

EFFICACY OF A BRIEF INTERVENTION TARGETING A LOW RESPONDER
PROFILE IN PRESCHOOLERS WITH AUTISM SPECTRUM DISORDER

by

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This dissertation is dedicated to my sweet, sweet boy,
my son, Charles Hunter Cormier.

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Abstract

Background: Autism spectrum disorder (ASD) is characterized by impaired social communication, and repetitive behavior and restricted interests. The need for effective service delivery models is urgent, as rates of ASD diagnosis continue to be high (Ofner et al., 2018). However, heterogeneity of symptom severity and functional impact makes effective treatment for all preschoolers difficult. Individual-level data from Nova Scotia’s early intervention program based on Pivotal Response Treatment (PRT; Bryson et al., 2007) show that some children make minimal treatment gains (Smith, Flanagan, Garon, & Bryson, 2015). These “low responders” have a distinct behavioral profile that includes low levels of *toy contact* and *positive affect* (Fossum, Williams, Garon, Bryson, & Smith, 2018)—potentially modifiable variables.

Objective: The study aim was to generate efficacy data for a brief therapist- and parent-implemented intervention targeting these skills that low responders lack, the Pivotal Response Intervention-Minimal Responder (PRIMeR) Intervention.

Methods: Four preschoolers with ASD (aged 3.5–4.5 years) participated in this single case experimental design study. The 12-week intervention, using Reciprocal Imitation Training (Ingersoll, 2010) to target toy play, and responsiveness training (Landry, Smith, & Swank, 2006) to boost shared positive affect, was delivered in each child’s natural environment, at 2–3 hours per week, plus a one-month follow-up. Parents were coached in responsiveness intervention strategies.

Results: An intervention effect was obtained for shared positive affect, variety of functional play actions, and variety of functional play objects, but not frequency of functional play. This study provides initial support for the efficacy of this partially parent-mediated naturalistic and developmental treatment package in shifting preschoolers’ low responder profiles by boosting skills in areas that are theoretically important for optimal response to PRT-based programs such as NS EIBI.

Conclusions/Importance: The current intervention remediates behavioral deficits associated with poor progress in treatment. Individualization of children’s programming within PRT, by including PRIMeR, may optimize children’s progress and thereby produce positive outcomes for a wider range of children.

Keywords: autism spectrum disorder; early intervention; imitation; affect; single case experimental design.

List of Abbreviations Used

AAC	Augmentative alternative communication
ABA	Applied behavior analysis
ADI-R	Autism Diagnostic Interview – Revised
ADOS-2	Autism Diagnostic Observation Schedule, 2 nd Edition
APA	American Psychiatric Association
ASD	Autism spectrum disorder
BAP	Broader autism phenotype
CBQ	Children’s Behavior Questionnaire
CI	Confidence interval
DSM	Diagnostic and Statistical Manual of Mental Disorders
DTT	Discrete trial training
EIBI	Early intensive behavioral intervention
EMT	Enhanced Milieu Teaching
ESDM	Early Start Denver Model
FQOL	Family quality of life
ICC	Intraclass coefficient
ImPACT	Improving Parents as Communication Teachers
IOA	Inter-observer agreement
IQ	Intelligence quotient
JASPER	Joint Attention Symbolic Play Engagement and Regulation
MB-CDI	MacArthur-Bates Communicative Development Inventory
M-P-R	Merrill-Palmer-Revised

NDBI	Naturalistic developmental behavioral intervention
NRC	National Research Council
NS	Nova Scotia
PACT	Preschool Autism Communication Trial
PEAR	Play, Engagement, and Affect Ratings (protocol)
PECS	Picture Exchange Communication System
PLS-5	Preschool Language Scales, 5 th edition
PRIMeR	Pivotal Response Intervention Minimal Responders
PRT	Pivotal Response Treatment
RCT	Randomized controlled trial
RIT	Reciprocal Imitation Training
RPMT	Responsive Education and Prelinguistic Milieu Teaching
RRB	Restricted, repetitive behavior
SCED	Single case experimental design
SD	Standard deviation
SGD	Speech-generating device
SMART	Sequential multiple assignment-randomized trial
TAU	Treatment as usual
UCLA	University of California Los Angeles
VABS	Vineland Adaptive Behavior Scales
WWC	What Works Clearinghouse

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Chapter 1: Introduction

Autism spectrum disorder (ASD) is a conceptual model and diagnostic label used to describe a pattern of developmental differences that manifest early in childhood. These differences are related to social communication and social interaction, as well as to patterns of behavior and interests which tend to be restricted and repetitive. Criteria in the Diagnostic and Statistical Manual of Mental Disorders (DSM-5; American Psychiatric Association [APA], 2013) pertaining to differences in communication and behavior are sufficiently sensitive and specific such that ASD can be reliably diagnosed by experienced clinicians using a combination of an observation schedule (e.g., Autism Diagnostic Observation Schedule, 2nd edition, ADOS-2; Lord et al., 2012), a caregiver interview (e.g., Autism Diagnostic Interview, Revised, ADI-R; Rutter, LeCouteur, & Lord, 2003) and clinical expertise (Falkmer, Anderson, Falkmer, & Horlin, 2013).

Heterogeneity in symptoms experienced by individuals who have ASD is omnipresent. Children are affected, to varying degrees, by the symptom clusters that characterize this neurodevelopmental disorder: impairments in social communication and social interaction such as differences in social approach; impaired back-and-forth conversation; reduced sharing of interests, emotions, and affect; poor nonverbal communication; poor imaginative play; as well as difficulty understanding and maintaining relationships, and making friends (APA, 2013). Children with ASD also experience different levels of severity of repetitive behavior and restricted interests and activities, e.g., insistence on sameness; rigid thinking patterns; perseverative interests; and hyper- and/or hypo-reactivity to sensory input (APA, 2013). Commonly observed are stereotyped movements and use of objects, such as lining up toys, and/or stereotyped

speech, such as echolalia. The impact of symptoms varies with respect to adaptive functioning, self-regulation of emotion and behavior, as well as cognitive and language development (APA, 2013). Early intervention programs are regarded as important for mitigating disability (Elder, Kreider, Brasher, & Ansell, 2017); however, outcomes for children are highly variable. Understanding this variability is of critical importance for clinical, research, and policy reasons. As the prevalence of ASD is high, estimated to be 1 in 66 in Canada among youth aged 5–17 (Ofner et al., 2018), the need is urgent for effective service delivery models that can be individualized to each child’s specific needs.

The recognized need to optimize children’s outcomes by delivering intervention based on children’s individual characteristics serves as the motivation for my dissertation. Using a specific early intervention model, I address the challenge of variability in treatment progress, and contribute to the growing literature on individualizing children’s treatment paths based on children’s pre-treatment individual characteristics/symptom profiles.

In Chapter 2 of this dissertation, I will summarize the literature on the heterogeneity observed in ASD etiology and symptomatology, as well as in developmental pathways of individuals with ASD. I will then provide an overview of research pertaining to early intervention for ASD. I will first discuss conventional early intensive behavioral intervention (EIBI), its components and variations, as well as the evidence supporting its use in the population of preschoolers with ASD. I will then provide background on early intervention programs that are based on the same behavioral principles as EIBI, but which are also founded in developmental science, hence named

naturalistic developmental behavioral interventions (NDBIs). After describing the evidence for the use of NDBIs, my focus will then shift to the discussion of the variability observed in children's outcomes following participation in established interventions, as well as individual predictors of treatment progress. As part of the background on NDBIs, I will include an overview of the specific intervention model used in relation to my study intervention, Pivotal Response Treatment (PRT). An empirically derived predictor profile associated with differential outcomes within a PRT-based program served as the specific impetus for my dissertation. I will then outline the "active ingredients" that have been identified for some treatment models for ASD before discussing individualizing children's treatment to enhance outcomes. Based on this literature, I will discuss the selection of intervention targets as part of the development of an intervention package designed with the PRT predictor profile in mind. Concluding my background section (Chapter 3), I will provide an overview of the context for my intervention package, which is designed to enhance response to a specific publicly-delivered model of PRT-based intervention. I will describe its development, including rationale for its components, before offering my dissertation's purpose, specific objectives, and hypotheses. As part of Chapter 4, I will describe my participants, measures used, dependent variables, and procedure, as well as my single case experimental design (SCED) and corresponding data analytic strategy. In Chapter 5, I will describe my results and report statistics related to inter-rater reliability and fidelity of implementation. Lastly, as part of Chapter 6, I will discuss my findings in the context of the early intervention literature, provide study limitations, as well as offer steps for further research.

Chapter 2: Background

Heterogeneity in ASD

Heterogeneity in etiology and symptomatology. Autism spectrum disorder (ASD) is widely accepted as a complex neurodevelopmental disorder. There is little dispute among researchers that ASD is inherently heterogeneous and multifaceted, encompassing multiple etiologies and developmental trajectories (Geschwind, 2011). Although a common *pattern* of differences (i.e., in communication and behavior) is observed as part of the ASD behavioral phenotype, heterogeneity is observed both *intrapersonally*, among features manifested and their level of severity, which also changes over time (Georgiades, Bishop, & Frazier, 2017), and *interpersonally*, reflecting important differences between individuals with ASD. Heterogeneity is evident at multiple levels of analysis – in genetics, neurology, cognition, emotion, behavior, and adaptive functioning (Lombardo, Lai, & Baron-Cohen, 2019). Not surprisingly, as individuals with ASD present differently to the world from one another, they thus have different needs.

The factors that produce observed heterogeneity are multifarious. Heterogeneity stems from multiple etiological pathways related to genetic predispositions, environmental impacts, and the interaction of the two, i.e., epigenetics (Siu & Weksberg, 2017). Between 400 and 1,000 genes have been implicated in susceptibility to ASD, with genetic abnormalities contributing to approximately 20% of diagnosed ASDs (Bergbaum & Ogilvie, 2016; Rylaarsdam & Guemez-Gamboa, 2019; Yoo, 2015). Through large-scale genetic studies of thousands of individuals with ASD, we know that specific genetic mutations differentially influence ASD symptoms (Webb et al., 2017). The sequencing of

DNA of children with ASD has shown to be a fruitful avenue of research. For example, Autism Speaks has organized the sequencing of 7,000 genomes, to date, in an effort to identify subtypes of ASD so that more personalized and effective treatments can be developed (MSSNG, 2020). Research using this genomic database has thus far identified 61 genetic variations that affect risk for ASD. Recently, as part of the largest whole genome study of ASD, investigators identified 18 new autism-linked genes, some of which affect biochemical pathways in the brain and may be future targets for medicine (Yuen et al., 2017). Environmental factors that have been identified as conferring risk of ASD include maternal immune activation in response to an infection including fever (Spann, Sourander, Surcel, Hinkka-Yli-Salomaki, & Brown, 2017), decreased levels of neurotrophic factors in the neo-natal period (Abdallah et al., 2013a; 2013b), and environmental exposure to high levels of air pollution (Goodrich et al., 2018; Kim et al., 2017). Consequently, variability in behavioral phenotype is prevalent and affects response to treatment, which will be discussed in detail later.

Heterogeneity in developmental pathways/outcomes (natural history).

Longitudinal studies of individuals with ASD indicate that developmental trajectory varies, as do prognoses. By virtue of being a neurodevelopmental disorder, ASD is considered to be a lifelong condition, characterized by persistent differences in language, social skills, communication, adaptive functioning, and educational attainment. However, variability is great, to the extent that for a small proportion of cases, symptoms become so mild that individuals no longer meet diagnostic criteria (“optimal outcome”; Fein et al., 2013). Efforts are needed to better understand the common patterns of change across children with ASD and the paths of children who appear to do better or worse than

expected. We know that some children who are severely affected when young fare better than anticipated later in life in terms of adaptive functioning. Conversely, some children who do not appear to be as strongly affected as young children have more difficulty than expected later in adolescence (Szatmari et al., 2015).

Large, longitudinal studies have statistically identified subgroups of individuals with ASD who differ with respect to developmental trajectories (Fountain, Winter, & Bearman, 2012; Lord, Bishop, & Anderson, 2015; Pickles, Anderson, & Lord, 2014; Szatmari et al., 2015). Variability is evident in developmental pathways spanning toddlerhood to adolescence. Fountain et al. (2012) followed almost 7,000 children with ASD from diagnosis at approximately 2 years of age until these individuals were 14 years old. Using group-based latent trajectory modelling, they identified six heterogeneous subsets of symptom trajectories, each subset with respect to a) communication abilities, b) social functioning, and c) level of repetitive behavior over time. With respect to the six communication trajectories, approximately 30% of the sample fell into each of the “low functioning” and the “low-medium functioning” trajectories. A nearly identical proportion of the sample fell into these two lowest (both ‘low’ and ‘flat’) trajectories for social functioning. The authors found substantial heterogeneity in social and communication development, as some children improved much more than others. Interestingly, they identified a “bloomers” group along both the communication and social dimensions. Children in this group started with low scores, comparable to those on the low functioning trajectories, yet improved quickly. Investigators concluded that intellectual disability and socioeconomic factors separated this group from other children. Those most likely to “bloom” were without intellectual disability and with more

educated, non-minority mothers (perhaps reflecting more advantaged home/neighborhood environments and corresponding quality and intensity of supports available). Notably, the most rapid development with respect to communication and social functioning occurred before age 6 years, after which trajectories tended to flatten. This finding highlights the importance of intervening early in development to influence later outcomes.

Other groups have also reported clear diversity in children's developmental trajectories in the critical period of early childhood. Szatmari et al. (2015) from the Canadian "Pathways in ASD" study examined trajectories of children's symptom severity and adaptive functioning from age of diagnosis (at 3.25 years, on average) to 6 years, using four measurement occasions. With respect to severity of symptoms over time, two trajectories were found. Approximately 89% of children had more severe symptoms and a stable trajectory, while approximately 11% showed an improving trajectory (and less severe symptoms). For adaptive functioning, three trajectories were observed: approximately 29% had lower functioning and a worsening trajectory; almost 50% had moderate functioning and a stable trajectory [associated with higher baseline intelligence quotient (IQ) and language], and 20% had higher functioning and an improving trajectory (associated with earlier age at diagnosis). These findings add to the body of literature that recommends early intervention (i.e., prior to age 6 years) tailored to each child's needs and strengths (Szatmari et al., 2015). Heterogeneity in developmental trajectory, especially with respect to optimal outcomes, suggests that children's phenotypes are sensitive to environmental variables (see Szatmari et al., 2015). The goal of early intervention is to influence children's trajectories to enhance prognosis.

Early Intervention for ASD

Early intervention, i.e., delivered during the preschool period, is important for children's acquisition of language, communication skills, and adaptive functioning skills (Narzisi, Costanza, Umberto, and Filippo, 2014; Reichow et al., 2018). Early intervention is also key to the prevention of the development of secondary symptoms such as aggression, behavioral dysregulation, and self-injurious behavior. This can be accomplished, for example, by teaching children functionally equivalent replacement behaviors to communicate their wants and needs (Horner, Carr, Strain, Todd, & Reed, 2002). Ameliorating core ASD symptoms through early intervention, as well as preventing secondary symptoms, reduces the need for some behavioral, psychological, and other supportive services later in life, rendering early intervention potentially cost effective over the lifetime of the individual (Rogge & Janssen, 2019). In this way, early intervention increases the likelihood of improved long-term outcomes for children and their families.

Conventional model of early intensive behavioral intervention (EIBI). Early intensive behavioral intervention (EIBI) implemented early in the preschool period is considered to be a well-established form of treatment for reducing behavioral symptoms associated with ASD and improving functional abilities (Reichow et al., 2018). Early intensive behavioral intervention is a specific model of behavioral intervention, rooted in principles of applied behavior analysis (ABA), that uses a highly structured teaching approach for children usually younger than 5 years. Applied behavioral analysis constitutes a formal scientific discipline surrounding the application of techniques based on the principles of learning, to change socially significant behavior (Cooper, Heron, &

Heward, 2019). The empirical origins of EIBI can be traced to the work of Ivar Lovaas, who developed and tested the University of California at Los Angeles (UCLA) Young Autism Project model, or “Lovaas model” (Lovaas, 1981; Lovaas, 1987), as an intervention protocol for children with ASD. Defining features of the UCLA/Lovaas model include 1) a highly specific teaching procedure that involves systematically following a treatment curriculum/manual dictating specific tasks to be taught and in what order (e.g., Lovaas, 2003); 2) the use of a 1:1 adult-to-child ratio; 3) implementation, either at home or school, for 20 to 40 hours per week for one to four years (Eikeseth, 2009; T. Smith, 2010); 4) systematic application of behavioral principles, e.g., with respect to antecedents and consequences of children’s behavior, prompts, prompt fading over successive presentations of a learning task until error-free learning has occurred, and task analysis (breaking down complex behaviors into component behaviors) across a range of domains; and 5) discrete trial training (DTT) spanning various targets. Discrete trial training procedures include a specific teacher-child instructional sequence whereby an operationally defined learning target is identified (e.g., imitation, expressive language, self-help skills; see Klintwall & Eikeseth, 2014) and behavioral procedures are systematically repeated until the target is mastered, i.e., the skill can be performed without error to a pre-specified criterion level. Reinforcers for correct performance typically involve an item or activity that the child enjoys such as stickers, food, or listening to music. Once the child has acquired the skill, it is then combined with other targets to produce more complex tasks, or the complexity of the original target behavior is increased in a stepwise fashion (Klintwall & Eikeseth, 2014). Guidelines for comprehensive treatment for ASD that were outlined by the National Autism Center

(2015) map on to EIBI, in that it a) addresses the core differences as part of ASD; b) is delivered in structured, predictable settings, c) has a low child-to-therapist ratio, d) includes programming for generalization and maintenance of acquired skills, e) involves families in planning and sometimes implementation, f) uses a functional behavioral approach to address children's challenging behavior, and g) monitors progress over time using ongoing data collection.

Variations of conventional EIBI. Conventional EIBI as described above, i.e., intervention that uses highly structured ABA procedures such as DTT, has limitations. For example, the generalization of behavior taught within the structured DTT settings to natural situations can be limited (Klintwall & Eikeseth, 2014). Incidental teaching is often used to complement DTT (Delprato, 2001; Goldstein, 2002; McGee & Daly, 2007); it follows the same learning principles but occurs in children's natural environments such as community settings (park, stores, school) and allows learning opportunities to be initiated by the child's interest in an object or activity, i.e., naturally occurring "incidents". Incidental teaching helps to prepare children for natural environment teaching during the maintenance phase of conventional EIBI programs (Klintwall & Eikeseth, 2014). Acknowledging that teaching within the routines in which children will be required to use acquired skills (i.e., in natural environments) is important, ABA procedures such as prompting, prompt fading, and behavioral reinforcement, are utilized during children's daily routines such as mealtimes, getting dressed, and play.

