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**Behavioral Phenotypes as Contextual Factors for Problem Behavior in Individuals**

**with Developmental Disabilities**

A Dissertation Presented

by

**Megan Robinson Joy**

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**Megan Robinson Joy**

We, the dissertation committee for the above candidate for the Doctor of Philosophy degree, hereby recommend acceptance of this dissertation.

**Susan O’Leary, Ph.D. – Dissertation Advisor**  
**Professor**  
**Department of Psychology**

**E. David Klonsky, Ph.D. – Chairperson of Defense**  
**Assistant Professor**  
**Department of Psychology**

**Hoi-Chung Leung, Ph.D.**  
**Assistant Professor**  
**Department of Psychology**

**Joel D. Bregman, M.D.**  
**Assistant Clinical Professor of Child Psychiatry**  
**Yale University**

This dissertation is accepted by the Graduate School.

Lawrence Martin  
Dean of the Graduate School

Abstract of the Dissertation

**Behavioral Phenotypes as Contextual Factors for Problem Behavior in Individuals  
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Individuals with developmental disabilities frequently engage in problem behavior such as aggression, tantrums, self-injury, and disruptive behavior. While research has indicated that problem behavior is frequently associated with aversive contexts, it is often difficult and time consuming to determine which contexts may be problematic for a given individual. The literature on behavioral phenotypes is a potentially rich source of information that may be used to identify contexts that are aversive for individuals with specific developmental disability syndromes. The goal of this study was to demonstrate how information derived from behavioral phenotypes can be incorporated into functional assessment and intervention for problem behavior for individuals with two specific developmental disability syndromes: autism spectrum disorder and Fragile X syndrome. Problematic contexts for each syndrome were identified from the literature on the behavioral phenotype for that syndrome. Direct observation of these contexts in natural settings confirmed that the phenotype-relevant contexts were associated with problem behavior. Using a multiple baseline design, evidence-based interventions were implemented to effectively reduce problem behavior in the phenotype-relevant contexts. The results indicate that information regarding behavioral phenotypes can be systematically integrated into evidence-based functional assessment and intervention for problem behavior displayed by individuals with developmental disabilities.

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## Introduction

Problem behavior, such as aggression, tantrums, self-injury, and property destruction are commonly displayed by people with developmental disabilities (Emerson et al., 2001). Problem behavior compromises the physical safety of the person and those around him or her, and presents a major barrier to successful education, socialization, community inclusion and employment (Bruininks, Hill, & Morreau, 1988; Koegel, Koegel, & Dunlap, 1996). Given the significant impact on quality of life, problem behavior has been a major focus of research and intervention. The reduction of problem behavior is essential for individuals with developmental disabilities to achieve valued outcomes and an optimal quality of life.

### *Functional Assessment*

In order to develop interventions that reduce problem behavior, it is useful to have information regarding the variables that evoke and maintain such behavior. The process of functional assessment identifies the antecedents and consequences that reliably trigger and maintain problem behavior (O'Neill et al., 1997). Meta-analyses of the effectiveness of interventions based on functional assessments have documented substantial reductions or elimination of problem behavior in 50% or more of the cases examined (Carr et al., 1999; Didden, Duker, & Korzilius, 1997; Scotti, Evans, Meyer, & Walker, 1991). Significantly, these meta-analyses have also noted that interventions based on a functional assessment are twice as likely to succeed as interventions not based on a functional assessment.



### *Contextual Factors and Problem Behavior*

A growing literature emphasizes the concept that the function of problem behavior varies according to the context in which it occurs (Luiselli & Cameron, 1998). Contextual factors include both discriminative stimuli and setting events. A discriminative stimulus is an event that predicts that reinforcement is likely to occur when behaviors from a given response class are emitted in the presence of the stimulus. This relationship is due to a history in which behaviors from the response class have been systematically and reliably followed by reinforcers (Skinner, 1938). A setting event is a contextual variable that can influence the probability of specific ongoing stimulus-response relationships (Bijou & Baer, 1978). For example, anxiety may function as a setting event for an individual. If a task demand (discriminative stimulus) is presented when the individual is anxious (setting event), the individual becomes aggressive. In contrast, if the same task demand (discriminative stimulus) is presented when the individual is not anxious (absence of the setting event), the individual responds with cooperation and no aggression. Clearly, all contextual factors, both discriminative stimuli and setting events, must be examined to obtain a complete understanding of the function of problem behavior.

Research has shown that problem behavior is often associated with aversive contextual factors. In a meta-analysis of 536 functional analyses, Hanley, Iwata, and McCord (2003) found that 34.2% of functional analyses demonstrated that the function of problem behavior was to escape from or avoid aversive situations. While research has focused on identifying the *types of* contextual factors that trigger behavior, few researchers have attempted to determine *why* certain contextual factors or environments

are aversive. The answer to this question is likely to depend on a close examination of the interaction between characteristics of the individual and characteristics of the context (environment).

One area of the research literature that has focused on the interaction between the individual and the environment relates to the concept of contextual fit. Contextual fit refers to the degree of match between an individual's competencies and the performance requirements of the environment (Carr, Carlson, Langdon, Magito McLaughlin, & Yarbrough, 1998). Carr and colleagues hypothesized that problem behavior may result when individuals are placed in contexts that require them to use skills in areas of weakness. The model of contextual fit is useful because it suggests that one can intervene with the individual (skill building) or with respect to the environment (context modification).

The concept of contextual fit provides insight into the relationship between aversive contexts and problem behavior for individuals with developmental disabilities. However, there exists a very large set of contexts that are potentially problematic for any given individual. Therefore, from an assessment perspective, it is difficult to know where to begin when determining which contexts may be difficult for a particular individual with a developmental disability. There is a clear need to develop efficient strategies for identifying which contexts might be problematic for a given individual based on his/her weaknesses. The literature on behavioral phenotypes is a potentially rich source of information that could be used to identify the areas in which an individual may lack competency and, therefore, what contexts may constitute a poor fit for that individual.

### *Behavioral Phenotypes*

A behavioral phenotype is defined by the characteristic behavioral repertoire exhibited by individuals with a specific genetic or chromosomal disorder (Flint & Yule, 1994). It includes a wide range of developmental and behavioral features related to cognitive, language, and social profiles (Finegan, 1998). The information regarding behavioral phenotypes for individuals with developmental disabilities is generally found in the biomedical/genetic research literature. Traditionally, behavioral psychologists have not focused on this literature but have tended to focus, instead, on specific instances of problem behavior irrespective of phenotype. Such methodology misses the potentially valuable information that the literature on behavioral phenotypes could provide. The body of research on behavioral phenotypes could be used to identify specific areas of skill weakness for individuals with a given syndrome. This information, in turn, could be used to identify particular contexts that may be especially problematic for the individual. With this knowledge, a clinician could narrow down the number of possible aversive contexts that merit further evaluation during functional assessment.

The present study sought to establish the relationship between behavioral phenotypes, contextual fit, and problem behavior. Two developmental disability syndromes, autism spectrum disorder (ASD) and Fragile X syndrome (FXS), were selected for investigation. Given the scope and prevalence of developmental disabilities, several inclusion criteria were used to facilitate the selection of relevant diagnostic categories. First, the specific syndrome had to be relatively common among the array of developmental disabilities. Second, the research pertaining to the specific syndrome had to be robust with respect to identifying consistent behavioral profiles (behavioral

phenotypes). Autism spectrum disorder and Fragile X syndrome fit these two inclusion criteria. Both syndromes are relatively common with respect to prevalence. Autism spectrum disorder has a prevalence rate of 1 in 150 individuals (Centers for Disease Control and Prevention, 2009). Fragile X syndrome has a prevalence rate of 1 in 4,000 males and at least half as many females (Turner, Webb, Wake, & Robinson, 1996; Sherman, 2002). Fragile X syndrome is the most common inherited form of mental retardation. Each syndrome has a research base that documents a consistent behavioral phenotype.

For each syndrome, one behavioral phenotype characteristic was chosen for investigation. The characteristic chosen for each group is described below.

*Autism spectrum disorder (ASD)*. The phenotype characteristic chosen for individuals with ASD was *impairment in social interaction*. Indeed, impairment in social interaction is the cardinal feature of autism (American Psychiatric Association, 2000; Kanner, 1943; Volkmar, Carter, Grossman, & Klein, 1996). Abnormalities in social interactions are present in individuals with autism across developmental stages and regardless of cognitive ability (Bauminger, 2002). Research has demonstrated that individuals with ASD have weaknesses in many of the key skill areas that comprise social interaction, including: social recognition (Wing, 1998); perspective taking and theory of mind (perception of the needs, interests, and feelings of others) (Baron-Cohen, 1998); emotion recognition (Celani, Battachi, & Arcidiacono, 1999; Loveland et al., 1997); and understanding and use of interpersonal gestures (Attwood, Firth, & Hermelin, 1988). Deficits in all of these areas significantly impact the ability of individuals with ASD to interact with others.

*Fragile X syndrome (FXS)*. The phenotype characteristic chosen for individuals with Fragile X syndrome was *social anxiety or hyperarousal in social situations*. Most males with FXS display a range of difficulties related to social behavior, including anxiety, gaze avoidance, hyperarousal, and problems relating to others (Hagerman & Hagerman, 2002; Hessel, Glaser, Dyer-Friedman, & Reiss, 2006; Roberts et al., 2006; Spencer, Alekseyenko, Serysheva, Yuva-Paylor, & Paylor, 2005). Parents of children and adolescents with FXS rate their children as more anxious and withdrawn, and showing more avoidant behaviors than typical controls (Lesniak-Karpiak, Mazzocco, & Ross, 2003). Many researchers have focused on the arousal levels of people with FXS in social situations, as the cognitive component of anxiety may be difficult to assess in individuals with communication difficulties. Current research has highlighted the distinction between these avoidant behaviors and the social motivation of people with FXS. Individuals with FXS often display a behavior pattern of approach/withdrawal in social situations (Belser & Sudhalter, 2001). These individuals will often indicate that they are interested in interacting with others, but will also demonstrate avoidant behaviors such as gaze aversion and withdrawal. Despite the display of avoidant behaviors, individuals with FXS appear interested in others and are able to interact with others in safe and familiar settings, a behavior profile consistent with social anxiety. Rather than lack of motivation, it is anxiety and hyperarousal in social situations that produces avoidant behaviors (Bailey et al., 1998; Bregman, Leckman, & Ort, 1988; Cohen, Vietze, Sudhalter, & Jenkins, 1989; Sullivan, Hooper, & Hatton, 2007). This behavior profile distinguishes individuals with FXS from individuals with ASD. Individuals with ASD are typically

socially disinterested, whereas individuals with FXS are typically socially interested, but their anxiety significantly interferes with their ability to interact with others.

### *The Present Study*

While some researchers have highlighted the importance of considering behavioral phenotypes when designing interventions (e.g., Dykens, Hodapp & Finucane, 2000; Hagerman, 2009), no one has articulated a model for how to systematically incorporate knowledge of behavioral phenotypes into assessment and intervention. The present study examined a heuristic model for how research on behavioral phenotypes could be used to inform a functional assessment of problem behavior. In this model, potential aversive situations for an individual could be identified by reviewing the behavioral phenotype for the individual's developmental disability syndrome. The behavioral phenotype would predict skill areas that are likely to be weak for the individual. Based on the contextual fit theory, contexts that require the individual to use skills in areas of weakness would be expected to evoke problem behavior. Once the association between these aversive contexts and problem behavior is established, empirically supported interventions could be implemented to reduce problem behavior.

This heuristic model was evaluated with a small group of individuals with autism spectrum disorder and Fragile X syndrome. The study consisted of two phases. In Phase 1, *contextual assessment*, information about competency weaknesses associated with ASD and FXS was used to predict contexts that would constitute a poor fit for individuals with each given syndrome. For participants with ASD, the phenotype-relevant context selected required the use of social interaction skills; for participants with FXS, the phenotype-relevant context selected involved situations that provoked social anxiety or

hyperarousal. These contexts, selected because they were anticipated to be a poor fit for the participants, were expected to be associated with problem behavior. The association with problem behavior was evaluated using an ABAB design with naturally occurring control contexts.

Phase 2, *intervention for problematic contexts*, was initiated after the association between phenotype-relevant contexts and problem behavior was validated in Phase 1. In Phase 2, specific situations, derived from the phenotype-relevant contexts, were selected for intervention. A brief functional assessment of problem behavior within the selected context was conducted during baseline observations. Based on this assessment, a multicomponent intervention package comprised of empirically supported interventions was developed for each participant. The effectiveness of these interventions was evaluated through a multiple baseline design. The goal of this phase was to demonstrate how empirically supported interventions could be used to reduce problem behavior in contexts that were aversive for the individuals due to deficits reflective of their syndrome's behavioral phenotype.

STUDY 1: SOCIAL SKILLS: AUTISM BEHAVIORAL PHENOTYPE AS A  
CONTEXTUAL FACTOR FOR PROBLEM BEHAVIOR

Method

*Procedure Overview*

Participants were recruited from local agencies in New York and Massachusetts that served individuals with developmental disabilities. To be enrolled in the study, participants were required to meet the following inclusion criteria: (1) diagnosis of an autism spectrum disorder (ASD), such as Autistic Disorder or Pervasive Developmental Disorder-Not Otherwise Specified (PDD-NOS), (2) reside at home with family of origin, (3) history of problem behavior (e.g., aggression, self-injury, property destruction, noncompliance, tantrums) in the home and/or community, and (4) family reports that problem behavior restricts family lifestyle (e.g., family is not able to engage in typical home and community activities) or the child's ability to succeed in school. The decision tree for determining whether or not the potential participant met the inclusion criteria is presented in Appendix A.

