



## Temperament factor structure in fragile X syndrome: The Children's Behavior Questionnaire



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### ABSTRACT

Early patterns of temperament lay the foundation for a variety of developmental constructs such as self-regulation, psychopathology, and resilience. Children with fragile X syndrome (FXS) display unique patterns of temperament compared to age-matched clinical and non-clinical samples, and early patterns of temperament have been associated with later anxiety in this population. Despite these unique patterns in FXS and recent reports of atypical factor structure of temperament questionnaires in Williams Syndrome (Leyfer, John, Woodruff-Borden, & Mervis, 2012), no studies have examined the latent factor structure of temperament scales in FXS to ensure measurement validity in this sample. The present study used confirmatory factor analysis to examine the factor structure of a well-validated parent-reported temperament questionnaire, the *Children's Behavior Questionnaire* (Rothbart, Ahadi, Hershey, & Fisher, 2001), in a sample of 90 males with FXS ages 3–9 years. Our data produced a similar, but not identical, three-factor model that retained the original CBQ factors of negative affectivity, effortful control, and extraversion/surgency. In particular, our FXS sample demonstrated stronger factor loadings for fear and shyness than previously reported loadings in non-clinical samples, consistent with reports of poor social approach and elevated anxiety in this population. Although the original factor structure of the *Children's Behavior Questionnaire* is largely retained in children with FXS, differences in factor loading magnitudes may reflect phenotypic characteristics of the syndrome. These findings may inform future developmental and translational research efforts.

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## 1. Introduction

### 1.1. Statement of Problem

For decades, researchers have studied temperament as a means to investigate the biobehavioral processes underlying individual differences. Rothbart and Derryberry (1981) describe temperament as relatively stable individual differences in

Abbreviations: CBQ, Children's Behavior Questionnaire; FXS, Fragile X syndrome; CFA, Confirmatory Factor Analysis.

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reactivity and self-regulation; processes influenced by genetics, experience and development. The psychosocial relationships of temperament to cognitive and behavioral outcomes are believed to be rooted in neurobiological development reflecting gene–brain–behavior relationships, particularly in prefrontal and limbic regions of the brain (see Whittle, Allen, Lubman & Yücel, 2006, for review). From this psychobiological perspective, the study of temperament involves examining and appreciating strengths and limitations of an individual's response to the environment. Moderate variability in temperament patterns is expected across individuals and reflects complex interactions of biological, environmental, and experiential factors that result in a good or poor “fit” in terms of the match between temperament traits and environmental demands. Extreme patterns of temperament often reflect maladaptive reactivity or regulatory abilities and have been linked to a host of pathological outcomes including conduct problems, hyperactivity, anxiety disorders and withdrawal (Rothbart & Bates, 2006; Rothbart, Posner, & Hershey, 1995). Thus, the study of temperament “allows for a view of the tremendous diversity and individual integrity of functioning existing even from the earliest months of life” (Rothbart & Derryberry, 1981, p. 39).

Rothbart et al. (2001) describe fifteen clusters of temperament characteristics (Table 1) that may be measured over time using developmentally specific versions of parent-rated temperament measures. These measures differentiate infant, toddler and early childhood as distinct developmental stages in which temperament characteristics vary in terms of their emergence and the quality of behaviors that vary over time. For example, negative affect and surgency are present in early infancy; however, infants present reactive affiliation and orienting characteristics that mature into self-regulatory effortful control during toddlerhood (Putnam, Rothbart, & Gartstein, 2008).

In contrast to earlier work that focused on the individual temperament subscales, recent research has examined the latent structure and interrelationship among these temperament subscales in greater detail. Several factor analytic studies of typically developing children using temperament measures have consistently yielded a three-factor structure consisting of negative affectivity, surgency (also termed “extraversion”), and effortful control (Rothbart, Ahadi, & Evans, 2000). These factors are generally stable over time. Negative affect reflects negative emotional reactivity and includes constructs such as fear, approach, soothability, sadness, anger, discomfort, and motor activity (Putnam et al., 2008). Surgency characterizes a child's desire for warmth and closeness with others and is reflected in both positive affective responsiveness to stimulation and novelty and speed of response and level of gross motor activity (Rothbart, 2007). Effortful control includes the capacity to focus and maintain short attention, control or inhibit impulses, and to derive pleasure from low-intensity stimuli (Kochanska, Murray, & Harlan, 2000). These factors have been increasingly conceptualized as interrelating risk and protective factors for psychosocial development (e.g. Fox, Russo, Bowles, & Dutton, 2001; Putnam et al., 2008; Rothbart, 2007).

### 1.2. Temperament in clinical populations

In light of the well-established relationship between temperament and psychosocial outcomes in typically developing children, a growing body of work has begun examining the relationship between temperament and atypical development in clinical pediatric samples. Children with Williams syndrome, a neurodevelopmental disorder caused by the deletion of ~25 genes on chromosome 7q11.23 (Klein-Tasman & Mervis, 2003; Tomc, Williamson, & Pauli, 1990). In the only published study to investigate the factor structure of temperament in neurodevelopmental disorders that we are aware of, Leyfer et al. (2012) reported a four-factor model of temperament that differentiated children with Williams syndrome from typically developing norms. The four-factor solution for Williams syndrome included constructs from both infant and early childhood stages, potentially reflecting the delayed developmental changes observed in Williams (Leyfer et al., 2012). Interestingly, the authors also identified differential associations between specific temperament factors and diagnoses of anxiety and attention deficit hyperactivity disorder in this sample.

### 1.3. Fragile X syndrome

In addition to this work in Williams syndrome, several studies have examined atypical patterns of temperament in young children with fragile X syndrome. Fragile X syndrome (FXS) is the most common inherited form of intellectual disability and

**Table 1**  
Participant characteristics.

Variable	N	M	SD	Min	Max
Age (months)	90	70.2	17.63	36.44	118.08
VABS <sup>a</sup>	90	53.34	11.23	30	94
Race					
White	76 (84.44%)				
African American	8 (8.89%)				
Hispanic	1 (1.11%)				
Other	5 (5.56%)				

<sup>a</sup> Vineland Adaptive Behaviors Composite Scaled Score.

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