



## INTERACTIONAL STYLE AND SUBJECTIVE STRESS IN MOTHERS OF YOUNG CHILDREN WITH FRAGILE X SYNDROME, DOWN'S SYNDROME OR TYPICAL DEVELOPMENT

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### Abstract:

This study examined the interactional style of mothers of boys with Fragile X syndrome (FXS) and mothers of children with Down's syndrome (DS) or developmentally typical children ( $N=33$  mother-child-dyads) in different situations (matching task; playing with toys; request). The average developmental age of the children was 29 months. Mothers completed a self-report scale for maternal stress. Directive and restrictive parenting behaviors were used more frequently by mothers of children with FXS. Mothers of children with DS tended to use more confirming and praising behaviors. Directive and restrictive behaviors were negatively associated with children's attention and positively associated with child behavioral problems and maternal stress. Implications of these findings are discussed.

**Keywords:** maternal interactional style, Fragile X syndrome, Down's syndrome, maternal stress

### Introduction

#### Factors that Influence Child Development

Vygotskij theorized that early reciprocal interactions between mother and child play an important role in child development. When playing with a 'competent partner' a child is supported in regulation processes and may thus succeed at tasks that he or she could not manage alone (Vygotskij, 2010). According to the transactional model, (Sameroff & Chandler, 1975) development is shaped not only by the extent to which the child's

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environment satisfies his or her needs, but also by the extent to which the child can influence his or her environment. Guralnick's (2011) 'developmental systems approach' identifies three main, interacting influences on child development: child characteristics, family interaction patterns and parental resources. Guralnick stressed that alongside parental intellectual capacities and educational background, family wellbeing and parental locus of control were important factors in child development. Ispa et al. (2002) revealed a positive correlation between maternal alienation and stress reactivity. Further studies have shown that maternal proneness to stress is negatively associated with sensitivity and warmth in maternal interactions with the child (Belsky et al., 1995; Mangelsdorf et al., 1990).

Research findings suggest that it is not simply the mother's presence that provides support for the child; how she interacts with the child is also important. Roggman et al. (2013) identified four aspects of supportive parenting:

- *"Affection: Warmth, physical closeness, and positive expressions towards child*
- *Responsiveness: Responding to child's cues, emotions, words, interests, and behaviors*
- *Encouragement: Active support of exploration, effort, skills, initiative, curiosity, creativity, and play*
- *Teaching: Shared conversation and play, cognitive stimulation, explanations, and questions"*

(Roggman et al., 2013, p. 2)

Mothers who show these or similar qualities in interactions with their children tend to have children who achieve language milestones earlier (Tamis-LeMonda et al., 2011) and display better cognitive (Bernier et al., 2010; Hubbs-Tait et al., 2002) and social (Ispa et al., 2004; Kelley et al., 2000) skills.

### **Knowledge of Syndrome-Specific Interactions**

Down's syndrome (DS) and Fragile X syndrome (FXS) are the main genetic causes of intellectual disability. Both syndromes are associated with a specific behavioral phenotype. Children with FXS have mild to moderate intellectual disability, an inhibitory control deficit and show hyperactivity and gaze avoidance (Backes et al., 2000; Hatton et al., 2002; Kau et al., 2004; Langthorne & McGill, 2012). These specific behaviors may be caused by deficits in executive functions (Cornish et al., 2004). Children with DS often have problems with emotion regulation. Children with DS show lower task motivation and reduced tolerance of frustration compared with children with other forms of an intellectual disability. There is also evidence that children with DS have relatively good

social skills (Berger & Cunnigham, 1981; Dykens, 1994; Fidler et al., 2006). As a consequence, children with DS are more likely to seek help from adults when confronted with challenging tasks (Jahromy et al., 2008; Kasari & Freeman, 2001; Pitcairn & Wishart, 1994).

Hodapp (1997) distinguished between the direct and indirect effects of a genetic disposition: direct effects are behaviors which are more probable in people with the syndrome whereas indirect effects can be described as reactions of the environment to a person with the syndrome. Although there is strong evidence that parent-child-interactions have a great impact on child development little is known about specific relationships between behavioral phenotypes and parenting styles.

Until now there has been little data available on the indirect effects of genetic disorders on mother-child-interactions. Wheeler et al. (2007) logged the frequency of maintaining behavior, directive behavior, scaffolding and restriction in 24 mother-child-pairs observed in a 10-minute play context and a 1-hour unstructured, everyday context. The mothers primarily used maintaining behaviors. They made less use of directive behavior in the unstructured, naturalistic context than in the structured play context. In another study Wheeler et al. (2010) observed 46 children with FXS and their mothers in a frustrating task situation. The children were given a box containing an attractive toy which could only be opened using a key which was given to the mother. Mothers were instructed only to intervene when the child got too frustrated. The mothers mainly showed encouraging and directive behaviors, making demands and suggestions. The only investigation to compare the interactional style of mothers with children with FXS with developmentally typical children (Sterling et al., 2012) used a within-family design. Sterling et al. compared how mothers interacted with their children with FXS and with their siblings without disability. Mothers used a responsive style with both children but used more behavior management strategies and a less conversational style with the child with FXS. These findings indicate that mothers react to the child's inhibitory control deficits by regulating task performances more closely.

Considerably more studies have investigated the interaction of mothers and their children with DS. Roach et al. (1998) compared the interactions of 28 mothers and their children with DS with those of mothers and their developmentally typical children. Mothers of children with DS were more directive but also more supportive. In a longitudinal study Blacher et al. (2013) examined parenting behavior during structured and unstructured activities. The 183 participants included mothers of children with autism spectrum disorders, cerebral palsy, DS, undifferentiated developmental delay and typical cognitive development. The group of mothers with children with DS used more positive,

encouraging behaviors. These findings suggest that mothers of children with DS combine a directive interactional style with supportive and motivating behaviors.

