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S.I. : ANXIETY IN AUTISM SPECTRUM DISORDERS

Intervention for Anxiety and Problem Behavior in Children with Autism Spectrum Disorder and Intellectual Disability

Lauren J. Moskowitz[1] · Caitlin E. Walsh[2] · Emile Mulder[3] · Darlene Magito McLaughlin[4] · Greg Hajcak[5] · Edward G. Carr[5] · Jennifer R. Zarcone[6]

Abstract There is little research on the functional assessment and treatment of anxiety and related problem behavior in children with autism spectrum disorder (ASD), particularly those with intellectual and developmental disability (IDD). In a recent study, we evaluated a multimethod strategy for assessing anxiety in children with ASD and IDD (Am J Intellect Dev Disabil 118:419–434, 2013). In the present study, we developed treatments for the anxiety and associated problem behavior in these same children. A multiple baseline design was used to evaluate the effectiveness of a multicomponent intervention package, incorporating individualized strategies from Positive Behavior Support and Cognitive Behavioral Therapy. During intervention, all three participants showed substantial decreases in anxiety and problem behavior and significant increases in respiratory sinus arrhythmia in the situations that had previously been identified as anxiety-provoking.

Keywords Autism · Anxiety · Intellectual disability · Applied behavior analysis · Positive Behavior Support · Cognitive Behavioral Therapy · Respiratory sinus arrhythmia

✉ Lauren J. Moskowitz
Moskowil@stjohns.edu

[1] St. John's University, Queens, NY, USA

[2] University of Colorado School of Medicine, Denver, CO, USA

[3] The Shoreline Center, Grand Haven, MI, USA

[4] Positive Behavior Support Consulting, Centerport, NY, USA

[5] Stony Brook University, Stony Brook, NY, USA

[6] Johns Hopkins School of Medicine, Baltimore, MD, USA

Introduction

Recent research suggests that anxiety-related concerns are among the most common presenting problems for children and adolescents with autism spectrum disorders (ASD) (White et al. 2009). Fear and anxiety are reported to be more prevalent in children with ASD than in neurotypical (NT) children (Kim et al. 2000; Leyfer et al. 2006; Weisbrot et al. 2005) as well as those with intellectual and developmental disabilities (IDD) (Bradley et al. 2004; Brereton et al. 2006; Evans et al. 2005). However, despite this research and longstanding clinical concerns regarding anxiety in children with ASD (e.g., Kanner 1943), behavior analytic assessment and intervention research in children with ASD has often neglected the role of anxiety, particularly its potential role in contributing to problem behavior. This is largely because assessing anxiety in children with ASD, particularly those with comorbid IDD, is challenging due to the communication deficits inherent in ASD, the difficulty differentiating symptoms of anxiety disorders from symptoms of ASD, and the idiosyncratic behavioral expression of anxiety in this population (see Hagopian and Jennett 2008, and; White et al. 2009, for a review).

In the traditional clinical psychology literature, anxiety is a multi-component construct involving *affective* states (e.g., subjective fear), *cognitions* (e.g., thoughts, beliefs), *behavioral* patterns (i.e., avoidance), and associated *physiological arousal* (e.g., increased heart rate) (Barlow 2000; Wolpe 1958). Groden et al. (1994) make the point that behavior analytic researchers have been reluctant to use the construct of anxiety to either describe or explain behavior when discussing those with ASD and IDD because, unlike behaviors, cognitions, subjective state, and physiological arousal often cannot be directly observed. This makes the assessment of anxiety in children with ASD more difficult,

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particularly for those who also have IDD, given that they often cannot report or articulate their emotional states, or may express their thoughts or emotions in idiosyncratic ways (Hagopian and Jennett 2008). Thus, in a previous study (Moskowitz et al. 2013), we operationally defined the construct of "anxiety" in three children with ASD and IDD by collecting data on its three components. (1) To measure the behavioral component of anxiety, we collected data on idiosyncratic behaviors that were thought to indicate anxiety for each child. (2) To measure the physiological component of anxiety, we collected data on heart rate (HR) and respiratory sinus arrhythmia (RSA). (3) To index the cognitive/affective or "subjective" component of anxiety, we assessed the context in which the behaviors and physiological arousal occurred (using parent-report and blind observer Likert-type ratings), given 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). The rationale for our multimethod approach was that, although behaviors on their own do not necessarily indicate anxiety (e.g., a child may cry because he is feeling afraid, sad, angry, in pain, or ill), just as physiological arousal on its own does not necessarily indicate anxiety, when multiple sources are taken together, converging data may support the idea that the behavior and/or arousal is indicative of anxiety. Once the presence of anxiety has been identified using a multi-method assessment, a treatment can be developed to address the anxiety. This was the focus in the present study.

Research has shown that biological factors such as pain, illness, and fatigue can function as setting events for problem behavior in individuals with ASD and IDD (Carr and Smith 1995; Carr et al. 2003; Smith et al. 2016). In the presence of these factors, problem behaviors have been shown to be more likely to occur. Given these findings, it seems plausible that anxiety might serve as a setting event or establishing operation (Freeman et al. 1999), making individuals with ASD more likely to display problem behavior when they are anxious (e.g., child is feeling anxious and thus, when asked to do his homework, he is more likely to hit himself). Additionally, it is possible that anxiety may also serve as a discriminative stimulus (S^D) for problem behavior (e.g., child is feeling anxious and bites himself to reduce the anxiety). Given either scenario, individuals with ASD may engage in problem behavior to reduce their anxiety by escaping/avoiding an anxiety-provoking situation (i.e., negative reinforcement) and/or by seeking reassurance, comfort, or access to self-soothing stimuli (i.e., positive reinforcement). This hypothesis was supported by our previous study in which we conducted a functional assessment of problem behavior during high-anxiety and low-anxiety conditions and found that, for three children with ASD and IDD, substantially more problem behaviors occurred in high-anxiety than in low-anxiety contexts (Moskowitz et al. 2013). The goal of the present study was to treat the identified anxiety and associated problem behavior in these same participants. Specifically, we examined whether a multicomponent behavioral intervention package, incorporating principles and strategies from the anxiety treatment literature for neurotypical (NT) children with principles and strategies from applied behavior analysis (ABA) and Positive Behavior Support (PBS), would reduce anxiety and associated problem behavior in three children with ASD and co-occurring IDD.

There is extensive research supporting the effectiveness of cognitive behavioral therapy (CBT) in reducing anxiety in NT children (e.g., Velting et al. 2004), but there is not as much research evaluating CBT in the treatment of anxiety with children with ASD, particularly those with ASD and IDD (Rosen et al. 2016). Several randomized clinical trials indicate that CBT with modifications (e.g., visual supports, more concrete language, greater parental involvement, incorporating special interests) can reduce symptoms of anxiety in children with high-functioning ASD (HFA) (e.g., Chalfant et al. 2007; Reaven et al. 2012; Sofronoff et al. 2005; Storch et al. 2013; Wood et al. 2009). In fact, rates of positive treatment response in these studies have been over 50%, which is comparable to the treatment response for NT children with anxiety disorders (e.g., Kendall 1994). Further, the moderate to large effect sizes found in a meta-analysis of CBT in youth with HFA (Ung et al. 2015) are in line with the findings of meta-analyses examining CBT in NT youth without ASD. However, it has not been examined whether these same treatments can be used with individuals who display more severe symptoms of ASD or with those children with ASD who have an IDD (Moree and Davis 2010; White et al. 2009).

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 de-emphasized, simplified, modified, adapted to the child's level, or excluded altogether so that the intervention is "behavioral" (i.e., ABA/PBS) rather than "cognitive-behavioral." Only a handful of studies have used behavioral interventions to treat specific fears, "fearful responses," or "avoidance behavior" in children with ASD and IDD (Cale et al. 2009; Ellis et al. 2006; Koegel et al. 2004; Love et al. 1990; Luscre and Center 1996; Rapp et al. 2005; Ricciardi et al. 2006; Schmidt et al. 2013; Shabani and Fisher 2006; for a review, see; Rosen et al. 2016). These single-subject design studies incorporated a variety of behavioral intervention procedures including graduated exposure or systematic desensitization (Ellis et al. 2006; Love et al. 1990; Luscre and Center 1996; Schmidt et al. 2013), modeling (Ellis et al. 2006; Luscre and Center 1996), positive reinforcement (Love et al. 1990; Luscre and Center 1996; Schmidt et al. 2013), hierarchy/

stimulus fading (Ricciardi et al. 2006; Shabani and Fisher 2006), stimulus control procedures (Luscre and Center 1996), blocking plus reinforcement for approach behavior (Rapp et al. 2005), progressive relaxation training (Mullins and Christian 2001), and presenting the child with a choice between the feared stimulus and an alternative stimulus matched on generic content (Cale et al. 2009).