To confirm each child's diagnosis, the parents of the child provided documentation of their child's diagnostic evaluation. Parents needed to provide documentation that their child received a diagnosis of an ASD from a behavior evaluation conducted by a psychiatrist, psychologist or neurologist. The behavioral evaluation needed to include satisfaction of the DSM-IV-TR criteria as well as use of the Autism Diagnostic Observation Schedule-Generic (ADOS-G; Lord et al., 2000). Participants needed to receive a classification of *autism spectrum* on the ADOS-G (cut-off score of at least 7) to satisfy the inclusion criteria for the study. To confirm the diagnosis of an ASD,



the principal investigator completed the Childhood Autism Rating Scale (CARS; Schopler, Reichler, & Renner, 1980) after the first observation of the child. Since clinicians have speculated that the CARS may over-identify children with autism (Lord et al., 1997), a more conservative cut-off score of 32 was used for the current study. If the child did not receive a score of at least 32 on the CARS, s/he was excluded from the study.

To verify the presence of problem behavior, parents completed the Aberrant Behavior Checklist-Community (ABC-Community; Aman & Singh, 1994). The ABC-Community provides a rating of the severity of the participant's problem behavior across several different topographies of behavior. Once it was determined that the child met the four inclusion criteria, informed consent was obtained from the parents and the child was enrolled in the study.

In the first stage of the study, the participant's parent(s) were interviewed by the researcher. The goal of the interview was to identify the location for the study (home, school, community) and to verify that the child had weaknesses in the behavioral phenotype area targeted for investigation. The parents were asked to describe their child's problem behavior and their primary areas of concern. Through this discussion, the principal investigator and the participant's parent(s) determined whether the study should be conducted in the home, in community settings, or at the participant's school. If the parent and the principal investigator felt that the school would be an appropriate venue for the study, a plan was made to contact the child's school to obtain informed consent from the child's teacher.

Through the initial interview, the principal investigator also verified that the participant had skill weaknesses in the targeted area of the behavioral phenotype for the child's diagnosis. To verify deficits in social interaction skills, parents completed the Socialization Domain of the Vineland Adaptive Behavior Scales, Second Edition, (Vineland-II) Parent Rating Scale (Sparrow, Balla, & Cicchetti, 2005). If the study was to take place solely at the child's school, the participant's teacher completed the Socialization Domain of the Teacher Rating form of the Vineland Adaptive Behavior Scales, Second Edition (Vineland-II). This measure provides an assessment of the participant's social behavior skills compared to a national sample of peers the same age. To confirm weaknesses in social skills, participants needed to score at least one standard deviation below the national average for children the same age (e.g., standard score < 85). Once weaknesses in the target phenotype area were verified, the principal investigator initiated Phase 1 of the study.

The goal of Phase 1 was to determine if phenotype-relevant contexts, anticipated to be a poor contextual fit with the participant's skills, were associated with problem behavior. Participants were observed across a target context and a naturally occurring control context. The purpose of these observations was to see if there was a unique relationship between the target context identified through the analysis of the behavioral phenotype and problem behavior. The association with problem behavior was evaluated using an ABAB design. If the participant did not show problem behavior in the target context, the child's participation in the study ended. If problem behavior was observed in the target context, the participant moved on to Phase 2 of the study.

In Phase 2, a specific problematic situation, based on the phenotype-relevant context from Phase 1, was identified for intervention. For example, if, in Phase 1, it was determined that social interaction contexts were associated with problem behavior, then, in Phase 2, the principal investigator worked with the child's family to identify a specific social interaction context for intervention. The process of identifying the specific context for intervention involved collaboration between the principal investigator and the participant's parent(s). The goal of this collaboration was to identify a high priority context, specific to the target phenotype-relevant context, that was significantly impacting the family's quality of life. Once the specific situation was identified, researchers conducted baseline observations of the identified context for intervention. During baseline observations, the researchers recorded the latency to problem behavior or the duration of the successfully completed routine. The researchers also recorded antecedent-behavior-consequence (ABC) data as part of a brief functional assessment within the problematic context.

At the conclusion of baseline data collection, the principal investigator reviewed the data with the family. Following guidelines for best practices when working with families, parents were included in the process of creating interventions for their child (e.g., Lucyshyn, Albin, & Nixon, 1997). The principal investigator presented intervention suggestions to the family based on the literature of empirically supported interventions. The principal investigator described the interventions that had empirical support and then discussed with the parent(s) how these strategies could be incorporated into the family's routine. Parents were included in this process since they were the most knowledgeable about what would be feasible within their family situation. Through this discussion, the

principal investigator and the family developed a multicomponent intervention package that would be used during intervention in Phase 2. If the intervention was to take place at school, the principal investigator met with the participant's teacher and engaged in a similar process to develop an intervention package that would be feasible to deliver in the child's classroom. Following the development of the intervention package, the parent(s) or teacher were trained to implement each intervention component.

A nonconcurrent multiple baseline across three participants (Hersen & Barlow, 1976; Watson & Workman, 1981) was used to evaluate the effectiveness of the multicomponent intervention on reducing problem behavior. Recall that, while the specific situation for intervention was individualized for each participant, the target context was the same for all three participants with ASD: situations that required social interaction skills.

### *Participants*

#### *Participant 1: Grace*

Grace was a five-year-old girl diagnosed with Pervasive Developmental Disorder-Not Otherwise Specified (PDD-NOS) with an IQ of 100 (Wechsler Intelligence Scale for Children-Fourth Edition). She attended a half-day integrated preschool classroom at her local elementary school. Grace lived at home with her mother, father, and younger brother, who was also diagnosed with an autism spectrum disorder. Grace was verbal and communicated in sentences and phrases. While her vocabulary was advanced for her age, Grace's parents reported that she was frequently unable to communicate her needs. Her language was odd and idiosyncratic, often leaving her communication partners confused and unable to interact with her. Grace's standard score of 80 on the Socialization Domain

of the Vineland Adaptive Behavior Scales (Vineland-II) indicated that her social skills were below those of her peers (one standard deviation below what is typical for children her age).

Grace's parents reported that she engaged in problem behavior (e.g., tantrums, aggression, bolting) at home, at school and in community settings. The family and the principal investigator decided that Grace's participation in the present study would take place in all three settings.

*Participant 2: Edward*

Edward was a 2.5 year-old boy diagnosed with Pervasive Developmental Disorder-Not Otherwise Specified (PDD-NOS) who lived at home with his mother and father. While Edward had not received formal intelligence testing, his IQ was estimated to be in the average range. He received applied behavior analysis (ABA) therapy as a part of early intervention services. Edward received 18 hours a week of ABA and 2 hours a week of speech therapy. He communicated with simple phrases and gestures. Edward received a standard score of 75 on the Socialization Domain of the Vineland Adaptive Behavior Scales (Vineland-II). This score indicates that his social skills were less developed than others of the same age. Edward's parents reported that he frequently displayed problem behavior at home that included tantrums, aggression and throwing objects. Throwing objects was of particular concern to his parents since this behavior had resulted in injury to his family members (e.g., bruising, chipped tooth). Edward's participation in the study occurred with his family in home and community settings.

*Participant 3: Matthew*

Matthew was a 4-year-old boy diagnosed with Pervasive Developmental Disorder-Not Otherwise Specified (PDD-NOS). Matthew's IQ could not be assessed due to his problem behavior, but it was estimated that he met the criteria for Mild Mental Retardation using the DSM-IV-TR narrative criteria. Matthew attended a specialized school for children with developmental disabilities. In his classroom, he participated in discrete trial training with a one-to-one instructor and group activities with other students.

Matthew lived at home with his mother, father and infant sister. He communicated through the use of single words, simple phrases and gestures. Matthew received a score of 65 on the Socialization Domain of the Vineland Adaptive Behavior Scales (Vineland-II). This score indicates that Matthew's social skills were below average when compared to a national sample of typical peers. Matthew's parents reported that he frequently engaged in tantrums and aggression (e.g., hitting, kicking, hair pulling, biting) at home and at school. After meeting and reviewing the study protocol, the principal investigator and the parents determined that Matthew's participation in the study to occur at school. Matthew's school administrators and teachers agreed to implement the study protocol at school. Matthew's participation in the study occurred at school in his classroom.

*Phase 1: Assessment of the Target Context Associated with the Behavioral Phenotype*

*Target Context*

The phenotype-relevant target context for participants with ASD was tasks that required social interaction skills. Social interaction contexts were selected for observation through a discussion with the participant's parent(s) or teacher. Parents and teachers were

asked to identify a social interaction context that occurred with regularity and was relatively consistent from day to day. The goal of this discussion was to identify an ongoing social interaction context that would be suitable for observation across several occurrences. As previously stated, the researcher did not “set up” or contrive situations for observation. However, the researcher attempted to identify contexts that naturally had consistency in order to minimize confounding variables during the observations. The target context was developmentally appropriate (based on the developmental age of the participant) and occurred in natural settings (home and school). For all three participants, the target context involved unstructured free play with at least one additional peer. For Grace and Matthew, free play contexts occurred at school. For Edward, the free play context occurred at a community early intervention group. All three ASD participants were observed in the target context on six occasions. The specific target contexts observed for each participant are listed in Appendix B.

#### *Control Context*

In addition to the target context, all participants were observed in a “control context.” The control context was identified as a task similar in nature to the target context, but one that did not require use of skills in areas of identified weakness (based on the behavioral phenotype). Since the target context for participants with ASD was contexts that required the use of social interaction skills, the control context was solitary in nature (thus not requiring social interaction skills). In the control context, the participant was engaged in the same setting with the same materials as the target context. In contrast to the target context, the participant was alone rather than engaged in the materials with peers. The control context, like the target context, was individualized for

each participant. This individualization allowed the principal investigator to select a control context with maximum similarity to the target context while eliminating the demands of the targeted skill area. Individualization also allowed the principal investigator to ensure that the target context and control context were developmental equivalent and appropriate for the given participant. The control context was observed for each participant on four occasions. The specific control contexts observed for each participant are listed in Appendix B.

### *Observation Procedure*

To ensure the safety of the child, specific guidelines were developed to address the occurrence of problem behavior. Sessions were terminated contingent on the display of problem behavior defined as either: (1) the occurrence of a single instance of “untolerated” problem behavior, specifically aggression (i.e., hitting, kicking, throwing objects toward a person), bolting (i.e., leaving a designated room or area without adult permission), or more than five seconds of screaming (Carr & Carlson, 1993) or (2) three instances of “tolerated” problem behavior, defined as a short episode of screaming (less than five seconds), verbal protests, and/or stomping feet on floor (Carr & Carlson, 1993). Tolerated problem behavior was seen as less serious by the participants’ parents and, therefore, up to three instances were permitted before the session was terminated.

During observations, the researcher, and, when possible, a second observer, recorded the latency to session termination due to problem behavior or successful completion of the context. Latency to session termination was defined as the amount of time that elapsed between the discriminative stimulus that signaled the onset of the



context and the termination of the session, either due to the occurrence of problem behavior or the successful completion of the activity.

### *Interrater Reliability*

A second observer was present and made recordings during 20% of the sessions for Grace, 30% of the sessions for Edward, and 30% of the sessions for Matthew. For all three participants, the second observer was a research assistant with previous education in the area of psychology. Research assistants had received, or were studying to receive, a bachelor's degree in a psychology related field. The research assistant received training on data collection from the principal investigator. Prior to data collection in the natural environment, the research assistant demonstrated that s/he could obtain acceptable interrater reliability with the observations of the principal investigator.

During observations, the second observer was informed as to the nature of the discriminative stimulus that signaled the onset of the context. The second observer was not told whether or not problem behavior was expected in that context. That is, the second observer was "blind" as to whether the context being observed was the target context expected to be associated with problem behavior for that participant or the control context.

A binary reliability index was used to evaluate agreement on latency to session termination and reason for session termination. For each session, reliability was scored as either agreement or no agreement. For latency to session termination, agreement was defined as both observers recording latency within five seconds of one another. As to the reason for session termination, agreement was defined as using the same descriptor for

session termination (i.e., occurrence of tolerated problem behavior, occurrence of intolerated problem behavior, or successful completion of routine).

For Grace, agreement on latency to session termination and reason for session termination was 100%. For Edward, agreement on latency to session termination and reason for session termination was 100%. For Matthew, agreement on latency to session termination was 86%. Agreement on reason for session termination was 100%.

*Phase 2: Intervention for the Problematic Context Associated with the Behavioral Phenotype*

Once the occurrence of problem behavior during the target context was verified in Phase 1, the principal investigator met with the family to discuss identifying a specific context for intervention in Phase 2. During this process, the principal investigator and the family reviewed the participant's data from Phase 1 and discussed how a specific phenotype-relevant context associated with problem behavior was impacting the family's quality of life. From this clinical interview, the principal investigator was able to identify a specific high priority situation, within the broader problematic context, that would be targeted for intervention.

*Context Selection*

*Participant 1: Grace.* Grace's family reported that she engaged in problem behavior in a variety of settings that involved social interactions with her peers. Of particular concern to Grace's mother was her problem behavior at a community ballet class. The ballet class was a high priority for Grace's family as it provided her with an opportunity to engage in an activity that she enjoyed with typical peers. Prior to this study, Grace's behavior had become so difficult in this setting that her mother reported

that if her behavior did not improve, she would need to remove Grace from the class. Therefore, the community ballet class was chosen by the family as the context for intervention in Phase 2.

*Participant 2: Edward.* Edward's mother reported that mealtime was the top priority social interaction context for her family. She chose this context because (1) mealtime was frequently associated with problem behavior (i.e., Edward throwing his cup, bowl and food) and (2) mealtime was important to their family, as it was often the only time of the day when members of the family could sit down and interact.

*Participant 3: Matthew.* Since Matthew's family had requested that his participation take place at his school, the process of identifying the context for intervention involved the head teacher in his classroom. Matthew's teacher expressed concern about his ability to participate in the "tabletops" activity in their classroom. During "tabletops," all of the children in the classroom sat at a circular table and played with a common toy/activity. Activities included playing with blocks, play-doh, coloring, and arts and crafts. During this time, the teachers in the classroom prompted the students to interact and share toys. The goal of "tabletops" was to begin to build basic collaborative play skills. Matthew's teacher identified this context as a priority for intervention because (1) the intensity of problem behavior that occurred during this activity (e.g., tantrums that involved flopping to the ground, hitting, kicking and biting) disrupted the class and (2) "tabletops" was a common activity in inclusion kindergarten classes and, if Matthew was unable to successfully participate in this activity, it would limit his integration with typical peers.