What data we have indicate that mothers of children with a specific syndrome adapt to the behavioral characteristics of their child. Mothers of children with FXS are more likely to use behavior management strategies, presumably in response to their children's impulsivity, whereas mothers of children with DS seem to encourage their children more often to try to increase their motivation and endurance.

The aim of the study was to examine maternal parenting in three different groups of mother-child dyads: mothers and their children with FXS, mothers and their children with DS and mothers and their developmentally typical (TYP) children. The conceptual framework for the study recognizes various influences on maternal interactional style: situation or context (matching task; free play; request), child characteristics during the various situations (attention; behavioral adaptation) and maternal wellbeing (parenting stress).

The following research questions guided the study:

- *Do mothers of boys with FXS use a different interactional style from mothers of children with DS or TYP children? If so, how does their interactional style differ?*
- *What influence does situation have on maternal parenting?*
- *Is maternal parenting style correlated with child behavior variables?*
- *Does maternal wellbeing of mothers with FXS boys differ from maternal wellbeing of mothers of children with DS or TYP children?*
- *Is maternal wellbeing correlated with parenting style?*

## **Material and Methods**

### **Participants**

The participants were 11 mothers of children with FXS, 11 mothers of children with DS and 11 mothers of developmentally typical children. Families were recruited from Fragile X and Down's syndrome listservs and websites in Germany and Switzerland. The families of the control group were contacted via postings in day care centers. None of the mothers of the children with FXS had full mutation status. Demographic information for all groups is provided in Table 1.

Children's developmental age was determined from a child development questionnaire (*Elternfragebogen zur kindlichen Entwicklung*; Brandtstetter, Bode & Ireton, 2003, the German version of the Child Development Inventory, Ireton, 1992) which was filled out by the mother. As there is evidence that children with DS have significantly lower expressive language skills than receptive skills (Sarimski, 2014), developmental age

was determined without considering score on the expressive language scale. The Shapiro-Wilk test was used to confirm that developmental age was normally distributed in all groups. ANOVA confirmed that there were no group differences in developmental age.

	FXS	DS	Typ
<b>Number of mother-child-pairs</b>	n = 11	n = 11	n = 11
<b>Maternal age in years M</b>	34	41	33
<b>German ethnicity</b>	10	10	9
<b>Other ethnicity</b>	1	1	2
<b>In a stable relationship</b>	10	7	11
<b>Single parent</b>	1	4	0
<b>Ø Number of children in family</b>	2.2	2.2	1.9
<b>Maternal educational attainment: junior high school</b>	2	0	1
<b>Maternal educational attainment: Middle / High school</b>	5	3	0
<b>Maternal educational attainment: Matriculation standard</b>	4	8	10
<b>Full-time employment</b>	1	2	0
<b>Part-time employment</b>	7	3	8
<b>Homemaker</b>	3	5	3
<b>In further education or training</b>	0	1	0
<b>Number of boys</b>	11	6	3
<b>Number of girls</b>	0	5	8
<b>Child's age in months, M (SD)</b>	57 (13.71)	51 (9.70)	32 (5.03)
<b>Child's developmental age (excluding expressive speech) in months, M (SD)</b>	28 (5.21)	27(4.62)	31(5.03)

**Table 1:** Demographic profile of mother-child dyads (n=33)

#### Procedures

Mother-child dyads were visited in their homes. Behavior was videotaped in three situations: a structured 10-minute session in which the mother was instructed to explain a matching task to her child, a 10-minute play session for which toys (vehicles, a house, Lego and figurines) were available and, finally, a request situation, in which mothers were instructed to ask the child to tidy up the toys. After the observation period mothers were asked to fill out questionnaires about the child's development and their own wellbeing. The average length of visit was 2 hours.

## Instrumentation

**Maternal measures.** Maternal behavior was observed and coded using an adaptation of the coding system developed by Wheeler and colleagues (2007). There were four main categories, each containing four or five items: *maintaining behaviors*, *regulation of emotions*, *directive behaviors* and *restriction*. A rest category served to code maternal behaviors which did not involve interaction with the child (e.g. cleaning one's nose). Maintaining behaviors tell the child that his or her interests, thoughts and actions are important (Wheeler et al., 2007). This category was used when the mothers made comments on the child's activities or made suggestions about how the child could extend his or her activity. Regulation of emotions refers to maternal attempts to increase the child's positive affect or reduce or prevent negative affect e.g. encouraging, praising or comforting the child or supporting the child to handle toys. Directive behaviors serve to structure the situation and to focus the child's attention on what is relevant. Directive behaviors can be described as responsive to the child's behavior or unresponsive if the mother is not taking into account her child's activity (Landry et al., 2000). Maternal demands or requests for information were coded as directive behaviors. Restrictions are another way of directing or restricting the child's actions but this term is used for behaviors which cannot be described as helpful to the child's development. This category was used when the mother limited the child's actions, e.g. by holding the child, or when the mother disciplined the child. All videos were coded using a time-sampling method with 5-seconds segments.

We also used the PICCOLO rating scale (Roggman et al., 2013) to rate four aspects of supportive parenting: *affection*, *responsiveness*, *encouragement* and *teaching*. Each domain consists of 7-8 behavioral items. Items are rated as not observable (0), partly observable (1) or full observable (2). PICCOLO was developed for play situations with pretend play toys or manipulative toys and picture book situations. Although PICCOLO was only appropriate to our play situation according to the PICCOLO Handbook (Roggman et al., 2013), maternal behavior in all three situations was rated using it. PICCOLO is a reliable instrument. Based on data from over 2000 families the inter-rater reliability for pairs of observers averaged 75% across all four domains; the internal consistency (average Cronbach's  $\alpha = .78$ ) is also very good.