Many of these therapeutic approaches are similar to those used when implementing an ABA and/or PBS approach to treatment. Practitioners of PBS (Carr et al. 2002) use the principles of ABA to understand the full range of variables that impact problem behavior; develop multicomponent intervention plans (based on those variables) that emphasize prevention and teaching new skills; and provide parents, teachers, and other stakeholders with behavior support that is acceptable and effective in natural settings (Lucyshyn et al. 2007; Neufeld et al. 2014). The majority of aforementioned studies targeting fearful/avoidant responses in children with ASD and IDD have focused more on consequence-based intervention strategies (e.g., reinforcement) than on prevention or antecedent-based strategies. With the exception of one study that used the antecedent intervention of providing choices (Cale et al. 2009) and a recent study by Neufeld and colleagues (2014) that integrated components of CBT with PBS to treat the fear of riding elevators for a child with autism, previous research has not used antecedent-based interventions to treat anxiety and problem behavior in individuals with ASD, nor has it incorporated stakeholder participation or ecological validity (core features of PBS) in the selection of goals and strategies. Further, in most of the aforementioned studies (with the exception of Neufeld et al. 2014), intervention was not informed by a functional assessment, even though research has shown that interventions based on assessment of the function of problem behavior are about twice as likely to succeed as those that are not (see meta-analysis by Carr et al. 1999). In short, most of the intervention research targeting fear/anxiety in children with ASD and IDD has not incorporated key principles or procedures of PBS.

In the present study, we combined aspects of CBT with aspects of ABA and PBS to treat anxiety in specified contexts for three children with ASD and IDD. Our multicomponent intervention plan incorporated three of the main components of CBT programs for anxiety in NT youth (e.g., Kendall 1994) and those with HFA (e.g., Reaven et al. 2012): graduated exposure, psychoeducation, and cognitive restructuring. Our intervention plan also incorporated antecedent-based strategies from ABA/PBS that have been shown to reduce problem behavior (although not anxiety) in individuals with ASD and IDD including increasing predictability (Flannery and Horner 1994), providing choices (Shogren et al. 2004), generalized reinforcement (Magito McLaughlin and Carr 2005), and using a child's obsessive or perseverative interests as reinforcers (Charlop Christy and Haymes 1996) as well as consequence-based strategies including contingent positive reinforcement (e.g., Luscre and Center 1996), differential reinforcement (e.g., Athens and Vollmer 2010), and escape extinction (e.g., Carr et al. 1980). In terms of antecedent-based strategies to increase predictability, the use of visual schedules (Mesibov et al. 2002), Social Stories (Kokina and Kern 2010), and priming (Schreibman et al. 2000) to signal the sequence of upcoming activities have all been shown to reduce problem behavior in individuals with ASD and/or IDD. Many of these ABA/PBS strategies are consistent with the modifications suggested by previous CBT studies for children with HFA (i.e., incorporating special interests, increased parental involvement, and simplified cognitive restructuring using visual supports and more concrete language). Whereas existing studies of CBT to treat anxiety in ASD have used manualized treatments and group designs, PBS emphasizes individualized treatments. This is significant, given that the structured, linear format of manualized CBT, particularly group CBT, may limit the ability to match intervention strategies to the individual characteristics of a child with ASD (Wood et al. 2009). Wood and colleagues (2009) noted that, given the heterogeneity of youth with ASD, individualized interventions tailored to a child's specific characteristics may be particularly effective. Further, given the heterogeneity of setting events and antecedents of anxious behavior in children with ASD, the different functions of such behavior, the idiosyncratic fears that children with ASD possess, and the idiosyncratic ways in which they might express their fear and anxiety (Kerns et al. 2016; Moskowitz et al. 2017), individualized treatments tailored to each specific child may be that much more powerful for youth with ASD and IDD. Thus, in accordance with the tenets of PBS, we designed individualized interventions tailored to specific children with ASD and IDD based on the function(s) of the child's anxious behavior, the specific antecedents that evoked this behavior and the setting events that increased the likelihood of this behavior, the idiosyncratic fears and preferences/interests of each child, and the goals and values of the child's family.

Method

Participants and Setting

Participants were three school-aged children, Jon, Sam, and Ben, ages 6, 8, and 9 years old, diagnosed with autism spectrum disorder (ASD), according to criteria specified in the diagnostic and statistical manual of mental disorders (text revision; DSM-IV-TR 2000). Participants were recruited through a local listserv for parents of children with ASD. The parents were asked a series of questions about the frequency, intensity, and behavioral indicators of anxiety as well as problem behaviors that might be associated with

anxiety. 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; and (c) had a history of behavior problems and reported anxiety that became more frequent when specific stressors were present, as confirmed by direct observation. All three were identified as functioning in the low range of intellectual and/or adaptive functioning; Jon's adaptive behavior on the Vineland was in the first percentile (low range), Ben's IQ was 61 (first percentile) on the Stanford-Binet and his adaptive behavior was 62 (first percentile, low range) on the Vineland, and Sam's IQ was 66 (second percentile) on the Stanford-Binet and his adaptive behavior was 70 (second percentile, low range) on the Vineland (see Moskowitz et al. 2013 for more information about participants). None of the participants were on medication, and one participant (Sam) dropped out of treatment after six intervention sessions. All sessions were conducted within the natural context of the participants' ongoing routines at home or in the community.

Response Definition and Measurement

All intervention sessions were videotaped and scored for anxious behavior and problem behavior at a later time. Data were collected on the frequency of anxious behaviors and problem behavior across 10-s intervals by two independent observers. For Jon, anxious behaviors included clinging (holding onto his mother), crying/tearfulness, cowering (e.g., turning into corner), and anxious vocalizations (e.g., whimpering, moaning, or idiosyncratic throat noises). For Ben, anxious behaviors included repetitive questioning about his parents' whereabouts (e.g., "Where are you going?", "Where is Mommy?"), verbal pleading (e.g., "I really want Mommy now!"), running to the front door, and watching out the front door. Finally, for Sam, anxious behaviors were defined as plugging his ears, eyes darting back and forth, mouthing his finger, and turning his head around to look out the rear window of the car. (See Moskowitz et al. 2013 for how these behavioral indicators of anxiety were identified.) Problem behavior was defined for Jon as yelling/screaming, elopement, pushing others, and pulling his mother's hair. For Ben, these included whining, yelling/screaming, tantrum behavior, and running out the front door of his house. For Sam, these included laying down in the car seat, reaching for his parents while they were driving, and displaying tantrum behavior (e.g., yelling "No left!").

In addition to recording problem behavior and anxious behavior, in order to index the contextual or cognitive/affective component of anxiety, we used parent-report on the contexts that evoke anxiety as well as subjective ratings of anxiety. Regarding the latter, we asked blind observers to rate appearance of anxiety from videos of sessions using a four-point scale with three indicating high anxiety and zero indicating no anxiety. These ratings were made after the study by undergraduate research assistants, who had no direct involvement in the study. Finally, to index the physiological component of anxiety, we measured HR and RSA. See Moskowitz et al. (2013) for details regarding how HR and RSA were analyzed using QRSTool and CMetX (Allen et al. 2007).

In order to keep the intervention sessions comparable to baseline sessions, we recorded frequency of anxious behaviors and problem behavior, as well as HR/RSA and ratings of appearance of anxiety, beginning from when the anxiety-evoking discriminative stimulus (S^D) was first presented (i.e., for Jon, when the birthday cake was presented along with people singing "Happy Birthday" song; for Ben, when his parents said they were leaving the house; for Sam, when the car ride began) and ending when the anxiety-evoking situation ended (i.e., for Jon, when the song was over and the candles blown out; for Ben, when his parents returned home; for Sam, when the car ride ended). This is the data that is presented in the intervention sessions for Figs. 1, 2, 3, 4 and 5.

Procedure

Using the Functional Assessment Interview (FAI; O'Neill et al. 1997), the first author assisted parents in prioritizing a "high-anxiety" context that was most likely to evoke both anxiety and problem behavior and most reduced family quality of life. To be considered a "high-anxiety context," (1) the context needed to be rated by parents as a "5" on a scale of 1 to 5 (1 = no anxiety, 5 = severe anxiety), (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 consistently rated as either highly anxious (a "3") or moderately anxious (a "2") on the four-point rating scale by an undergraduate rater (see Moskowitz et al. 2013). For Jon, the High-Anxiety context was birthday parties (lighting candles while singing "happy birthday"), for Ben, it was separating from his parents (leaving Ben at home with another adult) and, for Sam, it was making left or right turns while riding in the car. All identified contexts were confirmed by direct observation.

