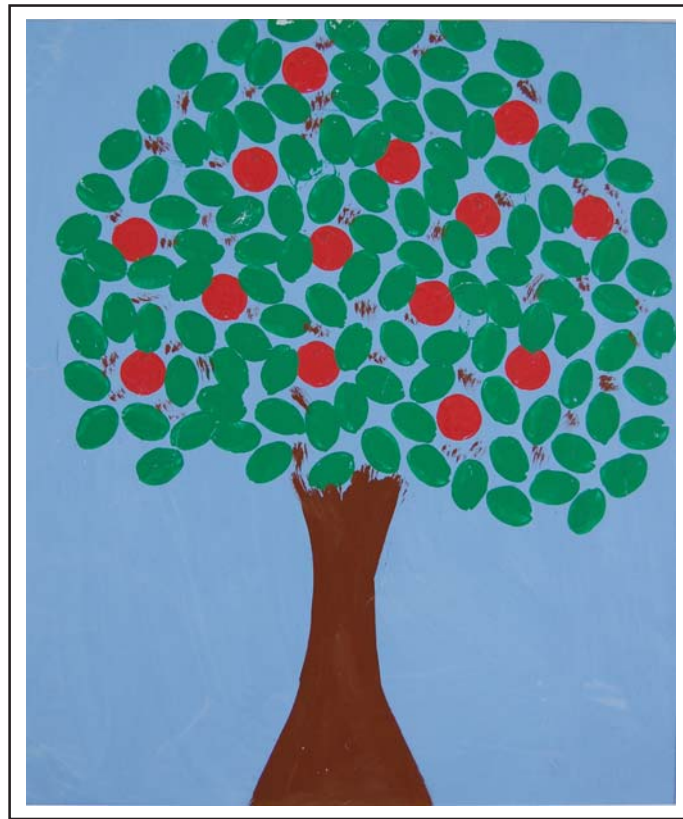


Waisman Center  
University of Wisconsin – Madison

*Dedicated to the advancement of knowledge about human development, developmental disabilities, and neurodegenerative disease.*

## Study on Family Adaptation to Fragile X Syndrome



### Report 4: Fragile X Syndrome in Adolescents and Adults

- Principal Investigators -

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## Table of Contents

<b>Section I: Introduction .....</b>	<b>1</b>
<b>Section II: Change in Adolescent and Adult Behavioral Profiles .....</b>	<b>2</b>
Daily Living Skills .....	3
Behavior Problems .....	5
Autism Symptoms .....	7
Health Conditions .....	8
<b>Section III: Conclusions .....</b>	<b>9</b>
<b>Section IV: Future Work with Families .....</b>	<b>10</b>

## **Section I: Introduction**

Prior to the start of our longitudinal study in 2008, most of the research conducted about individuals with fragile X syndrome (FXS) had focused on children. Little was known about the lives of adolescents and adults with FXS and their families, and what factors might be important for promoting positive development during these periods in the lifespan. Our study began to address this gap by gathering data across three time points from families of adolescent and adult children with FXS. Throughout the five years of the study, participants provided us with very valuable information about the strengths and challenges experienced by individuals with FXS during adolescence and adulthood. We also have learned a great deal about the factors that are associated with positive outcomes for individuals with FXS as they grow older.

At each of the three waves of data collection, mothers participated in telephone interviews and reported on multiple aspects of their children's lives and behavioral profiles. This report focuses on the data regarding the son or daughter's daily living skills, behavior problems, psychological symptoms, autism symptoms, and overall health. This report also discusses recent findings regarding the child and family factors that support these skills and behaviors.

We are very grateful to the families who have provided us with this in-depth information about the lives of their adolescent and adult children. We hope our research will continue to inform our understanding not only of the patterns of development for individuals with FXS but also will provide data to help shape new services and supports for adolescents and adults with FXS and their families. It is only through longitudinal research that the natural history of FXS and the impact on the family can be fully understood. For this reason, we look forward to the opportunity to continue to partner with your family in this work in the future.