Maternal stress was measured by using the Elterliches Belastungsinventar (EBI; Tröster, 2011), a German version of the Parenting Stress Index (PSI; Abidin, 1995). The EBI consists of 48 items organized into two domains: stress linked with the child's characteristics (child domain) and parental distress (parent domain). In this study the EBI had satisfactory internal consistency (average of Cronbach's  $\alpha$  for both domains = .92).

**Child measures.** Children's behavior was coded using two categories: *attention* and *behavioral adaption*. *Attention* was coded as *on-* and *off-task behaviors* in relation to engagement with the materials available in the situation (matching task or toys). Children's behavior was coded as on-task when the child was dealing with the material in a non-specific way. *Behavioral adaption* was coded as *rule-consistent* or *not rule-consistent*. The child's behavior was coded as rule-consistent when it was in accordance with the object of the task (matching task situation) or when it was non-destructive (play situation). In the request situation behavior was coded as rule-consistent when the child obeyed the mother's instruction to tidy up. When the child was coded as *off-task* his or her behavior was by definition *not rule-consistent*. An event-sampling method was used to code children's behavior.

## Data Analysis

Two researchers observed all three situations and inter-rater agreement were calculated for 12% of the video data. The two researchers were trained in use of the coding system and the PICCOLO rating scale. A minimum inter-rater agreement coefficient of .70 was maintained for all interaction behaviors across all the data.

As a 5s coding segment could contain behaviors from several categories the total number of codes assigned was not the same for all situations or dyads and so inter-group-comparisons could not be presented as percentages. We therefore used the average number of instances of a given code per minute of maternal behavior in all analyses. This was a way of transforming observations of specific behaviors into meaningful, comparable data. Since the data were not normally distributed we used a non-parametric test, the Kruskal-Wallis test. This analysis shows via post hoc pairwise comparisons which groups differ from each other.

Spearman's rank correlations were run to identify relationships among the variables (e.g. maternal stress and interactional style). Effects were rated as follows:  $.0 \leq r_s \leq .2$ , no correlation;  $.2 < r_s \leq .5$ , little correlation;  $.5 < r_s \leq .8$ , clear correlation and  $.8 < r_s \leq 1.0$ , high correlation.

## Results

### Maternal Behavior

Analysis of the distribution of maternal interactive behaviors indicated that in all groups the most common behaviors were maintaining behaviors (range: 35-49% across all groups) and directive behaviors (range: 37-47% across all groups). Regulation of the child's

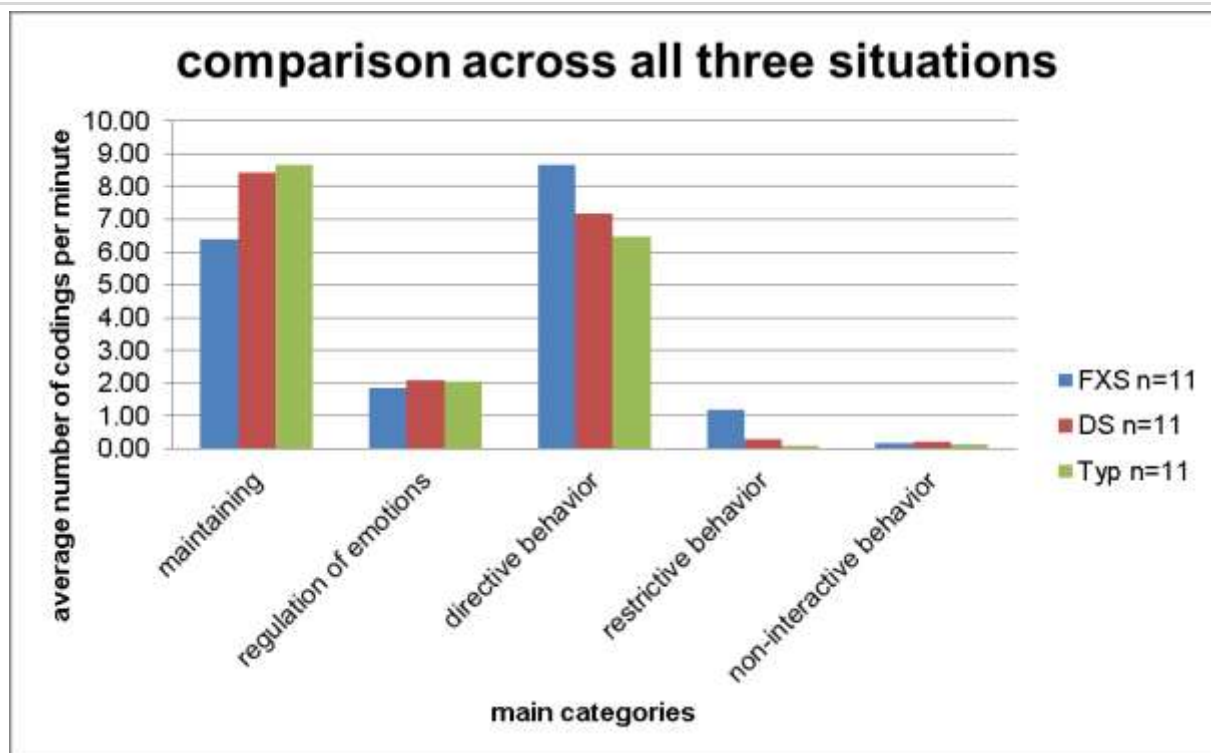
emotions (range: 10–12% across all groups) was less frequent and restrictive behaviors (range: 1–7% across all groups) were rarely observed. Over all situations mothers of children with FXS or DS made greater use of more directive behaviors than mothers of developmentally typical children.

