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

Study on Family Adaptation to Fragile X Syndrome



Report 3: Daily Health Symptoms and Emotional Well-Being of Mothers Who Have a Child with Fragile X Syndrome

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Section I: Introduction

In the two years since the beginning of the study, participants have provided us with a great deal of information about the daily lives of families of adolescents and adults with fragile X syndrome (FXS). This knowledge has informed our understanding of the strengths, experiences, and needs of families at later points in the lifespan.

As part of the first wave of data collection, mothers participated in an eight-day diary study, providing information on their daily activities, time use, positive and stressful events, and physical health symptoms. This report focuses on the data regarding daily health symptoms, including pain symptoms, gastrointestinal symptoms, dizziness, and menstrual-related problems. This report also discusses the data pertaining to the daily psychological well-being of mothers.

We are very grateful to the families who have provided us with this in-depth information on their health and well-being. From the ongoing participation of families, we will continue to gain insights about the health of parents and the impact of having a child with FXS on the family. We look forward to using this knowledge to inform and enhance comprehensive services and supports for families.

Section II: Daily Health of Mothers

This report includes data from the 112 mothers who participated in the daily diary study and whose son or daughter with FXS was living at home. Mothers ranged in age from 37 to 70 years, with an average age of 49 years. The target sons and daughters with FXS (i.e., son or daughter who was the focus of the study) were between the ages of 12 and 44 years, with an average age of 20 years. Eighty-six percent of target children were sons.

Over the course of 8 days, mothers were called each evening and asked about their experiences that day. Questions related to mothers' daily activities, health symptoms, and the types of behavior problems their child with FXS displayed.

We also selected two comparison groups of mothers to serve as benchmarks for the health symptoms of mothers of children with FXS. The first comparison group was drawn from a nationally representative study of adults in their midlife (Midlife in United States [MIDUS]), and consisted of mothers of children *without disabilities*. The second comparison group was drawn from the Adolescents and Adults with Autism (AAA) study and consisted of mothers of a son or daughter with an *autism spectrum disorder* (ASD). Many of the questions that were asked of the mothers in our study also were asked in the MIDUS and AAA studies, allowing comparisons.

The comparison sample from MIDUS included 230 mothers who did not have a child with a disability or a mental health problem and who had at least one child still living at home. Mothers in the MIDUS sample were similar to mothers in the FXS study in terms of age, education, ethnicity, and marital status. The comparison sample from AAA included 96 mothers of adolescent and adult children who had a diagnosis of ASD, but not FXS. Mothers of children with ASD were selected as a comparison group because they share similarities with mothers of children with FXS such as having difficulty gaining accurate diagnoses and trouble accessing appropriate services. Although mothers of children with ASD were older than mothers in the FXS study, family characteristics (e.g., income, maternal age) that differed among the three groups of mothers were statistically taken into account in all analyses.

If you are interested, you can learn more about MIDUS by visiting the study website (<http://midus.wisc.edu>).

Similarly, you can learn more about the AAA study by visiting the study website http://www.waisman.wisc.edu/family/study_autism.html).

