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**Assessment and Treatment of Anxiety and Problem Behavior in Children with  
Autism Spectrum Disorders and Intellectual Disability**

A Dissertation Presented

by

**Lauren Jill Moskowitz**

to

The Graduate School

in Partial Fulfillment of the

Requirements

for the Degree of

**Doctor of Philosophy**

in

**Clinical Psychology**

Stony Brook University

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**Stony Brook University**  
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Abstract of the Dissertation

**Assessment and Treatment of Anxiety and Problem Behavior in Children with  
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The relationship between anxiety and problem behavior has not been systematically investigated in children with ASD. Although there are numerous studies demonstrating successful treatment of anxiety in neurotypical populations, there is little research on the functional assessment and treatment of anxiety in children with autism spectrum disorder (ASD) and intellectual disability (ID). Thus, the present study evaluated a multimethod strategy for the assessment and treatment of anxiety and problem behavior in three children with comorbid ASD and ID. In Study 1, anxiety was operationally defined, a functional analysis of problem behavior was conducted in High-Anxiety versus Low-Anxiety conditions, and heart rate data was collected across conditions. Results indicated that, in High-Anxiety conditions, levels of problem behavior were high and, in Low-Anxiety conditions, problem behavior was low. Two of the three participants also exhibited a significantly higher heart rate in the High-Anxiety than in the Low-Anxiety conditions. In Study 2, a multiple baseline design was used to evaluate the effectiveness of a multicomponent intervention package, incorporating strategies from the Cognitive Behavioral Therapy (CBT) literature for neurotypical children with strategies from Positive Behavior Support (PBS) for children with DD. Following intervention, all three participants showed substantial decreases in anxiety and associated problem behavior in the situations that had previously been identified as anxiety-provoking.

## Dedication Page

I dedicate this dissertation to my former mentor, the late Dr. Edward (Ted) Carr, for his pioneering work in the field of functional behavior assessment and intervention for problem behavior in individuals with autism spectrum disorders and other developmental disabilities. Ted was a true visionary. I was immediately drawn to his view of the world and of life: about how clinicians sometimes have a tendency to impose societal values on the people we are trying to “help,” rather than working with the individuals to explore to what extent their environment can be modified to suit their individual preferences, skills, and personalities. During our first meeting, Ted explained his view maintaining the notion that problem behavior only served four main functions was outdated, and instead that problem behavior could serve many other functions, such as escaping/avoiding attention or social interaction, or escaping/avoiding pain or discomfort. I recall relating to Ted about the profound anxiety exhibited by many of the children with autism and fragile X syndrome with whom I had worked, and how I believed that a main function of problem behavior could also be to escape, avoid, reduce, or alleviate anxiety. Ted had an incredible enthusiasm, and it was on full display during that initial conversation and our many conversations thereafter. I was enthralled with Ted’s passion for improving the lives of others through his research. I sincerely hope I can honor Ted’s legacy by conducting clinical work, teaching, and applied research that have a meaningful impact on people’s lives and change the way people think, as he forever changed how I think – perhaps most of all by teaching me that the goal of therapy should be more than the absence of problem behavior; it should be the presence of a good quality of life.

I also dedicate this dissertation to my late grandmother, Beverly Wallach. Her unflinching (almost delusional) belief in me while I was growing up helped me to believe in myself. She gave me the strength to make it through difficult times. I would like to think I inherited her patience, compassion, and her relentless dedication to caring for other people; I could not perform this type of work without those qualities. When my grandmother was young, everyone called her “Pollyanna” because she was so benevolent – kind, good-natured, optimistic, and trusting. She showed me how to always see the best in other people. She always made me feel like the most special person in the world, and I hope that I help make other people feel the way she made me feel. If I do have that gift, it is because of my grandmother, Beverly Wallach.

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## **List of Abbreviations**

Autism Spectrum Disorders = ASD

Developmental Disability = DD

Intellectual Disability = ID

Pervasive Developmental Disorder = PDD

High-functioning autism = HFA

Asperger's syndrome = AS

Typically developing = TD

Autonomic nervous system = ANS

Heart Rate = HR

Heart Rate Variability = HRV

Respiratory Sinus Arrhythmia = RSA

Discriminative Stimulus = S<sup>D</sup>

Contextual Assessment Inventory = CAI

Stress Survey Schedule = SSS

Self-injurious behavior = SIB

Interobserver agreement = IOA

BL = Baseline

INT = Intervention

Cognitive behavioral therapy = CBT

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This dissertation could not have been conducted without the help and support of several important people. First, I would like to thank my doctoral advisor, Dr. Greg Hajcak, for graciously stepping in as my advisor after Dr. Ted Carr passed away, and for working with me to pursue the dissertation topic of my choice. I am grateful for his mentorship and guidance, both as a research advisor and as a clinical supervisor in Stony Brook University's Anxiety Clinic, which Greg created as a platform to train students in evidence-based assessment and treatment. I would not have been able to assess and treat the anxiety of the participants in this study without the foundation of knowledge I learned from Greg about assessing and treating anxiety. Our conversations have critically shaped my thinking as a clinical scientist. In addition, I have always felt that Greg treated me as a colleague, considering my opinions as important contributions to our conversation and respecting my judgment; I have learned that this combination of humility, openness, and collaboration is a rare quality, particularly in someone as intelligent as Greg.

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## Anxiety and Problem Behavior in Children with Autism Spectrum Disorders

Recent research suggests that anxiety-related concerns are among the most common presenting problems for children and adolescents with autism spectrum disorders (ASD) (White, Oswald, Ollendick, & Scahill, 2009). However, despite this research and longstanding clinical accounts of anxiety in children with ASD (e.g., Kanner, 1943, 1951), applied assessment and treatment research in individuals with ASD has focused almost exclusively on behaviors without acknowledging the role of affect and cognitions in the lives of these individuals. Thus, affective states such as fear, anxiety, or sadness are rarely discussed or acknowledged in this population. In fact, symptoms of anxiety generally go unrecognized in individuals with ASD and anxiety disorders are seldom diagnosed due to a general clinical consensus that symptoms of these disorders are “better explained by the ASD itself” (White et al., 2009). However, there is increasing recognition that negative affect is an important factor in understanding problem behavior in individuals with ASD and intellectual disabilities (ID), although negative affect is usually attributed to anger or frustration rather than anxiety (Murphy, 1997). As a result, behavioral assessment and intervention research in children with ASD has often neglected the role of anxiety, particularly its potential role in contributing to problem behavior.

Problem behavior (e.g., aggression, self-injury, tantrums, property destruction, disruptive behavior, noncompliance, elopement) is commonly displayed by individuals with ASD and other developmental disabilities (DDs) (Emerson et al., 2001). This problem behavior is a major barrier to education, social development, integration, employment, and community adaptation (Bruininks, Hill, & Morreau, 1988; Janney & Meyer, 1990; Koegel et al., 1992; Lucyshyn, Dunlap, & Albin, 2002; Turnbull & Ruef, 1996). Due to its negative impact on quality of life, problem behavior has been a major focus of research and intervention in the field of ASD/DD.

Research has demonstrated that intervention targeting problem behavior in children with DD is about twice as likely to succeed if it is based on a functional assessment, or assessment of the factors controlling problem behavior, than if it is not (Carr et al., 1999). Problem behaviors can function to gain attention from others (Carr & McDowell, 1980; Durand, Crimmins, Caulfield, & Taylor, 1989; Mace et al., 1986), escape from aversive task demands (Carr, Newsom, & Binkoff, 1980; Carr & Newsom, 1985; Steege et al., 1989), escape from social interaction (Taylor & Carr, 1992), gain preferred tangible items (Durand & Crimmins, 1988a; Sasso et al., 1992), gain sensory reinforcement (Favell, McGimsey, & Schell, 1982), and escape from aversive sensory stimuli (O'Reilly, 1997). However, it is not always possible to identify the function(s) of problem behavior in individuals with ASD and DD (McGill, 1999). In fact, in approximately 30% of cases, researchers using functional assessment could not identify a socially mediated function (i.e., to gain attention, to escape demands) that was controlling problem behavior (Derby et al., 1992; Iwata et al., 1994; Kennedy, 1994; Vollmer, Marcus, & LeBlanc, 1994), requiring clinicians to design an intervention based on indeterminate assessment results (Vollmer et al., 1994). Likewise, anecdotally, parents and teachers of children with ASD frequently report that the children's problem behavior often seems to come out of nowhere, with no reliable antecedents and no apparent function.

One explanation for this unpredictable behavior is that psychological distress, in particular anxiety, may be an internal, and thus often unobservable, antecedent to problem behavior (Romanczyk & Mathews, 1998). Anxiety could either be the discriminative stimulus ( $S^D$ ) that directly and immediately leads to problem behavior (e.g., child feels anxious so he hits his mother) or the setting event that sets the stage for problem behavior to be triggered by another  $S^D$  (e.g., child feels anxious so, when asked to do homework, he is more likely to hit)

(Freeman, Horner, & Reichle, 1999). Thus, behaviors that appear to be out of the blue may actually be influenced or controlled by internal factors such as pain, illness, fatigue (Carr & Smith, 1995), anxiety, or another emotional state that the individual with ASD is unable to communicate. The present study proposes that one of the functions of a child's problem behavior could be to escape, avoid, or alleviate his distress, in particular to escape/avoid or alleviate his anxiety. Although the difference between escaping/avoiding an anxiety-provoking situation or stimulus or internal state and escaping/avoiding a disliked or non-preferred task or demand may at first glance appear to be only a semantic distinction, there are intervention implications; conceptualizing problem behavior as due to fear or anxiety rather than due to noncompliance, disobedience, anger, or irritability may lead to very different interpretations and attributions by parents, teachers, and therapists, as well as different intervention strategies to reduce or prevent the problem behavior.

### **Background: Anxiety in ASD**

Recent research has shown that anxiety, as a specific form of affective distress, appears to be particularly prevalent in children with ASD (for a review, see White et al., 2009). In fact, as many as 80% of children with ASD present with clinically significant symptoms of anxiety (Muris, Steerneman, Merckelbach, Holdrinet, & Meesters, 1998) and approximately 42% to 84% of children with ASD meet criteria for at least one anxiety disorder (Muris et al., 1998; de Bruin, Ferdinand, Meester, de Nijs, & Verheij, 2007; Simonoff et al., 2008; Sukhodolsky et al., 2008). This is likely an underestimate, given the difficulties of assessing anxiety in this population. Nevertheless, fear and anxiety are reported to be more prevalent in children with ASD than in typically developing (TD) children (Matson & Love, 1990; Kim, Szatmari, Bryson, Streiner, & Wilson, 2000; Bellini, 2004; Kuusikko et al., 2008; Leyfer et al., 2006; Weisbrot, Gadow,

DeVincent, & Pomeroy, 2005) as well as those with other DDs (Bradley, Summers, Wood, & Bryson, 2004; Brereton et al., 2006; Evans, Canavera, Kleinpeter, Maccubbin, & Taga, 2005; Gilliot, Furniss, & Walter, 2001). Further anxiety appears to be associated with increased impairment in social functioning in children and adolescents with ASD (e.g., Bellini, 2004).

Despite the fact that clinicians have long asserted that individuals with ASD experience high levels of anxiety (e.g., Attwood, 2000; Kanner, 1943, 1951), and that investigators have recently called for treatments that address anxiety-related symptoms in ASD (e.g., Sofronoff, Attwood, & Hinton, 2005; White et al., 2009), behavior analytic research has largely neglected the potential role of anxiety in contributing to problem behavior in individuals with ASD. Anxiety involves *affective* states (e.g., subjective fear and panic experienced), *cognitions* (beliefs, thoughts, and images, such as worry and dread), *behavioral* escape or avoidance of the feared situation (and nonverbal behaviors such as crying, whining, and visible muscle tension), and associated *physiological arousal* (Barlow, 2000, Wolpe, 1958). Hagopian and Jennett (2008) described an anxiety disorder as a fear response that is “out of proportion relative to the actual threat, and/or extreme in its intensity to the extent that it significantly disrupts the individual’s functioning.” Groden, Cautela, Prince, and Berryman (1994) make the point that operant researchers have been reluctant to use the construct of anxiety either as an explanatory concept or in a descriptive manner when discussing those with ASD and DD. This may be because, unlike behaviors, the cognitions, subjective state, and physiological variables that are part of the construct of anxiety often cannot be directly observed. Further, anxiety in neurotypical or “typically developing” (TD) children is usually assessed by asking the children and their caretakers to report on their thoughts, feelings, and behaviors. Given that children with ASD often cannot report or articulate their emotional states, traditional assessment of anxiety using



paper-and-pencil self-reports or verbal self-reports (interviews) is often difficult or impossible (Hagopian & Jennett, 2008). After all, communication in autism is universally impaired to some degree; approximately one-half of individuals with autism are functionally nonverbal and those who have verbal language often have difficulty describing their mental states, mental experiences, and daily life experiences (Leyfer et al., 2006) and tend not to signal their emotional states to others (Rogers, 1998). In addition to the limitations of self-report due to the communication deficits of children with ASD, the fact that children with ASD may express their thoughts or emotions in idiosyncratic ways might cause caretakers to be unaware of their children's thoughts, feelings, and behaviors, which could also limit the usefulness of informant reports and interviews. Thus, anxiety is often overlooked or unrecognized in children with ASD.

### **Why are Individuals with ASD more susceptible to Fear and/or Anxiety?**

Although many of the factors involved in contributing to or maintaining anxiety in TD children are likely to be involved in the development of anxiety in children with ASD, there is a lack of research to support this. There are several mechanisms that have been proposed regarding what may make children with ASD anxious. Broadly speaking, anxiety could be caused by external triggers (e.g., change, unpredictability, transitions, new person, loud noise, crowds) or internal triggers (i.e., centrally-driven, such as interoceptive cues, autonomic arousal, somatic sensations, or intrusive images). For example, changes in routines and interruption of stereotyped behaviors may increase anxiety, tension, and emotional upset in children with ASD (Volkmar, Cook, Pomeroy, Realmuto, & Tanguay, 1999), who often show acute symptoms of anxiety and panic in response to environmental changes and transitions (Steingard, Zimnitzky, DeMaso, Bauman, & Bucci, 1997).

On a biological level, limbic system dysfunction (Hutt & Hutt, 1970) and/or abnormal features of the amygdala (Baron-Cohen et al., 2000), which is involved in both social and emotional processes and plays an important role in moderating fear and anxiety, may contribute to co-occurring ASD and anxiety (Amaral et al. 2003). In fact, structural and functional abnormalities in the amygdala have been connected to both ASD (Nacewicz et al., 2006) and anxiety disorders (Rooszendaal, McEwen, Chattarji, 2009). Juranek et al. (2006) reported a significant association between anxiety and larger amygdala volumes in 49 children with ASD. In a study on abnormal fear conditioning and amygdala processing in an animal model of autism, Markram, Rinaldi, La Mendola, Sandi, and Markram (2008) suggested that it is possible that abnormal processing in the amygdala, in particular a hyperreactive amygdala, “might be central to the generation of these excessive fears observed in autism.” In addition, abnormal serotonin (5HT) neurotransmission could be a shared causal factor in ASD and anxiety (Chugani et al. 1999). Some researchers have suggested a shared heritability between ASD and anxiety disorders (Smalley, McCracken, & Tanguay, 1995). There is preliminary evidence that several genetic markers (i.e., gene polymorphisms) associated with anxiety in TD individuals are also associated with anxiety in individuals with ASD (Gadow, Roohi, DeVincent, Kirsch, & Hatchwell, 2009, 2010).

On a physiological level, it has been hypothesized that many of the symptoms of ASD may stem from atypical functioning of the autonomic nervous system (ANS) (Anderson & Colombo, 2009). Specifically, in comparison with controls, heightened autonomic responses at rest have been found in individuals with ASD, including higher skin conductance, HR, blood pressure, respiratory rate, and larger pupil size (Anderson & Colombo, 2009). Anderson and Colombo proposed that structural and/or neurochemical impairment in the ANS could play a

major role in the heightened baseline (i.e., at rest) autonomic responses in individuals with ASD. However, there is evidence for both hyperactive and hypoactive sympathetic activity in children with autism (Hirstein, Iversen, & Ramachandran, 2001). It may even be the case that the same child with autism can have either hyperactive or hypoactive sympathetic activity, depending on what the child is doing (Hirstein et al., 2001). In a study of children with autism, Hirstein et al. found that most of the children (26 out of 37) had abnormally high electrodermal activity and appeared to use “calming” self-stimulation activities (e.g., immersing their hands in dry beans, eating, sucking on sweets, being wrapped in a heavy blanket, deep pressure massage) to calm hyper-responsive sympathetic activity. However, they also found a subgroup of children (4 out of 37) who showed a very flat response, with either a complete absence of skin conductance responses (SCRs) or SCRs produced only by extreme activities, such as self-injurious behavior (which occurred in 2 out of the 4 children). Hirstien et al. suggested that this subgroup of children may engage in self-injurious behavior or risk-taking behavior to produce *more* autonomic activity. Thus, it may be that many children with autism need to engage in calming or relaxing activities when their arousal levels become too high in order to bring them down to a manageable level, whereas other children with autism may need to engage in self-injurious behavior or other high-intensity behavior if their arousal levels are too low in order to bring them up to an optimal level. Abnormally high or low levels of autonomic arousal could be a trigger of anxiety in children with ASD and cause them to engage in behaviors to increase or decrease their level of arousal in order to maintain homeostasis.

On a psychological level, Baron-Cohen (1995) suggested that the child’s deficit in making sense of people’s actions and intentions may trigger high levels of anxiety, which the child with autism may seek to control by “retreating into the predictable world of things and

systems.” Another theory espoused by some researchers (Chalfant, Rapee, & Carroll, 2007) is that children with ASD may be vulnerable to experience increased anxiety because they are over-selective in the way they process information, focusing on small details rather than the whole context (see “central coherence theory,” e.g., Frith, 1989, “stimulus overselectivity,” e.g., Lovaas & Schreibman, 1971). As Chalfant and colleagues point out, TD children with anxiety disorders are thought to have a similar information processing bias whereby they selectively attend to threat-related information and, as a result, misinterpret ambiguous situations as threatening because they fail to attend to the global context (e.g. Daleiden & Vasey, 1997; Kendall, 1985). If overselectivity contributes to anxiety, it may help explain why children with ASD appear particularly prone to heightened anxiety. After all, children with autism often respond to only one component of a stimulus, which appears to prevent them from acquiring and generalizing behaviors (Lovaas, Schreibman, Koegel, & Rehm, 1971). Thus, Koegel, Openden, and Koegel (2004) hypothesized that overselectivity may have caused the children with autism in their study who appeared “hypersensitive” to auditory stimuli to attend only to the auditory stimuli and prevented them from responding to more relevant aspects of their environment. Similarly, Groden, Baron, and Groden (2006) noted that the cognitive features of ASD (e.g., rigidity) appear to place children with ASD at a disadvantage in forming effective emotion coping skills. Further, Groden, LeVasseur, and Baron (2002) hypothesized that, due to the nature of autism, individuals with autism do not have the ability to cognitively appraise situations they may find anxiety-provoking or the coping skills to handle those anxiety-provoking situations. Overall, it is likely that there are many general characteristics inherent to autism – such as the need to maintain sameness as well as difficulties in communication, social interaction, forming or sustaining friendships, and understanding other people’s actions and intentions – that predispose

individuals with ASD to be more anxious than those without ASD, or that can predispose them to stressful experiences that lead to anxiety.

Children with ASD could also experience increased anxiety when their behaviors are in conflict with social expectations or demands or when their behaviors cause punishing reactions from others (Wood & Gadow, 2010). Based on research and their clinical experience, Wood and Gadow proposed the following possible sources of elevated stress in children with ASD: (1) repeated demands of teachers and others to conform and engage in assigned activities rather than in preferred routines and circumscribed interests; (2) difficulty understanding the perspectives of others, making daily social interactions unpredictable and at times overwhelming; (3) sensitivity to sound, touch, or light; and (4) teasing and rejection related to the social, communicative, and behavioral features of ASD (e.g., acting immaturely according to group standards). Wood and Gadow further postulated that stresses related to ASD could either contribute to increased global negative affectivity (a generalized, nonspecific risk factor for developing anxiety disorders) or, through learning processes, become specific foci of fear and anxiety.

### **Purpose of the Present Study**

Although the notion of anxiety in individuals with ASD has significant face validity, it has not been adequately operationalized to lend itself to experimental analysis. Wood and colleagues (2009) as well as other researchers have suggested that diagnosing anxiety in ASD should involve evidence of behaviors that are not part of the core domains of ASD, evidence that is consistent with the physiological, behavioral, or affective features of anxiety disorders (e.g., sympathetic nervous system arousal, fears), and evidence that distinguishes impairment in functioning due to symptoms of anxiety from impairment due to symptoms of ASD (e.g., a lack of participation in class due to social anxiety rather than to due to communication deficits)

(Leyfer et al., 2006; Matson & Nebel-Schwalm, 2007; Reaven & Hepburn, 2003). Moreover, Wood and Gadow (2010) called for the use of “objective measures of anxiety,” such as physiological arousal, as “benchmarks” for further validating diagnostic interviews and anxiety rating scales in ASD. They also recommended establishing convergent validity of purported measures of anxiety in ASD using a “heteromethod assessment strategy.”

Given the limitations of self-report in children with ASD, the difficulty distinguishing symptoms of anxiety disorders from symptoms of ASD (e.g., compulsions vs. repetitive behaviors), and the idiosyncratic behavioral expression of anxiety in this population, I aimed to evaluate a multimethod strategy for assessing anxiety in children with ASD which includes behavioral, psychophysiological, and contextual data to tap into the multiple components that make up the construct of anxiety. Specifically, in the present study, I assessed the behavioral component of anxiety by identifying the particular idiosyncratic behaviors that indicate anxiety (e.g., crying, pacing, freezing, following parent, reassurance-seeking), unique to each participant. Although any behavior on its own does not necessarily indicate anxiety (e.g., a child may cry because he is feeling afraid, sad, frustrated, tired, ill or in pain), multiple sources of converging data may suggest that the behavior is a sign or marker of anxiety. I indexed the physiological component of anxiety by examining heart rate and Respiratory Sinus Arrhythmia (RSA) as well as observable indicators of physiological arousal (e.g., sweating, flushed face, visible muscle tension). Finally, I evaluated the affective component of anxiety (i.e., subjective fear or panic experienced) by examining parent-reports on the contexts that elicit anxiety using the Stress Survey Schedule (SSS; Groden et al., 2001) and the Contextual Assessment Inventory (CAI; McAtee, Carr, & Schulte, 2004). The rationale for using these contextual measures, the SSS and CAI, is that the process of labeling one’s state of affective arousal as “anxiety” or any other

emotion is highly influenced by the situational context in which the arousal occurs (Bandura, 1988). For example, if one's heart were racing while exercising, the arousal would not likely be interpreted as anxiety, whereas if one's heart were racing while taking an exam, the arousal might be interpreted as anxiety because of the context in which the arousal occurs. Defining anxiety by collecting multiple converging pieces of evidence that point toward anxiety is a critical first step in determining whether a relationship exists between anxiety (a biological setting event) and a problem behavior in children with ASD. Should it be the case that anxiety is associated with elevated levels of problem behavior, then it becomes important to design interventions that are effective in reducing anxiety in this population.

### **The Clinical Utility of Physiological Measures**

This paper proposes that physiological measures can provide information about arousal, a main component of anxiety, in children with ASD. Although physiological measures might currently be difficult or impractical for use in clinical practice, many researchers recommend using physiological measurement for the assessment of anxiety in particular (e.g., King, Ollendick, & Murphy, 1997). After all, as previously mentioned, limitations of traditional methods for assessing anxiety (questionnaires/rating scales, interviews, direct observations) make it difficult to establish if and when anxiety is occurring and why it is occurring in children with ASD. The use of physiological measures such as heart rate (HR), heart rate variability (HRV), respiration, blood pressure, or skin conductance can inform us of the types of situations and stressors that cause increased arousal for individuals with ASD and the nature of varying stress responses, especially for those individuals who are not able to complete self-reports or reliably articulate their thoughts and feelings due to deficits in language and communication (Freeman et al., 1999; Romanczyk, Lockshin, & O'Connor, 1992). Even children without ASD

or intellectual disability (ID) may verbally report lower than actual levels of anxiety, but their physiological and behavioral responses more accurately reflect their levels of distress (Velting, Setzer, & Albano, 2004). Thus, using physiological measures may elucidate the meaning of behavioral data, which can often be difficult to interpret on its own. Converging data can guide interpretation of both physiological and behavioral measures. In particular, assessment of physiological arousal using telemetric monitoring (wireless technology that can collect and transmit data remotely) allows us to capture proximal events and behaviors closer to their actual occurrence, which can help in establishing temporal precedence (i.e., allowing causal influence more than a traditional correlational, cross-sectional design) (Goodwin, Velicer, & Intille, 2008). Further, using unobtrusive equipment to measure physiological arousal in naturalistic environments can lead to less behavioral reactivity than might occur with direct observation in laboratory studies or clinical settings. Overall, investigations of individual differences are often enhanced by considering concurrent physiological and behavioral responses, and data from a variety of sources can often strengthen behavioral data (Boccia & Roberts, 2000). Physiological measures of arousal thus have the potential to improve our understanding of overt behavior in children with ASD and can ultimately lead to improved prevention strategies (Grodén et al., 2005). In the present study, I examined not only mean HR, a general measure of arousal, but also respiratory sinus arrhythmia (RSA), an index of vagal activity which reflects beat-to-beat changes in HR coupled to the respiratory cycle. Incorporating physiological measures into functional assessments could be useful in identifying internal precursors to problem behavior that are not directly observable and helping to predict when problem behaviors are more likely to occur. This could then help to prevent problem behaviors before they do occur.

### **Background: Physiological Arousal in ASD**



The autonomic nervous system (ANS) is generally thought to have two branches: the sympathetic system, associated with energy mobilization (“fight-or-flight”), and the parasympathetic system, associated with vegetative and restorative functions (“rest-and-digest”). Whereas the sympathetic system is excitatory, the inhibitory parasympathetic system works to slow the heart, which creates more beat-to-beat variability in HR; this variability is considered to reflect vagal tone. Although heart rate variability (HRV) results from the relationship between sympathetic and parasympathetic influences, RSA indexes solely the parasympathetic nervous system, which is controlled by the vagus nerve (Porges, 1995). Specifically, heart rate increases when one is breathing in and decreases when one is breathing out; RSA is the difference between the heart rate during inspiration versus expiration, which reflects the influence of the vagus nerve in regulating the heartbeat, with a larger RSA indicating greater vagal activity and a smaller RSA reflecting lesser vagal activity.

It has been hypothesized that many of the symptoms of ASD may stem from atypical functioning of the autonomic nervous system (ANS) (Anderson & Colombo, 2009). Although some older studies did not find differences in mean heart rate between children with autism and their TD peers (e.g., Hutt, Forrest, & Richer, 1975; Lake, Ziegler, & Murphy, 1977), several recent studies have reported a higher heart rate in children with autism under resting conditions (Bal et al., 2010; Goodwin et al., 2006; Kootz & Cohen, 1981; Ming, Julu, Brimacombe, Conner, & Daniels, 2005). Further, in comparison with controls, heightened autonomic responses at rest have been found in individuals with ASD, including higher HR, skin conductance, blood pressure, respiratory rate, and larger pupil size (Anderson & Colombo, 2009). Anderson and Colombo proposed that structural and/or neurochemical impairment in the ANS could play a major role in the heightened baseline (i.e., at rest) autonomic responses in individuals with ASD.

Interestingly, results from a study by Ming and colleagues found that, compared with a group of TD children, there was elevated sympathetic activity (resting HR, diastolic blood pressure, and mean arterial blood pressure) and low baseline parasympathetic activity (vagal tone) in the majority of children with autism in their study, whether or not they had symptoms or signs of autonomic abnormalities. They suggested that deficits in parasympathetic function in children with ASD may result in relatively unrestrained sympathetic activity. Other studies have also found that children with ASD show reduced levels of baseline RSA compared with TD children (Bal et al., 2010; Vaughan Van Hecke et al., 2009). In addition to higher sympathetic activity and lower parasympathetic activity while at rest, physiological arousal or “autonomic defensiveness” (i.e., acceleration in HR, respiration, pupil dilation) in response to environmental stimulation has been shown to occur in individuals with autism (e.g., Goodwin et al., 2006; Groden et al., 2005; Kootz, Marinelli, & Cohen, 1982). Further, Corbett, Mendoza, Wegelin, Carmean, and Levine (2008) found that, in comparison with TD children, evening values of salivary cortisol for children with autism tended to be consistently elevated, which they proposed could reflect a greater responsivity to the events of the day.

**The relationship between stereotypic behavior and HR.** Hutt and colleagues originally proposed that individuals with ASD may engage in problem behavior to achieve homeostatic regulation (Hutt & Hutt, 1965, 1968, 1970). Sroufe, Stuecher, and Stutzer (1973) found evidence in support of this hypothesis when they examined the covariation between self-stimulatory behavior (e.g., finger-flicking), HR, and behavioral indices of stress (e.g., muscular tension, facial expression) in a 6-year-old boy with autism. They found a significant association between HR accelerations and episodes of body rocking; in new situations and during new tasks, finger-flicking was preceded by HR acceleration. Similarly, Hutt et al. (1975) found that, for a group of

children with autism, HR decreased significantly 5 seconds after engaging in repetitive motor movements. Further, they found that the children with autism had higher resting HR than a group of younger TD children and a group of age-matched TD children. The authors interpreted these findings as evidence for heightened levels of arousal in ASD as well as support for the hypothesis that engaging in problem behavior leads to a decrease in arousal. They hypothesized that the monotony produced by repetitive motor movements and the corresponding blockage of novel sensory input might be the mechanisms which lead to the decrease in arousal. Lewis et al. (1984) correlated stereotyped body rocking with cardiac activity in 17 adults with autism and severe to profound ID. Although they found increases in mean HR and HRV during periods of stereotypical behavior, in contrast to Sroufe et al., they did not find a significant positive correlation between HR and body rocking. However, increased body-rocking rate was significantly correlated with an increase in HRV. Lewis et al. interpreted the findings as evidence for “cardiac-somatic coupling” (Obrist, Webb, Sutterer, & Howard, 1970), in which a hypo-aroused individual increases motor movement to maintain optimum metabolic functioning.

Willemsen-Swinkels, Buitellar, Dekker, and van Engeland (1998) measured HR changes around the onset of stereotypical behaviors associated with distress, elation, and composure in 26 children, 14 with a pervasive developmental disorder (PDD). Each time a stereotypic behavior was performed, the observer judged (based on information other than the stereotypic behavior itself) whether the child was experiencing positive excitement or negative excitement. Behavioral indicators for elation were, for example, laughing or other clear positive facial expressions and the content of verbal or nonverbal communication (such as requests for the repetition of an event). Behavioral markers for distress were, for example, crying, screaming, aggressive actions towards parent or objects, attempts to escape, and negative verbal or

nonverbal communicative messages. If there were no clear signs of excitement, the stereotypic behavior was coded as “composure.” Results revealed a positive correlation between HR increases and stereotypical behaviors associated with distress just before the onset of the behavior. Specifically, in 85% of the stereotypic behaviors associated with distress, the peak value in HR had occurred just before the onset of the behavior. Their findings suggest that individuals with PDD may engage in stereotypical behavior while distressed as a functional response for decreasing distress or arousal. The authors concluded that their results supported the homeostasis interpretation that stereotypic behaviors “compensate for an overstimulating environment and serve a calming function.”

In spite of this evidence that self-stimulatory behavior serves a calming or de-arousing function, recent evidence suggests a more complicated picture. Specifically, there is evidence for both hyperactive and hypoactive sympathetic activity in children with autism, suggesting that self-stimulatory behavior can either serve a de-arousing or arousing function (Hirstein et al., 2001). It may even be the case that the same child with autism can have either hyperactive or hypoactive sympathetic activity (i.e., fluctuate between over-arousal and under-arousal) depending on the context, suggesting that the child may engage in self-stimulatory behavior to either increase or decrease arousal, and thus maintain homeostasis. Hirstein et al. found that most children with autism had abnormally high electrodermal activity and appeared to use “calming” self-stimulation activities (e.g., immersing their hands in dry beans, eating, sucking on sweets, being wrapped in a heavy blanket, deep pressure massage) to calm hyper-responsive sympathetic activity. However, they also found a subgroup of children who showed a very flat electrodermal response, with either a complete absence of skin conductance responses (SCRs) or SCRs produced only by extreme activities, such as self-injurious behavior (SIB). Hirstien et al.

suggested that this subgroup of children may engage in SIB or risk-taking behavior to produce *more* autonomic activity. Thus, it may be that many children with autism need to engage in calming activities when their arousal levels become too high in order to bring them down to a manageable level, whereas other children with autism may need to engage in SIB or other high-intensity behavior if their arousal levels are too low in order to bring them up to an optimal level. Abnormally high or low levels of arousal could thus be a trigger for problem behavior in children with ASD in that it may cause them to engage in problem behavior to increase or decrease their level of arousal (to maintain homeostasis).