Mothers of children with FXS made less use of maintaining behaviors than mothers in the other two groups but made more frequent use of directive and restrictive behaviors. The interactional behavior of mothers of children with DS and mothers of developmentally typical children was similar with respect to the main categories; however mothers of children with DS tended to use encouraging behaviors, such as praising and confirming their children, more often. Table 2 gives the average number of instances of each code per minute for mothers for all the situations combined (also displayed in Figure 1).

Category	FXS M (SD)	DS M (SD)	Typ M (SD)	Pairs	Significance	Test statistics
<b>Maintaining behavior</b>	6.38	8.44	8.66	FXS – DS	.006	-11.273**
	(1.83)	(1.37)	(0.58)	FXS – Typ	.003	-12.455**
				DS – Typ	.774	-1.182
<b>Regulation of emotions</b>	1.85 (0.65)	2.10 (0.90)	2.07 (0.58)	n.s.	.565	1.143
<b>Directive behavior</b>	8.64	7.20	6.49	FXS – DS	.103	6.727
	(1.67)	(1.31)	(1.42)	FXS – Typ	.004	11.818**
				DS – Typ	.217	5.091
<b>Restrictive behavior</b>	1.20	0.28	0.11	FXS – DS	.018	9.773*
	(1.21)	(0.39)	(0.09)	FXS – Typ	.001	13.818**
				DS – Typ	.326	4.045
<b>Non-interactive behavior</b>	0.17 (0.14)	0.20 (0.14)	0.14 (0.11)	n.s.	.651	.858

**Table 2:** Group comparisons (n = 11 per group) for main categories; average instances of each code per minute across all situations; \*p < .05; \*\*p < .01; n.s. = not significant; FXS = Fragile X; DS = Down’s syndrome; Typ = developmentally typical





**Figure 1:** Group comparisons of maternal behavior (n = 11 per group) for main categories; average instances of each code per minute across all situations; FXS = Fragile X; DS = Down's syndrome; Typ = developmentally typical

In all three groups the distribution of behavior varied according to situation. Directive behavior was used more frequently in the more challenging situations (matching task and request situation) (range: 51–57%) than in the play situation (range: 19–30%). For reasons of space detailed situation-specific data are not presented.

Scores from the PICCOLO rating scale indicated that mothers of children with DS or FXS were less likely to support their children to do things on their own than the mothers of developmentally typical children. Mothers of children with FXS had lower scores for the item *Adjusts pace or activity according to child's interests or needs*, which is part of the responsiveness category. This group also had lower score for the items *Speaks in a warm tone of voice* and *Shows emotional warmth*. During the matching task situation mothers of children with FXS spent less time following what their child was trying to do. Table 3 presents data for a selection of the PICCOLO items.

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Item (Domain)	Situation	FXS M (SD)	DS M (SD)	Typ M (SD)	Pairs	Significance	Test statistics
Speaks in a warm tone of voice (Affection)	S1	1.45 (0.52)	1.91 (0.30)	2.00 (0.00)	FXS – DS	.010	-7.500*
					FXS – Typ	.002	-9.000**
					DS – Typ	.608	-1.500
	S2	1.64 (0.51)	2.00 (0.54)	2.00 (0.00)	FXS – DS	.010	-6.000*
					FXS – Typ	.010	-6.000*
					DS – Typ	1.000	.000
	S3	1.36 (0.67)	1.91 (0.30)	2.00 (0.00)	FXS – DS	.010	-7.591*
					FXS – Typ	.002	-9.045**
					DS – Typ	.620	-1.455
Shows emotional warmth (Affection)	S1	1.45 (0.52)	2.00 (0.00)	2.00 (0.00)	FXS – DS	.001	-9.000**
					FXS – Typ	.001	-9.000**
					DS – Typ	1.000	.000
	S2	1.55 (0.52)	1.91 (0.30)	2.00 (0.00)	FXS – DS	.029	-6.000*
					FXS – Typ	.006	-7.500**
					DS – Typ	.586	-1.500
	S3	1.45 (0.69)	2.00 (0.00)	2.00 (0.00)	FXS – DS	.003	-7.500**
					FXS – Typ	.003	-7.500**
					DS – Typ	1.000	.000
Adjusts pace or activity according to child’s interests or needs (Responsiveness)	S1	1.27 (0.65)	1.91 (0.30)	2.00 (0.00)	FXS – DS	.003	-9.091**
					FXS – Typ	.001	-10.545**
					DS – Typ	.636	-1.455
Follows what the child is trying to do (Responsiveness)	S1	0.45 (0.52)	1.00 (0.45)	1.00 (0.63)	FXS – DS	.029	-7.636*
					FXS – Typ	.024	-7.909*
					DS – Typ	.938	.273
Supports the child to do things on his or her own (Encouragement)	S1	0.55 (0.52)	0.73 (0.79)	1.55 (0.52)	FXS – DS	.586	-2.091
					FXS – Typ	.002	-11.955**
					DS – Typ	.010	-9.864*
	S2	1.45 (0.52)	1.73 (0.47)	2.00 (0.00)	FXS – DS	.157	-4.500
					FXS – Typ	.005	-9.000**
					DS – Typ	.157	-4.500
	S3	0.36 (0.67)	0.45 (0.69)	1.18 (0.75)	FXS – DS	.744	-1.227
					FXS – Typ	.011	-9.545*
					DS – Typ	.027	-8.318*

**Table 3:** Scores per group (n = 11 per group); \*p < .05; \*\*p < .01; n.s. = not significant; FXS = Fragile X; DS = Down’s syndrome; Typ = developmentally typical; S1 = matching task situation, S2 = play situation, S3 = request situation

### Children's Behavior

On average off-task behaviors were most frequent in children with FXS. Children with FXS spent more time in off-task behaviors during the matching task situation and the play situation than developmentally typical children. Both groups of children with disability spent a lower proportion of their time on task than the group of developmentally typical children. For all groups the percentage of on-task behavior was highest in the play situation. Table 4 presents data for on-task behavior.