Evidence for efficacy/effectiveness of conventional EIBI. Expert consensus on the importance of early intervention has been long established (National Research Council [NRC], 2001). Reichow and Wolery (2009) published a meta-analysis specific to

13 studies of EIBI based on the UCLA Young Autism Project (i.e., conventional EIBI); these studies varied in experimental design. The authors concluded that, based on generally positive effect sizes, EIBI was an effective treatment for children with ASD to address multiple dimensions of functioning. Eldevik and colleagues (2009) replicated and extended Reichow and Wolery's (2009) meta-analysis, compiling data from nine studies and using stricter inclusion criteria and a more precise definition of EIBI. Their findings also supported EIBI's effectiveness. Contrary to these two studies' findings were results of a systematic review and meta-analysis of ABA-based intervention using DTT with preschool children with ASD published the same year (Spreckley & Boyd, 2009). Included were randomized controlled trials (RCTs) and quasi-RCTs as compared to standard care. Spreckley and Boyd (2009) reported that there was inadequate evidence that ABA-based intervention had better outcomes than standard care for children with ASD, in part due to the fact that only four of the 13 reviewed studies were included in their meta-analysis (because of insufficiently powered samples – reflecting the sparse state of the literature just a decade ago). Virues-Ortega (2010) investigated the effectiveness of comprehensive ABA-based treatments through meta-analysis of 22 intervention studies (RCTs and quasi-RCTs; half of studies were based specifically on the UCLA model). Virues-Ortega (2010) concluded that EIBI led to positive, medium-to-large effects on language development, and intellectual, social, and adaptive functioning in children with ASD. Lastly, Makrygianni and Reed (2010) published a meta-analytic review of the effectiveness of conventional EIBI ('behavior analytic treatment' or replications of the UCLA model). They included more studies than Spreckley and Boyd (2009), and more developmental outcomes and measures of effectiveness than Eldevik et

al. (2009). Further, this review limited inclusion to moderate and high quality group-design studies and excluded single case experimental designs (SCEDs). Makrygianni and Reed (2010) found that EIBI was effective for children with ASD (54 months old at treatment onset, on average), that children's intellectual and adaptive functioning, language, and social abilities improved post-intervention (moderate to high effects), and that EIBI was more effective than comparison eclectic intervention programs with respect to those same outcomes. Similarly, Klintwall, Eldevik, and Eikeseth (2015) analyzed data from group studies comparing EIBI and controls (from the database used by Eldevik et al., 2010). Participants in the EIBI group (N = 295) exhibited 75% faster learning rates than those in the control group (N = 135) with respect to mean IQ (changes in IQ age-equivalents between intake and follow-up divided by the duration of the intervention, i.e., the slope of the developmental trajectory), and 38% faster learning rates (N = 284) as compared to the control group (N = 112) for mean adaptive behavior learning rate. Reichow, Barton, Boyd, and Hume's (2012) Cochrane review of some of the aforementioned meta-analyses demonstrated overall positive effects for EIBI treatment compared to treatment as usual (TAU) for all outcomes: adaptive behavior, IQ, expressive language, receptive language, and daily communication skills (Vineland Adaptive Behavior Scales [VABS] Communication domain). Authors highlighted that most studies exploring EIBI have been non-randomized, and therefore indicated that the overall quality of evidence was 'low'. Reichow, Hume, Barton, and Boyd (2018) updated their Cochrane meta-analytic review to examine whether additional evidence could be identified on the effect of EIBI on young children with ASD using five RCTs and controlled clinical trials. Their results provided weak evidence that EIBI improves

adaptive behavior and autism symptom severity, IQ, expressive and receptive language, everyday communication skills, everyday social competence, daily living skills, and problem behavior. Evidence was described as ‘weak’ due to the quality of the evidence (rated as low to very low). In sum, empirical evidence culminating in several independent meta-analyses generally supports that early intervention (i.e., prior to age 4 years) leads to gains in many areas of functioning and that intense intervention, i.e., 20-40 hours per week for one to two years, leads to beneficial outcomes (Makrygianni & Reed, 2010; Peters-Scheffer, Didden, Korzilius, & Sturmey, 2011; Reichow, 2012; Virues-Ortega, 2010). However, caution is warranted in our confidence of strong associations between EIBI and outcomes, since the quality of evidence is not high (Reichow et al., 2018). Behavioral interventions based on the principles and techniques of ABA have been repeatedly cited as having strong evidence for effectiveness in ASD treatment (NRC, 2001; Weitlauf et al., 2014), despite the caveats just described. Moreover, gains in cognitive and adaptive functioning during EIBI have been shown to be maintained 10 years after EIBI has ended (T. Smith, Hayward, Gale, Eikeseth, & Klintwall, 2019).

Naturalistic developmental behavioral interventions (NDBIs). Evidence-based early behavioral interventions for preschoolers with ASD are increasingly also founded in developmental theory and combine naturalistic and behavioural methods (Dawson & Bernier, 2013; Schreibman et al., 2015). Through iterative processes of synthesizing research and clinical expertise within the domains of early behavioral intervention research and development sciences, a considerable number of manualized interventions that target the social communication and play skills of young children with ASD have been created and subsequently empirically validated. Many such early intervention

packages share the same theoretical underpinnings and thus have been classified as naturalistic developmental behavioral interventions (NDBIs; Schreibman et al., 2015; T. Smith & Iadarola, 2015). These interventions draw upon evidence-based practices such as use of strategies based in ABA, e.g., prompting, prompt fading, the three part contingency (antecedent, behavior/response, consequence such as reinforcement), and incidental teaching techniques such as capitalizing on child-initiated learning opportunities and using natural consequences (Odom, Collet-Klingenberg, Rogers, & Hatton, 2010).

Naturalistic interventions are more consistent with the tenets of developmental science and early intervention, including an emphasis on children's learning within actively engaged, socially laden, play-based environments that are inherently less restrictive than conventional EIBI instructional settings (Iovannone, Dunlap, Huber, & Kincaid, 2003; NRC, 2001; Schreibman et al., 2015). A common goal of NDBIs is to help the child to develop interpersonal connections that will lead to the mastery of skills/enhanced capacities in, e.g., engagement, play, and social communication. Developing such interpersonal connections is accomplished using a relationship-based model focused on interpersonal processes such as reciprocity and affective sharing. The idea behind developmental treatment approaches is that the development of a child with ASD can be positively influenced by fostering strong interpersonal relationships through natural play (NRC, 2001). In this way, it is believed that children with ASD can learn to engage appropriately with others, in part by building affective sharing and reciprocity (Rogers & Dawson, 2009). Developmental interventions derive from a philosophy whereby strategies are implemented within the context of child-directed interactions and

center around a child's interests, i.e., techniques are implemented within the context of following a child's lead based on their preferences and selected activities. Further, techniques are used in a naturalistic way, with learning opportunities embedded within children's typical environments and following everyday routines, in addition to using natural rewards to foster a child's motivation to communicate. Part of the success in creating natural learning opportunities for implementation of intervention is organizing the environment to facilitate communicative and social interactions.

Most NDBIs meet many criteria considered for qualification as a comprehensive intervention program according to the National Autism Centre's (2015) criteria, as described in an earlier section pertaining to conventional EIBI. Some NDBIs are more focused on social-communication and target these skills rather than a wider range of abilities. However, changes in behaviors not directly targeted during intervention, i.e., collateral gains, are often observed (Koegel, Koegel, & McNeerney, 2001; Ledbetter-Cho, Lang, Watkins, O'Reilly, & Zamora, 2017). Common elements across NDBI models include parent involvement/parent-mediated aspects of intervention, and parental sensitivity and responsiveness to children's communication (Brian, Smith, Zwaigenbaum, Roberts, & Bryson, 2016; Dawson et al., 2010; Ingersoll & Dvortcsak, 2010; Kasari et al., 2006; Koegel & Koegel, 2006; Prizant, Wetherby, Rubin, & Laurent, 2003; Rogers, Dawson, & Vismara, 2012). Naturalistic developmental behavioral intervention models are typically designed for young children (48 months of age and younger), and feature naturalistic environments, use of natural reinforcers, active family involvement, intensity of 12–36 hours per week, a focus on developmental skills, use of methods to promote generalization and maintenance of skills, individualized goals based on individual

strengths and needs, and planned transitions to school environments (see Schreibman et al., 2015, for an overview of NDBI). See Table 1 for a representative list of NDBIs for children with ASD considered to be evidence-based (APA Presidential Task Force, 2006).

Many of the interventions classified as NDBIs include a parent coaching component whereby parents learn to implement the intervention strategies with their children; some NDBIs are developed as exclusively parent-mediated models (see Table 1 for such NDBIs). Parent-implemented interventions constitute an established evidence-based practice (Odom et al., 2010). Results of RCTs suggest that children's developmental progress is accelerated when an intervention includes parent coaching (Rogers et al., 2019; Wetherby et al., 2014). Coaching involves actively engaging the parent in promoting skill acquisition or behavior change in their child—either as part of exclusively parent-mediated interventions or complementary to therapist-delivered interventions. Coaching may involve education and in-home sessions with therapist-guided parent coaching during parent-child interactions across a span of several weeks (when involved as an adjunct to therapist-mediated intervention; Bearss, Burrell, Stewart, & Scahill, 2015; Schultz, Schmidt, & Stichter, 2011). Parents are coached within a therapeutic approach to interact positively with and respond sensitively to their children. Such parental responsiveness is intended to be used in daily natural interactions and routines. Parent coaching and parent use of intervention strategies differs from the *training* of parents within conventional EIBI programs to act as their children's co-therapists (T. Smith, Groen, & Wynn, 2000).

Table 1

Widely Known, Empirically Supported NDBI Models for Children With ASD

Model	Component(s)/intervention target(s)
Pivotal Response Treatment (PRT; Koegel, Koegel, Harrower, & Carter, 1999; Koegel & Koegel, 2006)	<ul style="list-style-type: none"> - Pivotal skills as targets of treatment; following the child’s choice of activities; natural reinforcers - May include parent coaching
Project ImPACT (Improving Parents as Communication Teachers; Ingersoll & Dvortcsak, 2010; Ingersoll & Wainer, 2013)	<ul style="list-style-type: none"> - Parent training curriculum to improve social communication skills - Parent-mediated
Reciprocal Imitation Training (RIT; Ingersoll, 2010; Ingersoll & Schreibman, 2006)	<ul style="list-style-type: none"> - Children’s imitation and social engagement as targets
Learning Experiences and Alternative Program for Preschoolers and Their Parents (LEAP) and Denver Model (Rogers & Lewis, 1989; Strain & Bovey, 2011)	<ul style="list-style-type: none"> - Immersing children with ASD into preschools with typically developing peers who are trained on strategies to use in communicating and interacting with children with ASD
Early Start Denver Model (ESDM; Dawson et al., 2010; Rogers & Dawson, 2009)	<ul style="list-style-type: none"> - Positive affect via interpersonal exchange, shared engagement with materials/activities as targets, and use of adult responsivity and sensitivity to child cues - Parent intervention: Rogers et al. (2012)
Social Communication, Emotional Regulation, and Transactional Support (SCERTS; Prizant, Wetherby, Rubin, Laurent, & Rydell, 2003)	<ul style="list-style-type: none"> - Children’s communication and social-emotional functioning as targets and accompanying goal to support family interaction
Parent-Mediated Communication-Focused (Preschool Autism Communication Trial [PACT]) intervention (Green et al., 2010)	<ul style="list-style-type: none"> - Parental sensitivity and responsiveness to child communication - Includes a parent coaching component

Model	Component(s)/intervention target(s)
Joint Attention Symbolic Play Engagement and Regulation (JASPER; Kasari et al., 2006, 2008, 2010)	<ul style="list-style-type: none"> - Joint attention, symbolic play, social engagement, emotion regulation - Parent-implemented version: Kasari, Gulsrud, Wong, Kwon, & Locke (2010); Kasari, Paparella, Freeman, & Jahromi (2008)
Advancing Social-communication and Play (ASAP; Boyd et al., 2018): based on Kasari et al.'s interventions (2006; 2008) for implementation specifically within preschools	<ul style="list-style-type: none"> - Social-communication and play skills applicable to school settings
Social ABCs (Brian, Smith, Zwaigenbaum, Roberts, & Bryson, 2016)	<ul style="list-style-type: none"> - Language/communication skills and positive emotion sharing as targets - Parent-mediated

Evidence for efficacy/effectiveness of NDBIs. Naturalistic developmental behavioral interventions for children with ASD have garnered empirical attention. Although naturalistic interventions have been deemed an “evidence-based practice” for social and communication outcomes (Odom et al., 2010), the National Autism Center (2015) categorized social communication interventions, “Developmental Relationship-based Treatments”, and imitation-based intervention as “emerging” in their level of evidence review. Although effectiveness has not been established using multiple RCTs for some programs, an impressive evidence base has been documented for specific NDBI models. For example, a review (Rogers & Dawson, 2010) of 15 effectiveness studies of the Early Start Denver Model (ESDM) reported significant improvements in IQ, receptive and expressive language, as well as adaptive behavior in young children with ASD.

Inclusion of NDBIs in meta-analyses or reviews of efficacious (e.g., Seida et al., 2009) and effective interventions for young children at risk for or diagnosed with ASD

(Narzisi et al., 2014; Vismara & Rogers, 2010) has typically been combined with conventional EIBI and they comprised a minority of the studies reviewed. One review, however, by Yoder, Bottema-Beutel, Woynaroski, Chandrasekhar, and Sandbank (2013), included a majority proportion of NDBIs; 75% of the 24 reviewed studies used interventions that have been classified as NDBIs. Findings showed that treatments demonstrated strong evidence of effects for social communication outcomes in 54% of the 60 social-communication-dependent variables examined (across 23 independent samples). The likelihood that an intervention led to improvements in social communication varied as a function of two aspects: gains were more probable a) when social communication was targeted and b) when social communication was measured in contexts similar to the treatment sessions. In a second review, Fuller and Kaiser (2020) included both conventional EIBI and NDBIs in their meta-analysis of the effects of early interventions on social communication outcomes for preschoolers with ASD. Analysis of 29 studies (group experimental designs) comprising 1,442 children (786 intervention participants, 656 control participants) yielded significant effect sizes associated with social communication outcomes. It is important to note that documented positive effects within the early intervention literature typically relate to improvement of *short-term* outcomes; generalization of such effects within long-term development is lacking, as highlighted in a research review/commentary of psychosocial interventions for preschoolers with ASD (Green & Garb, 2018). To my knowledge, only one meta-analysis has been conducted looking exclusively at NDBIs. This meta-analysis included 27 experimental group-design studies (i.e., excluding SCEDs and pre-post designs; Tiede & Walton, 2019). Small, significant positive effects of NDBIs were found for play skills,

expressive language, and symptom reduction. Large effects were found for cognitive development and social engagement (Tiede & Walton, 2019). In sum, more effectiveness studies for NDBIs are needed, in addition to NDBI-specific meta-analyses. However, research in this area is progressing and results are promising.

Variability in Outcomes of Established Interventions and Individual Predictors of Progress During Treatment

Conventional EIBI. Although a range of therapeutic approaches has shown positive outcomes for children with ASD with respect to communicative and social abilities, not all children benefit from any single intervention model. Reports abound of significant differences in outcomes for individual children following intervention (Schreibman & Anderson, 2011; Yoder, 2010). As expected, individual pre-treatment differences in behavioral, communicative, and intellectual functioning contribute to variability in response to early intervention (Lord et al., 2005). Variability was highlighted in Eldevik et al.'s (2010) meta-analysis (discussed earlier with respect to effectiveness of conventional EIBI). They used data from 16 group outcome studies (from a total of 453 children, 68% of whom received behavioral intervention; almost 9% received comparison interventions of similar intensity and 23% were in a control group/TAU). Eldevik et al. reported that approximately 30% of the children who received intensive behavioral intervention achieved reliable change in IQ [compared to approximately 2.5% in the comparison (eclectic treatment) and 8.5% in the control groups], while 21% achieved reliable change in adaptive behavior (approximately 5.5% in comparison and 5% in the control groups). The majority of the sample was comprised of either “moderate” learners or those who showed little or no improvement. Another

way of examining outcome is to assess learning rate, e.g., the change in age-equivalents over time, which can then be displayed graphically as developmental trajectories. Klintwall and colleagues (2015) studied a subgroup of the sample receiving EIBI described by Eldevik et al. (2010). The sample was smaller (N = 244) due to inclusion criteria of intake age of 5 years and younger, as well as having both IQ and VABS data available. They analyzed intellectual ability and adaptive behavior using age-equivalents and changes in these scores over time. They noted considerable variability in individual learning rates (using a mean of IQ and adaptive behavior learning rates for each child). Higher intake intellectual ability, but not younger age, predicted higher individual learning rates. Significant gains in treatment outcome variables are often reported at the group level; however, considerable differences exist at the individual level, e.g., in the rate of change over time and in statistically reliable increases in scores between pre- and post-treatment.

Some of the most-researched predictors of (conventional) EIBI treatment outcomes include pre-treatment variables such as children's IQ, age, adaptive functioning, ASD symptom severity (Howlin, Magiati, & Charman, 2009; Virues-Ortega, Rodriguez, & Yu, 2013; Zachor & Ben-Itzhak, 2017), and language and communication abilities (Magiati, Moss, Charman, & Howlin, 2011; Virues-Ortega, Rodriguez, & Yu, 2013; Zachor & Ben-Itzhak, 2017). Intervention intensity (Howlin et al., 2009; Klintwall, Eldevik, & Eikeseth, 2015) has also been associated with treatment progress. In meta analyses, pre-treatment IQ has been identified as a strong predictor of outcome (Howlin et al., 2009), with some studies reporting IQ to have accounted for the majority of the variance observed in treatment outcomes (e.g., Perry et al., 2011). Not all reviews

document such a relation (e.g., Makrygianni & Reed, 2010; Virues-Ortega, 2010). With respect to Makrygianni and Reed's (2010) meta-analytic review of the effectiveness of EIBI, children's intellectual abilities at intake had little impact on the effectiveness of the intervention programs, as pre-treatment intellectual ability was not correlated with pre-post treatment effect sizes (however, pre- and post-treatment intellectual ability estimates were statistically related). In this review, pre-treatment adaptive behavior was significantly correlated with effect sizes but not language ability. Likewise, age at intake has been identified as a predictor in some reviews (Makrygianni & Reed, 2010) but not others (e.g., Eldevik et al., 2010; Klintwall, Eldevik, & Eikeseth, 2015; Reichow & Wolery, 2009). With respect to treatment dosage, T. Smith, Klorman, and Mruzek (2015) showed that increased number of treatment hours did not predict better outcomes. However, meta-analyses of EIBI (e.g., Eldevik et al., 2010; Virues-Ortega & Rodriguez, 2013) and other treatment studies (Fey, Yoder, Warren, & Bredin-Oja, 2013) have suggested that more hours are associated with larger gains. Inconsistencies in findings may be attributable to variation in such variables as fidelity of implementation of the intervention (and other aspects of quality such as therapist training, supervision, and frequency thereof), intervention frequency and duration, intervention delivery setting, intervention implementer (e.g., parent, therapist, researcher), as well as children's diverse profiles of symptoms and abilities. Indeed, family- and intervention-related variables have been examined as predictors of differential outcome in treatment for preschoolers with ASD. These include family characteristics such as parents' responsivity to their children, parents' attained level of education, and parental attitudes, as well as

intervention variables such as timing, intensity/dosage, techniques used, and fidelity of implementation (Stahmer, Schreibman, & Cunningham, 2011).

Beyond the aforementioned long-standing predictors of outcome in conventional EIBI, social behavior-related predictors of preschoolers' outcomes have been examined. Social engagement variables identified as plausible predictors of treatment outcome have been highlighted by Vivanti, Prior, Williams, and Dissanayake (2014). However, studies examining early social behavioral characteristics associated with EIBI outcomes are scant. Children's pre-treatment levels of imitation and social responsiveness (measured using the mean of parent- and teacher-report VABS Socialization subscale scores and the Social Incompetence factor from the Personality Inventory for Children) were found to predict outcome in one EIBI study (Sallows & Graupner, 2005). More recently, T. Smith and colleagues (2015) reported on children's social behaviors that predicted outcome to community-based EIBI across five sites (using a pre-post design, $n = 71$). They defined social engagement as social approach, joint attention, and motor imitation, which loaded onto a single factor in a principal component analysis. Social engagement predicted higher post-treatment IQ and adaptive behavior—accounting for 20% of the variance of IQ scores and adaptive behavior at Years 1 and 2 (after adjusting for age, IQ, baseline scores on predictors, treatment dosage, and site). T. Smith and colleagues' (2015) findings were important as they highlighted that specific behavioral pre-treatment variables are associated with EIBI outcome, in addition to IQ, age, adaptive functioning, and ASD symptom severity.