**HR in response to stressors.** Although Hirstein et al. suggested that self-stimulatory behavior may serve a calming function in children with ASD, they did not actually expose the children in their study to stressors. In a more recent series of studies, Groden and colleagues (Groden et al., 2005; Goodwin et al., 2006) assessed cardiovascular responses to stressors in children with ASD. Specifically, Goodwin et al. used the LifeShirt to compare HR in response to environmental stressors in five boys with autism and five TD children. Each session began with a 5-minute baseline phase (sitting quietly with a familiar person), followed by six potentially stressful situations that were alternated with 2-minute rest phases. The stress phases were from the *Stress Survey Schedule for Persons with Autism and Developmental Disabilities* (SSS; Groden et al., 2001) and included: 1) Loud noise (sensory/personal contact), 2) Remote robot (anticipation/uncertainty), 3) Unstructured time (anticipation/uncertainty), 4) Eating a preferred food (pleasant event), 5) Difficult task (changes/threats), 6) Change in staff (unpleasant event), and 7) Transition (time between stressors & rest phases). The findings did not support the hypothesis that the group with autism would show significant HR responses to a greater number of stressors than the TD group; out of the 35 opportunities for each group to show a significant

mean HR response to a stressor, the group with autism showed significant responses only 22% of the time, compared with the TD group, which showed significant responses 60% of the time. Although these results make it appear that the group with autism is less aroused by environmental stressors than the TD group, Goodwin et al. pointed out that this reduced reactivity to potential stressors in the group with autism may be due to their higher baseline HR and reduced variance in responsivity, or to being overly aroused by the testing situation, or to being in a general state of autonomic defensiveness. The authors suggested three possible explanations for why findings of physiological overarousal do not always replicate in individuals with autism, citing Zahn (1986): it is possible that (a) most individuals with autism have chronically high levels of autonomic activity, but that a subgroup of individuals with autism may respond at normal levels; (b) individuals with autism may exhibit heightened autonomic activity only some of the time; and (c) physiological studies requiring interaction with people or that create confusion about what the participant is being asked to do can cause the individual to display high arousal.

I also propose that – given that two children with autism in the Goodwin et al. (2006) study showed no HR changes to any of the potential stressful situations, two others showed changes to two stressors, and one child showed changes to four stressors – this supports the need to take a more idiographic approach to physiological arousal in autism, and to the study of anxiety in ASD in general, rather than simply examining group differences. For instance, it is possible that the analog tasks used in the study by Goodwin et al. (e.g., vacuum cleaner, remote-control robot) may have been anxiety-provoking, stressful, and/or physiologically arousing to one child with autism, but not to another child. Similarly, Groden et al. (2005) found that each of the four stress domains chosen from the SSS and their corresponding stress situations elicited

statistically significant changes in HR across some of the participants with autism and DD, but not others. Specifically, the stress domain of *changes/threats* (assessed by engagement in a difficult task) elicited significant HR changes in 40% of the sample, the stress domain of *pleasant events* (assessed by eating a preferred food) elicited significant HR changes in 60% of the sample, the stress domain of *anticipation/uncertainty* (assessed by unstructured time) elicited significant HR changes in 60% of the sample, and the stress domain of *unpleasant event* (assessed by a change in staff) elicited significant HR changes in only 20% of the sample. Thus, it is important for researchers and clinicians to examine idiosyncratic stimuli or situations that have been reported by parents or teachers to make particular children anxious or aroused, rather than standardized laboratory situations that may make some children with ASD anxious but not others. After all, Reese, Richman, Zarcone, and Zarcone (2003) found that sensory responses to idiosyncratic stimuli may play a role in increasing the probability of disruptive behavior exhibited by individuals with autism. In addition, Carr, Yarbrough, and Langdon (1997) compared rates of problem behavior in sessions that included idiosyncratic stimulus variables with rates of problem behaviors in sessions that did not include those idiosyncratic variables. Carr et al. (1997) found that the presence or absence of these idiosyncratic stimulus variables could dramatically alter the outcome of the functional analysis; the role of negative reinforcement (i.e., escape from demands) and positive reinforcement (i.e., social attention) in maintaining the problem behavior was not clear until the authors analyzed the effect of idiosyncratic stimulus variables. This supports the need to examine physiological arousal in response to idiosyncratic stimuli and situations in individuals with ASD.

**Parasympathetic activity.** Whereas several studies have examined heart rate (HR) in children with ASD, very few studies have analyzed heart rate variability (HRV), which reflects

the degree to which heart activity can be modulated to meet changing situational demands. HR, which is the number of beats per minute, is a general measure of arousal that has both sympathetic and parasympathetic influences. In contrast, analysis of HRV, which is the variation in the time interval between heart beats (or variation in the beat-to-beat interval), allows us to separate the sympathetic and parasympathetic contributions to variability. This is because HRV is mediated by the vagus nerve and controlled primarily by the parasympathetic branch of the ANS. Due to its parasympathetic influence, high vagal tone is thought to reflect the ability to regulate emotions, or the ability to self-soothe when stressed (Porges, 2007). It is thought that, in individuals with poor vagal regulation, sympathetic influences to the heart will be “unchecked” and the individuals will therefore be unable to attenuate the naturally occurring sympathetic reactivity to stressful challenges (Bal et al., 2010). This balance between the effects of the sympathetic and parasympathetic systems is reflected in the beat-to-beat changes of the cardiac cycle. Metrics of HRV allow us to examine the activity of the two branches of the ANS in conjunction, as the sympathetic and parasympathetic system can covary reciprocally, independently, or nonreciprocally (Bernston, Cacioppo, & Quigley, 1993). As such, measures of HRV can provide an important window into the central control of autonomic processes and by inference, the central processes necessary for organized behavior. Although RSA is not a direct measure of fear or anxiety, research has demonstrated that inhibited children who are low on approach and high on fear exhibit low RSA (Kagan, Reznick, & Snidman, 1987), whereas higher RSA indicates appropriate engagement and emotion regulation, which manifests as social competence in older children (Doussard-Roosevelt, Porges, Scanlon, Alemi, & Scanlon, 1997). Research further suggests that baseline deficiencies in parasympathetic tone are related to negative emotional traits (e.g., depression, anxiety, aggression) and that excessive vagal



withdrawal is related to negative emotional states (i.e., panic and anger) (Beauchaine, 2001). Overall, shifts in RSA in response to environmental demands appear to reflect attentional focus, emotion regulation, and mood state (Beauchaine, 2001).

As mentioned, although several studies have examined sympathetic activity in children with autism, research examining parasympathetic activity is more limited. Several recent studies reported significantly lower levels of RSA in children with ASD than TD children (Bal et al., 2010; Ming et al., 2005; Vaughan Van Hecke et al., 2009). Toichi and Kamio (2003) examined heart activity based on Cardiac Vagal Index (CVI), which reflects parasympathetic activity, and Cardiac Sympathetic Index (CSI), which reflects sympathetic function, in response to mental tasks requiring sustained attention in individuals with autism and age- and ability-matched controls. The authors found no significant differences in mean R-to-R intervals or HRV between the group with autism and the TD group under resting conditions. Further, while the TD group showed a significant decrease in the parasympathetic function during mental arithmetic tasks, the group with autism showed no significant changes in autonomic function. However, when examined individually, parasympathetic function was suppressed (marked decrease in CVI) during mental arithmetic in all but two participants in the control group, whereas parasympathetic function was activated (increase in the CVI) in half of the participants with autism. The paradoxical autonomic response suggested to the authors that some participants with autism were more stressed under “resting” conditions than while performing mechanical or repetitive mental tasks. The results seem to support autonomic hyperarousal in some people with autism, but not in others. This lends further support to the need to examine physiological arousal in response to idiosyncratic stimuli and situations in individuals with autism rather than standardized laboratory situations. Toichi and Kamio also suggested that the elevated

sympathetic function found in some studies might be partly attributable to differences in the characteristics of participants and/or experimental conditions during measurements. They also noted that lower-functioning individuals with autism (who purportedly have a larger region of abnormality in the limbic system) tend to show features of excited states, such as hyperactivity, self-stimulation, and other emotional instabilities, more frequently than higher-functioning individuals, and that these features are more common in children than in older individuals. Thus, the authors reasoned that elevation in sympathetic function is more likely to occur in lower-functioning children with ASD than the higher-functioning individuals with ASD they studied.

To assess whether children with high-functioning ASD (HFA) and TD controls displayed the adaptive response of suppressing vagal activity during a challenge or stressor, Vaughan Van Heck et al. (2009) measured RSA while the children viewed videos of a familiar and an unfamiliar person reading a story. Vaughan Van Hecke et al. found that the HFA group had lower overall RSA levels than the TD group and exhibited decreased RSA to the video of the unfamiliar person, whereas the TD group maintained baseline levels of RSA to the unfamiliar person. It should be noted, however, that the HFA group returned to baseline RSA during the familiar person and moving objects videos. Surprisingly, the TD group showed a more dampened RSA reactivity to the task demands. Nevertheless, the HFA group showed, overall, lower levels of RSA across conditions in comparison with the TD group (in support of findings by Ming et al., 2005). Given that lower RSA reflects increased arousal and poorer control of the vagus nerve over the systems of fight, flight, and immobilization (Porges, 2007), Vaughan Van Hecke et al. suggested that children with autism may be in a chronically “mobilized” state compared with TD children. Further, because the children with HFA showed a specific RSA reaction to a video of an unfamiliar person (whereas the TD children did not show decreased

regulation of the heart while viewing an unfamiliar person), the authors proposed that unfamiliar people may be particularly “threat-inducing” stimuli for children with autism. The authors reasoned that children with ASD may react to unfamiliar people with a precautionary mobilization to fight or flee, in contrast to TD children’s increase in HR regulation in response to unfamiliar people. Of note, results also indicated that higher RSA (better regulation of RSA) was related to higher social skills ratings and fewer problem behaviors (as rated by caregivers) in the children with HFA. In sum, the authors concluded that, considering the pattern of RSA suppression to the videos of unfamiliar people, it may be that unfamiliar people elicit an “anxiety-like” response in children with ASD.

In contrast to findings of lower levels of RSA (Ming et al., 2005; Toichi & Kamio, 2003), Watson, Roberts, Baranek, Mandulak, and Dalton (2011) did not find differences in RSA between children with ASD and their peers during either nonsocial or social stimuli. However, they found that children with ASD exhibited shorter IBIs than chronological age-matched TD children during exposure to both nonsocial and social stimuli, possibly suggesting a delayed or deviant autonomic system, or specific endogenous factors interacting with environmental conditions. These results are consistent with several previous studies that found higher heart rates for children with ASD than controls (e.g., Bal et al., 2010). Watson et al. (2011) noted that their findings that children with ASD had lower mean IBIs (i.e., increased heart rate) than TD children even during periods of sustained attention is consistent with an interpretation that children with ASD may have overactive systems or underactive parasympathetic systems, or both. However, the authors stated that their findings related to RSA do not support an underactive parasympathetic system (although they cannot completely rule out that possibility) and are consistent with the possibility that at least some children with ASD have overall higher levels of

sympathetic activity (as suggested by Hirstein et al., 2001). They did acknowledge, however, that there were a relatively small number of children with RSA data in each group and that RSA was calculated over relatively brief time periods, which could have contributed some instability to the RSA measurement. In addition, given that the children in their study (ages 29-42 months) were younger than in previous studies, it is possible that differences in RSA may not appear until later in development.

### **Physiological Predictors of Problem Behavior**

The aforementioned research has assessed the physiological reactivity of individuals with DDs to a variety of behaviors (e.g., Kootz & Cohen, 1981; Kootz et al., 1982; Lewis et al., 1989; Sroufe et al., 1973; Willemsen-Swinkels et al., 1998) and to a variety of environmental stressors (e.g., Goodwin et al., 2006; Vaughan Van Hecke et al., 2009) using cardiovascular measures. However, whereas most of these studies have examined the association between heart activity and stereotypic/self-stimulatory behavior, there is a paucity of studies that have formally linked physiological measures to problem behavior (e.g., aggression, self-injury, destructive behavior, disruptive behavior/tantrums). Further, while the aforementioned studies have examined arousal in *response* to behaviors or stressors, only three studies to date (Barrera, Violo, & Graver, 2007; Freeman, Grzymala-Busse, Riffel, & Schroeder, 2001; Freeman et al., 1999) have examined physiological measures *preceding* problem behavior.

Barrera et al. (2007) exposed three adults with DDs who exhibited severe, chronic self-injurious behavior (SIB) to functional analog experimental and control conditions while recording HR. Results demonstrated a reliable and consistent HR pattern across all participants for all conditions (Attention, Demand, Alone, and Control), consisting of an increase in HR immediately before SIB and quickly followed by a temporary drop in HR during or after SIB.

These patterns were evident regardless of SIB topographies, SIB durations, body positioning, movement, respiratory action, or baseline HR activity. The authors interpreted their results to suggest that internal or endogenous mechanisms of SIB were functioning in these participants (that autonomic arousal was a physiological precursor of SIB), with operant contingencies playing little or no role in the subsequent shaping or maintenance of SIB, although some additive effects may have accumulated. Barrera et al. thus proposed that SIB functions as a negative reinforcement mechanism that terminates, reduces, or allows escape from arousal.

Whereas the study by Barrera et al. used an analog design, Freeman et al. (1999) collected naturalistic data on the rate and covariation of problem behavior, HR, and environmental activities in two adult males with severe intellectual disability (ID). Using 15-s interval recording during periods of problem behavior and no problem behavior, they found that, for both participants, HR generally increased in the 15-s *following* problem behavior (i.e., self-bite, bang/slap), yet there was a low likelihood of HR increase in the 15-s *preceding* problem behavior. Thus, the HR data did not yield any information about physiological *precursors* that could be used to reliably predict the onset of problem behavior. This could be because self-injurious behavior (SIB) was combined with aggression and property destruction and/or because HR was averaged over only 15-s intervals and the researchers may have missed subtler variability-related changes preceding the occurrence of SIB. (The authors also noted that 15-s intervals may not have been a sensitive enough timescale to identify physiological precursors of problem behaviors). Freeman et al. (2001) further analyzed data obtained from one of the participants in the Freeman et al. (1999) study at longer time scales and found that SIB was more likely to occur in the presence of high HR 30-seconds before SIB and during engagement in SIB. The authors noted that future HR measures will need to be more sensitive, recording interbeat

intervals (IBIs) for more accurate rule sets to be identified. The present study recorded IBIs during 30-s intervals.

In another step toward establishing a link between physiological measures and problem behavior, Calamari, McNally, Benson, and Babington (1990) reported a positive relationship between high tonic (resting) HR and engagement in problem behavior (aggression and SIB) in Ms. M., a 23-year-old woman with severe ID. They found that administration of propranolol, a non-selective beta-blocker used in the treatment of hypertension, reduced resting HR and lowered the rate and severity of problem behavior. Specifically, initiation of treatment with propranolol was associated with a cessation in the dramatic escalation in aggressive behavior, and systematic increases in propranolol were correlated with progressive decreases in aggression. Ms. M.'s SIB also decreased as a function of propranolol dosage, although not as dramatically as the aggression decreased. In addition, increases in resting HR were associated with a worsening in Ms. M.'s behavior, especially the significant increase in aggression, and progressive increases in propranolol dosage were correlated with reductions in HR and in significant behavior improvement. In sum, there was a strong association between increases in propranolol, decreases in resting pulse rate, and decreased aggression for Ms. M. Conversely, increases in resting pulse rate were correlated with development of increased aggression and SIB. These data provides evidence for a relationship between autonomic arousal (i.e., HR and blood pressure) and problem behavior (i.e., aggression and SIB). The results of this study support the hypothesis that increased arousal could lead to or be associated with increases in problem behavior, and decreased arousal could lead to or be associated with decreases in problem behavior.

**Measuring physiological activity and problem behavior in the present study.** Taken together, this small but promising body of research suggests that problem behavior may be

linked with autonomic measures in individuals with ASD, and that these individuals may engage in problem behavior to achieve homeostatic regulation. However, there are several issues that the proposed study aims to address. First, whereas prior research has studied physiological measures in individuals with ASD and other DDs, the present study aims to take one step closer toward assessing the latent construct of “anxiety” in children with ASD by combining physiological data with detailed behavioral data and contextual assessment. Second, with only four exceptions (Barrera et al., 2007, Calamari et al., 1990, and Freeman et al., 1999, 2001), the majority of aforementioned studies examined self-stimulatory and stereotypic behavior rather than a wider range of problem behavior. Thus, the present study will examine the relationship between physiological measures and a wide range of problem behavior (e.g., disruptive behavior/tantrums, elopement, aggression, SIB). Third, previous research (with the exception of Freeman et al., 1999, 2001) has been conducted in analog (i.e., laboratory) settings rather than the real-life contexts in which stressors occur. Therefore, the proposed study will examine behavioral and physiological measures in naturalistic settings (e.g., home, school, community) rather than laboratory settings in an attempt to evaluate clinical utility. Relatedly, and more importantly, whereas previous research (e.g., Goodwin et al., 2006) has used standardized stressor tasks to attempt to evoke physiological reactions in children with ASD, the present study will examine idiosyncratic stimuli and situations that have been reported by parents and teachers to evoke anxiety and that naturally occur in the child’s environment. Finally, although the majority of the aforementioned studies (with the exception of Lewis et al., 1984, Ming et al., 2005, Toichi & Kamio, 2003, and Van Hecke et al., 2009) used HR as the measure of physiological arousal, it is possible that HR alone may be too general a measure and thus may not be the most useful index of physiological arousal, stress, and/or anxiety in terms of

predicting problem behavior in this population. After all, HR is a general measure of arousal which does not separate sympathetic and parasympathetic activity. Since the heart is dually innervated by the ANS, and the sympathetic and parasympathetic branches can have opposing effects on HR, this means that reductions in HR (which might be interpreted as evidence of under-arousal) can actually arise from increased vagal modulation or reduced sympathetic modulation. Likewise, increases in HR can arise either from heightened sympathetic or diminished vagally mediated influences. In other words, an increase in HR could result from either increased sympathetic activity *or* decreased parasympathetic inhibition (vagal withdrawal). Therefore, the proposed study will include not only HR but also RSA to allow a more fine-grained analysis of cardiovascular arousal, including the assessment of parasympathetic activity. Further, the present project will examine the interaction between heart activity, problem behavior, and environmental stressors in children with ASD.

### **Conceptualizing problem behavior functionally**

Carr and Smith (1995) outlined a contextual model of problem behavior in which problem behavior is viewed as a function of two contextual variables: *discriminative stimuli* and *setting events*. A discriminative stimulus ( $S^D$ ) is an event that sets the occasion for, or is correlated with, reinforcement of behavior (Skinner, 1938). Given that an  $S^D$  signals that problem behavior is likely to be reinforced, it therefore serves as a “trigger” for problem behavior (i.e., problem behavior occurs immediately after the presentation of the  $S^D$ ). Setting events are broad contextual factors that alter the relationship between  $S^D$ 's and responses (Bijou & Baer, 1978), thus influencing whether the  $S^D$  will or will not evoke problem behavior. A setting event can also alter the value of the consequences of behavior by making them more reinforcing or more aversive (Michael, 1982). For example, a child may typically find academic



demands aversive and periodically display problem behavior to escape from the demands. However, when the child is ill, the same demands become much more aversive, thereby increasing the reinforcing properties of escaping from the demand. In this case, illness would constitute a setting event that makes problem behavior more likely to occur in the presence of demands (because such behavior is now associated with strong reinforcers).

Taken together, discriminative stimuli and setting events form the *context* for problem behavior. Context variables consist of antecedent discriminative stimuli and setting events, and a given context variable can function as either an  $S^D$  or a setting event. Cale, Carr, Blakeley-Smith, and Owen-DeSchryver (2009) illustrated an example of how teasing could function as either an  $S^D$  (i.e., if a boy is teased by his peers and aggresses against them, then the teasing may stop) or a setting event (i.e., if the boy were teased earlier in the day and then given a task demand by his teacher much later in the day, he may aggress against the teacher, and the teacher may respond by removing the task). In the former example, teasing was the  $S^D$  in that it directly triggered problem behavior whereas, in the latter example, teasing functioned as a setting event that increased the aversiveness of the task, making aggressive behavior more likely in the presence of the  $S^D$  (i.e., the task demand).

Setting events include activities and routines (e.g., transitions, noisy or crowded environment), social/interpersonal factors (e.g., lack of attention, recently teased), and biological factors (e.g., illness, pain, discomfort, fatigue). Research has demonstrated that biological or internal variables such as fatigue (e.g., O'Reilly, 1995), menstrual pain (e.g., Carr, Smith, Giacini, Whelan, & Pancari, 2003), allergies (Kennedy & Meyer, 1996), and physical illness (e.g., Carr & Owen-DeSchryver, 2007) can be setting events for problem behavior. In illustration, consider a child with autism who occasionally shows problem behavior when his parents ask him to board

the school bus. On some days, his parents ask him to board the school bus ( $S^D$ ) and he complies (response), which results in his parents praising him (consequence). However, on other days, his parents ask him to board the bus ( $S^D$ ) and he exhibits aggression (response), which results in his parents withdrawing the demand and driving the child to school instead (consequence). Further assessment reveals that the child is experiencing fatigue on the days he shows aggression. In this scenario, fatigue functions as a setting event, increasing the aversiveness of the  $S^D$  (makes boarding the bus more aversive), and also increasing the reinforcement value of escaping from boarding the bus. Thus, on days in which the child is both fatigued (setting event) and asked to board the bus ( $S^D$ ), he is more likely to exhibit aggression and be allowed to escape from boarding the bus (consequence of negative reinforcement). On days in which he is not fatigued, the  $S^D$  is not as aversive, parental praise has a greater reinforcement value than escape, and the child is more likely to comply with boarding the bus. As illustrated in this example, a biological variable (fatigue) can be a setting event for problem behavior and substantially impact the display of this behavior.

**Anxiety as a context variable for problem behavior.** Based on the evidence that other biological factors can function as setting events for problem behavior (Carr & Smith, 1995), it is also possible that fear or anxiety can function as a setting event for problem behavior, making it more likely that individuals will display such behavior (Freeman et al., 1999). Additionally, since a context variable can function as either a setting event *or* an  $S^D$ , it is also possible that fear/anxiety can function as either an  $S^D$  or a setting event for problem behavior. In fact, it has been noted that anxiety-producing events often precede problem behavior in individuals with ASD (Grodén et al., 1994) and that arousal/anxiety can be an internal antecedent to problem behavior in these individuals (Romanczyk & Mathews, 1998), although these observations are

generally based on clinical observations rather than empirical studies. However, in a data-based study using parent-report measures of fear and problem behavior, Evans et al. (2005) found that the fears of children with ASD were more related to externalizing behavior problems than the fears of children with Down syndrome as well as mental-age-matched and chronological-age-matched TD children. Similarly, using a parent questionnaire to assess children's anxiety, Kim et al. (2000) reported that children with ASD who had anxiety and mood problems were more aggressive, limited their parents' social activities, and had poorer relationships with teachers, peers and family members than those children with ASD who had low scores on anxiety and mood problems. Further, using a hierarchical multiple regression, Rzepecka, McKenzie, McClure, and Murphy (2011) found that medication, sleep problems and anxiety accounted for 42% of the variance in challenging behavior in children with ID and/or ASD, with a large effect size. Although these studies are correlational and thus we cannot infer causation, it is plausible to suggest (based on Barrera et al., 2007; Calamari et al., 1990, Freeman et al., 1999, 2001, Groden et al., 1994, and Romanczyk & Mathews, 1998) that fear or anxiety may be causally or functionally related to problem behavior in many children with ASD, in that they engage in problem behavior to reduce their anxiety or escape/avoid an anxiety-provoking situation.

In support of the argument that engaging in problem behavior may serve to escape or reduce anxiety, Joosten, Bundy, and Einfeld (2009) added four items to the Motivation Assessment Scale (MAS; Durand & Crimmins, 1988b) to assess anxiety as an intrinsic motivator of stereotypical and repetitive behavior in children with ASD and ID and children with ID alone. The added items were based on the symptoms of anxiety reported in individuals with autism and included: (1) resistance to change, (2) being easily upset, (3) presence of tantrums, fearfulness, tenseness, and (4) agitation and irritability. Joosten et al. found that anxiety was a stronger

intrinsic motivator for stereotypy and repetitive behavior than sensory-seeking for children with dual diagnoses (both ASD and ID), whereas the reverse was true for children with ID only. These findings provide support for the notions that children with ASD may experience anxiety as a result of unpredictable sensory input or that children with ASD who are anxious may use sensation or familiar repetitive behavior as a way to calm themselves (Joosten et al., 2009). In addition to engaging in repetitive behavior as a way to calm themselves, it is plausible that many children with ASD may also engage in other problem behavior (e.g., tantrums, self-injury, aggression) to avoid, escape, reduce, or otherwise alleviate their anxiety. The hypothesis that anxiety may be causally linked to problem behavior is also supported by several case studies documenting a reduction of problem behavior following intervention to reduce anxiety (Davis, Kurtz, Gardner, & Carman, 2007; Luscre & Center, 1996; Mullins & Christian, 2001; Rapp, Vollmer, & Hovanetz, 2005). It is reasonable to propose, therefore, that many children with ASD may sometimes engage in problem behavior *because* they are anxious and do not know how to cope with this anxiety, or because problem behavior *is* in fact their way of coping with anxiety.

Specifically, given the contextual model of problem behavior described above (Carr & Smith, 1995), I propose that anxiety could either be the  $S^D$  that directly and immediately leads to the occurrence of problem behavior (e.g., child is feeling anxious and thus bites himself to reduce the anxiety) or the setting event that sets the stage for problem behavior to be triggered by another  $S^D$  (e.g., child is feeling anxious and thus, when asked to board the bus, he is more likely to bite himself).

Given the plausibility of an association between anxiety and problem behavior, I aimed to address the question of whether a multi-method assessment strategy that examines the construct of anxiety based on four sources of data can be used to reliably predict the likelihood of problem

behavior in children with ASD: (a) behavioral indicators of anxiety (e.g., avoidance, pacing, fidgeting, crying) and of physiological arousal (e.g., sweating, flushed face, visible muscle tension); (b) parent-report of contexts which elicit anxiety; (c) subjective ratings of anxiety by individuals with no knowledge of the purpose of the study; and (d) physiological data derived from measures of HR. Specifically, the present study aimed to address three research questions. First, Study 1 investigated whether, under conditions of maximum anxiety (i.e., known context for anxiety present, behavioral indicators of anxiety present, observable physiological indicators of anxiety present), we would observe more problem behavior than under conditions in which these indicators of anxiety were not present. Second, in Study 1, I aimed to examine whether behavioral and contextual indicators of anxiety would co-occur with physiological indices; whether, under conditions of maximum anxiety (i.e., behavioral indicators of anxiety present, observable physiological indicators of anxiety present, known context for anxiety present), I would find changes in patterns of HR and/or RSA in contrast to non-anxiety-provoking situations. Finally, in Study 2, I investigated whether a multicomponent behavioral intervention would reduce anxiety (e.g., frequency of observable anxious behaviors, subjective ratings of anxiety) and problem behavior in three children with ASD and co-occurring ID.

### **STUDY 1: ASSESSMENT**

In this first study, the construct of “anxiety” was operationally defined for each participant, and “High-Anxiety” and “Low-Anxiety” contexts were identified on the basis of parent interviews, subjective rating scales, and direct observation (as in Magito McLaughlin & Carr, 2005), in addition to heart rate for Ben and Jon. Next, I assessed the effects of anxiety on problem behavior. Specifically, for each participant, the effects of an anxiety-provoking (High-

Anxiety) versus non-anxiety provoking (Low-Anxiety) condition on the level of problem behavior were examined.

## **Method**

### **Identifying Participants (Selection of Participants)**

Participants were three school-aged children, ages 6 to 9 years old, diagnosed with an autism spectrum disorder, according to criteria specified in the *Diagnostic and Statistical Manual of Mental Disorders (DSM-IV, 2000)*. Participants were required to meet the following inclusion criteria: (a) diagnosis of an autism spectrum disorder using *DSM-IV* criteria, (b) presence of problem behavior in the home, school, and/or community, (c) presence of anxiety in the home, school, and/or community, and (d) parental consent. IRB approval and parental consent were obtained for all participants. Selection of participants was made on the basis of interviews with parents who contacted the author (L.J.M.) after receiving the recruitment letter through a local listserv for parents of children with ASD. The parents were asked a series of questions that can be found in the screening questionnaire in Appendix A. These interview questions were developed to aid in the identification of those children whose problem behavior appears to increase in frequency or intensity when they are anxious. Children were included in the study if they: (a) met *DSM-IV* diagnostic criteria for Autistic Disorder, Asperger's syndrome, or Pervasive Developmental Disorder – Not Otherwise Specified (PDD-NOS), (b) met *DSM-IV* diagnostic criteria for an anxiety disorder, including Generalized Anxiety Disorder, Obsessive-Compulsive Disorder, Panic Disorder, Specific Phobia, Social Phobia, Separation Anxiety Disorder, or Anxiety Disorder NOS; (c) had a history of behavior problems and reported anxiety, as determined by the interview questions in Appendix A; (d) had a history of behavior problems that became more frequent when specific stressors were present, as determined by ratings of 5 on

one or more items on the Activities and Routines section of the Contextual Assessment Inventory (CAI; Appendix B) or on the Stress Survey Schedule (SSS; Appendix C), and as confirmed by direct observation. If the participant met these inclusion criteria, the participant was retained, and several additional questions were asked. Specifically, another question posed was designed to identify the situations that were associated with anxiety so that I could more efficiently plan subsequent direct observations (i.e., “Are there certain events, people, or times of the day that are reliably associated with anxiety?”). I screened a total of eight participants, five of whom were excluded because parent interview and/or direct observation failed to confirm the presence of either problem behavior or anxiety. The remaining three participants satisfied the inclusion and exclusion criteria and were thus selected for the study.

**Participant 1.** Jon was a 6-year-old boy of Jamaican descent, diagnosed with autism, who was educated in a special education classroom and lived at home with his mother, father, and two younger brothers. Jon did not take any medications. Review of a prior psychoeducational evaluation of Jon indicated cognitive abilities in the low range. Specifically, on the Vineland Adaptive Behavior Scales (VABS-II; Sparrow, Balla, & Cicchetti, 1984), Jon’s overall level of adaptive functioning was in the low range; he received a VABS-II Adaptive Behavior Composite score of 56 (<1<sup>st</sup> percentile, low range), with a score of 74 on the Communication domain (4<sup>th</sup> percentile, moderately low range), 48 on Daily Living Skills (<0.1 percentile, low range), 49 on Socialization (<0.1 percentile, low range), and 64 on Motor Skills (1<sup>st</sup> percentile, low range). His score on the VABS-II Maladaptive Behavior Index was elevated, with a clinically significant Internalizing score and an elevated Externalizing score. Jon communicated through the use of 1-2-word phrases (e.g., “want cookie”).

**Participant 2.** Ben was a 9-year-old boy diagnosed with autism who lived at home with his mother, father, and maternal uncle. Ben received the educational classification of “autistic” and received Physical Therapy, Occupational Therapy, Speech and Language Therapy, and Social Skills Training. Ben did not take any medications. Review of a prior psychoeducational evaluation of Ben indicated that he falls within the mildly delayed range of intellectual functioning. Specifically, on the Stanford-Binet Intelligence Scales – Fifth Edition (Roid, 2003), Ben received a Full-Scale IQ (FSIQ) of 61 (1<sup>st</sup> percentile, mildly impaired range), with a Nonverbal IQ score of 70 (2<sup>nd</sup> percentile, borderline range) and a Verbal IQ score of 56 (0.2 percentile, moderately delayed range). On the VABS-II, Ben’s overall level of adaptive functioning was in the low range; he received a VABS-II Adaptive Behavior Composite score of 62 (1<sup>st</sup> percentile, low range), with a score of 67 on the Communication domain (1<sup>st</sup> percentile, low range), 63 on Daily Living Skills (1<sup>st</sup> percentile, low range), and 57 on Socialization (1<sup>st</sup> percentile, low range).

**Participant 3.** Sam was an 8-year-old boy diagnosed with PDD-NOS who lived at home with his mother, father, and younger brother. Sam received the educational classification of “autistic” and was educated in a self-contained special education classroom (8:1:1). Sam did not take any medications. Review of a prior psychoeducational evaluation of Sam indicated cognitive abilities in the extremely low range. Specifically, on the Weschler Intelligence Scale for Children-IV (WISC-IV; Weschler, 2003), Sam received a FSIQ score of 50, which is in the extremely low range (<0.1 percentile), with a standard score of 45 on the Verbal Comprehension Index (<0.1 percentile, extremely low); a standard score of 67 on the Perceptual Reasoning Index (1<sup>st</sup> percentile, extremely low); a standard score of 54 on the Working Memory Index (0.1 percentile, extremely low); and a standard score of 68 on the Processing Speed Index (2<sup>nd</sup>



percentile, extremely low). Sam was primarily a verbal communicator, using words, phrases, and sentences. On the Stanford-Binet, Sam received a FSIQ of 66, with a Nonverbal IQ score of 70 (borderline range) and a Verbal IQ score of 56 (extremely low). On the VABS-II, Sam's overall level of adaptive functioning was in the low range; he received a VABS-II Adaptive Behavior Composite score of 70 (2<sup>nd</sup> percentile, low range), with a score of 74 on the Communication domain (4<sup>th</sup> percentile, moderately low range), 79 on Daily Living Skills (8<sup>th</sup> percentile, moderately low range), and 59 on Socialization (<1<sup>st</sup> percentile, low range).