Situation	FXS M (SD)	DS M (SD)	Typ M (SD)	Pairs	Significance	Test statistics
Matching task	88%	91%	97%	FXS – DS	.929	-.364
	(15)	(7)	(10)	FXS – Typ	.007	-10.955**
				DS – Typ	.009	-10.591**
Play situation	95%	97%	100%	FXS – DS	.033	-7.909*
	(6)	(7)	(1)	FXS – Typ	.014	-9.136*
				DS – Typ	.740	-1.227
Request situation	83%	95%	93%	n.s.	.223	3.003
	(19)	(11)	(16)			

**Table 4:** Children's on-task behavior by group (n = 11); \*p < .05; \*\*p < .01; n.s. = not significant; FXS = Fragile X; DS = Down's syndrome; Typ = developmentally typical

There were also situational differences in *behavioral adaption*: in all groups rule-consistent behavior was more common in the play situation than the other situations. Children with FXS were less rule-consistent than the other groups. There were group differences in behavioral adaptation in the play situation. Table 5 presents data on rule-consistent behavior.

Situation	FXS M (SD)	DS M (SD)	Typ M (SD)	Pairs	Significance	Test statistics
Matching task	78%	90%	94%	n.s.	.449	1,600
	(31)	(8)	(6)			
Play situation	88%	96%	100%	FXS – DS	.005	-10,864**
	(19)	(10)	(1)	FXS – Typ	.001	-13,136**
				DS – Typ	.557	-2,273
Request situation	55%	66%	63%	n.s.	.656	.842
	(34)	(32)	(27)			

**Table 5:** Children's rule-consistent behavior by group (n = 11); \*p < .05; \*\*p < .01; n.s. = not significant; FXS = Fragile X; DS = Down's syndrome; Typ = developmentally typical

### Relationship between Child's and Mother's Behavior

In order to identify relations between maternal and child variables Spearman's rank correlations were used to evaluate relationships between maternal and child variables. There was a small positive correlation between children's on-task behavior and maternal maintaining behaviors ( $r = .370, p < .05$ ) and a clear correlation between on-task behavior and maternal encouragement ( $r = .581, p < .01$ ). On-task behavior was clearly negatively correlated with maternal restriction ( $r = -.618, p < .01$ ). Rule-consistent behavior was positively correlated with maternal maintaining behavior ( $r = .473, p < .01$ ), regulation of emotion ( $r = .475, p < .01$ ) and encouragement ( $r = .676, p < .01$ ). Children's rule-consistent behavior was negatively correlated with maternal directive ( $r = -.522, p < .01$ ) and restrictive ( $r = -.656, p < .01$ ) behaviors.

### Maternal stress

The EBI (the German version of the PSI) differentiates between stress caused by the child's behavior (child domain) and parental distress (parent domain). Total parental stress levels were similar in mothers of children with FXS and DS. Mothers with developmentally typical children reported the lowest levels of stress. Mothers of children with FXS reported high stress arising from the *hyperactivity or distractibility* of their child. Mothers of children with DS reported more stress related to how demanding their child was than mothers in the other groups. Table 6 shows the differences between the three groups. Both groups of mothers of children with disability reported high levels of stress in the *role restriction* subscale of the parent domain. Mothers of children with FXS also reported high levels of stress in the *competence* and *depression* subscales. Mothers of children with FXS reported more *competence-related* stress than mothers of children with DS. These data are presented in Table 6 and Table 7.

EBI Subscales	FXS	DS	Typ	Pairs	Significance	Test statistics
Child domain	M (SD)	M (SD)	M (SD)			
<b>Hyperactivity or distractibility</b>	8.36	6.64	4.73	Typ – DS	.038	8.318*
	(1.12)	(1.96)	(1.35)	Typ – FXS	.000	16.227***
				DS – FXS	.049	7.909*
<b>Mood</b>	6.73	4.55	4.27	Typ – DS	.798	1.045
	(2.15)	(1.57)	(1.90)	Typ – FXS	.009	10.682**
				DS – FXS	.018	9.636*
<b>Acceptability</b>	8.09	7.45	5.00	Typ – DS	.001	13.227**
	(0.30)	(1.04)	(0.89)	Typ – FXS	.000	18.237***
				DS – FXS	.197	5.045
<b>Demandingness</b>	8.55	7.91	6.45	Typ – DS	.005	10.864**
	(0.93)	(2.02)	(1.21)	Typ – FXS	.001	13.409**
				DS – FXS	.515	2.545

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<b>Adaptability</b>	7.18	5.91	5.27	Typ – DS	.263	4.545
	(1.47)	(2.34)	(1.55)	Typ – FXS	.012	10.182*
				DS – FXS	.165	5.636

**Table 6:** Group comparisons of maternal stress (EBI subscales, child domain); Stanines are presented; \* $p < .05$ ; \*\* $p < .01$ , \*\*\* $p < .001$ ; n.s. = not significant; FXS = Fragile X; DS = Down’s syndrome; Typ = developmentally typical

