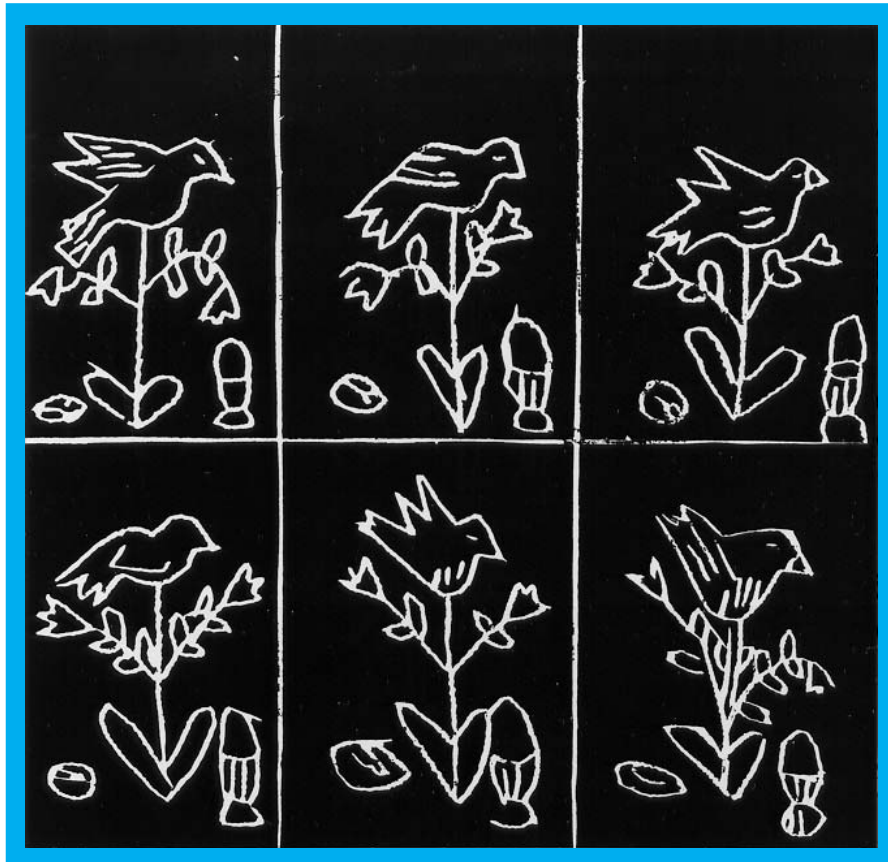


Waisman Center  
University of Wisconsin – Madison

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

## Study on Family Adaptation to Fragile X Syndrome



### Report 2: Daily Experiences and Biological Markers of Stress in Mothers of Adolescents and Adults with Fragile X Syndrome

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## Table of Contents

<b>Section I: Introduction .....</b>	<b>2</b>
<b>Section II: Daily Experiences .....</b>	<b>3</b>
<b>Fatigue and Work Schedules .....</b>	<b>3</b>
<b>Stressful Daily Events.....</b>	<b>4</b>
<b>Positive Daily Events .....</b>	<b>6</b>
<b>Social Support .....</b>	<b>7</b>
<b>Behavior Problems of Son/Daughter .....</b>	<b>8</b>
<b>Section III: Cortisol .....</b>	<b>10</b>
<b>Section IV: Conclusions.....</b>	<b>12</b>

## **Section I: Introduction**

Over the past year, we have learned a great deal about the experiences of adolescents and adults with fragile X syndrome (FXS) and their families. This knowledge has informed our understanding of how FXS both affects, and is affected by, families across the lifespan.

As part of the first interview, we gathered information about the “day-to-day” experiences of mothers who have an adolescent or adult with FXS. Mothers participated in a daily diary study that involved telephone interviews on 8 consecutive days. Mothers also provided salivary samples on 4 of these days, which allowed us to measure their cortisol (a stress hormone). This study has helped us better understand what life is like for these mothers and the biological impact of these daily experiences. In this report, we summarize some of our findings.

We are grateful to the families who have and continue to participate in our research. Our research would not be possible without their generous support. We look forward to the opportunity to use findings from our project to inform interventions and public education programs for adolescents and adults with FXS and their families.

## **Section II: Daily Experiences**

This report includes data from the 134 mothers from our project who participated in the daily diary study. Mothers ranged in age from 35 to 79 years, with an average age of 50 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 48 years, with an average age of 20 years. About one-third (34%) of families had more than one child with FXS, and an additional 21% of families had at least one child with another type of disability.