### *Baseline Observations*

Once the priority context for intervention had been selected, the principal investigator, and when possible, a second observer directly observed the context. Guidelines for session termination were the same as those established for Phase 1 of the investigation. Prior to the observations, the principal investigator and the parent or teacher determined the discriminative stimulus that would signal the beginning of the context. During baseline observations, the observers recorded data on the following dependent variables: (1) latency to session termination (either due to the occurrence of problem behavior or successful completion of the activity), (2) number of sessions terminated due to problem behavior. Observers also made antecedent-behavior-consequence (ABC) recordings for each incident of problem behavior (Carr et al., 1994). Baseline observations were naturalistic and unobtrusive. The context participants (e.g., parent, teacher, other children) were instructed to engage in the activity as they typically would have if the researcher(s) were not present.

In ABC recording, the observer recorded the *antecedent* (the event that happened immediately before the behavior), the *behavior*, and the *consequence* (the event that happened immediately after the behavior). The descriptions recorded included environmental antecedents (e.g., noisy classroom, transition between activities), social antecedents (e.g., teacher gave a direction, peer initiated a social interaction), and biological antecedents (e.g., student complained of illness). Behavior recordings included a description of the topography of the behavior (e.g., hitting head with a closed fist). Consequence descriptions included environmental, social, and biological consequences

for the behavior (e.g., current activity was terminated, student was told to stop doing behavior, student was given medication).

ABC recordings were made in an effort to ascertain the function of the problem behavior within the specific context. The hypothesized function of the behavior was determined by analyzing the pattern of the ABC recordings. For example, consider a student who repeatedly engages in a behavior (hitting head) when his teacher is working with another student (antecedent). When he hits his head, his teacher turns around and tells the student to stop hitting his head (consequence). This pattern of behavior suggests that the student may be engaging in the behavior in order to access attention from his teacher. Consider another student who repeatedly tears up his worksheet (behavior) when he is given math work (antecedent). When he tears up his math sheet, the teacher stops the task sends him to a timeout in the hall (consequence). This student is plausibly engaging in the behavior to escape the math work. Analysis of the pattern of ABC recording allowed the researchers to develop hypotheses regarding the function of the participants' behavior within the given context.

#### *Development of Intervention*

Following baseline observations, the principal investigator met with the family to review the assessment information obtained during the observations and to design an intervention package to reduce the occurrence of problem behavior. The development of the intervention package was a collaborative process between the principal investigator and the family. The principal investigator presented the family/teacher with several empirically supported interventions relevant to the problematic context based on the research literature. With feedback from the family, the principal investigator developed a

multicomponent intervention package designed to address the possible functions of the problem behavior in the target context. Intervention strategies focused on techniques that altered the stimulus properties of the context (context modification) and/or taught skills to the child (skill building) so that s/he could more successfully participate in the context.

*Participant 1: Grace.* Grace participated in an hour long ballet class which involved multiple group instructions through drills, exercises and dance routines. During these activities, the children in the class were expected to follow verbal instructions given to the group and interact quietly in between activities (during “down time”). Grace exhibited problem behavior in ballet class that consisted of yelling, stomping her feet, and tantrums that involved dropping to the floor. Grace’s mother was presented with several intervention possibilities. The principal investigator and her mother discussed context modification strategies, such as training the teacher of the class and implementing visuals in the room. Skill building strategies, such as teaching target social skills required by the class, were also reviewed. Through this discussion, Grace’s mother and the principal investigator chose to implement the skill building strategy of using a social story. This intervention was identified as the best option given the circumstances of the ballet class. The teacher was a college student who taught the class in her spare time. Grace’s mother was reluctant to ask her to participate in time consuming training sessions or alter the physical aspects of the ballet room. Thus, her mother chose a strategy that would build Grace’s skills so that she could better participate in the class as it was already constituted.

Social stories have been demonstrated to reduce problem behavior associated with social routines for individuals with developmental disabilities (Gray & Garand, 1993;

Sansosti, Powell-Smith, & Kincaid, 2004). Clinicians have suggested that social stories are effective because they provide the individual with predictability and concrete expectations for behavior in the particular social situation. Children with ASD have a strong preference for predictability in routines and research has demonstrated that enhancing predictability is associated with lower rates of problem behavior (Dettmer et al., 2000).

Based on observations of the ballet class and discussions with Grace's mother, the principal investigator was able to outline the typical order of activities for the class. The social story described each activity (e.g., drills, stretching, dancing) as well as the behavior Grace needed to exhibit during each activity. Overall, three behavioral expectations were emphasized: (1) quiet voice, (2) look at the teacher, and (3) listen to the teacher. These expectations were repeated throughout the story. In addition, these expectations were depicted with pictures on a separate visual support sheet that was reviewed with Grace before class. The script for the social story is presented in Appendix C. When the social story was presented to Grace, it had illustrations of each activity. For simplicity, the illustrations have been removed from Appendix C.

ABC data collection suggested that the function of Grace's problem behavior in ballet class was to gain *access to attention* and to *escape the activity*. The social story also described functionally equivalent responses so that Grace could meet her needs. Extant research has demonstrated that problem behavior can be reduced by teaching individuals appropriate responses that serve the same function as problem behavior (e.g., Carr & Durand, 1985; Wacker et al., 2005). Once the individual is able to meet her/his needs with the functionally equivalent response, s/he no longer needs to engage in

problem behavior. The response targeted to teach *access attention* was raising her hand. Through the social story, Grace was instructed that if she wanted to get the teacher's attention, she needed to raise her hand and wait to be called on by the teacher. The response targeted to address the *escape* function was asking for help. The researchers hypothesized that Grace was engaging in escape motivated problem behavior because the activity was difficult. Asking for assistance from the instructor would mitigate the difficulty of the task. Importantly, it was thought that Grace would engage in less escape-motivated behavior once she had a better understanding of the class' behavioral expectations through the social story.

The social story was initially introduced to Grace one day prior to her ballet class. When it was first introduced, Grace's mother read the story to her. After the introduction, Grace was permitted to read the story as many times as she requested. Grace's mother brought the social story in the car on the way to ballet class and reviewed it during the car ride before each class. Before entering class, Grace's mother reviewed the three behavioral expectations (e.g., quiet voice, look at teacher, listen to teacher) with the accompanying visual support.

*Participant 2: Edward.* During baseline observations, ABC data collection suggested that Edward's problem behavior served the function of gaining *access to attention* during mealtime. During meals, Edward's parents conversed with each other and frequently moved to and from the table. Edward typically did not receive attention during these conversations until he screamed or threw his cup or bowl. When Edward engaged in problem behavior, his parents would reprimand him. These baseline observations indicated that Eric did not have the social interaction skills to solicit his



parents' attention during this busy family routine. Based on the results of the ABC data collection, the intervention package developed for mealtime had two components: (1) structured social reinforcement (context modification) and (2) teaching Edward to solicit his parents' attention (skill building).

During baseline, the researchers observed that Edward was most likely to receive attention from his parents when he engaged in problem behavior. One of the goals of intervention was to modify the context so that Edward received social attention every 2-3 minutes for engaging in appropriate mealtime behavior. Regular attention undermines the need to engage in problem behavior to receive attention (Hanley, Piazza, & Fisher, 1997). In order to structure social attention, the principal investigator outlined a concrete behavioral expectation for Edward's mealtime behavior: keeping Edward's cup and bowl "in their place." This phrase referred to keeping the bowl and cup on the table when not being used for their intended purpose (e.g., cup could come off the table when Edward was taking a drink). This expectation was chosen as a competing, social appropriate alternative to Edward throwing his bowl and cup (problem behavior most frequently observed during baseline).

To promote Edward's success following this expectation, a visual support in the form of a placemat was used during mealtime. The placemat displayed a small red circle for the cup and a larger red circle for the bowl. Edward was instructed to put his cup and bowl on the red circles. When the cup and bowl were on the circles, they were "in their place". This strategy was implemented because research has demonstrated that individuals with autism are visual learners (Hodgon, 1995) and provision of a visual support during intervention can facilitate skill acquisition (Quill, 1995).

Edward's parents were instructed to give him positive reinforcement every 2-3 minutes for keeping his bowl and cup "in their place." Reinforcement was in the form of positive social attention, such as, "Nice job keeping your cup in its place!" and "I love the way you are following the rules!" These phrases were paired with physical attention in the form of hugs, "high fives" and pats on the head. In addition to the social attention, Edward received a token for each interval he successfully kept his bowl and cup on the table. The token was in the form of a puzzle piece. When Edward earned all six puzzle pieces (the length of time it took to complete the meal), he was allowed access to a reward. Through a preference assessment interview conducted with Edward's mother (Matson et al., 1999), it was determined that Thomas the Tank Engine was Edward's most preferred toy character. Thus, when Edward earned all six token pieces he was allowed access to a special Thomas the Tank Engine toy. Edward was only able to earn all six pieces of the puzzle when he completed the meal, keeping his cup and bowl in their place for the duration of the meal (typically 12-15 minutes). If Edward had any instances of throwing, a puzzle piece was neutrally removed from his token board. While Edward could continue to earn tokens throughout the meal (and receive praise), he did not earn the toy at the end of the meal if he had any instance of problem behavior. This token system was implemented to ensure that Edward's parents delivered regular, concrete, positive attention to Edward for engaging in socially appropriate behavior during meals.

During baseline, Edward did not attempt to solicit his parents' attention through socially appropriate means. Through ongoing ABA instruction, Edward was taught to say, "Mommy" and "Daddy" to solicit attention from his parents. This skill was then

transferred to the mealtime setting. Edward's parents were taught how to prompt him to say "Mommy" or "Daddy" when he looked toward them, indicating that he wanted attention. His parents were also instructed to respond to Edward every time he solicited their attention (schedule of reinforcement: fixed ratio 1). Reinforcement was provided at this high level to increase the likelihood that Edward would demonstrate this behavior.

*Participant 3: Matthew.* Through baseline observations and discussions with Matthew's teacher, the principal investigator hypothesized that the social nature of "tabletops" was the aversive component of the activity. The actual toys/materials did not appear to be aversive as discrete trial data indicated that Matthew was proficient at using the toys. During individual training sessions with these toys, Matthew did not engage in problem behavior. However, when Matthew was prompted to use these toys in collaboration with peers during "tabletops", he was likely to engage in problem behavior.

The intervention developed for Matthew had three main components. First, to decrease the likelihood of problem behavior, positive mood induction strategies were implemented prior to "tabletops" (context modification). Second, to decrease the association between the activity and problem behavior, a highly preferred toy was introduced into the "tabletops" activity (context modification). Third, since ABC recordings indicated that Matthew engaged in problem behavior to *escape the activity*, he was taught a socially appropriate response that could be used to access escape from the activity (skill building).

The first component of the intervention was to incorporate positive mood induction strategies into Matthew's daily routine prior to "tabletops." Carr, Magito McLaughlin, Giacobbe-Grieco, and Smith (2003) demonstrated that there is a

relationship between mood and problem behavior. They showed that mood can be altered through mood induction strategies, thus decreasing the likelihood of problem behavior occurring. To induce a positive mood prior to the “tabletops” activity, Matthew was given access to a preferred snack he brought from home. Matthew was also provided with 2-5 minutes of noncontingent praise and affection from a preferred staff person in the classroom. Praise and affection was in the form of verbal praise, “high fives” and tickles.

The second component of the intervention was to introduce a highly preferred toy to the tabletops activity. While it was not believed that Matthew disliked the toys/activities used during “tabletops,” they were not among his most highly preferred toys. Research has shown that introducing a highly preferred object into a non-preferred activity decreases problem behavior associated with that activity (Cameron, Luiselli, McGrath, & Carlton, 1992). To identify the preferred toy, a forced choice preference assessment was conducted with a variety of toys that were identified by Matthew’s mother and teacher as highly preferred. During the preference assessment, the principal investigator recorded which toy Matthew selected from the forced choice array of two toys. In addition, the principal investigator observed and recorded how long Matthew engaged with the toy, as well as other indicators that Matthew liked the toy (e.g., smiling, laughing). Through this preference assessment, the preferred toy identified was a Sesame Street letter game. This letter game was placed on the table, in Matthew’s view, prior to the instruction for him to sit at the table for “tabletops.”

The third component of the intervention was to teach Matthew an efficient functional communication response to allow him escape from the activity without engaging in problem behavior. Matthew’s teacher had already started a program to teach

him to request escape from activities by touching a “break” card. When Matthew touched the card, he was immediately allowed to leave the activity. The act of touching the “break card” was an efficient response for Matthew. That is, it was relatively easy for him to do. For functionally equivalent responses to effectively replace problem behavior, they must be more efficient than the problem behavior (Carr & Durand, 1985). While Matthew’s team had started to teach him to use this “break” card to request escape from activities, the card was not being used during “tabletops.” Thus, the decision was made to introduce the “break” card into the “tabletop” activity as part of the intervention.

It is important to note that the intervention package for Matthew was slightly different in focus from the other participants in Study 1. For the other participants, the intervention involved strategies to teach the participants social interaction skills so they could more successfully participate in the target social context. For Matthew, the intervention package focused on reducing the overall aversiveness of the target context by introducing positive stimuli (e.g., mood induction, preferred toy) into the context. This strategy was adopted due to Matthew’s lack of participation in “tabletops” during baseline. During the baseline observations, Matthew frequently began to engage in problem behavior as soon as he was cued to sit at the table for “tabletops”. In many instances, he flopped to the floor and began to tantrum before he sat at the table. These baseline data suggested that the overall aversiveness of the activity needed to be reduced in order to get Matthew to approach the table and sit through the activity. Once Matthew was able to sit at the table with his peers, his teachers could begin to prompt social interaction to build his social skills. In this way, the intervention targeted the prerequisite

skills for Matthew learn social skills in the target context, namely, his ability to sit and engage in parallel activities his peers.