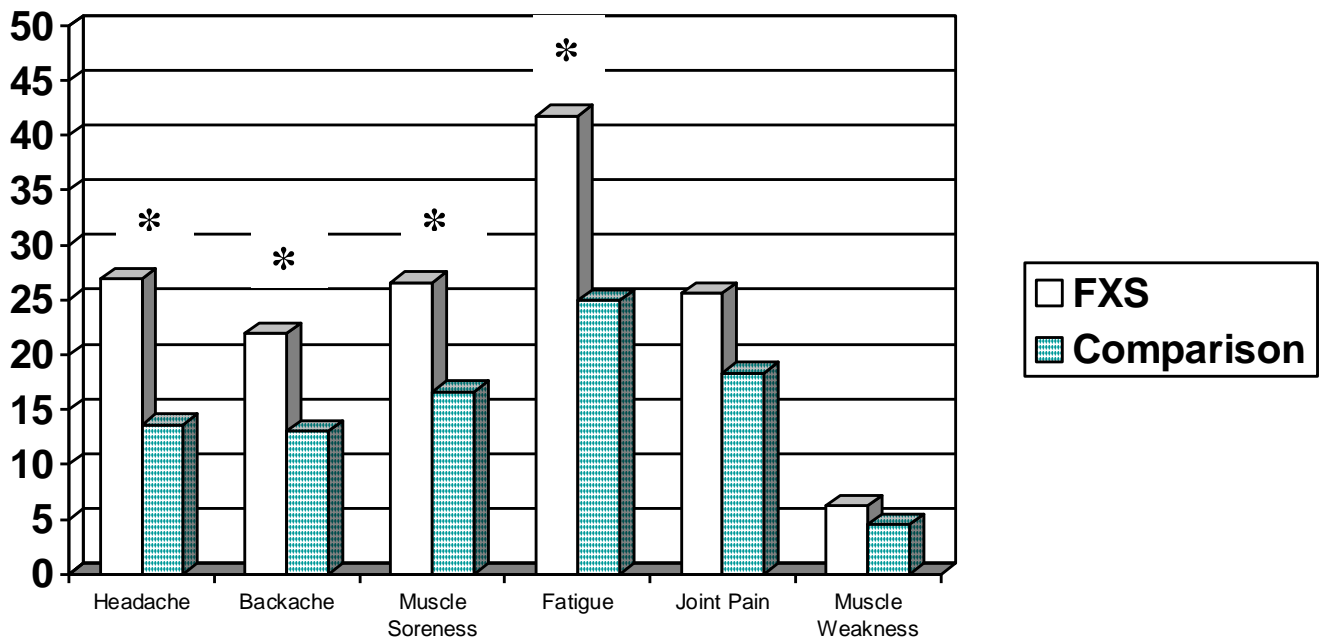
Differences in Pain Symptoms

On each day of the daily diary study, mothers were asked questions about their health during the previous 24 hours. Some of these questions asked about symptoms of pain and weakness. Mothers responded *yes* or *no* to whether they had experienced any of the following pain symptoms during the day:

- Headache
- Backache
- Muscle soreness
- Fatigue
- Joint pain
- Muscle weakness

Mothers of adolescents and adults with FXS reported significantly more days of with headaches, backaches, muscle soreness, and fatigue than the comparison group of mothers who had an adolescent or adult *without a disability*. As shown in Figure 1, mothers in our FXS sample reported headaches and muscle soreness on 27% of days compared with less than 17% of days for mothers in the comparison group. Similarly, mothers in the FXS sample reported backaches on 22% of study days, whereas the comparison group of mothers reported backaches on only 13% of study days. Most notably, mothers of a son or daughter with FXS reported experiencing fatigue on 42% of study days compared to only 25% of days for mothers of children *without disabilities*. There were no differences between the groups for joint pain or muscle weakness.

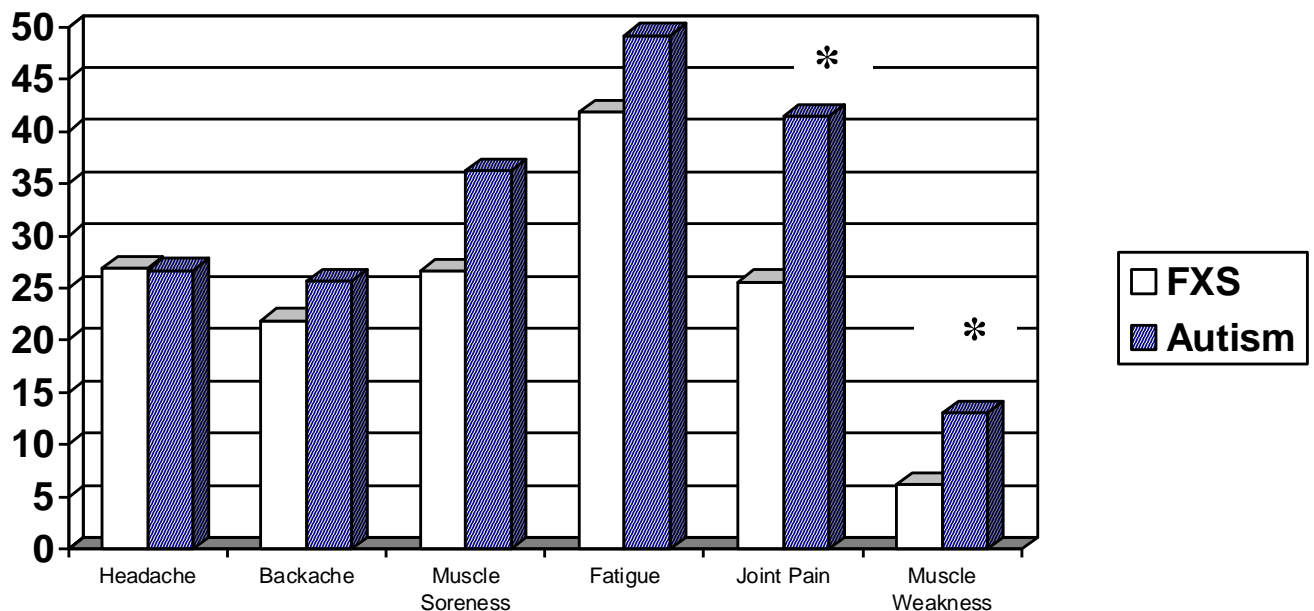
Figure 1. Percent of Days with Pain Symptoms Compared to Mothers of a Son or Daughter *without Disabilities*



Note: Asterisk indicates statistically significant differences between groups.