## **Measures**

**Contextual Assessment Inventory (CAI).** Parents who nominated their children for the study were asked to complete the CAI (McAtee et al., 2004). The CAI is a rating scale designed to help families and teachers identify the contexts (i.e., discriminative stimuli and setting events) that evoke problem behavior in home, school, and community settings. The CAI contains 24 items that make up three general categories of contextual factors (i.e., social, activities/routines, biological), with eight exemplars of each category (e.g., “hurried or rushed” for social category, “difficulty transitioning between settings or activities” for activities/routines category, “feeling tired” for biological category). Parents were asked to endorse the likelihood of problem behavior for each context using a 5-point Likert-type scale, with “1” indicating that problem behavior is not likely to occur in that context, and “5” indicating that it is very likely to occur. The CAI is efficient, comprehensive, comprehensible, and reliable (McAtee et al., 2004) and has both convergent and predictive validity (Carr, Ladd, & Schulte, 2008). Informants were only asked to complete the 8 items in the “Activity and Routines” category (#9-16). These items can be found in Appendix B.

### **Stress Survey Schedule for Persons With Autism and Other Pervasive**

**Developmental Disabilities (SSS).** Parents who nominated their children for the study were also asked to complete the SSS (Grodén et al., 2001). The SSS is a rating scale containing 49 events that relate to eight dimensions of stress: anticipation/uncertainty, changes and threats, unpleasant events, pleasant events, sensory/personal contact, food-related activity, social/environment interactions, and ritual-related stress. The SSS also contains six items that pertain to fears and seven items that pertain to life stressors. Parents were asked to rate the intensity of their child's stress reaction to the given events as a '1' (none to mild), '2' (mild to moderate), '3' (moderate), '4' (moderate to severe), or '5' (severe). The SSS also contains space to respond to the following questions: "Please list any other stressors on the lines below" and "Which do you consider the most significant stressors of those you have identified? Why?" Results of a study by Goodwin, Grodén, Velicer, and Diller (2007) demonstrated that the SSS is a valid tool for identifying which dimensions of stress are perceived to be the most and least stressful for subgroups of individuals with autism. The SSS can be found in Appendix C.

### **Identifying Behavioral Indicators of Anxiety**

To generate possible anxious behaviors that we could record as present or absent within each 10-s interval, I initially created a comprehensive list of behavioral indicators of anxiety (Appendix D). Parents, teachers, staff members, or other caregivers identified behaviors the child typically displays when he is anxious (i.e., behaviors which indicate to the parent/teacher that the child is anxious) from the list of behavioral descriptors in Appendix D and/or identified idiosyncratic behaviors (which were not on the list) that the child displays that indicate anxiety. The coders then recorded the occurrence or nonoccurrence of these behaviors (and any additional behaviors they noticed that were not reported by caregivers but appeared to indicate anxiety)

from videotapes during each 10-second interval. The list of behavioral descriptors in Appendix D was originally derived from a variety of sources, including the Cues for Tension & Anxiety Survey Schedule (CTASS; Cautela, 1977), the Affex Facial Coding System for Negative Facial Expressions (Izard, Dougherty, & Hembree, 1989), the Behavioral Relaxation Scale (BRS; Poppen, 1998), behavioral indicators from Lesniak-Karpiak, Mazzocco, and Ross (2003), Richards, Moss, O'Farrell, Kaur, and Oliver (2009), and Sullivan, Hooper, and Hatton (2007), as well as from clinical observations. These original scales can be found in Appendix E.

Direct observations revealed that Jon exhibited the following behaviors that indicated anxiety: crying, distress vocalizations/noises, clinging to his mother, perseverative phrases (“Here we go!”), verbal protest (e.g., “No!”), and cowering or hiding (e.g., turning into corner). Direct observations revealed that Ben exhibited the following behaviors that indicated anxiety: jumping up from his chair and running to the front door when his parents left the house, watching out the window of the front door while his parents were gone, inquiring about his parents' whereabouts when they were leaving or after they had left (e.g., “Mommy, where are you going?”, “Where is she?”), asking questions about his parents (e.g., “Do you see Mommy?”), verbalizing that he wanted or needed his parents (e.g., “I really want my Mommy!”), “Mommy, I need you!”), simply calling “Mommy!” repeatedly, and downturned mouth (frowning) and downward sloping eyebrows (in inverted V shape). Direct observations revealed that Sam exhibited the following behaviors that indicated anxiety: crying or tearfulness, plugging or covering his ears, rapid eye movements (e.g., eyes rapidly darting back and forth), a fearful facial expression (e.g., eyebrows raised and drawn together in an upside down V shape, wide open eyes with tense lower eyelids and stretched lips), and mouthing his finger.

**Frequency of anxious behavior (i.e., percentage of intervals with anxious behavior).**

The videotaped sessions were coded using a 10-s partial-interval recording procedure. The presence or absence of the anxious behaviors previously defined was recorded for each 10-s interval. Agreement occurred when both observers recorded the same anxious behavior within the same interval. Interobserver agreement was calculated by dividing the total number of agreements by the total number of agreements plus disagreements and multiplying the sum by 100. For Jon, “anxious behavior” was defined as clinging (holding onto his mother), crying (shedding tears), eyes wide open or eyes rapidly darting back and forth, cowering, freezing (a lack of movement except for respiration), frowning (turning down of the mouth), eyebrows raised and sloping down in an inverted V shape, vocalizations of fear/anxiety (e.g., whimpering, moaning, or distinct noises that he appeared to make in the back of his throat and were separate from the yelling that was categorized as problem behavior), and perseverative verbalizations that were reported to indicate anxiety (e.g., “C'mon everybody!”). For Ben, “anxious behavior” was defined as inquiring about his parents' whereabouts (e.g., “Where are you going?”), calling his mother (e.g., “Mommy!”), verbal pleading (e.g., “I really want Mommy now!”), jumping up from the chair when he realizes they are leaving, running to the front door, watching out the front door, eyebrows raised and downturned in inverted V shape, and the corners of his mouth downturned. For Sam, “anxious behavior” was defined as touching his ears (e.g., plugging his ears, covering his ears, holding his ears), eye movements (e.g., eyes rapidly darting back and forth, blinking, eyes wide open, closing eyes), crying or tearfulness, mouthing his finger, and turning his head around to look out the rear window of the car.

**Appearance of fear/anxiety.** We also obtained an overall rating of fear/anxiety, completed via videotape at the conclusion of each session, in which the child's behavior (e.g.,

visible muscle tension, heavy or rapid breathing, freezing, flushed face, trembling) was rated on a 4-point Likert-type scale, with 3 being high fear/anxiety and 0 indicating no fear/anxiety (see Appendix F). Our rating scale is based on that of Love, Matson, and West (1990), who used a 5-point Likert-type scale (with 5 being most fearful and 1 indicating no fear) as well as Koegel et al. (2004), who used a 4-point scale to rate the children as appearing *comfortable* (i.e., absence of anxiety), exhibiting *mild anxiety* (e.g., short whining or whimpering), exhibiting *High-Anxiety* (e.g., slowly moving away from the sight or sound of the stimulus), or appearing as if the auditory stimulus was *intolerable* (e.g., running away from the activity, crying or screaming). Two undergraduate research assistants (who had no direct involvement in the study and were unaware of the study design or purpose) independently rated each child's anxiety from videotaped sessions using the 4-point rating scale. Observer ratings for each session were given numeric values (no anxiety=0; mild anxiety=1; moderate anxiety=2, High-Anxiety=3). Averages of these scores were calculated across sessions to obtain a mean anxiety score for all High-Anxiety sessions and Low-Anxiety sessions for each participant. Interobserver agreement was defined as two ratings that fell within one point of each other.

### **Identifying High-Anxiety and Low-Anxiety Contexts**

Selection of anxiety-provoking contexts was based on (a) ratings of a '5' (very likely to show problem behavior) on the CAI and/or a '5' (severe stress reaction) on the SSS, (b) interview with informants (parents), and (c) direct observation (live and via videotapes). To be considered a "High-Anxiety context," the following three criteria were required to be met: (1) the given context needed to be rated as a '5' on the SSS and/or CAI, (2) the child had to exhibit anxious behavior in at least 50% of the 10-s intervals that made up the given context, and (3) the child had to be rated as either highly anxious (a '3') or moderately anxious (a '2') on a Likert-type

rating scale ranging from 0 to 3 (no anxiety to maximum anxiety). To be considered a “Low-Anxiety context,” the following three criteria were required to be met: (1) the given context needed to be rated as a '1' on the SSS and/or CAI, (2) the child could not exhibit anxious behavior in > 10% of the 10-s intervals that made up the given context, and (3) the child had to be rated as either not anxious (a '0') or mildly anxious (a '1') on the Likert-type rating scale.

**Participant 1.** On the CAI, Jon's mother rated the following contexts as a '5' (very likely to show problem behavior): a preferred activity ends or is no longer possible; activities or routines that are difficult, frustrating, disliked, or boring; activity is too long; activity is too noisy and/or crowded; having to wait; transitions between settings or activities. She rated “changes in routine, or has to deal with new and unfamiliar situations” as a '4.' On the SSS, Jon's mother rated the intensity of Jon's stress reaction as a '5' (severe) for the following situations: waiting for preferred events; waiting on line; being unable to communicate needs; waiting at a restaurant; needing to ask for help; participating in group activity; having a change in staff, teacher, or supervisor; waiting for reinforcement; feeling crowded. For fears, she rated “fear of animals” and “fear of the dark” as a '5.' During the follow-up interview, Jon's mother reported that the most anxiety-provoking context for Jon was a social event with a lot of children, such as a birthday party. She noted that he became particularly anxious and fearful upon hearing the song “happy birthday,” which she hypothesized was because the activity was noisy, crowded, overstimulating, and unfamiliar for Jon. Jon's classroom teacher also completed the SSS and rated a '5' for “having a change in environment from comfortable to uncomfortable” and “being unable to communicate needs.” She rated a '4' for several situations such as “participating in a group activity” and “having a change in staff, teacher, or supervisor.” Under “additional stressors,” his teacher added, “singing happy birthday song.” When asked what she considered to be the most

significant stressor of the ones she identified, she responded, “happy birthday and candles due to being in a classroom setting where birthdays are celebrated.”

During the postassessment interview (following CAI and SSS administration), Jon’s mother indicated that he was most likely to exhibit anxiety and problem behavior when participating in a group activity, in particular an unfamiliar or non-routine group activity that did not occur on a regular basis. Specifically, Jon's mother reported that he frequently engaged in anxious behavior (i.e., crying, yelling) when birthday candles were lit and people started singing the “happy birthday” song at home, in the classroom, or in a community setting. Jon's father, classroom teacher, and his one-to-one classroom paraprofessional confirmed this report, as did direct observations made by the first author (L.J.M.).

**Participant 2.** On the CAI, Ben's mother rated medical appointments or medical settings as a ‘5.’ She rated the following contexts as a ‘4’: frustrated because he has trouble communicating with you about what he wants or needs; denied access to what he wants. She rated the following contexts as a ‘3’ (somewhat likely to show problem behavior): activities or routines that are difficult, frustrating, disliked, or boring; activity is too long; activity is too noisy and/or crowded; she rated the following as a ‘2’ changes in routine, or has to deal with new and unfamiliar situations; transitions between settings or activities. On the SSS, Ben's mother rated the intensity of Ben's stress reaction as a ‘5’ (severe) for the following situations: waiting to talk about a desired topic; waiting for preferred events, having personal objects or materials missing; being prevented from completing a ritual; having a change in environment from comfortable to uncomfortable; transitioning from a preferred to non-preferred activity; being interrupted while engaging in a ritual; having to engage in not-liked activity; needing to ask for help; having a

change in staff, teacher, or supervisor; losing at a game. On the SSS, in terms of Fears, Ben's mother rated “fear of being left alone” as a '5' (severe).

During the postassessment interview, Ben’s mother stated that “fear of being left alone,” in particular being left at home by his parents, was Ben's greatest fear. She reported that her son was most likely to exhibit problem behavior (e.g., crying, screaming/yelling) in the context of separating from his parents, in particular when his mother and/or father left the house and left Ben alone with another adult (e.g., his uncle, his grandparents, a baybsitter). Ben's father and uncle confirmed this report, as did direct observations.

**Participant 3.** On the CAI, Sam's mother rated the following contexts as a '5' (very likely to show problem behavior): a preferred activity ends or is no longer possible; activities or routines that are difficult, frustrating, disliked, or boring; activity is too long; activity is too noisy and/or crowded; changes in routine, or has to deal with new and unfamiliar situations; transitions between settings or activities. On the SSS, Sam's mother rated a '5' (severe) for having a change in schedule or plans; having a change in task to a new task with new directions; receiving a reprimand, transitioning from a preferred to non-preferred activity; being told “no,” receiving criticism; having something marked incorrect; being unable to communicate needs; and participating in a group activity.

During the postassessment interview, Sam’s mother stated that her son was most likely to exhibit problem behavior (e.g., crying, screaming/yelling) while riding in the car, in particular when his parent turned on the blinker and made a left or right turn. She reported, “He can't always have control. People need to turn and use the blinker.” Sam's father and two home-teachers confirmed this report, stating that Sam displayed the most anxiety when riding in the



car, particularly when the driver made a left or right turn. Direct observations made by the first author (L.J.M.) confirmed this report.

## **Procedure**

Once the participants were identified, items from the Contextual Assessment Inventory (CAI) and Stress Survey Schedule (SSS) were then used to help identify anxiety-provoking contexts or situations. Items rated as a ‘5’ on the CAI (very likely to elicit problem behavior) and as a ‘5’ on the SSS (severe stress reaction) were initially used to select the “High-Anxiety” conditions. Items rated as a ‘1’ on the CAI (not likely to elicit problem behavior) and as a ‘1’ on the SSS (none to mild stress reaction) were initially used to select the “Low-Anxiety” conditions. Specific “High-Anxiety” and “Low-Anxiety” contexts were then selected, for each participant, on the basis of these SSS and CAI items, interviews with relevant stakeholders (e.g., parents and teachers), and direct observation (quantitative ratings of anxious behaviors and qualitative Likert-type ratings of anxiety). Parents were assisted in prioritizing a “High-Anxiety” context for assessment (and intervention, in Study 2) based on the context that was most likely to elicit both anxiety and problem behavior and most reduced family quality of life. This context was chosen as the focus of experimental assessment and, in Study 2, intervention.

Following administration of the CAI and SSS, interview questions based on the Functional Assessment Interview (O’Neill et al., 1997) were conducted with the parent(s) to identify, in greater detail, the specific events or situations that predicted the occurrence of both anxiety and problem behavior. For example, since Jon's mother selected “participating in group activity” as a context that was very likely to elicit a severe stress reaction on the SSS, follow-up questions assessed (a) the specific type of group activity that was most likely to trigger problem behavior (e.g., birthday parties), (b) with whom problem behavior was most likely to occur (e.g.,

parents or teachers), (c) in what setting this activity was most likely to be associated with problem behavior (e.g., home, classroom, or community setting), (d) the specific trigger or discriminative stimulus that was most likely to be associated with problem behavior (e.g., after the candles were lit and/or when the song “happy birthday” begins), (e) the parental response to the problem behavior (e.g., parent or teacher holds and comforts Jon or allows him to escape the situation by leaving the room), and (f) the child’s reaction to the parental response (e.g., Jon no longer displays problem behavior after leaving the room). For each child, following consultation with caregivers, a priority context based on the interview was identified for assessment (baseline observations) and intervention.

**Functional Analysis Probes.** During these observations, the investigator (senior author) and a second observer, who was a graduate student in clinical psychology, directly observed the contexts identified to confirm that the situations identified in the questionnaires and interviews were indeed associated with the occurrence of anxious behavior. For Jon, data were collected in his home. For Ben, data were collected in his home, his front yard, and in his backyard. For Sam, data were collected in his home and in the car.

The purpose of the Functional Analysis Probes (Carr, Magito McLaughlin, Giacobbe-Grieco, & Smith, 2003) was to probe the effect of anxiety-provoking stimuli or situations on problem behavior. During this phase, to assess the effect of the purportedly anxiety-provoking situations on problem behavior, I examined, in a reversal design, High-Anxiety versus Low-Anxiety conditions. Each child participated in a randomized ABAB design, in which B represented exposure to the stimulus or situation that was rated by parents as anxiety-provoking (e.g., singing happy birthday, car ride, parents leaving) and A represented a period that was rated as Low-Anxiety (e.g., watching television, playing with balls or a toy, playing a handheld game).

Each condition or session lasted for approximately 3 to 5 minutes (i.e., the period of time needed to carry out the situation, such as driving to a particular errand location) and occurred during the course of normal routines that parents identified (during the earlier participant selection interview) as being anxiety-provoking or non-anxiety-provoking. Each of the High-Anxiety conditions for Jon lasted from 3 minutes and 0 seconds to 3 minutes and 36 seconds (mean duration = 3 minutes and 18 seconds). Each of the High-Anxiety conditions in Ben's baseline sessions lasted from 1 minute and 33 seconds to 6 minutes and 55 seconds (mean duration = 3 minutes and 58 seconds). Each of the High-Anxiety conditions in Sam's baseline sessions lasted from 4 minutes and 37 seconds to 6 minutes and 53 seconds (mean duration = 5 minutes and 51 seconds). The order of the High-Anxiety and Low-Anxiety conditions was presented in a randomized design. Because observations were naturalistic, it was not possible to achieve exact counterbalancing of the High-Anxiety and Low-Anxiety conditions (as in Magito McLaughlin & Carr, 2005).

**High Anxiety condition.** In the High-Anxiety condition, the participant was observed during a situation previously identified as most likely to evoke high levels of anxiety by at least two informants (e.g., two parents, or a parent and teacher). The situation was chosen from the information generated from prior questionnaires and prior parent interviews in which parents reported that these stimuli or situations were highly anxiety-provoking. For example, if the context “waiting in line” was identified as stressful (i.e., rated as a ‘5’ on the SSS), the “High-Anxiety” condition might involve observing the child while waiting in line at the grocery store. For Jon, the High-Anxiety condition was being required to remain in the room with candles lit atop a birthday cake while other people are singing the “happy birthday” song; for Ben, the High-Anxiety condition was staying in his house, or in the backyard or front yard, while his

parents left the house and drove away (leaving him with the researcher); for Sam, the High-Anxiety condition was riding in the car while the driver (his mother or father) made left and right turns. During the High-Anxiety condition, data was collected on observed signs of physiological arousal (e.g., sweating, flushed face, visible muscle tension) and observable behaviors indicating anxiety (e.g., crying, avoidance, cowering, pacing, excessive talking, unusual movements, vocalizations or verbalizations) to verify the presence of anxiety. Data was also collected on problem behavior (e.g., aggression, self-injury, disruptive behavior such as screaming/yelling) and on heart rate (using a heart rate monitor). Heart rate data was collected for each of the children, though was only usable for Ben.

**Low Anxiety conditions.** In the Low-Anxiety condition, the participant was observed during a situation identified as typically eliciting low levels of anxiety or none at all. For example, if the context “playing with others” was identified as eliciting no stress, the “Low-Anxiety” condition might involve observing the child playing a game with his home-teacher or parent. In the Low-Anxiety condition, the participant was allowed to engage in a leisure activity of his choosing (e.g., watching TV, listening to music). The Low-Anxiety condition was intended to approximate those periods of the day when the participants were not engaged in any structured activity and no demands were made of them. Invariably, Jon would typically watch television, play with a toy, listen to music, or play music on his keyboard; Ben would typically play a hand-held video game, watch television, or swing on the swings in his backyard; Sam would typically play with his toys. During the Low-Anxiety condition, data were collected on observed signs of physiological arousal and observable behaviors indicating anxiety to verify the absence of anxiety. The child was observed for observable signs of anxiety, along with the

occurrence of problem behavior. Heart rate data was also collected for each of the children, though was only usable for Ben.

Session length was yoked to the duration of previously run High-Anxiety sessions so that, for every B session (High-Anxiety), there was an A session (Low-Anxiety) of the same duration. For example, if a previous High-Anxiety condition (e.g., riding in the car) lasted for 5 minutes then, the participant was likewise observed for 5 minutes in the Low-Anxiety condition (e.g., playing a videogame). This procedure was used so that session lengths were approximately equal across both High-Anxiety and Low-Anxiety sessions and, therefore, session length could be ruled out as a variable controlling problem behavior. To control for possible order effects, it was randomly determined each day, when possible, whether an A or B session would be conducted first (similar to Carr, Smith, et al., 2003). This procedure was followed to achieve approximate counterbalancing so that a High-Anxiety condition followed a Low-Anxiety condition about as often as a Low-Anxiety condition followed a High-Anxiety condition (similar to Magito McLaughlin & Carr, 2005).

All sessions were conducted within the natural context of the participant's ongoing routine. During baseline observations, parents, relatives, home-aides, or teachers were free to interact with the participant in a manner that was consistent with the natural context in which the observation occurred (e.g., if the participant was watching television, his mother might comment on the program).

Electrocardiogram (ECG) measurement was recorded using the Alive Heart and Activity Monitor (Model HM131 made by Alive Technologies) and processed using QRSTool and CMET software (Allen, Chambers, & Towers, 2007). During all sessions, participants wore the Alive monitor, a portable, wireless device with electrode transmitters that adhered to the

participant's chest and a receiver that was placed in a small pack worn by the participant. Similar monitors have been comfortably worn and viably used to measure HR in individuals with ASD and other DDs in previous studies (e.g., Boccia & Roberts, 2000; Freeman et al., 1999; Groden et al., 2005; Roberts, Boccia, Bailey, Hatton, & Skinner, 2001; Roberts, Boccia, Hatton, Skinner, & Sideris, 2006; Watson et al., 2011). To help participants adapt to the equipment and increase their comfort and compliance with the procedure, participants were introduced to the heart monitor prior to the experimental session and practiced wearing the electrodes ("stickers") on their chest as needed prior to the first assessment. As in Goodwin et al. (2006), participants were given a rationale for measuring HR that was appropriate to their developmental level (e.g., "We are interested in seeing how fast your heart beats when you are doing activities").

All sessions were recorded with a small, portable video camera (Microsoft LifeCam, a webcam measuring 2 inches in length and 1.5 inches in height), which was plugged into a small, portable laptop (MSI U110 Netbook, measuring 10.2 x 7.1 x 1.2 inches and weighing 3.2 pounds). Sessions were recorded for the purpose of collecting behavioral data, analyzing physiological measures, and collecting interobserver agreement on observed behaviors.

All baseline and intervention sessions were videotaped and coded for anxious behavior and problem behavior by the senior author and another graduate student in clinical psychology. Problem behaviors were defined as behaviors that would be considered disruptive or problematic in the home, classroom, car, or community setting. Examples included tantrum behavior and elopement. Approximately 50% of the videotaped assessment sessions were coded for interobserver agreement (IOA) by the senior author and another graduate student in clinical psychology: 60% of sessions (6 out of 10 sessions) for Ben, 63% of sessions (5 out of 8 sessions) for Jon, and 33% of sessions (2 out of 6 sessions) for Sam. Two undergraduate research

assistants, who had not received any formal training, also coded the videotapes for subjective ratings of anxiety using a Likert-type rating scale; they double-coded at least 33% of the sessions for each child for IOA.

## **Design**

The effects of anxiety on problem behavior were studied using a within-subject reversal design whereby conditions in which an anxiety-evoking stimulus was presented (“High-Anxiety”) alternated with conditions in which an anxiety-evoking stimulus was withheld (“Low-Anxiety”), similar to the alternation between the “demands” and “no demands” conditions in Carr et al. (1980).

Each session began with either a condition that was likely to evoke “Low-Anxiety” (e.g., watching television, playing with toys) or a condition that was likely to evoke “High-Anxiety” (e.g., singing happy birthday, riding in car, parents leaving the house) for each participant. The order of the conditions was presented in a randomized design, when possible. Given that observations were naturalistic (occurring within the context of participants' daily routines at home), it was not always possible to achieve exact counterbalancing of the Low-Anxiety and High-Anxiety conditions; therefore, I aimed to achieve an approximate counterbalancing so that each type of study condition followed every other type of condition about equally often (similar to Carr, Magito McLaughlin et al., 2003).

## **Response Recording**

Three dependent variables were recorded. The first dependent variable was a behavioral measure: the frequency of problem behavior (percentage of intervals containing problem behavior). Two physiological measures were also used as dependent variables: (1) mean heart rate (HR) and (2) respiratory sinus arrhythmia (RSA).

**Frequency of problem behavior (percentage of intervals with problem behavior).**

For Jon, “problem behavior” was defined as yelling/screaming (loud, vocal noises or speech produced at a very high intensity), elopement (running away; leaving the room, attempting to leave the room, or having left the room), pushing (moving or attempting to move his parent, the researcher, or another person by force), and pulling his mother's hair. For Ben, “problem behavior” was defined as whining (high-pitched complaining, begging, or acting as if crying, but without tears), yelling/screaming (loud, vocal noises or speech produced at a very high intensity), tantrum behavior (any yelling, screaming, or loud vocalization or verbalization accompanied by crying, whining, or stomping his feet on the floor), verbal protest (verbal utterances that suggested resistance to the situation, such as “No!”), and running out of the front door of his house after his parents (as well as remaining outside in the front yard rather than in the house). For Sam, “problem behavior” was defined as verbalizations (e.g., yelling or whining "No left!" or "Not on!" "This way"), laying down in the car seat while his parent was driving the car, reaching for his parent who was driving the car, tantrum behavior (defined as yelling, screaming, or any loud vocalization accompanied by crying), and noncompliance (refusal to comply with instructions or directives, such as his mother’s request to get out of the car in the parking lot).

The videotaped sessions were coded using a 10-s partial-interval recording procedure. The occurrence or nonoccurrence of the problem behaviors previously defined was recorded for each 10-s interval. Agreement occurred for each interval in which both observers recorded the presence or absence of the same problem behavior(s). Interobserver agreement was calculated by dividing the total number of agreements by the total number of agreements plus disagreements and multiplying the sum by 100.



The two heart activity variables in this study were (a) mean heart rate (HR); and (b) respiratory sinus arrhythmia (RSA).

**Mean heart rate (HR).** Mean HR was derived from the inter-beat-interval (IBI), a measure of heart activity that represents the time between successive peak R waves. I chose to analyze mean HR in the present study, given that previous research has examined mean HR in children with ASD (Goodwin et al., 2006; Groden et al., 2005) and adults with ASD (Barrera et al., 2007) and ID (Freeman et al., 1999). Continuous records of IBI were recorded throughout the experimental sessions. The IBIs of the child's heartbeat were detected by the electrodes and transmitted to a receiver. Mean IBI was computed during the High-Anxiety and Low-Anxiety conditions. Raw digitized ECG signals were analyzed off-line. An IBI series was generated to a file using a peak detection algorithm, after which the series was screened by hand and corrected for artifacts (missed or erroneous beats) using QRSTool software (Allen et al., 2007). IBI was then extracted using CMetX software (Allen et al., 2007), which produces several indices of heart activity, one of which is mean HR. Thus, mean HR was calculated for each recording period by transforming each IBI to HR and averaging across all values. Each of the metrics produced by CMetX (i.e., meanHR, meanIBI, logRSA) are derived from a single electrocardiographic channel.

**Respiratory Sinus Arrhythmia (RSA).** Log-transformed total cardiac variance was extracted as an index of HRV. Specifically RSA reflects changes in beat-to-beat intervals of the heart, and was used as the measure of parasympathetic activity. The validity of using RSA as an index of parasympathetic activity has been demonstrated in previous research (e.g., Berntson et al., 1997), including children with ASD (Vaughan Van Hecke et al., 2009; Watson et al., 2011). In this study, RSA was quantified during each sequential 30-second epoch, and was computed

from participants' ECG data using the software and procedure described by Allen et al. As previously mentioned, IBI series were first derived from raw ECG, using an R-spike detection algorithm, followed by hand screening with correction for artifacts. Heart rate variability in the high frequency band (.12-1.0 Hz) was extracted using CMetX software (Allen et al., 2007), which produces an estimate of RSA as the natural log of the variance of the filtered waveform. Although .12 to .4 Hz is the default frequency band in CMetX (for adults), I customized the frequency so that it corresponded to the frequency bands associated with spontaneous breathing, which, given the age range of the participants in this study, was 0.12–1.0 Hz. (I based this on a study by Vaughan Van Hecke et al., who used a frequency band of 0.12 to 1.0 Hz in their study of 8-to-12-year-old children with ASD).

For any condition, if the amount of heart activity data requiring editing exceeded 20%, data were excluded from analyses (as in Watson et al., 2011). Some data were missing due to technical problems during physiological data collection and excessive artifacts; much of the data was not useable or required > 20% editing for two children, Jon and Sam.

### **Interobserver Agreement (IOA)**

Approximately 50% of sessions across the three participants were coded by this author and another observer (a graduate student in clinical psychology) for IOA on percentage of intervals containing anxious behavior and problem behavior; 5 out of 8 sessions were coded for Jon, 6 out of 10 sessions were coded for Ben, and 2 out of 6 sessions were coded for Sam. IOA for anxious behavior was scored for each 10-s interval in which the two researchers agreed on the presence or absence of anxious behavior during each interval. IOA for problem behavior was scored for each 10-s interval in which the two researchers agreed on the presence or absence of problem behavior during each interval. A binary reliability index (i.e., the observers scored either

perfect agreement or no agreement) was used for occurrence/nonoccurrence of problem behavior or anxious behavior during the given 10-s interval. Exact IOA was compared on an interval-by-interval basis; if anxious behavior or problem behavior did occur, both observers had to agree on the type of anxious behavior or problem behavior observed within the given 10-s interval. Mean IOA for percent of intervals containing anxious behavior was above 80%. Mean IOA for percent of intervals containing problem behavior was also above 80%. For appearance of anxiety using the Likert-type rating scale, IOA was defined as two ratings that fell within one point of each other. Mean IOA for appearance of anxiety was 100% across the three participants. IOA is generally considered acceptable if average agreement between two observers is at least 80% (Page & Iwata, 1986) across at least 20% of sessions (Cooper, Heron, & Heward, 1987).

## **Results**

### **Behavioral Measures**

Recall that, to be considered a “High-Anxiety context,” the following three criteria were required to be met: (1) the given context needed to be rated as a '5' on the SSS and/or CAI, (2) the child had to exhibit anxious behavior in at least 50% of the 10-s intervals that made up the given context, and (3) the child had to be rated as either highly anxious (a '3') or moderately anxious (a '2') on a Likert-type rating scale ranging from 0 (no anxiety) to 3 (maximum anxiety). These criteria were met for the “happy birthday” context for Jon, for the “separation” context for Ben, and for the “car ride” (left/right turn) context for Sam.

**Defining anxiety: anxious behaviors.** For all three participants, anxious behavior occurred in over 50% of intervals for the High-Anxiety context, averaged over sessions (see Figure 1). For Ben, anxious behavior occurred in an average of 83% of High-Anxiety sessions (SD = 22.6%, range = 50% to 100%) versus 1% of Low-Anxiety sessions (SD = 2.1%, range =

0% to 5%). For Jon, anxious behavior occurred in an average of 53% of High-Anxiety sessions ( $SD = 8\%$ , range = 43% to 60%) versus 5% of Low-Anxiety sessions ( $SD = 9\%$ , range = 0% to 18%). For Sam, anxious behavior occurred in an average of 80% of High-Anxiety sessions ( $SD = 16\%$ , range = 64% to 96%) versus 0% of Low-Anxiety sessions.

**Defining anxiety: subjective ratings.** Using the Likert-type rating scale, on a scale of 0 (no anxiety) to 3 (high anxiety), Sam's anxiety was rated an average of 2.8 ( $SD = 0.29$ , range = 2.5 to 3) for each of the car rides ("High-Anxiety" contexts) and an average of 0 for each of the playing game and worksheet ("Low-Anxiety") contexts. Jon's appearance of anxiety was rated an average of 2.75 ( $SD = 0.5$ , range = 2-3) for each of the happy birthday ("High-Anxiety") contexts and a 0 for each of the watching TV/playing with a toy ("Low-Anxiety") contexts. Ben's appearance of anxiety was rated an average of 3 ( $SD = 0$ , range = 3-3) for each of the separation ("High-Anxiety") contexts and a 0.4 ( $SD = 0.55$ , range = 0 to 1) for each of the playing game ("Low-Anxiety") contexts. Ratings are shown in Figure 2.