### *Intervention Agent Training*

The principal investigator trained the parent or teacher to implement the multicomponent intervention package in the specific phenotype-relevant context. Training consisted of four stages and occurred over the course of 1-2 sessions. First, the principal investigator explained the rationale for each intervention strategy. Second, the principal investigator demonstrated how to implement the intervention strategy through a role play. When possible, a second researcher participated in the role play. Parents and teachers were given opportunities to ask questions during the role play. Third, the parent or teacher was asked to demonstrate the intervention strategy in a role play with the researcher. Fourth, the parent or teacher implemented the intervention strategy during the natural target context with feedback from the researcher. During the fourth stage, the researcher completed the intervention fidelity checklist (described below). Parent training sessions ended when the parent or teacher was able to correctly implement all the components of the intervention package in the natural context. If, at any time during Phase 2, the principal investigator determined that the parent or teacher was not correctly implementing all the components of the intervention package, the four stages of the training sequence were reinitiated.

### *Intervention Fidelity*

An intervention fidelity checklist, based on the intervention components for each participant, was developed to evaluate intervention integrity (see Appendix D, which lists the intervention components for the Phase 2 interventions for Study 1 and Study 2). The

intervention fidelity checklist was completed by the primary observer (e.g., researcher or mother) for 80% - 100% of the intervention sessions. The intervention fidelity checklist was completed by a second observer in 30% of the sessions for Grace, 33% of the sessions for Edward, and 56% of the sessions for Matthew. The checklist was completed by placing a checkmark whenever a specific component of the intervention package was implemented.

### *Interrater Reliability*

Since intervention occurred in home, school, and community settings with natural intervention agents (e.g., mother, teacher), the primary observer for each participant varied depending on the nature of the setting. For Grace and Edward, the intervention package was delivered in the community and home settings, respectively. For these two participants, the primary observer was the parent, as it was difficult to have a researcher present for every community ballet class (Grace) and every family meal (Edward). The intervention package for Matthew occurred in the school setting. In this setting, it was relatively easy to introduce a researcher unobtrusively into the environment. Therefore, for Matthew, the principal investigator was the primary observer.

To assess interrater reliability, a second observer was present during some of the baseline and intervention sessions. For all three participants, the second observer was a researcher. For Grace and Edward, the second observer was the principal investigator. For Matthew, the second observer was an undergraduate research assistant. Prior to data collection in the natural environment, the research assistant underwent research training and demonstrated that he could obtain acceptable interrater reliability with the observations of the principal investigator.

A binary reliability index was used to evaluate agreement on intervention fidelity, latency to session termination, and reason for session termination. For each session, reliability was scored as either agreement or no agreement. For intervention fidelity, agreement was defined as all the steps of the intervention package checked as implemented correctly on the intervention fidelity checklist. For latency to session termination, agreement was defined as both observers recording a time within five seconds of one another. For reason for session termination, agreement was defined as identifying the same description for session termination (i.e., occurrence of tolerated problem behavior, occurrence of intolerated problem behavior, or successful completion of routine).

For Grace, two observers completed reliability checks on 20% of the baseline sessions and 30% of the intervention sessions. Agreement on intervention fidelity, latency to session termination, and reason for session termination was noted in 100% of reliability sessions. For Edward, two observers completed reliability checks in 25% of baseline sessions and 33% of intervention sessions. Agreement on intervention fidelity, latency to session termination, and reason for session termination was noted in 100% of reliability sessions. For Matthew, two observers completed reliability checks on 27% of baseline sessions and 56% of intervention sessions. Agreement on intervention fidelity occurred in 80% of reliability sessions. Agreement on latency to session termination and reason for session termination occurred in 100% of reliability sessions.



## Results

### *Phase 1: Assessment of the Target Context Associated with the Behavioral Phenotype*

For all three participants, six observation sessions were conducted for the target context and four observation sessions were completed for the control context.

#### *Participant 1: Grace*

Figure 1 shows the latency to session termination for the contexts observed for Grace. For the target context, social contexts that required social skills for participation, 0% were completed successfully. That is, 100% were terminated due to tolerated problem behavior. The mean latency to session termination was 16 min, 7 s (range: 9 min, 24 s to 24 min). For the control context, 100% of sessions were completed successfully. The mean duration of the control context was 21 min, 30 s (range: 15 min, 30 s to 30 min).

#### *Participant 2: Edward*

Figure 2 shows the latency to session termination for the contexts observed for Edward. For the target context, 17% were completed successfully. That is, 83% were terminated due to untolerated problem behavior. The mean latency to session termination was 6 min, 16 s (range: 2 min, 24 s to 9 min, 8 s). For the control context, 100% were completed successfully. The mean duration of the control context was 15 min, 15 s (range: 13 min to 17 min).

#### *Participant 3: Matthew*

Figure 3 shows the latency to session termination for the contexts observed for Matthew. For the target context, 0% were completed successfully. In other words, 100% of these tasks were terminated due to untolerated problem behavior. The mean latency to session termination was 1 min, 50 s (range: 3s to 4 min, 15 s). For the control context,

100% of the observed sessions were completed successfully. The mean duration of the control context was 4 min, 45 s (range: 3 min, 30 s to 6 min).

*Phase 2: Intervention for the Problematic Context Associated with the Behavioral Phenotype*

Figure 4 presents data on the amount of time that elapsed (latency) before the session was terminated (due to problem behavior or the successful completion of the context) for the three participants with ASD. Sessions could be terminated due to the presence of intolerated problem behavior (solid black bars), tolerated problem behavior (grey bars), or successful completion of the context (open bars). For Grace, all five baseline sessions were terminated due to tolerated or intolerated problem behavior. The mean latency to session termination during baseline was 10 min, 36 s. During intervention, Grace did not display problem behavior. All intervention sessions were successfully completed. The mean duration of successfully completed sessions was 60 min. For Edward, 7 of the 8 baseline sessions (86%) were terminated due to intolerated problem behavior. The mean latency to session termination during baseline was 5 min, 42 s. During intervention, 11 of the 12 sessions (92%) were completed successfully. The mean duration of successfully completed sessions was 15 min, 49 s. For Matthew, 10 of the 11 baseline sessions (91%) were terminated due to intolerated problem behavior. The mean latency to session terminated during baseline was 1 min, 15 s. During intervention, Matthew did not display problem behavior; all sessions were completed successfully. The mean duration of successfully completed sessions was 9 min, 32 s.

STUDY 2: SOCIAL ANXIETY: FRAGILE X SYNDROME BEHAVIORAL  
PHENOTYPE AS A CONTEXTUAL FACTOR FOR PROBLEM BEHAVIOR

Method

*Procedure*

The general procedure used in Study 2 was the same as the procedure described for Study 1. Study 2 investigated the behavioral phenotype for Fragile X syndrome (FXS) as a contextual factor for problem behavior.

To confirm the diagnosis of FXS, parents of potential participants needed to provide a diagnostic evaluation that indicated a diagnosis of FXS. Since this diagnosis requires medical testing, the evaluation needed to include reporting of relevant genetic testing. If the family of a potential participant with FXS could not supply a report that included genetic testing, the child was excluded from the study. Potential participants were also excluded from the study if their evaluation indicated any comorbid diagnosis. This criterion was especially important since children with FXS may also be diagnosed with an ASD (Bailey, Mesibov, Hatton, Clark, Roberts & Mayhew, 1998; Lewis et al., 2006).

The targeted behavioral phenotype characteristic for FXS was *social anxiety* or *hyperarousal in social situations*. To confirm that participants with FXS did, in fact, exhibit social anxiety and/or hyperarousal, the principal investigator administered the social phobia (social anxiety disorder) subscale of the Anxiety Disorders Interview Schedule for DSM-IV, Parent Interview Schedule (ADIS; Silverman & Albano, 1996). The ADIS is a semi-structured interview for parents that evaluates DSM-IV criteria for a variety of anxiety disorders. For social anxiety disorder, the ADIS highlights three

criteria relevant to making a clinical diagnosis: (1) individual fears that s/he will act in a way that is embarrassing or humiliating in social situations, (2) exposure to social situations causes fear and social situations are either avoided or endured with distress, and (3) this avoidance or distress interferes with the child's ability to carry out normative daily activities. Since participants in the current study had impaired cognitive ability due to their developmental disability, it was often not possible for parents to assess whether their child feared embarrassment. As such, to confirm the presence of social anxiety in the current study, parents did not need to indicate that their child met the first criterion. They did, however, need to report that exposure to at least one situation caused distress (rated 4 or more on a scale of 1-8) and that this distress interfered with the child's daily life (rated 4 or more on a scale of 1-8).

### *Participants*

#### *Participant 1: Jonathan*

Jonathan was a 19-year-old man diagnosed with FXS who lived at home with his parents. Jonathan had an IQ 49 (Wechsler Adult Intelligence Scale). As part of his self-determination plan, Jonathan participated in various community jobs and volunteer experiences, including working at a local convenience store, animal shelter and radio station. Jonathan was verbal and communicated in sentences and phrases. Jonathan's parents identified six items on the ADIS as associated with high levels of anxiety and hyperarousal. Items endorsed as anxiety provoking included working or playing in a group, speaking to new or unfamiliar people, and attending parties.

Jonathan had a history of exhibiting tantrums (e.g., yelling and screaming), property destruction (e.g., throwing objects), and verbal perseveration. His parents

reported that verbal perseveration was the problem behavior of most concern to them. His verbal perseveration took the form of repetitive question asking or stating the same thing over and over again, such as “You hate me.” Verbal perseveration is a behavior frequently exhibited by people with FXS, and has been identified as a defining characteristic of the syndrome (Roberts et al., 2007). Research has suggested that verbal perseveration is a manifestation of the hyperarousal that individuals with FXS experience in social situations (Abbeduto, Brady, & Kover, 2007; Belser & Sudhalter, 2001; Roberts et al., 2007). Since this behavior is verbal, it is not dangerous in the way that aggression, self-injury and property destruction can be dangerous. However, it can be highly disruptive to those who interact with the individual. Jonathan’s parents reported that his verbal perseveration was negatively impacting their family’s quality of life. Jonathan’s participation in the study occurred at home and in community settings.

*Participant 2: Jacob*

Jacob was a six-year-old boy diagnosed with FXS who lived at home with his parents and his nine-year-old sister. Jacob had a Mental Development Index of 57 on the Bayley Scales of Infant Development – II. He was verbal and communicated with simple 2-3 word phrases. Jacob attended school in a substantially separate classroom for children with developmental disabilities. His parents reported that he frequently displayed problem behavior such as tantrums, property destruction, bolting, and aggression toward his parents and sister. Dangerous problem behavior occurred at least once per day. Jacob’s mother identified seven items on the ADIS as associated with high levels of anxiety and hyperarousal. Items endorsed as anxiety provoking included working or playing in a group, speaking to new or unfamiliar people, gym class, inviting a friend for

a get together, and attending parties. Jacob's participation in the study occurred at home and in community settings.

*Phase 1: Assessment of Contexts Associated with Behavioral Phenotypes*

*Target Context*

Recall that, for Phase 1, participants were observed in contexts that were anticipated to be a poor contextual fit for the individual, based on the literature for the behavioral phenotype for the participant's disability. The target context in Phase 1 for FXS was situations that provoked social anxiety/hyperarousal. Since the literature on FXS suggests that situations involving novel people and environments may be anxiety provoking (Belsler & Sudhalter, 1995), the target context for both participants in Study 2 involved novel environments or interactions with novel people.

The principal investigator worked with both Jacob's and Jonathan's parents to identify situations in which the participants naturally encountered novel people or new environments. Social interactions with novel people were matched to the developmental level of the participant. For Jacob, the target context involved meeting a new therapist, going to a new community location, and meeting a friend of his mother's for the first time. For Jonathan, the target context involved meeting new members of his self-determination team, meeting typical peers at a community integration "teen night", and having his home routine taped by a documentary film crew making a movie about self-determination. For both participants, each novel situation was used for 1-2 observations. Specific contexts were not used more than twice because the researchers felt that after two exposures the context would no longer be novel, and, therefore, would no longer fit

the criteria for the target context. The target context was observed on six occasions for each participant. The target contexts observed for Study 2 are listed in Appendix B.

### *Control Context*

As with Study 1, the control context was an activity that was similar in nature to the target context, but did not require use of skills selected from the behavioral phenotype. For participants with FXS, the control contexts involved a similar social activity that was not anxiety provoking, namely, a context that involved routine interactions with familiar people in familiar settings. For Jacob, the control context occurred with a neighborhood ice cream man. The man delivered ice cream to Jacob's house every day and engaged in a routine, predictable conversation with Jacob. For Jonathan, the control context was one of his volunteer jobs. Twice a week Jonathan went to a community animal shelter with his job coach and volunteered to care for the animals. During this time Jonathan interacted with his job coach in a predictable manner. The control context was observed on four occasions for each participant. The control contexts observed for each participant are listed in Appendix B.

### *Observation Procedure*

The guidelines for session termination were the same as Study 1. Sessions were terminated following a single occurrence of "untolerated" problem behavior or three instances of "tolerated" problem behavior. During the observations, the researcher, and, when possible, a second observer, recorded the latency to the first instance of problem behavior or successful completion of the context.

### *Interrater Reliability*

A second observer was present and made recordings during 60% of the sessions for Jonathan and 20% of the sessions for Jacob. The second observer in Study 2 was a clinical psychology doctoral student. Prior to data collection, the second observer demonstrated acceptable reliability with the observations of the principal investigator.

The same binary reliability index used in Study 1 was used in Study 2 to evaluate agreement on latency to session termination and reason for session termination. For both Jonathan and Jacob, agreement on latency to session termination and reason for session termination was 100%.

### *Phase 2: Intervention for the Problematic Context Associated with the Behavioral Phenotype*

After Phase 1 was completed, the principal investigator met with the family to discuss identifying a problematic context for intervention in Phase 2. From this clinical interview, the researcher was able to identify a specific situation, within the phenotype-relevant context, that would be targeted for intervention. A nonconcurrent multiple baseline across two participants (Hersen & Barlow, 1976; Watson & Workman, 1981) was used to evaluate the effectiveness of the multicomponent intervention on reducing problem behavior.