Naturalistic Developmental Behavioral Interventions (NDBIs). Variability within treatment outcomes and progress made during NDBI has been predominately

examined in the context of individual treatment studies, rather than in reviews and meta-analyses. Meta-analyses that have included NDBIs (e.g., Fuller & Kaiser, 2020; Tiede & Walton, 2019) have not reported on individual variability in NDBI outcomes. Specific treatment studies documenting longer term outcomes, i.e., two years post-intervention, tend to report gains at the group level without assessing variability at the individual level (e.g., ESDM follow-up study, Estes et al., 2015). Due to the paucity of large-scale studies examining treatment outcome and variability therein, I offer discussion of the variability reported as part of specific NDBI models. One such well-studied NDBI model is ESDM. Using a pre-post group design examining outcomes in children with ASD one year post-ESDM intervention, Contaldo, Colombi, Pierotti, Masoni, and Muratori (2020) reported significant differences, at the *group* level, between pre- and post-treatment scores on their curriculum checklist. They noted, however, important differences in skill acquisition at the *individual* level, in that the number of learning objectives acquired during treatment ranged across participants from 13.5 to 186.5. Results such as these motivate researchers to identify moderators of treatment as well as to delineate the behavioral differences between those children who make favorable versus minimal gains, and to establish profiles of lesser versus greater response to specific treatment models.

Parent-Mediated Communication-Focused Treatment (PACT; Green et al., 2010) constitutes another highly studied NDBI model. In a follow-up study of the original PACT RCT that included 121 participants from intervention and control groups at a median of 5.75 years post-intervention, Pickles and colleagues (2016) reported group differences in favor of the PACT intervention, notably in children's social initiations. In 2018, Hudry and colleagues from the PACT Consortium performed a secondary analysis

of variability in children's gains in social communication within the PACT trial (Green et al., 2010) in an effort to identify predictors of reliable symptom change over the one-year intervention (as well as to compare this group to TAU). Two subgroups of children were identified—those who showed reliable gains (Improvers) and those who did not (Non-Improvers). They found that 43 children (30% of the PACT sample) showed statistically reliable social-communication symptom improvement (using the social-communication algorithm score of the ADOS-Generic; Lord et al., 2000), whereas 41 children (28% of sample) were Non-Improvers. Sixty-two children (42% of the sample) showed intermediate, non-reliable change (and were omitted from further analysis). The investigators subsequently examined which baseline factors varied as a function of children's outcome status. Results showed that children's baseline nonverbal intellectual ability was a significant indicator of prognosis for those with ASD in both the PACT and TAU groups. Further, parent synchrony presented as a marginal predictor of differential outcome (in that a trend toward significance was observed). Parent synchrony referred to the proportion of parent communication behaviors that were supportive of the child's attentional focus and involved commenting on the child's play/activity (versus asynchronous behaviors, i.e., directive or placing demands on the child's attention/behavior). The investigators showed an association between lower parent synchrony and non-improver status for children in the TAU group (but not PACT). These findings provide a representative example of individual variability in response to treatment, despite significant group-level improvement through intervention. The findings also highlight the importance of parents' behavior during dyadic interactions as predictors of their children's progress.

Pivotal Response Treatment (PRT) is another NDBI that is considered to be an established effective model of intervention (Wong et al., 2014); however, variable individual outcomes have also been documented following PRT. In a community-based intervention using PRT, preschoolers with lower pre-treatment IQ (< 50) were shown to make somewhat smaller gains in expressive and receptive language age equivalents, compared to those with higher IQ (> 50; mean differences of 8.8 months and 11.1 months for expressive and receptive language, respectively; I. Smith et al., 2010). In a more recent pre-post group design study of this same PRT-based intervention program, I. Smith, Flanagan, Garon, & Bryson (2015) reported statistically reliable increases in IQ for 14% of the sample, and 16% for adaptive functioning, leaving a substantial proportion of children who did not make such significant improvements following intervention. When data from a subset of these children were analyzed (Fossum, Williams, Garon, Bryson, & Smith, 2018), diverse outcomes were noted with respect to expressive language (targeted outcome of the intervention), with gains varying from a mean of two standard score points for the lowest performing third of children to a 20-point mean gain for the highest performing third. Children's higher pre-treatment IQ and language level were associated with greater gains in expressive and receptive communication, cognitive and adaptive skills, as well as improvements in severity of ASD symptoms and challenging behavior.

Aside from established predictors of treatment progress, such as children's pre-treatment age, IQ, and cognitive ability, research has examined baseline prelinguistic, early social behavioral child characteristics for a variety of NDBI models. Children's pre-treatment initiating joint attention has been shown to predict differential treatment effects

on outcome levels of this variable as it related to the Picture Exchange Communication System (PECS; Bondy & Frost, 2001) versus another treatment, Responsive Education and Prelinguistic Milieu Teaching (RPMT; Yoder & Stone, 2006). Joint attention is typically achieved when one individual uses eye contact and gaze, pointing, or some other verbal or nonverbal means to alert the other's attention to an object of focus before returning their gaze to the individual (Mundy et al., 2007). Children's imitation of gestures and actions on objects has also been associated with treatment outcome (in the ESDM model; Vivanti et al., 2013). Relatedly, toy play skills/functional use of objects (Ingersoll, 2010; Kasari, Gulsrud, Freeman, Paparella, & Helleman, 2012; Kasari, Paparella, Freeman, & Jahromi, 2008; Vivanti et al., 2013), and interest in objects (Carter et al., 2011; Schreibman, Stahmer, Barlett, & Dufek, 2009; Yoder & Stone, 2006) have been shown to predict preschoolers' treatment progress within NDBIs.

Vivanti and colleagues (2014) have argued that it is important to continue to look beyond broad child characteristics such as baseline IQ, language ability, and adaptive functioning that have been found to predict better outcomes. Their argument was that these characteristics are not specific enough to serve as treatment targets amenable to change. Instead, the potential role of specific, modifiable predictors of social-communication skills such as imitation, toy play, engagement/social approach behaviors, reciprocity, and joint attention should be emphasized.

Researchers are also beginning to examine constellations of child characteristics that may predict progress in treatment. By shifting focus from the identification of individual predictors and, instead, examining groupings of child characteristics that predict progress, interventions can be tailored to children of varying profiles. In a SCED

study, Sherer and Schreibman (2005) examined a set of characteristics associated with response to PRT. Children who responded favorably to PRT had higher pre-treatment levels of appropriate toy contact and social approach, and lower levels of verbal and non-verbal self-stimulatory behavior and social avoidance, compared to children who showed minimal change in response to PRT. Fossum and colleagues (2018), using a group pre-post design, empirically derived a profile of predictors of PRT progress. Consistent with the broader literature, they found high levels of expressive language and cognitive abilities to be part of the responder profile. Building on the findings of Sherer and Schreibman (2005), they also found that higher responders exhibited increased levels of toy contact and low levels of social avoidance at baseline compared to other children. However, whereas Sherer and Schreibman's (2005) responders showed lower pre-treatment verbal self-stimulatory behavior, Fossum et al.'s (2018) high responders showed higher stereotyped/repetitive vocalizations before treatment. Fossum and colleagues also examined children's expressed positive affect; higher levels of positive affect constituted part of the responder profile—the first study to demonstrate the prominent role of positive affect in response to PRT.

Identification of 'Active Ingredients' of Treatment Models for ASD

Research to date has primarily focused on the investigation of predictors of treatment outcome, i.e., factors that have a main effect on outcomes. However, researchers are also acutely aware of the need for research to examine specific components of interventions that can be linked to greater social-communication gains (Fuller & Kaiser, 2020). These are baseline characteristics that influence the strength of intervention effects or predict differential response to interventions, i.e., moderators

(Kraemer & Gibbons, 2009), and the mechanisms responsible for those effects including processes that occur during treatment that contribute to change in target outcomes, i.e., mediators. Investigating moderators and mediators of treatment is important for developing more targeted treatments.

Recently, Fuller (2018; dissertation) investigated potential moderators of response to an adaptive communication intervention (n = 34) compared to TAU (n = 34). The intervention, J-EMT, was a blend of JASPER (Kasari, Freeman, & Paparella, 2006) and Enhanced Milieu Teaching (EMT; Kaiser, 1993) interventions aimed at teaching basic communication/expressive language. Therapists implemented J-EMT during play and routines with the child. The intervention also included parent coaching and DTT to teach foundational skills such as joint attention, receptive language, and imitation for those who showed skill deficits in these areas upon assessment. Additionally, use of a speech-generating device (SGD, using an iPad application) was incorporated to model and prompt language during the DTT and J-EMT sessions. Treatment as usual involved training parents to use the SGD for communication during daily routines, but not how to use it during play with their children. Fuller found that lower pre-treatment object interest (i.e., toy play behaviors) significantly moderated the effect of group assignment on initiating joint attention. Children in the intervention group who had lower pre-treatment object interest showed more post-intervention initiations of joint attention (but no main effect of object interest on initiating joint attention). Frequency of escape behaviors also significantly moderated the effects of intervention on initiating joint attention, such that children in the intervention group with higher pre-treatment frequency of escape behaviors showed more initiations of joint attention post-intervention. Again, no main

effect of escape behaviors was observed on initiating joint attention. Moderating variables have been also recently reported within other NDBIs. In an effort to investigate the large heterogeneity in outcomes within ESDM, Contaldo et al. (2020) examined pre-treatment variables that moderated outcome for 32 children with ASD after one year of community-based ESDM intervention. Pre-treatment characteristics were measured using scores on the parent-reported MacArthur-Bates Communicative Development Inventory (MB-CDI; Fenson et al., 2007): word comprehension (used to calculate receptive lexical quotient), word production, and production of action and gestures (first communicative gestures, actions with objects, and imitating other adult actions). Children's initial repertoire of first communicative gestures (both verbal/expressional and representational) moderated communication outcomes (a single score representing receptive and expressive domains on the ESDM Curriculum Checklist completed by interventionists) and socialization outcomes (a single score representing imitation, joint attention, and social skills items of the same checklist). Further, level of first communicative gestures, actions with objects, and lexical comprehension on the MB-CDI moderated gains in socialization and cognition/play (and was also associated with rate of learning). Studies such as these highlight the potential benefit of uncovering children's characteristics that may influence the strength of an intervention's effects.

Recent research also aims to identify specific developmental mechanisms that may underlie the predictive power of children's baseline behavior. To date, few researchers have attempted to isolate and empirically test potential mechanisms of treatment response by testing mediation within the context of an RCT. Studies measuring mediators of treatment outcome are scant for NDBIs, in large part due to the limited

numbers of RCTs. French and Kennedy (2018), in a systematic review of RCTs of early intervention for infants and young children diagnosed with or at risk of ASD, concluded that few published RCTs of behavioral, developmental, and multicomponent treatment approaches identify the active ingredients, i.e., mediators, in the diverse range of named treatment models. Complicating matters is the diversity of treatment approaches in the ASD intervention literature that are developed separately, distinctly named, but potentially incorporate similar treatment targets and intervention strategies. French and Kennedy (2018) identified 32 treatment models among the 48 RCTs reviewed, rendering it difficult to derive the successful (potentially common) active ingredients of intervention. For example, a plethora of terms appear in the literature that likely represent similar constructs related to affective social reciprocity; these include “social engagement”, “social interest”, “social reciprocity”, “responsivity”, “engagement in reciprocal interaction”, “social synchronous engagement”, “parent-infant synchrony”, “interpersonal synchrony”, “shared positive affect”, “affect sharing”, “positive emotionality”, and “socially engaged imitation”. Such variations in terminology make it difficult to delineate common mediators of treatment across NDBI models.

Gulsrud, Helleman, Shire, and Kasari (2016) tested the relation between core components of their parent-mediated social-communication intervention (JASPER; Kasari et al., 2006) for toddlers with ASD (mean age: 2.5 years) and the primary outcome, joint attention. Via mediation analyses on two strategies, they ascertained that “mirrored pacing” (and not “environmental arrangement”) was an active ingredient in JASPER. Mirrored pacing was measured by scoring conceptually linked parent behaviors: imitation of appropriate and functional play acts, and the timing and

positioning of these mirrored actions. Timing consisted of the contingency and rapidity of the imitative act; positioning included whether the adult displayed the act in the child's line of view. Similarly, in the large PACT RCT for preschoolers with ASD (Pickles et al., 2016; n = 59 in PACT group and n = 62 in TAU), parent synchronization to child activity was found to mediate children's short-term outcomes related to engagement and communication within the dyadic context. Green and Garg (2018) highlighted in their review that research investigating target mechanisms of intervention remains a pressing issue. Consistent with the studies reviewed here, they offered that dyadic factors such as joint engagement, joint attention, shared affect, parental behavior such as responsiveness and synchrony, and children's behavior such as communication initiation should be further studied as mechanisms indicating potential treatment targets within NDBIs.

Individualizing Children's Treatment to Enhance Outcomes

Once heterogeneity of children's treatment response is better understood by way of identifying moderators of treatment outcome and interventions' active ingredient(s), we can work towards boosting children's skills in these specific areas so that children are more capable of participating in the aspect of intervention most likely to lead to optimal outcomes. Personalizing intervention in this way will help to ensure that children maximally benefit from intervention. Investigators have begun to examine the impact of changing the intervention approach for children showing no or slow response to a particular treatment or method. Adaptive treatment designs are a research method well suited to address questions related to the personalization of interventions, for example the sequential multiple assignment-randomized trial (SMART) design. This systematic approach is possible within the context of large-scale intervention studies in which the

components of intervention have been well-studied, for instance JASPER (Kasari et al., 2006). Researchers using SMART designs aim to systematize evidence-based practices by constructing an optimal adaptive intervention so that children's treatment can be more personalized (and treatment uptake enhanced). These designs are used in personalized intervention research whereby the targeted outcomes attributed to each clinical decision rule can be empirically evaluated (Lei, Nahum-Shani, Lynch, Oslin, & Murphy, 2012) following random assignment of children to relevant conditions (Kasari, Sturm, & Shih, 2018). In this way, it is possible that clinicians can then modify prospective children's treatment paths based on children's initial responsivity to the program.

Other empirical approaches to augmenting response to treatment involve examining the effect of a modular treatment component accompanying an intervention program. For example, Landa, Holman, O'Neill, and Stuart (2011) investigated the effects of an "Interpersonal Synchrony" module to a six-month intervention comprising DTT (Lovaas, 1987), PRT (Koegel et al., 1989), augmentative communication systems (as needed), visual cues (Carr, Binkoff, Kologinsky, & Eddy, 1978), and visually-based organizational strategies (Lord, Bristol, & Schopler, 1993) provided to 50 toddlers with ASD (21 to 33 months). Outcomes between children receiving this intervention alone and those receiving the intervention with the Interpersonal Synchrony module were compared. The module, which targeted socially engaged imitation, joint attention, and affect sharing, showed a significant treatment effect for socially engaged imitation (imitative acts paired with eye contact). This study provided preliminary evidence for the efficacy of an intervention module overlaid on an existing group-based comprehensive intervention.

Research has also explored matching treatment approaches to individual children's treatment needs. For example, Schreibman and colleagues (2009) showed that their behavioral profile of responders to PRT was *specifically* predictive of response to PRT and not DTT. To better understand distinct responses to PRT and DTT, Jobin (2020) conducted a follow-up study comparing the relative efficacy of these treatment methods for teaching language, play, and imitation to children under 3 years of age and at risk for ASD. Using a SCED (alternating treatments design) for 12 weeks each, Jobin demonstrated that PRT and DTT was each more effective for some children and some learning domains. Individual children responded uniquely to PRT and DTT and one child responded to each treatment based on the skill examined. For example, one child showed a superiority of PRT for *expressive* language while another benefitted more from PRT for *receptive* language. Two other children who made more gains in DTT (for imitation, receptive language, and play) nonetheless showed superiority of PRT for expressive language. Jobin (2020) concluded that a hybrid approach (implementing both interventions) may be optimal for some children. Unfortunately, the factors that predisposed a child to benefit from one intervention over another, and for which learning domains, could not be determined in this small study.

Additional studies have examined whether specific behavioral profiles may be useful in identifying which children will likely respond to different treatments. Schreibman and Stahmer (2014) performed a randomized comparison study to investigate differential effects of PRT (a language-based approach) and PECS (Bondy & Frost, 2001) on preschoolers' communication, social, and cognitive outcomes. For every two children assessed and matched on variables of interest, one child was randomly assigned

to PRT or PECS and the other to TAU. Children were matched on three factors: word use (no words or 1 to 9 functional words), age (18-32 months or 33-47 months), and cognitive functioning (low: age-adjusted visual reception score of ≤ 50 on the Mullen Scales of Early Learning [Mullen, 1995], or high: adjusted score of ≥ 50). Results showed that nonverbal and minimally verbal children were equally likely to develop verbal communication skills in either treatment program (and early word use was highly predictive of verbal gains in either PRT or PECS). In another study comparing differential effects of communication interventions linked to child characteristics (discussed earlier in a different context), Yoder and Stone (2006) compared children's response to PECS versus a vocally based naturalistic intervention: responsive education and prelinguistic milieu teaching (RPMT). Children who initially had low levels of object exploration benefitted more (in development of expressive language) from RPMT, and those with higher levels of object exploration benefitted more from PECS. It was highlighted that although both treatments rely on access to objects as rewards (for communication), RPMT also teaches play with objects, which appeared to be important for children with lower baseline object exploration skills.

In summary, various NDBIs have shown beneficial results with respect to gains made during treatment and maintained at short-term follow-up. Children's individual outcomes, however, remain variable. Given the heterogeneity in children's abilities, symptoms, and severity of impairment, there is a need to identify factors that could explain the observed variability in treatment response with respect to particular interventions. Identification of key variables for targeted study, including children's profiles predictive of treatment progress, constitutes an important, continued focus of

research efforts.

Selecting Intervention Targets

Based on the previous review, toy play and positive affect may be two child variables that are especially promising modifiable predictors of progress in PRT. I will provide an account of typical and atypical development in these areas, and summarize research establishing them to be key modifiable predictors that could be targeted to enhance expressive language response to PRT.

Toy play. Children's functional toy play has been revealed as a predictor of progress in PRT (Fossum et al., 2018; Sherer & Schreibman, 2005). In typically developing infants/toddlers, four hierarchical levels of play are considered to emerge between the ages of 2 and 24 months: sensorimotor-exploratory (e.g., holding and looking at objects, hitting objects; emerges at 2–4 months), relational - non-functional (e.g., stacking, nesting, pushing objects; emerges at 6–10 months), functional-conventional (e.g., putting a cup on a saucer, drinking from a cup, pushing toy car, scribbling with a crayon; emerges at 10–12 months), and symbolic (e.g., pretend play; emerges at 12–30 months; Casby, 1992; 2003). Based on a typical developmental sequence of play with objects (Lifter, Foster-Sanda, Arzamarski, Briesch, & McClure, 2011), the most basic level of play involves performing indiscriminate actions on objects, i.e., treating all toys in the same manner: spinning, licking, banging, or throwing them. Next-level play involves performing discriminate actions and thereby showing an understanding of different physical properties of toys, e.g., squeezing a plush toy. Children will then typically learn to take apart components of toys and put them back together, build them to produce new configurations, and use toys based on physical

construction (e.g., stacking cups). These actions develop before the propensity to engage in pretend play with toys, ultimately leading to the enactment of multi-scheme sequences and engagement in socio-dramatic play.