Over the course of 8 days, mothers were called each evening and asked about their experiences that day. Questions asked about how mothers spent their time during the day, the occurrence of any stressful or positive events, and the types of behavior problems their son or daughter with FXS displayed.

In order to better understand the daily lives of families with adolescents and adults with FXS, a sample of mothers was drawn from a nationally representative study of adults in their midlife (Midlife in United States [MIDUS]), none of whom had a son or daughter with a disability. Many of the questions that were asked of the mothers in our study were also asked in the MIDUS study, allowing us to compare groups.

The comparison sample from MIDUS included 230 mothers who were similar to the mothers in our project in terms of age, education, and the age of their son or daughter. Family characteristics that differed among our sample of mothers and the MIDUS comparison group 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>).

### **Fatigue and Work Schedules**

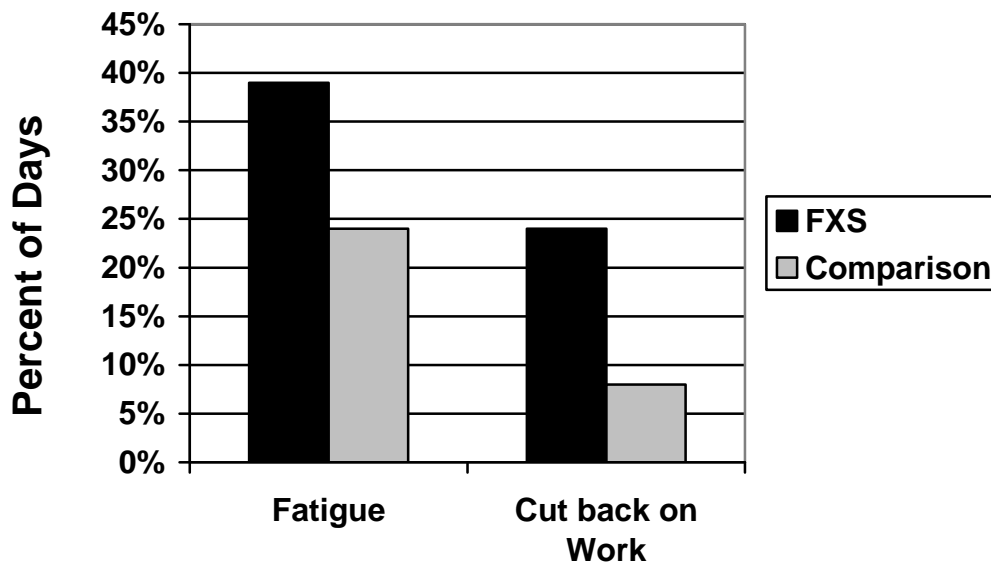
On each day of the daily diary, mothers were asked questions about their energy level and work schedule during the previous 24 hours. Mothers responded *yes* or *no* to the following items:

- Did you experience fatigue during the day?
- Did you cut back on normal work?

Mothers of adolescents and adults with FXS reported significantly more days when they felt fatigued and more days when they needed to cut back on work 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 fatigue on 39% of the study days as compared to 24% of days for mothers in the comparison group. Similarly, mothers in the FXS sample reported having to cut back on work on 24% of study days, whereas the comparison group of mothers reported having to cut back on work on only 8% of study days.