### *Context Selection*

*Participant 1: Jonathan.* The high priority context identified for intervention by Jonathan's family was his morning routine of getting dressed and ready to leave his house. Jonathan's parents reported that he exhibited a high rate of verbal perseveration during this activity. They also stated that his verbal perseveration was typically



associated with times when he was anxious. The researchers identified this specific context as a social anxiety context because they hypothesized that Jonathan was experiencing anticipatory anxiety about his day. This anxiety was social in nature as Jonathan was scheduled for a variety of social activities in the community each day.

*Participant 2: Jacob.* Jacob's mother reported that he exhibited problem behavior such as aggression, property destruction, and bolting in many social situations. She reported that problem behavior most frequently occurred at home when Jacob was asked to interact with someone other than his parents or familiar adults. Jacob did not play or have extended interactions with his sister; these interactions usually resulted in Jacob being aggressive toward his sister. Jacob's mother attributed his difficulty in these situations to his level of hyperarousal, which became elevated in unfamiliar social situations. Although Jacob was familiar with his sister, he rarely interacted with her beyond casual greetings due to parental concerns for his sister's safety. Since this lack of interaction seriously impacted the family's quality of life, playing with his sister was chosen as the target context for Jacob.

#### *Baseline Observations*

Once the specific context for intervention had been identified, the researcher, and when possible, a second observer directly observed the context selected for intervention. Guidelines for session termination were the same as those established for Phase 1 of the investigation. However, in contrast to Study 1, researchers recorded the latency until the *first instance* of problem behavior, irrespective as to whether or not the session needed to be terminated. This change was made after interviews with FXS participants' parents revealed a high level of concern for tolerated problem behavior, such as verbal

perseveration. Given this concern, the decision was made to record the latency until the first instance of any problem behavior.

During baseline observations, the observers recorded data on the following dependent variables: (1) latency to the first instance of problem behavior (either tolerated or intolerated problem behavior) or successful completion of the context, and (2) number of sessions terminated due to problem behavior. Researchers also made ABC recordings for each incident of problem behavior (Carr et al., 1994). Baseline observations were naturalistic and unobtrusive.

#### *Development of Intervention*

Following baseline observations, the principal investigator met with the family to review the assessment information and to design an intervention package to reduce the occurrence of problem behavior in the identified context. As in Study 1, the development of the intervention package was a collaborative process between the principal investigator and the family. Intervention strategies focused on techniques that altered the stimulus properties of the context (context modification) and/or taught skills to the child (skill building) so that s/he could more successfully participate in the context.

*Participant 1: Jonathan.* During baseline observations, Jonathan exhibited verbal perseveration during his morning routine. Verbal perseveration typically occurred when he was cued to get dressed before leaving the house for the morning. ABC data collection and observations suggested that verbal perseveration served multiple possible functions. Two possible functions were to *access to attention* from his mother and to *escape his anxiety*. By asking the same question over and over again, Jonathan was able to garner attention his mother. Importantly, this attention had a predictable nature to it; he asked

the same questions with the same answers. In this way, repetitive question asking appeared to have anxiety-reducing properties similar to ritualistic behavior seen in individuals with obsessive-compulsive disorder.

A third possible function was to gain *access to sensory stimulation*. Baseline data indicated that the behavior could be maintained, in part, by automatic reinforcement. Researchers have proposed that repetitive verbal behavior may be maintained by the auditory stimulation it produces (Lovaas, Newson, Hickman, 1987). Rincover, Newson, and Carr (1979) suggested that ritualistic behavior may require interventions to address sensory reinforcement associated with the behavior. Given the complex nature of Jonathan's verbal perseveration, an intervention package was developed to address the various possible functions of his behavior. The intervention package included two context modification strategies: (1) preview Jonathan's daily routine, (2) schedule a time to allow verbal perseveration.

To address Jonathan's anticipatory anxiety associated with the morning routine, the researchers suggested use of a visual schedule. Research has indicated that visual schedules can provide predictability for individuals with developmental disabilities (Dettmer et al., 2000). While Jonathan's parents insisted that he was familiar with the activities he would be doing each day (each day was different), the researchers believed that providing a visual schedule would support his comprehension of his daily routine.

After much discussion with Jonathan's family, they ultimately decided that they did not want to use a visual schedule. They felt that a visual schedule underestimated their son's abilities. Instead, the family agreed to verbally review Jonathan's daily schedule with him when he woke up in the morning. Through this verbal rehearsal, the

intervention attempted to approximate the predictability that would be obtained with a visual schedule. In addition, the verbal rehearsal allowed Jonathan to access a predictable social interaction. As such, the verbal rehearsal of his day served as a plausible replacement for *accessing attention* and *escaping anxiety*.

The second component of Jonathan's intervention package involved bringing the verbal perseveration under stimulus control. The researchers hypothesized that Jonathan's behavior may have served an additional *sensory* function; that is, the behavior was maintained by automatic reinforcement. Since it was believed that Jonathan was deriving some reinforcement from the behavior itself, the researchers were concerned that the behavior could not simply be replaced. Jonathan needed a time when he could engage in the behavior, without it interfering with his daily routine. Therefore, one aspect of the intervention involved allowing Jonathan time to engage in the behavior during an identified time period. This schedule allowed Jonathan to obtain reinforcement from the behavior without having it interfere with his morning routine.

The time-frame in which to allow verbal preservation was carefully chosen to fit into the family's morning routine. Jonathan was allowed to engage in 30 minutes of verbal preservation *after* he completed his morning routine. If Jonathan attempted to engage in verbal perseveration prior to his morning routine, he was redirected to getting dressed and told that he could engage in the behavior from 8:30-9:00 am. If he was dressed at 8:30 am, his mother went into Jonathan's room and offered to answer any of this questions. If Jonathan was not dressed at 8:30 am, he was given the amount of time available between when he finished getting dressed and 9:00 am to engage in repetitive questions asking with his mother. In addition to allowing Jonathan an appropriate time to

access reinforcement for the behavior, the scheduled verbal perseveration time also added another predictable element to Jonathan's morning routine.

*Participant 2: Jacob.* In baseline observations, Jacob exhibited a number of aggressive behaviors toward his sister during play scenarios, including hairpulling, hitting, kicking and pushing. When Jacob was given the opportunity to initiate play activities (e.g., when his sister asked "What do you want to play?"), he would not identify or initiate an activity to play. When his sister attempted to initiate a play activity (e.g., "Jacob, let's play with the trains"), Jacob typically aggressed toward his sister and ran from the room. At that time, Jacob's mother would intervene and the activity would terminate. Jacob's problem behavior was hypothesized to serve the functions of *escape from the activity* and a means to *access attention* in the form of a reaction from his sister and mother. The goal of the multicomponent intervention package was to reduce the anxiety/hyperarousal associated with playing with his sister and eliminate the factors currently maintaining the problem behavior. The intervention package was comprised of two context modification strategies: (1) training his sister to be an effective play facilitator, and (2) visual supports for play initiation.

Peer interventions have been used successfully to train siblings to effectively structure play sessions to better meet the needs of children with developmental disabilities (Strain & Schwartz, 2001). Peer training has been associated with increased participation and decreased problem behavior exhibited during play. Given the research support for training siblings to facilitate play, one component of the intervention involved training Heidi, Jacob's sister, to effectively engage Jacob during play situations.

Training sessions occurred prior to each intervention session and involved two researchers and Heidi. The first session training lasted 15 minutes, each subsequent session training last 5-10 minutes. During training, the researchers focused on teaching Heidi four strategies: (1) use indirect social overtures, (2) give frequent and enthusiastic verbal praise, (3) prompt functional communication responses, and (4) limit reactions to problem behavior. Teaching occurred in three phases: (1) the researchers explained the strategy and the rationale for the strategy, (2) the researchers demonstrated how to implement the strategy in a role play (one instructor pretended to be Jacob and the other pretended to be Heidi), and (3) Heidi was asked to demonstrate the strategy with one of the researchers in a role play. Initially, during the third phase, one researcher sat with Heidi and gave her corrective feedback as she implemented the strategy in the role play with the other researcher. As Heidi became more proficient with the strategy, the second researcher faded corrective feedback during the role play. The training sessions was terminated when Heidi was able to demonstrate each strategy independently (e.g., without researcher prompts) in the role play scenario. In total, five training sessions were conducted.

Heidi was taught three strategies that were expected to reduce Jacob's anxiety/hyperarousal around play scenarios and one strategy that would eliminate one of the factors maintaining aggression. First, Heidi was taught to use indirect social overtures by creating play situations in which she "needed help". For example, she would start to put a puzzle together and say, "I wish I knew where to put this next piece" or "I hope that someone can help me with this puzzle." These statements were intended to be social overtures to encourage Jacob to join the situation. However, in contrast to baseline, these

social overtures were indirect in nature. During baseline, Heidi would direct Jacob to join the play by saying, “Jacob, come put the puzzle together” or “Put this piece over here.” The goal of implementing indirect social overtures was to reduce the demand characteristics of the situation (thus reducing Jacob’s arousal level). He could enter into the play situation when he was comfortable and on his terms.

Second, Heidi was taught to use frequent and enthusiastic verbal praise when Jacob was engaging in appropriate behavior. Jacob enjoyed it when people clapped for him and cheered, “Yeah, Jacob!” Heidi was taught to use this enthusiastic reinforcement every time Jacob followed the rules of the game, waited his turn without a tantrum, or shared his toy. The goal of this strategy was to create a positive play atmosphere that was motivating for Jacob, thus reducing his anxiety. An additional goal was to reinforce and strengthen appropriate play skills, allowing him to be more successful in the target context.

Third, Heidi was taught to prompt Jacob to use functional communication responses if he appeared frustrated. This strategy was described to Heidi as, “help Jacob find words”. The researchers explained to Heidi that Jacob’s problem behavior often functioned as a way for him to communicate. She could help him communicate by finding the words he needed to say. For example, if Jacob grabbed a toy, she could model the words, “Can I have the toy?” If Jacob tipped over a board game, she could model for him, “I am all done”. This strategy was practiced extensively during the training, as it was the most complex and difficult for Heidi to implement. By the end of training, she was able to effectively prompt the two phrases just described, the most common functional communication responses required during their play.

Lastly, Heidi was taught to reduce her verbal reactions to Jacob's problem behavior. The purpose of this strategy was to eliminate one of the maintaining variables for Jacob's behavior, attention from his sister and mother. During baseline, Heidi typically reacted to Jacob's aggression by screaming and running out of the room. In training, Heidi was taught to verbally ignore Jacob's attempted aggression and move out of Jacob's physical proximity. Jacob's mother was also instructed to verbally ignore the behavior. If problem behavior required immediate intervention, Jacob's mother was instructed to neutrally block aggression (by moving Jacob's arms down to his side) and prompt a functional communication response. Heidi was told that she could leave the play situation at any time if she felt uncomfortable. If she wanted to leave, she was instructed to get up quietly and exit the room.

In addition to the sibling training, the researchers introduced visual supports into the play situation in the form of a choice board. The goal of the choice board was to increase predictability for the interaction and reduce the demands associated with initiating play. As such, it was expected that the choice board would reduce Jacob's anxiety/hyperarousal associated with initiating play. The choice board contained digital pictures of Jacob's preferred games and activities. Prior to engaging in play, Jacob's sister would choose four activities she would like to play. She would then show Jacob the choice board with the four activities and allow him to choose an activity. The choice board allowed Heidi to have input into which activity they played (since she picked the four activities) and allowed Jacob to initiate the activity with support for his verbal communication.



### *Intervention Agent Training*

The principal investigator trained the natural intervention agents (Jonathan's and Jacob's parents) to implement the multicomponent intervention package. Prior to intervention data collection, the parents were required to meet criteria for the four stages of training. If, at any point during intervention, the parents did not demonstrate 100% fidelity, training was reinitiated by the principal investigator.

### *Intervention Fidelity*

Appendix D displays the intervention fidelity checklist related to the intervention components in Phase 2. The intervention fidelity checklist was completed by the primary observer for 80% - 100% of the intervention sessions and by a second observer in 21% of sessions for Jonathan, and 71% of the sessions for Jacob.

### *Interrater Reliability*

As in Study 1, the primary observer for each participant varied depending on the nature of the setting. For Jonathan, the primary observer was the parent, as intervention occurred early in the morning in the family's home. For Jacob, the primary observer was the principal investigator. To assess interrater reliability, a second observer was present during some of the baseline and intervention sessions. For both participants, the second observer was a researcher. The researcher was a doctoral student who, prior to data collection, was able to achieve acceptable reliability with the principal investigator.

The same binary reliability index used in Study 1 was used in Study 2 to evaluate agreement on intervention fidelity, latency to the first instance of problem behavior, and reason for session termination. For each session, reliability was scored as either agreement or no agreement. For intervention fidelity, agreement was defined as all the

steps of the intervention package checked as implemented correctly on the intervention fidelity checklist. For latency to the first instance of problem behavior, agreement was defined as both observers recording a time within five seconds of one another. For reason for session termination, agreement was defined as identifying the same description for session termination (i.e., occurrence of tolerated problem behavior, occurrence of intolerated problem behavior, or successful completion of routine).

For Jonathan, two observers completed reliability checks on 17% of the baseline sessions and 21% of the intervention sessions. Agreement on intervention fidelity, latency to the first instance of problem behavior, and reason for session termination was noted in 100% of reliability sessions. For Jacob, two observers completed reliability checks in 44% of baseline sessions and 71% of intervention sessions. Agreement on intervention fidelity, latency to the first instance of problem behavior, and reason for session termination was noted in 100% of reliability sessions.

## Results

### *Phase 1: Assessment of Contexts Associated with Behavioral Phenotypes*

For each participant, six observation sessions were conducted for the target context and four observation sessions were completed for the control context.