We also compared mothers in our sample to mothers of adolescents and adults with ASD. In general, mothers of adolescents and adults with FXS did not differ from mothers of adolescents and adults with ASD in daily pain symptoms. They were similar in the percent of days with headaches, backaches, muscle soreness, and fatigue. However, mothers of adolescents and adults with FXS reported significantly *fewer* days when they felt joint pain or muscle weakness than mothers who had an adolescent or adult with ASD. As shown in Figure 2, mothers in our FXS sample reported joint pain on 26% of the study days as compared to 41% of days for mothers in the autism group. Similarly, mothers in the FXS sample reported experiencing muscle weakness on 6% of study days, whereas the mothers in the autism group reported muscle weakness on 13% of study days.

Figure 2. Percent of Days with Pain Symptoms Compared to Mothers of a Son or Daughter with ASD



Thus, although mothers of adolescents and adults with FXS experienced more daily pain symptoms than mothers whose children did not have disabilities, their frequency of pain symptoms was similar to or less than mothers of adolescents and adults with ASD.

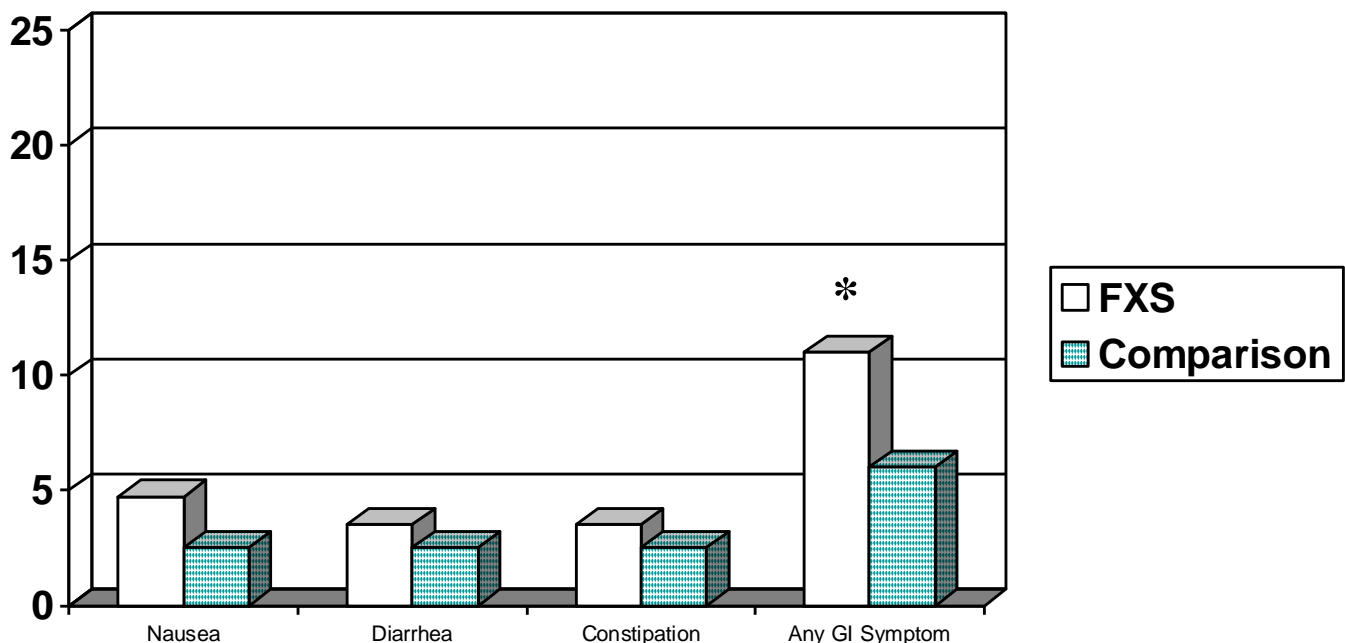
Differences in Gastrointestinal Symptoms