If you would like to learn more about the purpose of the study, our staff, and articles written about our Family Adaptation to FXS Study please visit the study website: [http://www.waisman.wisc.edu/family/study\\_frax.html](http://www.waisman.wisc.edu/family/study_frax.html)

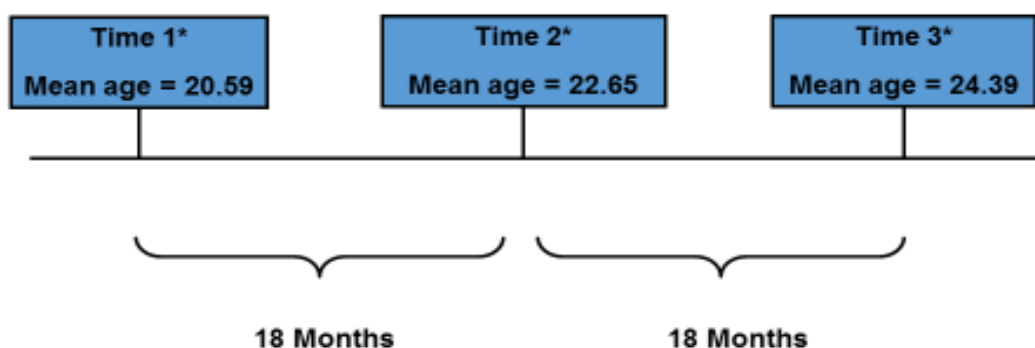
## **Section II: Change in Adolescent and Adult Behavioral Profiles**

This report includes data from the 147 adolescents and adults with FXS, and their families, who participated in our longitudinal study. Families in the sample resided in 38 US states and one Canadian province. Adolescents and adults ranged in age from 12 to 48 at the start of the study (average age = 20.6 years) and 17.9% were female. Almost half of the females (45.6%) and more than four-fifths (88.2%) of the males had a diagnosis of intellectual disability, and one-third (32.3%) of the adolescents and adults had a co-occurring diagnosis of an autism spectrum disorder.

Mothers ranged in age from 35 to 79 at the start of the study (average age = 50.3 years). The majority of mothers had a college degree or greater (57.8%) and were married (80.9%) at the start of the study. The number of children per family ranged from 1 to 6, with an average of 2.4 children per family. About one-third (34%) of families had more than one child with FXS, and an additional 21% of families had at least one other child with another type of disability.

For this study, mothers participated in telephone interviews and completed mail-back questionnaires at three time points separated by approximately 18 months (see diagram below). At each interview, mothers reported on their child's daily living skills, challenging behaviors, and physical and psychological health. We note that 122 of the 147 families (83%) participated in all three waves of the study.

### **FXS Study Timeline**



\* At each time point, we measured the symptoms, behavior, and functioning of the adolescents and adults with FXS.