All sessions were conducted within the natural context of the participant's ongoing routine. During all sessions, participants wore the Alive Heart and Activity Monitor (Model HM131 made by Alive Technologies), 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. There were four baseline sessions conducted for Jon, five baseline sessions for Ben, and seven baseline sessions for Sam. During baseline observations, parents, relatives, or home-teachers were

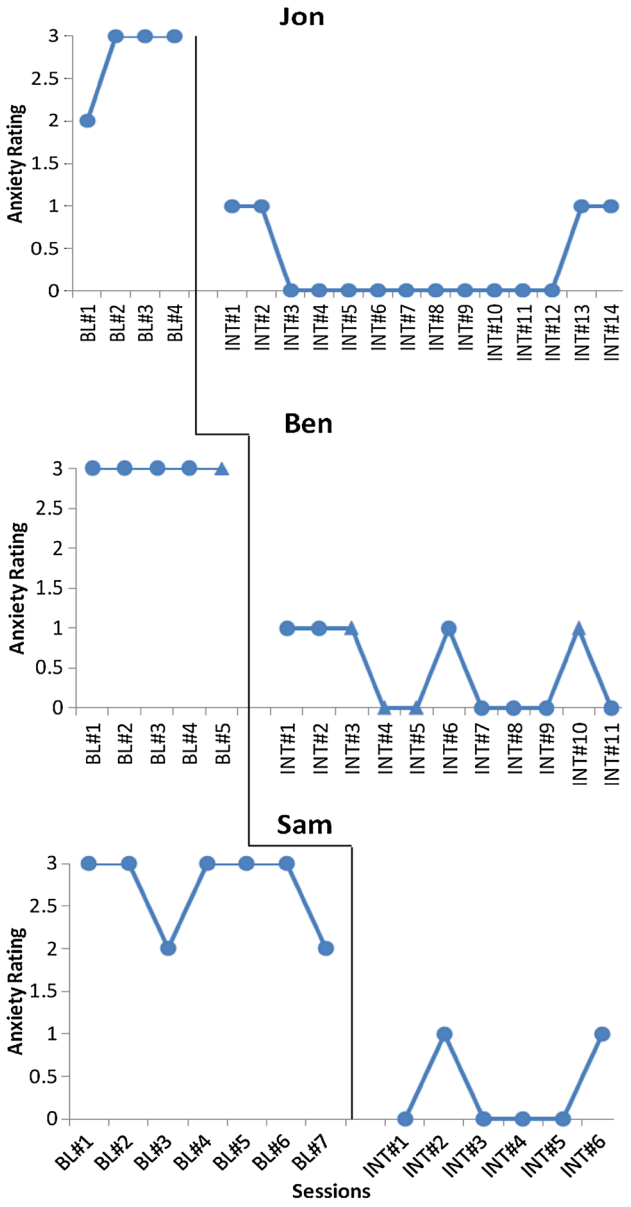

Fig. 1 Subjective ratings of anxiety on Likert-type rating scale for baseline (BL) and intervention (INT) phases. Raters were blind to whether session was a BL or INT session. The *closed triangle* symbol for Ben denotes the times that he stayed home alone with someone else (e.g., his grandparents, his uncle) other than the primary researcher (L.J.M.), while the researcher left the house with his parents

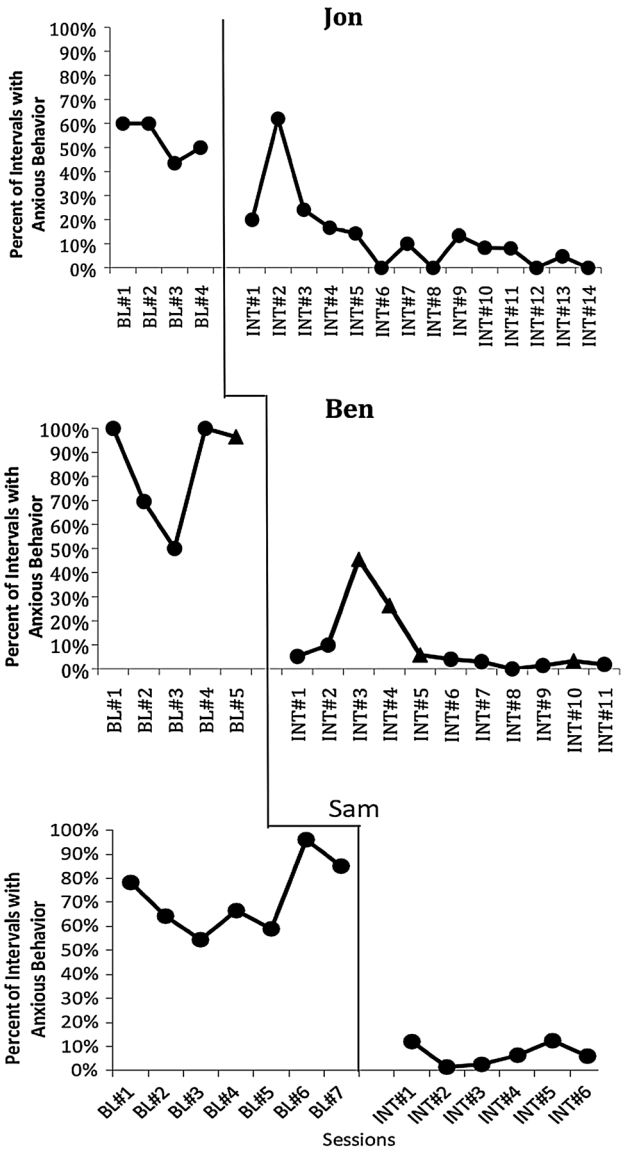

Fig. 2 Frequency of anxious behavior (percent of 10-s intervals with anxious behavior) for the three participants during baseline (BL) and intervention (INT) phases. The *closed triangle* symbol for Ben denotes the times that he stayed home alone with someone else (e.g., his grandparents, his uncle) other than the primary researcher (L.J.M.), while the researcher left the house with his parents

free to interact with the participant in a manner that was consistent with the natural context in which the observation occurred. They were asked to respond as they typically would to their child's target behaviors and were not provided with any feedback from the researcher about how to manage the behaviors during baseline.

Based on caregiver report and direct observations, it appeared that the primary function of each child's problem behavior was to escape or avoid an anxiety-provoking situation. For Jon, this involved attempting to leave the room. For Ben, this involved preventing his parents from leaving the house. For Sam, this involved intervening with his parents while driving, and attempting to convince them to drive straight and/or to avoid making turns.

After the final baseline observation, the first author met with the parents to discuss the results of the assessment and to propose various intervention strategies derived from both the CBT literature on treating anxiety in NT children and those with HFA, and from the ABA and PBS literature on treating avoidance and problem behavior in children with

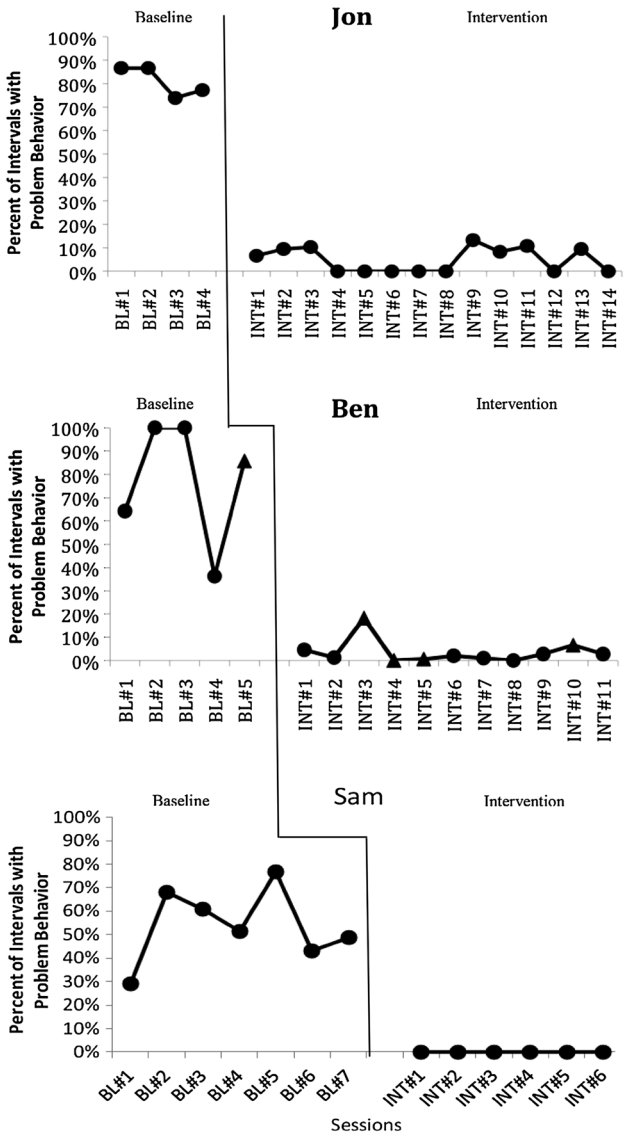

Fig. 3 Frequency of problem behavior (percent of 10-s intervals with problem behavior) for the three participants during baseline (BL) and intervention (INT) phases. The *closed triangle* symbol for Ben denotes the times that he stayed home alone with someone else (e.g., his grandparents, his uncle) other than the primary researcher (L.J.M.), while the researcher left the house with his parents