Mothers were asked questions about whether they had experienced any gastrointestinal (GI) symptoms during the previous 24 hours. Mothers responded *yes* or *no* to if they had experienced any of the following three GI symptoms during the day:

- Nausea
- Diarrhea
- Constipation

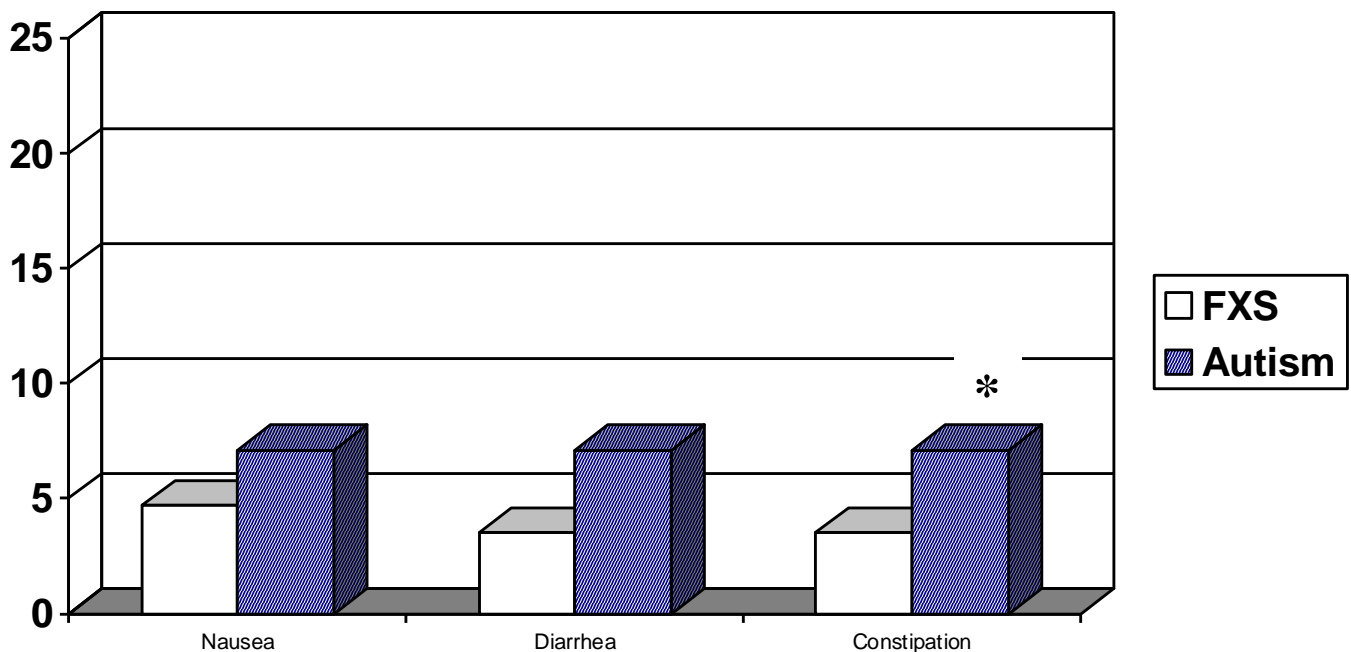
Although mothers of a son or daughter with FXS reported more days with each type of GI symptom (nausea, diarrhea, and constipation) than mothers of similarly-aged children *without disabilities*, none of these differences were statistically significant (shown in Figure 3). However, mothers of adolescent and adult children with FXS experienced at least one GI symptom on over 11% of the study days, compared to mothers of children *without disabilities*, who experienced GI symptoms on only 6% of the study days. This was a statistically significant difference.

Figure 3. Percent of Days with GI Symptoms Compared to Mothers of a Son or Daughter *without Disabilities*



Next we compared mothers of adolescents and adults with FXS to mothers of similarly-aged children with ASD. As shown in Figure 4, mothers of a son or daughter with FXS reported significantly *fewer* days with constipation than mothers of a son or daughter with ASD (3% vs. 8% of the study days). Mothers in both groups were similar in terms of nausea and diarrhea.

Figure 4. Percent of Days with GI Symptoms Compared to Mothers of a Son or Daughter with ASD



In summary, although mothers of adolescents and adults with FXS experienced more overall GI symptoms than mothers of similarly-aged children *without disabilities*, these differences were small. Mothers in our FXS sample also experienced similar levels of GI symptoms as mothers in the ASD sample, although constipation was significantly lower among mothers in the FXS group compared to mothers in the ASD group.

