Research Brief

A SUMMARY OF A PUBLISHED ARTICLE

Child and Genetic Variables Associated with Maternal Adaptation to Fragile X Syndrome

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FINDINGS FROM FAMILY ADAPTATION TO FXS, A STUDY CONDUCTED AT THE UNIVERSITY OF NORTH CAROLINA AT CHAPEL HILL

Families of children with fragile X syndrome (FXS) often face caretaking challenges for reasons that include: learning and behavior problems present in many children with FXS, stress associated with finding appropriate services, and managing daily routines. Researchers have been particularly interested in the psychological and emotional adaptation of mothers who have children with FXS. Given the inheritance pattern of this disorder, mothers are often carriers of FXS or may have the full mutation themselves. Therefore, they may be dealing with stressors associated with raising an affected child as well as emotional and psychological symptoms associated with having the pre-or full mutation themselves.

Past studies have focused on different emotional and psychological aspects, such as stress, depression, and anxiety disorders in female carriers. Studies have also examined stress in carrier mothers of children with FXS and reported an association between high levels of stress and child behavior problems. Other research studies focusing on depression in female carriers have provided evidence for high rates of depressive symptoms in women with the FMR1 premutation. In addition to stress and depression, high rates of social phobia and obsessive compulsive symptoms have been reported in women with the FMR1 premutation.

Although the majority of research has focused on social and environmental factors, genetic status (i.e. premutation vs. full mutation) has also been examined as a potential factor that may affect emotional and psychological adaptation in mothers of children with FXS. The few studies that have investigated the relationship between maternal adaptation and genetic status have not found differences between women with the pre- or full mutation in regards to emotional functioning. Other studies have focused on the relationship between CGG repeat length and adverse mental health outcomes in women with the premutation. Results from these studies have been mixed. One study found no relationship between CGG repeat length and increased psychological symptoms while another study reported that women with CGG repeats above 100 scored higher on depression scales.

The goals of this study were to examine a wider range of positive and adverse mental health outcomes, including stress, depressive symptoms, anger, anxiety, hope, quality of life, and optimism. The researchers aimed to (1) determine the percent of mothers who meet or exceded a clinically significant threshold on one or more mental health outcomes, (2) examine interrelationships among mental health outcomes, (3) compare mental health outcomes

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STRESS

30% of mothers had clinically significant scores that indicated elevated levels of stress.

DEPRESSION

35.5% of mother endorsed no symptoms of depression.

15.9 % had depressive symptoms that were clinically significant.

ANXIETY

17% of mothers were experiencing clinically significant levels of anxiety.

HOPE

11.2% of mothers had low levels of hope.

OPTIMISM

8 mothers' scores reflected low optimism.

ANGER

12.2% of mothers scored in the high anger to very high anger range.

QUALITY OF LIFE

76% of mothers reported an average to high quality of life.

24.1% of mothers was classified as having a low quality of life.

13% of mother reported having a very low quality of life.

of mothers with the premutation to those with the full mutation, (4) examine the influence of the child's severity of delay, and (5) test whether CGG repeat length in mothers was associated with mental health outcomes in mothers with the premutation.

One hundred and eight mothers were enrolled in the study, each of whom had at least one child with the full mutation. Eighty-eight percent of mothers had the FMR1 premutation and 12 percent had the full mutation.

Mothers were asked to take 4 surveys to measure negative adaptation (i.e. stress, depressive symptoms, anger, and anxiety) and 3 surveys to examine positive adaptation (i.e. hope, optimism, and quality of life). Mothers were also asked to answer two additional questionnaires, the Vineland Adaptive Behavior Scale and the Child Behavior Checklist, to asses their child's developmental delays and problem behavior.

The findings of this study suggest that poor adaptation associated with raising a child with a disability is not universally experienced by mothers. Indeed, most mothers in this sample scored in the normal or clinically non significant range on most measures of mental health. Moreover, the majority of mothers reported high levels of hope and optimism and had an average to high quality of life. When looking at all mental health outcomes, the highest proportion of clinically significant scores was on the stress measure (30%). This finding is consistent with the current literature that suggests that families of children with FXS are likely to experience high levels of stress.

Findings associated with genetic status were also consistent with past research. Mothers with the full mutation did not differ significantly from mothers with the premutation on mental health outcomes with the exception of depressive symptoms. In addition, CGG repeat length was not associated with maternal adaptation.

Child behavior problems were also associated with maternal adaptation. Behavior problems were associated with maternal stress, depressive symptoms, anxiety, anger, and quality of life. The severity of the child's delay, however, was not associated with maternal adaptation.

Overall, the findings suggest that most mothers scored in the normal range on most of mental health outcomes examined in this study. However, it is important to note that about half of the mothers experienced difficulties in at least one area of mental health. Therefore, intervention and prevention programs should be developed to assist mothers of children with FXS with healthy adaptation.

This research summary is based on the following published article: Bailey DB, Sideris J, Roberts JE, Hatton DD. Child and genetic variables associated with maternal adaptation to fragile X syndrome: A multidimensional analysis. American Journal of Medical Genetics, Part A. 2008; 146(6): 720-729. This summary was prepared by the Fragile X Research Registry. If you have any questions or would like to contact the researchers of this study, please send an email to info@FragileXRegistry.org.