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Physiological Correlates of Maternal Responsivity in Mothers of Preschoolers with Fragile X Syndrome

Abstract

The present study examined the relationship between salivary cortisol and maternal responsiveness in mothers of boys with fragile X syndrome (FXS). Maternal responsivity is strongly associated with child outcomes, and children with FXS are at risk for compromised development due to intellectual disabilities and problem behavior. Increased understanding of the nature and underlying mechanisms of maternal responsivity in FXS is important to optimize outcomes in children with FXS and contribute to improved family cohesion. Data from 36 mother-child dyads indicated a complex age effect with elevated cortisol levels associated with high maternal responsivity scores when children are young and low responsivity scores when children are older. Implications of findings and directions for future research are discussed.

Keywords

fragile X syndrome; maternal responsivity; parenting behaviors; physiological arousal; salivary cortisol

The present study examined physiological correlates of maternal responsivity in mothers of young boys with fragile X syndrome (FXS). Fragile X syndrome is the leading known inherited genetic cause of autism and intellectual disabilities, affecting approximately 1:4000 males. Consistent with Sameroff's transactional model (Sameroff, 1975), developmental outcomes in children with FXS are affected by multiple factors including endogenous and exogenous child, parent, and environmental factors, that interact and vary over time.

Maternal Responsivity and Child Outcomes

Evidence clearly establishes that higher levels of maternal responsivity are associated with improved social, emotional, communication, and cognitive competence in young children with and without disabilities. Further, the literature documents a relationship between less responsive parenting and slower skill development (Landry, Smith & Swank, 2006; Warren, Brady, Sterling, Fleming, & Marquis, 2010). Research on maternal responsiveness in populations with disabilities suggests that frequent and positive maternal-child interactions are positively related to cognitive, social-emotional, and communicative functioning (Brady, Warren, Fleming, Keller & Sterling, 2014; Hauser-Cram, Warfield, Shonkoff, & Krauss, 2001; Landry, Smith, Swank, Assel, & Vellet, 2001; Sterling, Warren, Brady, & Fleming, 2013; Warren et al., 2010). The most clearly established parental factors associated with a reduction in parental responsivity are affective conditions such as stress, anxiety, and depression (Essex, Marjorie, Cho, & Kalin, 2002; Sarimski, 1997).

Specific to child factors, developmental research shows that child age influences parental stress and parenting behaviors across time. While some studies have shown that parents of young children report higher levels of stress (Barker, Oliver, Viding, Salekin, & Maughan, 2011; Gray, 2002) that increase with child age (Tehee, Honan, & Hevey, 2009), other work suggests a decline in parental stress levels over time as child problem behavior improves (Bopp, 2007). Furthermore, Orr and colleagues (1993) found a curvilinear relationship between child age and stress in mothers of children with developmental delays, suggesting that maternal stress was highest during middle childhood (6-12 years of age) when compared to preschool and adolescence (2-5 years and 13-18 years, respectively). Thus, the relationship of parental stress and responsivity to the developmental stage of the child appears complex, likely reflecting multiple co-occurring and dynamic factors.

Studies examining the relationship between parental stress and parenting behavior increasingly recognize the contribution of biomarkers to the understanding of this complex relationship (Foody, James, & Leader, 2014; Hartley et al., 2012). Salivary cortisol is reflective of the hypothalamic–pituitary–adrenal (HPA) system and is a physiological marker of stress. High levels of salivary cortisol are correlated with experienced stress (Hackman et al., 2013) and blunted cortisol responsivity has been shown to signal social and behavioral problems (Ouellet-Morin et al., 2011). Research suggests that both high and low baseline cortisol levels are related to insensitive and negative parenting behaviors, implying that these behaviors are associated with physiological indicators of stress (Giardino, Gonzalez, Steiner, & Fleming, 2008; Kiel & Buss, 2013; Martorell & Bugental, 2006; Mills-Koonce et al., 2009; Schechter et al., 2004). In research on mothers of children with difficult temperaments, high rates of harsh and intrusive parenting are correlated with elevated parental cortisol reactivity (Kiel & Buss, 2013; Martorell & Bugental, 2006). However, intrusive and insensitive parenting is also correlated with lower cortisol levels in mothers with post-traumatic stress disorder (Schechter et al., 2004).

