Assessing Psychiatric Trait Burden In FXS Families: The Potential Benefits of Whole Family Intervention

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URES 295

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Introduction

Fragile X Syndrome (FXS) is the most common single gene disorder and leading cause of intellectual and cognitive disability (Hunter 2021). It is highly associated with comorbid disorders such as autism and anxiety.

Familial factors are highly implicated in anxiety, with several studies associating maternal anxiety disorders with children's behavioral inhibition and anxiety (Hunter 2021). Behavioral studies affirm that children begin to model the anxious responses they observe in parents. Maternal anxiety may also result in parental over-involvement and reduced child encouragement, each associated with elevated child anxiety symptoms (Potter, 2022). Genetic factors are additionally implicated, as both behavioral inhibition and anxiety disorders are considered highly heritable, and distinct patterns of temperament have been defined in disorders such as FXS (Hunter 2021, Potter 2022).

Currently, studies show that parents do not have access or are not frequently recommended services to help alleviate or garner coping skills related to parental stress (Potter, 2022).

Aims

1. Generate family pedigrees to describe the rates of FXS and FXS-associated disorders in a sample families
2. Generate correlations between parent traits and child traits to provide insight into how these features are related across parent-child dyads as a function of premutation and full-mutation status.
3. Ascertain the usefulness of parent traits in identifying intervention targets for FXS children, as well as the potential benefits of wholistic family intervention approach.

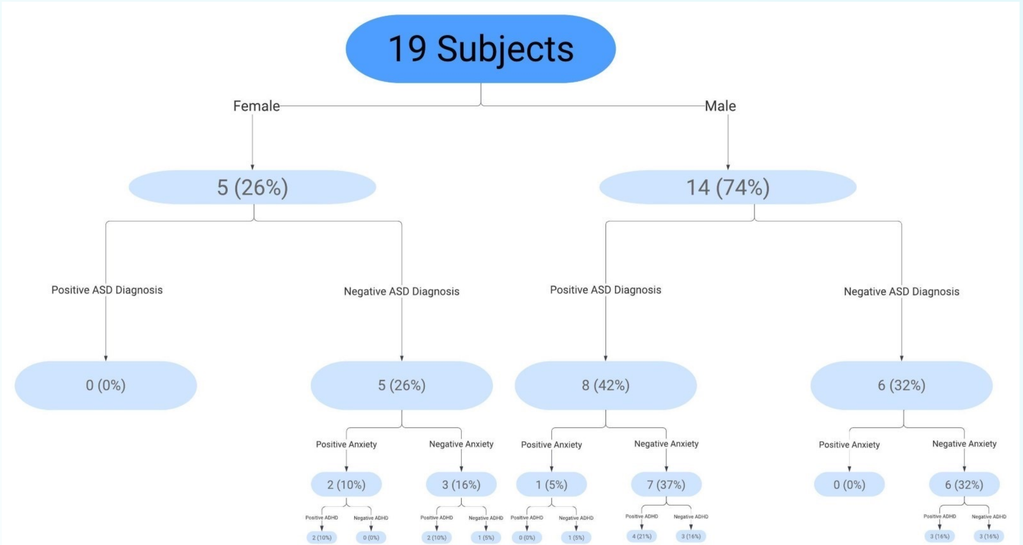
Methods

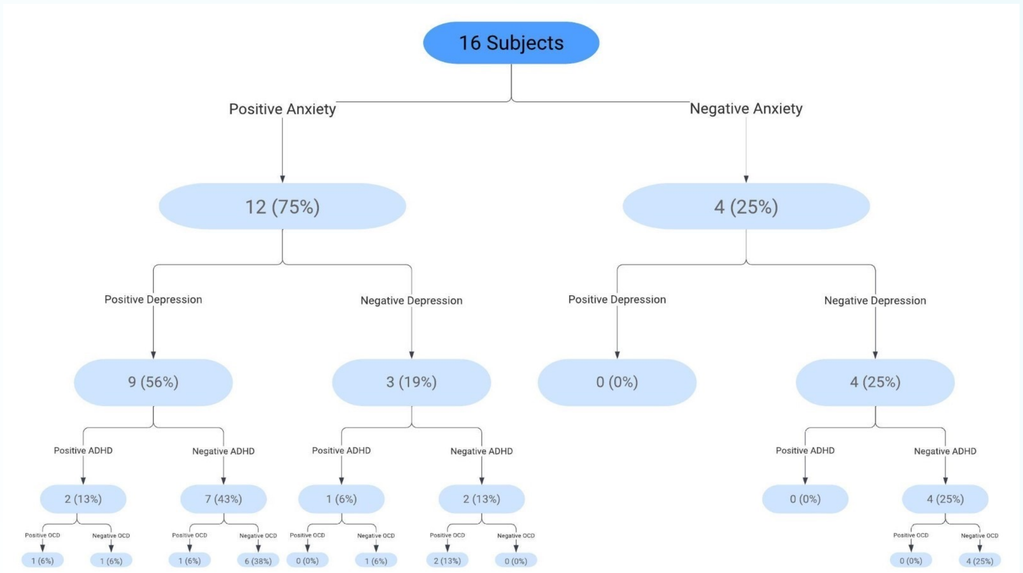
Pedigrees were curated using Quickped online Pedigree creator (https://magnusdv.shinyapps.io/quickped). Charts were edited in Adobe Acrobat to attribute predetermined genetic and medical history to family members based on completed forms.