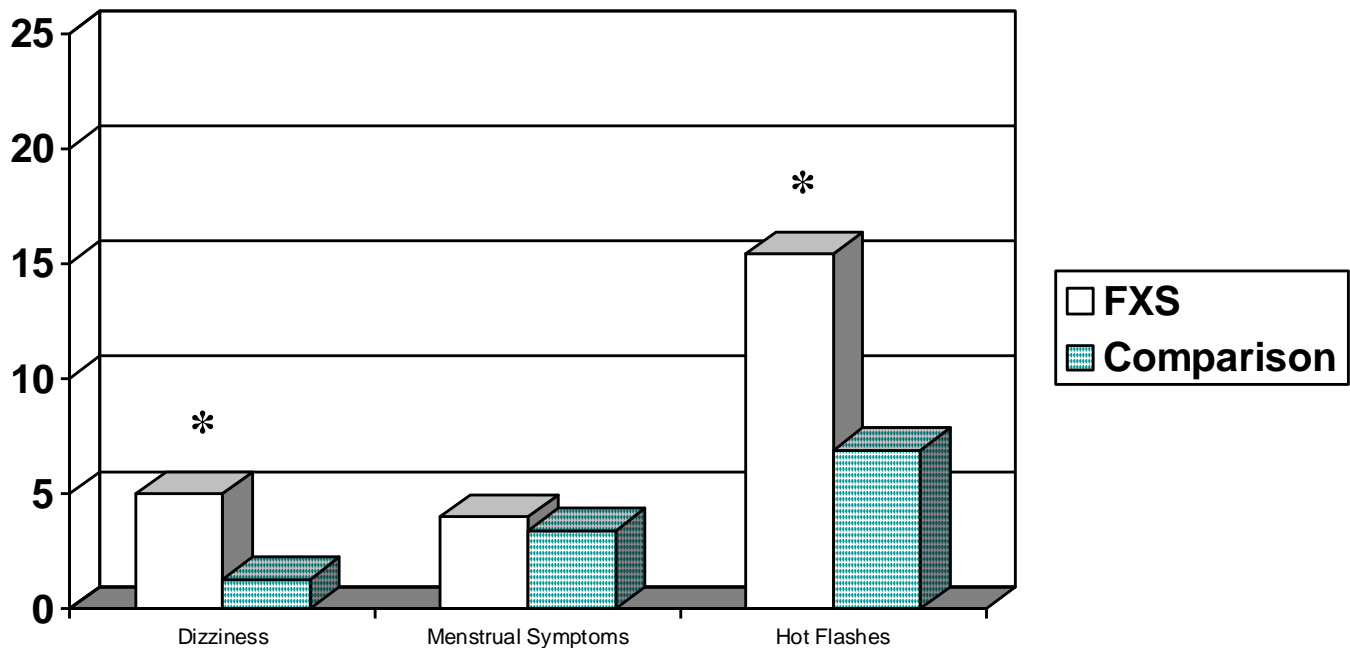
Differences in Other Health Symptoms

Mothers also were asked questions about whether they had experienced any other health symptoms during the previous 24 hours. Mothers responded *yes* or *no* to if they had experienced any of the following symptoms during the day:

- Dizziness
- Menstrual-related symptoms
- Hot flashes or flushes

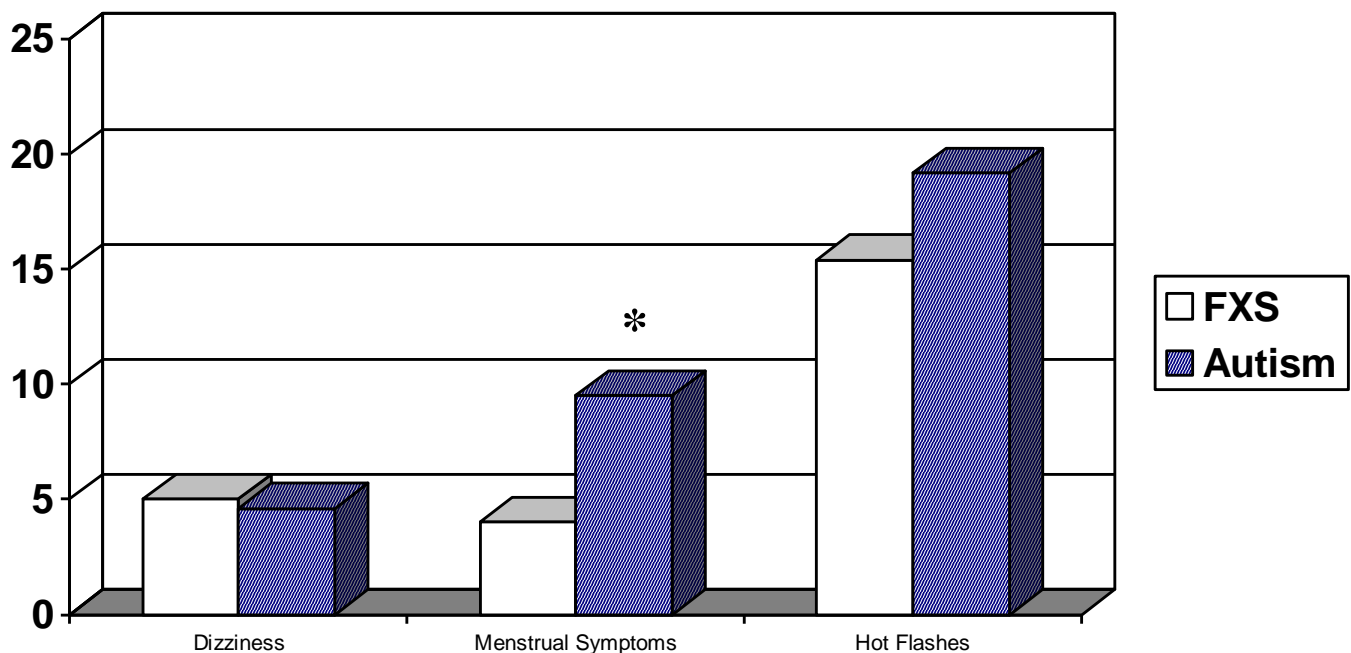
Mothers of adolescents and adults with FXS reported significantly more days with dizziness than the comparison group of mothers who had a son or daughter *without a disability*. As shown in Figure 5, mothers in our FXS sample reported dizziness on 5% of the study days as compared to only 1% of the study days for mothers in the comparison group. Mothers in the FXS sample also reported hot flashes on 15% of the study days, whereas the comparison group of mothers reported hot flashes on 7% of the study days. There were no statistical differences between the groups on the percent of days with menstrual-related symptoms.

Figure 5. Percent of Days with Other Symptoms Compared to Mothers of a Son or Daughter *without Disabilities*



Mothers of adolescents and adults with FXS were similar to mothers of adolescents and adults with ASD in terms of the percent of the study days that they experienced dizziness. In contrast, as shown in Figure 6, mothers of a son or daughter with FXS reported fewer days with menstrual symptoms than mothers of similarly-aged children with ASD (4% of days vs. almost 10% of days). There were no statistically-significant differences between these two groups in the experience of hot flashes.

Figure 6. Percent of Days with GI Symptoms Compared to Mothers of a Son or Daughter with ASD



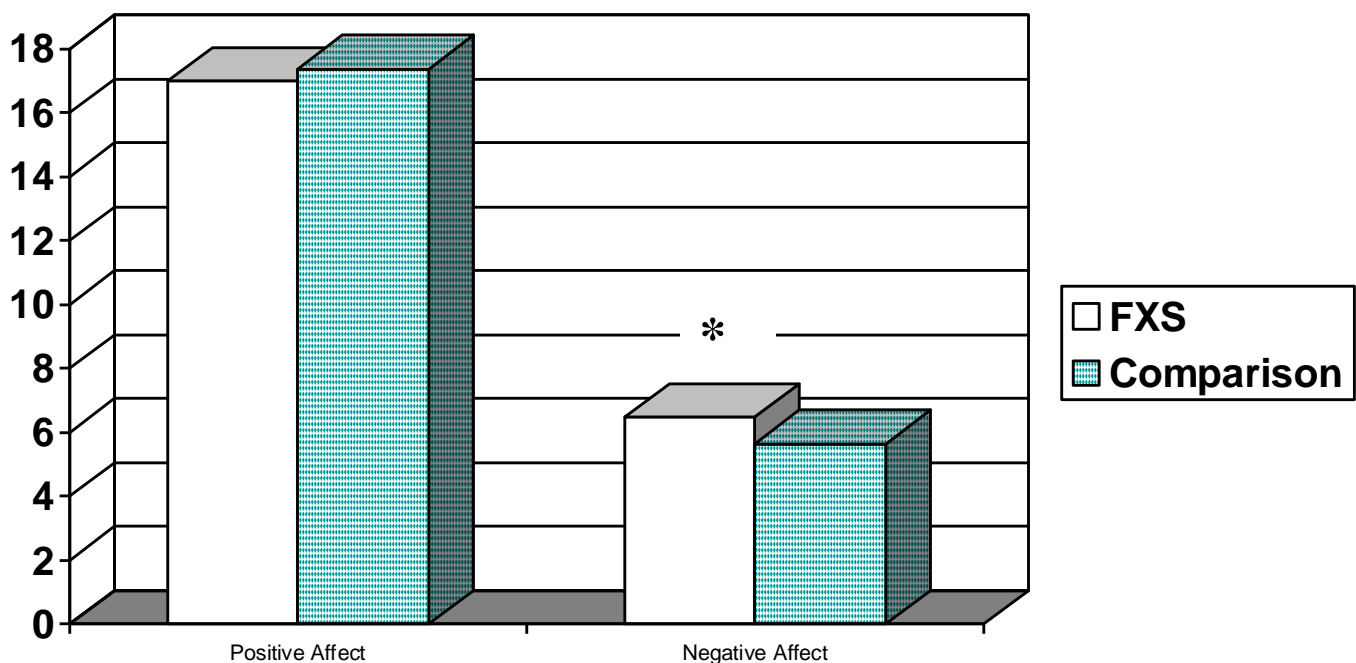
Although mothers of adolescents and adults with FXS experienced more days with dizziness and hot flashes than mothers whose children did not have disabilities, their frequency of these symptoms was similar to mothers of adolescents and adults with ASD. Additionally, mothers in the FXS group reported a similar proportion of days with menstrual symptoms as mothers of children *without disabilities*, but a lower proportion of symptomatic days compared to mothers in the ASD group.