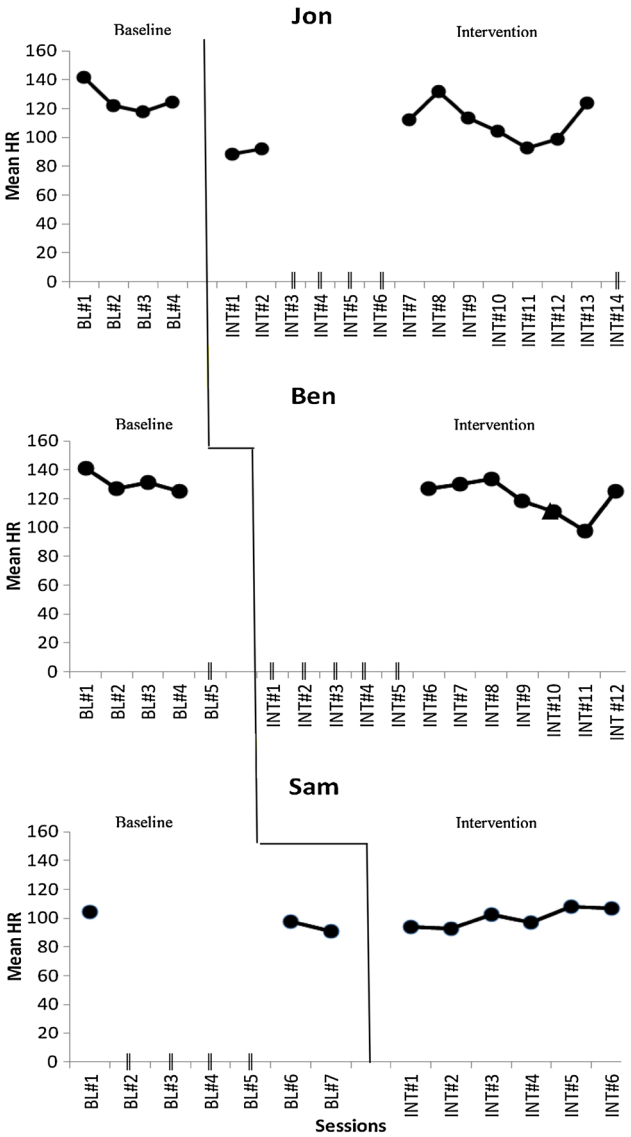

Fig. 4 Mean heart rate (HR) (averaging each 30-s epoch within a session) for the three participants during baseline (BL) and intervention (INT) phases. The *closed triangle* symbol for Ben denotes the times that he stayed home alone with someone else other than the primary researcher (L.J.M.), while the researcher left the house with his parents. The ‖ symbol denotes that the HR monitor did not record IBI for that session or that there was not enough usable IBI data in the session to analyze HR

IDD. Parents selected the options that they felt represented the best fit for their family and were trained to implement the intervention strategies using in vivo modeling and coaching and problem-solving discussions (Lucyshyn et al. 2007). The treatment included both strategies that were used *during* the intervention sessions (i.e., graduated exposure, counterconditioning/generalized reinforcement, incorporating perseverative interests, escape extinction) in addition to strategies that were used *prior* to the intervention sessions in preparation for those sessions (i.e., increasing predictability, psychoeducation/cognitive restructuring). Intervention sessions spanned across 10 weeks for Jon, 9 weeks for Ben, and 4 weeks for Sam. The treatment components are summarized below (and see Appendix A).

CBT Treatment Components

Graduated Exposure

Treatment involved gradual exposure to increasing proximity, intensity, or amounts of feared stimuli or situations. For

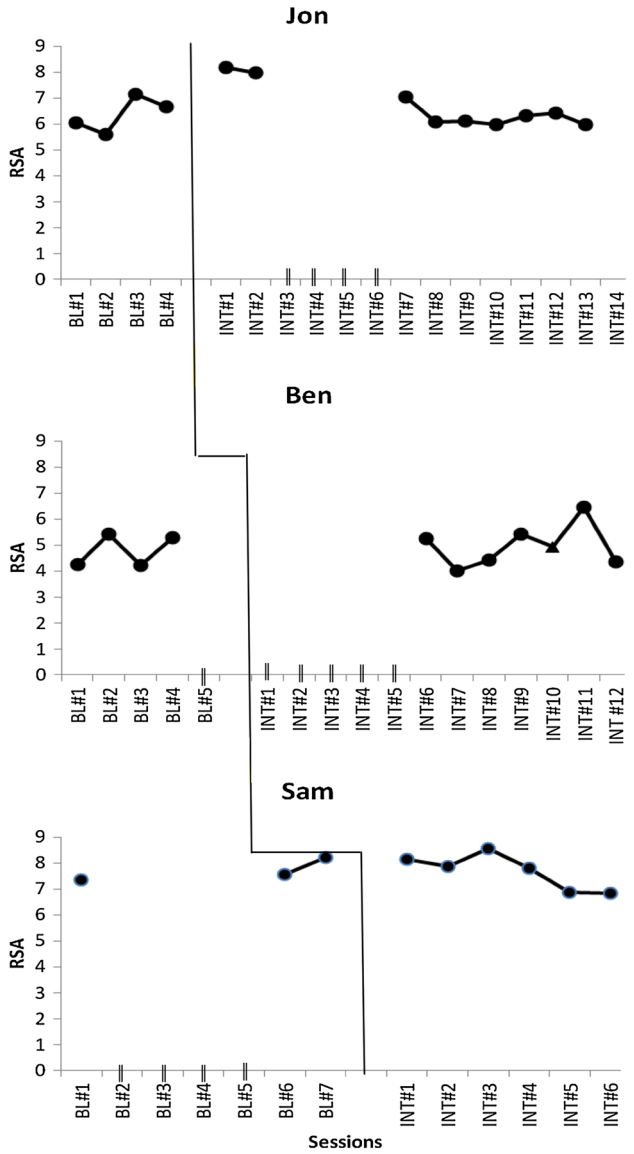

Fig. 5 Mean respiratory sinus arrhythmia (RSA) (averaging each 30-second epoch within a session) for the three participants during baseline (BL) and intervention (INT) phases. The *closed triangle* symbol for Ben denotes the times that he stayed home alone with someone else other than the primary researcher (L.J.M.), while the researcher left the house with his parents. The || symbol denotes that the HR monitor did not record IBI for that session or that there was not enough usable IBI data in the session to analyze RSA

Jon, exposure involved singing happy birthday, for Ben it involved repeated exposure to having his parents leave the house to go run errands, and for Sam, it involved left/right turns while riding in the car. These exposures were conducted in a hierarchical fashion from situations that were rated by parents as less anxiety-provoking (e.g., singing happy birthday with the candles in the cake unlit) to situations that were rated as more anxiety-provoking (e.g., candles lit, increasing proximity to cake). For example, Sam was exposed to activities that his parents and home teachers predicted would provoke relatively low levels of anxiety: listening to audio recordings of the sound of directional signals and then watching videos of cars making left and right turns on YouTube. Sam then engaged in an activity that was predicted to evoke moderate levels of anxiety: listening to the blinker in the car when the car was stationary (parked in the driveway), initially with the car door open and then with the door closed. Finally, Sam engaged in activities that his caretakers predicted would evoke high levels of anxiety: 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 turns (e.g., major intersections with traffic lights).

Of note, anxious behavior, problem behavior, subjective ratings, and HR/RSA were measured in the full target situation (i.e., same as baseline) rather than a less anxiety-provoking version for all of the intervention sessions, with the exception of the first intervention session for Jon (in which the musical candles were presented unlit on the birthday cake). For Jon's intervention session #2 and all of the following Intervention sessions, the candles on the cake were lit, which was identical to the baseline conditions. For the other two participants, there was a relatively constant level of exposure across all sessions; the graded or lower-level exposures were conducted prior to the first intervention session. For example, Sam engaged in the activities that his caretakers rated as evoking low and moderate levels of anxiety (e.g., listening to blinkers on YouTube) immediately *prior* to the first intervention session when we began to collect data.

Psychoeducation

Prior to the first intervention session, participants' parents were provided with information about how anxiety is learned and maintained, and the rationale for various treatment techniques (Velting et al. 2004). Immediately prior to the first few intervention sessions, Social Stories (for Ben and Sam) and video modeling (for Jon) were used as a way to provide a simplified visual format for psychoeducation to teach the children about anxiety and/or the anxiety-provoking situation. For example, Ben's Social Story (a) normalized anxiety; (b) explained that anxiety has a function or purpose; (c) described the specific nature of Ben's anxiety; (d) explained the concept of habituation; and (e) provided Ben with replacement behaviors that he could engage in 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) (see Appendix B for text of story).