**Figure 1. Percent of Days with Fatigue and Cutting Back at Work**

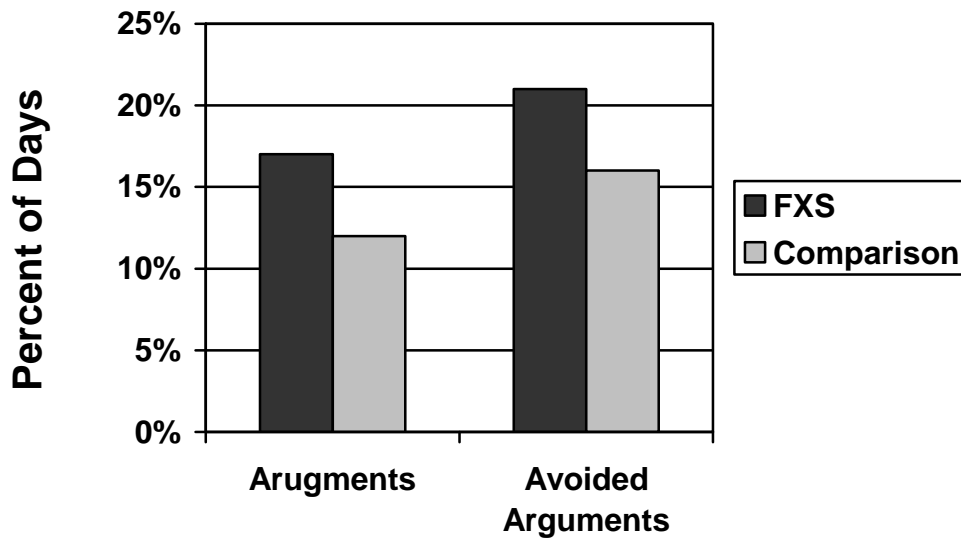


### **Stressful Daily Events**

On each day of the daily diary, mothers were asked to indicate whether they had experienced the following types of stressful events in the previous 24 hours.

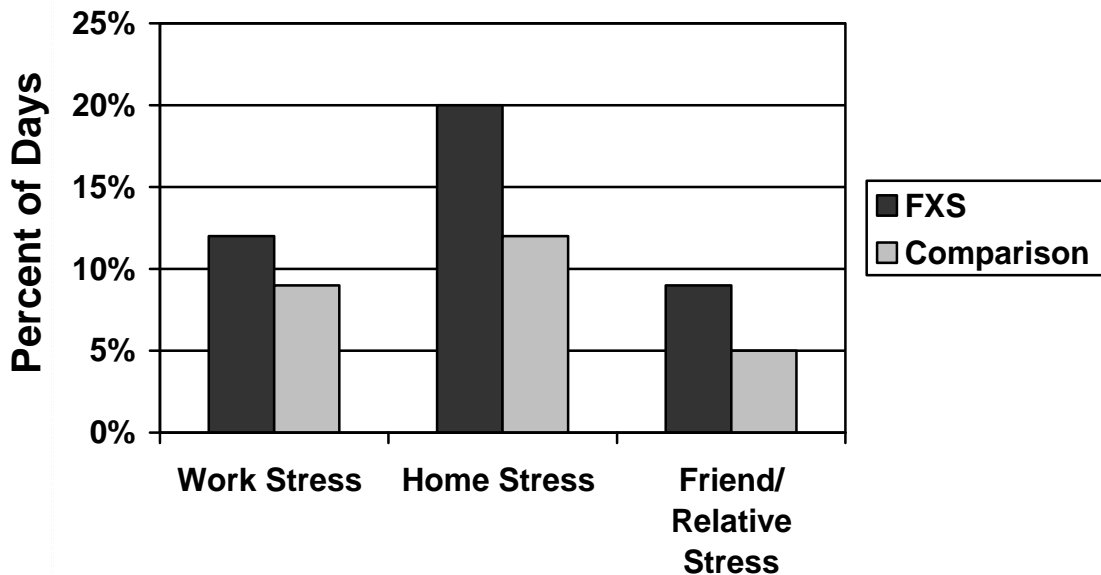
- Arguments: Did you have an argument or disagreement with anyone?
- Avoided Arguments: Did anything happen that you could have argued about but you decided to LET PASS in order to AVOID a disagreement?
- Work Stress: Did anything happen at work or school that most people would consider stressful?
- Home Stress: Did anything happen at home that most people would consider stressful?
- Friend/Relative Stress: Did anything happen to a close friend or relative that turned out to be stressful for YOU?

**Figure 2. Percent of Days with Arguments and Avoided Arguments**



As shown in Figure 2, mothers of adolescents and adults with FXS experienced more days with arguments and more days during which they could have had an argument but chose not to, as compared to the comparison group.