Subscales EBI	FXS	DS	Typ	Pairs	Significance	Test statistics
Parent domain	M (SD)	M (SD)	M (SD)			
<b>Parental attachment</b>	7.00	6.45	5.00	Typ – DS	.069	7.364
	(1.34)	(2.12)	(1.55)	Typ – FXS	.009	10.636**
				DS – FXS	.419	3.273
<b>Isolation</b>	6.09	6.82	4.18	Typ – DS	.003	12.045**
	(2.47)	(1.66)	(1.08)	Typ – FXS	.031	8.818*
				DS – FXS	.429	-3.227
<b>Competence</b>	8.00	5.73	4.82	Typ – DS	.332	3.955
	(0.89)	(2.20)	(1.83)	Typ – FXS	.000	14.455***
				DS – FXS	.010	10.500*
<b>Depression</b>	8.00	6.64	6.73	n.s.	.078	5.102
	(1.34)	(2.58)	(1.10)			
<b>Health</b>	6.73	6.55	5.36	n.s.	.215	3.075
	(1.01)	(2.25)	(2.06)			
<b>Role restriction</b>	7.27	7.09	4.82	Typ – DS	.004	11.727**
	(1.74)	(1.38)	(1.25)	Typ – FXS	.001	12.955**
				DS – FXS	.763	1.227
<b>Spouse</b>	6.00	6.44	5.73	n.s.	.790	.472
	(1.55)	(1.88)	(1.35)			

**Table 7:** Group comparisons of maternal stress (EBI subscales, parent domain); Stanines are presented; \* $p < .05$ ; \*\* $p < .01$ , \*\*\* $p < .001$ ; n.s. = not significant; FXS = Fragile X; DS = Down’s syndrome; Typ = developmentally typical

**Maternal Stress and Parenting Style**

There was a negative correlation ( $r = -.463, p < .01$ ) between total EBI score and maternal maintaining behaviors. Total EBI score was positively correlated with maternal directive behavior ( $r = .502, p < .01$ ) and restrictive behavior ( $r = .502, p < .01$ ). EBI score was also related to the PICCOLO ratings. EBI score was clearly negatively correlated with maternal responsiveness ( $r = .482, p < .01$ ) and encouragement ( $r = .582, p < .01$ ).

## Discussion

### Mother-Child Interaction

The goal of the study was to explore patterns of maternal behavior in mother of children with two different genetic syndromes. We found that mothers of children with FXS or DS adapt their behavior to the syndrome-specific characteristics of their child.

Wheeler et al. (2007) showed that mothers of children with FXS tend to use an interactive style that combines high proportions of maintaining (67%) and directive (27%) behaviors; we corroborated this finding in a play context; however we found that in a matching task situation and a request situation these mothers made much more use of directive behavior compared to the play situation. Wheeler et al. (2010) confronted children with FXS with an unsolvable task; in this situation 26% of the mothers used negative behavioral control and 20% used only directive and negative control behaviors. Taken together these findings suggest that in challenging situations mothers of children with FXS make more extensive use of directive and controlling behaviors than mothers of children with DS or mothers of developmentally typical children. We found that compared with mothers of children with DS and mothers of developmentally typical children the mothers of children with FXS used a generally more controlling style during interactions with their children. Based on observations of mothers with children with FXS and their developmentally typical sibling Sterling et al. (2012) reached a similar conclusion; their mothers were responsive to both children but were less flexible when interacting with the child with FXS. Sterling et al. characterized the behavior of mothers towards the child with FXS as less conversational and more directive. Our results, based on the coding system and the PICCOLO rating scale confirm the earlier findings. One possible explanation for the pattern of maternal interactions with children with FXS is that these children tend to be impulsive and mothers feel a greater need to structure and direct their child's activity. The results of this study indicate that mothers of children with DS made more use of directive behaviors than mothers of developmentally typical children; however on the whole there were few differences between the interactions of mothers of children with DS and mothers of developmentally typical children. Directive and responsive behavior are not mutually exclusive and both types of behavior were used extensively by mothers of children with DS. Tannock (1989) and Roach et al. (1998) reported similar findings. We also observed that mothers of children with DS confirmed and praised their children more often than mothers in the other groups. This may be because children with DS often have motivational problems and can be described as externally oriented. Confirming and praising the child continuously may be the mother's

way of trying to motivate her child to persist. It is likely that children with DS will remain dependent on this form of external regulation.

Analysis of the relationship between child and maternal behavior indicated that maternal restrictive behaviors were associated with off-task behavior in the child, whereas on-task behavior was related to maternal behaviors that encouraged the child to handle toys or to extend his or her activity. Children with FXS tend to have problems switching attention within a task (Cornish et al., 2004), so it is possible that if a mother directs the child's attention too forcibly by offering new toys or making demands the child's attentional system may be overloaded and consequently he or she may turn away. Attentional problems may also arise if the mother does not respond to the child's interests. In this study the mothers of children with FXS followed their child's goals and activity to a lesser extent than mothers in the other groups. This may be because they felt they needed to be more controlling in order to reduce their child's impulsiveness.

### **Maternal Wellbeing and Maternal Behaviors**

Both groups of mothers with children with disability reported stress related to parenting and child care. Mothers of both groups reported experiencing stress caused by role restriction (parent domain). Mothers of children with FXS also reported high stress related to child behaviors such as hyperactivity. This finding has been reported previously (Sarimski, 2010; Wheeler et al., 2008). A study based on qualitative interviews (Wheeler et al., 2008) concluded that mothers of children with FXS experienced stress when dealing with their child's challenging behaviors and when they did not know how to help or control their child. Moreover, one third of mothers reported that parenting and family life in general were stressful. Amongst the reasons given were having too little time and the feeling of being pulled in too many directions (Wheeler et al., 2008).

In this study both groups of mothers of children with disabilities had similar EBI scores. The findings that mothers of children with FXS made more use of restrictive behaviors and that restrictive behavior was positively correlated with maternal stress suggest that in mothers of children with FXS child characteristics are the main source of stress.