Self-reported family psychiatric history forms were compiled and assessed to assess additional comorbid psychiatric disorders. The top four disorders were charted for FXS subjects and mothers alike. General correlative relationships were drawn.

Additional analysis was completed in looking at specific case-study examples of mothers with high trait burden or low trait burden and the current outcome of trait burden on their FXS proband.

Results





None of the female probands received a positive ASD diagnosis., ADHD was more common in the female population (4/5) than anxiety (2/5). Only 10% of the female probands had 2 diagnoses, whereas 20% of the female probands had at least 1 psychosocial comorbid diagnosis. 5% of the female FXS subjects had no formal diagnosis.

42% of the male subjects had a positive ASD diagnosis with 26% of that population having one additional psychosocial diagnosis. Of the ASD positive males, 16% were negative for additional psychosocial disorders. Half of ASD negative males had at least one additional psychosocial disorder, whereas the other half had no other diagnosis.

If a mother did not have anxiety, they also did not have any other comorbid disorder. Mothers with positive anxiety (75%) had at least one other diagnosis. Only 6% of mothers positively reported all 4 psychosocial disorders.

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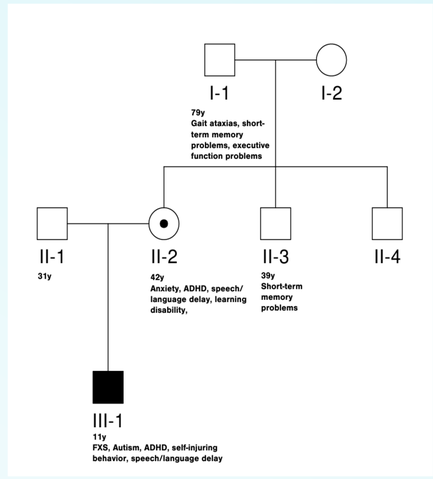
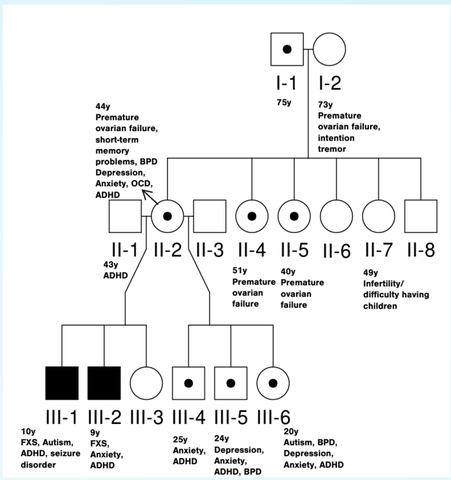
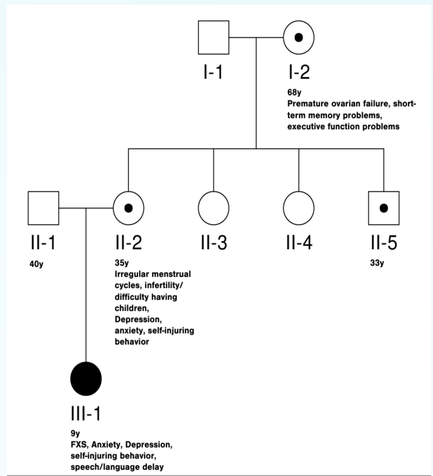
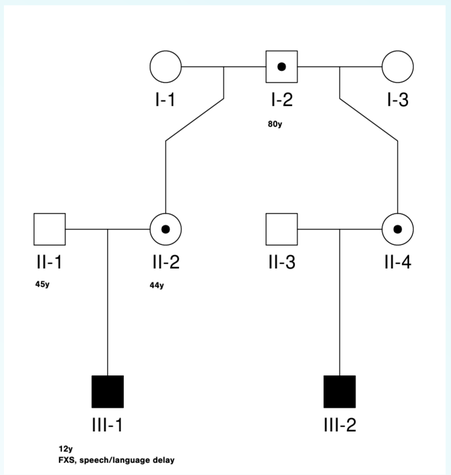
Pedigrees









Conclusion and Future Directions

Preliminary results on a small sample of mothers and children reveal *proof of principle for establishing whole family intervention with families of children with FXS.*

These intergenerational relationships could represent a novel link between comorbid psychiatric disorders and FXS- child functioning outcome. This could also be compounded by FXTAS/ FXPOI symptom presence. Data collection is ongoing and future updates will include more subjects to increase statistical power.

Intergenerationally delineating phenotypic and genetic profiles can contribute to understanding child outcome as it relates to psychiatric predisposition.