Cognitive Restructuring

Cognitive restructuring involves identifying anxious thoughts, challenging those anxious thoughts, and teaching coping-focused thinking or coping self-talk. Although we did not address identifying or disputing anxious thoughts, we taught coping statements to one participant (Ben). For Ben, cognitive restructuring took the form of prompting coping self-talk through the use of a Social Story. The Social Story provided Ben with sentences or phrases that he could repeat to himself when he was feeling anxious (e.g., "I am going to be brave"). Before and/or during exposures, the researcher and/or caregiver modeled the coping statements that were written in the Social Story, repeated variations of the coping statements (such as referring to Ben as a superhero, "Super-Ben"), or provided indirect references to the coping statements (e.g., "Were you brave, Super-Ben?"). Social Stories (for Sam) and video modeling (for Jon) were also used as a way to provide very simplified cognitive restructuring to the children in terms of presenting the anxiety-provoking situation in a non-anxiety-provoking light (for Sam), and modeling non-anxious behavior in the targeted context (for Jon) in a visual format that could more easily be comprehended.

ABA/PBS Antecedent-Based Strategies

Increasing Predictability

One PBS antecedent strategy entails providing information proactively to reduce anxiety (Lucyshyn et al. 2007); this can be done using visual schedules, Social Stories, or priming, which are all ways to increase predictability by manipulating antecedent events or setting up establishing operations.

Visual Schedules

A visual schedule informs an individual about the upcoming sequence of events (Mesibov et al. 2002). For one participant (Sam), pictures were created to represent the most common locations where his parents drove and those pictures were used to construct a visual schedule. Before entering the car during treatment, 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. The visual schedule was used only in two intervention sessions for Sam, as his parents felt that it was no longer necessary or useful and therefore they discontinued using it.

Social Stories

Social Stories are used to visually depict the sequence of events involved in a routine or situation (Gray and Garand 1993). Individualized Social Stories were created for Ben and Sam to help them know what to expect during anxiety-provoking situations (see Appendix B for the text of these stories). The Social Story was used only in two intervention sessions for Sam and three sessions for Ben, as Sam's parents felt that it was initially helpful but no longer necessary and they discontinued using it, whereas Ben's parents felt as though reading the story prior to their departure may have made him more anxious, so they discontinued reading it to him (although Ben still carried the story around with him). Of note, a meta-analysis of Social Stories by Kokina and Kern (2010) found that Social Stories were more effective when they are read in fewer sessions rather than mores sessions (perhaps because intervention effects may wear off as a result of longer intervention duration) and that, in many cases, Social Stories seemed to produce immediate changes in the levels of targeted behaviors.

Priming/Video Priming

In priming, an individual previews future events so that they become more predictable (Wilde et al. 1992). Prior to the first intervention session, Sam was exposed to audio-recordings of blinkers and videos of cars making left and right turns on YouTube (video priming), while Jon viewed multiple internet videos of his favorite characters from Sesame Street singing happy birthday and blowing out candles on a birthday cake (video modeling). For Ben, priming involved behavioral rehearsal (i.e., "practicing" remaining inside the house with another adult while his parents left the house) immediately prior to the first intervention session, before his parents actually left in the car. As with the Social Story, priming was used in only three sessions for Ben, as his parents felt that rehearsing the separation was actually increasing his anticipatory anxiety, so this procedure was stopped.

Providing Choices

Sam was provided with 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, whenever possible, Sam's parents attempted to provide him with a choice of the destinations. Similarly, Ben was given the opportunity to choose preferred activities that he could engage in while his parents were out.

Generalized Reinforcement/Counterconditioning

Generalized reinforcement involves pairing the aversive situation with many strongly preferred reinforcers (Magito McLaughlin and Carr 2005). For Jon, the presentation of the birthday cake and the happy birthday song were paired with his most highly preferred and otherwise inaccessible positive stimuli (e.g., Sesame Street pop-up toy or Sesame Street CD player). For Ben, his parents' departure was paired with his most highly preferred items and activities (e.g., Muppet Movie, swings, perseverative counting). Finally, for Sam, riding in the car was paired with his most highly preferred and otherwise inaccessible positive stimuli (e.g., audio recording of Dr. Seuss' book "The Sneetches"). We chose reinforcers that participants did not otherwise have access to outside of the targeted context. This strategy of pairing aversive (or anxiety-producing) stimuli with competing positive (or anxiety-reducing) reinforcers noncontingently stands in contrast to the consequence-based strategy of positive reinforcement contingent upon displaying approach 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.

Incorporating Perseverative Interests

These positive stimuli that were paired with the anxiety-provoking stimuli were not just preferred reinforcers (such as pretzels, as in Magito-McLaughlin and Carr 2005) but were the participants' most obsessive or perseverative reinforcers (e.g., The Muppet Movie) or reinforcers that incorporated their perseverative interests (e.g., Dr. Seuss). Participants' perseverative or circumscribed interests were also incorporated into other intervention procedures (e.g., Social Story, video modeling) in addition to being used in counterconditioning as described above and incorporated into the exposure itself (e.g., Sesame Street birthday candles in the cake for Jon).

Prompting an Approach Response (Jon Only)

Jon was initially provided with a highly preferred object (e.g., Sesame Street pop-up toy) noncontingently for two sessions. Then, he was presented with a general verbal prompt (e.g., "Jon, do you want to play with this? If you blow out the candle, you can play with this!") and contingent reinforcement upon approach behavior.

Consequence-Based Strategies

Positive Reinforcement Contingent upon Approach (Jon Only)

Starting the third session, if Jon responded by approaching the birthday cake for the preferred item and attempting to blow out the candle, Jon's perseverative reinforcer was immediately delivered and he received positive feedback. If Jon did not approach the cake, he was presented with a general cue (e.g., "Jon, do you want to play with this?", while showing him the toy) to encourage an approach response. Following each successful approach, Jon's proximity to the cake was gradually increased in order to strengthen the approach response. This procedure was not conducted with Sam or Ben in that they were not required to approach a feared stimulus.

Differential Reinforcement of Alternative Behavior (DRA)

Ben's parents, grandparents, and uncle were coached to promote Ben's brave behaviors by differentially responding to Ben's brave versus anxious behavior. Attention for Ben's anxious behavior was minimized, while Ben was provided with praise or attention for brave behavior when he was engaging in a behavior that was incompatible with anxious behavior (e.g., sitting in a chair in the living room, versus standing by the front door watching to see his parents' car) or when he was behaving in a calm and/or quiet manner (i.e., not compatible with anxious behavior).

Escape Extinction

Escape extinction was implemented for all three participants in which their problem or anxious behaviors no longer resulted in reinforcement. For each child, this meant continuing with the anxiety-provoking situation regardless of the child's behavior.

In summary, the four intervention strategies that were used consistently across all three participants in every intervention session were: graduate exposure, counterconditioning/generalized reinforcement, incorporating the participant's perseverative interests, and escape extinction of problem behavior or anxious behavior (when such behavior was present). The treatment strategies that were only used prior to the first few intervention sessions were the strategies that involved increasing predictability (i.e., visual schedule, Social Story, priming, video modeling). As we explained above, for these increasing predictability strategies, the parents either felt they were no longer necessary after the first two or three intervention sessions or that they made the children more anxious, so we discontinued using them. Likewise, parents were only provided with

psychoeducation, another treatment component, prior to the first intervention session, and the psychoeducation and cognitive restructuring (that was conveyed to the children via the Social Stories or video modeling/priming) was only included at the beginning.

Design

A nonconcurrent multiple baseline design (Hersen and Barlow 1976) across three participants with ASD was used to evaluate the impact of the multicomponent intervention packages on anxious behavior and problem behavior that occurred during the target contexts.

Interobserver Agreement (IOA)

Frequency of anxious behaviors and problem behavior were coded for IOA by the first and second author in 67% (30 out of 45) of the videotaped sessions, distributed evenly across baseline (BL) and intervention (INT) sessions. IOA for anxious and problem behavior was scored for each 10-s interval in which the two researchers agreed on the presence or absence of each 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; agreement occurred when both observers recorded the same anxious behavior(s) or problem behavior(s) within the same interval. 12 out of 18 of Jon's sessions were coded for IOA (4 BL and 8 INT), 10 out of 15 of Ben's sessions were coded for IOA (4 BL and 6 INT), and 9 out of 13 of Sam's sessions were coded for IOA (3 BL and 6 INT). For Jon's 12 sessions, IOA was 95% ($SD=6$%, range 80–100%) for problem behavior and 88% ($SD=7$%, range=80–100%) for anxious behavior. For Ben's 10 sessions, IOA was 97% ($SD=4$%, range 91–100%) for problem behavior and 93% ($SD=7$%, range 83–100%) for anxious behavior. For Sam's 9 sessions, IOA was 98% (SD=4%, range 91–100%) for problem behavior and 95% (SD=6%, range 85–100%) for anxious behavior. Two undergraduate research assistants who were blind to the purpose of the study and blind to study conditions also coded the videotapes for appearance of anxiety using a four-point rating scale; they double-coded 33% of the sessions for each child for IOA. For appearance of anxiety using the 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.