### **Limitations**

Our results only provide a glimpse into how different genetic syndromes influence interactional style in mother-child dyads, owing to the use of a small, selective sample and a single observation session per dyad. It is also important to bear in mind that mothers participated voluntarily; it seems likely that those who participated were probably relatively secure in their parenting competence. Another critical issue is that data on

children's developmental age were based on a questionnaire rather than direct testing, which would have had greater validity. The matching task may not have been similarly demanding for all the children as their chronological age varied. Given that behavior may be modified under open observation it would have been useful to get more detailed information about the children's behavior, e.g. via the Child Behavior Check List (Achenbach & Rescorla, 2000). Finally, sequence analysis is needed to determine whether specific maternal behaviors tend to be followed by specific child behaviors (and *vice versa*); such analysis would have provided more insight into interaction patterns.

### **Conclusion**

These limitations notwithstanding, this study provides important information about how maternal interaction behavior is related to factors such as situation, intellectual disability, behavioral phenotype and maternal stress.

Firstly, in all three groups maternal behavior was influenced by the context or situation. Secondly, both groups of mothers of children with disability made more use of directive behaviors than mothers of developmentally typical children. This finding makes it clear that some differences in maternal behavior are related to having a child with an intellectual disability. Our third finding was that maternal behaviors can be related to the behavioral phenotype associated with the child's genetic syndrome. Mothers of children with DS made heavy use of confirmation and praise and this could be linked to the motivational problems and external orientation which are known to characterize DS. The extensive use of directive and restrictive behaviors by mothers of children with FXS may be due to the inhibitory control deficit and hyperactivity these children display. Finally, the study demonstrated that maternal stress affects mother-child interactions.

These findings have implications for practice and research. Families who need help with interacting with a child with a disability should be offered syndrome-specific guidance. There also seems to be a need to find ways of reducing parental stress and the first step towards this is to identify the main sources of stress. Further research might include an investigation with a larger sample and extending the comparisons to include other syndromes, e.g. Prader-Willi syndrome. Finally, a longitudinal investigation could be used to determine how parental factors influence child development.

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Dissertation an der Pädagogischen Hochschule Heidelberg. Online in: <http://opus.ph-heidelberg.de/frontdoor/index/index/docId/146>

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## References

1. Achenbach T, Rescorla L, 2000. Manual for the ASEBA Preschool forms and Profiles, Burlington, VT, University of Vermont Department of Psychiatry
2. Backes M, Genc B, Schreck J, Doerfler W, Lehmkuhl G, von Gontard A, 2000. Cognitive and behavioral profile of Fragile X boys: correlations to molecular data. *American Journal of Medical Genetics* 95(2): 150–156. doi: 10.1002/1096-8628(20001113)95:2<150::AID-AJMG11>3.0.CO;2-1
3. Belsky J, Crnic K, Woodworth S, 1995. Personality and parenting: exploring the mediating role of transient mood and daily hassles. *Journal of Personality* 63(4): 905–929. doi: 10.1111/j.1467-6494.1995.tb00320.x
4. Berger J, Cunningham C, 1981. The development of eye contact between mothers and normal versus Down's syndrome infants. *Developmental Psychology* 17(5): 678–689. doi: 10.1037/0012-1649.17.5.678
5. Bernier A, Carlson S, Whipple N, 2010. From external regulation to self-regulation: early parenting precursors of young children's executive functioning. *Child Development* 81(1): 326–339
6. Blacher J, Baker B, Kaladjian A, 2013. Syndrome specificity and mother–child interactions: examining positive and negative parenting across contexts and time. *Journal of Autism and Developmental Disorders* 43(4): 761–774. doi: 10.1007/s10803-012-1605-x
7. Brandstetter G, Bode H, Ireton H, 2003. Elternfragebogen zur Entwicklung im Kleinkindalter (EFkE). Manual, Augsburg
8. Cornish K, Sudhalter V, Turk J, 2004. Attention and language in Fragile X. *Mental Retardation and Developmental Disabilities Research Reviews* 10(1): 11–16. doi: 10.1002/mrdd.20003
9. Dykens E, Hodapp R, Evans D, 1994. Adaptive behavior and Down syndrome. *American Journal on Mental Retardation* 98(5): 580–587
10. Fidler D, Hepburn S, Rogers S, 2006. Early learning and adaptive behaviour in toddlers with Down syndrome: Evidence for an emerging behavioural phenotype? *Down Syndrome Research and Practice* 9(3): 37–44. doi: 10.3104/reports.297