**Figure 3. Percent of Days with Stress at Work, Home, and with Friends or Relatives**



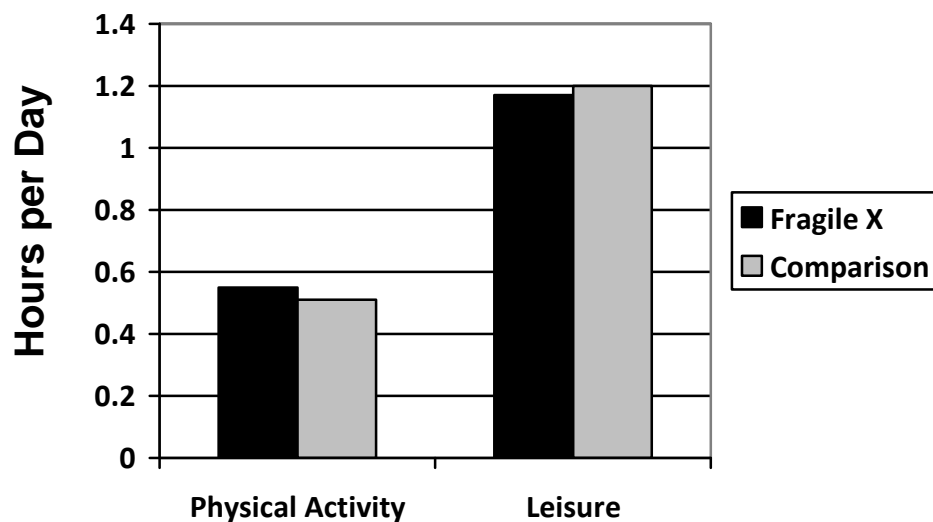
As seen in Figure 3 on the previous page, in contrast to mothers in the comparison group, mothers in the FXS sample reported significantly more days with something stressful happening at work, at home, and to a friend or relative.

## Positive Daily Events

Across the 8 days in the daily diary, mothers also responded with a *yes* or *no* to each of the following questions about positive events:

- How many hours did you spend engaged in vigorous physical exercise?
- How many hours did you spent engaged in a leisure activity?
- Did you have an interaction with someone that most people would consider PARTICULARLY positive since this time yesterday?
- Since this time yesterday, did anything happen to a close friend or relative that turned out to be particularly positive for YOU?

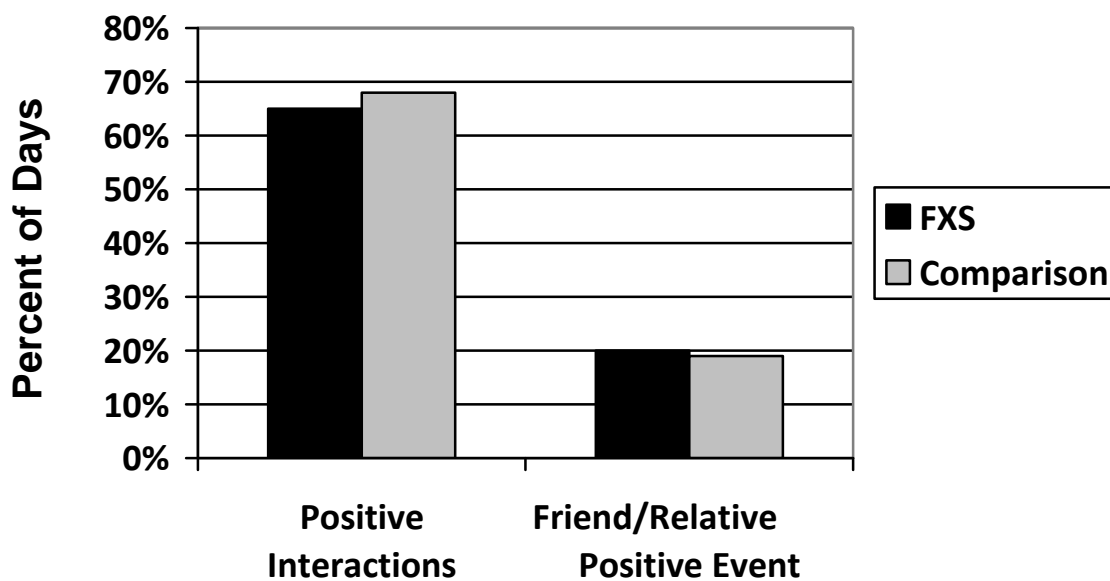
**Figure 4. Average Time (in Hours) Spent in Physical Activity and Leisure**



As shown in Figure 4, mothers in the FXS sample reported *similar* levels of daily physical activity and time spent in leisure activities as mothers in the comparison group. On average, both mothers in the FXS sample and mothers in the comparison group engaged in physical activity for about a half an hour per day and spent more than one hour a day in leisure activities.