Children with ASD differ in the quantity and quality of play compared to both typically developing children (Charman & Baron-Cohen, 1997) and children with other developmental disabilities (Rutherford, Young, Hepburn, & Rogers, 2007). Play research with young children who have ASD has often focused on exploratory, relational, and functional play skills, which are often present by 12 months of age in typically developing children. Research shows that young children with ASD show reduced levels of object exploration (Pierce & Courchesne, 2001) as well as fewer novel play acts (Charman & Baron-Cohen, 1997). Children with ASD also demonstrate fewer play behaviors, less frequent spontaneous play, and less varied play (reduced diversity in toys and play actions) than children with typical development (Barton, Gossett, Waters, Murrari, & Francis, 2019; Kasari, Chang, & Patterson, 2013; Lifter, Mason, & Barton, 2011). The play intervention literature with children with ASD is peppered with studies by researchers whose aims consisted of increasing children's frequency and duration of functional play behaviors (Jung & Sainato, 2013), symbolic and pretend play, and use of play scripts and actions associated with play scenarios such as cooking (see Kossyvakis & Papoudi, 2016, for a review). Increasing the *diversity* of play behaviors remains a specific area worthy of continued study given the idiosyncratic and often repetitive ways in which some children with ASD use objects (Barton et al., 2019).

Play interventions can be classified as behavioral: focused on teaching new skills in a structured environment where systematic reinforcement follows a correct response to

a clear, antecedent stimulus, and developmental/relationship-based: focused on learning through strong affect-laden relationships between child and adult. This latter type of play intervention assumes that children with ASD follow the developmental trajectories of their typically developing peers and thus teaches skills underlying missed milestones. Some play interventions combine these two approaches, e.g., combining imitation and following a child's lead with behavioral strategies such as prompting and modelling (Ingersoll & Dvortcsak, 2006). A relatively recent review that included interventions to improve play performance in children with ASD reported improvement in children's play skills following adult modelling and prompting but concluded that evidence was limited (Tanner, Hand, O'Toole, & Lane, 2015).

Children's skill in toy play has implications for the successful implementation of many strategies used within NDBIs. Communication exchanges are often made within the context of play, and toy play predicts rate of development of communication skills, from age 4 to 6.5 years, in young children with ASD (Toth, Munson, Meltzoff, & Dawson, 2006). Research also shows that treatment focusing on play skills of pre-school-aged children with ASD is important for long-term functional, spoken language outcomes (at 5-year follow-up; Kasari et al., 2012). Language development occurs through a transactional process involving non-linguistic aspects of development, such as imitation (Tomasello & Farrar, 1986). Intervention targeting imitation and play enhances effects on communication development in children with ASD (play: Kasari et al., 2006; 2012; Stahmer, 1995; imitation: Charman et al., 2003; Ingersoll & Schreibman, 2006). Sherer and Schreibman (2005) and Fossum et al. (2018) found toy contact to be one of the variables that predicted progress made in PRT (i.e., expressive language outcome).

Moreover, increasing toy contact led to improved PRT outcomes in a SCED study (Schreibman et al., 2009). Targeting toy contact is theoretically important because toy play is often the vehicle that sparks interaction between a child and therapist (or parent). It is within the context of these dyadic interactions, when a child is socially motivated because of a desire for access to an object/toy, that initiating/maintaining a social interaction involving the toy (perhaps by speaking) may occur. Thus, functional toy play (toy contact) is a skill that warrants attention in low responders to PRT and possibly other NDBI models.

In sum, quality and quantity of toy play of young children with ASD during the preschool period is related to later language skills (Lewis, 2003, 2000; Toth et al., 2006) as well as later social peer play (Sigman & Ruskin, 1999). Given that play-based interactions comprise a primary context for the acquisition of new skills (e.g., as in PRT to learn spoken language), shifting children's play profiles so that children become more interested in toy play and in joint play interactions is important. Play also comprises a critical developmental context for social engagement with same-aged peers; targeting preschoolers' toy play skills can contribute to the child's future success in learning and socialization.

Positive affect. A second promising modifiable predictor of progress in PRT is children's positive affect (i.e., happiness or enjoyment) when interacting with others. Positive affect emerged as part of Fossum et al.'s (2018) behavioral profile predictive of progress in PRT. In typical development, early attentional preference for social stimuli (Reynolds & Roth, 2018) promotes the development of social communication. Social communication is fostered by mutual gaze and shared positive affect between a child and

caregiver (Parladé & Iverson, 2011) as well as social engagement and parental scaffolding (Hustedt & Raver, 2002). These dyadic behaviors emerge within the parent-child relationship within the first year of life. Parents' and infants' early dyadic interactions involve nonverbal responses to each other's emotional expressions (McDonald & Messinger, 2012; McQuaid, Bibok, & Carpendale, 2009). During the first six months of typical development, an infant and their parent become increasingly responsive to one another's expressions of positive affect, which usually take the form of smiles (Parladé et al., 2009). Mutual and responsive dyadic social interactions provide the foundation for the healthy development of social and emotional reciprocity (e.g., Kochanska, 2002; Leclère et al., 2014). Typically, 9- to 10-month-old infants understand that others' pointing gestures and direction of gaze is pertinent social information to which to pay attention (Crais, Douglas, & Campbell, 2004); consequently, infants learn joint attention: shifting their attention from an interaction partner to an object of reference and back. Inherent in this process is the discovery that words refer to objects, thereby facilitating the development of language (Baldwin & Moses, 2001). Early joint attention abilities have been shown to be predictive of later language functioning in the typically developing population (Tomasello & Todd, 1983) as well as in individuals with ASD (Charman et al., 2003; Dawson et al., 2004).

Positive affect is itself linked to language in typical development. Laake and Bridgett (2014) showed that positive affect measured at 10 months of age predicted expressive language at 14 months in typically developing children. More recently, Laake and Bridgett (2018) further explored the link between positive affect and later language outcomes. They demonstrated that maternal support (characterized by parent

responsiveness techniques) when typically developing infants were 10 months old was positively linked to infants' expressive language development at 14 months specifically for those infants rated higher in positive affect.

Early social-communication skills and behaviors are impaired in children with ASD, and delays are apparent towards the end of the first year (Bryson et al., 2007; Colgan et al., 2006; Osterling, Dawson, & Munson, 2002). An early sign of delayed social development in young children with ASD is reduced expression of positive affect (e.g., smiling or laughter; Filliter et al., 2015). Affective responding serves to draw the attention of adults and peers and, thus, is vital for increasing social interaction opportunities. Children with ASD, however, tend to show little interest in engaging in social reciprocity with others (see Dawson et al., 2004). Young children with ASD often show disruptions in positive emotional responsiveness, leading to atypical social-emotional outcomes (see Lambert-Brown et al., 2015).

Naturally, children and their parents both influence the parent-child relationship and are, in turn, influenced by the relationship. The ways in which parents interact with their children who have ASD could influence the children's ongoing social experiences and, subsequently, their developmental outcome. In a bidirectional manner, early social-communicative differences in young children with ASD affect their ability to engage in social interactions with their parents (i.e., lower responsiveness to parents' smiles and social approaches, less integration of smiles with eye contact towards parent), which may then affect a parent's social behavior within the parent-child relationship (Dawson, 2008). Complicating matters, parents of children with ASD are more likely to manifest social-communication difficulties (e.g., as part of the 'broader autism phenotype'; Flippin

& Watson, 2018) than are parents of typically developing children. They are also at increased risk for experiencing high levels of stress and low levels of self-efficacy (Estes et al., 2009; Meirsschaut, Roeyers, & Warreyn, 2010). These factors necessarily also have the potential to alter the parent-child relationship. Evidence suggests differences between parent-child interactions in families with children with ASD as compared to interactions in families without ASD (Freeman & Kasari, 2013; Yirmiya et al., 2006). Therefore, providing intervention to enhance the quality of the parent-child relationship in the context of families with children who have ASD is important.

Early mother-infant interactions are thought to be especially salient for children who are at risk for social difficulties (Schwichtenberg, Kellerman, Young, Miller, & Ozonoff, 2019). Fathers (and, thus, father-child interactions) are under-represented in research on young children with ASD; research highlights the role that maternal gaze, vocalizations, and responsiveness can play in the social development of preschoolers with ASD (Bang & Nadig, 2015; Bottema-Beutel et al., 2014; Flippin & Watson, 2015; McKean et al., 2017; Northrup & Iverson, 2015; Stern, Maltman, & Roberts, 2017). Wan, Green, and Scott (2019) conducted a systematic review with the goals to a) identify a developmental timeline for parent-infant interactions within the child's first 2 years of life, in children at familial risk for, and in emerging, ASD, and b) examine parent-infant interaction associations with later social-communicative outcomes. Their findings showed that the interactions of children with eventual ASD and their primary caregivers deviated from a typical trajectory in the latter months of the children's first year. Specific dyadic variables predicted ASD outcome later in the preschool period. Most notably, high-risk siblings and the eventual-ASD group of children showed differences in the

dyadic qualities of their parent-child interactions from those without eventual ASD. Children in the high-risk/eventual-ASD group were a) less socially reciprocal at 11-12 months (but no group differences in degree of positive affect in these studies), b) demonstrated fewer “show-and-give” gestures at 11 months, and c) were less attentive to their parents at 14 months (Wan et al., 2019). Following their systematic review, Wan et al. (2019) highlighted the role of parent-infant interaction in early parent-mediated intervention.

Since interactions of primary caregivers and their children with eventual ASD have been shown to deviate from a typical trajectory as early as during infancy (Wan et al., 2019), coaching parents to implement intervention strategies targeting social, dyadic aspects reflective of typical reciprocal parent-child interactions is important. The quality of parent-child interactions has been targeted, explicitly or inadvertently, in parent-mediated NDBI models (e.g., explicitly in ESDM; Dawson et al., 2010). Schwichtenberg and colleagues (2019) examined indices of maternal behavior within parent-mediated intervention designed to mitigate risk for ASD and promote children’s prosocial behaviors. Mothers’ dyadic play-based interactions with their high-risk infants (siblings of children with ASD) were coded when infants were 6, 9, and 12 months of age. Maternal social responsiveness was associated with increases in infants’ positive affect and responsiveness. Schwichtenberg and colleagues (2019) concluded that maternal social responsiveness behaviors could, therefore, be highly important to children’s outcomes. Indeed, research supports that increased attention sharing and parental responsiveness within the parent-child relationship of children with ASD is positively related to children’s social, behavioral, and cognitive outcomes (Clifford & Dissanayake,

2009; Haebig, McDuffie, & Weismer, 2013; Ruble, McDuffie, King, & Lorenz, 2008). Parents' difficulties with social responsiveness and communication have been documented within families raising children with ASD (Parr, Gray, Wigham, McConachie, & Couteur, 2015; Virkud, Todd, Abbacchi, Zhang, & Constantino, 2008). It has been suggested that 'enrichment' in parent-child interactions may open opportunities for early intervention (Bradshaw, Steiner, Gengoux, & Koegel, 2015). Research demonstrates that boosting the quality of parent-child interaction (higher levels of joining the child, enjoying the child, parental sensitivity and reciprocity, responsiveness, and synchrony) leads to positive social-communicative and language outcomes in children with ASD (Casenhiser, Shanker, & Stieben, 2013; Green et al., 2010; Mahoney & Perales, 2003; Siller & Sigman, 2002). The importance of targeting parents' social responsiveness (of which sharing positive affect is a part) to their children has been acknowledged as part of some NDBIs, e.g., ESDM (Dawson et al., 2010) and Social ABCs (Brian et al., 2016). Boosting shared positive affect may enhance a child's potential for a stronger learning experience in PRT-based intervention in which the target is expressive language.

Chapter 3: Rationale for the current study

NS EIBI and Present Context

The Nova Scotia (NS) EIBI program is a one-year publicly funded community-based program in which all preschoolers with ASD in the province are eligible to participate prior to school entry. The NS EIBI model was designed with PRT (Koegel & Koegel, 2006; Koegel, Koegel, Harrower, & Carter, 1999) as the foundation of treatment (Bryson et al., 2007). As noted, PRT is classified as an NDBI, as the model highlights social motivation and includes techniques that are embedded within motivating, naturally occurring situations. The NS EIBI program's explicit targeted outcome is expressive language, with additional goals aimed at improving children's communication, play, and other functional skills (Bryson et al., 2007). The program involves a parent-coaching component in PRT strategies (see Coolican, Smith, & Bryson, 2010). The overall effectiveness of the NS EIBI program has been documented in pre-post group design studies over 15 years (I. Smith, Flanagan, Garon, & Bryson, 2015; I. Smith et al., 2019). Although group-level data from NS EIBI have shown significant developmental gains for children (moderated by IQ), variability in *individual* progress during treatment has been observed. Such variability served as the springboard for my dissertation.

As noted earlier, our team previously established a behavioral profile that predicted levels of progress within NS EIBI in order to better understand the observed heterogeneity in expressive language outcome (Fossum et al., 2018), building on work by Sherer and Schriebman (2005). As described earlier, two variables, toy play and affect, were especially promising as key modifiable treatment targets that could enhance treatment progress.

Development of the Intervention

The focus of my dissertation was to design a booster intervention with the goal of amplifying responsiveness to PRT by enhancing pre-treatment levels of toy play and shared positive affect in children who were of lower cognitive ability and lower language level. This booster is the Pivotal Response Intervention Minimal Responders (PRIMEr) intervention. The goal of PRIMEr was to prepare children to benefit from PRT-based intervention such as NS EIBI by boosting preverbal skills that are related to poorer progress during PRT-based intervention (Fossum et al., 2018) and that are purported to form part of the foundation upon which expressive language develops. The PRIMEr intervention has a theoretical basis for shifting children's behavioral profiles so that low responders may benefit from NS EIBI to a greater degree. The success of PRT depends on use of strategies that include following the child's lead (i.e., capitalizing on their interests/play ideas), gaining control of whatever object or social routine motivates them, and inserting a language opportunity therein (i.e., making access to the desired object or routine contingent upon the child's vocalization, the explicit targeted outcome; Koegel, Koegel, Harrower, & Carter, 1999; Koegel, Koegel, Shoshan, & McNERney, 1999). The interventionist relies on cues from the child to guide when language opportunities are inserted, typically within the context of toy play and enjoyable social routines. Finding opportunities to motivate children to speak is more challenging if the child is a) not interested in, or not making contact with, toys and b) not interested in social interactions characterized by positive emotion and mutual enjoyment. I therefore selected intervention methods for use in PRIMEr that were intended to facilitate growth in children's level of toy play and propensity to share positive affect with another person.

Reciprocal imitation training (RIT) for toy play. A key component of the PRIMeR intervention was Reciprocal Imitation Training (RIT; Ingersoll, 2010), included to enhance toy play. Reciprocal Imitation Training was developed to teach young children with ASD to imitate an adult partner during play (Ingersoll, 2012). It uses a blend of naturalistic behavioral and developmental strategies to teach imitation within ongoing play interactions with an adult. It incorporates strategies that have been shown to increase reciprocity [e.g., following the child's lead, contingent imitation (i.e., imitating the child's play with objects), modelling actions for the child to imitate, and linguistic mapping (narrating actions using simple language)] as well as naturalistic behavioral strategies to promote acquisition and maintenance of skills, e.g., explicit prompting and direct response-reinforcer relationship. Object imitation teaching follows the typical developmental progression, beginning with recognition of being imitated by another, followed by the imitation of familiar actions, and ending with the imitation of novel actions.

Imitation is an important early developmental skill; children learn about the world and communicate social interest via imitation. Imitation is a vital avenue through which toy play learning and language occur (Dawson & Galpert, 1990; Ingersoll & Schreibman, 2006; Toth et al., 2006). Poor imitation is common among individuals with ASD (Rogers & Williams, 2006; I. Smith & Bryson, 1998; Williams, Whiten, & Singh, 2004) and is observed in children as young as 20 months of age (Charman & Baron-Cohen, 1997). Impaired early ability to imitate others is related to language functioning later in the preschool years (Stone & Yoder, 2001; Toth et al., 2006). Reciprocal imitation training has been shown to lead to gains in language (Ingersoll & Lalonde, 2010; Ingersoll &

Schreibman, 2006). Moreover, adults' use of imitation with a child has been identified as an active ingredient in JASPER (i.e., mirrored pacing; Gulsrud et al., 2016). Labelled "contingent imitation" in RIT, an adult's imitation of a child's behavior serves to acknowledge the child's action and confirm that the adult's attention is on the child and what they are doing (i.e., joint attention and joint engagement). The contingent imitative act by the adult should then summon the child to respond and to also pay attention to the therapist and their actions. Through this process, children may be encouraged to learn functional actions on objects (i.e., toy play). Ingersoll (2010) reported significantly greater gains in elicited and spontaneous imitation in a group of pre-verbal preschoolers with ASD who received RIT.

In published studies, RIT has been delivered to 2- to 4-year-old children with ASD for an hour per session, one to two days per week for 10–12 weeks (e.g., Ingersoll, 2010). Teaching children to imitate object play should theoretically motivate them to contact toys and other objects spontaneously during daily activities/routines. Therefore, RIT was selected to boost toy play.

Responsiveness training for shared positive affect. The other key component of the PRIMeR intervention was responsiveness training. The goal of responsiveness training was to enhance shared positive affect between parent and child. Generally, responsive parenting emphasizes an affective–emotional style with positive affection and high levels of warmth and nurturance, parental responses that are contingently linked to children's signals, and acceptance of children as unique individuals. Specific responsiveness techniques include: a) display of positive affect (e.g., smiling, laughing, and facial animation); b) positive emotion sharing; c) warm sensitivity (acceptance of

child's interests and needs, physical affection, enthusiasm in activities, positive tone of voice); d) verbal encouragement (praising efforts, encouraging activity involving reciprocity in emotion sharing); e) prompt and sensitive responses to children's signals (including strategic use of tone of voice, pacing, positive affect, interacting without being physically intrusive); and f) verbalizations that engage the child's attention. Infants (13 months of age, born at term and born at very low birth weight) have been shown to increase their displayed positive affect in response to parent-mediated intervention using parent responsiveness training strategies (Landry, Smith, & Swank, 2006). Moreover, parents' use of such "responsive parenting" techniques have been shown to contribute to typically developing preschoolers' foundation for the development of social and communication skills between 6 and 13 months of age (Landry et al., 2006).

Parent responsiveness principles and techniques have been incorporated into play routines commonly represented within empirically supported NDBIs. For example, Social ABCs, a caregiver-mediated, PRT-based intervention for toddlers showing signs of ASD, uses parent responsiveness training procedures. In an RCT, Brian et al. (2017) showed significant increases in shared smiling (i.e., shared positive affect) and social orienting between parents and 12- to 30-month-old toddlers with early signs of ASD, following 12 weeks of caregiver-mediated intervention using these principles. The Early Start Denver Model (ESDM; Dawson et al., 2010) is another NDBI (for children with ASD younger than 3 years) that uses parent responsiveness techniques as a component of intervention. Responsive parenting techniques used in these NDBIs include an emphasis on observing and responding to child cues, the nondirective use of language and play models / expansions, contingent imitation of spontaneous child behaviors, and following

the child's lead in their selection of activity. By following the child's lead, one is able to learn what the child likes, enjoys, or is interested in—and, in this way, may build a repertoire of activities to enhance the child's motivation to socially respond to the adult, e.g., with a smile that serves to share/communicate emotion. Use of sensory social routines (e.g., Dawson et al., 2010) allows the focus of the interaction to be on the affective and relationship-based aspects of the dyadic social experience between the adult and child and facilitates use of parent responsiveness strategies. Such routines are believed to increase the salience of social rewards for children while enhancing their social attention and motivation for interaction (Dawson et al., 2010).