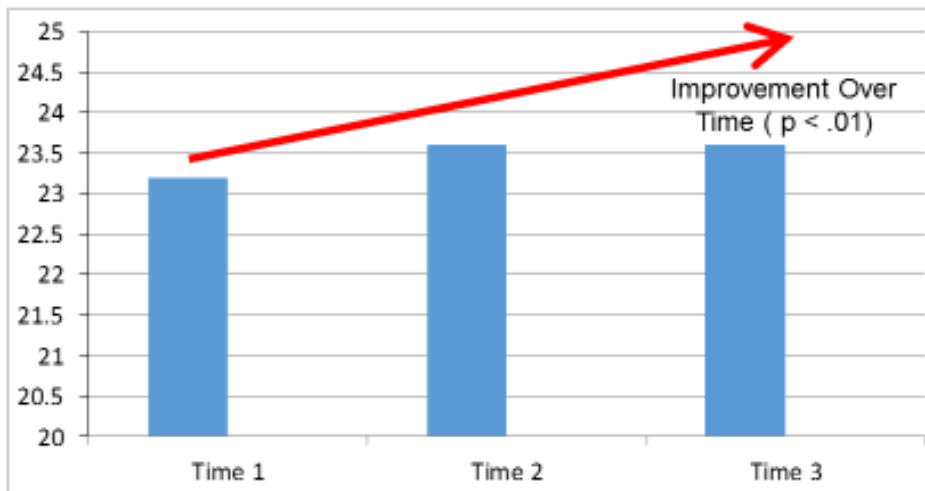
## Change in Daily Living Skills

Mothers reported on their adolescent or adult child's independence in daily living skills at each of the three data collection interviews. Skills included personal care (bathing, toileting), housekeeping (making bed, cleaning) and meal-related activities (make a sandwich, cook simple foods). Higher scores (shown in the left hand column in Figure 1 below) indicated more independence in completing daily tasks.

As shown in Figure 1, on average, there was a steady and statistically significant improvement in daily living skills for adolescents and adults with FXS across the study period.

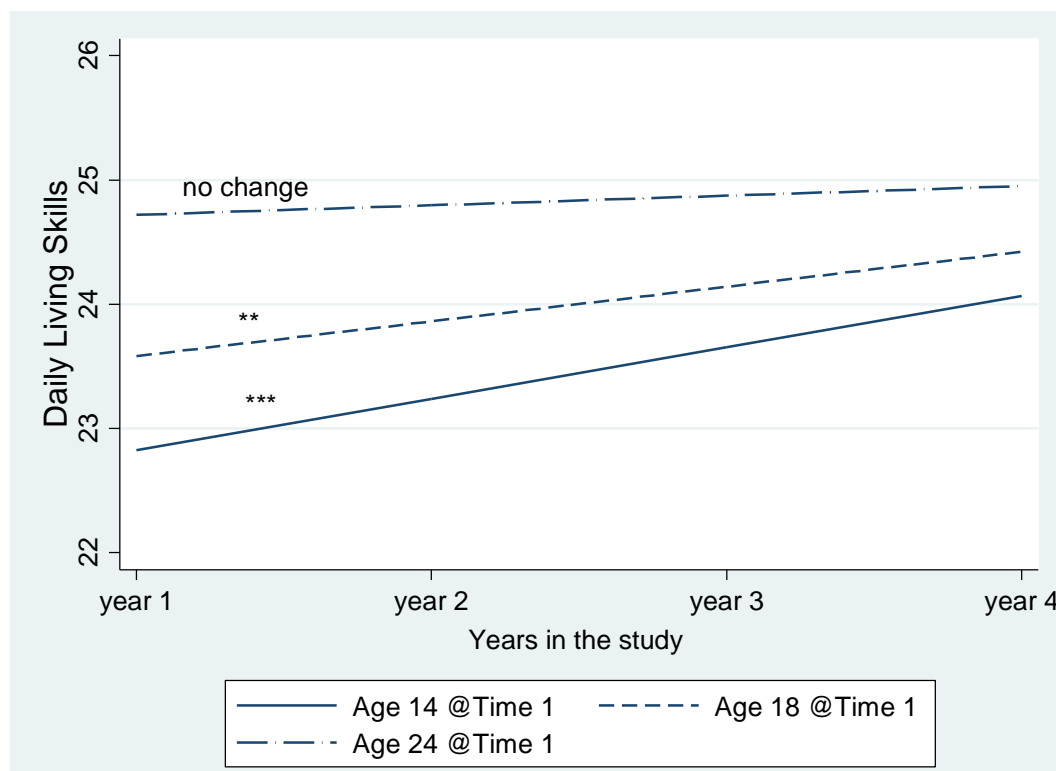
**FIGURE 1.**

### Daily Living Skills



However, we also saw a difference in the amount of change in daily living skills depending on the age of the child at the start of the study. As shown in Figure 2, individuals with FXS who were young teens at the beginning of the study had the fastest rate of improvement. Individuals who were older teens at the start of the study also showed improvement, but not as much of a rapid rate. Finally, individuals who were adults at the start of the study did not show statistically significant gains in daily living skills during the course of the study. Rather, they maintained their skills over time. This means that although on average adults were not gaining additional new daily living skills, they had the highest overall levels of daily living skills.