Fragile X Syndrome

Fragile X syndrome is one of several disorders (Chonchaiya, Schneider, & Hagerman, 2009) involving *FMRI* gene dysfunction. The full mutation (>200 CGG repeats) in males is linked to intellectual disabilities and problem behaviors including autism, anxiety and attention deficits (Roberts et al., 2009b; Sarimski, 2010; Woodcock, Oliver, & Humphreys, 2009). The premutation (55 – 199 CGG repeats) is common in females with up to 1:150 in the general community having the *FMRI* premutation (Seltzer et al., 2011). A subset of women with the *FMRI* premutation appear at risk for stress and psychopathology with 30% of mothers with the *FMRI* premutation falling in the clinically significant range for parenting stress (Bailey, Sideris, Roberts, & Hatton, 2008), and 57% meeting DSM-IV diagnostic criteria for a disorder (Roberts et al., 2009a). Contributing factors associated with higher levels of parenting stress and psychopathology include child problem behavior, negative life events, marital status, social support, and genetic markers (i.e., activation ratio and CGG repeat length) (Hartley et al., 2012; Roberts et al., 2009a; Seltzer et al., 2011).

Using a diathesis-stress model, recent evidence indicates that mothers with the *FMRI* premutation display a suppressed awakening cortisol expression in response to elevated

child problem behavior that is associated with a lower activation ratio (Hartley et al., 2012). Likewise, CGG repeat length and maternal age may influence the relationship between the cortisol awakening response, the experience of negative life events, and psychopathology (i.e., depressive symptoms and anxiety) in mothers of children with FXS (Seltzer et al., 2011). These studies suggest that the caregiving experience of mothers with the *FMRI* premutation is complex, involving endogenous genetic and physiological factors affected by child behavior. Given evidence that responsive parenting in mothers with the *FMRI* premutation is associated with better behavior regulation and language outcomes in young boys with FXS (Wheeler et al., 2010; Warren et al., 2010), increased understanding of the mechanisms associated with maternal responsivity in mothers with the *FMRI* premutation may be important for enhancing responsivity associated with positive child outcomes. This enhancement may be particularly salient during the early childhood years when early intervention is the most effective.

Present Study

To date, there are no studies investigating physiological correlates of maternal responsivity in mothers of young children during experimentally derived mother-child interactions in FXS or any other developmental disability. The current study represents an examination of extant data available as part of a larger research project focused on family adaptation to FXS. While this study is considered preliminary, we believe it contributes to the literature by highlighting the importance of integrating biomarkers and serving to generate hypotheses for future, more detailed studies of this nature.

The over-arching aim of this study is to examine salivary cortisol as a physiological correlate of maternal responsivity in mothers of young boys with FXS. Given that a typical cortisol levels are related to sub-optimal parenting behaviors in typically developing children (Giardino et al., 2008; Mills-Koonce, et al., 2009), it is hypothesized that salivary cortisol levels will be associated with maternal responsivity scores. Additionally, based on the trajectories of parenting stress across age groups, it is hypothesized that child age will influence the relationship between physiological arousal and maternal parenting behaviors (Bopp, 2007; Orr, Cameron, Dobson, & Day, 1993).

Method

Participants

Participants included 36 mother-child dyads recruited through parent list serves, a research registry, and collaboration with studies at the University of North Carolina - Chapel Hill. All 36 mothers had the *FMRI* premutation and a male son with FXS. The mean maternal IQ was 110 (range: 73 – 130). Annual household income was reported by parents and low SES was defined as falling below the 200% poverty guideline (www.aspe.hh.gov/poverty/). Race, education and income are represented in Table 1. Participants completed one (n=18), two (n=16), or three (n=2) assessments across 18-month intervals as part of the larger research project, resulting in 56 observations. Mean ages of children at assessment time points were 30 months at Time 1 (range = 11– 40 months), 46 months at Time 2 (range = 29 – 63

months), and 63 months at Time 3 (range = 40 – 68 months). Given the exploratory nature of this study, we included all participants for whom there were complete data.

Measures

Measures used in this study included maternal responsivity, salivary cortisol, child age, parental stress, child problem behavior, and maternal medication history. Data were collected concurrently for each measure at each assessment period.

Maternal Responsivity—Maternal responsivity scores were derived from 25-minute maternal-child interactions across four activities including reading a book, making and eating a snack, an unstructured play with the child's choice of toys, and an extended interaction period where the mother-child dyad participated in an everyday activity (i.e., laundry, cleaning, or playing a game). See Warren et al. (2010) for a detailed description of data collection, coding, and reliability procedures. Scores on the maternal responsivity composite were calculated based on frequencies of specific maternal behaviors including gesture use, requests for verbal complies, comments, and recodes. A recode was defined as a maternal interpretation of the child's attempt to communicate. For example, after the child says “ju”, the mother may say “Do you want your juice?”.