Treatment Fidelity

Reliability data were also collected on fidelity of implementation (also referred to as "intervention integrity"), or the accurate use of each treatment component. Two observers, both of whom were doctoral students with no prior knowledge of the study, independently completed intervention integrity checks on 100% of baseline and intervention sessions for each child. The observers were blind to whether sessions were baseline or intervention sessions. Integrity was scored on a trial-by-trial basis for the presence of each treatment component for each child. For Jon's treatment, fidelity was 33% during BL and 94% during INT. For Ben's treatment, fidelity was 38% during BL and 94% during INT. For Sam's treatment, fidelity was 40% during BL and 100% during INT. IOA on integrity was 96% for Jon (100% for BL and 95% for INT), 88% for Ben (86% for BL and 88% for INT), and 100% for Sam (for both BL and INT). Intervention integrity checklists for each participant are available from the first author upon request.

Data Analysis Procedures

To further quantify treatment outcomes, two measures of nonregression effect size were calculated for each participant: the percentage of non-overlapping data (PND) and mean baseline reduction (MBLR). PND was determined by calculating the number of intervention data points that fell 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 et al. 1987). A PND of 90–100% indicates that the treatment was highly effective in behavior reduction and a PND of <50% indicates that the treatment was ineffective. We also calculated 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. We calculated MBLR for all data points and for the last three in each phase.

Results

As shown in Fig. 1, using the Likert-type rating scale, on a scale of 0 (no anxiety) to 3 (high anxiety), blind undergraduate observers rated each child's anxiety during baseline and intervention sessions. All three participants showed clear reductions in ratings once intervention was in place. Jon's anxiety in the high range ($M=2.8$, $SD=0.5$, range 2 to 3) during baseline sessions and close to the "no anxiety" range ($M=0.21$, $SD=0.38$, range 0 to 1) during intervention sessions. Ben's anxiety was rated as high ($M=3$, $SD=0$) during

baseline sessions and in the "no anxiety" to "mild anxiety" range ($M=0.46$, $SD=0.52$, range 0-1) during intervention sessions. Sam's anxiety was rated in the moderate-to-high range during baseline sessions ($M=2.67$, $SD=0.4$, range 2–3) and close to the "no anxiety" range ($M=0.17$, $SD=0.26$, range 0–0.5) during intervention sessions.

Figure 2 shows the frequency of anxious behavior for the three children in the anxiety-provoking contexts during the baseline (BL) and intervention (INT) phases. There was an immediate level change in anxious behavior between the baseline and intervention phase for all three participants. For Jon (upper panel), the mean percentage of intervals in which anxious behavior occurred during the happy birthday context was 53% ($SD=8$%, range 43–60%) during BL and 13% ($SD=16$%, range 0–62%) during INT, a 76% mean baseline reduction (MBLR) in the frequency of anxious behavior. When considering just the last three sessions of baseline and the last three sessions of intervention, the MBLR substantially increases, resulting in a 97% reduction in the frequency of anxious behavior for Jon. The PND for Jon's anxious behavior equaled 93% (13 of 14 INT data points did not overlap with BL data points), which indicates that the treatment was highly effective in reducing anxious behavior. For Ben (middle panel), 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% ($SD=23$%, range 50–100%) during BL and 10% ($SD=14$%, range 0–45%) during INT, an 88% mean baseline reduction (MBLR) in the frequency of anxious behavior. When considering just the last three sessions of baseline and intervention, the MBLR increases, resulting in a 97% reduction in the frequency of anxious behavior for Ben. The PND for Ben's anxious behavior was 100% (11 of 11 INT data points did not exceed the BL data points), indicating the treatment was highly effective in reducing anxiety. For Sam (bottom panel), 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 73% ($SD=14$%, range 58–96%) during BL and 7% ($SD=5$%, range 3–13%) during INT, which represents a 91% mean baseline reduction (MBLR) in the frequency of anxious behavior. When considering just the last three sessions of baseline and intervention, the MBLR remains a 90% reduction in the frequency of anxious behavior for Sam. The PND for Sam's anxious behavior was 100% (i.e., 100% of the INT data points did not overlap with BL data points), indicating the treatment was highly effective in reducing anxiety.

Figure 3 shows the frequency of problem behavior for the three children in the anxiety-provoking contexts during the baseline and intervention phases. There was an immediate level change in problem behavior between the baseline and intervention phase for all three participants. For Jon (upper panel), the mean percentage of intervals in which problem behavior occurred during the happy birthday context was 81% ($SD=7$%, range 74–87%) during BL and 2% ($SD=4$%, range 0–13%) during INT, a 97% mean baseline reduction (MBLR) in frequency of problem behavior when considering all baseline and intervention data, and a 98% MBLR when considering just the last three sessions of baseline and intervention. The PND for Jon's problem behavior was 100% (14 of 14 INT points did not overlap with BL points), which indicates that the treatment was highly effective in reducing problem behavior. During BL, none of the happy birthday sessions were completed without any problem behavior. During INT, 50% of happy birthday sessions were completed successfully without any problem behavior and 86% were completed with $\leq$10% of intervals containing problem behavior.

For Ben (middle panel), 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 36–100%) during BL and 3% during INT ($SD=5$%, range 0–18%), an 96% mean baseline reduction (MBLR) in the frequency of problem behavior (whether considering all baseline and intervention data points, or just the last three sessions of baseline and intervention). The PND for Ben's problem behavior was 100% (11 of 11 INT data points did not overlap with BL points). During BL, none of the sessions in which his parents left the house was completed without any problem behavior. During INT, 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 $\leq$8 instances of problem behavior.

For Sam (bottom panel), 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 54% ($SD=16$%, range 29–77%) during BL and 0% during INT, a 100% mean baseline reduction (MBLR) in the frequency of problem behavior (whether considering all baseline and intervention data points, or just the last three sessions of baseline and intervention). The PND for Sam's problem behavior was 100% (6 of 6 INT data points did not overlap with BL points). During BL, none of the sessions was completed without any problem behavior. During INT, 100% of sessions were completed successfully without any problem behavior.

In order to test overall effects of HR and RSA, we conducted a mixed effects model using the xtmixed command in the Stata statistical software (StataCorp 2011). The two models that we tested contained two levels of observations (level 1—representing each 30-s epoch measured) and participant (level 2—representing the three individual participants measured). We centered session numbers individually so that zero represents the first day of intervention (and

all baseline measurements are negative values). We then regressed RSA on Treatment Condition, Session Number (each session represented one observation or exposure session) and their interaction, and repeated the same model using HR as a dependent variable instead of RSA.

A Shapiro-Francia test of normality revealed that level 1 residuals were significantly positively skewed for RSA ($Z=3.55$, $p<0.01$). Multiple transformations were attempted; however, the residuals remained non-normal. As a result, specific parameter estimates should be interpreted with caution.

The RSA model showed good fit with log-likelihood ratio of −380.95 and Wald χ^2 (6, $N=240$, $n=3$) = 14.11, $p<0.01$. Within the model, treatment condition [$\beta=0.36$, $Z=1.13$, $p=0.26$, 95% CI (−0.26, 0.98)] and Session Number [$\beta=0.10$, $Z=1.65$, $p=0.10$, 95% CI (−0.18, 0.21)] did not significantly predict RSA. However, their interaction [$\beta=-0.19$, $Z=-2.99$, $p<0.01$, 95% CI (−0.32, −0.07)] did significantly and negatively predict RSA. A subsequent interaction effect analysis revealed that, when session number was recentered one SD above the mean (at 5.54 sessions), Treatment Condition [$\beta=1.42$, $Z=3.66$, $p<0.01$, 95% CI (0.66, 2.18)] did significantly and positively predict RSA, while Session Number [$\beta=0.10$, $Z=1.65$, $p=0.10$, 95% CI (−0.18, 0.21)] did not. That is, on the first treatment session, RSA did not change, but after 5 treatment sessions, RSA increased.

The same analysis was conducted with mean HR as the response variable and showed good fit with a log-likelihood ratio of -995.42 and Wald χ^2 (6, $N=240$, $n=3$) = 13.89, $p<0.01$. Within this model, Treatment Condition [$\beta=-0.31$, $Z=-0.07$, $p=0.94$, 95% CI (−8.73, 8.11)] did not significantly predict HR, nor did the interaction term [$\beta=1.48$, $Z=1.70$, $p=0.09$, 95% CI (−0.22, 3.18)]. However, Session Number [$\beta=-1.60$, $Z=-2.04$, $p=0.04$, 95% CI (−3.12, −0.06)] did significantly and negatively predict HR in this model. In order to further understand this result, we examined a simpler model with only Session Number (un-centered) predicting HR. This model had a log-likelihood ratio of −1000.09 and performed significantly better than a null model, Wald χ^2 (3, $N=240$, $n=3$) = 10.69, $p<0.01$. Within the model, Session Number [$\beta=-0.66$, $Z=-3.27$, $p<0.01$, 95% CI (−1.05, −0.26)] significantly and negatively predicted HR, indicating that HR decreased across sessions.