#### *Participant 1: Jonathan*

Figure 5 shows the latency to session termination due to problem behavior or successful session completion for the contexts observed for Jonathan. For the target context, social tasks that provoked anxiety/hyperarousal, 0% of the sessions were completed successfully. Specifically, 5 of the 6 target sessions were terminated due to intolerated problem behavior. The remaining target session was terminated due to tolerated problem behavior. The mean latency to session termination due to problem behavior was 1 min, 59 s (range: 3 s to 10 min) across all six target context sessions. All control sessions (100%) were completed successfully. The mean length of the control context session was 14 min, 58 s (range: 10 min to 15 min).

#### *Participant 2: Jacob*

Figure 6 shows the latency to session termination due to problem behavior or successful session completion for the contexts observed for Jacob. For the target context, social tasks that provoked anxiety/hyperarousal, 0% of the sessions were completed successfully. All six target sessions were terminated due to intolerated problem behavior. The mean latency to session termination due to intolerated problem behavior was 1 min, 41 s (range: 18 s to 4 min, 24 s). All control context sessions (100%) were completed successfully. The mean length of the control context session was 4 min, 53 s (range: 4 min, 30 s to 5 min, 30 s).

*Phase 2: Intervention for the Problematic Context Associated with the Behavioral Phenotype*

Figure 7 presents data on the amount of time that elapsed before the first instance of problem behavior or successful completion of the session (no problem behavior observed) for each participant. Latency to the first instance of intolerated problem behavior is represented by solid black bars, latency to the first instance of tolerated problem behavior is represented by grey bars, and successful completion of the session is represented by open bars. If intolerated problem behavior occurred, the sessions were terminated at the first instance of behavior. For Jonathan, tolerated problem behavior occurred in all six baseline sessions. The mean latency to the first instance of problem behavior was 4 min 13 s (range: 3 s to 12 min). During intervention, Jonathan did not display problem behavior; all 14 sessions were completed successfully. The mean duration of successfully completed sessions during intervention was 14 min, 50 s (range: 12 min, 45 s to 15 min).

For Jacob, 89% of baseline sessions were terminated due to intolerated problem behavior. The mean latency to session termination during baseline was 1 min, 13 s (range: 30 s to 2 min, 18 s). The remaining baseline session, which was successfully completed, lasted 2 min, 24 s. During intervention, 86% of sessions were completed successfully, without problem behavior. The mean duration of successfully completed intervention sessions was 5 min, 17 s (range: 3 min, 54 s to 6 min, 12 s). Tolerated problem behavior occurred in one intervention session. The latency to problem behavior in that session was 8 min, 45 s.

## Discussion

The present study demonstrated preliminary support for behavioral phenotypes as contextual factors for problem behavior. For both participants with ASD and FXS, problem behavior was associated with target contexts that were based on the behavioral phenotype for the participant's developmental disability syndrome. In contrast, problem behavior was not associated with the control contexts, which did not require the participants to use skills in areas of weakness. Empirically supported interventions were effective at reducing problem behavior in the target contexts. This unique association between the target contexts and problem behavior suggests that behavioral phenotypes may play a significant role in the conceptualization of problem behavior. The literature on behavioral phenotypes indicates areas in which there may be a mismatch between an individual's skills and the demands of the environment. The heuristic model described in this study suggests that analysis of this mismatch may lead to more efficient and effective models for assessment and intervention for problem behavior.

### *Behavioral Phenotypes and Functional Assessment*

Within the behavioral field, there exists a substantial literature documenting that the function of problem behavior is associated with aversive situations (Carr et al., 1999; Hanley, Iwata, & McCord, 2003). Unfortunately, there are a large number of factors that may be aversive for a given individual. Traditional behavioral assessment methods are undertaken without regard for the individual's specific developmental disability syndrome. That is, individuals showing problem behavior receive a generic behavioral assessment in which the features of the behavioral phenotype are *not* considered. As a result, clinicians may spend substantial amounts of time assessing factors that are not

related to the individual's problem behavior, or they may miss important factors related to such behavior.

A more efficient model for assessment might involve the systematic evaluation of factors likely to be associated with problem behavior based on the individual's specific developmental disability syndrome. In this model, knowledge of the behavioral phenotype associated with the syndrome could be used to narrow down the areas of relative skill weaknesses from the universe of all possible areas to a select and plausible few. The clinician could then undertake the process of evaluating the contextual factors most likely to be associated with problem behavior for the individual.

Although the current study focused on ASD and FXS, the proposed model could be extended to other disabilities to illustrate its breadth. Thus, consider the case of a clinician who is conducting an assessment of a child with Down syndrome. Using the proposed model, the clinician would examine the literature on the behavioral phenotype of Down syndrome to focus assessment efforts on specific phenotype features as possible contextual factors for problem behavior. For Down syndrome, potentially relevant features would include the following: deficits in short term verbal memory, associated medical conditions, and difficulty communicating needs (Chapman & Hesketh, 2000; Dykens, Hodapp, & Finucane, 2000). Using this knowledge, the clinician could systematically ask specific questions about the potential relationship between these factors and problem behavior. For example, to address weaknesses in short term verbal memory, the clinician might ask, "Does your child have difficulty in situations where s/he needs to respond after s/he is given large amount of verbal information? If so, when in these situations, does your child show problem behavior?" To address associated

medical conditions, the clinician might ask, “Does your child experience any of the medical conditions commonly associated with Down syndrome, such as congenital heart defects, ophthalmic conditions, endocrine conditions, or dental conditions? If so, does your child suffer from pain or discomfort associated with these conditions? Does s/he display problem behavior when experiencing such pain? Also, does your child need to frequently visit the doctor to address these conditions? When you visit the doctor, does your child show problem behavior?”

Importantly, by using information derived from the behavioral phenotype, the clinician would not invest valuable time assessing tasks or situations that are not likely to be associated with problem behavior. The clinician would know that the child most likely has deficits in auditory short term memory (*not* visual-spatial processing). The clinician would also know, in contrast to children with FXS, that the child would not be anxious in social situations. In fact, the literature on Down syndrome indicates the children with this syndrome are often very social and do well in social situations (Dykens, Hodapp, & Finucane, 2000). In this way, the clinician would be able to efficiently fine tune his/her assessment to the factors *most likely* to be associated with problem behavior.

#### *Context-Based Intervention*

The contextual fit theory suggests that problem behavior results from a poor fit between the individual’s competencies and the performance demands of the environment. This theory suggests two possible intervention points: improving the competencies of the individual (building skills) and changing the performance demands of the environment (context modification).

Having two potential intervention points provides the clinician with flexibility when designing intervention packages that are consistent with the family's values and feasible in the real world. Recall the intervention for Grace, the young girl with ASD who displayed problem behavior at her community ballet class. The best intervention package for Grace would have incorporated changing the demands of the environment in the ballet class by working with the ballet teacher. However, given that the teacher was a college student who taught the class in her spare time, it was not feasible to modify the context by training the ballet teacher. Based on contextual fit theory, there was another possible intervention point – building Grace's skills to better match the demands of the environment. By working with Grace's mother, a social story intervention was implemented to provide Grace with consistent expectations and teach her skills to succeed in the environment.

In addition to feasibility, contextual fit theory allows flexibility when designing interventions that are desirable for the family to implement. Recall Jonathan, the young man with FXS who exhibited problem behavior in the morning when getting dressed. Jonathan experienced anticipatory anxiety about his day. Research has demonstrated that visual schedules may be helpful at reducing anxiety by providing the individual with predictability. When this idea was presented to the family, they did not seem comfortable with the idea of a visual schedule. As such, we ultimately decided on verbal rehearsal of his day as the intervention strategy. The family approved of this intervention and was able to consistently implement the strategy.



### *Behavioral Phenotypes and Context-Based Intervention*

Clear understanding of the variables that control problem behavior from a functional assessment informed by knowledge of behavioral phenotypes may lead to more effective interventions for problem behavior. Consider the examples of ASD and FXS. Individuals with both of these diagnoses have difficulties in social situations. More sophisticated analysis of the behavioral phenotypes for these two disabilities reveals that individuals with ASD are often not socially motivated and display poor social skills. In contrast, the social difficulties experienced by individuals with FXS are often a product of anxiety or hyperarousal in social situations. These two behavioral profiles suggest different intervention strategies. For example, the work of Strain and colleagues has demonstrated that teaching peers and siblings to persist with social initiations toward children with autism has been effective at increasing the number of social interactions (Strain & Danko, 1995). During peer training, peers are taught to repeatedly attempt social initiations, even when the child with autism does not respond. This type of intervention, which has been demonstrated to be successful with children with autism, may actually heighten anxiety in individuals with FXS. Frequent initiation attempts may increase the individual's perceived pressure to perform, thus increasing their anxiety and potentially leading to an increase in problem behavior.

A more phenotypically sensitive intervention for individuals with FXS would involve creating an environment that would decrease anxiety associated with social interaction. Such an intervention was demonstrated in Study 2 with Jacob and his sister. Prior to intervention, Jacob's sister would attempt to interact with Jacob by repeatedly asking him to play. Jacob typically responded by ignoring her or hitting her. For the

intervention, Jacob's sister was taught to solicit participation in play through incidental overtures (e.g., "I wish someone would help me put this puzzle together") instead of direct initiations (e.g., "Come put together the puzzle with me"). Through these overtures, Jacob was able to enter the social situation when he was comfortable, thus reducing his anxiety and problem behavior. Knowledge of the behavioral phenotype resulted in the development of a more effective intervention.

#### *Future Directions*

The present study presents preliminary support for a heuristic model incorporating the literature on behavioral phenotypes into assessment and intervention with participants with ASD and FXS. This study had several limitations. The small sample size limits the scope of the implications that can be drawn from the results. Target and control contexts selected for observation were not set up or manipulated by the researcher. As such, causal associations between the characteristics of the contexts and problem behavior cannot be determined. During the intervention phase, multicomponent intervention packages were implemented to reduce problem behavior. Because multiple interventions were used, it is difficult to evaluate which interventions made the most significant impact on the rates of problem behavior. The naturalistic nature of these methods highlights the external validity of the study, but limits the precision of the analysis among the variables under investigation.

Despite these limitations, the results of this study lay the groundwork for programmatic research that can continue to integrate knowledge about behavioral phenotypes with behavioral assessment and intervention. Future research should investigate the relevant variables within the relationship between behavioral phenotypes,

contextual fit and problem behavior. Analog assessments, in which the relevant features of behavioral phenotypes are selected and manipulated, may be used to evaluate causality in these relationships. For example, the current study suggested an association between deficits in social interaction for individuals with ASD and problem behavior. Future research could systematically evaluate the impact of possible moderating variables, such as age, sex, proximity, and number of peers, on problem behavior. Other relevant variables in this relationship may include the length of the interaction, the social skills of the peers, the familiarity of peers, and the conditions of the environment. To maximize external validity, these variables could be manipulated within natural settings.

Another direction for future research involves evaluation of the model of assessment and intervention proposed in this study. Is this model, which incorporates knowledge of behavioral phenotypes into the assessment process, more efficient and effective than other models of assessment? Efficiency and effectiveness could be evaluated by comparing the results of assessment and intervention with this model to more traditional models of behavior assessment. To evaluate breadth, efficiency and effectiveness should also be evaluated with other developmental disabilities, including, but not limited to, Down syndrome, Prader-Willi syndrome and Williams syndrome.

#### *Concluding Comment*

The goal of the present study was to connect two scientific traditions, biomedical and behavioral, within the field of developmental disabilities. Historically, these two fields have remained independent of one another. This study presents a model for how to integrate the literature of behavioral phenotypes to generate new systematic assessment and intervention options for problem behavior. Future research should continue to

evaluate these relationships and the clinical utility of incorporating knowledge of behavior phenotypes into the assessment and intervention process. In this manner, a broader understanding of problem behavior and greater clinical effectiveness in remediating such behavior could be achieved.

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## Appendix A

### Decision Tree for Study Inclusion and Participation

- 1. Does the child have a diagnosis of an autism spectrum disorder (ASD) or Fragile X syndrome (FXS)?**
  - a. Can the family provide a diagnostic evaluation to confirm the child's diagnosis?
    - i. If the child is diagnosed with FXS, does the evaluation include necessary genetic testing?
    - ii. If the child is diagnosed with an ASD, does the evaluation include:
      1. Satisfaction of DSM-IV criteria
      2. Use of Autism Diagnostic Observation Schedule-Generic (ADOS-G; Lord, Rutter, DiLavore, & Risi, 2000)
      3. If the child is diagnosed with an ASD, did s/he receive a score of at least 32 on the researcher completed Child Autism Rating Scale (CARS; Schopler, Reichler, & Renner, 1980). If **No**, exclude from study.
    - iii. Is the child diagnosed with other comorbid disorders? If **Yes**, exclude from study.
- 2. Does the child live at home with his or her family?**
- 3. Does the child display problem behavior?**
  - a. Is the presence of problem behavior confirmed by the child's scores on the Aberrant Behavior Checklist-Community (ABC-Community; Aman & Singh, 1994)?
- 4. Do the child's parents report that problem behavior interferes with the family's quality of life, ability to participate in typical family routines, or the child's ability to participate successfully in school?**

**If all four inclusion criteria are met, child may be considered for participation in the study.**

Appendix B

Participant	Phase 1 Contexts	Context Observed	Rationale
Study 1: Autism Spectrum Disorder (ASD)			
Grace	Social Skills Context*	“Centers” (free play) during preschool class	Interaction with typical peers during free play
	Control Context	Leisure activity alone	Same skill involved in target context without social interaction component
Edward	Social Skills Context*	“Center” (free play) during EI play group	Interaction with peers during free play
	Control Context	Leisure activity alone	No social interaction required during a typical routine
Matthew	Social Skills Context*	“Centers” (free play) during preschool class	Interaction with peers during free play
	Control Context	Leisure activity alone	Same skill involved in target context without social interaction component
Study 2: Fragile X Syndrome (FXS)			
Jacob	Social Anxiety Context*	Novel person present/novel routine	Common trigger associated with social anxiety
	Control Context	Interaction with familiar person (Ice cream man)	Social interaction without anxiety provoking component
Jonathan	Social Anxiety Context*	Novel person present/novel routine	Common trigger associated with social anxiety
	Control Context	Interaction with familiar person (Animal shelter job)	Social interaction without anxiety provoking component

Note: Asterisks denote target context based on the behavioral phenotype for the diagnosis.