**FIGURE 2. Change in Daily Living Skills for Adolescents and Adults with FXS**



We also were interested in what factors might predict improvement in daily living skills over time for adolescents and adults with FXS. To explore this question, we examined a range of factors related to the individual with FXS and factors related to the family.

Our results indicated that higher levels of daily living skills for adolescents and adults with FXS were predicted by:

- Being female
- Being older
- Taking fewer psychotropic and non-psychotropic medications
- Experiencing more family warmth (meaning kindness, concern, and empathy expressed towards the individual with FXS)

### **Change in Behavior Problems**

At each time point, mothers also reported on the severity and frequency of the following 8 behavior problems for their son or daughter:

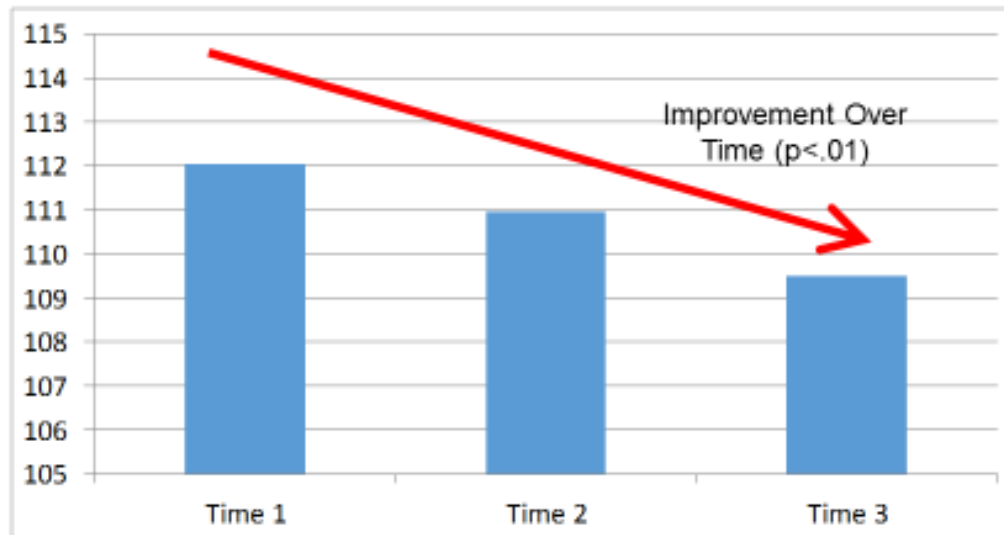
- Disruptive behavior
- Uncooperative behavior
- Socially offensive behavior
- Hurtful to self
- Hurtful to others
- Hurtful to property
- Repetitive or unusual behavior
- Withdrawn or inattentive behavior

Severity and frequency ratings of each behavior problem were combined to give a total behavior problems score at each wave of data collection. As shown in Figure 3, on average there was a steady and statistically significant decline in behavior problems across the study period, suggesting an improvement in overall behavioral functioning as individuals with FXS grew into later adolescence and adulthood.



FIGURE 3.