**Figure 5. Proportion of Days experienced Positive Interaction or Friend/Relative experienced Positive Event**



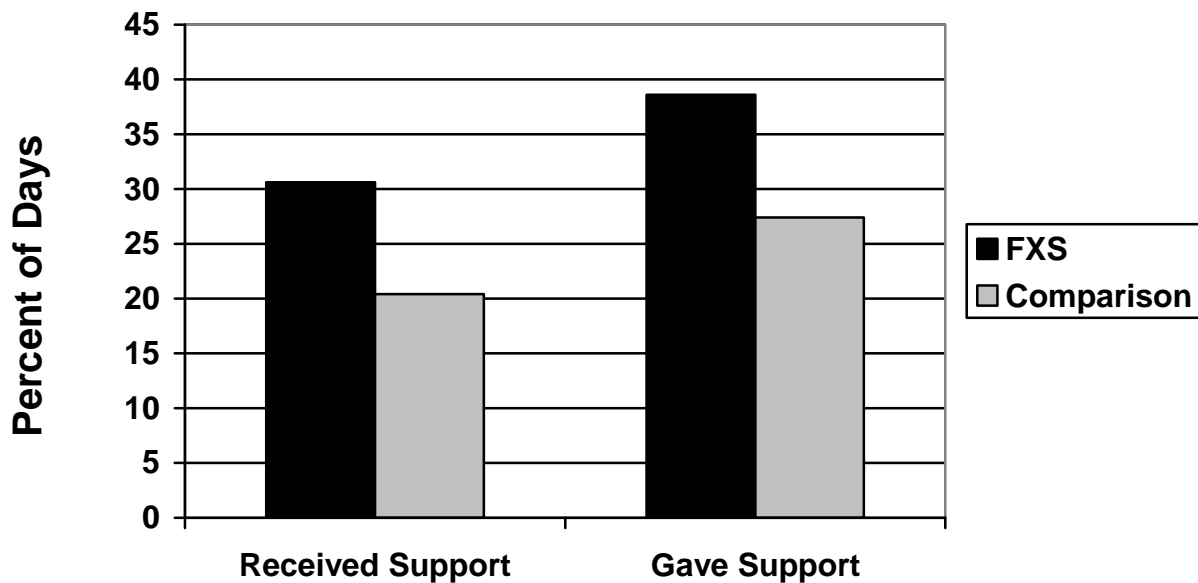
In addition, as shown in Figure 5, mothers in the FXS sample were *similar* to mothers in the comparison group in terms of the percent of days that they had positive interactions or reported positive events for a friend/relative. Mothers with an adolescent or adult with FXS reported having positive interactions on 67% of the days studied and indicated that a positive event occurred for a friend/relative on 20% of the days, similar to the comparison group.

### **Social Support**

Additionally, mothers reported on giving and receiving emotional support.

- Since this time yesterday, did you RECEIVE any EMOTIONAL SUPPORT from anyone or any organizations?
- Not counting work you might do as part of your job, did you spend any time GIVING EMOTIONAL SUPPORT to anyone?

**Figure 6. Percent of Days with Emotional Support Given and Emotional Support Received**



Mothers in the FXS sample reported that they gave more emotional support to others, but also received more emotional support from others, than did the comparison group of mothers. Mothers in the FXS sample reported that they gave emotional support to others on 38% of the days and received emotional support on about one-third of the days.

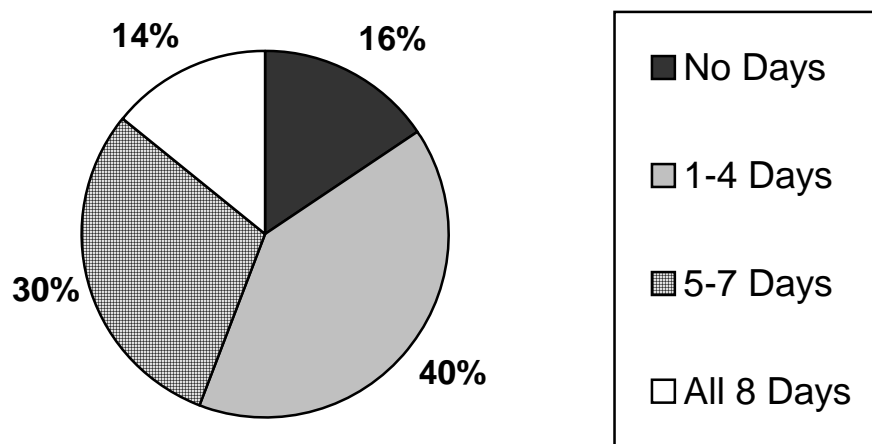
### **Behavior Problems**

Mothers were also asked to report whether the target son or daughter with FXS exhibited various behavior problems during the previous 24 hours. These questions were not asked of the comparison group.