Research shows that parents' use of responsiveness during social interactions with their children with ASD (in the context of parent-implemented interventions) is linked to increased social engagement by the child (Patterson, Elder, Gulsrud, & Kasari, 2014), social initiations (Ruble et al., 2008), and language acquisition (McDuffie & Yoder, 2010; Siller & Sigman, 2008). Therefore, boosting parent responsiveness and social-affective engagement within socially rewarding activities for the child should facilitate increases in children's shared positive affect. Techniques associated with parent responsiveness (Landry et al., 2006), as well as sensory social routines, were therefore selected to facilitate children's (and their parents') growth in sharing of positive affect. The combination of responsiveness techniques and sensory social routines was designed to engage the child in positive emotional experiences with another person, to draw the child's attention to social stimuli, to make such stimuli rewarding for the child, and to foster the child's motivation to continue such activities. In developing the intervention, a goal was that social engagement would become intrinsically rewarding for the child over

time. Enhancing children's level of sharing of positive affect should lead to increases in their motivation/ability to benefit from PRT (Vivanti et al., 2013), thereby optimizing treatment progress in NS EIBI.

The main strategy to increase children's shared positive affect was to first increase the parent's sharing of positive affect with the child, i.e., directing positive emotion such as a smile toward their child, using eye contact. Therefore, the PRIMeR intervention involved parent coaching in responsiveness strategies and use of sensory social routines.

Parent coaching. The PRIMeR module incorporated parent coaching. Use of a family-centered approach has been associated with positive outcomes for children and families (Dunst, Bruder, & Espe-Sherwindt, 2014). The National Research Council (NRC, 2001) has long considered parental involvement in early intervention to be essential to effective treatment and optimal outcome for children with ASD. Empirical support has been documented for parent coaching programs (social-communication based, or naturalistic developmental behavioral) for parents of young children with ASD (see Nevill, Lecavalier, & Stratis, 2018, for meta-analysis). As previously described, early research involved *training* parents as therapists/co-therapists in conventional EIBI; however, within the context of NDBIs, the framework has shifted to an emphasis on *coaching* the parent to mediate aspects of the intervention with their children. It is ideal and feasible for interventionists to support parents in learning to use research-based intervention strategies (Coogle, Floyd, Hanline, & Hiczewski, 2013; Coolican et al., 2010). Engaging families in this way serves to strengthen the parent-child relationship, increase parents' responsivity to their child, and empower parents to help their children (Koegel, Bimbela, & Schreibman, 1996; Minjarez, Mercier, Williams, & Hardan, 2012).

Coaching parents also results in parents' increased self-efficacy and perceived self-competence (Brian et al., 2016), reduction in parent stress (Baker-Ericzén, Brookman-Frazee, & Stahmer, 2005), increased sense of family cohesion and family quality of life (Dawson & Burner, 2011; Rogers et al., 2012), as well as increased treatment intensity, maintenance, and generalization of learned skills (Bryson et al., 2007; Kaiser, Hancock, & Nietfeld, 2000). Research shows that parents successfully learn to implement strategies related to child-responsive engagement when parent coaching is utilized in intervention programs. Further evidence supports the combination of parent-mediated and therapist-delivered intervention to maximize developmental gains (Landa, 2018). For these reasons, parent coaching was an important element of the PRIMeR intervention package, distinct from programs that are exclusively therapist-implemented. This approach is also consistent with NS EIBI practices (Bryson et al., 2007; I. Smith et al., 2019).

Altogether, the PRIMeR treatment package incorporated elements that separately meet criteria for an established level of evidence (behavioral interventions, natural teaching strategies, modelling, and parent training; National Autism Center, 2015).

Pilot work. An abbreviated version of the PRIMeR intervention was implemented with one 4-year-old child who had been recently diagnosed with ASD. The child was flagged by a clinical psychologist as potentially fitting the 'low responder' profile (i.e., low cognitive ability, low level of expressive language, low frequency of functional toy play, and low positive affect). With parental consent, and following screening and eligibility assessments, the child received six weeks of intervention at daycare, including one week of home-based parent coaching in the responsiveness strategies utilized by the researcher-therapist. This pilot intervention was intended to inform feasibility of

objectives and to identify the need for modifications to the study protocol prior to beginning the 4-month, multi-phase study intervention.

The Current Study - Purpose and Specific Objectives

My specific aim was to develop an intervention that targets toy play and positive affect for preschoolers with ASD and the PRT low responder profile, and to evaluate the intervention's short-term efficacy in improving the behavior represented by these targets. My research question centered on whether I could shift preschoolers' behavioral response profile by increasing their levels of toy play and shared positive affect. I sought to a) bolster the development of children's toy play by teaching object imitation using RIT, and b) foster shared positive affect, using responsiveness strategies and parent coaching.

Specific objectives. I sought to accomplish this aim by addressing the following specific objectives, using a multiple baseline across participants SCED (Gast, 2009) whereby four children participated, semi-concurrently, in this 12-week intervention.

Specific objectives were 1) to determine the efficacy of a brief therapist- and parent-mediated packaged intervention, PRIMeR (using RIT and parent responsiveness training) for increasing level of toy play and shared positive affect in children diagnosed with ASD who exhibited the PRT low responder profile; and 2) to ascertain whether this brief intervention could enhance family well-being/quality of life as well as parents' perceived self-efficacy.

I hypothesized that (1) for children diagnosed with ASD who showed a low responder profile (low language, low cognitive ability, low frequency of functional toy play, low level of positive affect), the PRIMeR intervention would increase children's levels of toy play and shared positive affect, and (2) the intervention would increase

parent-reported self-efficacy and family quality of life.

Importance of the Current Study

Ensuring optimal benefit for all children is important. Intervention programs based on PRT offer children an important opportunity to develop key functional skills. Findings may inform the design of specific treatment pathways within PRT-based programs for children who exhibit particular PRT responder profiles, perhaps most importantly children with the low responder profile—as they are the most vulnerable. This approach to treatment modification for low responders to PRT has some empirical support (Koegel, Koegel, Shoshan, et al., 1999; Koegel, Vernon, & Koegel, 2009).

Chapter 4: Methods

All procedures were in accordance with the ethical standards of the IWK Health Centre and were approved by the IWK Research Ethics Board.

Participants

Recruitment and inclusion/exclusion criteria. Four preschoolers with ASD participated, aged between 3.5 and 4.5 years. Two of the children were diagnosed with ASD by a multi-disciplinary team with expertise in ASD at the local children's hospital. Diagnostic assessment included the Autism Diagnostic Observation Schedule, 2nd edition (ADOS-2; Lord et al., 2012) and the Autism Diagnostic Interview-Revised (ADI-R; Rutter et al., 2003). One child was diagnosed privately by a clinical psychologist using the same diagnostic measures. At the time of the study, the fourth child was on the wait-list for a diagnostic assessment by the children's hospital autism team but was given a diagnosis by a co-investigator (student's supervisor), a clinical psychologist with expertise in diagnosis of ASD.

Inclusion criteria for the study required that children have no or minimal spoken language. Our definition of 'minimally verbal', consistent with Goods, Ishijima, Chang, and Kasari (2013), was based on a combination of observational and standardized assessments. Spontaneous functional use of 10 or fewer words was reported by parents and confirmed with an expressive language score of 15 months or less on the Preschool Language Scale, 5th edition (PLS-5; Zimmerman, Steiner, & Pond, 2011), and fewer than 10 words expressed in a 10-minute period in which the investigator 'pulled' for communication during a standardized play protocol, the *Play, Engagement, and Affect Ratings (PEAR) protocol* (Flanagan, 2015; see description below). Another inclusion

criterion for participation in the study was low cognitive ability, initially indexed by an intellectual ability assessment if completed as part of the child's diagnostic assessment. This criterion was confirmed during our eligibility screening using a standardized measure, the Merrill-Palmer-Revised Scales of Development (M-P-R; Roid & Sampers, 2004); a child was eligible for participation if he/she obtained a ratio score (Developmental Index age equivalent estimate/chronological age x 100) below 55—within two standard deviations of the mean for cognitive ratio scores of low responders (Fossum et al., 2018). The next criterion was low frequency of functional toy play, measured during the PEAR protocol, which involves engaging the child in toy play using a specific set of pairs of toys for 10 minutes. The administrator joins in play with the item paired to the child's choice, and models 10 novel play actions, adapted to the child's skill level (See Appendix A). The requirement was that the proportion of total intervals (30-second intervals in the 10-minute period) in which functional toy play occurred was less than 90%, informed by the low responder profile. Inclusion criteria also required that children exhibit a low level of positive affect, also measured during the play protocol. Average scale ratings (on a 5-point scale) for the same 10-minute period in which toy play was measured needed to be 3 or less (rated at 1-minute intervals; see Appendix B for child affect coding scheme). Other criteria for enrollment in the study required that participants lived (and attended daycare, if intervention was to be provided there) within 35 kilometers of the local children's hospital. Children could not be expected to begin the provincial EIBI program within the four months following commencement of participation in the intervention study. Additionally, parents needed to possess the capacity to provide informed consent for her/his and the child's participation. Parents

also needed to be comfortable with me entering their homes to video-record behavioral samples of their children every couple of days during the baseline phase (which lasted between one and four weeks), and behavioral samples of parent-child interactions during natural routines once every 3–4 weeks (for 10 minutes).

Exclusion criteria for children’s participation included receiving another form of treatment based on ABA, including private behavioral therapy (but with the exception of a communication-based program for caregivers of children under 6 years of age who experience social communication difficulties, delivered by speech-language pathologists as part of public speech-language therapy service). Many families are referred to this program by their family physicians at the first sign of expressive language delay, i.e., before or when ASD is flagged as a possible diagnosis. Other exclusion criteria included having a major sensory, motor, or neurological impairment/disorder (e.g., uncorrected visual or hearing loss, or physical impairment limiting mobility and/or hand use), and use of an augmentative alternative communication (AAC) device or program such as PECS (Frost & Bondy, 2002) – due to the potential for harm of the researcher supporting social interaction without reinforcing AAC use. Parents could make an informed choice to pause use of the AAC for the three months of the study. The first four children to meet all criteria, and whose parents provided informed consent, were enrolled in the study.

Participant descriptions. Four young children with ASD, three boys and one girl, met the eligibility criteria and parents provided consent for research participation. Pseudonyms have been used for the children.

Participant #1. Rose was a Caucasian girl, 4 years, 5 months old, who lived with her older sibling and parents. She was diagnosed by a hospital-based multidisciplinary

team. Her cognitive assessment at the time of diagnosis placed her intellectual skills at the 1st percentile, as did confirmatory standardized testing during eligibility assessment (see Table 2 for more descriptive information). Her language level (on the PLS-5) fell below the 1st percentile; her speech was limited to one-syllable vocalizations including just one consonant-vowel combination, “ba”. She primarily communicated by leading people by the hand to what she wanted (occasionally pairing this action with a non-discriminant vocalization). Rose was not using PECS or any other AAC device, although her parents had attempted PECS in the last year. Her parents were involved in Hanen’s ‘More Than Words’ program (Carter et al., 2011), delivered privately. Shortly after Rose’s diagnosis, her parents received parent training in PRT, privately, which involved two hours of training, two years previous to participation in this study. Rose was receiving speech-language therapy (and a play program as part of these services) and non-ASD specific early intervention at the time of the study. Before the study intervention, Rose lacked basic toy play skills; she enjoyed toys with music and lights that she would recruit adults to activate for her. She also enjoyed having adults read books to her. Rose had received occupational therapy and physiotherapy services in the past. Some fine motor difficulties were present but not severe enough to limit toy play. No vision or hearing problems were reported by Rose’s parents. She attended an inclusive preschool classroom full-time, with the help of a full-time aide. Therefore, study treatment took place at her childcare centre; sessions occurred during the morning, in an empty classroom, with a familiar staff member present.

Rose’s mother rated aspects of Rose’s temperament, using the Childhood Behavior Questionnaire (Rothbart, Ahadi, Hershey, & Fisher, 2001); see Appendix C for

subscale labels and descriptions. Maternal ratings yielded scores reflective of low levels of high intensity pleasure, approach/positive anticipation, smiling/laughter, and activity.

Participant #2. Eli was 4 years, 5 months old when the study began. He had been diagnosed with ASD one month before his 4th birthday by a hospital-based multidisciplinary assessment team. He was the only child of a Caucasian family. On the M-P-R, which was administered by the author, Eli's intellectual functioning was below the 1st percentile. Using the PLS-5, Eli's expressive language skills were at the 1st percentile (see Table 2 for more descriptive statistics). He was reported by his parents to have approximately 10 words at the onset of the study, including a few well-practiced 'chunks' technically comprised of two words (e.g., "all done"), but no meaningful two-word combinations. His speech was unintelligible to the researcher. His family had used PECS for a brief period of time, in the past, but not consistently. Before intervention, his toy play was limited; for example, he enjoyed lining up cars and watching them go around on a track. In terms of developmental history, Eli was born two weeks prematurely and had hydrocephalus at birth (with a shunt inserted shortly thereafter). No hearing or vision problems were reported. He was delayed in walking and continued to have an awkward gait. The family had not been trained in treatment strategies as part of another behavioral intervention; however, parents participated in a publicly-delivered program similar to Hanen's 'More Than Words' (Carter et al., 2011). Eli participated in ASD-non-specific early intervention and had received speech-language therapy prior to participating in the current intervention. Eli had never attended a child-care centre. His intervention took place at his home. Data were collected in the living room or outdoors, with both parents present.

In terms of temperament, both of Eli's parents independently rated Eli's activity level, falling reactivity/soothability, high intensity pleasure, and inhibition as falling at levels observed in the typically-developing population for his age group. While Eli's father rated his son's anger/frustration comparable to his same-aged peers, Eli's mother rated it higher. Paternal ratings of Eli's shyness also fell within typically developing limits whereas his mother rated Eli as shyer than same-aged, typically developing peers. Both of Eli's parents rated his approach/positive anticipation and smiling/laughter below that of his same-aged, typically developing peers. Eli's father rated Eli's low intensity pleasure as lower than typically developing 4-year-olds, while Eli's mother's rating fell within the range of typically developing children.

Participant #3. Austin, the only child in his Caucasian family, was 4 years and 2 months old at the onset of the study. He was diagnosed with ASD by a clinical psychologist in private practice shortly after he turned 3 years of age. Austin was given a diagnosis of Global Developmental Delay before his ASD diagnosis. His intellectual functioning was below the 1st percentile (M-P-R), as was his language (PLS-5; see Table 2 for more information). Austin had two reported words, one of which was "no." The rest of his verbal expression constituted "gibberish" and was unintelligible by the researcher and parent. Austin received publicly delivered individual speech-language therapy for two years. Speech-language pathologists introduced PECS, but the family reported that they were not committed and did not use the system. Before intervention, Austin's play skills largely consisted of throwing toys, dropping them (apparently to hear the sounds), as well as flicking and spinning them, despite introduction of an informal play skills program by speech therapists. Austin's parents had not been trained in behavioral

treatment strategies as part of another intervention. Prior to the study, Austin had attended daycare three days per week for 3 hours, for less than a year, but withdrew for the summer months during which this study commenced. Therefore, treatment took place at home, in his living room, with usually only his mother present.

Both of Austin's parents independently rated aspects of Austin's temperament. Both parents rated Austin's anger/frustration, falling reactivity/soothability, inhibition, low intensity pleasure, as well as high intensity pleasure as falling within limits reported by parents of same-aged, typically developing peers. While Austin's father rated Austin's shyness as falling within the normal range, his mother's ratings were higher. Maternal ratings placed Austin's approach/positive anticipation and smiling/laughter within the normal range, whereas paternal ratings placed both of these aspects of his temperament below what is observed in the typically developing population of 4-year-olds. Both of Austin's parents rated Austin's activity level as low.

Participant #4. Sam was the first of two children born to a Caucasian family; he was aged 3 years and 9 months at the time that the study began. He was given an ASD diagnosis by a registered clinical psychologist, co-investigator with expertise in ASD-specific diagnostic assessment; Sam had been on a waiting list to be seen by the multidisciplinary diagnostic team. Sam scored below the 1st percentile on a standardized measure of intellectual functioning (M-P-R) and at the 1st percentile on a standardized measure of expressive language (see Table 2 for more information). Prior to this study, Sam had previously received non-ASD-specific early intervention as well as occupational therapy (biweekly for 3 months) for specific behavioral challenges such as difficulty transitioning between activities. Sam's mother reported that Sam's toy play was a

concern and that his occupational therapist, on one occasion, had attempted to show Sam how to play with his toys. While involved in this study intervention, Sam participated in private music therapy (once or twice, monthly, 30-minute sessions) and ongoing public speech-language therapy (two appointments per month, on average). Sam was very vocal, but not yet using words consistently. His mother reported that Sam had said 10 to 20 words in the last few years (at the single-word level) but used far fewer words consistently. She reported incidents when Sam would say a word and then never say it again, e.g., “go.” The author heard no words spoken by Sam throughout the three months of intervention. Sam’s parents had not yet decided whether to use PECS. To communicate, Sam’s mother reported that Sam brought objects to his parents or led his parents to desired objects. At the beginning of the study, Sam’s object play involved flipping pages of books, doing jigsaw puzzles (4-6 pieces), and building blocks. He did not play with any other objects in a functional manner. Sam’s parents had not participated in any ASD intervention prior to participation in this study intervention. Treatment took place at home, where Sam stayed full-time with his mother and younger sibling.

Both of Sam’s parents independently rated the following aspects of his temperament as being comparable with that of other children of similar age: anger/frustration, falling reactivity/soothability, high intensity pleasure; approach/positive anticipation, inhibition; and shyness. Both parents’ ratings yielded scores reflective of lower levels of low intensity pleasure, smiling/laughter, and activity.

Table 2

Participants' Pre-Treatment Assessment Results

	Auditory Comprehension (PLS-5)			Expressive Communication (PLS-5)			Total Language (PLS-5)			Intellectual Functioning (M-P-R)		
	SS	AE	%ile	SS	AE	%ile	SS	AE	%ile	SS	AE	%ile
Rose	50	0:11	1	50	0:5	1	50	1:9	1	<15	< 0:11	<1
Eli	75	2:11	5	53	1:3	1	62	2:1	1	38	2:6	<1
Austin	50	0:6	1	51	1:7	1	50	0:11	1	<15	< 0:11	<1
Sam	50	1:3	1	50	1:5	1	50	1:3	1	<15	< 0:11	<1

Note. “%ile” denotes percentile.

Measures

Child characterization measures. The following measures were used to collect demographic information, capture baseline abilities, and inform whether or not a child matched the target profile. Aside from the ADOS-2, administration of the child measures took approximately an hour and 45 minutes to complete.

The Autism Diagnosis Observation Schedule (ADOS-2). The ADOS-2 (Lord et al., 2012) is an observational assessment in which a trained examiner engages a child in activities designed to assess social and communication behavior indicative of ASD symptoms. This measure was administered by a clinical psychologist as part of each child’s diagnostic assessment. The use of Module 1 in the diagnostic assessment served as the initial flag that a child was potentially eligible to participate, as it means that the child is not yet using flexible word combinations.