**Functional analysis results.** Figure 3 shows the results of the functional analysis of the relationship between anxiety and problem behavior. Specifically, Figure 3 shows the percentage of intervals in which problem behavior occurred during each session for the three children, demonstrating that problem behaviors were high in the High-Anxiety condition and near zero in the Low-Anxiety condition. This is indicated by the large percentage of 10-s intervals in which problem behavior occurred when presented with the High-Anxiety condition and the low percentage of 10-s intervals in which problem behavior occurred when presented with the Low-Anxiety condition.

For Ben, problem behavior occurred in an average of 80% of the five High-Anxiety sessions ( $SD = 26.2\%$ , range = 36% to 100%) versus 0% of the five Low-Anxiety sessions. For

Jon, problem behavior occurred in an average of 83% of the four High-Anxiety sessions ( $SD = 8.8\%$ , range = 74% to 93%) versus 1% of the four Low-Anxiety sessions ( $SD = 2.3\%$ , range = 0% to 5%). For Sam, problem behavior occurred in an average of 48% of the three High-Anxiety sessions ( $SD = 22\%$ , range = 30% to 72%) versus 1% of the three Low-Anxiety sessions ( $SD = 1\%$ , range = 0% to 2%).

**Interobserver agreement (IOA).** Of the sessions that were coded for IOA by the senior author and another graduate student in clinical psychology, the two observers scored an average of 93.82% agreement for Sam's problem behaviors ( $SD = 1.9\%$ , range = 92% to 94%) and 84.36% agreement for Sam's anxious behaviors ( $SD = 3.4\%$ , range = 82% to 87%). The two observers scored an average of 96.16% agreement for Ben's problem behaviors ( $SD = 4.41\%$ , range = 91% to 100%) and 90.02% agreement for Ben's anxious behaviors ( $SD = 8.68\%$ , range = 78.95% to 100%). Finally, the two observers scored an average of 90.69% agreement on Jon's problem behaviors ( $SD = 7.75\%$ , range = 80% to 100%) and 87.09% agreement on Jon's anxious behaviors ( $SD = 7.83\%$ , range = 80% to 100%).

### **Physiological Measures**

Visual analysis of the data reveals that Jon and Ben had higher mean heart rate and lower parasympathetic estimates (Figures 4-7), in the High-Anxiety conditions (happy birthday, parents leaving the house) than in the Low-Anxiety conditions (e.g., watching TV, playing a handheld game). Repeated measures (paired)  $t$ -tests also revealed that these differences between the High-Anxiety and Low-Anxiety conditions were significant.

**Heart rate.** For Jon, across thirteen 30-second epochs (four 30-s epochs from Session #1, three 30-s epochs from Session #2, three 30-s epochs from Session #3, and three 30-s epochs from Session #4), a paired  $t$ -test indicated that the mean HR during the High Anxiety condition

( $M=127.83$ ,  $SD=15.40$ , range=109.11-162.19) was significantly higher than during the Low Anxiety condition ( $M=97.39$ ,  $SD=11.17$ , range=88.11-119.84),  $t(12) = 6.5302$ ,  $p < 0.0001$ , which reflects higher levels of heart activity in the High Anxiety condition. There were not sufficient analyzable 30-s intervals in each session to conduct a t-test for each individual session.

For Ben, across twenty-six 30-second epochs (six 30-s epochs from Session #1, seven 30-s epochs from Session #2, ten 30-s epochs from Session #3, and three 30-s epochs from Session #4), a paired  $t$ -test indicated that the mean HR during the High Anxiety condition ( $M=131.875$ ,  $SD=23.21$ , range=88.9 to 171.82) was significantly higher than during the Low Anxiety condition ( $M=90.12$ ,  $SD=5.98$ , range=80.71 to 112.88),  $t(25) = 8.0622$ ,  $p < 0.0001$ , which reflects higher levels of heart activity in the High Anxiety condition. Examining each session individually, for Session #1 for Ben, a paired  $t$  test indicated that mean HR during the High Anxiety condition (141.22 BPM) was significantly higher than during the Low Anxiety condition (84.39 BPM),  $t(5) = -11.12$ ,  $p < .001$ . Similarly, for Session #2 for Ben, a paired  $t$  test indicated that mean HR during the High Anxiety condition (127.04 BPM) was significantly higher than during the Low Anxiety condition (90.63 BPM),  $t(6) = -3.7449$ ,  $p < .01$  ( $p = 0.0047834506$ ). For Session #3 for Ben, a paired  $t$  test indicated that the mean HR during the High Anxiety condition (131.61 BMP) was significantly higher than during the Low Anxiety condition (93.08 BMP),  $t(9) = -3.61$ ,  $p < 0.01$ . There were only three 30-s intervals in Session #4 for Ben, which was not sufficient to conduct a t-test.

For Sam, in the two sessions that yielded usable data, a dependent  $t$  test failed to show a significant difference between mean HR during the High Anxiety and Low-Anxiety condition.

**RSA.** For Jon, across thirteen 30-second epochs, a paired  $t$ -test indicated that the mean RSA during the High Anxiety condition ( $M=6.365$ ,  $SD=1.812$ , range=109.11-162.19) was not

significantly lower than during the Low Anxiety condition ( $M=7.512$ ,  $SD=0.539$ , range=88.11-119.84),  $t(12) = 2.0799$ ,  $p = 0.0596$ . There were not sufficient analyzable 30-s intervals in each session to conduct a t-test for each individual session.

For Ben, across twenty-six 30-second epochs, a paired  $t$ -test indicated that the mean RSA during the High Anxiety condition ( $M=4.67$   $SD=1.82$ , range=1.31 to 6.94) was significantly lower than during the Low Anxiety condition ( $M=7.51$ ,  $SD=0.70$ , range=6.07 to 8.69),  $t(25) = 6.9782$ ,  $p < 0.0001$ . Examining each session individually, for Session #1 for Ben, a dependent  $t$  test indicated that mean RSA during the High Anxiety condition (4.25) was significantly lower than during the Low Anxiety condition (7.43),  $t(5) = 4.75$ ,  $p < .001$  ( $p = 0.0025$ ). Similarly, for Session #2 for Ben, a dependent  $t$  test indicated that mean RSA during the High Anxiety condition (5.407) was significantly lower than during the Low Anxiety condition (7.167),  $t(6) = 2.507$ ,  $p < 0.05$  ( $p = 0.023$ ). For Session #3 for Ben, a paired  $t$  test indicated that the mean RSA during the High Anxiety condition (4.23) was significantly lower than during the Low Anxiety condition (7.58),  $t(9) = 4.067$ ,  $p < 0.01$ . There were only three 30-s intervals in Session #4 for Ben, which was not sufficient to conduct a t-test.

For Sam, in the two sessions that yielded usable data for both conditions, a dependent  $t$  test failed to show a significant difference between mean RSA during the High Anxiety and Low-Anxiety condition.

## **STUDY 2: INTERVENTION**

The purpose of Study 2 was to examine whether a multicomponent behavioral intervention incorporating principles and strategies from the anxiety literature for TD children with principles and strategies from the PBS literature for children with ASD/DD would reduce anxiety and problem behavior in three children with ASD and co-occurring ID. Although there is

extensive research supporting the effectiveness of CBT in reducing anxiety in TD children (Velting et al., 2004), there is comparatively little research on psychosocial interventions for the treatment of anxiety in children with ASD, particularly those with both ASD and ID. Indeed, randomized clinical trials (RCTs) examining the effectiveness of CBT generally exclude children with ASD (e.g., Kendall, 1994). Although probably efficacious cognitive behavioral interventions have been developed for TD children with anxiety disorders (e.g., Kendall, 1994), the communicative, social, and cognitive characteristics inherent in ASD may make these standard treatments less effective for children with ASD (Wood et al., 2009). In spite of this, only a handful of studies have examined the efficacy of modified CBT (i.e., increased use of visual supports, increased parental involvement) to treat anxiety in children with ASD.

Specifically, several case studies (e.g., Lehmkuhl, Storch, Bodfish, & Geffken, 2008; Reaven & Hepburn, 2003; Sze & Wood, 2007, 2008), exploratory clinical trials (e.g., Chalfant et al., 2007; Reaven et al., 2009; Sofronoff et al., 2005), and three recent RCTs (Reaven, Blakeley-Smith, Culhane-Shelburne, & Hepburn, 2012; Sung et al., 2011; Wood et al., 2009) have suggested that CBT, with ASD-relevant modifications, can reduce symptoms of anxiety in children with ASD. In fact, rates of positive treatment response in these studies have been over 50%, which is comparable to the treatment response for TD children with anxiety disorders (e.g., Kendall, 1994; Walkup et al., 2008).

However, it is important to note that these recent RCTs examining the effectiveness of CBT in children with ASD excluded children with a below-average IQ. Specifically, in Reaven et al. (2012), the children with high-functioning autism spectrum disorder (HFA) had a verbal IQ above 70, in Sung et al. (2011), the children with ASD were required to have a Verbal Comprehension of  $\geq 80$  and Perceptual Reasoning skills of  $\geq 90$  and, in Wood et al. (2009),



children with ASD were excluded if they had a verbal IQ less than 70. Similarly, children with ASD and impaired cognitive functioning were excluded in the exploratory clinical trials; in Chalfant et al. (2007), children who had intellectual delay were not included, in Reaven et al. (2009), the children with HFA were required to have an overall IQ above 70 and be verbally fluent and, in Sofronoff et al. (2005), the children with AS all had an IQ above 80. Likewise, in a pilot study by White, Ollendick, Scahill, Oswald, and Albano (2009) examining the preliminary efficacy of a manual-based intervention targeting anxiety and social competence in four adolescents with HFA, the participants had an  $IQ \geq 70$ . Even in the case studies using CBT, participants were of average or above-average intellectual functioning; in Lehmkuhl et al. (2008), the participant with HFA had an IQ in the average range (92) and, in Reaven and Hepburn (2003), the participant with AS had an IQ in the gifted range (135-145). Further, Moree and Davis (2010) noted that all of the studies they reviewed that used modified CBT for anxious children with ASD have been conducted with children who have either Asperger syndrome (AS) or HFA. They further noted that it has not been examined whether CBT with the ASD-relevant modifications they described (i.e., disorder-specific hierarchies, use of more concrete, visual tactics, incorporation of child-specific interests, and incorporation of parents) could be used with more severe cases of ASD or in those children with ASD who have an ID.

When children have a cognitive deficit or are impaired in their ability to understand and/or express language, the cognitive components of CBT are often downplayed, de-emphasized, modified, adapted to the child's level, or excluded altogether so that the intervention is "behavioral" rather than "cognitive-behavioral." Behavioral treatments for children who are nonverbal, minimally verbal, and/or cognitively impaired typically include systematic desensitization (also referred to as "contact desensitization" or "in vivo desensitization"),

reinforced practice, and modeling approaches. With *systematic desensitization*, approach/acceptance responses are positively reinforced while fearful/avoidant responses are extinguished as the individual is gradually exposed to increasing proximity, intensity, or amounts of the feared stimulus or situation. *Contact desensitization* refers to exposing an individual to the phobic (avoided) stimulus by gradually shaping approach responses. *Reinforced practice* refers to positively reinforcing an individual's repeated tolerance of fear-provoking stimuli or situations. Although these labels differ, the actual procedures are essentially the same in function and practice (Luiselli, 2011) in that they all involve graduated exposure. *Modeling* involves the individual observing another person engaging with or approaching the feared stimulus appropriately (i.e., with calm or non-anxious behavior). Several behavioral procedures, such as systematic desensitization (Wolpe, 1990), contact desensitization (Ricciardi, Luiselli, & Camare, 2006), covert reinforcement (Cautela, 1970), participant modeling (Matson, 1981), and relaxation training (Cautela & Groden, 1978) have been used to reduce behaviors considered to indicate anxiety in individuals with DD. These procedures have been shown to reduce verbal statements indicating fear or anxiety and overt behavioral avoidance of anxiety-provoking stimuli (Groden et al., 1994) in individuals with DD.

### **Behavioral Treatment of Avoidance Behavior in Children with ID**

Few controlled studies describe effective treatment of fear or anxiety in individuals who have DDs (cf. Erfanian & Miltenberger, 1990; Rapp et al., 2005; Ricciardi et al., 2006), and there are even fewer controlled studies documenting the effective treatment of fear/anxiety in children with co-occurring ASD and ID. Jennett and Hagopian (2008) reviewed the literature regarding behavioral treatment of “phobic avoidance” in individuals with ID, some of whom were children with autism. The authors identified seven main treatment components in the

studies they reviewed: (1) in-vivo exposure, (2) hierarchy (also referred to as shaping or stimulus fading), (3) contingent reinforcement, (4) prompting (verbal or physical), (5) modeling, (6) extinction/blocking, and (7) use of distracting stimuli. In vivo exposure and reinforcement were used in all of the single-case studies. Twelve of the 13 single-case experimental design studies they reviewed demonstrated treatment efficacy through the use of good experimental design and two of the four group designs they reviewed reported significant differences between treatment and control groups. The authors concluded from their results that behavioral treatment can be considered a well-established treatment for “phobic avoidance” in individuals with ID. They noted, however, that a limitation of their review was that the description of clinical problems they called “phobic avoidance” differed between studies (and few used DSM diagnoses), most likely because of the difficulty in assessing and differentially diagnosing anxiety disorders within this population. There are few instruments for assessing anxiety in adults with ID (and none designed for assessing anxiety in children with ASD and/or ID), particularly for those individuals with limited verbal abilities, and there is a lack of research on whether the standard instruments for assessing anxiety in TD populations are valid for assessing anxiety within this population (Jennett & Hagopian, 2008). Of note, of the 13 single-case studies and 4 group studies of individuals with ID that they reviewed, only four single-case studies examined children with autism and ID (Love et al., 1990; Luscre & Center, 1996; Rapp et al., 2005; Ricciardi et al., 2006).

### **Behavioral Treatment of Avoidance Behavior in Children with ASD and ID**

Only a handful of case studies (Jackson & King, 1982; Luiselli, 1978) and single-subject experimental designs (Ellis, Ala'i-Rosales, Glenn, Rosales-Ruiz, & Greenspoon, 2006; Koegel et al., 2004; Love et al., 1990; Luscre & Center, 1996; Rapp et al., 2005; Ricciardi et al., 2006)

were identified that have used behavioral interventions to treat specific fears, “fearful responses,” or “avoidance behavior” in children with autism who also had ID. (As previously noted, fear or anxiety may be more difficult to ascertain in children with below-average cognitive abilities, and can often only be identified when overt “avoidance behavior” is displayed.) Specifically, these studies used behavioral procedures such as graduated exposure, modeling, and positive reinforcement (e.g., contingent social attention) (Ellis et al., 2006), systematic desensitization (Koegel et al., 2004), graduated exposure, participant modeling, and positive reinforcement contingent upon approach (Love et al., 1990), gradual exposure with reinforcement and stimulus fading (Luiselli, 1978), desensitization with guided mastery, video peer modeling, and reinforcement (Luscre & Center, 1996), blocking plus reinforcement for pool approach and occupancy (Rapp et al., 2005), and contact desensitization (shaping or reinforcing approach responses) (Ricciardi et al., 2006). As noted by Rapp et al., however, few studies on anxious/avoidant or “phobic” behavior in individuals with ASD or DD have utilized direct measurement of the target behavior and systematic replication of behavior change, with most studies using either indirect or ordinal measures of avoidance behavior (e.g., steps completed in a hierarchy, such as in Luscre & Center, 1996) and attempting to demonstrate functional control of the intervention on “phobic” behavior with inadequate designs (e.g., A-B designs; Luiselli, 1977). Conversely, only a handful of studies have directly and repeatedly measured overt escape/avoidance behavior when presented with the aversive S<sup>D</sup> (Rapp et al., 2005).

The majority of the aforementioned studies have focused more on consequence-based intervention strategies (e.g., positive reinforcement contingent upon approach) than antecedent-based strategies (e.g., providing choice). However, a recent study by Cale, Carr, Blakeley-Smith, and Owen-DeSchryver (2009) demonstrated that problem contexts, one such context involving

“the presence of a feared stimulus,” produced problem behavior and that, by altering the problem context, they reduced or eliminate the problem behavior within those contexts. Specifically, in the study by Cale et al. (2009), parents of three children with autism nominated “the presence of a feared stimulus in school” as the context most likely to be associated with problem behavior, which was corroborated in a postassessment interview with the children’s teachers. The authors noted that, although fear can be difficult to assess in young children with autism, the teachers of the three children noted that each child, when confronted with the identified “feared stimulus,” reliably exhibited one or more nonverbal behaviors consistent with fear: startle response, gasping, hyperventilation, trembling hands, shielding face with hands, closing eyes and turning away from the stimulus, and wincing. One of the children (FSIQ=108) was reported by her teacher to display problem behavior (e.g., screaming, crying) during group reading activities in which the other children read stories that contained specific onomatopoeic sounds (e.g., sneezing: “a choo,” and “huff and puff”). The second child (FSIQ=77) exhibited problem behavior (e.g., screaming, pushing) when presented with math activities that contained materials with sea creatures. The third child (FSIQ=104) displayed problem behavior (e.g., screaming, dropping to the floor) when informed that the class would watch the “Arthur” video collection. Intervention consisted of presenting the child with a choice between the feared stimulus and an alternative stimulus (or alternative stimuli) that were matched with the feared stimulus relative to generic content category (i.e., choice between a book without onomatopoeic sounds and a book with them, choice between math worksheets without sea creatures versus ones with them, choice between Rugrats video and Arthur video). Following intervention, the problem behavior of all three children substantially improved (dependent variables were percentage of tasks steps completed, latency to session termination, and number of sessions terminated). It was proposed

that allowing the children to choose among academic stimuli gave them the opportunity to avoid the feared stimulus without compromising the instructional goal of the task at hand. Although the results of the study by Cale et al. are promising, it may not always be possible to avoid anxiety-provoking stimuli or situations; at times, it may be beneficial or even necessary to mitigate the anxiety-provoking context or teach the individual to cope with the anxiety-provoking context (see “avoid-mitigate-cope strategy” in Carr, Ladd, & Schulte, 2008). In addition, although the teachers had reported nonverbal behaviors consistent with fear, these behaviors were not measured in the study.

A close examination of this small body of research suggests a number of areas in which the systematic application of behavioral intervention to assess and treat fear and anxiety in children with ASD and ID could be improved. First, some studies used either indirect or ordinal measures of avoidance behavior (e.g., steps completed in a hierarchy, such as in Luscre & Center, 1996, or percentage of task steps completed, as in Cale et al., 2009) or were case studies using potentially insufficient designs (Jackson & King, 1982; Luiselli, 1978). Second, some studies only included a single child with autism (Rapp et al., 2005) or two children with autism (Love et al., 1990), whereas a multiple baseline lag across 3 or 4 children is generally preferable. Third, in some studies, specific fear responses were not recorded (e.g., Luscre & Center, 1996; Ricciardi et al., 2006). Fourth, only two studies used a Likert-type rating scale to judge participants' level of fear or anxiety (Koegel et al., 2004 used a 4-point rating scale to assess “mean level of anxiety per session”; Love et al., 1990 used a 5-point rating scale to assess “overall rating of fear” at the end of each learning trial). Fifth, some investigators did not video-record or audio-record their baseline or intervention sessions (Cale et al., 2009; Luscre & Center, 1996; Ricciardi et al., 2006). Sixth, none of the aforementioned studies used a multimethod

assessment of fear or anxiety, relying only on overt avoidance behaviors or other observable behaviors. Finally, no study to date has integrated evidence-based treatments for childhood anxiety disorders in TD children (Albano & Kendall, 2002) with the best-practice features of Positive Behavior Support (Carr et al., 2002) to focus on assessing and treating anxiety and/or problem behavior related to anxiety in children with ASD and ID.

### **Positive Behavior Support**

Core features of Positive Behavior Support (PBS) include ecological validity (how applicable science is to real-life settings), stakeholder participation (e.g., collaborating with family members, teachers, and other relevant stakeholders), designing interventions based on the results of functional assessments, a focus on quality of life, and multicomponent intervention plans that emphasize prevention and teaching new skills (Carr et al., 2002; Lucyshyn et al., 2007). Although the aforementioned studies targeting fearful/avoidant responses in children with ASD and ID used behavioral procedures such as graduated exposure/systematic desensitization, modeling, and reinforcement, most of these studies did not incorporate features of PBS. In particular, with the exception of using gradual exposure (which could be conceptualized as an antecedent strategy that reduces the aversiveness of the anxiety-provoking stimulus), most of these studies did not use PBS antecedent-based interventions such as increasing predictability (e.g., visual schedules, priming), choice-making, noncontingent presentation of positive reinforcers, presenting discriminative stimuli for nonproblem behavior, and incorporating preferences or interests into disliked or difficult activities. Rather than imposing a consequence following the occurrence of problem behavior (reactive), antecedent-based interventions are preventative (proactive) in that they aim to reduce the likelihood of the problem behavior initially occurring by eliminating or modifying the  $S^D$ s that trigger the problem behavior or

altering the setting event(s) that make the problem behavior more likely to occur. Further, in previous studies, intervention was not informed by a functional assessment. This is a significant point, as a meta-analysis demonstrated that interventions based on assessment of the function of problem behavior (i.e., assessment of the factors controlling problem behavior) are about twice as likely to succeed as those that are not (Carr et al., 1999). Therefore, using functional assessment to inform intervention has become a best practice in the field of ASD and DD.

### **The Present Study**

Wood et al. (2009) noted that the structured, linear format of group therapy (and manualized CBT) may limit the ability to match intervention techniques to the individual characteristics of a child with ASD and that, given the heterogeneity of phenotypes in ASD (and, most likely, the related heterogeneity in underlying pathology), individualized interventions tailored to a child's specific characteristics may be particularly effective. Further, I propose that, given the heterogeneity of setting events and antecedents of fearful/anxious behavior and the different functions of such behavior, as well as given the idiosyncratic fears that children with ASD possess and the idiosyncratic ways in which they express their fear and anxiety, individualized treatments tailored to each specific child would be that much more powerful. In the present study, I designed individualized interventions tailored to participants and their families based on the function(s) of the child's problem behavior (i.e., to escape/avoid an anxiety-provoking stimulus or situation, to obtain comfort/reassurance), the specific antecedents that triggered the problem behavior and the setting events that increase the likelihood of the problem behavior (e.g., unpredictability), the idiosyncratic fears and preferences/interests of each child, and the needs, resources, abilities, goals, values, and priorities of the child's family. The purpose of Study 2 was to provide participants and their parents with strategies to use that would



alter the anxiety-provoking context or help the participants cope with their anxiety, thereby making the situation less anxiety-provoking.

## **Method**

### **Procedure**

The participants, High-Anxiety conditions, and measures were the same in Study 2 as in Study 1. The study began with a baseline (i.e., preintervention) phase, in which all procedures and measures were identical to those previously described in the assessment study for the High-Anxiety condition (the only condition relevant to intervention). During baseline observations, the investigator directly observed the target context to confirm that the identified stimulus or situation was indeed associated with the occurrence of anxious behavior. Baseline was followed by a series of intervention sessions carried out over a period of 4 weeks across participants. The baseline observations (along with the interview information from Study 1) produced assessment information that was used to form a hypothesis about the functions of the problem behavior (e.g., to escape anxiety-provoking stimulus or situation, to obtain comfort or reassurance from parent) and develop a relevant multicomponent intervention plan.

After the baseline observations and prior to developing the intervention plan, I formulated hypotheses about the functions of the children's problem behavior and any relevant antecedents and setting events. To initiate the hypothesis generation process, I also asked the child's parents (and, in the case of Jon, his classroom teacher and, in the case of Sam, his home-teachers) why they thought the particular child displayed problem behavior (as in Carr & Carlson, 1993). For each child, the relevant stakeholders (parents and teachers) consistently hypothesized that the child's problem behavior (in the targeted experimental context) served the function of avoiding or escaping from an anxiety-provoking stimulus or situation. During

baseline, I conducted an additional descriptive observational assessment as an aid to subsequent treatment planning (as in Carr & Carlson, 1993). The purpose of this informal functional assessment was to collect information that could be used to generate plausible hypotheses concerning the variables that maintained each child's problem behavior (as in Carr & Carlson, 1993). Based on our direct observations, it appeared that the primary function of the children's problem behavior was to avoid or escape an anxiety-provoking situation by leaving the room or attempting to leave the room for Jon, attempting to prevent his parents from leaving for Ben, and attempting to cause his parents to drive straight instead of make a left/right turn for Sam. I also used the assessment information to develop an intervention plan to mitigate the impact of the anxiety-provoking stimulus or situation.

After the final baseline observation, the researcher met with the parents to discuss the results of the assessment and the researcher's suggestions for intervention strategies. This meeting was a collaborative negotiation process in which the needs and concerns of the parents were given equal consideration to those of the researcher (Lucyshyn et al., 2007). A treatment strategy was proposed because it addressed a function of the child's problem behavior by focusing on a discriminative stimulus ( $S^D$ ) for the child's problem behavior, altering a relevant setting event, providing a response alternative to problem behavior, or changing a consequence (i.e., removing reinforcement for problem behavior, providing reinforcement for alternative behavior). Several treatment options were discussed and offered to each child's parents, who selected the option that they felt represented the best fit for their family (Lucyshyn et al., 2007).

Prior to implementing treatment procedures, participants' parents received training in which the rationale for each procedure was explained and the procedure itself was modeled. Parents were then required to demonstrate use of the procedure on the author, after which they

received corrective or supportive feedback as appropriate (as in Carr & Carlson, 1993). During the first intervention session, the author prompted the parent on what to do when the anxiety-provoking stimulus was presented or the anxiety-provoking situation occurred. In subsequent sessions, the author provided prompts only if the parent failed to implement the required treatment within 5 s of the onset of anxiety-provoking stimulus or situation or, if a problem behavior arose, within 5 s of the onset of problem behavior (as in Carr & Carlson, 1993). Next, I describe each of the procedures that constituted the multicomponent intervention package.

**Psychoeducation.** Psychoeducation is one of the central components in CBT for childhood anxiety disorders in TD populations. Participants' parents were provided with information about the nature of anxiety, the ways in which anxiety is learned and maintained, and the rationale for various treatment techniques (Velting et al., 2004). Social Stories (for Ben and Sam) and video priming (for Jon) were also used as a way to provide psychoeducation to the children in a visual format that could more easily be comprehended. For example, Ben's Social Story (a) normalized anxiety (i.e., “Everyone feels worried or afraid or anxious sometimes – kids and grownups too”); (b) explained that anxiety has a function or purpose (i.e., “If a lion is chasing you, it is okay to feel afraid, because your fear will make you run from the lion”); (c) described the specific nature of Ben's anxiety (i.e., “I am very, very afraid when Mommy or Daddy leave the house and go out without me. I am afraid because I really want to be with them.”); (d) explained the concept of habituation (i.e., “At first, when Mommy and Daddy leave, I will feel scared. Then, after a while longer, I will feel less scared. Then, after a while longer, I won't be scared anymore. I will see that my anxiety goes down after a while, even when Mommy and Daddy are not home”); and (e) provided Ben with replacement behaviors that he could do instead of crying, screaming, and trying to run after them (i.e., coping self-statements such as

“This is just my anxiety talking” or “Mommy and Daddy always come home, so I won't be afraid,” relaxation techniques such as deep breathing, and activities such as playing, watching a movie, and playing video games). The complete text of the story can be found in Appendix G.

**Increasing predictability (visual schedule, social story, video modeling, priming).**

Unpredictability was judged to be a setting event for anxiety and problem behavior in all three participants. Research has demonstrated that increasing predictability reduces problem behavior in individuals with ASD. For example, Flannery and Horner (1994) manipulated the predictability of both familiar and unfamiliar events for two students with moderate-to-severe intellectual disabilities and autism; in the predictable condition, they described and modeled all the steps involved in an upcoming task (either familiar or unfamiliar). They found that problem behavior was lower in predictable conditions and conversely was higher when the upcoming tasks were un signaled (i.e., unpredictable). Although Flannery and Horner (1994) did not conceptualize the reduction in problem behavior as being due to a reduction in “anxiety” per se (they hypothesized that signals providing information about the environment could serve as an establishing operation for appropriate behavior by making the event more predictable), studies suggest that predictability is a moderator of anxiety in that the ability to predict aversive events attenuates anxious responses (Grillon, 2008). For example, using the startle reflex as a measure of aversive states, Grillon et al. (2004) found greater anxiety during anticipation of unpredictable shocks compared with predictable shocks.

A visual schedule is one way to reduce the unpredictability associated with transitions by informing children about the upcoming sequence of events. Research suggests that the use of visual schedules and cues to signal the sequence of upcoming activities reduces problem behavior during transitions (Krantz, MacDuff, & McClannahan, 1993; Mesibov, Browder, &

Kirkland, 2002). Therefore, to ensure the predictability of Sam's transition, pictures were created to represent the most common locations to which Sam's parents drove, and those pictures were used to construct a visual schedule. Before entering the car, Sam was presented with a portable board that contained pictures and words representing the locations he would be traveling to in the community as well as anchor pictures of his home on each end of the schedule.

Social stories (Gray & Garand, 1993) are another way to increase predictability. Social Stories are individualized narratives that visually depict the sequence of events involved in a routine or situation and describe appropriate behavior relevant to the situation, thereby decreasing unpredictability and providing a model for socially acceptable behavior. These stories are intended to adopt the perspective of the child for whom the story is written. Social stories establish increased predictability regarding upcoming events and, in so doing, have been shown to generate decreased levels of problem behavior (Ozdemir, 2008; Sansosti, Powell-Smith, & Kincaid, 2004). Social Stories can be used for a multitude of purposes such as explaining new situations or changes in routine, describing situations in a way that is non-intimidating, teaching adaptive skills, and dealing with challenging behaviors, including emotional expression, aggression, or obsessive behavior. Gray (2000) described several types of sentences to use when creating Social Stories including *descriptive* statements (define the who, what, where, what, when, and why of a situation), *perspective* statements (explain the behaviors and feelings of others), *directive* statements (describe what the child is expected to say and/or do), and *control* statements (provide guidance for the child on how to remember what to do and how to understand the situation). Gut and Safran (2002) proposed that, since some children with disabilities exhibit anxiety when routines are changed, providing advance information about this type of situation can reduce anxiety and provide alternative coping strategies to deal with such a

situation. To increase predictability for Sam and provide guidance on how he could understand the situation, a Social Story was created about riding in the car with his parents which included information about what would happen in the car ride, how he should behave, what positive activities he would engage in while riding in the car, and why his parents needed to make left/right turns to get to where they needed to go. In addition, to capitalize on Sam's special interest in Dr. Seuss books, a second Social Story was created about making left turns and right turns while driving, which was written in the format of Dr. Seuss' "The Foot Book." (The texts of these Social Stories can be found in Appendix G).

"Priming" is another way to increase predictability by manipulating antecedent events, or setting up establishing operations; in priming, an individual previews future events so that they become more predictable (Schreibman, Whalen, & Stahmer, 2000; Wilde, Koegel, & Koegel, 1992). Video priming is one way to do this. Schreibman et al. (2000) videotaped upcoming events or activities in which the participants exhibited problem behaviors and showed these videos prior to the event to help the children predict the event. Their results suggested that informing participants of upcoming events was associated with decreases in disruptive behavior. As Schreibman et al. noted, video permits the presentation of future events in a manner that allows for many of the cues associated with the primed situation (e.g., sight, sound, movement, ancillary features of the environment), which is not possible with verbal description or pictorial representation. The use of video also allows for priming with children who are nonverbal or limited in their ability to comprehend verbal descriptions. Thus, it was decided that video priming would be a more effective method of increasing predictability for Jon than using a visual schedule (as done for Sam) or a Social Story (as done for Sam and Ben). Specifically, Jon viewed a video about a birthday party in a relaxed environment at home. Capitalizing on Jon's

perseverative interest in Sesame Street, Jon viewed the DVD “Elmo and Abby's Birthday Fun!” (© 2009). He also viewed multiple internet videos of his favorite characters (e.g., Ernie and Bert, Alvin and the Chipmunks) singing happy birthday and blowing out candles on a birthday cake. They were novel videos that he had not seen prior to intervention. Of note, this procedure with Jon may be more appropriately characterized as “video modeling,” which typically involves the child observing a videotape of a model engaging in a target behavior and subsequently imitating (Charlop-Christy, Le, & Freeman, 2000) than “video priming,” in which no models appeared in the videos (Schreibman et al., 2000), given that Jon's videos featured characters in them. Video modeling has been shown to result in quicker acquisition of skills and greater generalization than in vivo modeling (Charlop-Christy et al., 2000), perhaps in part because watching videos or television may be automatically reinforcing for some children with autism, such as Jon.