Section III: Daily Well-Being of Mothers

As part of the daily interview, mothers were asked about their experience of different types of emotions, both positive and negative, in the past 24 hours. Each day, mothers indicated how frequently they felt each emotion over the past 24 hours on a 5-point scale from “none of the time” to “all of the time.” A scale of positive affect was created from 5 items, including feeling enthusiastic, attentive, proud, active, and confident; each item was summed, resulting in possible range of 6 to 25, with higher scores reflecting higher levels of positive affect. A negative affect scale was comprised of 5 items, including feeling afraid, jittery, irritable, ashamed, and upset; items were summed to create a possible score of 6 to 25, with higher scores indicating greater negative affect. We averaged the positive and negative affect scores over the course of the eight-day study to create an overall composite of positive and negative emotions during the study.

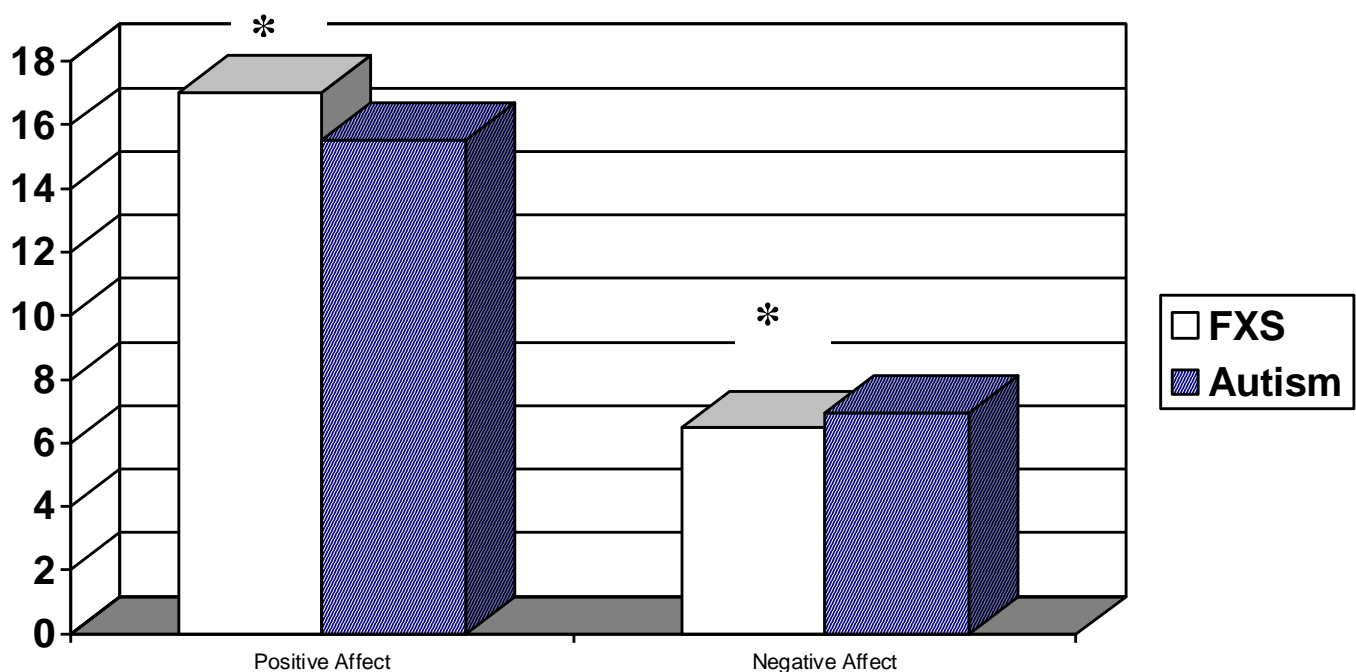
As shown in Figure 7, mothers of a son or daughter with FXS reported similar levels of positive affect compared to mothers of children *without disabilities*. In contrast, however, mothers of adolescent and adult children with FXS reported significantly higher levels of negative affect than comparison mothers of children *without disabilities*.