The Preschool Language Scales – Fifth Edition. The primary (receptive and expressive) language measure was the Preschool Language Scale-Fifth Edition (PLS-5; Zimmerman, Steiner, & Pond, 2011). The PLS-5 is considered to be a sound measure for examining the receptive and expressive language abilities of children with ASD of varying developmental levels from birth to age 6 years, 11 months (Volden et al., 2011). The Total Language score was used as a measure of both language comprehension and expressive communication. An age-equivalent score of 15 months or less at baseline, served as the ‘low expressive language’ inclusion criterion. This measure took approximately 45 minutes to complete.

The Merrill-Palmer-Revised (M-P-R). Baseline cognitive abilities (using the Developmental Index age equivalent) were measured with the M-P-R (Roid & Sampers, 2004). The M-P-R is a measure of intellectual development for children aged one month to 6 years, 6 months. It demonstrates strong psychometric properties and is highly correlated with the Mental Score from the Bayley Scales of Infant Development, Second Edition (Bayley, 1993; $r = .92$). Its validity for assessing cognitive skills specifically in preschoolers with ASD has also been documented; it shows good concurrent and predictive validity with relevant measures (Dempsey et al., 2018). This scale was administered at baseline and took approximately 45 minutes to complete.

Family Background Questionnaire. Parents completed a brief demographic questionnaire about their child and family, including socio-demographic variables as well as the child’s previous intervention experiences and current services. This questionnaire took each child’s parents 2-3 minutes to complete.

Children's Behavior Questionnaire. Each child's temperament was measured prior to intervention using the parent-report Children's Behavior Questionnaire (CBQ, 3-7 years of age; Rothbart et al., 2001). Parents rated aspects such as "anger/frustration", "low intensity pleasure", and "shyness". The CBQ is a highly differentiated assessment of temperament and is commonly used in developmental research.

Family characterization and outcome measures. The following measures were used to describe each family and also to measure parents' impressions of treatment effects post-intervention. Together these forms took approximately 25 minutes to complete:

The Beach Center Family Quality of Life Scale (FQOL). The FQOL (Beach Center on Disabilities, 2006) served as a measure of family and emotional wellbeing. Specifically, it assessed families' perceptions of, and satisfaction with, different aspects of family quality of life specific to families with children with disabilities, ages birth to 21 years. The scale contains five subscales: Family Interaction, Parenting, Emotional Wellbeing, Physical/Material Wellbeing, and Disability-related Support. Test-retest reliability (i.e., correlations) for importance and satisfaction responses for each subscale has shown to be significant. The *item-level* overall scale, and the *subscale-level* overall scale have both been shown to have acceptable and excellent fit, respectively (Hoffman, Marquis, Poston, Summers, & Turnbull, 2006). Further, it has significant convergent validity with relevant existing measures (e.g., the Family Resource Scale; Van Horn, Bellis, & Snyder, 2001). This scale was completed pre- and post-intervention. It contains 25 items and took approximately 10 minutes to complete.

Parenting Efficacy Questionnaire. This form was completed by parents, pre- and post-intervention, and served as a measure of the extent to which parents felt competent and confident in using skills and strategies to help their children. It contains 21 items and took approximately 5 minutes to complete.

Parent Satisfaction Survey. This form was completed by parents, post-intervention, as a measure of the extent to which they felt either satisfied or dissatisfied with different aspects of their experience (related to their child's therapist, the treatment, etc.). There was space for open-ended comments. This form contains 10 items and took fewer than 5 minutes to complete.

Acceptability, Feasibility, and Social Validity Survey. To assess the acceptability, feasibility, and social validity of the intervention, parents rated 15 items on a 7-point Likert scale (1 = strongly disagree and 7 = strongly agree), post-intervention. Items reflected the extent to which parents agreed that the intervention a) was valuable; b) was a positive experience; c) provided a significant positive change for their child; d) was something that parents would recommend to others; etc. Parents were also asked to indicate a) how helpful the intervention was on a 5-point Likert scale (1: "not at all helpful" to 5: "extremely helpful") and b) how likely they were to continue using the intervention strategies learned, also on a 5-point scale (1: "not at all likely" to 5: "extremely likely"). There was also space on this form to offer comments. This form took fewer than 5 minutes to complete.

Dependent (intervention) measures and data collection. Toy play was measured in three different ways in order to capture play diversity. I not only measured children's frequency of functional toy play per session, but also the number of unique

functional actions they performed using toys as well as the number of unique toys they played with in a functional manner. Functional play was coded as present when the child performed an action physically independent of the researcher-therapist (i.e., when not physically prompted using hand-over-hand). All instances of spontaneous and imitated functional actions were considered to be functional play.

Frequency of Functional Play. The definition of toy play was based on Fossum's (2014) coding scheme (adapted from Sherer & Schreibman, 2005; Schreibman, Stahmer, Barlett, & Dufek, 2009; see Appendix D for coding scheme, also used by second rater, blind to study phase, for inter-rater reliability). Functional toy play was coded as having occurred when the child interacted with a toy according to its function (e.g., rolling train along the floor) or used the toy to represent another object in play (e.g., using a toy banana as a phone). Consistent with the RIT protocol, the set of duplicate toys used for RIT (the intervention context within which toy play was coded) included cars, trains, blocks, animals, kitchen utensils and serving ware, gardening tools and plants, musical instruments, nesting cups, picnic baskets with food, among other similar items. The total number of intervals in which appropriate behavior occurred was summed, and the proportion of total intervals in which appropriate behavior occurred was calculated.

Functional toy play was measured during the pre-intervention assessment, coded from the 10-minute video-recorded administration of the PEAR by the researcher-therapist. Functional toy play was then measured during the baseline phase, coded from video-recorded parent-child interactions before which the parent was told to play with their child as they typically would. These videos were recorded in the family's home (prior to parent training in responsiveness techniques). Toy play was measured during the

intervention phase, coded from video-recorded researcher-child interactions during which the researcher implemented RIT (also in the family's home for Eli, Austin, and Sam; at daycare, for Rose). The first 10 minutes of each RIT session were video-recorded and functional toy play was coded in 30-second intervals. Finally, toy play was also measured at the follow-up appointment, using the PEAR, administered by the researcher-therapist.

Number of Unique Functional Actions on Objects. Hereafter, this variable will be referred to as *variety of functional play actions*. Every instance that a child played with a toy functionally (using the definition above) was recorded. The number of unique actions on objects performed by each child was tallied for each 10-minute video-recorded session in which the child participated. Similar to the process of data collection for frequency of functional play, measurements took place pre-intervention, coded from the same 10-minute video-recorded administration of the PEAR; during the baseline phase, coded from the series of 10-minute video-recorded parent-child interactions; during the intervention phase, coded from 10-minute video-recorded researcher-child interactions during which the researcher implemented RIT; and, lastly, measured at the follow-up appointment, using the PEAR.

Number of Unique Objects Played with Functionally. Hereafter, this variable will be referred to as *variety of functional play objects*. As described for the aforementioned outcome variable, every instance that a child played with a toy functionally was recorded. The number of unique objects played with by the child was recorded and tallied for each 10-minute video-recorded session: at the pre-intervention time point, during the baseline and intervention phases, and at follow-up, as described above.

Positive affect. Positive affect was coded from the video-recorded PEAR administration to confirm the child's eligibility to participate (i.e., the child displayed low levels of positive affect, as per the 'low responder' profile, Fossum et al., 2018). Positive affect during this screening assessment was coded in one-minute intervals for a total of 10 intervals per (10 minute) video of the PEAR. The total score was an average of interval scores (i.e., interval 1 rating + interval 2 rating + ... + interval 10 / total number of scorable intervals). Videos were coded using Noldus The Observer behavioral coding software (Noldus, 2014).

Shared positive affect. The measurement of shared (dyadic) positive affect took place in the context of parent-child interactions coded from video-recorded behavior samples, using Noldus The Observer behavioral coding software (Noldus, 2014) during the baseline and 3-month intervention phases. During the baseline phase, shared positive affect was coded from interactions in which the parent was instructed to play with their child as they typically would. These videos were recorded in the family's home, prior to parent training in parent responsiveness techniques. Throughout the intervention phase, video-recorded behavior samples of parent(s) and their child were taken every three to four weeks, and parents were again instructed to simply play with their child. Duration data (in seconds) were collected from each 10-minute video. Positive affect was coded as having occurred and having been directed towards the social interaction partner if a child: smiled; laughed; jumped to express joy; or gestured (or vocalized) to express happiness, show interest, or request continuation of the dyadic interaction, *while also making eye contact or in response to an adult's action* (reflecting reciprocity). *Shared* positive affect was considered to have been present if the parent and child were *simultaneously* directing

positive affect towards one another. The duration of such events was measured in order to derive a proportion score by dividing said duration by the total duration of the video-recording (i.e., 600 seconds / 10-minutes). See Appendix E for the coding scheme.

Procedural reliability measure. The researcher-therapist's fidelity (i.e., adherence to pertinent aspects) of RIT implementation was measured by a naïve rater on 20% of the 10-minute video-recorded intervention sessions.

Fidelity of RIT Implementation form. This fidelity form (see Ingersoll & Lalonde, 2010; see Appendix F) included ratings of an implementer's behavior that ranged from 1 (Low Fidelity) to 5 (High Fidelity) for pertinent aspects of this training protocol: Contingent imitation, Linguistic mapping, Modelling action, Prompting, and Praising. A description of each rating (i.e., 1 through 5) for each of these five variables was included for the naïve rater. This form was used by the rater to code and assess the researcher-therapist's implementation of RIT during 10-minute video-recorded intervention sessions, for a subset of the intervention sessions for each child. Ratings of 3 or 4 were considered correct implementation of that skill area for fidelity summary scoring. The fidelity criterion was 80% correct implementation, i.e., the interventionist was considered to have met fidelity criteria if she demonstrated 80% correct implementation of each of the five RIT treatment techniques.

Procedure

Eligibility screening. Two children were identified via the NS EIBI waiting list (families who had given written consent to be contacted by our research team) and were subsequently enrolled in the study intervention. Two personal contacts from one of these families requested the investigator's email address and inquired about the opportunity to

participate in the research study (i.e., the latter two families recruited). A brief telephone interview with parents was undertaken to confirm each child's potential eligibility to participate, based on preliminary criteria (e.g., age, geographical location, services received, absence of impairment/disorder by parent report).

Pre-intervention assessment. To confirm if the child matched the profile of the children to whom this intervention was targeted, a series of measures was used to characterize the child's abilities during a screening assessment of language (PLS-5) and intellectual (M-P-R) abilities as well as levels of toy play and positive affect (PEAR). Families visited the Autism Research Centre at the local children's hospital for this appointment, where first they provided written, informed consent. After confirming that a child met these final eligibility criteria, parents and their children enrolled in the study.

The study intervention. The study intervention consisted of baseline, intervention, and follow-up phases.

Baseline. The purpose of baseline sessions was to record typical rates of parent and child behaviors. As appropriate for a multiple-baseline across-participant design, the baseline condition varied such that the experimental condition (i.e., intervention) was introduced after different baseline lengths for each child (ranging from 1 to 4 weeks, selected in random order for participants; Gast, 2010). A researcher visited the children's homes to video-record each child and their parent playing for 10 minutes; parents were instructed to play with their children as they normally would. Baseline sessions took place every two to three days for the 1- to 4-week baseline phase (whichever length was assigned to the family). Baseline videos were coded for each child's toy play skills and children's and parents' shared positive affect.

During this baseline phase, parents completed the Family Background Questionnaire, the Children's Behavior Questionnaire, the Beach Center Family Quality of Life Scale, and the Parenting Efficacy Questionnaire (see measures).

Parent coaching. Parent coaching took place following the baseline phase and before intervention commenced. It comprised approximately two hours of psychoeducation and coaching in responsiveness (not RIT) strategies that the researcher-therapist would use for the duration of the intervention phase. The didactic portion of parent coaching sessions included coverage of concepts and strategies used in parent responsiveness training and an overview of the platform for which these strategies could be used/practiced, e.g., sensory social routines. Related techniques were described by the researcher-therapist and practiced by the parent with in vivo coaching by the researcher-therapist. Techniques included responding promptly and sensitively in ways that were linked to their children's signals including tone of voice, pacing, positive affect, interacting without being physically intrusive, and verbalizing in ways that engaged the children's attention. Parent coaching was available throughout the 3-month intervention if requested by the parent or deemed advisable by the researcher-therapist (judged by parent-child video-recorded interactions every three weeks). Generally, the researcher-therapist checked in with the family at least once per week about their experiences implementing responsiveness strategies, their children's responses to these interpersonal strategies, what social routines seemed to elicit favorable responses, and to trouble-shoot any perceived barriers.

Intervention. After the target behaviors (toy play and shared positive affect) showed a stable pattern in the baseline phase, intervention was introduced. Intervention

was delivered in the children's natural environments (e.g., home, daycare) by the researcher-therapist, for 12 weeks, at 2-3 hours (i.e., sessions) per week. Each hour was divided into two 30-minute periods, with each 30-minute period dedicated to one of the treatment targets: toy play via RIT (Ingersoll, 2010; See Appendix G for protocol description), and shared positive affect via responsiveness training (Landry et al., 2006; See Appendix H for protocol description).

Post-intervention. Following 12 weeks of intervention, each child visited the Autism Research Centre at the local children's hospital so that the researcher-therapist could measure the child's toy play skills using the PEAR protocol (Flanagan, 2015) and assess any collateral gains in language using the PLS-5 (Zimmerman et al., 2011). The child's parent(s) were asked to complete the Beach Center Family Quality of Life Scale, the Parenting Efficacy Questionnaire, the Parent Satisfaction Questionnaire, and Acceptability and Feasibility Survey (see Measures).

Follow-up phase. At approximately one month post-intervention, a 10-minute video-recorded behavior sample between researcher-therapist and child was obtained to assess maintenance of children's treatment gains in toy play. A parent-child video-recorded behavior sample was obtained for the measurement of the child's shared positive affect. These behavior samples took place in the family's home.

Study design. A semi-concurrent (i.e., two children at a time, but offset by several weeks), multiple-baseline, single-case series experimental research design was used (Dugard, File, & Todman, 2012). Within such a design, the "case", i.e., participant, provides its own control for purposes of comparison. Each participant's behaviour for each outcome variable pre-intervention is compared to the outcome variable during and

post-intervention. Outcome variables are measured repeatedly within and across different conditions or “phases”, i.e., the baseline phase and the intervention phase. Single-case experimental design (SCED, also commonly referred to as single-subject research design or “SSRD”) standards are designed to address common threats to internal validity such as temporal precedence, history, maturation, etc.

The SCED is a rigorous scientific methodology often employed in applied psychology to establish evidence-based practices by documenting functional relations between independent and dependent variables. Typically, SCEDs are used in the development and evaluation of interventions designed to alter a specific human behavior (Kazdin, 2011) and are well suited to the study of highly heterogeneous disorders such as ASD. It is common in intervention research to progress from small-scale efficacy studies (including using SCEDs), to more controlled, robust, and larger experimental studies including RCTs. The SCED is suitable to address the current research questions for several reasons. First, establishing treatment efficacy for children exhibiting a specific behavioral profile demands a research design capable of providing fine-grained, time-series analysis of change in dependent variables across the systematic introduction of an independent variable (intervention). Second, the current study emphasizes dependent variables of high social importance. Concordant with research goals suited to SCEDs, this study hoped to demonstrate that the intervention produced an effect related to clinical need and could be applied with fidelity in typical intervention contexts. Further, I hoped to show that intervention agents (e.g., parents) report the procedures to be acceptable, feasible within available resources, and effective, and that parents report commitment to

their use following study participation. These study aims reflect the social validity of research goals inherent in a SCED.

Methodological rigor of the current experimental design was assessed using the “What Works Clearinghouse” (WWC) Single-Case Design Standards (Kratochwill et al., 2013), which were specifically developed to evaluate the internal validity of single-case designs. The WWC Design Standards consist of five criteria, four of which are coded on a dichotomous scale: a) systematic manipulation of the independent variable, b) dependent variable measured by more than one observer for at least 20% of sessions, c) inter-observer agreement (IOA) is at least 80%, and d) at least three attempts to demonstrate a basic effect at three different points in time (determined by the number of phases and phase contrasts). Each criterion is coded as present or absent for each case and all have to be coded “present” for a case to meet standards. The fifth WWC Design Standard criterion pertained to the number of data points per phase and is coded on a 3-point scale (meets standards, meets standards with reservations, or does not meet standards). To meet standards, each phase must have at least five data points per phase. To meet standards with reservations, each phase must have three or four data points per phase. If any phase has fewer than three data points, the case does not meet standards. A SCED can be evaluated, altogether, as meeting evidence standards, meeting evidence standards with reservations, or not meeting evidence standards.

The current study met WWC standards with reservations (Kratochwill et al., 2013). Consistent with criteria for SCED studies: the independent variable was systematically manipulated (i.e., the researcher determined when and how the independent variable conditions changed); the outcome variable was systematically

measured (i.e., measurement occurred over time, inter-rater reliability was included for every outcome variable, in every phase, for at least 20% of the sessions across all study conditions); and, meeting standards for experimental control, the study included at least three attempts to demonstrate an intervention effect [i.e., replication; see criteria in Byiers, Reichle, & Symons (2012)]. Although most phases included data from at least five measurement occasions, one phase provided three measurement occasions. In any case that a multiple baseline design study has fewer than five data points (but not fewer than three) to demonstrate an effect, the study is considered to *Meet Evidence Standards with Reservations*.

Given that one of my participants contributed just three data points across intervention targets (during her baseline phase), I offer discussion of an alternative view as it relates to implications, despite SCED guidelines that stipulate that more than three data points per phase are needed. Bartlett, Rapp, and Henrickson (2011) created 6,000 simulated multi-element design graphs and used visual analysis to determine the rate of false positives in graphs that contained two to five data points per data path. After applying visual analysis guidelines that dictated that researchers specify, a priori, the direction of behavior change, they found false positives in less than 5% of all graphs, with the exception of those graphs that included only two data points. Given this finding, Bartlett and colleagues (2011) proposed that researchers should conduct a minimum of three sessions per condition and specify the intended direction of behavior change.

Data analytic strategy. Analysis of SCED data involves graphical representation of the data and systematic visual analysis (Parsonson & Baer, 1978), as well as measures of effect size (Cohen, 1998). The multiple-baseline across participants design requires a

specific data pattern to demonstrate experimental control and to allow for claims that change in the dependent variables (toy play and shared positive affect) was a function of manipulating the independent variable (i.e., intervention). Visual analysis involves determining a) the immediacy of effects following intervention onset; b) the proportion of data points in adjacent phases that overlap in level; c) the magnitude of changes in dependent variables; and (d) the consistency of data patterns across multiple presentations of intervention and baseline conditions. Single case experimental design studies require that functional effects are documented with at least three participants (Gast, 2010).