Regardless of whether it was video priming or video modeling, Jon watched the same “happy birthday” videos repeatedly, which served to increase the predictability of the “happy birthday” routine. As another form of priming, Jon practiced blowing out make-believe birthday candles on a toy birthday cake (the Leap Frog Learning Path Counting Candles Birthday Cake). Video priming also allowed Sam to preview the anxiety-provoking situation of riding in the car (while making left turns/right turns) in a one-on-one basis under relaxed conditions. He first listened to audio recordings of the sound of directionals (blinkers) on YouTube and then watched internet videos of cars making left and right turns on YouTube. In addition, Ben’s parents rehearsed leaving the house for a few moments, while Ben “practiced” remained inside with the researcher, before they actually left. For the three participants, the use of visual schedules, Social Stories, priming, and video priming likely functioned to reduce the aversiveness (or anxiety-provoking quality) inherent in unpredictable transitions.

**Providing choices.** In the present study, implementation of choice procedures was employed because allowing individuals to choose activities and reinforcers has been demonstrated to increase task engagement while minimizing escape-motivated problem behaviors (Shogren, Fagella-Luby, Jik Bae, & Wehmeyer, 2004). After all, many researchers have suggested that a sense of unpredictability and uncontrollability is at the heart of anxiety and that the development of coping responses that impart a sense of control also buffers anxiety (Barlow, 2000). For example, research has shown that fear is conditioned at a much higher level in animals that are exposed to uncontrollable shock (Mineka, Cook, & Miller, 1984) than in those who are able to control the shock. Thus, increasing control or choice-making is often an important component of treating anxiety. In fact, treatments for childhood anxiety disorders, such as Parent-Child Interaction Therapy (PCIT) for separation anxiety (Choate, Pincus, Eyberg, & Barlow, 2005), directly target parents' overcontrolling behavior and provide the child with choices and control. Given that problem behavior can be a way of exerting control over one's life (Wehmeyer, 1999), providing opportunities for choice-making may provide children with ASD, especially those who are anxious, with an increased sense of control over their environments (Dattilo & Rusch, 1985), thus reducing their anxiety and allowing them to communicate their wants and needs in more appropriate ways. As such, Sam was given the opportunity to choose several preferred items (e.g., balls, books) to pack in a "Car Bag" that he could bring with him while riding in the car. In addition, when creating the visual schedule, whenever possible, Sam's parents attempted to provide him with a choice of the location to which his parents would drive. In this way, Sam became an active participant in the process of constructing his schedule and selecting preferred items with which to engage himself on the car ride. Similarly, Ben was allowed to choose which preferred activities he could engage in while his parents were out.



**Incorporating the child’s perseverative interests.** Incorporating a child's preferences or interests into a “disliked” or “unpleasant” activity is another antecedent intervention that has been shown to effectively reduce escape-maintained problem behavior (Clarke et al., 1995). Given the level of anxiety that the targeted contexts appeared to provoke in the participants, I decided to incorporate not only high-interest or reinforcing activities, but *perseverative* items or activities that were highly salient and highly preferred, in an attempt to override or counteract the aversiveness of the anxiety-provoking contexts. Perseverative interests refer to an object, activity, or topic with which the child is intensely preoccupied. Several studies have shown that incorporating the perseverative interests of children with autism into behavioral interventions can serve to increase motivation/incentive and has positive outcomes on various aspects of the children's behavior (Baker, 2000; Baker, Koegel, & Koegel, 1998; Vismara & Lyons, 2007). For example, Baker et al. found that highly preferred topics (those in which children with autism showed perseverative interest) could be used to create intrinsically reinforcing and socially appropriate play activities. Vismara and Lyons incorporated perseverative items into the motivational techniques of Pivotal Response Treatment to increase joint attention initiations in children with autism. Furthermore, studies have shown that using obsessions as reinforcers provided contingent upon nonoccurrence of problem behavior were the most effective at reducing inappropriate behaviors (Charlop-Christy & Haymes, 1996). Similarly, Charlop, Kurtz, and Casey (1990) found that using a child’s “aberrant behaviors” (stereotypy, delayed echolalia, and perseverative behaviors) as reinforcers was superior to using food reinforcers, and O'Brien and Repp (1990) found that “stimulatory” reinforcers resulted in successful treatment 75-100% of the time, whereas edible reinforcers were only successful 50-70% of the time. This could be

because of the anxiety-reducing properties of a child's idiosyncratic perseverative, stimulatory, or obsessive reinforcers, in contrast to other tangible or social reinforcers.

Thus, beyond using a wide variety of highly preferred reinforcers, Sam's parents used his primary perseverative interest (Dr. Seuss) as the reinforcer to pair with riding in the car.

Although Sam's favorite book, "The Sneetches" by Dr. Seuss, was an item he owned before intervention and he could access it at home, the researcher created an audio-recording of herself reading this book and other Dr. Seuss books to use at the start of intervention. Sam was only allowed to listen to these Dr. Seuss audio-recordings on car rides. Similarly, Jon was only allowed access to his two favorite toys (Sesame Street pop-up toy and CD player, which were purchased by the researcher and he had not owned prior to this study), which he repeatedly asked for throughout the day, within the context of singing happy birthday. By pairing the activity of singing happy birthday (with the candles lit atop the cake) with an obsessive/perseverative reinforcer that Jon could not access at other times throughout the rest of the day, the formerly anxiety-inducing situation of singing happy birthday and the birthday cake then become an S<sup>D</sup> for approach rather than escape and avoidance through problem behavior. Finally, one way to make separation from his parents less anxiety-provoking for Ben was to pair his parents' departure with his favorite movies or videos that he asked for throughout the rest of the day (e.g., Muppet movie). In addition, while his parents were gone, the researcher engaged in perseverative or self-stimulatory behaviors along with him (e.g., counting with the specific hand motions that Ben directed the researcher to use, repeating the specific perseverative phrases that Ben used, or singing and dancing along with Ben to his favorite songs from the movie he was watching, as Ben directed the researcher to do). Ben's uncle or grandparents were coached to do the same during the sessions in which the researcher left the house along with Ben's parents.

**Graduated exposure.** The power of stimuli to evoke fear reactions can be reduced by exposing individuals to their feared stimuli, which is typically the most critical component of treatment for any anxiety disorder (Kendall, 1994). Habituation occurs when an individual remains in the presence of a feared stimulus or situation (e.g., birthday candles and song, making a left/right turn) until the situation no longer evokes a distressing level of arousal. Most researchers would agree that engaging in exposure is necessary for positive treatment outcome when treating anxiety, and that exposure to the feared stimuli is a crucial element of all CBT for anxiety in TD children (Kendall et al., 2005). Exposure has also been shown to reduce anxiety in children with ASD (Lehmkuhl et al., 2008) as well as fear and problem behavior in children with DD (Davis et al., 2007). With graduated exposure, “brave behavior” (i.e., approach/acceptance responses) is positively reinforced while fearful/avoidant responses are extinguished, as the child is gradually exposed to increasing proximity, intensity, or amounts of the feared stimulus or situation. Given that each of the participant's problem behavior appeared mainly to be escape-motivated (e.g., Sam's parents had avoided taking him on car rides or purposely took longer routes to avoid right/left turns as much as possible), prolonged exposure to the anxiety-provoking activities of singing happy birthday (for Jon), making turns while driving (for Sam), and his parents leaving the house (for Ben), was a key component of intervention. These exposures were conducted in a hierarchical fashion from situations that were rated by parents as less anxiety-provoking to situations that were rated as more anxiety-provoking. For example, Sam first engaged in activities that his parents and home teachers predicted would provoke relatively low levels of anxiety: he listened to audio recordings of the sound of directionals (blinkers) on YouTube and then watched internet videos of cars making left and right turns on YouTube. Sam then engaged in an activity that his parents and home teachers predicted might evoke moderate

levels of anxiety: Sam and his parents practiced listening to the real blinker in their car when the car was stationary (parked in their driveway), first with the car door open and then with the door closed. Finally, Sam engaged in activities that his parents and home teachers predicted would evoke high levels of anxiety, involving riding in the car, moving up the exposure hierarchy from right and left turns that were thought to be less anxiety-provoking (e.g., a small intersection on a side street with a stop sign) to more anxiety-provoking right and left turns (e.g., major intersections with traffic lights). Similarly, Jon was exposed to singing happy birthday with a cake with no candles in it, singing happy birthday with the candles in the cake unlit, singing happy birthday with the candles in the cake lit. Each exposure also attempted to increase Jon's proximity to the cake, little by little, until Jon was eventually sitting in a chair right next to the birthday cake and even blowing out the candles. In these contexts, exposure constituted a setting event that made problem behavior less likely to occur in the presence of the anxiety-inducing discriminative stimulus (the birthday cake, left/right turns, separation from parents).

**Counter-conditioning.** In counterconditioning, the conditioned stimulus (CS) is repeatedly paired with an unconditioned stimulus (US) having a valence opposite from the original US that was used for acquisition. Thus, counterconditioning entails presenting an item or activity that produces an emotional state that is incompatible with anxiety (i.e., an item or activity that induces a relaxed, positive state). For example, Luscre and Center (1996) used anti-anxiety stimuli (e.g., hand-held mirror, music, Play-doh) to elicit a positive response in children with autism and help counter a fear response to dental examinations. Although counterconditioning has not proven more effective than standard exposure (repeated presentation of the CS in the absence of the US) in the treatment of fear in humans, it should be noted that, in the present study, the participants' anxious behaviors did not decrease throughout the repeated

exposures of the baseline sessions. Thus, the researchers decided to use counterconditioning in the present study, pairing the anxiety-provoking situation (the CS) with a highly preferred and otherwise inaccessible positive reinforcer. For Jon, the presentation of the birthday cake and the song happy birthday were paired with his most highly preferred and otherwise inaccessible positive stimulus (e.g., Sesame Street pop-up toy or Sesame Street CD player) and other positive stimuli (e.g., musical candle). For Sam, riding in the car was paired with his most highly preferred and otherwise inaccessible positive stimulus (e.g., audio recording of Dr. Seuss' book "The Sneetches"). For Ben, his parents' departure was paired with his most highly preferred reinforcers (e.g., Muppet Movie, swings, perseverative counting).

**Generalized reinforcement (noncontingent presentation of positive reinforcement).**

Research shows that a child may engage in problem behavior to escape from a situation that signals something aversive. One way to weaken the conditioned aversiveness of this situation is to pair the situation with a wide variety of highly preferred tangible, activity, and social reinforcers (Magito McLaughlin & Carr, 2005), such as candy, books, and singing favorite songs. This pairing establishes the situation as a generalized reinforcer (Skinner, 1953). By being consistently paired with many strongly preferred reinforcers, the formerly anxiety-inducing situation (of singing happy birthday, making right/left turns, or parents leaving the house) then becomes a discriminative stimulus for approach rather than escape/avoidance through problem behavior (Magito McLaughlin & Carr, 2005). In this way, Magito McLaughlin and Carr (2005) trained staff members who had a "poor rapport" with participants to noncontingently present positive reinforcers, which strengthened participants' approach behavior toward the staff members. Similarly, in the present study, upon Ben's parents leaving the house, the researcher delivered his most highly preferred items or activities noncontingently (i.e., Ben was not required

to perform any behavior to receive the preferred item or activity, such as watching the Muppet Movie). Likewise, upon entering the car, Sam immediately was allowed noncontingent access to his most highly preferred items, including the Dr. Seuss book “The Sneetches,” along with listening to an audio-recording of the Sneetches. Sam was not required to perform any behaviors to receive the book or listen to the audio-recording. This antecedent-based strategy of pairing aversive (or anxiety-producing) stimuli or situations with positive (or anxiety-reducing) stimuli *before* the activity stands in contrast to the consequence-based strategy of positive reinforcement contingent upon displaying a certain behavior or completing a certain task, in which the child’s compliance with riding in the car and/or his appropriate behavior in the car ride would be positively reinforced *after* the car ride. Finally, for Jon, upon presentation of the birthday cake accompanied by the birthday song, Jon was immediately given his most highly preferred item, the Sesame Street pop-up toy, without him having to perform any action to receive the pop-up toy. This strategy can also be conceptualized as counterconditioning (see above).

**Contingent presentation of positive reinforcement (contingent upon approach).**

Although Jon was initially provided with the pop-up toy noncontingently (without him having to perform any action to receive it), when the birthday cake and birthday song were presented for a third time, the researcher prompted an approach response by instructing Jon to approach the birthday cake (based on the procedure of Magito McLaughlin & Carr, 2005) and attempt to blow out the candle. Hagopian and Jennett (2008) suggested including prompting as a way to help the child to comply with the steps of the exposure hierarchy, which may be especially important when the child is displaying highly intense anxiety behaviors or not approximating the targeted approach response. If Jon responded by approaching the birthday cake for the preferred item and making any attempt to blow out the candle, the researcher immediately delivered the item and

provided positive feedback. If Jon did not approach the cake, the researcher presented a general cue (e.g., “Jon, do you want to play with this?”, while showing him the Sesame Street pop-up toy or CD player) to encourage an approach response. Following each successful approach response, the researcher gradually increased the cake's proximity to Jon, using the prompt-fading sequence just described to strengthen the approach response. If Jon spontaneously approached the birthday cake and attempted to blow out the candles (with the candles unlit and then with the candles lit), he immediately received positive feedback along with the preferred item. This procedure was not conducted with Sam or Ben in that they were not required to approach a feared stimulus; they were only required to remain in a feared situation (i.e., for Sam, riding in the car while his parent made a left/right turn; for Ben, staying in the house or backyard without his parents).

**Escape extinction.** Even after the introduction of antecedent interventions (e.g., choice procedures), it was possible that participants would still engage, occasionally, in problem behavior during anxiety-provoking situations. In the event that such behavior occurred, parents were told to use escape extinction. Escape extinction involved maintaining the presentation of the anxiety-provoking stimulus or situation (e.g., turning right/left) even after a problem behavior (e.g., yelling) occurred, thereby ensuring that the participant did not escape from the stimulus or situation (Carr et al., 1980). For example, prior to involvement in this research study, when Sam was riding in the car with his parents, they often altered their driving route to avoid left/right turns; Sam learned that his problem behavior (e.g., crying, yelling “no left!” or “straight please!”) would sometimes result in his parents driving straight rather making a left or right turn. Given that Sam’s problem behavior had been maintained by intermittent negative reinforcement (i.e., he often escaped/avoided the anxiety-provoking situation of making a right/left turn), treatment required eliminating this source of reinforcement. Thus, one component of intervention

involved extinction, in which escape was no longer reinforced through task termination (driving back home), avoidance (allowing Sam not to ride in the car when he did want to or avoiding asking him altogether), or altering the driving route to accommodate Sam's anxiety (i.e., driving straight instead of turning right or left).

Similarly, during intervention for happy birthday, the intervention was conducted in an enclosed space with a door that could be closed or blocked so that Jon could not run out of the room. If Jon attempted to run out of the room once his family started singing "happy birthday," the researcher or his mother would block the door. However, his mother was instructed not to hold Jon in her lap near the birthday cake, as she did during baseline sessions, because providing Jon with some choice and control (allowing him to approach the cake on his own to receive the sesame street pop-up toy) was judged to be more effective in reducing his anxiety than holding him near the cake. During baseline, Jon's mother held him and the family continued to keep singing the song in spite of Jon's display of problem behavior (e.g., yelling/screaming). Use of escape extinction prevented Jon from receiving negative reinforcement (i.e., escape from anxiety-provoking stimulus) contingent on the display of problem behavior. However, use of all of the previously mentioned strategies increased the likelihood of subsequent reinforcement, thereby reducing the aversiveness of the situation and undermining the necessity for escape-motivated problem behavior.

**Differential reinforcement of alternative behavior (DRA).** DRA involves providing a reinforcing consequence for a desired response (socially appropriate behavior) that is incompatible with the problem behavior, along with extinction of problem behavior. Many studies have demonstrated the effectiveness of DRA in increasing positive behavior and reducing problem behavior (e.g., Hall, Lund, & Jackson, 1968). Recent studies of PCIT with anxious



children used differential reinforcement in teaching parents to ignore or minimize attention for fearful displays (Pincus, Eyberg, & Choate, 2005). Similarly, in the present study, parents were coached in promoting their children's brave behaviors (e.g., approaching a feared situation, such as approaching a birthday cake with lit candles) in increasingly challenging exposure tasks. Parents were taught to differentially respond to children's brave and anxious behavior by providing labeled praise for brave/approach responses and selective ignoring for anxious or avoidant responses. For example, in addition to Sam's behavior being negatively reinforced by escape/avoidance (primary source of reinforcement), it was hypothesized that his anxious behavior had also been positively reinforced by attention and reassurance (secondary source of reinforcement). Prior to intervention, it was observed that Sam received reassurance/comfort (attention) when he displayed anxious behavior (e.g., crying, yelling) during the car ride, whereas he generally did not receive any attention when he was sitting quietly in the back seat. Therefore, during intervention, his parents and home teachers were instructed to provide attention to Sam and praise him for brave/courageous behavior when he was riding in the car quietly/calmly (i.e., not displaying anxious behavior) and, conversely, to minimize attention for his anxious behavior.

**Introduce discriminative stimuli for nonproblem behavior.** One procedure for mitigating the aversiveness of a situation is to introduce a variety of positive reinforcers known to be strongly preferred by the child. By introducing stimuli that are discriminative for nonproblem behavior in otherwise problematic situations, one can prevent the emergence of problem behavior (Carr & Carlson, 1993). For example, the researcher (L.J.M.) noted that Ben never exhibited problem behavior while watching a video (one of his favorite activities), and his parents confirmed that he rarely if ever exhibited problem behavior while watching a favorite

video on television. The video was discriminative for nonproblem behavior (such as singing and dancing). Thus, Ben was allowed to watch one of his favorite movies (e.g., Muppet Movie) when his parents left the house. Similarly, Sam was offered access to an array of stimuli that were discriminative for appropriate behavior (e.g., Dr. Seuss books, balls) while riding in the car. These preferred items/activities (e.g., watching videos, swinging on swing, reading or listening to Dr. Seuss books) may have functioned as discriminative stimuli for non-problem behavior and/or as reinforcers whose presence helped to mitigate the aversiveness of the anxiety-provoking events (making left/right turns, separation from parents, singing happy birthday).

**Coping self-talk.** In addition to increasing predictability, Ben's Social Story provided him with “coping self-statements” or “coping self-talk” (e.g., Kendall, 1994) that he could tell himself when he was feeling afraid or anxious, such as “I am going to be brave” and “I am going to fight my anxiety and I will win!” Coping self-statements are used before exposures to help reduce anticipatory anxiety and during exposures to help reduce anxiety. Before and during exposures, the researcher and Ben often repeated the coping self-statements from the Social Story, or repeated variations of the coping self-talk, such as referring to Ben as the superhero “Super-Ben.” For example, during the second exposure, when Ben's parents left the house and Ben was alone with the researcher, Ben exclaimed, “That's what I am, Super-Ben!” The researcher responded, “That *is* what you are, Super-Ben! Super-Ben is so good at staying alone without his Mommy or Daddy!”

## **Design**

During the intervention phase, the multicomponent treatment plan was introduced in a multiple baseline design across the three participants in the three formerly anxiety-provoking contexts to evaluate the effects of the interventions. Multiple baseline designs are those in which

the treatment (experimental condition) is applied sequentially (i.e., in an AB fashion) across different individuals, behaviors, or settings (Hersen & Barlow, 1976; Kazdin, 1982). In multiple baseline designs, there is only one baseline (i.e., no removal of the treatment variable) and one intervention condition, and the power of these designs comes from demonstrating that change occurs when, and only when, the intervention is directed at the behavior, setting, or individual in question (Nock, Michel, & Photos, 2007; Rizvi & Nock, 2008). A multiple baseline design across individuals involves collecting repeated measurements of the same behavior in different individuals for varying lengths of time, followed by the intervention phase designed to affect the frequency or level of that particular behavior; as such, the duration of the baseline varies across different individuals (Rizvi & Nock, 2008). Rather than using a return to baseline (as in a reversal or ABAB design), multiple baseline designs replicate the intervention-behavior change relation in temporal sequence across different behaviors, settings, or individuals (Nock et al., 2007). This temporal sequencing (the staggered manner in which treatment is implemented) controls for threats to internal validity, or rules out the likelihood that extraneous factors could account for the observed behavior change (Nock et al., 2007).

Thus, following standard research practice in the field (e.g., Carr & Carlson, 1993; Magito-McLaughlin & Carr, 2005; Moes & Frea, 2002; Owen-DeSchryver, Carr, Cale, & Blakeley-Smith, 2008; Ozdemir, 2008), a multiple baseline design across the three participants with ASD was used to evaluate the impact of the multicomponent intervention packages on problem behavior that occurred during the experimental contexts. Consistent with the rationale of the multiple baseline design, the intervention was implemented for each participant following demonstration of high, stable baseline rates of anxious behavior displayed by the children with

ASD. Thus, the intervention occurred after varying baseline lengths: 4 sessions for Jon, 5 for Ben, and 6 for Sam.

### **Response Recording**

Three dependent variables were recorded: (1) frequency of anxious behaviors (percentage of intervals containing observed physiological signs of anxiety and/or observed behavioral indicators of anxiety), (2) frequency of problem behavior (percentage of intervals containing problem behavior), and (3) Likert-type ratings of appearance of fear/anxiety. Description of these variables can be found in Study 1.

### **Interobserver Agreement (IOA)**

Thirty out of 45 videotaped sessions (66.7% of the sessions), distributed evenly across the baseline and intervention sessions, were coded for IOA by the senior author and another graduate student in clinical psychology; 12 out of 18 of Jon's sessions were coded for IOA (4 baseline and 8 intervention), 10 out of 15 of Ben's sessions were coded for IOA (4 baseline and 6 intervention), and 8 out of 12 of Sam's sessions were coded for IOA (3 baseline and 5 intervention). IOA for problem behavior for Jon's 12 sessions was 94.88% ( $SD = 6.3%$ , range = 80% to 100%); IOA for anxious behavior was 88.16% ( $SD = 6.6%$ , range = 80% to 100%). IOA for problem behavior for Ben's 10 sessions was 97% ( $SD = 3.6%$ , range = 91% to 100%); IOA for anxious behavior was 93% ( $SD = 6.6%$ , range = 83% to 100%). IOA for problem behavior for Sam's 8 sessions was 98% ( $SD = 3.7%$ , range = 91% to 100%); IOA for anxious behavior was 94% ( $SD = 6.1%$ , range = 85% to 100%).

### **Data Analysis Procedures**

To further quantify treatment outcomes, two measures of nonregression effect size were calculated for each participant: the percentage of nonoverlapping data (PND) and mean baseline

reduction (MBLR). PND, a measure of the proportion of nonoverlapping data between baseline and intervention, is the percentage of data points in an intervention phase that are below the range of data for problem behavior in the baseline phase. PND is determined by calculating the number of intervention data points that fall below the lowest baseline data point and dividing that number by the total number of data points in the intervention phase, multiplied by 100 (Scruggs, Mastropieri, & Casto, 1987). PND scores can range from 0% to 100%, with higher scores indicating more effective treatments. A PND of 90% indicates that the treatment was highly effective in behavior reduction, a PND of 70-90% indicates that the treatment was moderately effective, a PND of 50-70% indicates that the treatment was minimally effective, and a PND of <50% indicates that the treatment was ineffective. I also calculated the average reduction of behavior from baseline (e.g., Kahng, Iwata, & Lewin, 2002), known as mean baseline reduction (MBLR) (Campbell, 2003), by subtracting the mean of the treatment data-point values from the mean of the baseline values, dividing the difference by the mean of baseline values, and multiplying by 100.

## **Results**

### **Frequency of Problem Behavior**

Data from Study 2 are presented in Figures 8-10. Figure 8 shows the frequency of problem behavior for the 3 children in the anxiety-provoking contexts during the baseline and intervention phases. For all three children, frequency of problem behavior decreased substantially from before to after intervention, with a 97%, 96%, and 100% reduction in frequency of problem behavior for Jon, Ben, and Sam respectively. Intervention (post-baseline data) lasted 10 weeks for Jon, 9 weeks for Ben, and 4 weeks for Sam. The average frequency of problem behavior (the average percentage of intervals with problem behavior) for baseline

versus intervention, respectively, was 81% versus 2% for Jon, 77% versus 33% for Ben, and 60% versus 0% for Sam.

For Jon, the mean percentage of intervals in which problem behavior occurred during the happy birthday context was 81.13% ( $SD = 6.5\%$ , range = 74% to 87%) during baseline and 2.4% ( $SD = 3.8\%$ , range = 0% to 13%) during intervention (see Figure 8), a 97% mean baseline reduction (MBLR) in frequency of problem behavior. The PND for Jon's problem behavior was 100% (14 of 14 intervention points did not overlap with baseline points), which indicates that the treatment was highly effective in reducing problem behavior. During baseline, none of the happy birthday sessions (0 of 4 sessions) were completed without any problem behavior. During intervention, 50% of happy birthday sessions (7 of 14 sessions) were completed successfully without any problem behavior and 86% (12 of 14) were completed with  $\geq 10\%$  of intervals containing problem behavior.

For Ben, the mean percentage of intervals in which problem behavior occurred from the time his parents first mentioned leaving the house or indicated that they were going to leave the house until the time they returned was 77% ( $SD = 27\%$ , range = 36% to 100%) during baseline and 3.2% during intervention ( $SD = 5.2\%$ , range = 0% to 18%), an 96% mean baseline reduction (MBLR) in the frequency of problem behavior. The PND for Ben's problem behavior was 100% (11 of 11 intervention data points did not overlap with baseline points). During baseline, none of the sessions in which his parents left the house (0 out of 5 sessions) was completed without any problem behavior. During intervention, 20% of sessions were completed successfully without any problem behavior, 82% of sessions were completed with 3 or fewer intervals containing problem behavior, and 100% were completed with less than 8 instances of problem behavior.

For Sam, the mean percentage of intervals in which problem behavior occurred from the time his parents started driving the car until the time he exited the car was 60% ( $SD = 13.6\%$ , range = 43% to 77%) during baseline and 0% during intervention (see Figure 10), a 100% mean baseline reduction (MBLR) in the frequency of problem behavior. The PND for Sam's problem behavior was 100% (6 of 6 intervention data points did not overlap with baseline points). During baseline, none of the sessions (0 of 6 sessions) was completed without any problem behavior. During intervention, 100% of sessions (6 of 6 sessions) were completed successfully without any problem behavior.

### **Frequency of Anxious Behavior**

Figure 9 shows the frequency of anxious behavior for the 3 children in the anxiety-provoking contexts during the baseline and intervention phases. For all three children, frequency of anxious behavior decreased substantially from before to after intervention, with a 76%, 88%, and 90% reduction in frequency of anxious behavior for Jon, Ben, and Sam respectively.

For Jon, the mean percentage of intervals in which anxious behavior occurred during the happy birthday context was 53.4% ( $SD = 8.1\%$ , range = 43% to 60%) during baseline and 12.97% ( $SD = 16.12\%$ , range = 0% to 62%) during intervention (see Figure 9), a 75.7% mean baseline reduction (MBLR) in the frequency of anxious behavior. The PND for Jon's anxious behavior equaled 92.86% (13 of 14 intervention data points did not overlap with baseline points), which indicates that the treatment was highly effective in reducing anxious behavior.

For Ben, the mean percentage of intervals in which anxious behavior occurred from the time Ben's parents first mentioned leaving the house or indicated that they were going to leave the house until the time they returned was 83% ( $SD = 22.5\%$ , range = 50% to 100%) during baseline and 9.6% during intervention ( $SD = 13.9\%$ , range = 0% to 45%), an 88% mean baseline

reduction (MBLR) in the frequency of anxious behavior. The PND for Ben's anxious behavior was 100% (11 of 11 intervention data points did not exceed the baseline data points).

For Sam, the mean percentage of intervals in which anxious behavior occurred from the time Sam's parents started driving the car until the time he exited the car was 71% ( $SD = 16.2\%$ , range = 54% to 96%) during baseline and 7% ( $SD = 6.6\%$ , range = 3% to 17%) during intervention (see Figure 9), a 90% mean baseline reduction (MBLR) in the frequency of anxious behavior. The PND for Sam's anxious behavior was 100% (i.e., 100% of the intervention data points did not overlap with baseline data points).

### **Subjective Ratings of Anxiety**

Using the Likert-type rating scale, on a scale of 0 (no anxiety) to 3 (high anxiety), observers rated Jon's anxiety as an average of 2.8 ( $SD = 0.5$ , range = 2 to 3) during baseline and an average of 0.21 ( $SD = 0.38$ , range = 0 to 1) during intervention (see Figure 10). Ben's anxiety was rated an average of 3 ( $SD = 0$ ) during baseline and an average of 0.46 ( $SD = 0.52$ , range = 0 to 1) during intervention. Sam's anxiety was rated an average of 2.67 during baseline ( $SD = 0.4$ , range = 2 to 3) and an average of 0.17 during intervention ( $SD = 0.26$ , range = 0 to 0.5).

## **Discussion**

Our results offer initial support for using a multimethod approach to assess anxiety and related problem behavior as well as a multicomponent behavioral intervention to treat anxiety and related problem behavior in children with ASD and ID.

### **Study 1**

Study 1 used a multimethod approach to address the difficulties inherent in the study of anxiety, similar to the multimethod approach used by previous research to examine the construct of rapport (Magito McLaughlin & Carr, 2005) and mood (Carr, Magito McLaughlin, et al., 2003).



Four sources of data – (1) contextual data from the SSS and CAI ratings made by parents, (2) subjective anxiety ratings made by research assistants using the Likert-type rating scale, (3) direct observation of frequency of anxious behaviors, and (4) heart activity as measured by mean HR and RSA – were examined in an attempt to evaluate the construct of “anxiety” in children with ASD. In the contexts that were rated as highly anxiety-provoking (a ‘5’ on a scale of 0 to 5) by parents, the participants (a) displayed behavioral indicators of anxiety in at least 50% of the intervals that constituted each context (53% for Jon, 83% for Ben, and 80% for Sam), (b) were judged to be highly anxious by undergraduate observers using a Likert-type rating scale (mean ratings of 2.8 for Sam, 2.75 for Jon, and 3 for Ben, on a scale of 0 to 3, in which 0 is no anxiety and 3 is maximum anxiety), and (c) displayed a higher HR (for Jon and Ben, not Sam) and lower RSA (for Ben) than in Low-Anxiety contexts.

Taken together, these findings suggest that the use of multiple methods of assessment may provide a reliable, valid, and clinically useful strategy for studying ambiguous context variables such as anxiety. The major objective of this first study was to explore anxiety from multiple perspectives. The inherent difficulties in studying anxiety in this population necessitated a combination of assessment strategies. Importantly, direct observation served as a means of producing quantitative information to validate and support the use of more qualitative (i.e., subjective ratings) procedures.

Study 1 also documented a relationship between anxiety and problem behavior. For all three participants, few problem behaviors occurred in Low-Anxiety contexts (those rated as a ‘1’ on the SSS and/or CAI, those consistently rated as a ‘0’ or ‘1’ on the subjective Likert-type rating scale, and those containing no or few anxious behaviors) as compared with High-Anxiety contexts (those rated as a ‘5’ on the SSS and/or CAI, those consistently rated as a ‘2’ or ‘3’ on the

subjective Likert-type rating scale, and those with >50% of 10-s intervals containing anxious behavior). Moreover, both Jon and Ben exhibited higher HR in the High-Anxiety conditions as compared with the Low-Anxiety conditions, and Ben exhibited significantly lower RSA in the High-Anxiety condition than in the Low-Anxiety condition (there were too many artifacts to analyze RSA for Jon). Although the two sessions that yielded usable data for Sam failed to show a significant difference in HR or RSA between the High-Anxiety and Low-Anxiety condition, this may be because Sam was engaging in self-stimulatory activity (playing with toy balls) and he may have been excited (physiologically aroused in terms of positive affect rather than negative affect), or it may be because Sam was still recovering from the car ride (as the ball play occurred after the car ride).

## **Study 2**

The results of Study 2 support the effectiveness of a multicomponent behavioral intervention plan for the treatment of anxiety in children with ASD and ID, a group that has been neglected in the literature. In Study 2, following the multicomponent intervention, all three participants showed a decrease in anxiety (i.e., anxious behaviors, subjective anxiety ratings) and even more of a decrease in problem behavior in contexts that were formerly anxiety-provoking. It is important to note that, although the participants still exhibited some anxious behavior during intervention (in an average of 13% of intervals across sessions for Jon, an average of 9.6% of intervals across sessions for Ben, and an average of 7% of sessions for Sam), the intervention procedures appeared to mitigate their anxiety substantially so that it rarely resulted in displays of problem behavior. Of note, in contrast to baseline, almost all of the anxious behaviors that remained during intervention were of the milder and/or subtler variety, such as eyes rapidly darting back and forth for Jon when the birthday cake was presented (even when he sat near the

cake quietly), asking his parents “Where are you going?” for Ben, and turning his head to look out the back of the car window for Sam.

As shown in Figures 8-9, visual inspection of the data demonstrates that each participant's problem behavior and anxious behavior decreased when *and only when* the intervention was applied to that participant. This pattern of results strongly suggests that the intervention caused the behavior change, rather than extraneous factors such as history, maturation, or statistical regression (see Nock, 2002). In other words, the large change in problem behavior *immediately* after the intervention was implemented across all three children, in three different homes and with three different sets of parents, makes it unlikely that some extraneous variable caused these changes.