## Appendix C

### Social Story Script for Ballet Class

I take a ballet class at (*name of ballet company*). (*peer name*) and (*peer name*) are in my ballet class. In ballet class I learn to dance like a ballerina. Ballet class can be so much fun!

In ballet class it is very important to: have a quiet voice, look at the teacher, listen to the teacher.

If my eyes are on the teacher and my mouth is quiet, the teacher will know that I am listening to her directions. I need to listen to the teacher's directions to learn to be a good ballerina.

The first thing we do at ballet class is stretch at the bar. It is important to keep both hands on the bar and look at the teacher. Then we do stretching on the floor. It is important to have a quiet voice and look at the teacher when I am stretching.

Sometimes it is hard to do the stretch. I may need the teacher to help me. I need to try to do the stretch first and then I can raise my hand to ask the teacher for help. I need to wait quietly for the teacher to help me.

After stretching, we will do some dances. The teacher will show us the moves for the dances. I need to look at the teacher and listen to her directions.

If I have a question, I can watch (*peer name*) or (*peer name*) to see what they are doing. I can also raise my hand and wait quietly for the teacher to help me. If I wait with a quiet voice, the teacher will be so proud of me!

Sometimes we dance all together and sometimes we dance one person at a time. When we dance one person at a time, we take turns going first. Sometimes I am the first person to dance and sometimes (*peer name*) or (*peer name*) is the first person to dance. It is good to take turns going first. Taking turns makes everybody happy!

If (*peer name*) or (*peer name*) is dancing I need to wait with a quiet voice. I need to stand up with my arms at my side. Soon it will be my turn to dance!

I need to have good behavior when I am in ballet class. Good behavior means that I have a quiet voice, look at the teacher, and listen to the teacher. Mom will watch me in the waiting room. If I am not having good behavior, she will come into class to remind me what I need to do. Mom will give me 2 warnings. After two warnings I will need to leave class.

Ballet class is so much fun when I have good behavior. Good behavior makes my teacher and my Mom proud of me!



## Appendix D

### Intervention Fidelity Checklists

#### Study 1: Autism Spectrum Disorder (ASD)

Grace		Edward		Matthew	
Date		Date		Date	
Social story read at least once before ballet class		Visual placement on table for the duration of the meal		Mood induction attempted – teacher attempts tickles or offers other preferred food or activity prior to tabletops	
Visual support with behavioral expectations shown to Grace and reviewed at least once before ballet class		Reinforcement system for following meal time rules implemented. One token provided every 2-3 minutes for following meal time rules.		Sesame Street (highly preferred toy) present during tabletops	
		Verbal praise for following meal time rules paired with tokens.		Break card present at tabletops and in Matthew's view	

#### Study 2: Fragile X Syndrome (FXS)

Jonathan		Jacob	
Date		Date	
Verbally review daily activities		Peer training with sibling prior to play. Sibling demonstrates following skills independently <b>during training</b> : (1) use indirect social overtures, (2) frequent and enthusiastic verbal praise, (3) prompt functional communication response (4) no reaction to problem behavior	
Verbal perseveration time offered from 8:30-9:00 am, or from time Jonathan is dressed until 9:00 am		Jacob prompted to choose activity from visual choice board.	
		Sibling demonstrates previously described skills <b>during play sessions</b> .	

# Appendix E

## Figures

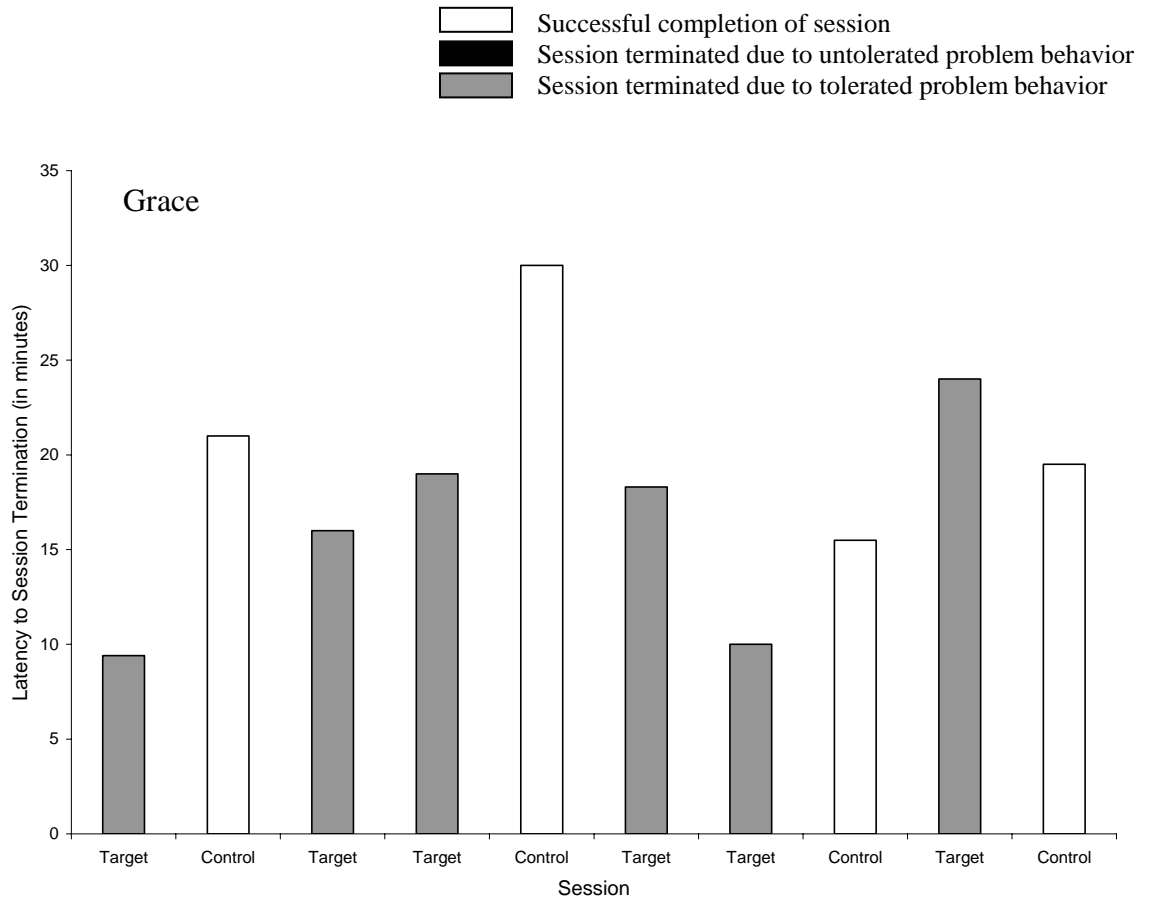


Figure 1. Latency to session termination for Grace during phase 1.

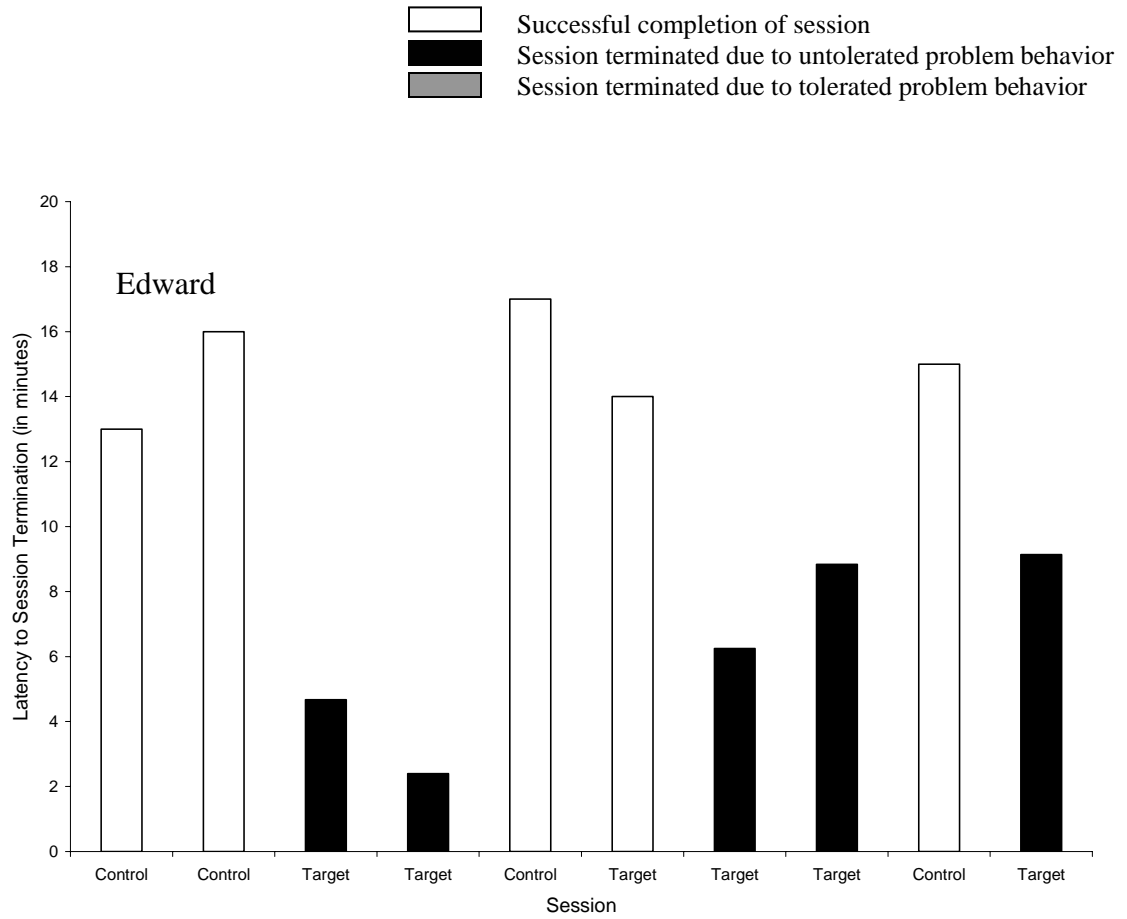


Figure 2. Latency to session termination for Edward during phase 1.

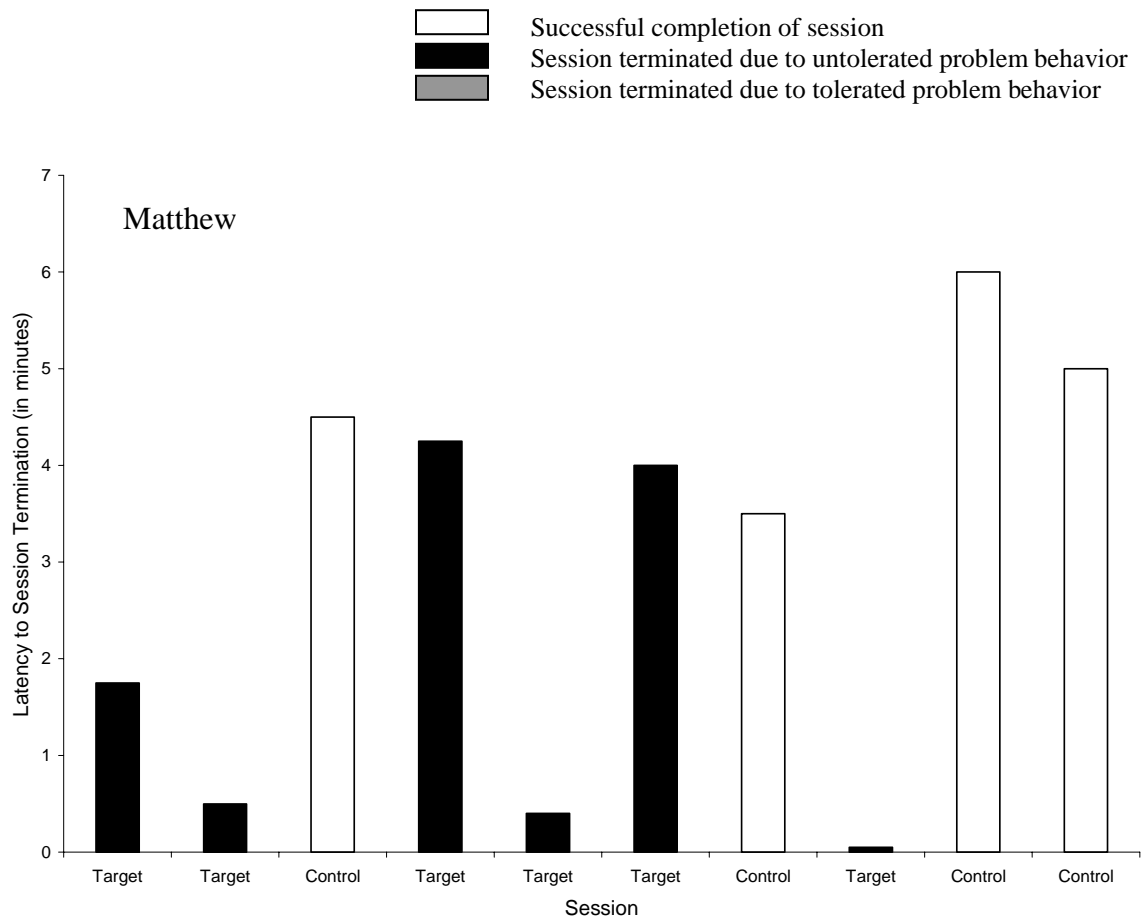


Figure 3. Latency to session termination for Matthew during phase 1.