Visual analysis (Kennedy, 2005; Lane & Gast, 2014) was conducted via investigation of the aforementioned parameters (i.e., variability, level, trend, immediacy of effect, and percentage of non-overlapping data between phases). Variability refers to the degree to which individual data points differ from the overall trend of the data. To determine the stability of level within each phase, a stability envelope was calculated; the stability envelope refers to those points within 25% of the baseline median. Subsequently, the percentage of data points within, or on, the stability envelope for each phase, was calculated. A change in level reflects a shift in the data at each point that the experimental conditions are changed (Kennedy, 2005). Absolute level change *within a phase* was determined by calculating the difference between the first and last value of that phase. Relative level change was determined by calculating the difference between the medians for each half of the data (within each phase). Trend refers to the best fit straight line that can be placed over the data within a phase in order to determine if the data are increasing or decreasing (i.e., improving or deteriorating) and to what extent; trend is assessed in

terms of slope/angle and magnitude. Trend, within each phase, was quantitatively estimated using the split-middle method of trend estimation. Relative level change *between phases* was determined by calculating the median value from the second half of the baseline phase from the median value from the first half of the intervention phase. Absolute level change (between phases) was determined by calculating the difference between the first value of the intervention phase and the last value of the baseline phase. Median and mean level changes were determined by calculating the difference between the median (or mean) of the baseline phase from the corresponding statistic of the intervention phase. Overlap of data refers to the degree to which data in adjacent phases (i.e., between baseline and intervention) are the same. Percentage of non-overlapping data between phases was calculated and reported. Immediacy of effect refers to the rapidity of change, i.e., the magnitude of change in level, trend, or variability between the last three to five data points in one phase and the first three to five data points in the next phase. How quickly the dependent variable changed after the introduction of the independent variable was examined. The *Standard Mean Difference* was used as a calculation of effect size for each variable—calculated by subtracting the mean of the baseline phase from the mean of the intervention phase and dividing by the standard deviation of the baseline (Busk & Serlin, 1992). Effect size conventions and criteria proposed by Cohen (1998) were used to differentiate small, medium, and large effects. An effect size was considered “small” if it was lower than 0.20, “moderate” if it was 0.20 to 0.60, “large” if it was 0.60 to 0.80, and “very large” if the value was greater than 0.80 (Vannest & Ninci, 2015).

Stability of baselines. A note is warranted about the feasibility of “stable” baseline phases of children with ASD, as it is often difficult to establish stable responding within this population at all (Brogan, Rapp, & Sturdivant, 2019). A steady state has been defined as a “pattern of responding that exhibits relatively little variation in its measured dimensional qualities over a period of time” (Johnston & Pennypacker, 1993, p. 370). Brogan et al. (2019) thoroughly discussed the parity in criteria for identifying adequately steady states in baseline data paths in order to evaluate an independent variable (e.g., intervention). Brogan et al. (2019) discussed the common occurrence of “transition states” between phases, i.e., when the first three or more consecutive data points in the intervention phase remain within the baseline level but there is an eventual demonstration of clear change in the dependent variable. A transition state may affect visual analysis by producing overlapping data points in the baseline and treatment phases, potentially increasing threats to internal validity (i.e., the lag in the effect of the independent variable; Kazdin, 2011). Identifying transition states and excluding those data points from calculations of percentage of non-overlapping data and other measures of effect size has its pitfalls and advantages (Brogan et al., 2019). Researchers who include transition states in their calculations may produce false negatives or smaller effect sizes for an independent variable. Unfortunately, it is also the case that data sets with transition states are not considered to show an effect and this may lead to publication bias (Brogan et al., 2019). In Brogan et al.’s examination of transition states in multiple baseline designs (using a total of 2,560 dependent variables), they found that transition states comprised an average of 4.7 data points (range = 3–19 data points, with a minimum of 3). Brogan et al. suggested omitting the first three (to five) sessions of the treatment phase and

collecting additional data points showing continued positive treatment effects. Of course, the authors highlighted the need for further research, especially as it relates to the use of this approach and its impact on effect size.

Altogether, some researchers acknowledge that high variability during the baseline phase may be more likely to produce a transition state, perhaps an inherent aspect of using SCEDs with developmental populations. Even though the current reported effect sizes are large for my dependent variables, this brief discussion is included to inspire flexibility in the interpretation of percentage of non-overlapping data between phases, given the underlying notion of a transition state. [In the current study, transition state data points were not omitted from any aspect of visual analysis, for any variable].

With regards to our secondary hypotheses, descriptive statistics were used to describe pre- and post- intervention parent-rated family quality of life and self-perceived parenting efficacy scores for each child's family.

Inter-rater reliability and treatment fidelity. Lastly, inter-rater reliability and treatment fidelity were calculated. The researcher-therapist served as the primary data collector throughout the study. An independent rater, an undergraduate student in Psychology blind to study phase, also collected data on the dependent variables for each participant, across study phases, and rated the researcher's treatment fidelity for 20-25% of RIT sessions. Intra-class correlation coefficients were used to assess inter-rater reliability for toy play variables. Kappa statistics were used to calculate inter-observer agreement for shared positive affect. Fidelity of implementation of treatment was calculated for each of five key RIT components by averaging ratings across all coded sessions.

Chapter 5: Results

Basic Effects

A “basic effect” is a change in the dependent variable (i.e., toy play variables and shared positive affect) when the independent variable is actively manipulated (i.e., introduction of an intervention). Documenting experimental control necessitates three demonstrations of an effect [via visual analysis (Lane & Gast, 2014), using adjacent phases from one participant’s data] at three times. Basic effects for variety of functional play actions were documented for Rose, Eli, and Austin, and for all participants’ variety of functional play objects. Basic effects were not observed for frequency of functional play. With respect to shared positive affect, basic effects were demonstrated for Rose, Eli, and Sam. These basic effects (and lack thereof for particular participants/variables) are explained in detail below.

Dependent Measures

Rose. Three baseline videos and 19 intervention sessions were coded for measurement of Rose’s frequency of functional play. Three baseline videos and five intervention sessions were coded for measurement of level of shared positive affect for Rose and her parents. As will be further described, below, the intervention served to increase Rose’s variety of functional play actions, variety of functional play objects, as well as her level of shared positive affect. Data did not indicate that the intervention increased the frequency with which Rose engaged in functional toy play.

Frequency of functional play. As indicated in Figure 1, during Rose’s baseline phase, she engaged in functional play with objects for 86.7% of a session, on average (range = 80-95%). When intervention began, Rose decreased the proportion of time she

spent engaging in functional toy play, to a mean of 58.9% intervals (range = 4-95%). The standard mean difference was used as an effect size and equaled -3.64 (as a result of the change in level *opposite* in direction to the intended and desired change). In terms of variability within the baseline phase, 100% of data points were within (or on) the stability envelope for the baseline phase. Data showed a slightly decreasing trend; it is important to note that the overall trend line was fitted on data from three measurement occasions that did not decrease in a linear fashion. The data within the intervention phase were more variable, with only 74% of the data falling within the stability envelope. Whereas the trend within the baseline phase appeared to be deteriorating, it was found to be improving within the intervention phase. Overall, there appeared to be a change in trend direction between baseline and intervention phases, from a slightly decelerating trend to a variable accelerating trend in a therapeutic direction. There was a relative and absolute deteriorating level change between baseline and intervention phases. There was also observed similar median and mean level changes in the contra-therapeutic direction. There were no overlapping data between phases, but *opposite* to the intended direction.

Variety of functional play actions. As indicated in Figure 2, during Rose's baseline phase, she performed an average of 2.7 unique and functional actions on objects (range = 2-4 actions). When intervention began, Rose increased her number of unique, functional actions on objects, performing an average of 7.9 actions (range = 2-13 actions). See Table 3 for an outline of Rose's functional toy play across phases. The standard mean difference was 4.5, representing a very large effect. Data within the baseline phase were somewhat variable: 67% of baseline data points fell within (or on) the stability envelope for that phase. Data within the intervention phase were more

variable; only 21% of intervention phase data points were within (or on) the stability envelope for that phase. Data showed an accelerating trend within both the baseline and intervention phases. There was a change in level between baseline and intervention phases. Although no *absolute* level change was observed (in part, due to the “transition state” observed at the beginning of the intervention phase; see Brogan et al., 2019), a *relative* improving change was observed (difference between the median of the second half of the baseline phase and the median value from the first half of the intervention phase). There were also observed similar median and mean level changes in the therapeutic direction. Due to the aforementioned transition state, immediacy of effect was lacking (i.e., delayed onset of effect was observed). There was 84% non-overlap of data between phases.

Variety of functional play objects. As indicated in Figure 3, during Rose’s baseline phase, she played functionally with an average of 4.3 objects (range = 4-5 objects). When intervention began, Rose increased the number of objects she interacted with appropriately, to an average of 7.4 objects per session (range = 2-12 objects). The standard mean difference was 5.26, representing a very large effect. In terms of variability within each phase, 100% of data points were within (or on) the stability envelope for the baseline phase, indicative of consistency among data points. The data within the intervention phase were more variable, with only 53% of the data falling within the stability envelope. The trend within the baseline phase appeared to be zero-accelerating, whereas it was improving within the intervention phase. Overall, there appeared to be a change in trend direction between baseline and intervention phases, from a stable, flat trend to a variable accelerating trend in a therapeutic direction. A relative

and absolute improving level change was observed between baseline and intervention phases. Similar median and mean level changes were also observed in the favorable direction. The level change was not considered to be immediate; 16% of data overlapped between phases (84% non-overlap of data between phases).

Semi-structured play assessment pre- and post-intervention. Rose's toy play skills were assessed using the PEAR before intervention began and one month after intervention ended as a measure of maintenance and generalization of learned skills. At both occasions, Rose was assessed with the researcher-therapist. Prior to intervention, Rose played functionally with toys for a total of 5% of the 30-second intervals within the 10-minute play period. She performed two unique, functional actions on objects and played functionally with two different toys. After intervention, Rose played with toys functionally as part of the PEAR assessment for 10% of the intervals within the 10-minute play period, doubling her pre-intervention rate. She performed equally to her pre-intervention level by displaying two different actions on objects and playing with two different toys.

Shared positive affect. As indicated in Figure 4, during Rose's baseline phase, she and her parent(s) shared positive affect for 7% of a session, on average (range = 2.5-15.5%). During the intervention phase, following parent training and the commencement of the child's intervention with the researcher-therapist, Rose and her parents increased the time they shared positive affect with each other, to an average of 37.2 within sessions (range = 4.3-69.0%). The standard mean difference was 4.10, representing a very large effect. Data within the baseline phase were more stable than in the intervention phase. Within the baseline phase, 66.7% of data were within (or on) the stability envelope,

whereas only 20% of data in the intervention phase were within the stability envelope. The trend within the baseline phase, as well as the intervention phase, appeared to be deteriorating; however, the data were quite variable. A level change was observed between baseline and intervention phases. Relative and absolute level improved between baseline and intervention, as did median and mean level changes to a similar degree. The level change was large and immediate. Finally, there was 80% non-overlapping data between phases.

Eli. Five baseline videos and 17 intervention sessions were coded for measurement of Eli's level of toy play. Five baseline videos and four intervention sessions were coded for measurement of level of shared positive affect for Eli and his parents. As will be described in detail, below, the intervention served to increase Eli's variety of functional play actions, variety of functional play objects, as well as his level of shared positive affect (same outcomes as Rose). Data did not indicate that the intervention increased the frequency with which Eli engaged in functional toy play.

Frequency of functional play. As indicated in Figure 1, during Eli's baseline phase, he engaged in functional toy play with objects for 73.4% of the session (range = 55-85%). During intervention, Eli increased the proportion of time he spent engaging in functional toy play to a mean of 83.8% intervals (range = 60-100%). The standard mean difference between phases was 0.81, representing a large effect. In terms of variability, 80% of data points were within (or on) the stability envelope for the baseline phase. For the intervention phase, 83% of the data fell within the stability envelope. The trend within the baseline phase appeared to be improving, whereas it was found to be deteriorating within the intervention phase; however, the data to which the trend line was fit were quite

variable. Overall, there appeared to be a change in trend direction between baseline and intervention phases, from a variable accelerating trend to a variable decelerating trend in a contra-therapeutic direction. However, relative and absolute level improved between baseline and intervention. There were similar median and mean level changes. The level change was considered to be immediate. There was 53% non-overlapping data between phases.

Variety of functional play actions. As indicated in Figure 2, during Eli's baseline phase, he engaged in an average of 3 unique, functional actions on objects (range = 1-5 actions). When intervention began, Eli increased the number of unique, functional actions on objects, performing an average of 11.8 actions (range = 6-18 actions). See Table 3 for an outline of Eli's functional toy play across phases. The standard mean difference was 5.50, reflecting a very large effect. Data within each phase were highly variable: 20% of baseline data points and 29% of intervention phase data points were within (or on) the stability envelope for that phase. Data showed a zero-celerating trend within the baseline phase and an accelerating trend in the therapeutic direction, within the intervention phase, indicating a favorable direction change between phases. There was a change in level between baseline and intervention phases. Relative and absolute level changes (as well as similar median and mean level changes) was seen between baseline and intervention, all in the therapeutic direction. The change in level was immediate and there was 100% non-overlap of data between phases.

Variety of functional play objects. As indicated in Figure 3, during Eli's baseline phase, he played functionally with an average of only 2.6 objects (range = 1-4 objects). During intervention, Eli increased the number of unique objects he appropriately played

with, to an average of 6.8 objects per session (range = 4-13 objects). The standard mean difference was 3.70, representing a very large effect. In terms of variability within each phase, 40% of data points were within (or on) the stability envelope for the baseline phase. The data within the intervention phase were also highly variable, with only 12% of the data falling within the stability envelope. There appeared to be no change in trend direction between baseline and intervention phases; the trend within both phases appeared to be accelerating in a therapeutic direction. There was, however, relative and absolute improving level changes between baseline and intervention phases, as well as similar median and mean level changes in the favorable direction. The level change was immediate and the percentage of non-overlapping data between phases was 77%.

Semi-structured play assessment pre- and post-intervention. In a structured play interaction with the researcher-therapist at baseline (PEAR), Eli played with toys for a total of 75% of the (30-second) intervals within the 10-minute period. He performed 9 unique, functional actions on objects and played, functionally, with 9 different toys. After intervention, Eli played with toys, functionally, for 90% of the intervals within the 10-minute assessment period. He exceeded his pre-intervention performance by performing 12 different actions on the toys and matched his pre-intervention performance by playing with 9 different toys.

Shared positive affect. As indicated in Figure 4, during Eli's baseline phase, Eli and his parent did not share positive affect on any of the five measurement occasions. Following parent training and intervention by the researcher-therapist, Eli and his parent spent an average of 41.5% of a session engaged in shared positive affect (range = 36.6-44.9%). The standard mean difference could not be calculated, as the difference between

means could not be divided by zero (i.e., the standard deviation of the baseline phase). In terms of variability, 80% of data points were within (or on) the stability envelope for the baseline phase. For the intervention phase, 83% of the data fell within the stability envelope (all baseline data points were identical, i.e., there was no variability). There was a change in trend between the baseline and intervention phase, from zero-celerating to slightly accelerating in a therapeutic direction. Relative and absolute level improved between baseline and intervention phases. Similar median and mean level changes were also observed. The level change was considered to be immediate and there was 100% non-overlapping data between phases.

Austin. Eight baseline videos and 18 intervention sessions were coded for measurement of Austin's level of toy play. Eight baseline videos and four intervention sessions were coded for measurement of his and his parents' level of shared positive affect. As will be described in detail, below, the intervention served to increase Austin's variety of functional play actions and variety of functional play objects. Data did not indicate that the intervention increased the frequency with which Austin engaged in functional toy play, nor his level of shared positive affect with his parents.

Frequency of functional play. As indicated in Figure 1, during Austin's baseline phase, he engaged in functional play with objects for 19.4% of the session, on average (range = 0-55%). During intervention, Austin increased the proportion of time he spent engaging in functional toy play to a mean of 44.6% of intervals (range = 25-72%). The standard mean difference was 1.10, representing a large effect. Data within each phase was highly variable; none of the data points within the baseline phase were within (or on) the stability envelope for that phase. Data showed an increasing trend. The data within

the intervention phase were also highly variable, with only 17% falling within the stability envelope. The trend within the intervention phase appeared to be slightly deteriorating, but overall variability was high across this phase. Overall, there appeared to be a change in trend direction between baseline and intervention phases, from a variable, accelerating trend to a variable, decelerating trend in a contra-therapeutic direction. With respect to change between baseline and intervention phases, relative and absolute improving level changes between baseline and intervention were seen. Similar median and mean level changes in the therapeutic direction were observed. There was only 17% non-overlap of data between phases, however.

Variety of functional play actions. As indicated in Figure 2, during Austin's baseline phase, he performed, on average, 1.9 unique and functional play actions with objects per session (range = 0-5 actions). During intervention, Austin increased the number of functional actions on objects to an average of 6.7 actions per session (range = 3-11 actions). See Table 3 for an outline of Austin's functional toy play across phases. The standard mean difference was 2.20, representing a very large effect. Data within each phase was highly variable: only 25% and 22% of data points for the baseline and intervention phases, respectively, fell within (or on) the stability envelope for that respective phase. Data showed a variable, decelerating (contra-therapeutic) trend within both the baseline and intervention phases, in part due to the variability displayed from one session to the next within either phase. Although there was a deteriorating relative level change for the intervention session, there was observed a small absolute *improving* change in trend within this phase (difference between the first and last intervention data points). There was ultimately no change in trend direction between baseline and

intervention phases; although, there was a change in level between baseline and intervention phases. There was observed a relative and absolute improving level change between baseline and intervention, as well as similar median and mean level changes in the therapeutic direction. The change in level was immediate. There was 72% non-overlap of data between phases.

Variety of functional play objects. As indicated in Figure 3, Austin played functionally with an average of only 1.5 objects per session during the baseline phase (range = 0-4 objects). During intervention, Austin increased the number of unique objects he appropriately played with, to an average of 7.1 objects per session (range = 4-10 objects). The standard mean difference was 3.50, representing a very large effect. The data within each phase were highly variable: 25% of data points in the baseline phase and 22% of data points in the intervention phase fell within (or on) the stability envelope for its respective phase. The trend within the baseline phase appeared to be deteriorating, but data were highly variable and the trend line did not reflect a consistent downward trend. Data in the intervention phase appeared to be accelerating in a therapeutic direction. Overall, there was a change from a variable deteriorating-decelerating trend in baseline to an improving-accelerating trend during intervention. Relative and absolute (improving) level changes between baseline and intervention phases were also observed, as well as similar median and mean level changes in the same, therapeutic direction. The level change was immediate and the percentage of non-overlapping data between phases was 94%, where the lowest intervention data point matched the highest baseline data point on one occasion.

Semi-structured play assessment, pre- and post-intervention. Prior to intervention, during the PEAR assessment, Austin played with toys for a total of 28% of the 30-second intervals within the 10-minute period, performed 3 unique, functional actions on objects, and played functionally with 4 different toys. After intervention, Austin engaged in functional toy play for 40% of the intervals within the 10-minute period. He performed 6 different actions on the toys and played with 8 different toys, doubling his pre-intervention rate.

Shared positive affect. As indicated in Figure 4, during Austin's baseline phase, he and his parent(s) shared positive affect with one another for 4.1% of a session, on average (range = 0.7 to 13.8% of a session). During the intervention phase, Austin and his parents increased the amount of time within a session during which they shared positive affect, to an average of 22.7% (range = 7.1-48.9%). The standard mean difference was 4.18, representing a very large effect. Data within the intervention phase were variable. The trend within the baseline phase was slightly accelerating-improving (but appeared flat to the naked eye); trend within the intervention phase appeared to be decelerating in a contra-therapeutic direction. This deteriorating trend within the intervention phase was in stark contrast to Austin's trend with respect to shared positive affect with the researcher-therapist. Shared positive affect between Austin and the researcher-therapist showed an accelerating-improving trend during the intervention phase, with an average of 36.27% of sessions characterized by shared positive affect and 100% non-overlap with baseline data. Mean and median level changes between baseline and intervention phases for Austin and the researcher-therapist was double the level changes observed between Austin and his mother. There was observed, however, a level

change between baseline and intervention phases for shared positive affect between Austin and his mother. Relative and absolute level improved, greatly, between baseline and intervention, and the level change was immediate. Median and mean level changes between phases were also observed. Finally, there was only 50% non-overlapping data between phases.