In terms of social validity, outside observers (undergraduate research assistants with no knowledge of the purpose of the study) corroborated these findings in that they judged Jon's level of anxiety to be in the “high” range (mean = 2.8 on a scale of 0 to 3) during baseline and close to the “no anxiety” range (mean = 0.2) during intervention. They judged Ben's anxiety to be in the high range (mean = 3) during baseline sessions and in the “none” to “mild anxiety” range (mean = 0.46) during intervention sessions. Finally, they judged Sam's anxiety to be in the moderate to high range (mean = 2.67) during baseline sessions and close to the “no anxiety” range (mean = 0.17) during intervention.

Importantly, the present investigation focused on anxious behaviors that occurred in natural social contexts, thereby addressing ecological validity issues relevant to typical home, school, and community settings. Although the generalizability of the current findings is subject to the limitations of a study involving only three participants, the results provide evidence that anxiety, and problem behavior related to anxiety, can be reliably assessed and treated in children

with ASD and ID. The results of Study 1 and 2 have several important theoretical and clinical implications with regard to the assessment of anxiety in children with both ASD and ID, identifying behavioral markers of anxiety in children with ASD, the applicability of *DSM-IV* diagnoses for children with ASD, and conceptualizing anxiety within a four-term contingency (i.e., as a setting event or discriminative stimulus).

## **Implications of Studies 1 and 2**

**The relationship between anxiety and IQ in children with ASD.** Many researchers have suggested that children with ASD who are higher functioning may experience more anxiety than those who are lower functioning (e.g., Weisbrot et al., 2005). For instance, Sudholsky et al. (2008) found that children without cognitive impairment ( $IQ \geq 70$ ) were rated by their parents to display more anxiety than were those without cognitive impairment ( $IQ < 70$ ). One common explanation for these findings is that children without cognitive impairment are likely to have a greater understanding of their condition, which leads to increased anxiety. However, I propose that another reason for these findings could be due to the difficulty of assessing or measuring anxiety in children with ASD who have a cognitive impairment, given that, as previously discussed, they often lack the ability to self-report or communicate their fear or anxiety, and may express their fear or anxiety in idiosyncratic ways. Thus, it may be that parents, teachers, or clinicians may not recognize their children's fear or anxiety, and may even attribute their problem behavior to noncompliance, disobedience, oppositionality, or anger/irritability rather than fear or anxiety. I would argue that the hypothesis that cognitively impaired children with ASD are less anxious than those with average IQ cannot be supported or disproved until we as a field can devise reliable, valid ways of measuring anxiety in “lower functioning” children with ASD who have cognitive and/or language impairments, and thus may be unable to express their

fears or anxiety. The present study aims to take a first step toward operationally defining and measuring fear or anxiety in this population.

**Identifying behavioral markers of anxiety in ASD.** The difficulty with assessing anxious behavior in children with ASD, beyond the fact that they may not be able to adequately express or even recognize their anxiety, is that it may not even be clear to parents, teachers, clinicians, or researchers which specific behaviors are indicative of anxiety. In fact, there may have been a selection bias in the present study in that, by recruiting children with ASD who were anxious, the parents who responded were those parents who already recognized their child's anxiety or who attributed their children's problem behavior to anxiety. There may be many parents of children with ASD who did not respond because they attribute their children's problem behavior to anger, irritability, frustration, boredom, noncompliance, disobedience, oppositionality, or other such factors. Indeed, researchers often disagree on which behaviors are manifestations of anxiety in ASD versus symptoms of ASD itself. For example, Howlin (1998) noted that high levels of obsessional behavior are often an indication of uncertainty, anxiety, or distress in individuals with autism. Brereton et al. (2006) stated that symptoms of anxious behavior in ASD include “fear of separation from familiar people, resistance to change, crying easily over small upsets, tenseness, shyness and irritability.” Khreim and Mikkelsen (1997) observed that fear in adults with ID may manifest itself as “agitation, screaming, crying, withdrawal, freezing, or regressive clingy behavior.” Although some or all of these behaviors might indicate anxiety in a child with ASD at certain times and in certain contexts, the same child might also cry, scream, withdraw, become tense, or behave irritably because he is tired, in pain, or feeling overstimulated, angry, frustrated, sad, or otherwise distressed.

This subjectivity in terms of behavioral indicators of anxiety in ASD is also an issue in informant-rating scales. For example, many items on parent-report measures of anxiety for TD children, such as the SCARED (Birmaher et al., 1999) (e.g., “My child is nervous” or “My child is a worrier”), are subjective items rather than observable, objective markers of anxiety. Even the seven “General Anxiety Items” in the Anxiety, Depression, and Mood Scale (ADAMS; Esbensen, Rojahn, Aman, & Ruedrich, 2003), which was designed as an observationally based informant rating scale of symptoms related to anxiety, depression, and mania in individuals with ID, are: *nervous, does not relax or settle down, tense, worried, anxious, experiences panic attacks, and trembles when frightening situations are not present*. Although a child with ASD might indeed be anxious, descriptors such as “nervous,” “worried,” and “anxious” are not necessarily objective, observable, measurable, behavioral indicators of anxiety. After all, an observer might rate the child as “nervous” or “anxious” *because* he is engaging in stereotyped behavior (which could be to gain sensory stimulation rather than reduce anxiety), aggression or SIB (perhaps due to gaining an attention or escaping a demand rather than anxiety), or frequently cries (perhaps due to sadness, frustration, pain, illness, or the inability to communicate), whereas another observer might attribute aggression, SIB, stereotypical behavior, or crying to factors other than anxiety. Cautela (1977) identified several symptoms of anxiety on the *Cues for Tension and Anxiety Survey Schedule* (CTASS), some of which are experienced states and some of which are observable behaviors, including *tenseness, sweating, face flushed or warm, skin cool and damp, trembling or shaking, tight grip, scratching a certain part of body, moving leg up and down, biting nails, grinding teeth, and trouble with speech*. Although these appear to be the most concrete, observable, behavioral descriptors of anxiety available to date, it is still possible that these behaviors could be associated with another internal state (e.g., anger/frustration,

excitement, pain/illness), could be context-dependent (e.g., the same behaviors could indicate different affective states depending on the context), or could be idiosyncratic (e.g., could indicate anxiety in one child with ASD but not in another). Further, Cooray, Gabriel, and Gaus (2007) noted, in modifying the diagnostic criteria for a panic attack for individuals with ID, that “extreme panic may result in irritability, aggression and destructive behavior and may also cause lashing out of arms and legs and head banging.” However, although aggressive, destructive, and self-injurious behavior may be functionally related to anxiety, these behaviors may also function to obtain attention, escape social interaction, obtain a tangible item or activity, or escape a disliked task or demand; context thus becomes critical. As a result, it is likely that identifying idiosyncratic markers of anxiety, or characteristics that are particular to each individual, might be the most reliable, valid, and clinically useful method of assessing anxiety in children with ASD. The present study aimed to identify idiosyncratic markers of anxiety in children with ASD, though future research should compare such markers in situations that are reportedly associated with other affective states – including anger, irritability, frustration, and excitement – to examine whether these states can be differentiated from anxiety.

**Applicability of DSM-IV diagnoses for ASD.** One difficulty with assessing anxiety in children with ASD, beyond their communication difficulties and idiosyncratic behavioral expression, is that most self-report questionnaires, informant-report questionnaires, and semi-structured interviews designed to assess DSM-IV criteria are normed and validated with TD children, not children with ASD. Although I initially attempted to use the SCARED-parent report version for the children with ASD in our study as a proxy for the cognitive/affective component of anxiety, it became clear to us that many of the items on the SCARED were not appropriate for the children in our study. For example, although items such as “My child follows

me wherever I go” were objective, observable, behavioral indicators of anxiety, some of the parents reported that they could not rate items such as “My child worries about other people liking him” or “My child worries about being as good as other kids” because they were not sure what their children were thinking. Thus, when in doubt, they rated such items as a zero (“Not true or hardly ever true”).

Although some researchers (e.g., Brown, Aman, & Lecavalier, 2004) have argued that *DSM-IV* criteria are applicable to children and adolescents with mild or moderate ID, other researchers have questioned the applicability of the existing diagnostic system to individuals with ASD, DD, and ID (e.g., Leyfer, Woodruff-Borden, Klein-Tasman, Fricke, and Mervis, 2008). For example, results of a study by Leyfer et al. (2008) suggested the need either to modify the definition of GAD or create a new category of anxiety to capture the “anticipatory” anxiety exhibited by a large proportion of children with Williams Syndrome. To begin to address the limitations in applying *DSM-IV* criteria to individuals with ID, Fletcher, Loschen, Stavrakaki, and First (2007) edited the *Diagnostic Manual – Intellectual Disability (DM-ID)*, which was created to adapt *DSM-IV* diagnostic criteria for individuals with ID. In the chapter on adapting anxiety disorder diagnoses, the authors stated that, in children and persons with Severe ID, anxiety pertaining to social phobia (for example) may be expressed by crying, tantrums, freezing, or shrinking from social situations with unfamiliar people (Cooray et al., 2007).

According to the American Psychiatric Association, the “central feature” of separation anxiety disorder (SAD) is “unrealistic and excessive anxiety upon separation or anticipation of separation from major attachment figures” (APA, 1994). Although Ben undoubtedly exhibited recurrent excessive distress when separation from his parents occurred or was anticipated, it was difficult for his parents to know whether he was worried about losing them or harm befalling



them (e.g., parents getting in a car accident), or worried that an untoward event would lead to separation from his parents (e.g., getting lost or being kidnapped), as he may have been unable to articulate such worries.

Similarly, the diagnosis of specific phobia in the *DSM-IV* may not be applicable to children with ASD and ID. The categories of specific phobia listed in the *ADIS-IV* are “animal type” (e.g., snakes, spiders, dogs, bees/insects, birds), “natural environment type” (high places, thunderstorms or lightning, water, darkness), “blood-injection or injury type” (getting shots or blood tests, seeing blood from cut or scrape), “situational type” (cars, planes, buses, trains, or any other way of traveling; elevators or small enclosed places), and “other type,” which includes loud noises (e.g., fireworks), doctors or dentists, vomiting, choking, or catching a disease. With the exception of fear of dental exams (Luscre & Center, 1996), swimming pools (Rapp et al., 2005), and riding a school bus (Luiselli, 1978), specific phobias for children with ASD often involve more idiosyncratic stimuli or situations not mentioned in the *ADIS-IV* or *DSM-IV*, including fear of the sound of flushing toilets (Jackson & King, 1982; Koegel et al., 2004), toys with animal sounds, vacuum cleaners, and hand mixers (Koegel et al., 2004), going into the front yard or backyard without a parent (Love et al., 1990), the sight and sound of a running bathroom shower (Love et al., 1990), animatronic objects (e.g., dancing Elmo doll) (Ricciardi et al., 2006), onomatopoeic sounds (e.g., “huff and puff”) (Carr et al., 2009), pictures of sea creatures and ocean scenery (Carr et al., 2009), the “Arthur” video collection (Cale et al., 2009) and, in the present study, fear of “happy birthday” and left/right turns. In addition, children with autism commonly display severe “transition-associated anxiety” (Steingard et al., 1997) that is not captured in the *DSM-IV* anxiety disorders diagnoses. Clinically, I believe that this is one of the reasons why we see so many patients with ASD who are diagnosed with “anxiety disorder

NOS,” because their fears and anxieties do not clearly fit into any *DSM-IV* category, although this hypothesis is in need of further examination. Of note, although the literature on psychopathology in adults with ASD and/or ID is beyond the scope of our study (which focuses on children with ASD and ID), Charlot et al. (2008) reported that many of the inpatients in their sample of adults with ASD and/or ID were diagnosed with anxiety disorder NOS or generalized anxiety disorder (62% of the ASD group and 38% of the ID group).

In a study by Cordeiro, Ballinger, Hagerman, and Hessler (2011), the authors administered the Anxiety Disorders Interview Schedule for DSM-IV: Parent Report Version (ADIS-IV) (Silverman & Albano, 2004) to assess the presence and severity of current anxiety disorders according to *DSM-IV* criteria in individuals with fragile X syndrome (ages 5.0-33.3 years). For social phobia, the authors examined an additional diagnostic category based on their determination that the screening question (“In social situations, does your child worry that they might do something that will be embarrassing?”) was beyond the cognitive or expressive language capacity of most of those with ID. Elimination of the screening question criteria resulted in an increased rate of social phobia among participants with ID (from 32.8% to 69.0%). As the authors noted, this single modification allowed for a diagnosis of social phobia in individuals who exhibited clinically significant impairment as a result of social phobia symptoms, but were unable to verbalize or explain “a worry that they might do something embarrassing.” As a field, we must consider similar modifications to other *DSM-IV* diagnostic categories in order to accurately assess anxiety in children with ASD and ID.

**Anxiety as a setting event versus discriminative stimulus.** Given the contextual model of problem behavior previously described (Carr & Smith, 1995), I proposed that anxiety could either be the S<sup>D</sup> that directly and immediately leads to the occurrence of problem behavior (e.g.,

child is feeling anxious and thus bites himself to reduce the anxiety) or the setting event that sets the stage for problem behavior to be triggered by another  $S^D$  (e.g., child is feeling anxious and thus, when asked to board the bus, he is more likely to bite himself). For each of the three participants in the present study, the anxiety-provoking objects or situations that triggered problem behavior appeared to function as discriminative stimuli for problem behavior in that the stimuli directly and immediately elicited problem behavior.

In future research, I hope to examine anxiety as a setting event. If anxiety is a setting event for problem behavior in a child with ASD then, when the child is already anxious and a certain  $S^D$  is presented (e.g., a difficult task, a demand, a new person, a non-preferred person), that  $S^D$  will likely result in problem behavior whereas, in the absence of anxiety (the setting event), problem behavior might *not* result when presented with the same  $S^D$ . For example, if a child experiences anxiety and exhibits an increase in sympathetic activity (and/or a decrease in parasympathetic activity) compared with baseline levels, this could be the setting event such that, when a certain  $S^D$  is presented (e.g., “Time to brush your teeth”), that  $S^D$  triggers problem behavior. Thus, the presence of a biological setting event, such as anxiety, can make a particular demand or situation more aversive than it normally would be, thereby increasing the likelihood of escape motivated problem behavior (Carr, Magito McLaughlin, et al., 2003; Carr, Smith, et al., 2003; Horner, Vaughn, Day, & Ard, 1996). Using this conceptualization, problem behaviors that are reported by parents and teachers to be unpredictable and “come out of nowhere” (e.g., “Some days he brushes his teeth right away without any problem behavior but, on other days when I ask him to brush his teeth, he starts hitting himself!”) may actually be precipitated by an internal physiological antecedent, such as anxiety/arousal, or by an external antecedent that elicits anxiety and thus makes a particular discriminative stimulus more aversive than it normally

would be.

In the present study, I only had two conditions: high anxiety and low anxiety. In future research, to assess the combined effect of anxiety (as a setting event) and a stressor (as a discriminative stimulus) on problem behavior, I plan to superimpose a stressor versus no stressor condition, in a reversal design, on the high anxiety and low anxiety contexts, similar to the design in Magito McLaughlin and Carr (2005), Carr, Magito McLaughlin, et al. (2003), and Carr, Smith et al. (2003). This procedure will yield four conditions: high anxiety plus stressor (HA+S), high anxiety plus no stressor (HA+NS), low anxiety plus stressor (LA+S), and low anxiety plus no stressor (LA+NS). For example, if the context “waiting in line” is identified as stressful (i.e., rated as a ‘5’ on the SSS), the high anxiety conditions (HA+S and HA+NS) might involve observing the child while waiting in line at the grocery store. For the HA+S condition, if “changes in routine” was identified as being problematic on the CAI, the child might be asked by his mother to return to an aisle to pick up an additional item. By contrast, in the HA+NS condition, I would only observe waiting in line (not returning to pick up an additional item). In the LA+S condition, I might observe the child playing on the playground and then observe a stressor, such as the child’s mother arriving to pick the child up from the playground and transition the child home. In the LA+NS condition, I would observe the child continue to play on the playground (no stressor would be added). This design would allow us to examine the effects of anxiety as a setting event versus a discriminative stimulus.

Related to the notion of anxiety as a discriminative stimulus versus setting event, the three children in the present study displayed “situational” anxiety that occurred predictably in the presence of specific discriminative stimuli. In future research, I aim to target anxious children with ASD whose problem behavior appears to be more unpredictable. Given that the function of

problem behavior cannot always be identified using a functional assessment (Vollmer et al., 1994), our ultimate goal is to be able to predict when problem behavior is more likely to occur in reportedly anxious children with inconclusive functional assessments, whose problem behavior often appears to “come out of the blue.” If future research can investigate whether, prior to the display of problem behavior, children with ASD show a particular pattern of physiological arousal (e.g., heightened sympathetic activity, reduced parasympathetic activity), perhaps we could ultimately be able to predict problem behavior before it actually occurs, and thus prevent it. However, it may make sense for future research to continue to examine patterns of physiological arousal and behaviors in situationally anxious children (whose anxiety is more predictable) in order to identify reliable and valid patterns before extending research to children with more unpredictable anxiety whose problem behavior reportedly “comes out of nowhere.”

### **Limitations and Future Directions**

Although the results of this study are encouraging, there are several limitations that should be addressed by future research. First, given that I did not, for purposes of this study, aim to differentiate “anxiety” from “fear,” I did not include assessment measures or methods to discriminate between anxiety and fear. Although there is no universally agreed upon distinction between fear and anxiety, “fear” is generally considered to be a reaction to an overt threatening stimulus (with escape or avoidance being the result of increased cue proximity), whereas “anxiety” is generally thought of as a more general state of distress that lasts longer, is evoked by less explicit or more generalized cues, and involves physiological arousal but often without organized functional behavior (Lang, Davis, & Öhman, 2000). Related to this, Barlow (2000) characterized “anxiety” as a *future*-oriented mood state in which one is ready or prepared to attempt to cope with upcoming negative events whereas in “fear,” the danger is *present* and

imminent. Of note, the children in the present study sometimes exhibited “fearful” behaviors *before* they were actually exposed to the feared discriminative stimuli (i.e., birthday cake with lit candles or singing happy birthday, right/left turns, parents leaving the house). For example, during Ben's 9<sup>th</sup> intervention session, his 6 instances of anxious behavior (all verbalizations such as “Where are you going Mommy?”) all occurred in the minute *before* his parents left the house (starting when his mother stood up from the chair in which she was sitting, which Ben interpreted as a cue that she would be leaving soon), with no instances of anxious behavior occurring after his parents actually left the house. Thus, Ben's anxious behavior occurred in anticipation of his parents leaving the house, not after they actually left, which is consistent with the future-oriented state of anxiety. Of course, it is possible that Ben's mother standing up from the chair in which she was sitting became the discriminative stimulus for fear (instead of Ben's mother walking out the front door, which was the original S<sup>D</sup>). However, it is also possible that the children were appraising novel stimuli as being *potentially* harmful in the future and therefore preparing themselves for an upcoming negative event (a hypothesis raised by Gallo, Klein-Tasman, Gaffrey, & Curran, 2008 regarding their children with Williams syndrome), which would constitute a general appraisal of environmental threat that is more consistent with Barlow's (2000) notion of anxiety or “anxious apprehension” than fear. Given that the children in this study were, at times, anticipating the occurrence of negative events before the original discriminative stimuli were actually present, this suggests that the term “anxiety” is in fact appropriate to apply to this study's participants. Nevertheless, further research is warranted to be able to discriminate between anxiety and fear in children with ASD and ID.

In addition to not discriminating between anxiety and fear, I did not include assessment measures or methods to discriminate between anxiety and other negative affective states such as

anger, irritability, frustration, sadness, or boredom. Because of this limitation, it is possible that other forms of negative affect may account for or partially account for the “anxious behaviors” and faster heart rate (and, for Ben, lower RSA) observed in this study. Although a functional analysis was conducted to compare the effects of anxiety-provoking versus non-anxiety-provoking situations on problem behavior, future research should conduct a more detailed functional analysis, comparing situations that are reported to make children with ASD afraid or anxious to other disliked or aversive situations. This can help determine whether the frequency, duration, intensity, or type of problem behavior children with ASD engage in when they are afraid or anxious is different from the problem behavior they engage in when they are angry, frustrated, irritated, bored, or experiencing some other negative emotional state (other than fear or anxiety). Clinically, we know there is a difference between when a child is afraid to do something versus when a child simply does not want to do something. In terms of research, however, this distinction is difficult to make, especially when the child with ASD cannot accurately communicate his thoughts or emotions, or cannot speak at all. Thus, future research should work to differentiate when a child with ASD is engaging in problem behavior to “escape anxiety” versus to “escape demand,” as it is traditionally termed in the applied behavioral analysis literature. Of note, preliminary psychometric evidence supports a distinct construct of anxiety in children with ASD; Lecavalier, Gadow, DeVincent, Houts, and Edwards (2009), using confirmatory factor analysis with 498 children with ASD, found a GAD factor that was separate from a depression or mood disorder factor and disruptive behavior disorder factors. Although Lecavlier et al. acknowledged that their sample was biased towards higher functioning children with ASD, their results suggest that the construct of anxiety can be defined and separated from other constructs (such as depression) in children with ASD. Regardless, further research is

necessary to be able to differentiate among anxiety, fear, and other negative affective states in children with ASD and ID.

A second limitation of this study is that, by implementing many interventions at the same time, we cannot ascertain the unique contribution made by each intervention in the multicomponent intervention plan. I believe that some elements of the intervention functioned as setting events that counteracted or attenuated the effects of the anxiety-inducing discriminative stimuli. However, given the multicomponent nature of the study, it was not possible in this study to attribute behavior change to a particular component or combination of treatments. It is unclear whether all of these interventions were necessary for change or whether a more limited treatment would have been as effective (Nock, 2002). This limitation occurs often in applied settings in which it is difficult to conduct a “pure” examination of the effects of a single treatment (Winn, Skinner, Allin, & Hawkins, 2004). As Winn et al. (2004) noted, when any behavior is targeted that is likely to be supported or reinforced in the child’s natural environment, the change in that behavior may be partially attributed to the intervention and partially attributed to this environmental support that is accessed as the behavior changes. This limitation of the present study could be addressed by future research that systematically dismantles the intervention package to determine which components are necessary and which are not. However, as noted previously, each of the intervention procedures employed in this study has been demonstrated, in previous research, to be effective in reducing either problem behavior (in individuals with ASD or DD) or anxiety (in TD children). Further, the use of multicomponent rather than single component interventions is currently considered to be best practice in the field of developmental disabilities (e.g., Carr & Carlson, 1993; Carr et al., 1999; Lucyshyn et al., 2007). As Carr and colleagues (2002) noted, “For any given individual, behavior challenges are likely to be



dependent on multiple functional and structural variables whose influence demands a multidimensional remediation strategy build on the assessment information.”

Related to the limitation that we do not know which treatment components were the “active” elements of treatment, it is important to note that most researchers would agree that exposure to the feared stimuli is a key element of all behavioral treatment and CBT for child anxiety and is necessary for positive treatment outcome (e.g., Kendall et al., 2005). Similarly, Jennett and Hagopian (2008) noted that exposure and reinforcement were treatment components common to all of the studies they reviewed pertaining to the treatment of “phobic avoidance” in individuals with ID, and could arguably be considered the primary treatment components. Although exposure and reinforcement may also be the primary treatment elements in the present study, it is worth noting that Jon, Ben, and Sam were exposed to their feared stimuli/situations during baseline observations for 4, 5, and 6 exposures, respectively, without showing significant habituation. Of course it is likely that, if they had been exposed to the feared stimuli/situations many more times (without the other components in the multicomponent intervention plan), they would have eventually habituated. However, ethically, more repeated trials in the presence of the feared stimulus would likely have been quite distressing to the children. Further, anecdotally, our clinical observations suggested that pairing the anxiety-provoking stimuli with equally potent or even more powerful perseverative stimuli (i.e., Sesame Street, Kai-Lan, Dr. Seuss; or “antianxiety stimuli,” as Luscre and Center called it) served to counteract the children's fearful/anxious responses in a way that exposure to the feared stimulus alone (without pairing it with antianxiety stimuli) may not have been able to accomplish, or at least may not have been able to accomplish as quickly. Future research must be conducted to examine this hypothesis in children with ASD, perhaps comparing an “exposure paired with antianxiety stimuli” (i.e.,

counterconditioning) to an “exposure-alone” condition. This is important to examine because using antianxiety stimuli would be considered “distraction” in the CBT literature, and Foa and Kozak (1986) posited that distraction interferes with the activation of fear by disrupting the match between aspects of the stimulus setting and the fear structure. In fact, Kamphuis and Telch (2000) found that fear reduction was hindered by having participants engage in a cognitively demanding distraction task. Although using antianxiety stimuli would be considered distraction or “safety signals” in the CBT literature and would thus be contraindicated for neurotypical adults with anxiety (and for TD children, though to a lesser extent than adults), this may not be as true for children with ASD, who may often lack the cognitive capacity to understand that their anxiety will eventually habituate if they simply remain in the situation for long enough without them having to do anything at all. Of note, in their treatment of anxiety in children with AS, Sofronoff et al. did not use exposure; they did, however, employ cognitive restructuring techniques (“thinking tools”) and use Social Stories to create an “antidote” to anxious thoughts. Thus, although standard exposure is typically thought to be a necessary component in behavioral interventions for reducing anxiety in neurotypical populations, more research needs to be conducted about the most effective treatments for anxiety in children with ASD.

A third limitation of the present study is the lack of a child self-report measure. Given that individuals with ASD often lack awareness of internal states or motivation to report their internal states (Lainhart & Folstein, 1994) and tend to provide less coherent representations of emotional experiences than their TD peers (Losh & Capps, 2006), I decided to rely on parent report measures rather than child report. Although the lack of a child-report measure is a limitation of the present study in attempting to validate the construct of anxiety (particularly the cognitive/affective component of anxiety), Wood and Gadow (2010) noted that, in TD children,

parent-reported symptoms are at least as strongly associated with child diagnostic status for both children and adolescents as are self-reported symptoms (e.g., Wood, Piacentini, Bergman, McCracken, & Barrios, 2002). Wood and Gadow speculated that this may be because many anxiety symptoms are behavioral in nature (e.g., psychosomatic symptoms) and are not restricted to private internal states. Thus, parent-reports may be a suitable proxy for child self-reports in assessing anxiety in children with ASD. However, conversely, in their pilot study of cognitive-behavioral group treatment for anxiety in children with HFA, Reaven et al. (2009) found that, while parent report of anxiety symptoms changed significantly post-treatment, child self-report did not. They noted that this discrepancy between parent report and child self-report raises important questions regarding the use of child self-report as a valid and reliable source of information concerning anxiety symptoms in this population. Their results also suggested that some of the children may have been underreporting their own symptoms of anxiety at pre-treatment. Likewise, in their RCT for children with HFA, Wood et al. (2009) found that child-report MASC scores did not yield a significant effect for treatment group, perhaps because the children's scores at baseline were relatively low on average. Similarly, White and Roberson-Nay (2009) found little agreement between parent and child self-reports of anxiety in a group of children and adolescents with HFA ( $IQ = 92.24 \pm 14.41$ ), and suggested that some children may have been confused by the items or responded randomly. Thus, one important direction for future research is to develop self-report measures of anxiety for children and adolescents with ASD that are easier to understand, and perhaps use pictorial or visual representations of abstract concepts. In general, further work in validating diagnostic interviews, parent-report questionnaires, and self-report questionnaires for children with ASD would be valuable in assessing the cognitive component of anxiety in this population and could help to inform treatment.

A fourth limitation of our study was that, given the inherent lack of experimental control in such an ecologically valid design, there were many instances of equipment malfunction (with the HR monitor, wireless webcam, and laptop computer) and substantial movement artifacts, which resulted in a lack of usable data (or ECG data that was too heavily edited) for the three children, in particular Jon and Sam. Anecdotally, Jon was the child who was the most physically active (e.g., jumping off the couch, rolling around on the floor) and who had the highest amount of movement artifacts. Sam was more stationary in the car, although his seat belt frequently pressed up against the heart rate monitor, causing artifacts and sometimes turning off the monitor. Related to this limitation, one possible influence on HR and RSA that was unaccounted for by our methods was motor movements (consistent with limitations of previous studies, such as Watson et al., 2011). Although Porges et al. (2007) reported that low-intensity motor movements did not affect RSA or IBI in school-age children, they found that more intense activity accompanying physical exercise did impact RSA and IBI. In the present study, although Sam was seated in the car seat with a seatbelt and thus relatively constrained in his physical activity (with the exception of occasionally laying down in the car seat), Jon and Ben were slightly more physically active during the anxiety-provoking conditions as compared with the non-anxiety-provoking conditions; Jon often stood up and attempted to leave the room when others were singing happy birthday (in contrast to sitting in a chair watching TV or playing the keyboard during the low-anxiety condition) and Ben generally stood up by the front door when his parents left the house (in contrast to sitting in a chair while playing his handheld video game in the low-anxiety condition). However, as these movements did not appear to constitute high-intensity motor activity, it is unlikely that motor movements had a major impact on our results. Nevertheless, perhaps future studies using a similar design should also include a physical

exertion task (as in Goodwin et al., 2006) and/or use an accelerometer to control for motor movements (as in Goodwin et al., 2006; Goodwin, Intille, Albinali, & Velicer, 2011). In addition, it should be noted that I analyzed the mean RSA across consecutive 30-s intervals, whereas a recent study by Watson et al. (2011) analyzed the mean RSA across three one-minute intervals, citing previous evidence that continuous short samples of heart activity are less reliable than either longer samples or multiple short samples separated by intervals of time (Richards, 1995). They noted, however, that the method of using longer samples (longer than one minute) was not feasible due to the ages and functioning levels of their participants, which was also the case in our study. Overall, due to the amount of missing physiological data, I acknowledge the possibility that our results might be impacted by patterns of missing data.

Fifth, it is unclear whether the interventions in the present study were successful in making longterm changes in the children's social environment and in their anxiety, as no follow-up data were collected. This is particularly important in terms of assessing the longterm effects of anti-anxiety stimuli, which may have functioned as distractions. After all, Craske, Street, and Barlow (1989) found that distracted exposure led to significantly less fear reduction at a 6-month follow-up compared with a focused exposure condition. That said, given the multicomponent nature of the present study, we would not be able to attribute a sustained improvement at follow-up, or a lack of a sustained improvement, to any one intervention component (such as the use of anti-anxiety stimuli in counterconditioning).

Finally, although the present study explored physiological arousal accompanying problem behavior and, anecdotally, much of the anxious behavior displayed by the children in our study appeared to precede problem behavior (often escalating into problem behavior), I did not formally analyze the patterns of behavior or physiological arousal preceding problem

behavior using a method such as time series analysis, sequential analysis, or multilevel modeling. Future research should be conducted using heart activity and other measures of physiological arousal (e.g., skin conductance) to analyze patterns of physiological arousal and behavior preceding problem behavior. A long-term goal is to use measures of physiological arousal in clinical settings to inform clinicians when a child is becoming aroused or anxious, the types of situations or stimuli that make the child aroused/anxious, and the nature of the child's physiological state prior to engaging in problem behavior. This information can ultimately be used to facilitate assessment in terms of identifying physiological precursors of problem behavior and, ideally, subtle behavioral precursors that are paired with the physiological precursors. This could also be used to facilitate treatment planning in children with ASD and DD in that, if the child is becoming physiologically aroused, the clinician could use an antecedent-based or setting-event-based strategy such as reducing demands, introducing discriminative stimuli for non-problem behavior, or prompting the use of a replacement skill (e.g., functional communication skills, relaxation exercises or another calming activity) to prevent problem behavior from occurring.

### **Concluding Comment**

As Friman, Hayes, and Wilson (1998) noted, "Despite its apparent technical opacity, the term *anxiety* does have well-established functional value for virtually all of the social-verbal community except behavior analysts." Thus, although behavior analysts historically use terms such as "avoidance behavior" (e.g., Rapp et al., 2005) in children with ASD and ID who cannot verbally express their fear or anxiety, I would argue that there is theoretical value and clinical value in applying the term "anxiety" to this population. Interventions to treat anxiety in TD children and interventions to treat avoidance behavior in children with ASD have largely

developed independently from one another, just as interventions for parents of children with Disruptive Behavior Disorders (DBDs) and ASD have largely developed independently from each other (Brookman-Frazee, Stahmer, Baker-Ericzén, & Tsai, 2006). Although the interventions for both populations share similar roots in operant and classical conditioning, the research regarding treatment efficacy has been reported in two separate literature traditions – mental health or clinical psychology literature for TD children with anxiety disorders or DBDs (e.g., *Journal of Consulting and Clinical Psychology*) and developmental disabilities literature for children with ASD (e.g., *Journal of Autism and Developmental Disabilities*) – which has resulted in little cross-fertilization between the two bodies of research (Brookman-Frazee et al., 2006). Beginning to use some of the same terminology, such as applying the term “anxiety” to children with ASD, may help facilitate cross-fertilization and the transfer of knowledge between these two bodies of research. As Brookman-Frazee et al. noted, traditional dissemination models in which one treatment protocol is prescribed for a specific diagnostic group may not be helpful in real world community practice given that, in clinical settings, providers often treat children and families with multiple needs, which may be best treated using a combination of intervention strategies. Given that children with ASD who present with anxiety and TD children with anxiety may share similar clinical characteristics when presenting for treatment in the “real world,” and may actually overlap in real world settings, future research may benefit from integrating the knowledge gained in these two bodies of literature (see Brookman-Frazee et al., 2006).

