Title: Understanding the Relationship Between Maternal Use of Verbally Responsive Language and Child Language Development in Fragile X Syndrome: The Impact of Maternal and Child Characteristics

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Introduction: Child spoken language is an important skill that shapes learning, social interactions, and academic success. Therefore, finding ways to support optimal language outcomes in youth with fragile X syndrome (FXS) is particularly important given their extensive delays in multiple domains of language (Abbeduto et al., 2007). Importantly, one such way to foster spoken language development in children is through advanced language input from a communicative partner, such as a parent (Landry et al., 2006). The current study explored the language input to children with FXS from their biological mothers. We hypothesized that maternal input could be less than optimal due to the challenging behaviors and limited skills of the child and the biological vulnerability of the mother to mental health challenges and parenting stress (Sterling et al., 2013). We examined both the relationship between the language used by the mother and metrics of child spoken language and how phenotypic characteristics of FXS commonly associated with the child and/or mother might impact the language from mothers.

Method: A total of 19 mother-child dyads completed the current study with child participants between the ages of 6 and 11 years old. The study employed multimodal methods for data collection in the home, all from a distance using secure video teleconferencing software. To assess the relationship between maternal language input and child spoken language, each mother-child dyad completed the telling of a wordless picture book together. These interactions were video recorded and later transcribed to derive the number of child story-related utterances, number of different words used by the child, and child mean length of utterance. Transcripts were also coded using a scheme adapted from Warren et al (2010), which codes for maternal language input that is more responsive in nature (i.e., using open-ended questions and expanding upon child utterances) as well input that was more consistent with behavior management (i.e., redirecting the child's focus of attention and asking the child to comply behaviorally). The former is thought to facilitate child language development. Child characteristics were assessed through the Child Behavior Checklist (CBCL) as a measure of child challenging behaviors, the Vineland-3 interview as a proxy for child developmental level, and the Childhood Autism Rating Scale-2 (CARS-2) coded from the mother-child interactions as a measure of autism symptom severity. For maternal characteristics, mothers self-reported on their mental health status, namely anxiety and depression levels, using the Symptom Checklist-90-Revised (SCL-90-R) and their feelings of parenting stress using the Parenting Stress Index-4 (PSI-4). We also included a more objective measure of stress by collecting the mother's heart rate variability (HRV) as detected through a wearable wristband.

Results: A series of Spearman correlations were conducted to assess the associations between maternal language input and child spoken language. Maternal responsive language was positively associated with child story-related talking and number of words used, whereas maternal behavior management input was negatively associated with child mean length of utterance. These relationships were moderated by child age in some cases. Next, we computed Spearman correlations to address the relationship between child and maternal characteristics on maternal language input. There were no significant associations between child characteristics, such as challenging behaviors or autism symptom severity, and maternal language input. Further, although the mothers had mental health and parenting stress profiles that were significantly higher than that of the general population, only maternal HRV collected just prior to the mother-child interaction was significantly associated with maternal language input during the interaction. Specifically, baseline HRV was positively associated with the subsequent use of verbally responsive language.

Discussion: Consistent with previous research, the present results confirmed the important relationship between maternal verbal input and child spoken language. Previous research, however, has typically focused on young children. Thus, findings from the present study suggest maternal responsivity is important for language development in school-age children with FXS as well. Further, we did find a significant association between baseline HRV and maternal responsive language, which is consistent with other physiological indices of stress such as cortisol being related to maternal language input in mothers of children with FXS

(Robinson et al., 2016). Given the association between HRV and overall responsiveness to one's environment (Thayer, & Brosschot, 2005), these findings could be indicative of our mother's readiness to engage with or respond to their child's communication acts. Findings from this study provide novel pathways for future clinical interventions.

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