Salivary Cortisol—Although circadian effects could not be controlled for due to lack of data on morning rise times for each participant, a standardized start (9:00 AM) and stop time (12:00 PM) was implemented across the sample. Also, Pearson correlations indicated no relationship between time of cortisol collected and cortisol for baseline and reactivity ($p > .05$). Cortisol was collected via cotton salivettes at two time points during each assessment. The pre-assessment baseline ($N = 56$) occurred within 15 minutes of the onset of the assessment ($M = .30$, $SD = .14$). The reactant cortisol sample ($N = 41$) was taken within 15 minutes of the end of the 3 hour assessment ($M = .17$, $SD = .09$). A cortisol change score ($N = 41$), a measure of regulated cortisol, was calculated by subtracting the baseline score from the reaction score ($M = -.14$, $SD = .12$).

The pre-assessment baseline cortisol value is considered indicative of the mother's emotional state prior to the challenge of completing the assessment. The reactivity score reflects the mother's response to the assessment process, which included the mother-child interactions. We recognize that the baseline value reflects the mother's anticipation of the assessment so it is not a “pure” baseline independent of context. The calculated change score allows for an index of the magnitude of the within-person variation in cortisol level resulting from the “challenge” of completing the research assessment, which is generally consistent with existing work examining HPA axis reactivity to stressful procedures (Dickerson & Kemeny, 2004; Gunnar, Talge, & Herrera, 2009).

Missing reactant and change ($n=15$) cortisol data were due to failure to collect the second sample, insufficient saliva for the analyses, or errors in saliva collection. The salivettes were processed using Salimetrics' Salivary Cortisol Enzyme Immunoassay kit (EIA) and levels were reported in micrograms/deciliter. Due to the non-normal distribution of the cortisol, the data were log transformed.

Child Problem Behavior—Problem behavior was assessed using The Child Behavior Checklist (Achenbach, 1991), a valid and reliable parent-report measure that has been used to assess behavioral and emotional difficulties in children with the full mutation (see Bailey et al., 2008). For the current study, T-scores on the internalizing, externalizing, and total problem behavior domains were used to examine if child problem behavior was associated with the relationship between maternal responsivity and cortisol levels.

Parenting Stress, Child Age, and Maternal Medication History—Mothers completed the Parenting Stress Inventory (PSI; Abidin, 1995) at each assessment time point to allow for an examination of the relationship between self-reported stress and physiological arousal. Child's age in months and maternal medication history were also reported by the child's mother at each assessment and confirmed by the experimenters. Medication was included as a covariate to control for the potential effects on the behavioral expression of maternal responsivity as well as physiological functioning. Of the mothers included in this study, 25% were on medication for symptoms of anxiety or depression for at least one assessment time point.

Procedure

Individual assessments were conducted in the families' home over a period of two days to allow the parent and child to acclimate to the procedures and examiners. On day 1, maternal responsivity data for the book reading, snack preparation, and unstructured play tasks were collected. On day 2, cortisol data and the unstructured child-maternal interaction were collected in addition to completing a developmental assessment with the child and multiple behavioral paradigms such as an experimental press of temperament.

Results

Initial Pearson correlations indicated that maternal medication history and total parenting stress were not related to maternal responsivity or cortisol scores so the variables were dropped from follow-up analyses. Eighteen mothers had a single cortisol data point while 16 mothers had two data points and 2 mothers had 3 data points for a total of 56 assessments. Interclass correlations were examined and values indicated dependence among observations from the same participant (i.e., ICC = .19; Srivastava & Keen, 1988). Because participants were assessed multiple times, observations were nested within individuals so multilevel models were used. Specifically, we included a random intercept to the model to control for within participant variability across observations. In the absence of this correction, the assumption of independent of errors using ordinary least squares regression is not reasonable.

Three multilevel models were tested wherein maternal responsivity scores were associated with baseline cortisol, reactant cortisol, and regulation cortisol. Child age was included as a covariate in all models. The models examining reactant and regulation cortisol were not significant. The model containing baseline cortisol indicated an interaction with child age, $B = -.12(.05)$, $t = -2.53$, $p < .05$. Details on model parameters are included in Table 2.