In sum, problem behavior that is traditionally viewed as noncompliant or disobedient behavior may in fact be a child’s way of reducing or alleviating anxiety rather than escaping a demand, gaining comfort or reassurance rather than gaining attention or tangible objects, or self-soothing rather than simply seeking sensory reinforcement. In short, problem behavior may often

serve to reduce, escape, or avoid feelings of anxiety or anxiety-provoking situations.

Recognizing anxiety in children with ASD can help parents, teachers, clinicians, and researchers to regard problem behavior as stemming from anxiety (i.e., escaping anxiety) rather than noncompliance, disobedience, or defiance (i.e., escaping demand). More important, identifying anxiety in children with ASD can help to inform treatment in terms of preventing problem behavior before it occurs, and reducing anxiety or teaching the child to cope with anxiety when it does occur.



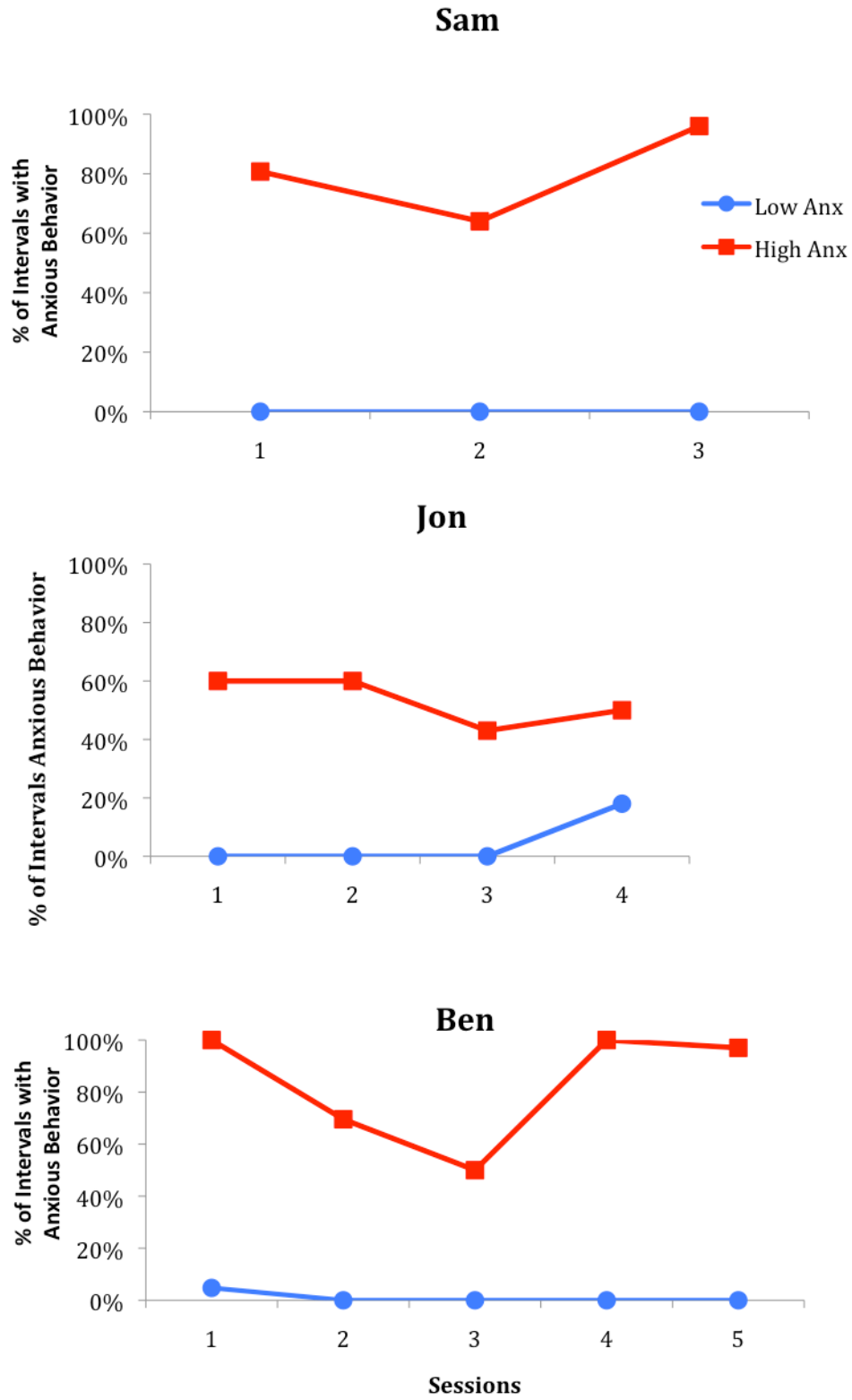


Figure 1. Percent of 10-s intervals containing anxious behavior in High-Anxiety vs. Low-Anxiety contexts, across three, four, and five sessions for Sam, Jon, and Ben respectively.

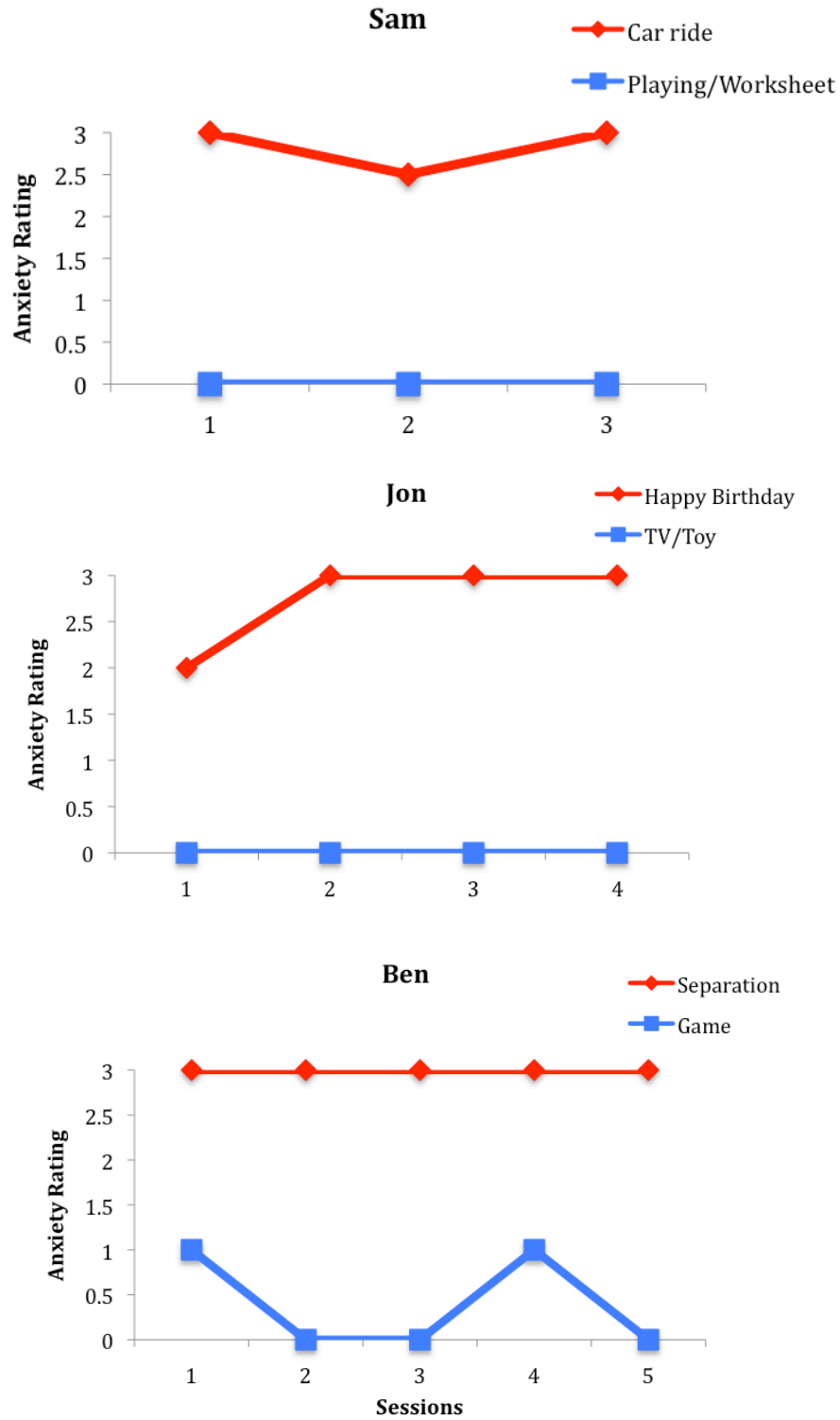


Figure 2. Subjective ratings of anxiety on Likert-type rating scale for High-Anxiety vs. Low Anxiety-Contexts, across 3, 4, and 5 sessions for Sam, Jon, and Ben respectively.

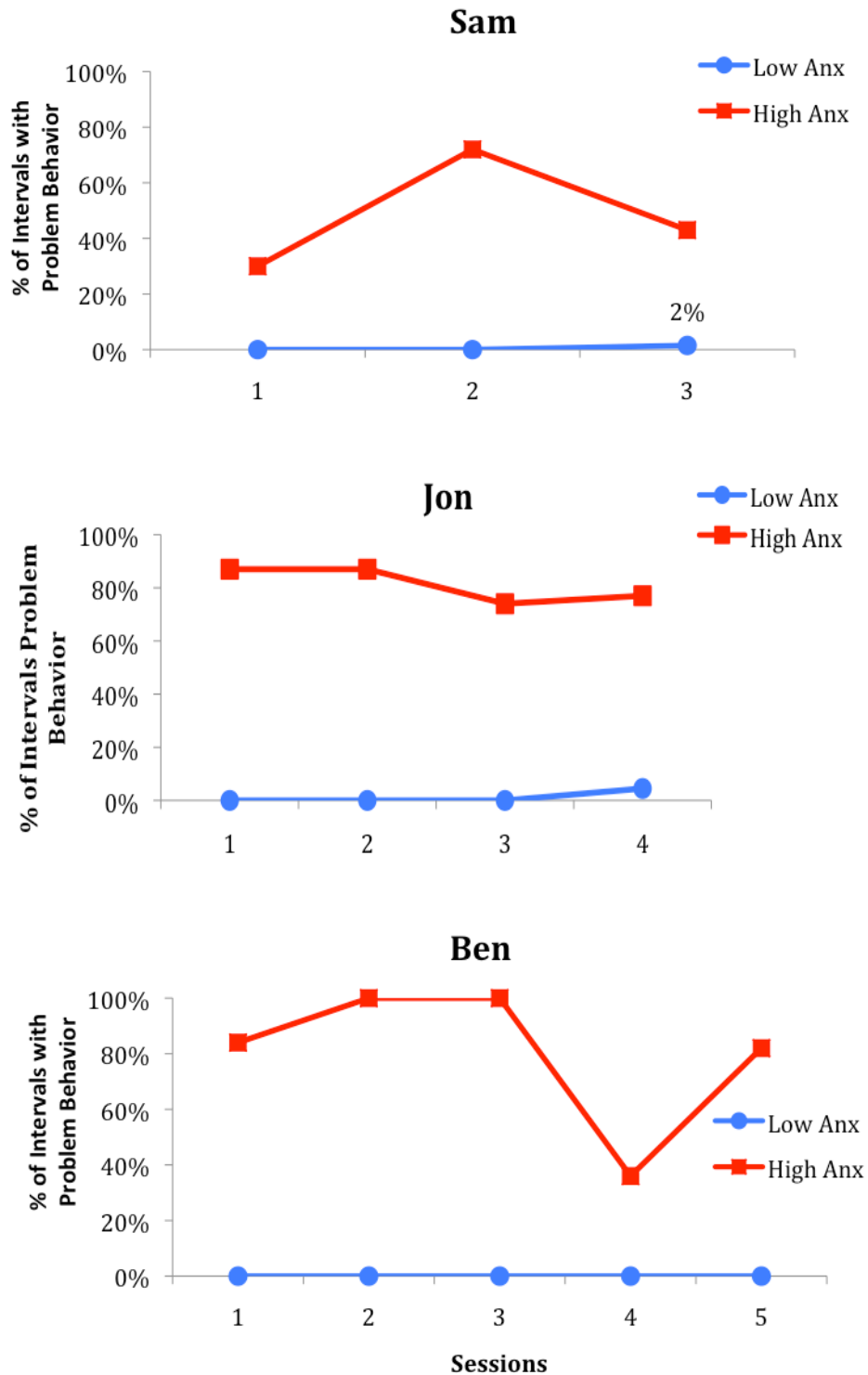


Figure 3. Frequency of problem behavior (percent of 10-s intervals with problem behavior) as a function of level of anxiety (High-Anxiety vs. Low-Anxiety contexts) across 3, 4, and 5 sessions for Sam, Jon, and Ben respectively.

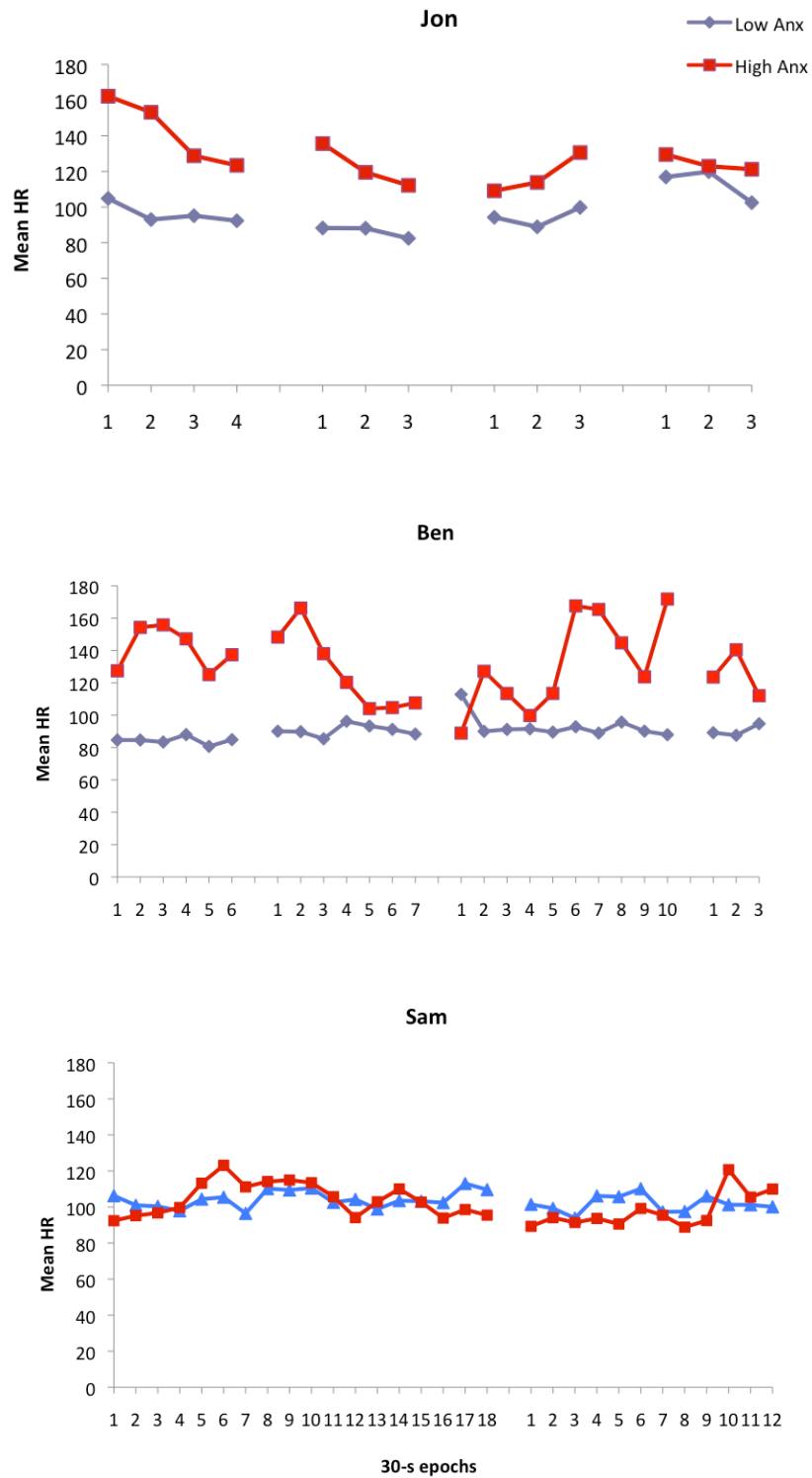


Figure 4. Mean heart rate (HR) for each 30-second period within a session, across four sessions for Jon and Ben, and across two sessions for Sam.

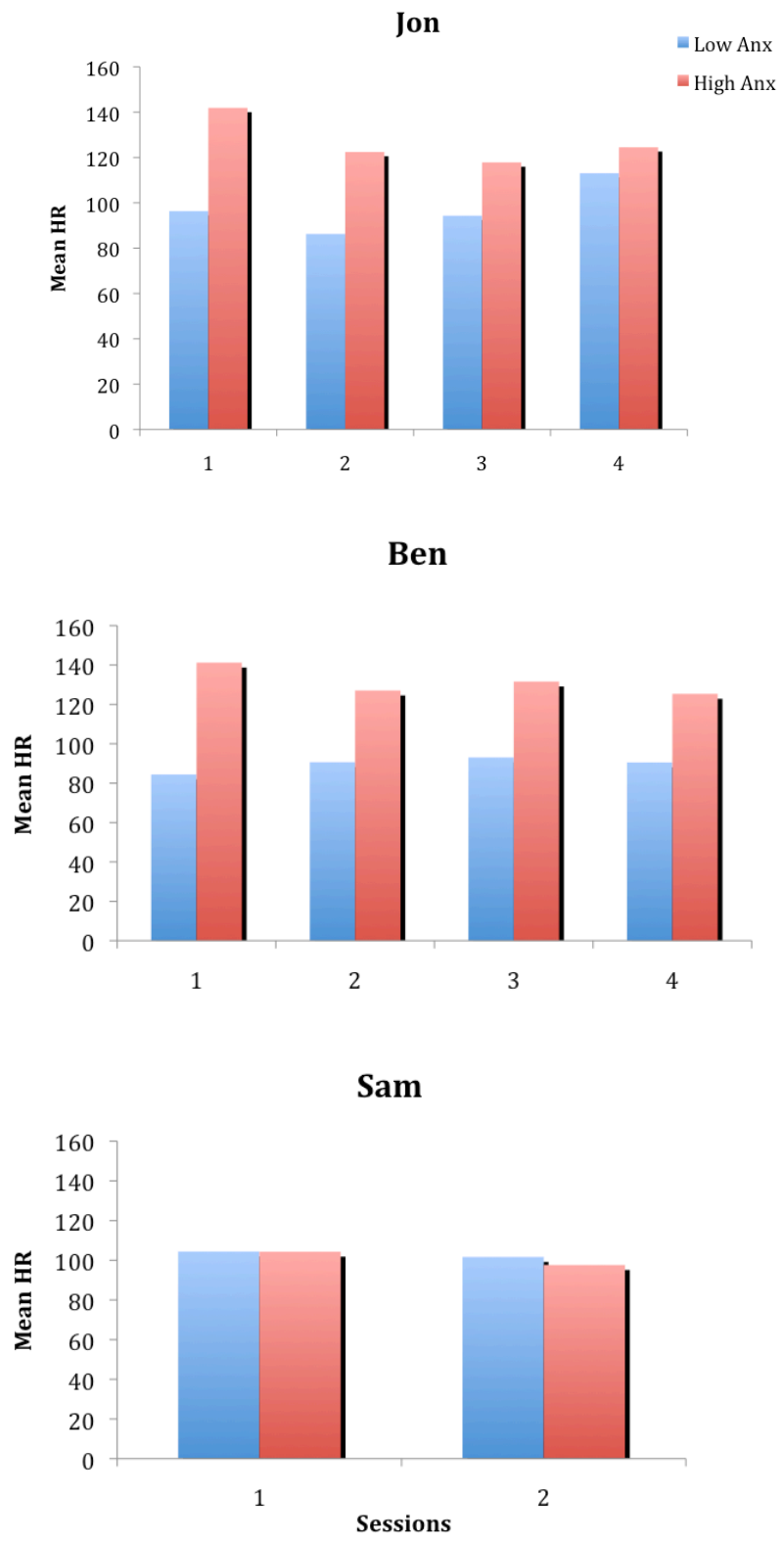


Figure 5. Mean heart rate (HR) for each session (average of 30-second epochs for each session).

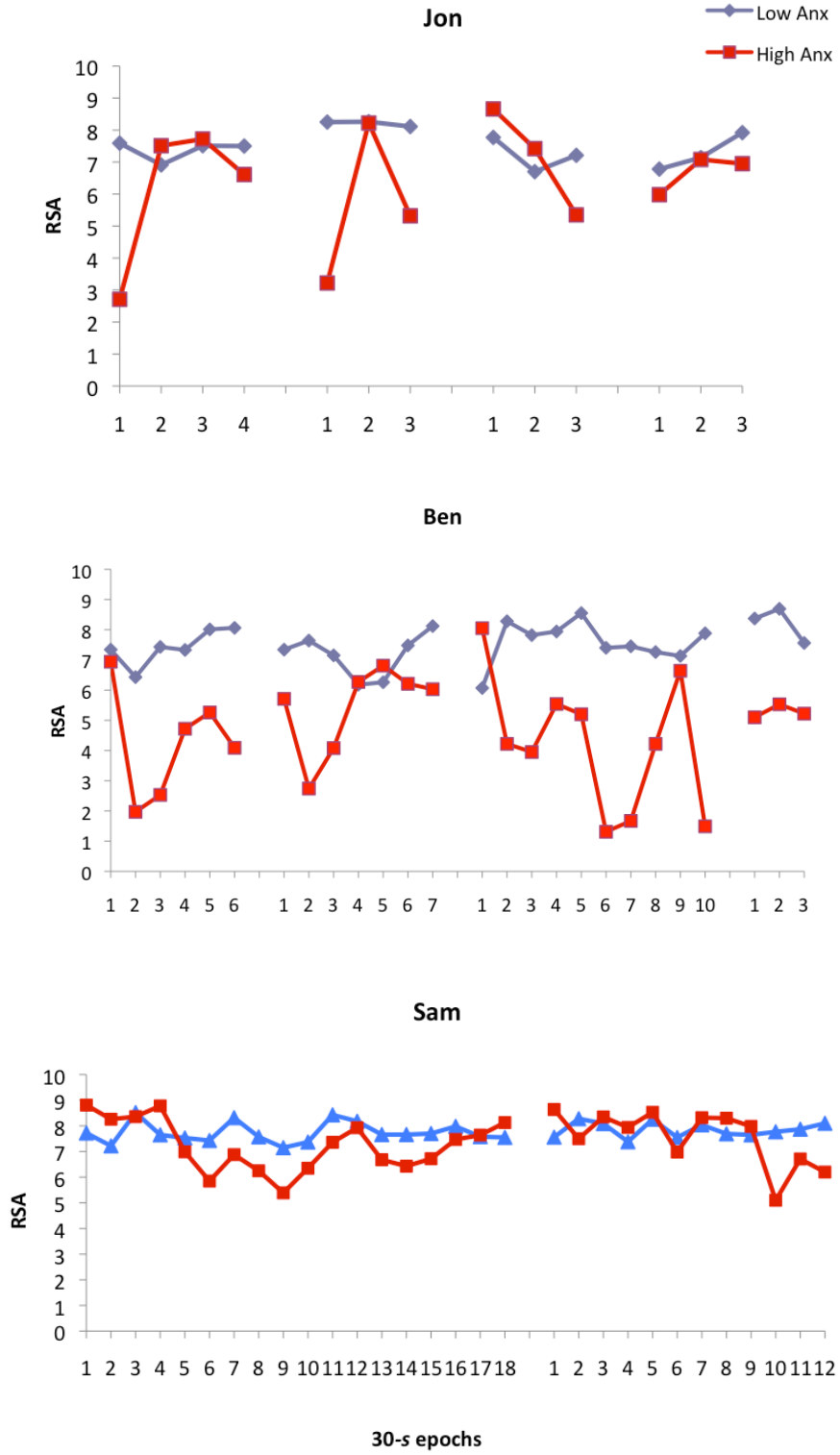


Figure 6. Mean respiratory sinus arrhythmia (RSA) for each 30-second period within a session, across four sessions for Jon and Ben, and across two sessions for Sam.

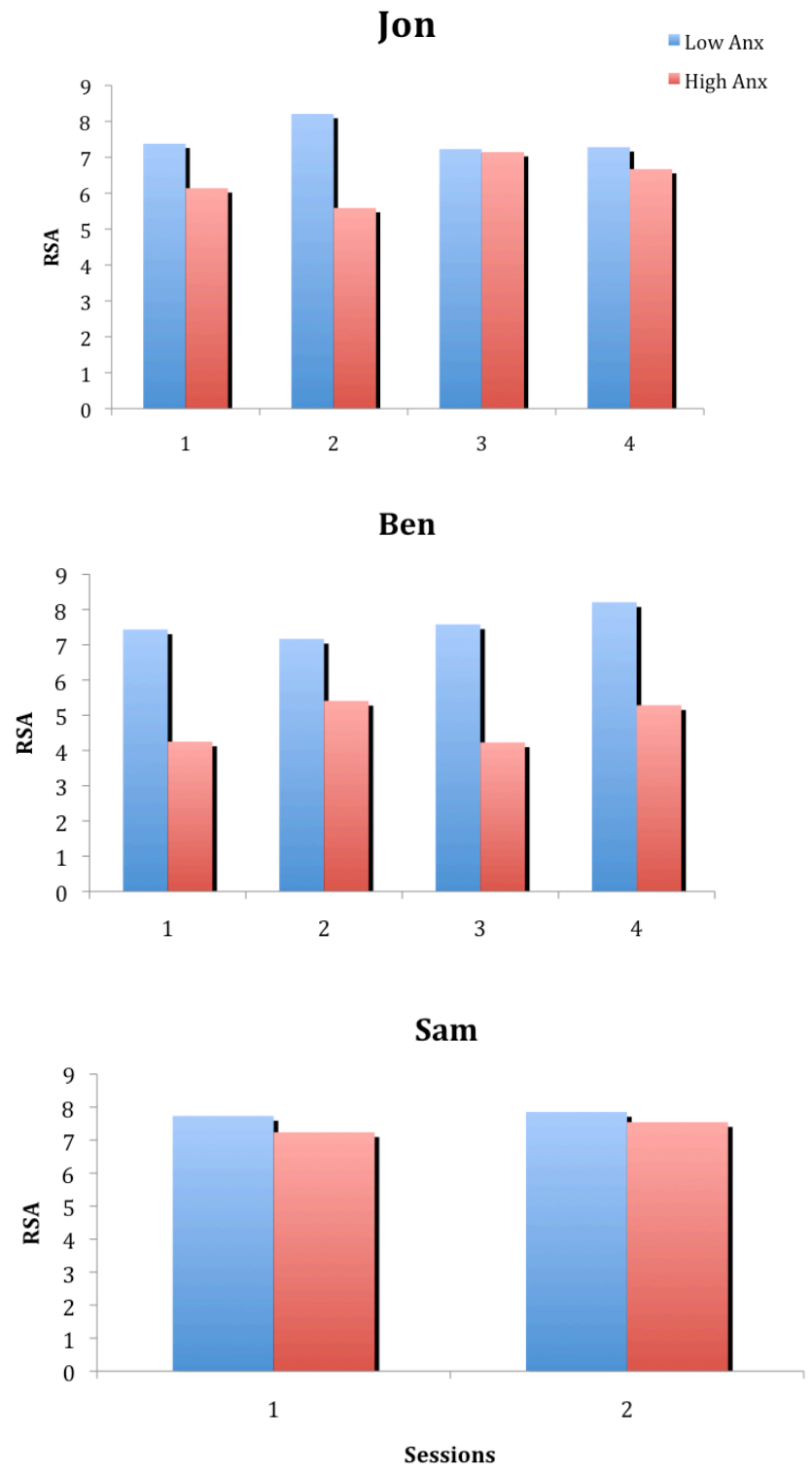


Figure 7. Mean respiratory sinus arrhythmia (RSA) for each session (average of 30-second epochs for each session).

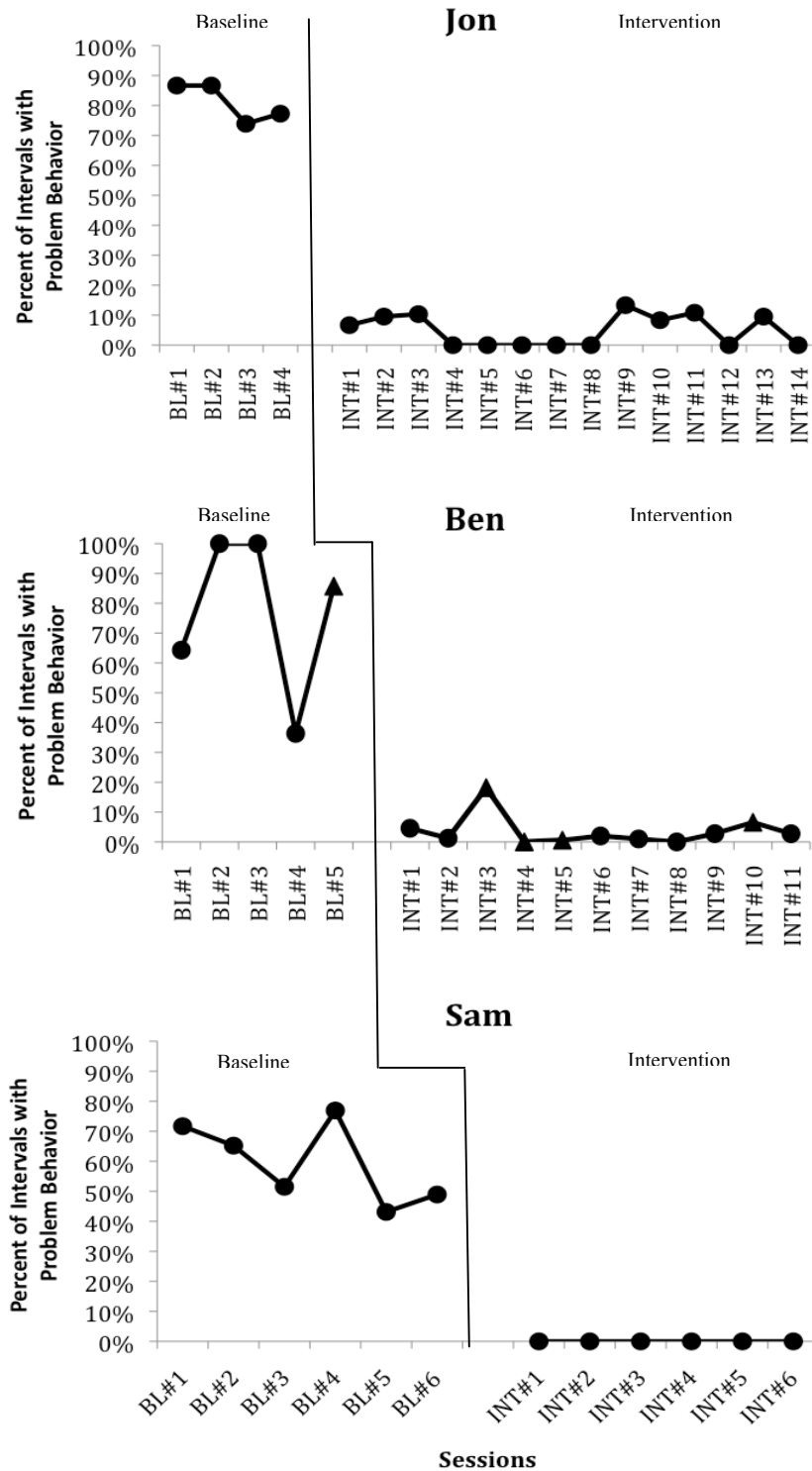


Figure 8. Frequency of problem behavior (percent of 10-s intervals with problem behavior) for the three participants during baseline (BL) and intervention (INT) phases. The ▲ symbol for Ben denotes the times that he stayed home alone with someone else (e.g., his grandparents, his uncle) other than the researcher, while the researcher left the house with his parents.