11. Gilmore L, Cuskelly M, 2011. Observational assessment and maternal reports of motivation in children and adolescents with Down syndrome. *American Journal on Intellectual and Developmental Disabilities* 116(2): 153–164. doi: 10.1352/1944-7558-116.2.153
12. Glenn S, Dayus B, Cunningham C, Horgan M, 2001. Mastery motivation in children with Down syndrome. *Down Syndrome Research and Practice* 7(2): 52–59. doi: 10.3104/reports.114
13. Guralnick M, 2011. Why early intervention works: a systems perspective. *Infants & Young Children* 24(1): 6–28. doi: 10.1097/IYC.0b013e3182002cfe
14. Hatton D, Hooper S, Bailey D, Skinner M, Sullivan K, Wheeler A, 2002. Problem behavior in boys with Fragile X syndrome. *American Journal of Medical Genetics* 108(2): 105–116. doi: 10.1002/ajmg.10216
15. Hodapp R, 1997. Direct and indirect behavioral effects of different genetic disorders of mental retardation. *American Journal of Mental Retardation: AJMR* 102(1): 67–79. doi: 10.1352/0895-8017(1997)102<0067:DAIBEO>2.0.CO;2
16. Hubbs-Tait L, McDonald Culp A, Culp R, Miller C, 2002. Relation of maternal cognitive stimulation, emotional support, and intrusive behavior during Head Start to children's kindergarten cognitive abilities. *Child Development* 73(1): 110–131. doi: 10.1111/1467-8624.00395
17. Ispa J, Fine M, Halgunseth L, Boyce L, Brooks-Gunn J, Brady-Smith C, 2004. Maternal intrusiveness, maternal warmth, and mother–toddler relationship outcomes: Variations across low-income ethnic and acculturation groups. *Child Development* 75(6): 1613–1631. doi: 10.1111/j.1467-8624.2004.00806.x
18. Ispa J, Fine M, Thornburg K, 2002. Maternal personality as a moderator of relations between difficult infant temperament and attachment security in low-income families. *Infant Mental Health Journal* 23(1-2): 130-144. doi: 10.1002/imhj.10008
19. Jahromi L, Gulrud A, Kasari C, 2008. Emotional competence in children with Down syndrome: negativity and regulation. *American Journal on Mental Retardation* 113(1): 32–43. doi: 10.1352/0895-8017(2008)113[32:ECICWD]2.0.CO;2
20. Kasari C, Freeman S, 2001. Task-related social behavior in children with Down syndrome. *American Journal on Mental Retardation* 106(3): 253–264. doi: 10.1352/0895-8017(2001)106<0253:TRSBIC>2.0.CO;2
21. Kau A, Tierney E, Bukelis I, Stump M, Kates W, Trescher W, Kaufmann W, 2004. Social behavior profile in young males with Fragile X syndrome: characteristics and specificity. *American Journal of Medical Genetics* 126: 9–17. doi: 10.1002/ajmg.a.20218

22. Kelley S, Brownell C, Campbell S, 2000. Mastery motivation and self-evaluative affect in toddlers: Longitudinal relations with maternal behavior. *Child Development* 71(4): 1061–1071. doi: 10.1111/1467-8624.00209
23. Landry S, Smith K, Swank P, Miller-Loncar C, 2000. Early maternal and child influences on children's later independent cognitive and social functioning. *Child Development* 71(2): 358–375. doi: 10.1111/1467-8624.00150
24. Langthorne P, McGill P, 2012. An indirect examination of the function of problem behavior associated with Fragile X syndrome and Smith-Magenis syndrome. *Journal of Autism and Developmental Disorders* 42(2): 201–209. doi: 10.1007/s10803-011-1229-6
25. Mangelsdorf S, Gunnar M, Kestenbaum R, Lang S, Debra A, 1990. Infant proneness-to-distress temperament, maternal personality, and mother-infant attachment: Associations and goodness of fit. *Child Development* 61(3): 820–831. doi: 10.2307/1130966
26. Pitcairn T, Wishart J, 1994. Reactions of young children with Down's syndrome to an impossible task. *British Journal of Developmental Psychology* 12(4): 485–489. doi: 10.1111/j.2044-835X.1994.tb00649.x
27. Roach M, Barratt M, Miller J, Leavitt L, 1998. The structure of mother-child play: young children with Down syndrome and typically developing children. *Developmental Psychology* 34(1): 77–87. doi: 10.1037/0012-1649.34.1.77
28. Roggman L, Cook G, Innocenti M, Norman V, Anderson S, Christiansen K, 2013. *Parenting Interactions With Children Piccolo Provider Starter Kit*, Baltimore
29. Sameroff A, Chandler M, 1975 *Review of child development research*, New York, pp 187-244
30. Sarimski K, 2010. Familienbezogene Lebensqualität bei Müttern von Jungen mit Fragilem-X-Syndrom im frühen Kindesalter. *Praxis der Kinderpsychologie und Kinderpsychiatrie* 59(5): 389–403. doi: 10.13109/prkk.2010.59.5.389
31. Sarimski K., 2014. *Entwicklungspsychologie genetischer Syndrome*, 4. überarbeitete und erweiterte Auflage, Göttingen
32. Sterling A, Barnum L, Skinner D, Warren S, Fleming K, 2012. Parenting young children with and without Fragile X syndrome. *American Journal on Intellectual and Developmental Disabilities* 117(3): 194–206. doi: 10.1352/1944-7558-117.3.194
33. Tamis-LeMonda C, Bornstein M, Baumwell L, 2001. Maternal responsiveness and children's achievement of language milestones. *Child Development* 72: 748–767. doi: 10.1111/1467-8624.00313

34. Tannock R, 1988. Mothers' directiveness in their interactions with their children with and without Down syndrome. *American Journal of Mental Retardation: AJMR* 93(2): 154–165
35. Tröster H, 2011. *Eltern-Belastungs-Inventar*, Göttingen
36. Vygotskij L, 2010 *Psychologie des Spiels*, Berlin, pp 441-465
37. Wheeler A, Hatton D, Holloway V, Sideris J, Neebe E, Roberts J, Reznick J, 2010. Maternal responses to child frustration and requests for help in dyads with Fragile X syndrome: Maternal responses to child behaviour. *Journal of Intellectual Disability Research* 54(6): 501–515. doi: 10.1111/j.1365-2788.2010.01269.x
38. Wheeler A, Skinner D, Bailey D, 2008. Perceived quality of life in mothers of children with Fragile X syndrome. *American Journal on Mental Retardation* 113(3): 159–177. doi: 10.1352/0895-8017(2008)113[159:PQOLIM]2.0.CO;2
39. Wheeler A, Hatton D, Reichardt A, Bailey D, 2007. Correlates of maternal behaviours in mothers of children with Fragile X syndrome. *Journal of Intellectual Disability Research: JIDR* 51: 447–462. doi: 10.1111/j.1365-2788.2006.00896.x

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