Discussion

Collectively, our results offer initial support for using a multimethod approach to assess anxiety and related problem behavior (in Moskowitz et al. 2013) in order to derive appropriate treatments for children with ASD and IDD. Moreover, the results of the present study support the effectiveness of a multicomponent behavioral intervention plan for the treatment of anxiety and related problem behavior in children with ASD and IDD, a population that has largely been neglected in the clinical literature on anxiety. Following the multicomponent treatment, all three participants showed substantial improvement, with a decrease in anxiety (i.e., anxious behaviors, subjective anxiety ratings) and even more of a marked 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 treatment (in an average of 13% of intervals across sessions for Jon, 10% for Ben, and 7% for Sam), the intervention procedures appeared to mitigate their anxiety substantially so that it rarely resulted in displays of problem behavior. In contrast to baseline, almost all of the anxious behaviors that remained during treatment were of the milder and/or subtler variety, such as eyes rapidly darting back and forth when the birthday cake was presented (even when he sat near the cake quietly) for Jon, asking his parents "Where are you going?" for Ben, and turning his head to look out the back of the car window for Sam.

There are several strengths of this study. First, this study incorporates the clinical psychology literature with ABA/PBS literature to treat anxiety in children with both ASD and IDD, supporting the results of the study by Neufeld et al. (2014) that integrated CBT with PBS to treat a fear of elevators in a child with ASD (who did not have IDD). Specifically, we incorporated evidence-based treatments for childhood anxiety disorders in neurotypical populations from the CBT literature (e.g., graduated exposure) with evidence-based practices from the ABA/PBS literature including antecedent-based strategies (e.g., visual supports, providing choices) and consequence-based strategies (e.g., positive reinforcement). Of note, several intervention strategies we used could be conceptualized in both a CBT framework (e.g., listening to blinkers on YouTube as a low-level exposure) as well as a PBS framework (e.g., listening to blinkers on YouTube as video priming). Other CBT intervention strategies (e.g., psychoeducation, cognitive restructuring) were presented or conveyed using a PBS method/modality (e.g., Social Stories, video modeling). Both the CBT strategies and most of the PBS strategies may have functioned as setting events that counteracted or attenuated the effects of the anxiety-evoking discriminative stimuli. From an ABA/PBS framework, almost all of the strategies we used can be viewed as methods for reducing the aversiveness of the feared context/stimulus (i.e., altering the motivating operation by reducing its aversiveness). Given the heterogeneity of children with ASD and the lack of intervention research targeting anxiety in those children with ASD who have IDD, it is likely that incorporating principles and procedures from these two bodies of literature will be necessary in developing effective treatments for this complex population.

In addition to incorporating procedures derived from ABA/PBS, we also incorporated the principles of

family-based PBS (Lucyshyn et al. 2007) in terms of addressing ecological and social validity, using family routines as the unit of analysis and intervention, assessing anxiety using a multi-method approach, developing a multi-component intervention plan, choosing intervention strategies based on the results of a functional assessment and parental preferences, attending to the family's goals, skills, and needs, and collaborating with the children's parents to ensure a good contextual fit. We collaborated with parents to identify the priority contexts for intervention, to determine which intervention procedures would be used, and to determine how and when interventions would be implemented. This is an important point, as contextualized approaches to intervention, rooted in a collaborative partnership with the family of the child with ASD (e.g., Moes and Frea 2002), may be more effective for some children with ASD and IDD than prescriptive approaches, such as manualized treatments, which are often driven by the interventionist.

A key concern of PBS is whether interventions are ecologically valid, meaning that the intervention effects from a laboratory or clinic-based study can be successfully transferred to real-world situations (Carr et al. 2002). Importantly, the present investigation focused on assessing and treating anxiety in naturalistic social contexts—in natural settings (home, car, backyard), within natural routines, and incorporating natural intervention agents (e.g., parents, grandparents, uncle, home teachers)—thereby addressing ecological validity issues relevant to home and community settings. In terms of social validity, outside observers (i.e., 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 during baseline and close to the "no anxiety" range during intervention. They judged Ben's anxiety to be in the high range during baseline and in the "no anxiety" to "mild anxiety" range during intervention. Finally, they judged Sam's anxiety to be in the moderate to high range during baseline and close to the "no anxiety" range during intervention.

Our findings that RSA was significantly higher in the intervention condition than in baseline, but only after five intervention sessions, and that number of sessions alone predicted HR, suggest the possibility of two parallel processes at work. RSA, which indexes parasympathetic arousal, is thought to reflect the ability to regulate emotions or self-sooth when stressed (Porges 2007). Our intervention involved helping children to cope through the use of anti-anxiety stimuli (counter-conditioning), versus graduated exposure without the use of anti-anxiety stimuli. The findings suggest that, over time, the intervention appeared to be successful in helping the children to increase their parasympathetic arousal and thus physiologically calm themselves or cope with their anxiety. This supports findings that higher RSA is associated with fewer problem behaviors in ASD (Van Hecke et al. 2009). It is important to note that both baseline and intervention sessions involved exposure to the feared stimulus. It is therefore not surprising that this exposure alone may have had its own treatment effect (even during baseline sessions). In this case, it appears that number of sessions negatively predicted HR, such that the more sessions a child participated in (whether other interventions were used or not), the lower the HR dropped in the presence of the feared stimulus. The lack of a significant interaction term in these models suggests that this was not significantly related to the presence of our intervention. Thus, it appears that it may be possible to reduce HR (and potentially anxiety) in children with ASD and IDD merely through repeated exposures to feared stimuli. However, our intervention may have promoted coping, reflected in improvements in RSA as well as substantial behavioral improvements.

One limitation of the present study is that we did not include assessment measures or methods to discriminate between anxiety and fear, or 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" observed in this study. 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 anxious behavior *before* they were actually exposed to the discriminative stimuli (i.e., birthday cake with lit candles or singing happy birthday, right/left turns, parents leaving the house). It is possible that the children were appraising stimuli as being *potentially* harmful in the future and therefore preparing themselves for an upcoming negative event (Gallo et al. 2008), which would constitute a general appraisal of environmental threat that is more consistent with anxiety. Given that the children in this study, at times, appeared to anticipate the occurrence of negative events before the discriminative stimuli were actually presented, this suggests that the term "anxiety" is in fact an appropriate construct to apply to participants in this study. Nevertheless, future research is warranted to study these constructs more carefully, particularly in children with ASD and IDD. Future studies using a multimethod assessment to evaluate anxiety might benefit from the additional inclusion of a standardized psychiatric questionnaire or interview that is validated in youth with HFA, such as the ASD addendum to the *Anxiety Disorders Interview Schedule* (*ADIS/ASA*; Kerns et al. 2016). However, the use of most questionnaires and interviews to assess anxiety may be limited to higher-functioning youth with ASD (Moskowitz et al. 2017) who have better communication skills than the individuals in the current study.

Another limitation of this study is that, in using multicomponent intervention plans in which we implemented many

interventions at the same time, we cannot ascertain the unique contribution made by each intervention. We did not attempt to address which intervention components were effective in reducing anxiety and problem behaviors. It is unclear whether all of the interventions were necessary for change or whether a more limited treatment package would have been as effective (Nock 2002). This limitation could be addressed in future research by conducting analyses such as a component analysis and dose analysis to determine which components are necessary and how much of each component is necessary, including an examination of the minimum number of components necessary to produce immediate change and yield long-term effects. That said, as we noted earlier, each of the intervention procedures we used in this study has been demonstrated in previous research to be effective in reducing problem behavior in children with ASD or IDD and/or reducing anxiety in NT children or those with HFA. Further, the use of multicomponent rather than single-component interventions is currently considered to be best practice for individuals with IDD (e.g., Carr et al. 1999; Lucyshyn et al. 2007).

Related to this question of which intervention components were necessary, most experts in the field of anxiety would agree that certain treatment components (e.g., gradual exposure to feared stimuli) are a critical element of CBT and are therefore necessary for positive treatment outcome (e.g., Kendall et al. 2005). Likewise, 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 IDD, and could arguably be considered the primary treatment components. In the present study, however, it is worth noting that Jon, Ben, and Sam were each exposed to their feared stimulus/situation during baseline observations for 4, 5, and 7 exposures, respectively, without showing significant habituation in terms of their anxious behaviors, problem behaviors, subjective ratings, or RSA. While additional repeated trials of exposure could have been implemented and may have eventually resulted in habituation, this strategy may have been quite distressing for the children. Instead, our clinical observations suggested that pairing the anxiety-provoking stimulus/situation with an equally potent or even more powerful perseverative stimulus (e.g., Sesame Street, Dr. Seuss) appeared to compete with or counteract the aversiveness of the anxiety-provoking situation in a way that exposure alone may not have, or at least may not have as quickly. Further, these effects were accomplished immediately within a short amount of time (i.e., limited number of sessions) and may therefore be considered a treatment of choice in a variety of real-world settings.