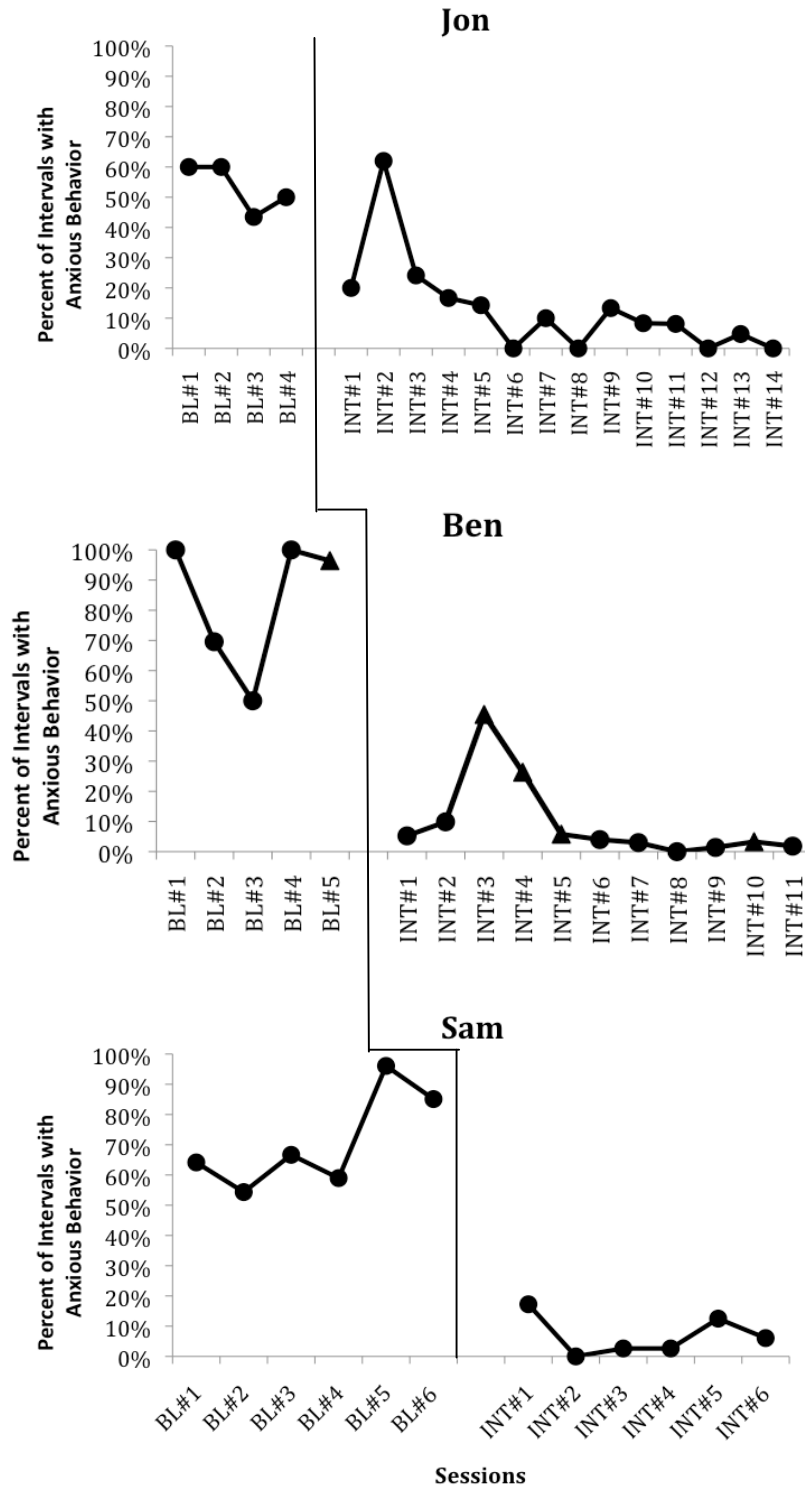


Figure 9. Frequency of anxious behavior (percent of 10-s intervals with anxious behavior) for the three participants during baseline (BL) and intervention (INT) phases. The ▲ symbol for Ben denotes the times that he stayed home alone with someone else (e.g., his grandparents, his uncle) other than the researcher, while the researcher left the house with his parents.

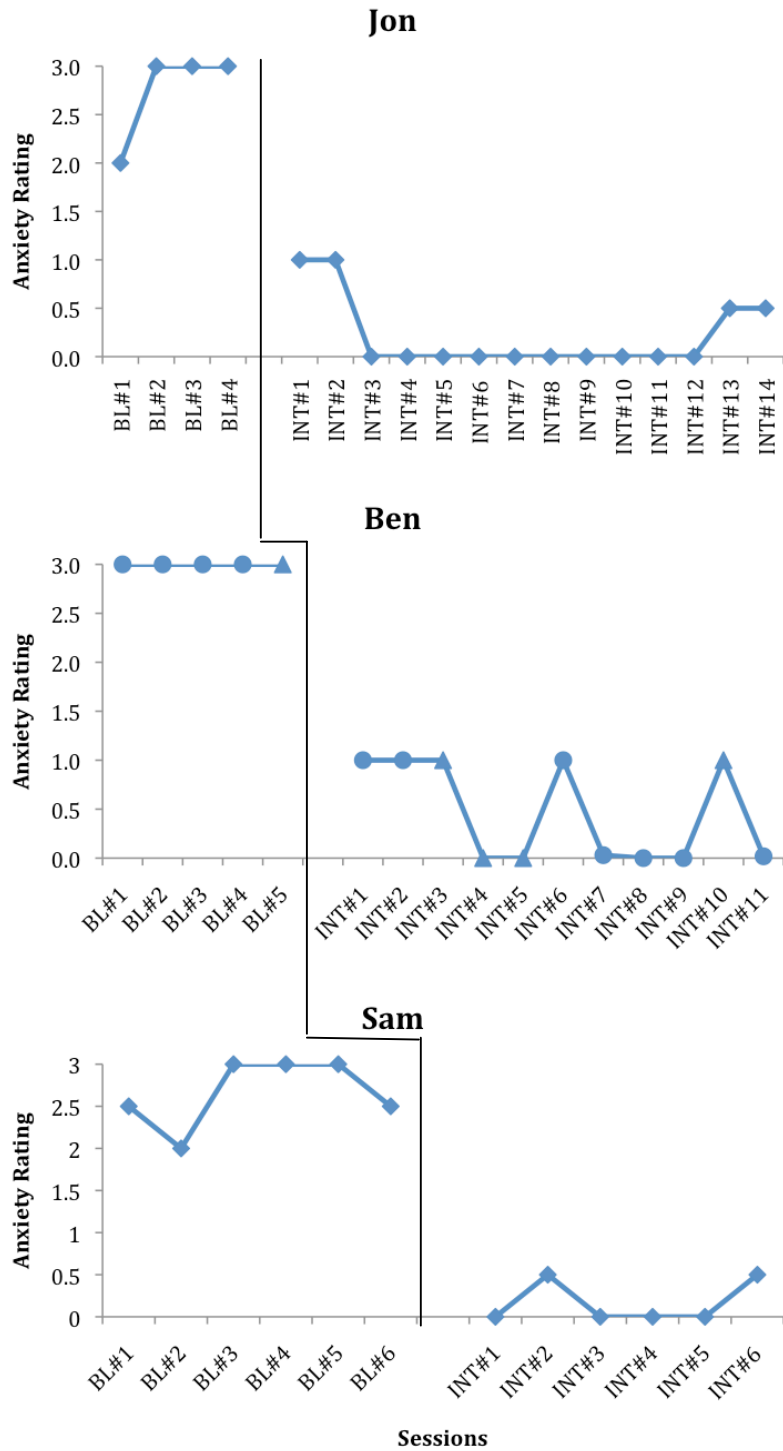


Figure 10. Subjective ratings of anxiety on Likert-type rating scale for baseline (BL) and intervention (INT) phases. Raters were blind to whether session was a BL or INT session. The ▲ symbol for Ben denotes the times that he stayed home alone with someone else (e.g., his grandparents, his uncle) other than the researcher, while the researcher left the house with his parents.

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Appendix A

Informant Screening Questionnaire

Date: \_\_\_\_\_ Target Individual: \_\_\_\_\_ Informant: \_\_\_\_\_

**1. Approximately how often does the target individual (e.g., your child or student) appear to experience fear, anxiety, or stress?**

Never \_\_\_\_\_ Once every few years \_\_\_\_\_ Once per year \_\_\_\_\_  
 Several times per year \_\_\_\_\_ Several times per month \_\_\_\_\_ Once per month \_\_\_\_\_  
 Once per week \_\_\_\_\_ Several times per week \_\_\_\_\_ Once per day \_\_\_\_\_  
 Several times per day \_\_\_\_\_

**2. What types of problem behavior (e.g., aggression, self-injury, tantrums, property destruction, perseveration) does the target individual typically display when he/she appears to be anxious/afraid/nervous/stressed?**

**3. Are there other types of problem behavior (e.g., aggression, self-injury, tantrums, and/or property destruction) that the target individual typically displays when he/she does NOT appear to be anxious/afraid/nervous/stressed? If so, what?**

**4. What specific modes of communication does the individual typically use to indicate anxiety/fear/stress?**

Verbal \_\_\_\_\_ Sign language or gestures \_\_\_\_\_ Picture communication system \_\_\_\_\_  
 Other: \_\_\_\_\_

**5. How is it determined that the individual is experiencing anxiety/fear/stress? In other words, what specific behaviors or symptoms lead you to think that the individual is experiencing anxiety/fear/stress?**

**5a. What physical symptoms are associated with anxiety?**

Behaviors	Engages in this behavior when anxious/afraid	Engages in this behavior when NOT anxious/afraid	Does not engage in this behavior at all
Rigidity or tenseness (visible muscle tension or stiffness)			
Hyperventilating, heavy breathing, rapid breathing, sharp intake of breath, gasping			
Sweating or perspiration			
Flushed face or neck			
Trembling or shaking			
Lips clenched			
Lips quivering			
Other behavior:			



Other behavior:			

**5b. What behavioral patterns are associated with anxiety?**

<b>Behaviors</b>	<b>Engages in this behavior when anxious/afraid</b>	<b>Engages in this behavior when NOT anxious/afraid</b>	<b>Does not engage in this behavior at all</b>
Withdrawal/avoidance			
Cowering			
Pacing			
Freezing			
Fidgeting			
Twitching or jerky/jumpy movements			
Frowning (turning down of mouth)			
Eyebrows raised in upside-down V shape			
Tears			
Rapid clenching and unclenching of fists			
Avoiding eye contact			
Difficulty maintaining eye contact (e.g., eyes rapidly darting back & forth)			
Unusual movements (e.g., eye blinking, twitching, lip licking, head jerking)			
Unusual vocal sounds (e.g., coughing, throat clearing, sniffing, grunting)			
Picking or scratching (e.g., picks nose, skin or other parts of body)			
Hand-wringing			
Teeth-grinding			
Compulsions (i.e., repeats certain acts, words, phrases, sentences, or movements over and over)			
Talking, singing, or vocalizing excessively (more than usual)			
Talking, singing, or vocalizing too loudly (more than usual) or for longer than usual			
High intensity movement			
Escalation (movements or vocalizations increase in volume, speed, or intensity)			
Stuttering or dysfluent speech, such as <i>repetitions</i> (e.g., “I w-want my box,” “I want – I want the box”), <i>interjections</i> (extra sounds, syllables, or words such as “uh,”			

“um,” “well”), <i>pauses/blocks</i> (child stops or pauses for more than 2 seconds between words), or <i>revisions</i> (child stops talking in the middle of a sentence and starts over)			
Other behavior:			
Other behavior:			

**6. Are there certain events, people, times of day, activities, or situations that are reliably associated with anxiety? \_\_Yes \_\_No**

**7. If yes, what types of contexts or situations? \_\_\_\_\_**  
 \_\_\_\_\_

**8. If this individual appears to be anxious, would you avoid exposing him/her to the situations mentioned in question #7? \_\_Yes \_\_No**

**9. If the individual is not anxious, would he or she probably be able to complete the same activity or enter the same situation without difficulty? \_\_Yes \_\_No**

**10. Are there certain events, people, times of day, activities, or situations that are reliably NOT associated with anxiety? \_\_Yes \_\_No**

**11. What, if anything, is done to eliminate or reduce the individual’s anxiety/fear/stress?**  
 \_\_\_\_\_  
 \_\_\_\_\_

**12. What, if anything, is done to control problem behavior associated with anxiety?**  
 \_\_\_\_\_  
 \_\_\_\_\_

Appendix B

Contextual Assessment Inventory for Families (CAI)

ACTIVITIES AND ROUTINES

Please rate how likely it is that your child will show problem behavior in the situations described.  
When completing the ratings, consider your child's problem behavior *over the past year*.

	<b>Not Likely</b>		<b>Somewhat Likely</b>		<b>Very Likely</b>	<b>Don't Know</b>	
9. A preferred activity ends or is no longer possible	1	2	3	4	5	DK	NA
10. Activities or routines that are difficult, frustrating, disliked, or boring	1	2	3	4	5	DK	NA
11. Activity is too long	1	2	3	4	5	DK	NA
12. Activity is too noisy and/or crowded	1	2	3	4	5	DK	NA
13. Having to wait	1	2	3	4	5	DK	NA
14. Medical appointments or medical settings	1	2	3	4	5	DK	NA
15. Changes in routine, or has to deal with new and unfamiliar situations	1	2	3	4	5	DK	NA
16. Transitions between settings or activities	1	2	3	4	5	DK	NA

Are there any other factors related to activities or your child's routine that make it more likely that he or she will show problem behavior?

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Appendix C

**THE STRESS SURVEY SCHEDULE FOR PERSONS WITH  
AUTISM AND DEVELOPMENTAL DISABILITIES  
The Groden Center, Inc.**

Please rate the intensity of the stress reaction to the following events by filling in the appropriate circle:

	None to Mild	Mild to Moderate	Moderate	Moderate to Severe	Severe
1. Receiving a present	1	2	3	4	5
2. Having personal objects or materials out of order	1	2	3	4	5
3. Waiting to talk about desired topic	1	2	3	4	5
4. Having a change in schedule or plans	1	2	3	4	5
5. Being in the vicinity of noise or disruption by others	1	2	3	4	5
6. Waiting for preferred events	1	2	3	4	5
7. Having a cold	1	2	3	4	5
8. Being touched	1	2	3	4	5
9. Having personal objects or materials missing	1	2	3	4	5
10. Having a change in task to a new task with new directions.	1	2	3	4	5
11. Going to the store	1	2	3	4	5
12. Being prevented from completing a ritual	1	2	3	4	5
13. Having a change in environment from comfortable to uncomfortable	1	2	3	4	5
14. Being prevented from carrying out a ritual	1	2	3	4	5
15. Moving from one location to the next	1	2	3	4	5
16. Playing with others	1	2	3	4	5
17. Having a change in environment from familiar to unfamiliar	1	2	3	4	5
18. Receiving activity reinforcement	1	2	3	4	5
19. Having something marked as correct	1	2	3	4	5
20. Being in the vicinity of bright lights	1	2	3	4	5
21. Following a diet	1	2	3	4	5
22. Having unstructured time.	1	2	3	4	5
23. Being allowed to attend a party or favored event.	1	2	3	4	5
24. Receiving a reprimand	1	2	3	4	5
25. Transitioning from preferred to non-preferred activity	1	2	3	4	5
26. Being told "no"	1	2	3	4	5
27. Receiving criticism	1	2	3	4	5
28. Having something marked incorrect	1	2	3	4	5
29. Being interrupted while engaging in a ritual	1	2	3	4	5
30. Receiving hugs and affection	1	2	3	4	5

31. Having to engage in not-liked activity	1	2	3	4	5
32. Waiting on line	1	2	3	4	5
33. Being unable to communicate needs	1	2	3	4	5
34. Waiting at a restaurant	1	2	3	4	5
35. Going home (from school, to visit parents)	1	2	3	4	5
36. Waiting for transportation	1	2	3	4	5
37. Being unable to assert oneself with others	1	2	3	4	5
38. Needing to ask for help	1	2	3	4	5
39. Participating in group activity	1	2	3	4	5
40. Having a change in staff, teacher or supervisor	1	2	3	4	5
41. Losing at a game	1	2	3	4	5
42. Waiting for reinforcement	1	2	3	4	5
43. Feeling crowded	1	2	3	4	5
44. Someone else making a mistake	1	2	3	4	5
45. Receiving tangible reinforcement	1	2	3	4	5
46. Waiting for food	1	2	3	4	5
47. Waiting for routine to begin	1	2	3	4	5
48. Having a conversation	1	2	3	4	5
49. Receiving verbal reinforcement	1	2	3	4	5

<b>FEARS</b>	<b>None to Mild</b>	<b>Mild to Moderate</b>	<b>Moderate</b>	<b>Moderate to Severe</b>	<b>Severe</b>
1. Fears of animals	1	2	3	4	5
2. Fear of water (e.g., pool, lake, ocean, etc.)	1	2	3	4	5
3. Fear of crowds	1	2	3	4	5
4. Fear of closed spaces	1	2	3	4	5
5. Fear of the dark	1	2	3	4	5
6. Fear of being left alone					

<b>LIFE STRESSORS</b>	<b>None to Mild</b>	<b>Mild to Moderate</b>	<b>Moderate</b>	<b>Moderate to Severe</b>	<b>Severe</b>
1. Going to the doctor or dentist	1	2	3	4	5
2. Having seizures	1	2	3	4	5
3. Having a new sibling	1	2	3	4	5
4. Moving to a new house	1	2	3	4	5
5. Moving to a new school	1	2	3	4	5
6. Having parents get divorced	1	2	3	4	5
7. Having a parent re-marry	1	2	3	4	5

**Please list any other stressors on the lines below:**

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**Which do you consider the most significant stressors of those you have identified? Why?**

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Appendix D

<b>Physical Symptoms Associated with Anxiety</b>	<b>Present</b>	<b>Absent</b>
Rigidity or tenseness (visible muscle tension or stiffness)		
Hyperventilating, heavy breathing, rapid breathing, gasping		
Sweating or perspiration		
Flushed face or neck		
Trembling or shaking		
Lips clenched		
Lips quivering		
Other:		
Other:		
Other:		
<b>Behaviors associated with anxiety</b>	<b>Present</b>	<b>Absent</b>
Withdrawal/avoidance		
Cowering		
Pacing		
Freezing		
Fidgeting		
Twitching or jerky/jumpy movements		
Frowning (turning down of mouth)		
Eyebrows raised in inverted V shape		
Tears		
Rapid clenching and unclenching of fists		
Avoiding eye contact		
Difficulty maintaining eye contact (e.g., eyes rapidly darting back & forth)		
Unusual movements (e.g., eye blinking, twitching, lip licking, head jerking)		
Unusual vocal sounds (e.g., coughing, throat clearing, sniffing, grunting)		
Picking or scratching (e.g., picks nose, skin or other parts of body)		
Hand-wringing		
Teeth-grinding		
Compulsions (i.e., repeats certain acts, words, phrases, sentences, or movements over and over)		
Talking, singing, or vocalizing excessively (more than usual)		
Talking, singing, or vocalizing too loudly (more than usual) or for longer than usual		
Escalation (movements or vocalizations increase in volume, speed, or intensity)		
Stuttering or dysfluent speech, such as repetitions, interjections, pauses/blocks, or revisions		
Reassurance-seeking		
Other behavior:		
Other behavior:		
Other behavior:		

## Appendix E

### Original Scales from which Behavioral Markers of Anxiety were Derived

#### Items from Form II: Cues for Tension & Anxiety Survey Schedule (CTASS) (Cautela, 1977)

1. Tense/rigid
2. Sweating
3. Face flushed or warm
4. Skin cool and damp
5. Tremble or shake
6. Tight grip
7. Scratch a certain part of body
8. Biting nails
9. Grinding teeth
10. Trouble with speech

#### Affex Facial Coding System for Negative Facial Expressions (Izard, Dougherty, & Hembree, 1989)

- *Sadness*: the inner corners of the eyebrows were raised, bulging or furrowing of the center of the forehead, the nasal root was narrowed, the corners of the mouth were drawn downward and outward, the chin may have pushed up the center of the lower lip
- *Anger*: eyebrows were sharply lowered and drawn together, vertical furrows or bulging between the eyebrows, the nasal root was narrowed, the mouth was open and angular or squarish in shape
- *Disgust*: the eyebrows were sharply lowered and drawn together, there were vertical furrows or bulging between the brows, the nasal root was broadened and/or bulged, the mouth was open and angular with the upper lip pulled up, the tongue forward beyond the gum line, the nasal bridge was furrowed, or there was a lumpy ridge running diagonally upward from the nasolabial fold
- *Contempt*: one eyebrow was raised higher than the other; the upper lip was raised on one side; the mouth corners may have been compressed against the teeth on one or both sides, causing the lower cheek to bulge; and dimpling may have been present
- *Fear*: the eyebrows were raised and drawn together in a straight or normal shape, short transverse furrows or thickening in the mid-region of the forehead were present, the nasal root was narrowed, the eye fissure was widened with the upper lid raised, the whites of the eyes showed more than normal, and the mouth was opened and tense, with the corners retracted straight back
- *Physical Distress or Pain*: the eyebrows were sharply lowered and drawn together, there were vertical furrows or bulging between the brows, the nasal root was broadened and bulged, the eyes were tightly closed, and the mouth was open in an angular or squarish shape

Potential indicators of social anxiety in females with fragile X syndrome  
(Lesniak-Karpiak, Mazzocco, & Ross, 2003)

- 1) Duration of silence, defined as the total time of non-interaction during the 105-second period of target role play
- 2) RT to first utterance: time to first utterance, defined as the amount of time elapsed after the examiner gave signal to initiate conversation until the participant first produced an utterance
- 3) Number of pauses, which reflected the frequency of breaks in conversations flow during the role play
- 4) Eye contact avoidance: defined as breaking eye contact with the examiner or by complete avoidance of eye contact during an interval
- 5) Rigidity: rigid body posture, defined as rigid or tense sitting with minimal changes in body positions
- 6) Fidgetiness: fidgeting, defined as frequent or abrupt changes in bodily position such as rocking or leaning in different directions
- 7) Wringing hands, which reflected repeated movements of the upper extremities in a rapid or jerky manner
- 8) Facial movements (such as jaw clenching, biting lips, or scowling) that reflected discomfort

Behaviors Indicative of Social Anxiety in Individuals with Cornelia de Lange Syndrome  
(Richards, Moss, O’Farrell, Kaur, & Oliver, 2009)

Behavior	Operational definition
Eye contact	Participant looks up/or at the examiner and fixates on the examiner’s eyes or face
Participant-communication	Any verbal communication or use of formal signs directed towards the examiner. This includes prompting, offering information and response to a question
Moving of hands	Moving of hands to face, head, or another part of the body. For example scratching or touching face, hair, arm, which has no obvious function. Excluding any forms of self-injury, or any communicative gestures

CBCL/TRF & CSI items as possible behavioural equivalents of anxiety  
(Sullivan, Hooper, & Hatton, 2007)

Anxiety Continuum Behaviours Domain

- Shows excessive fear to specific objects or situations (animals, heights, storms, insects, etc.) (from CSI)
- Cannot get distressing thoughts out of his or her mind (worries about germs or doing things perfectly, etc.) (from CSI)
- Feels compelled to perform unusual habits (hand washing, checking locks, repeating things a set number of times) (from CSI)



- Has experienced an extremely upsetting event and continues to be bothered by it (from CSI)
- Cannot get his or her mind off certain thoughts; obsessions (from CBCL/TRF)
- Repeats certain acts over and over; compulsions (from CBCL/TRF)

Behavioural Dysregulation Domain

- Talks excessively (from CSI)
- Does unusual movements for no apparent reason (eye blinking, twitching, lip licking, head jerking, etc.) (from CSI)
- Makes vocal sounds for no apparent reason (coughing, throat clearing, sniffing, grunting, etc.) (CSI)
- Bites fingernails (CBCL/TRF)
- Nervous movements or twitching (CBCL/TRF)
- Picks nose, skin or other parts of body (CBCL/TRF)
- Talks too much (CBCL/TRF)
- Nightmares (CBCL)
- Trouble sleeping (CBCL)
- Sleeps less than most kids (CBCL)
- Sleeps more than most kids (CBCL)

Anxiety Depression And Mood Scale (ADAMS) for Individuals with Developmental Disabilities  
(Esbensen, Rojahn, Aman, & Ruedrich, 2003)

General Anxiety Items

1. Nervous
3. Does not relax or settle down
7. Tense
11. Worried
15. Anxious
24. Experiences panic attacks
26. Trembles when frightening situations are not present

Behavioral Relaxation Scale (BRS)  
(Poppen, 1998)

*Behavioral Relaxation Scale (BRS) scoring criteria. One-minute intervals were divided so that the first 30 seconds of each minute was used to count the breathing rate, and the next 15 second period was used to observe the other nine behaviours. The last 15 seconds were then utilized to record on the data sheet.*

- (1) *Breathing* - Rate that is lower than that observed in baseline.
- (2) *Quiet* - No vocalizations.
- (3) *Body* - No trunk movement.
- (4) *Head* - In midline with the body.
- (5) *Eyes* - Closed with smooth eyelids.
- (6) *Mouth* - Lips parted in the centre.
- (7) *Throat* - No movement such as swallowing or vocalizations.
- (8) *Shoulders* - Sloped and even with no movement.
- (9) *Hands* - Curled in a 'clawlike' fashion, with fingers spread slightly apart.
- (10) *Feet* - Pointed away from each other at a 90 ° angle.

From Koegel et al. (2004)

<b>Comfortable</b>	<b>Mild anxiety</b>	<b>High anxiety</b>	<b>Intolerable</b>
<p>Absence of any anxiety relating to the stimulus.</p> <p>The child appeared to be relaxed, engaged happily in typical play and interactions, and unaffected by the sight or sound of the stimulus</p>	<p>Engagement in behaviors such as delaying attention to a game or activity, short whining or whimpering, and brief periods of the child covering his ears with his hands when the stimulus was introduced.</p> <p>These behaviors briefly delayed but did not interfere with the activity</p>	<p>The child exhibiting behaviors such as slowly moving away from the sight or sound of the stimulus, whining, or covering his or her ears to the extent that the child could not participate, play, or interact appropriately</p>	<p>The child exhibiting behaviors that suggested that the sight or sound of the stimulus was a painful experience: running away from the game or activity, crying or screaming, pushing his or her hands forcefully over his or her ears, sweating, shaking, and grinding the teeth.</p>

From Eisenberg, McCreath, and Ahn (1988)

Indicators of anxious expressions included grimacing, lips stretched back, tensing of lower eyelid, nervous mouthing, furrowing of brow and eye areas, eyebrows brought together somewhat, refusal to watch the film, and looking questioningly and nervously at experimenter

From Jackson and King (1982)

Child's phobic reaction consisted of pupil dilation, trembling, increased muscle tonus, screaming, crying, tantrums, and hyperventilation and flight.

From Chaplin, Cole, and Zahn-Waxler (2005)

Anxiety cues were mouth retracted; strained voice; or raised, stiff shoulders.

From Luiselli (1978)

When asked to board the bus, the child cried frantically, fell on the sidewalk, tantrumed excessively, and attempted to run in the house. When his mother lifted him onto the bus and rode with him to school, the child screamed and cried throughout the ride and hyperventilated.

From Harrigan and Dennis M. O'Connell (1996)

Muscle movements involved in fear include raising the eyebrows and drawing them together, and/or stretching the lips horizontally so that the lips form a rectangular mouth shape (Ekman & Friesen, 1975, 1978). In more extreme states of fear, a raised, tensed upper eyelid which widens the eye also is displayed (Ekman & Friesen, 1975, 1978). Partial fear actions involved only one part of the face (eye or mouth region) or less intense displays of fear facial actions than those displayed at the time the anxiety-producing incident actually occurred. Eye blinks, which have been shown to be related to anxiety and stress, also were recorded.

Appendix F

**Anxiety Rating Scale**

Please rate this child's anxiety by choosing the number that best described his state of anxiety in the activity you just observed.

<p><b>0</b> <b>No anxiety</b> <b>(i.e., comfortable)</b></p>	<p><b>1</b> <b>Mild anxiety</b></p>	<p><b>2</b> <b>Moderate anxiety</b></p>	<p><b>3</b> <b>High anxiety</b></p>
<p>Absence of any apparent anxiety relating to the stimulus or situation.</p> <p>The child appeared to be relaxed, engaged happily in typical play and interactions, and unaffected by the presence of the stimulus or situation.</p>	<p>The child displayed symptoms of anxiety – such as rigidity or tenseness (visible muscle tension), heavy or rapid breathing, freezing, flushed face, and trembling – to a <b>mild</b> degree, but these symptoms did not significantly interfere with the activity.</p> <p>The child <b>occasionally</b> displayed an anxious or fearful facial expression (e.g., lips stretched back, tensing of lower eyelid, furrowing of brow and eye areas, eyebrows brought together somewhat) or mild apprehension expression (e.g., eyebrow somewhat raised and pulled together in an inverted V shape)</p>	<p>The child displayed symptoms of anxiety – such as pulling/pushing away from the stimulus, cling to parent, cowering, rigidity or tenseness (visible muscle tension), heavy or rapid breathing, freezing, flushed face, trembling or shaking, tearfulness (eyes tearing or watering) – to a <b>moderate</b> degree, to the extent that he could <b>rarely</b> participate, play, or interact appropriately.</p> <p>The child <b>frequently</b> displayed an anxious or fearful facial expression (e.g., eyebrows raised and drawn together in an inverted V shape, wide open eyes, stretched lips).</p>	<p>The child exhibited symptoms of anxiety – such as crying, sobbing, or screaming, running away from the stimulus or situation, pulling/pushing away from the stimulus, clinging to parent, cowering, trembling or shaking, freezing, rigidity or tenseness (visible muscle tension), heavy or rapid breathing, and flushed face – to a <b>high</b> degree, to the extent that he could <b>not</b> participate, play, or interact appropriately.</p> <p>The child <b>constantly</b> displayed an anxious or fearful facial expression (e.g., eyebrows raised and drawn together in an inverted V shape, wide open eyes, stretched lips).</p>

## Appendix G

### **Text of Ben's Social Story (for Separation)**

Everyone feels worried or afraid or anxious sometimes – kids and grownups too. It is okay to feel worried or afraid or anxious sometimes. If a lion is chasing you, it is okay to feel afraid, because your fear will make you run from the lion! There are some things I'm afraid of and some things I am not afraid of. Some other kids are afraid of thunderstorms and lightening. Some kids are afraid of dogs. I am not afraid of thunder and lightening. I am not afraid of dogs. I am a little bit afraid of fireworks. I am very afraid of walking over railroad tracks. I am very afraid of bees. I am very, very afraid when Mom or Dad leave the house and go out without me. I am afraid because I really want to go with them. I really want Mommy and Daddy. But I don't have to be afraid when Mommy or Daddy leaves the house and I stay with Uncle Jimmy or Grandma and Poppi or Grandpa. I don't have to be afraid because Mommy and Daddy will always come back (picture of Mommy and Daddy walking through the front door and hugging Ben). At first, when Mommy and Daddy leave, I will feel scared. Then, after a while longer, I will feel less scared. Then, after a while longer, I won't be scared anymore. I will see that my anxiety goes down after a while, even when Mommy and Daddy are not home. I don't have to be afraid. My anxiety will go away. I will see that I am okay, even if Mommy and Daddy are not home. When Mommy and Daddy are not home, here are some things I can do when I feel afraid. When I feel worried or anxious, I can tell myself, "This is just my anxiety talking. I don't have to be afraid. I am okay. I can beat my anxiety." When Mommy and Daddy are gone, I can tell myself, "Mommy and Daddy always come home. So I won't be afraid." When Mommy and Daddy are gone, I will go do something fun, like play Leapster or Math Desk, or watch a movie, like the Muppet movie. When Mommy and Daddy are gone and I feel afraid, I can breathe nice and slow while I count to ten. This will help me feel calm. So, from now on, when Mommy and Daddy go out, I am going to be brave because I want to fight my anxiety and beat my anxiety. I am going to fight my anxiety and I will win (picture of Kai-Lan saying "We can do it!"). When Mommy and Daddy come home, they will bring me a special prize for being so brave. Mommy and Daddy will be so proud of me for being brave and staying with Uncle Jimmy or Grandma and Poppi or Grandpa.

### **Text of Sam's Social Story (for Left/Right Turns in Car Ride)**

Left turn, left turn, right turn, right. Turns in the morning, turns at night.  
Left turn, left turn, right turn, right. Turns in the morning, turns at night.  
Left turn, left turn, right turn, right. Wet turn, dry turn. Low turn, high turn.  
Front light, back light. Red light, green light.  
Left turn, left turn, right turn, right. How many, many turns can you learn?  
Slow turn, quick turn. Trick turn, sick turn.  
Up turn, down turn. Here comes the clown turn.  
Small turn, big turn. Here comes the pig turn.  
When you drive up to the light, you put on the brake. You turn on a street, and on a lake.  
How many, many turns you make.  
Up in the air turn, over a chair turn. More and more turns. Twenty-four turns.  
Here come more and more... and more turns!  
Left turn, right turn. Turns, turns, turns. Oh, how many, many turns you learn.