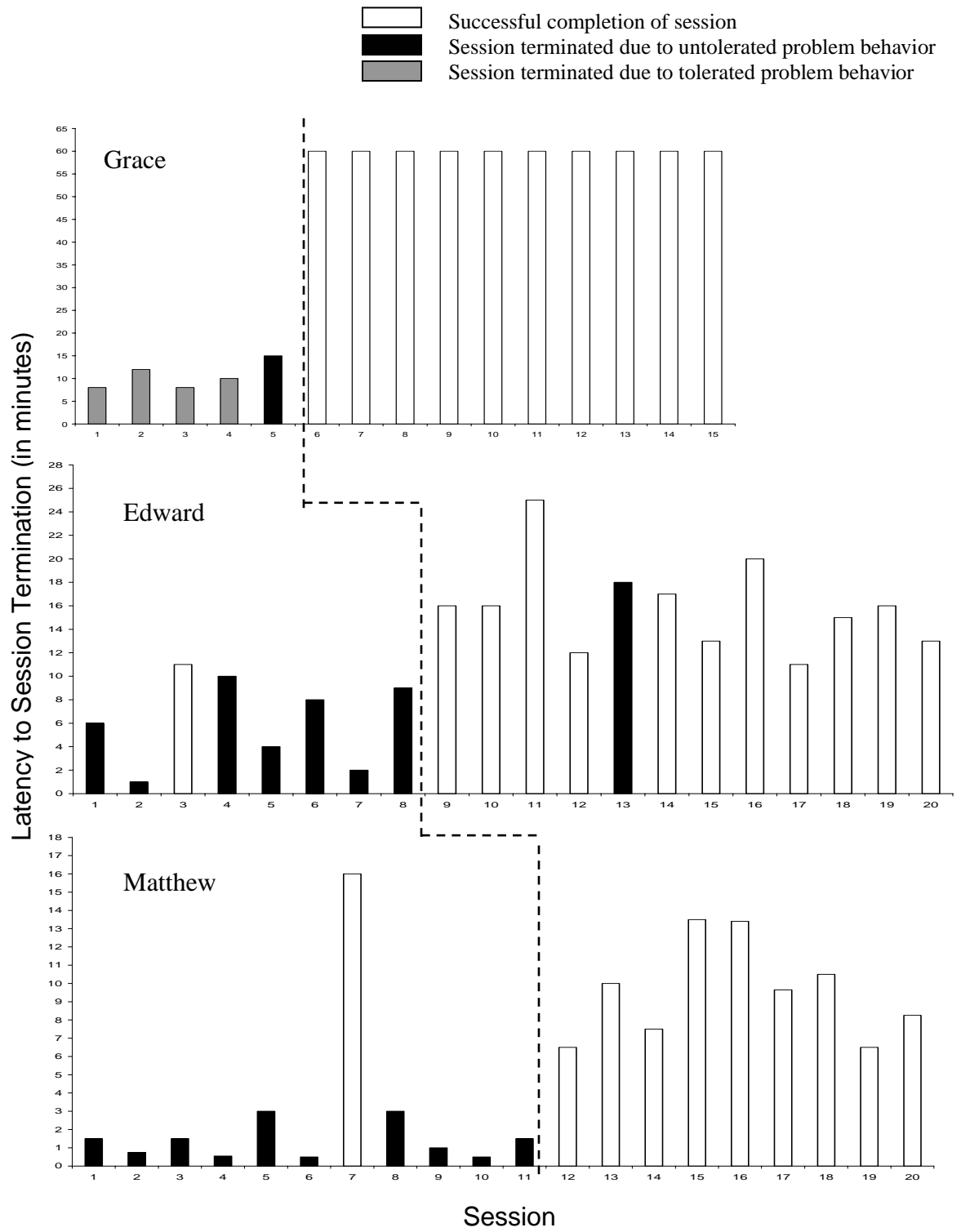


Figure 4. Latency to session termination for all three ASD participants during phase 2.

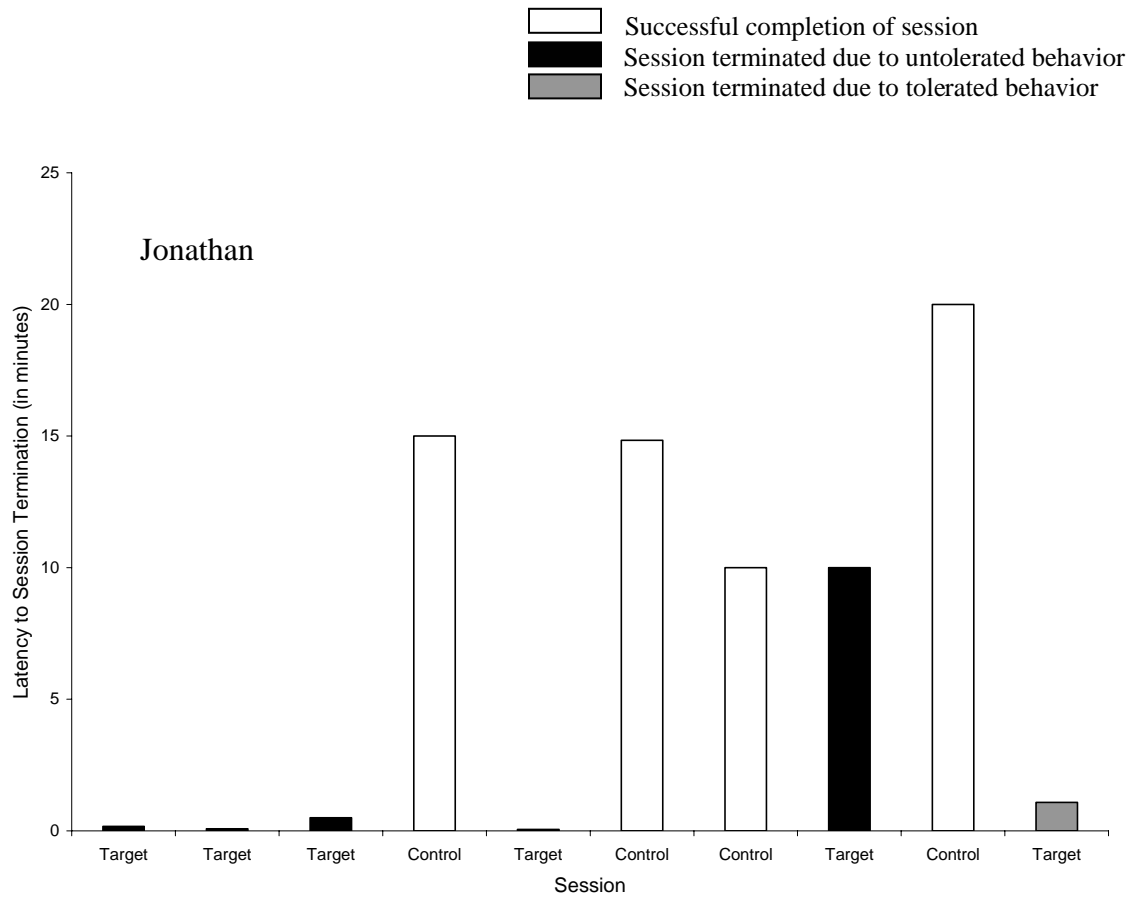


Figure 5. Latency to session termination for Jonathan during phase 1.

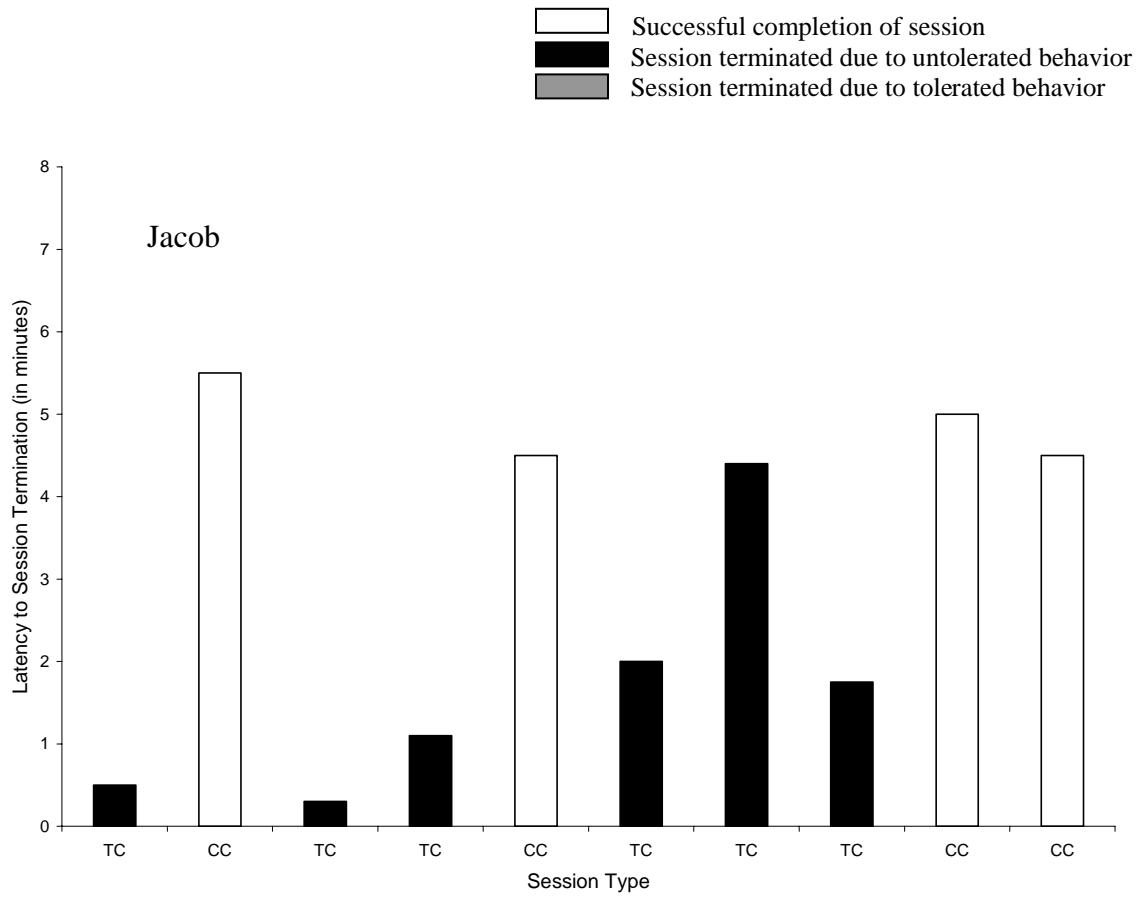


Figure 6. Latency to session termination for Jacob during phase 1.

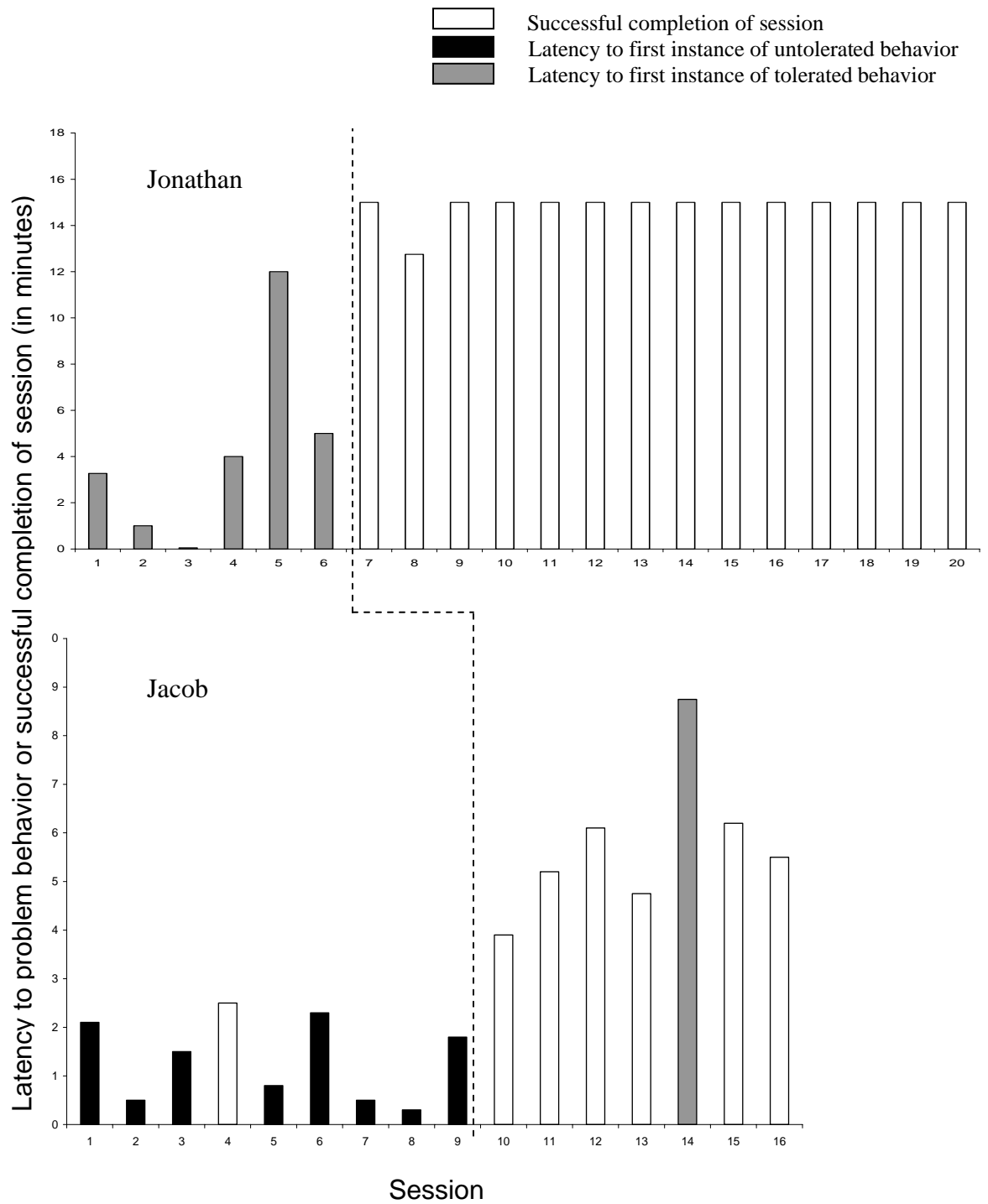


Figure 7. Latency to problem behavior or successful completion of session for both FXS participants during phase 2.