These results are in line with previous research that showed no effects from exposure alone for a 14-year-old neurotypical boy with an insect phobia, but a marked effect when exposure was combined with contingent rewards (Jones and Friman 1999). This suggests that positive reinforcement may sometimes be needed to "surmount the presumed negative reinforcement from escape or avoidance of phobic stimuli" (Jones and Friman 1999). Although counterconditioning using "antianxiety stimuli" (Luscre & Center, 1996) would be considered distraction in the CBT literature and therefore might be contraindicated in the treatment of anxiety for NT individuals (Foa and Kozak 1986), we do not know if this is as true for children with ASD and IDD, 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. Further, anecdotally, distraction did not appear to be a factor for the participants in this study. For example, when Sam's parents made a left or right turn in the car during intervention sessions, Sam often turned his head around to view the intersection where they had turned (which was coded as an anxious behavior), but he appeared otherwise calm and relaxed and did not display any problem behavior. This suggested that Sam was still aware of the left/right turn (i.e., was not distracted from the turn) but that his anxiety and problem behavior were in fact attenuated by the intervention underway. Future research should be conducted to compare an "exposure paired with anti-anxiety stimuli" (i.e., counterconditioning) to an "exposure-alone" condition in children with ASD and IDD.

Another limitation of the present study is that, although exposure and counterconditioning/generalized reinforcement using the participant's perseverative interests were implemented for all three participants in 100% of intervention sessions, and escape extinction was used in every intervention session for which problem behavior or anxious behavior was present, not all of the other intervention strategies were used in every intervention session across all three participants (see sect. "Method" and Appendix A). Further, because the treatments were individualized to each particular child and family, not all sessions were standardized to the same degree within each child, which may have weakened our internal validity. For example, for Ben, although there was a relatively constant level of exposure across all sessions, the person(s) with whom he was left at home varied, as did the duration of Ben's separation from his parents (depending on the location to which Ben's parents drove to run errands), given that we attempted to keep the situations as ecologically valid as possible. Future research may want to focus on using a standardized protocol across participants, but with modifications in the context, type of stimuli and reinforcers used to enhance effectiveness and strengthen the internal validity of the interventions.

While we collected data in naturalistic settings, which is a strength of our study, it is also a limitation in that variability in naturalistic settings is more difficult to control. There were many instances of equipment malfunction (with the webcam, laptop, and especially the HR monitor) and movement artifacts, which resulted in limited usable data

for the children, particularly limited HR/RSA data [i.e., five sessions for Jon, six sessions for Ben, and four sessions for Sam did not have enough usable interbeat interval (IBI) data to be analyzed for HR and RSA]. Related to this limitation, one possible influence on HR and RSA was motor movements, which we did not analyze. Although Porges et al. (2007) found that intense activity accompanying physical exercise did impact RSA and IBI in children, low-intensity motor movements did not affect RSA or IBI. Further, it is important to note that we found differences in all three components of anxiety (behavioral, physiological, and affective/contextual) for all three children between baseline and intervention, suggesting that the differences between baseline and intervention are not just due to physiological factors.

A final limitation of this study is that we did not assess generalization and maintenance of the intervention effects. Thus, it is unclear whether the improvements in the participants' responses to identified anxiety-provoking contexts would transfer to other anxiety-provoking situations. It is also unclear whether the interventions were successful in making long-term changes that ultimately impacted the family's quality of life. Related to this, although our study used PBS prevention strategies (altering setting events and antecedents) and consequence strategies, we did not use PBS teaching strategies in which we actively taught replacement skills, which could aid in maintenance and generalization. Although we provided Ben with models of coping statements, we never required him to use such coping self-talk or measured the use of his coping self-talk. Future research should assess the use of such replacement behaviors. We also did not teach replacement skills such as functional communication training (FCT) or relaxation skills. We decided not to use FCT as a first-line treatment because teaching children with ASD and IDD to ask for a break from the anxiety-provoking situation could possibly interfere with exposure and habituation to that situation. However, clinically, teaching children to ask for a break to escape an anxiety-provoking situation (i.e., FCT) and then fading the number of opportunities to ask for a break over time has been a helpful strategy for individuals with ASD and IDD whose severe aggression or self-injurious behavior prevented us from using standard exposure without providing the opportunity to ask for a break. More research into using FCT and other replacement strategies in the treatment of anxiety is warranted for individuals with ASD and IDD. Future studies should expand upon teaching coping skills by including FCT and self-management strategies (an active role) rather than having to wait for a parent or teacher to alter the environment or initiate an intervention (a passive role).

Friman, Hayes, and Wilson (1998) note that, "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, we would argue that there is theoretical value and clinical value in applying the term "anxiety" to this population. Interventions to treat anxiety in neurotypical 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 et al. 2006). Although the interventions for both populations share similar roots in operant and classical conditioning, the intervention research has been reported in two separate literatures—either the mental health or clinical psychology literature for NT children with anxiety disorders or DBDs (e.g., *Journal of Consulting and Clinical Psychology*) or the ASD/IDD literature (e.g., *Journal of Autism and Developmental Disabilities*) or ABA/PBS literature (e.g., *Journal of Applied Behavior Analysis, Journal of Positive Behavior Interventions*) for children with ASD. This has resulted in little cross-fertilization between the two bodies of research (Brookman-Frazee et al. 2006). Applying the term "anxiety" to children with ASD and IDD may help to facilitate cross-fertilization and the transfer of knowledge between these two important bodies of research. Recognizing anxiety in children with ASD and IDD can help parents, teachers, clinicians, and researchers to regard problem behavior as potential signs of anxiety (i.e., escaping anxiety) rather than noncompliance, disobedience, or defiance (i.e., escaping demand). More important, identifying anxiety in children with ASD and IDD 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.

Author Contributions Dr. Lauren Moskowitz conceptualized, designed, and implemented the study, analyzed the behavioral data, drafted the initial manuscript, reviewed and revised the manuscript, and approved the submission of the final manuscript. Dr. Caitlin Walsh was the primary coder of anxious behavior and problem behavior, assisted with conceptualizing the study, reviewed and revised the manuscript, and approved the submission of the final manuscript. Dr. Emile Mulder analyzed the HR and RSA data, carried out the statistical analyses for the physiological data, and drafted the results for the physiological data. Dr. Darlene Magito McLaughlin helped design and conceptualized the study as well as reviewed and revised the manuscript. Dr. Greg Hajcak supervised the physiological data collection and analysis and reviewed the manuscript. The late Dr. Edward G. Carr (Ted Carr) was instrumental in helping to design and conceptualize the study. Dr. Jennifer Zarcone helped to conceptualize the study and analyze the behavioral data as well as played a major role in reviewing and revising several drafts of the manuscript and approving the final manuscript as submitted.

Appendix A

See Table 1.

Table 1 Treatment Components For Each Participant

	Jon	Ben	Sam
CBT strategies			
Graduated exposure	Exposed to happy birthday	Exposed to parents leaving house	Exposed to left/right turns in car
Psychoeducation	Conveyed through video modeling	Conveyed through Social Story	Conveyed through Social Story
Cognitive restructuring	Conveyed through video modeling	Conveyed through Social Story	Conveyed through Social Story
ABA/PBS antecedent-based strategies			
Increasing predictability	Video modeling Priming	Social Story Priming	Visual schedule Social Story Video priming
Providing choices	N/A	Chose which activity to engage in while parents out	Chose items to pack in "Car Bag" and, when possible, destination
Generalized reinforcement/counterconditoning	Pairing presentation of birthday cake and song with Sesame Street	Pairing departure of parents with Muppet movie, swings, counting	Pairing riding in car with audio recording of "The Sneetches"
Incorporating perseverative interests	Incorporated Sesame Street	Incorporated Muppet movie, swings, & directing adult to engage in counting with specific hand motions	Incorporated Dr. Seuss
Prompting an approach response	After 3rd int. session, prompted to attempt to blow candle	N/A (no approach required for Ben)	N/A (no approach required for Sam)
ABA/PBS consequence-based strategies			
Positive reinforcement contingent upon approach	Provided with reinforcer when he attempted to blow out candles in cake	N/A (no approach required)	N/A (no approach required)
Differential reinforcement of alternative behavior (DRA)	N/A	Attention for anxious behavior minimized, while attention for brave behavior	N/A
Escape extinction	Parents no longer stopped singing happy birthday	Parents no longer terminated their departure	Parents no longer deviated from the left/right turn

Appendix B

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 lightning. 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 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 or Grandma and Grandpa.

Text of Sam's Social Story (for Left/Right Turns in Car Ride) Based on Dr. Seuss' *The Foot Book*

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.

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