Title: Family and Caregiver Characteristics Contribute to Caregiver Uptake of Strategies and Growth in Child Spoken Language in a Parent-Implemented Language Intervention in Fragile X Syndrome

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Introduction: Fragile X syndrome (FXS) is the leading inherited cause of intellectual disability (Hagerman et al., 2017) and is often characterized by significant delays across multiple domains of language (Abbeduto, Brady, & Kover, 2007). Therefore, there is a need for interventions focused on optimizing child language outcomes. One way to provide meaningful and prolonged treatment for these individuals is through parent-implemented interventions in which parents take on the role of a clinician. Distance teleconferencing can be utilized to deliver parent-implemented interventions, which increases the frequency and ease of training for parents without adding the burden of travel. Maximizing the efficacy of such interventions, however, may require additional support for parents to address factors such as parenting stress (Stahmer & Pellecchia, 2015). Further, biological mothers of individuals with FXS are at a higher risk of experiencing psychiatric disorders such as anxiety and depression compared to the general population (Bourgeois et al., 2011), suggesting an even greater need to consider maternal needs in intervention delivery in this population. Therefore, the present study focused on how family factors might relate to caregiver uptake of strategies and ultimately child growth in spoken language over the course of a parent-implemented language intervention (PILI) across four different treatment conditions. Such data are needed to develop comprehensive family system-focused interventions.

Method: A total of 47 parent-child dyads participated in two different studies looking at the benefits of a distance-delivered PILI in children with FXS between the ages of 10 and 17 years. The first study involved 19 parent-child dyads with 9 dyads randomly assigned to a treatment-as-usual group and 10 to the PILI group (described in McDuffie et al., 2018). For the second study, 28 parent-child dyads participated in a small-scale randomized controlled clinical trial during which all the parent-child dyads received PILI in addition to either a placebo pill (N=16) or the pharmaceutical, lovastatin (N=12). In both studies, PILI involved the parent and child telling the story from a wordless picture book while the parent was trained to use three primary strategies: (1) asking open-ended questions; (2) using intonation prompts; and (3) expanding upon child utterances. Parent use of these strategies was expected to lead to improvements in the child’s lexical diversity, grammatical complexity, and overall story-related talking. The family factors assessed immediately prior to starting intervention were ratings from the Parenting Stress Index, Fourth Edition Short Form (PSI-4-SF), Symptom Checklist-90-Revised (SCL-90-R), and the Parenting Sense of Competence scale (PSOC). The PSI-4-SF includes scores across three domains of parenting stress, including parental distress, parent-child dysfunctional interaction, and difficult child, as well as a measure of total parenting stress. The SCL-90-R measures psychological problems and symptoms of psychopathology; scores from the anxiety and depression domains were included in this study. The PSOC is self-report questionnaire measuring parenting efficacy and competence.

Results: All children who received PILI showed significant gains in their lexical diversity when compared to those who did not. Further, children who received PILI only or PILI + lovastatin, increased their overall story-related talking when compared to children who did not receive PILI, whereas children who received PILI + placebo were only marginally higher than those who did not receive PILI. With regard to parental uptake of strategies, all parents who received PILI showed significant increases in all of the targeted intervention strategies as well as in their overall story-related talking (i.e., total strategy use plus story-related comments) when compared to parents who did not receive PILI. Further, parents in the PILI only group had significantly greater improvements in their use of expansions when compared to parents who received PILI + placebo.

Next, we looked at potential barriers to treatment gains in this population by examining correlations between caregiver well-being and parenting stress prior to starting the intervention, and the change in both the use of parent strategies and child spoken language over the course of the treatment period. First, looking at change in parent strategy use, for the non-PILI group, parent use of open-ended questions was related to the difficult child rating as well as total parenting stress, whereas overall strategy use was related to the difficult child rating. For the PILI only group, parent use of intonation prompts was related to parent self-report of depression, parental distress, and total parenting stress and parent use of expansions and total strategies were related.
to parenting sense of competence. For the PILI + placebo group, parent-child dysfunctional interaction was related to parent use of intonation prompts, total strategy use, and story-related talking, with total parenting stress also relating to total strategy use. Interestingly, for this latter group, relationships indicated that the higher the level of parent-child dysfunctional interaction and total parenting stress, the more the parent used the intervention strategies. Lastly, for the PILI + lovastatin group, parent story related talking and total strategy use were correlated with parent self-report of depression. With regards to change in child performance, for the non-PILI group, changes in child story-related talking and linguistic diversity were related to the difficult child scores of the PSI with child story-related talking also relating to total parenting stress. For the PILI only group, parent self-report of depression, parental distress, and overall sense of parenting competence were significantly correlated with changes in child lexical diversity, with depression also relating to child story-related talking. Different patterns emerged for the PILI + placebo and PILI + lovastatin groups with no significant correlations for the former and difficult child ratings being significantly related to child grammatical complexity in the latter group.

Discussion: Our findings suggest that family factors, in particular parenting stress, influence the outcomes of parent-implemented interventions, affecting not only the uptake of the targeted parenting strategies, but also aspects of child treatment gains. More work is needed to further differentiate what family factors might be most critical to the efficacy of parent implemented interventions and ultimately, how those factors should be addressed within the interventions.

References:

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