Due to overlap in child age across assessment time points, this interaction was probed using age quartile splits, in which the data were separated into four age quartiles by child age (quartile 1 = 11-35 months (n=19), quartile 2 = 36-48 months (n=16), quartile 3 = 49-58 months (n=10), quartile 4 = 59-76 months (n=13)) and four bivariate regression models with maternal the relationship of baseline cortisol to maternal responsivity was examined. Results indicated a positive relationship of maternal responsivity in the first quartile, $B=2.78(.98)$, $t=2.83$, $p<.05$, with a negative relationship in the fourth quartile that approached significance, $B=-2.36(1.11)$, $t=-2.13$, $p=.06$, indicating a reversal of the effect. Refer to Figure 1 for a graph of the regression models of baseline cortisol and maternal responsivity by age quartile. There were no mothers/children represented multiple times within the quartiles with the exception of one pair in quartile 1. While we only had a subset of participants with longitudinal data (n=18), we ran a simple paired t-test and found no change in cortisol over time for these mothers ($p>.05$) with some mothers showing an increase (34%), others a decrease (45%) and some no change (21%) in baseline cortisol over time. To examine the influence of child problem behavior on the relationship between maternal responsivity and cortisol levels, we completed Pearson correlations, which indicated no association ($p>.05$).

Discussion

The goal of this study was a preliminary examination of the physiological correlates of maternal responsivity in mothers with the *FMRI* premutation. Our findings suggest a relationship between maternal responsivity and baseline salivary cortisol that is influenced by child age yet independent of child problem behavior. The results indicate that higher cortisol levels are associated with heightened maternal responsivity when children were three years old and younger; however, elevated cortisol scores are associated with dampened maternal responsivity when children were aged five years and older. One potential reason for the relationship between elevated levels of cortisol and high maternal responsivity with infants and toddler-aged children may be the heightened stress associated with receiving the FXS diagnosis and having a child with a disability (Roberts et al., 2009a; Wheeler et al., 2008). Elevated physiological arousal reflected in baseline cortisol may facilitate activation of heightened maternal responsivity as an initial “early response” to the challenges associated with caring for young children with FXS.

In contrast, our results show that heightened baseline cortisol is associated with reduced maternal responsivity with older school-aged children. Previous research indicates that problem behavior increases over the preschool years in children with FXS (Baranek et al., 2008). Further, the severity of child problem behavior predicts maternal stress, anxiety, and quality of life in mothers who are carriers of fragile X (Bailey et al., 2008; Wheeler et al., 2008), which may contribute to parental stress and influence caregiving behaviors. Of note, child problem behavior was not associated with maternal responsivity in our sample, which may indicate that maternal biological mechanisms have more of an influence on responsivity in mothers of school-aged children with FXS. Also, evidence suggests that these effects likely reflect relationships in a subgroup of families and not the entire sample as a whole. Recent evidence supports these hypotheses with elevated behavioral inhibition in young children with FXS (1 to 6 years of age) associated with higher parenting stress that appears

rooted in maternal genetic factors with a lower activation ratio associated with elevated parenting stress (Tonnsen, Cornish, Wheeler, & Roberts, 2014).

Our results show a profile of dampened cortisol in relation to less maternal responsiveness in a subgroup of mothers of school-aged children. The finding of a maternal dampened cortisol response that is associated with unexpected and non-optimal responses in a subgroup of mothers with the *FMR1* premutation is similar to maternal cortisol profiles identified within this population when examining maternal response to elevated rates of problem behavior in adult-aged children (Hartley et al., 2012). Our findings, however, were independent of child problem behavior. Consistent with work demonstrating a relationship between parental stress and child outcomes, we recently reported that elevated parenting stress in mothers with the *FMR1* premutation was associated with increased anxiety in their young children, which was also associated with the genetic status of the mother (Tonnsen et al., 2014). Additional research is needed to explore the relationship between maternal responsivity and maternal psychopathology, child psychopathology, and genetic status within this population.

Our analyses did not support a relationship between maternal responsivity and regulation or reactivity cortisol. The null findings could reflect low power given that these models had less data. Alternatively, these findings suggest that the relationship between maternal responsivity and HPA axis function may be rooted in the baseline HPA function. This can be viewed as a more state-like condition compared to the reactivity and change scores. Baseline scores are more reliant on the nature of the experimental condition, and may or may not induce a stress response. In our study, the reactivity measure reflected the mother's response to completing a research protocol consisting of a variety of direct child developmental and behavioral measures and maternal-child interaction that were designed and implemented in a way to not be particularly stressful to the participating families. Thus, a more discrete and intense inducement of stress may result in different findings. An alternative hypothesis is that our baseline measure reflected "anticipatory" participation in the research assessment, which could have induced stress for some participants and led to the stress reaction serving as a critical factor.