- Did your child hurt himself or herself today?
- Was your child destructive or hurtful to others?
- Was your child destructive to property today?
- Did your child show any unusual or repetitive behavior today?
- Did your child show any socially offensive behavior today?
- Was your child withdrawn or inattentive today?
- Was your child disruptive to others?
- Was your child uncooperative today?

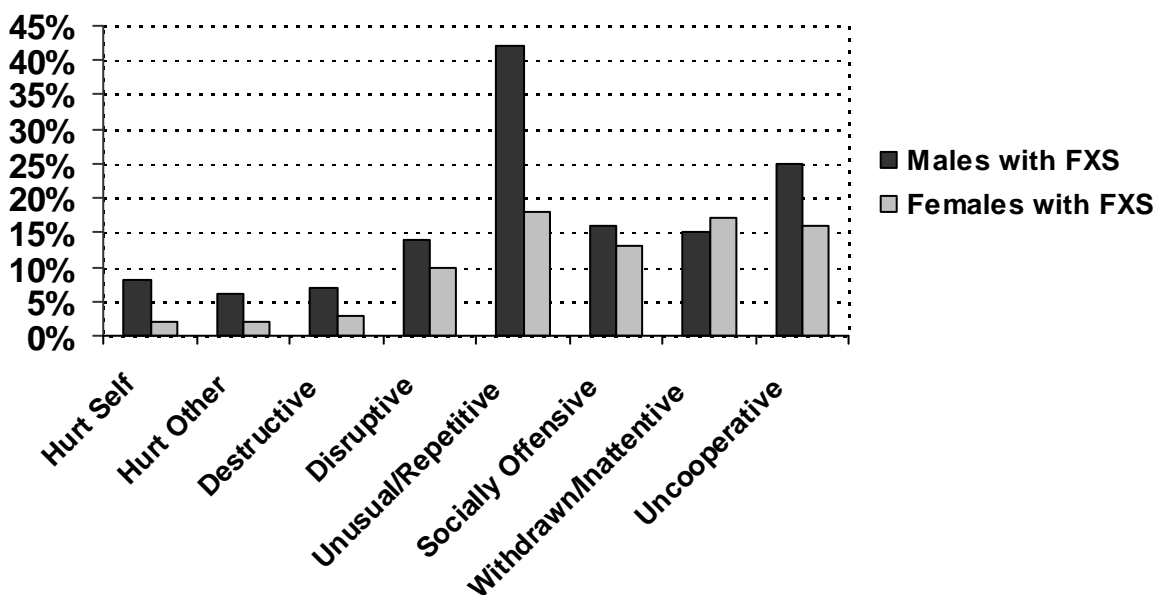
We calculated the number of days on which mothers reported that their son or daughter with FXS exhibited behavior problems. On average, mothers reported that their son or daughter exhibited at least one behavior problem on 4 days during the 8-day study period. As shown in Figure 7, only 16% of mothers reported that their son or daughter did not have any behavior problems during the study. The large majority (84%) of mothers reported behavior problems on at least one day, and 14% of mothers reported behavior problems on all 8 days.

**Figure 7. Percent of Mothers Reporting at Least One Behavior Problem**



We also calculated the number of days on which mothers reported that their son or daughter experienced each type of behavior problem.

**Figure 8. Percent of Days by Type of Behavior Problem**



As presented in Figure 8, the most common behavior problem for males was unusual or repetitive behavior, followed by uncooperative behavior. For females, the most common type of behavior problems were unusual or repetitive behaviors, withdrawn/inattentive behavior, and uncooperative behaviors. On average, behavior problems involving self-injury, aggression to others, or destroying property occurred on fewer than 8% of the days for both genders. Males exhibited unusual or repetitive behavior problems on more than twice as many days as females.