## Behavior Problems



We were also interested in what factors might predict a decline in behavior problems over time for adolescents and adults with FXS. We found that less severe behavior problems were once again predicted by:

- Being female
- Being older
- Taking fewer psychotropic and non-psychotropic medications
- Experiencing less family criticism of the child

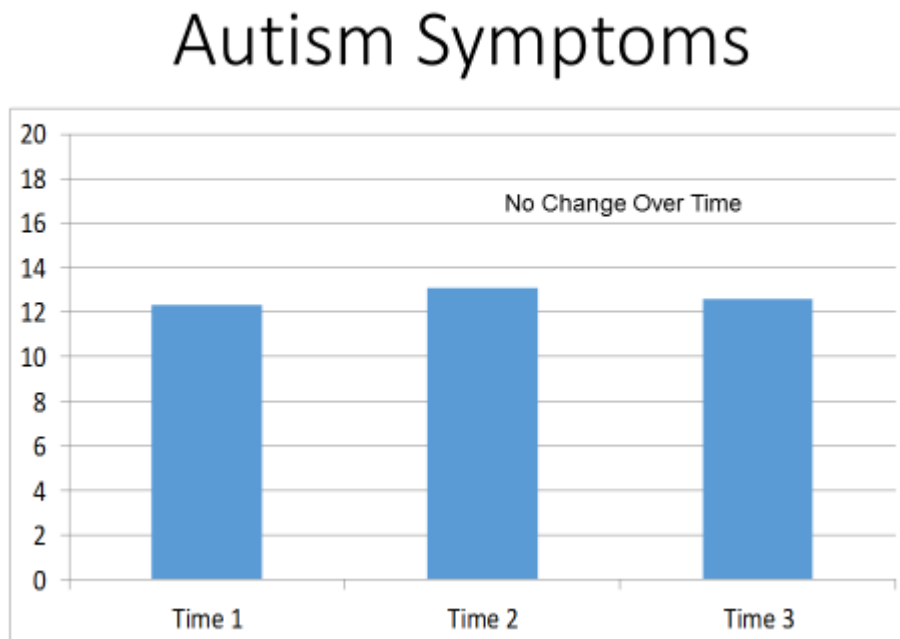
We also found that when there were increases in family warmth during the study period, the adolescents and adults in these families displayed more pronounced decreases in behavior problems.

## **Change in Autism Symptoms**

We know from past research that many individuals with FXS display behaviors or traits similar to autism. We were curious to see if autism symptoms were stable or if they changed over time in our sample. We asked mothers to report whether or not their son or daughter displayed specific behaviors related to autism such as problems with communication, social interactions, or intense interest in a particular topic at each wave of the study.

As shown in Figure 4, on average, autism symptoms remained very stable over time.

**FIGURE 4.**



We were also interested in what factors might predict the autism symptoms of adolescents and adults with FXS. Similar to the findings from the daily living skills section, we found that lower levels of autism symptoms were predicted by:

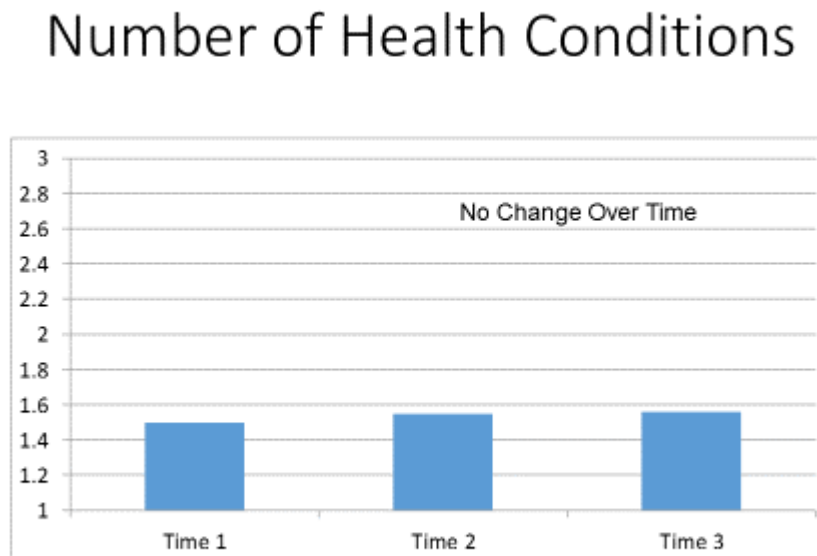
- Being female
- Being older
- Taking fewer psychotropic medications
- Experiencing more family warmth