Finally, no relationship between self-reported parenting stress and salivary cortisol emerged despite finding a relationship between a directly observed measure of parental responsivity and baseline salivary cortisol. This highlights the value of including multiple indices of parental stress, which allows consideration of differing sources and is particularly salient given the criticisms leveled at self-reports (Baker & Brandon, 1990; Morsbach & Prinz, 2006).

Summary and Implications

Results from this preliminary study suggest the possible existence of a complex relationship between maternal responsivity and baseline cortisol expression in mothers of young boys with FXS. This study contributes to previous literature examining parenting behaviors from a biosocial perspective and is the only known study to date to explore this relationship in mothers of boys with FXS or any other developmental disability. Our results provide

support for the inclusion of biomarkers; as such measures may be valuable when examining biobehavioral relationships for complex behaviors like caregiving.

These findings provide preliminary support for a pattern of positive parenting behaviors in mothers with the *FMR1* premutation that may be rooted, in part, in physiological arousal. Although this relationship is clearly complex and likely reflects a subgroup of mothers with the *FMR1* premutation, we highlight the importance of exploring the underlying mechanisms of maternal responsivity from a multidimensional perspective. Such an approach is particularly important given the positive outcomes associated with responsive parenting in children with and without FXS (Warren et al., 2010; Brady et al., 2014). Thus, this study provides initial insight into maternal responsivity in mothers of boys with FXS from a biosocial approach.

If replicated, our findings could have implications for the treatment of mothers and children with disabilities, particularly FXS. For example, Warren and Brady (2007) discuss parental responsivity training programs and conclude that they can effectively increase responsive parenting behaviors. As our data suggest, such parenting behaviors may also affect maternal stress levels and physiological arousal in mothers of children with disabilities.

Limitations and Future Research

This study has a number of limitations. First, because of the lack of a control group, we were unable to discern the degree to which our findings are unique to FXS. Second, we did not have a measure of stressful life events although we did include a measure of maternal total parenting stress, which was not related to maternal responsivity. Third, our sample was one of convenience and was skewed toward mothers with higher educational attainment. Fourth, our failure to control for circadian rhythm effects due to lack of data on wake times for each participant may have influenced baseline cortisol values. Finally, to limit the variables entered into our statistical models, child problem behavior and age were the only child variables included in this study. Future studies should explore the impact of additional child variables on the relationship between arousal and parenting behaviors across time.

Future research should consider examining the effect of maternal responsiveness interventions on physiological arousal, as this could provide important information into a potential causal relationship between parenting behaviors and physiological arousal. If a relationship were discovered between improvement in maternal cortisol levels in conjunction with the increases in maternal responsivity behaviors, such interventions could have potential maternal health benefits in addition to improving children's developmental and behavioral outcomes. This proposed outcome is consistent with recent evidence demonstrating a typical cortisol levels associated with elevated child problem behavior (Hartley et al., 2012).

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Figure 1.

Interaction between child age in months and maternal baseline cortisol predicting maternal responsivity scores. In the youngest age group, elevated levels of baseline cortisol are positively related to higher scores on maternal responsivity. At the oldest age quartile, high cortisol levels are negatively related to maternal responsivity.

Table 1
Descriptive Statistics of Maternal Sample

	N	%
Race/ethnicity		
White	33	91
African American	1	3
Latino	1	3
Other	1	3
Marital status		
Married	32	89
Divorced	2	5
Separated	1	3
Single, never married	1	3
Education		
Less than high school	2	6
High school and some training/college	9	24
College degree	11	31
Some post college or advanced degree	14	39
SES		
Low Income	5	17

Table 2
Baseline, Reactant, and Recovery Cortisol Predicting Maternal Responsivity Scores

Construct	Fixed effect				Random effect variance component	
Model/Variables	Coefficient	SE	t Ratio	σ^2	τ	
Baseline Cortisol						
Intercept	.17	.11	1.53	.21	.11	
Age	.01	.01	1.86		.00	
Medicine	-.15	.25	-.58		.00	
Cortisol	-.86	.89	-.97		12.19	
Cortisol * Age	-.12	.05	-2.53 *			
Reactant Cortisol						
Intercept	.15	.15	1.07	0.49	.00	
Age	.01	.01	.87		.00	
Medicine	-.13	0.31	-.41		.00	
Cortisol	-.23	1.89	-.12		19.40	
Cortisol * Age	-.06	.12	-.50			
Regulation Cortisol						
Intercept	.10	.15	.70	0.33	.12	
Age	0.00	0.01	.05		0.00	
Medicine	.18	.30	.60		.10	
Cortisol	-.07	1.19	-.06		7.46	
Cortisol * Age	0.11	0.06	1.78			

Note:

* p< .05