### **Section III: Cortisol**

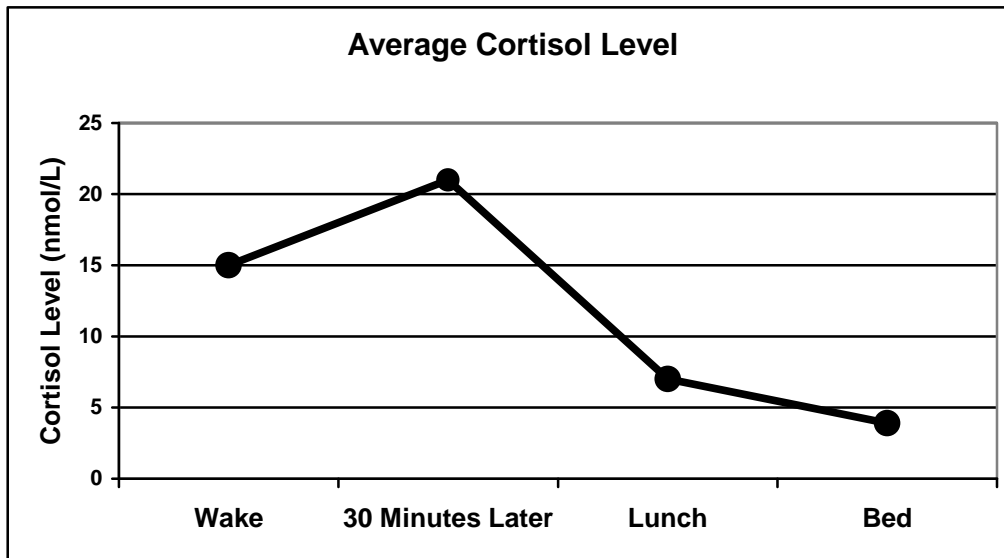
Cortisol is a hormone that is released by the body in response to stress. Researchers are interested in cortisol because it serves as a biological marker for stress. Cortisol has an important role in physiological systems that affect health, such as the body's immune response to the threat of illness.

Cortisol is contained in saliva. In our study, we measured the cortisol levels of mothers by asking them to provide saliva samples at four points during the day (waking up, 30 minutes after awakening, lunch, and bed time). Mothers provided saliva at these times on 4 days of the 8-day study. Thus, we were able to obtain 16 cortisol samples to measure how stress fluctuates from day to day.

Following a stressful day, individuals often show a different pattern of cortisol. A lower level of cortisol at awakening is a common response to chronic stress and is associated with feeling sleepy and tired upon waking up and anxiety, exhaustion, and poor health the previous day. Given the high level of daily stress of mothers of adolescents and adults with FXS, it is interesting to examine fluctuations in their daily patterns of cortisol.

The comparison group provides a benchmark of “average” levels of cortisol. Figure 9 shows the “average” cortisol pattern in the comparison group. As shown in this figure, levels of cortisol change throughout the day. The typical pattern is for cortisol to be high at awakening, with a subsequent increase within the first 30 minutes, and then a steady decline until the end of the day. It is thought that cortisol starts high and peaks early in the day in order to help us get going in the morning, and then declines throughout the day as our body winds down and prepares for sleep.

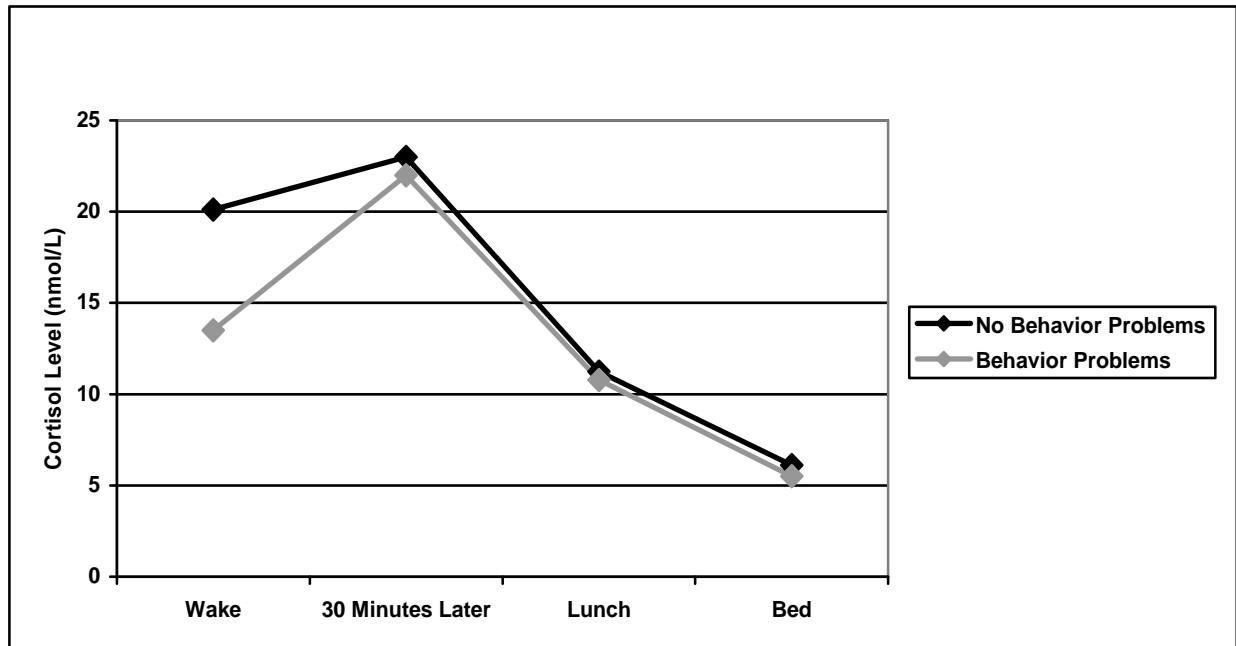
**Figure 9. Normal Daily Cortisol Rhythm**