Figure 7. Positive and Negative Affect Compared to Mothers of a Son or Daughter *without Disabilities*



Next, we examined possible differences between mothers of adolescent and adult children with FXS and mothers of similarly-aged children with ASD in terms of their daily well-being. As shown in Figure 8, mothers in the FXS group reported higher levels of positive affect compared to mothers in the ASD group. Mothers of adolescent and adult children with FXS also reported lower levels of negative affect than mothers of children with ASD. These were statistically significant differences.

Figure 8. Positive and Negative Affect Compared to Mothers of a Son or Daughter with ASD



In conclusion, although mothers of adolescents and adults with FXS reported higher levels of negative affect compared to mothers of similarly-aged children *without disabilities*, negative affect was still lower for mothers in the FXS group than mothers in the ASD group. Additionally, mothers of a son or daughter with FXS reported higher levels of positive affect than mothers of children with ASD and similar levels of positive affect as the non-disability control group, suggesting positivity may be a relative strength for mothers of adolescents and adults with FXS.

Section IV: Conclusions

This report has described the daily health symptoms and emotions of mothers of adolescents and adults with FXS. We have compared mothers in our FXS sample to two other groups of mothers: mothers of children *without disabilities* and mothers of children with ASD.

There were a number of differences between mothers of children with FXS and mothers of children *without disabilities* in terms of their daily physical health and psychological well-being:

- Mothers of a son and daughter with FXS reported *more* days with pain symptoms including headaches, backaches, muscle soreness, and fatigue than mothers of children *without disabilities*.
- Mothers of a son and daughter with FXS also reported experiencing *more* days with dizziness and hot flashes than did the comparison group of mothers.
- Mothers of a son or daughter with FXS reported *higher* levels of negative affect than mothers of children *without disabilities*.

We also found differences between mothers of sons and daughters with FXS and mothers of a son or daughter with ASD:

- Mothers of a son and daughter with FXS reported *fewer* days with joint pain, muscle weakness, constipation, and menstrual-related symptoms than mothers of similarly-aged children with ASD.
- Mothers of a son or daughter with FXS reported *lower* levels of negative affect and *higher* levels of positive affect compared to mothers of adolescent and adult children with ASD.

In summary, mothers of sons and daughters with FXS report more days with many different kinds of health symptoms compared to mothers of similarly-aged children *without disabilities*. Mothers of children with FXS also report a similar, and in some cases lower, level of health symptoms compared to mothers of children with ASD. These findings suggest that the stress of caring for a child with a disability may act as a potential contributor to health difficulties for mothers on a day-to-day basis. An exciting next step of our research will be to explore possible associations of health symptoms with CGG Repeat and Activation Ratio in mothers of children with FXS.

It is also important to highlight that mothers of adolescent and adult children with FXS reported similar levels of positive affect in comparison to mothers of children *without disabilities*, suggesting that despite experiencing more frequent physical health problems, mothers in our study are able to maintain a relatively high level of positivity. Future research will be needed to understand how positive affect in the face of daily challenges may serve to support good health in mothers of children with FXS.

We aim to share these findings, along with future data, with researchers and practitioners to inform them of what we have learned about the daily experiences of health and well-being of mothers of adolescent and adult children with FXS. We hope that our findings will inform interventions and programs for individuals with FXS and their families and provide evidence of the need for comprehensive, family-centered services not only during early childhood, but also during adolescence and adulthood.