19	0.29
	0.50	5.04	5.73	−6.21	16.29	0.38
	0.75	4.78	5.54	−6.09	15.65	0.39
	0.9	0.61	5.74	−10.66	11.87	0.92
Entry-3 Mo.						
	0.10	11.30	5.00	1.48	21.12	0.02*
	0.25	9.78	4.97	0.02	19.53	0.04*
	0.50	9.59	4.99	−0.19	19.38	0.05*
	0.75	10.31	5.00	0.49	20.12	0.04*
	0.90	6.23	5.26	−4.09	16.55	0.24
Entry-6 Mo.						
	0.10	9.81	5.20	−0.38	20.01	0.06
	0.25	5.27	5.00	−4.53	15.08	0.29
	0.5	5.31	5.13	−4.75	15.37	0.30
	0.75	0.63	5.28	−9.73	10.99	0.90
	0.90	−2.80	5.37	−13.34	7.75	0.60

Note

* $p \leq 0.05$

Table 4 outlines estimates of the parent domain model. Each estimate corresponds to the difference in slope of parental stress raw score for JASPER vs. PEI group from entry to either exit, 3-month follow-up, or 6-month follow-up. Positive values indicate an increase in parental stress. τ is the quantile of the distribution.