Now, let's turn to cortisol patterns in mothers who have a son or daughter with FXS. We found that there is a great deal of variation in cortisol levels both within and between mothers in our sample. This variation reflects that although all of these mothers share the common experience of parenting an adolescent or adult with FXS, each of these mothers approaches and responds to challenges a bit differently. One of our goals is to understand how *child-related* stress impacts the physiology of mothers of adolescents and adults with FXS. In this report, we focus on the impact of the adolescent or adult son or daughter's internalizing behavior problems on mother's cortisol levels the next day.

Internalizing behavior problems include self-injury, withdrawn/inattentive behavior, and unusual or repetitive behaviors. As shown in Figure 10, we found that on days after their son or daughter had an episode of internalizing behavior problems, mothers exhibited a much lower cortisol level at awakening than on days following no internalizing behavior problems. This finding suggests that mothers are sleepier and more fatigued and have a difficult time getting up and going the morning after a day when their son or daughter had an episode of internalizing behavior problems. A similar pattern of reduced cortisol has been reported for people who experience stress at work, and has also been found in our research on mothers of adolescents and adults with autism spectrum disorders.

**Figure 10. Average Cortisol Level on days following No Internalizing Behavior Problems vs. At least One Internalizing Behavior Problem**



## **Section IV: Conclusions**

This report has described the types of daily events experienced by mothers of adolescents and adults with FXS compared to mothers of adolescents and adults who do not have a disability. Within our FXS sample, we also reported on the behavior problems among individuals with FXS.

There were a number of differences between the two groups of mothers in terms of their stressful daily experiences:

- Mothers of sons and daughters with FXS reported more fatigue and had to cut back on work more often, likely as the result of their special parenting responsibilities, than did the comparison group of mothers.
- Mothers of sons and daughters with FXS reported experiencing (and avoiding) more arguments and had more stress at home, at work, and with friends than did the comparison group of mothers.

We did not find differences between mothers of sons and daughters with FXS and the comparison group of mothers in positive daily interactions and activities.

- Mothers of sons and daughters with FXS reported spending a similar amount of time in physical activity or leisure as the comparison group.
- There were no group differences in the experience of a positive interaction or having a positive event happen to a friend or relative.
- Although mothers of sons and daughters with FXS reported *giving* more emotional support than mothers in the comparison group, they also reported *receiving* more emotional support than did the comparison group.

Thus, despite experiencing a greater number of stressful events and having higher demands on their time, mothers of sons and daughters with FXS still find time for positive interactions and activities.

Behavior problems in the son or daughter with FXS were frequently reported by mothers across the 8-day period of data collection. Unusual and repetitive behaviors were the most commonly reported problem, whereas hurting self, hurting others and destructive behaviors were less frequent.

In this report, we have also provided a summary of our preliminary findings about cortisol, a hormone associated with stress. Our data suggest that the behavior problems of the adolescent or adult son/daughter with FXS are associated with mothers' physiological response.

- Mothers evidenced lower cortisol at awakening on days following an episode of an internalizing behavior problem by their son or daughter with FXS than on days following no internalizing behavior problems.

This pattern of reduced cortisol has also been shown in other groups of individuals experiencing marked stress and is associated with feelings of sleepiness and fatigue upon awakening, and anxiety and physical health symptoms the previous day.

We hope to share these and other findings with researchers and service providers to give them new insights into the daily lives of mothers of adolescents and adults with FXS and their unique and varied parenting experiences. We hope that our findings can inform policy and create new programs and services to support families affected by FXS, such as providing respite care programs, behavioral interventions, and developing family-friendly employee policies at the workplace. Our ultimate goal is to improve the quality of life and well-being of adolescents and adults with FXS and their families.