## **Change in Number of Health Conditions**

Finally, we were interested in how physical health might change over time for the adolescents and adults with FXS in our sample. At each time point mothers reported on the number of health conditions their son or daughter had been diagnosed with during the past 18 months. The most common health conditions for adolescents and adults were experiencing anxiety and depression (44%), being overweight (22%), and having allergies (20%).

Figure 5 shows the average number of health conditions for the sample. The adolescents and adults displayed an average of about 1.5 health conditions at each wave, suggesting that health remained relatively stable over the course of the study. Not surprisingly, individuals who took more non-psychotropic medications were those with higher numbers of health conditions. On average the adolescents and adults were taking 1.3 psychotropic medications and 0.6 non-psychotropic medications. There were no other significant predictors of health conditions.

**FIGURE 5.**



### **Section III: Conclusions**

In this report we have described the pattern of change and stability across key skills and behaviors of individuals with FXS as they move through adolescence and adulthood. We reported on daily living skills, behavior problems, autism symptoms, and health. We also reported on the factors that predict favorable outcomes, which included child age, gender, medication use, and the importance of a positive family environment.

We found that autism symptoms and health were relatively stable over time. In contrast, daily living skills improved over time for adolescents and young adults in the sample. Also encouraging was that behavior problems were declining over time for individuals of all ages. Notably, adults in the sample had higher levels of daily living skills and fewer behavior problems and autism symptoms than adolescents.

Females with FXS displayed higher levels of daily living skills and lower levels of behavior problems and autism symptoms than males in the sample. Medications were typically prescribed for those individuals who had fewer daily living skills and higher levels of behavior problems and autism symptoms.

We also found that experiencing higher levels of family warmth was associated with more independence in daily living skills, and lower levels of autism symptoms. Warmth was also related to declines in behavior problems over time.

In summary, we find patterns of both stability and improvement for adolescents and adults with FXS as they grow older. We also see that when parents provide a positive environment for the individual with FXS, it has a strong influence on outcomes during adolescence and adulthood. Families that expressed higher levels of warmth had children who consistently improved in their behavior problems over time, showing how positive relationships between family members can have lasting impacts on children's growth and well-being, and this impact lasts into adulthood.

Happily, our study's findings highlight that:

- Adulthood is a time when the gains made during adolescence in daily living skills are maintained.
- Health is stable during adolescence and adulthood for individuals with FXS.

- Behavior problems consistently become less severe as individuals age.
- The family can optimize the functioning of the adolescents and adults with FXS through expressions of warmth.

## **Section IV: Future Research with Families**

In this report we have outlined findings from our 5-year longitudinal study about outcomes for adolescents and adults with FXS. As the sons, daughters, and mothers in our longitudinal study become older, it is important to understand the overall impact of fragile X syndrome on the entire family. Of specific interest are the mothers in our study since they typically have multiple caregiving roles at this point in their lives with aging parents, children, and even perhaps grandchildren.

In 2016, mothers in our longitudinal sample will range in age from 43 to 87 years old. Important questions that come into focus are:

- What does the overall health profile look like for mothers at this point in their lives?
- What effect does stress and multiple caregiving roles have on over well-being?
- What factors contribute to or hinder overall quality of life for families affected by FXS?

We are very pleased to share that NICHD has awarded us a new grant to continue our research with the families in this study. Our future work will focus on various aspects of quality of life for mothers of adolescent and adult children with have FXS, and will deepen our understanding of the environmental factors that contribute to overall health, well-being, and daily functioning. An important purpose of this new study is to learn more about how to improve outcomes for all members of the families affected by FXS across the life course.