Sam. Eight baseline videos and 17 intervention sessions were coded for measurement of Sam's toy play. Eight baseline videos and 6 intervention sessions were coded for measurement of Sam's (and his mother's) level of shared positive affect. As will be described in detail, below, the intervention served to increase Sam's variety of functional play objects and his level of shared positive affect. Data did not indicate that the intervention increased his number of unique, functional play actions, nor the frequency with which Sam engaged in functional toy play.

Frequency of functional play. As indicated in Figure 1, during Sam's baseline phase, he engaged in functional play with objects for 83.8% of the session, on average (range = 36-100%). During intervention, Sam decreased the proportion of time he spent engaging in functional toy play to a mean of 65.9% of intervals (range = 30-94%, highly similar to the wide range of data observed in the baseline phase). Similar to Rose, this decrease from baseline to intervention level could be a function of the types of actions on objects displayed during the baseline phase as compared to the intervention phase when objects became more "toy-like"; see Table 3 for an outline of Sam's functional toy play across phases. The standard mean difference was used as an effect size and equaled -0.82 (again, as a result of the change in level *opposite* to intended and desired change). In terms of variability within each phase, 87.5% of data points were within (or on) the

stability envelope for the baseline phase and data showed a slightly accelerating trend. The data within the intervention phase were more variable, with only 70.6% of data falling within the stability envelope. The trend within the intervention phase appeared to be deteriorating but was highly variable from session to session. Overall, there appeared to be a change in trend direction between baseline and intervention phases, from a variable and slightly accelerating trend to a variable decelerating trend in a contra-therapeutic direction. There was observed a relative and absolute deteriorating level change between baseline and intervention phases. Congruous with these measures of level change, there was also observed similar median and mean level changes in the contra-therapeutic direction. There was 0% overlapping data between phases, but in the opposite direction to that intended (i.e., all intervention data points were within range of the maximum baseline value).

Variety of functional play actions. As indicated in Figure 2, during Sam's baseline phase, he performed, on average, 4.1 unique and functional play actions with objects per session (range = 2-7 actions). During intervention, Sam doubled the number of functional actions on objects to an average of 8.3 actions per session (range = 5-13 actions). The standard mean difference was 2.50, representing a very large effect. Data within each phase were variable: 62.5% and 35.3% of data points for the baseline and intervention phases, respectively, fell within (or on) the stability envelope for that respective phase. Data showed a variable, accelerating, therapeutic trend within both the baseline and intervention phases. There was ultimately no change in trend direction between phases, but there was a change in level between baseline and intervention. There was observed a relative and absolute improving level change between phases, as well as

similar median and mean level changes in the therapeutic direction. The change in level was immediate. Although the range of data points in each phase minimally overlapped with one another, in range, there was 53% non-overlap of data between phases.

Variety of functional play objects. As indicated in Figure 3, Sam played functionally with an average of only 3.8 objects per session during the baseline phase (range = 2-5 objects). During intervention, Sam increased the number of unique objects he functionally played with, to an average of 6.7 objects per session (range = 2-10 objects). The standard mean difference was 2.50, representing a very large effect. Data within each phase were variable: 75% of baseline data points and 59% of intervention phase data points were within (or on) the stability envelope for that respective phase. Data showed an accelerating trend within both the baseline and intervention phases. Although there was no change in trend direction between phases, there was a change in level. There was observed a small, relative improving level change between baseline and intervention (no absolute level change was observed, i.e., difference between the last value of baseline and the first value of intervention – in part, reflecting the lack of change in trend between phases). There was also observed similar median and mean level changes in the therapeutic direction. The change in level appeared to be immediate. There was 77% non-overlap of data between phases.

Semi-structured play assessment pre- and post-intervention. At baseline, during the PEAR protocol, Sam played with toys for a total of 45% of the (30-second) intervals within the 10-minute play period. He performed 8 unique, functional actions on objects and played functionally with 9 different toys. Sam maintained gains made during intervention and exceeded his pre-intervention play performance. After intervention,

during the play assessment (i.e., the PEAR), Sam played with toys, functionally, for 75% of the intervals within the 10-minute period. He performed 11 different actions on the toys and played with 11 different toys.

Shared positive affect. As indicated in Figure 4, during Sam's baseline phase, he and his mother shared positive affect with one another for an average of 2.2% of a session (range = 0.5-7.4%). During the intervention phase, Sam and his mother increased the amount of time within a session during which they shared positive affect with each other, to an average of 61.3% (range = 38-77.9%). The standard mean difference was 26.10, representing a very large effect. Data within the baseline and intervention phases were somewhat variable. A quarter of the data within the baseline phase fell within (or on) the stability envelope for that phase and 33.3% of data within the intervention phase fell within its stability envelope. The trend within the baseline phase appeared to be just slightly decelerating-deteriorating but resembled more of a flat (zero-celerating) trend line that hovered near zero. In sharp contrast, the trend within the intervention phase appeared to be accelerating in a therapeutic direction. There was observed absolute and relative level changes between baseline and intervention phases. Mean and median level changes between phases were also impressively high. The level change between baseline and intervention was immediate. Finally, there were no overlapping data between phases, i.e., percentage of non-overlap = 100%.

Table 3

Progression of Toy Play from Baseline to Intervention Phases for each Participant

	Examples of Toy Play Skills during Baseline	Examples of Toy Play Skills during Intervention
Rose	<ul style="list-style-type: none"> • Turned pages of book that parent was reading to her • Pressed a button on a light-up toy • Put a fish in a box tank • Popped bubbles <p>[exhaustive list]</p>	<ul style="list-style-type: none"> • Tapped the drum with a stick • Nested the cups • Placed one block on top of another • Jingled a musical ball • Rolled a train • Knocked over a wooden tower • Put a muffin in a muffin tray • Rolled a ball • Pushed a car • Took apart wooden blocks • Took a ring off/put a ring on plastic tower • Made a pony hop • Put a bowl in a picnic basket • Closed the basket lid • Brushed pony's hair • Shook a shaker egg and tambourine • Mixed food in pan with spatula
Eli	<ul style="list-style-type: none"> • Stuck magnets together • Filled a cup with water and poured it out • Sat on a bouncy ball and bounced • Pushed a button on a lamp • Rolled a car • Placed a sticker on his shirt • Locked locks on a busy board • Matched Paw Patrol characters to their picture cards • Put magnets on a board <p>[exhaustive list]</p>	<ul style="list-style-type: none"> • Played drums with sticks • Put muffins in a muffin pan • Raced cars with researcher • Built tower and fence using blocks • Watered plants with toy jug • Fed himself pretend food and water using toy dishes • Put frosting on muffin • Mixed food in a pan using spoon; put cover on pot; stirred soup using new pot • Loaded back of truck with strawberries, drove truck, unloaded truck • Pretended that his block was a rocket soaring through air • Made a bridge using wooden blocks; connected trains and pulled them through; parked trains in wooden parking spaces • Pretended trains were sleeping/waking • Made wooden block launch-pad for spaceship

Austin	Examples of Toy Play Skills during Baseline	Examples of Toy Play Skills during Intervention
	<ul style="list-style-type: none"> • Caught a ball and threw it in the air • Held and watched Elmo doll talk • Flipped a switch on the lights/sounds toy train • Put beads around his neck • Put a shape in a shape sorter <p data-bbox="412 772 602 800">[exhaustive list]</p>	<ul style="list-style-type: none"> • Banged on a drum using stick and hands • Stacked nesting cups • Built and disassembled two Mega Blocks • Rolled a car • Threw and caught a ball; bounced ball • Turned on a spinning light-up toy • Opened box, put a bunny in his home, closed box • Made bunnies hop on the ground • Filled picnic basket with “bread” • Spun the wheel of a well • Took food (muffin) out of pan/took dishes out of picnic basket • Brushed pony’s hair • Watered the plant with water jug • Cooked using pan and spatula • Put a bunny in his carrot home/put a spoon in a bowl/took lettuce out of bowl • Spun a fishpond around; removed fish from the pond • Fed toy corn to a horse
Sam	<ul style="list-style-type: none"> • Dipped his paintbrush in paint • Used finger to put paint on paper • Pressed shape into sand; filled shape with sand • Squished sand in his hand • Connected and took apart math cubes • Put cubes into a container and took them out • Put a block on a string/took a shape off a rod • Put marker to paper • Put marker back in tin can • Put puzzle pieces in an inset board 	<ul style="list-style-type: none"> • Put Mega Blocks together and took them apart • Banged drum with stick • Stacked nesting cups • Put the pony on top of a mountain • Took a bunny out of his home • Pushed a car • Stirred food using a drumstick and a pan • Built a tower and a fence using wooden blocks; knocked the tower down • Put a muffin in a pan and removed it • Placed a pot on the tray • Put picnic items in basket and closed lid • Pretended to pour water from one jug into another • Stood two ponies, upright, next to each other • Spun a fishpond and removed fish from the pond • Put a bowl on a plate; put a spoon in a bowl • Put the toy spoon to his mouth (pretended to eat)

Examples of Toy Play Skills during Baseline

Examples of Toy Play Skills during Intervention

- Took plant grass and strawberries out of its planter and put back in pot
 - Dug soil in potted plant, using garden fork
 - Put gnome on top of a “castle”
 - Took food out of a bowl using spoon and fed examiner with spoon
 - Took fish out of frying pan
-

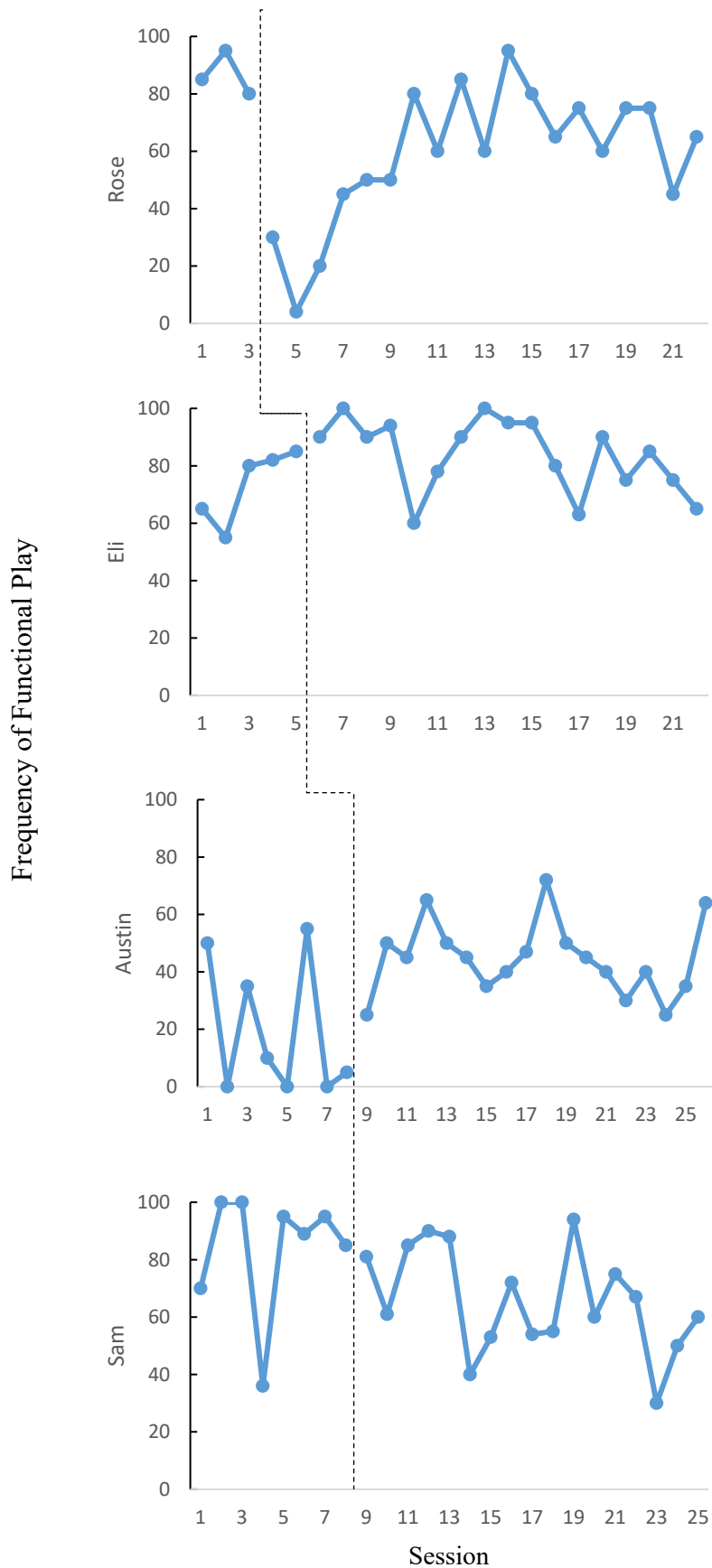


Figure 1. Percentage of session intervals during which the child engaged in functional toy play.

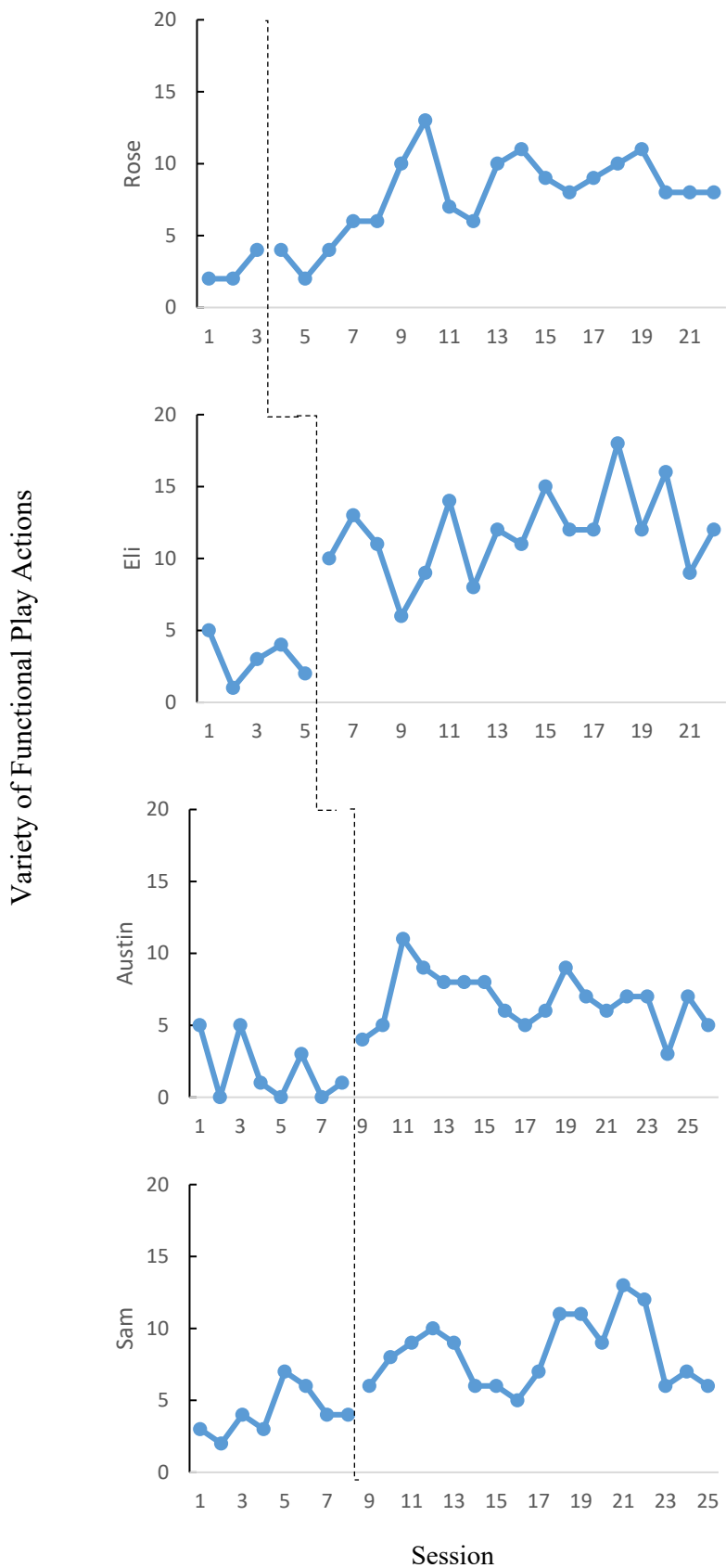


Figure 2. Number of unique functional actions on objects per session.

Variety of Functional Play Objects

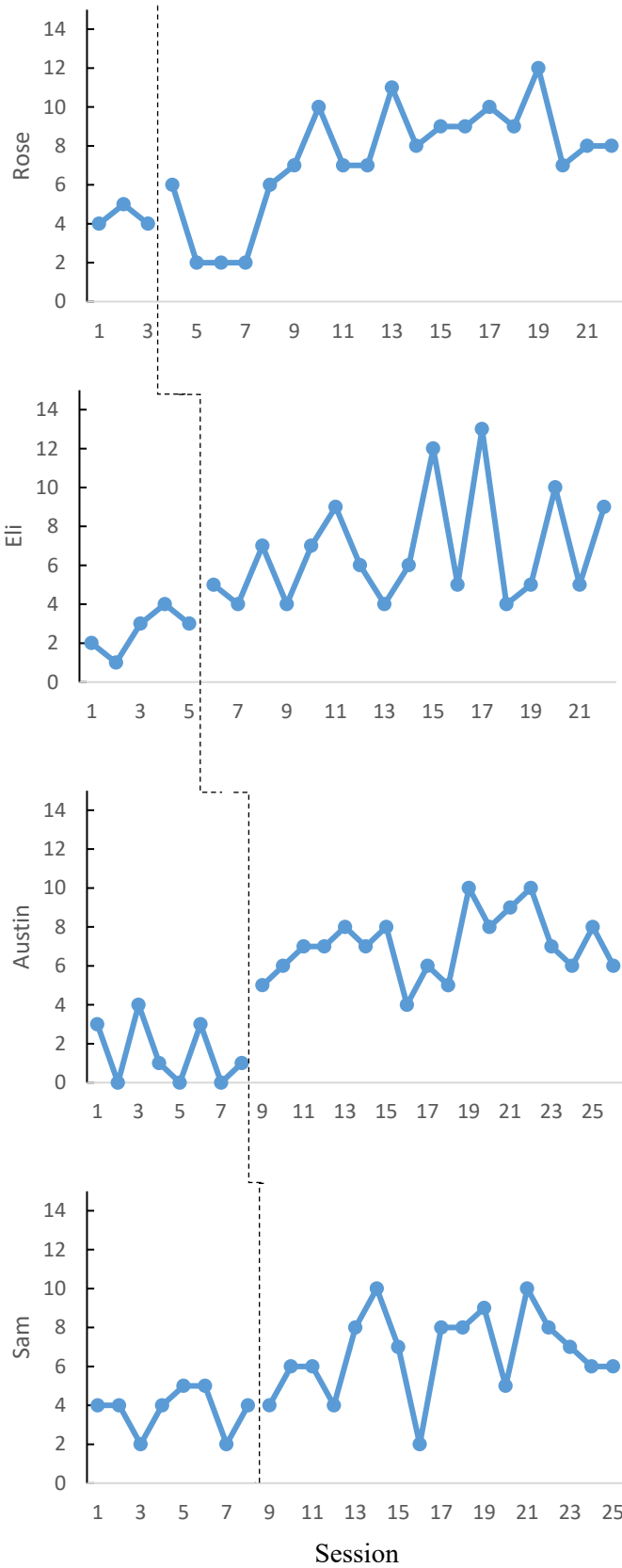


Figure 3. Number of unique objects interacted with functionally per session.

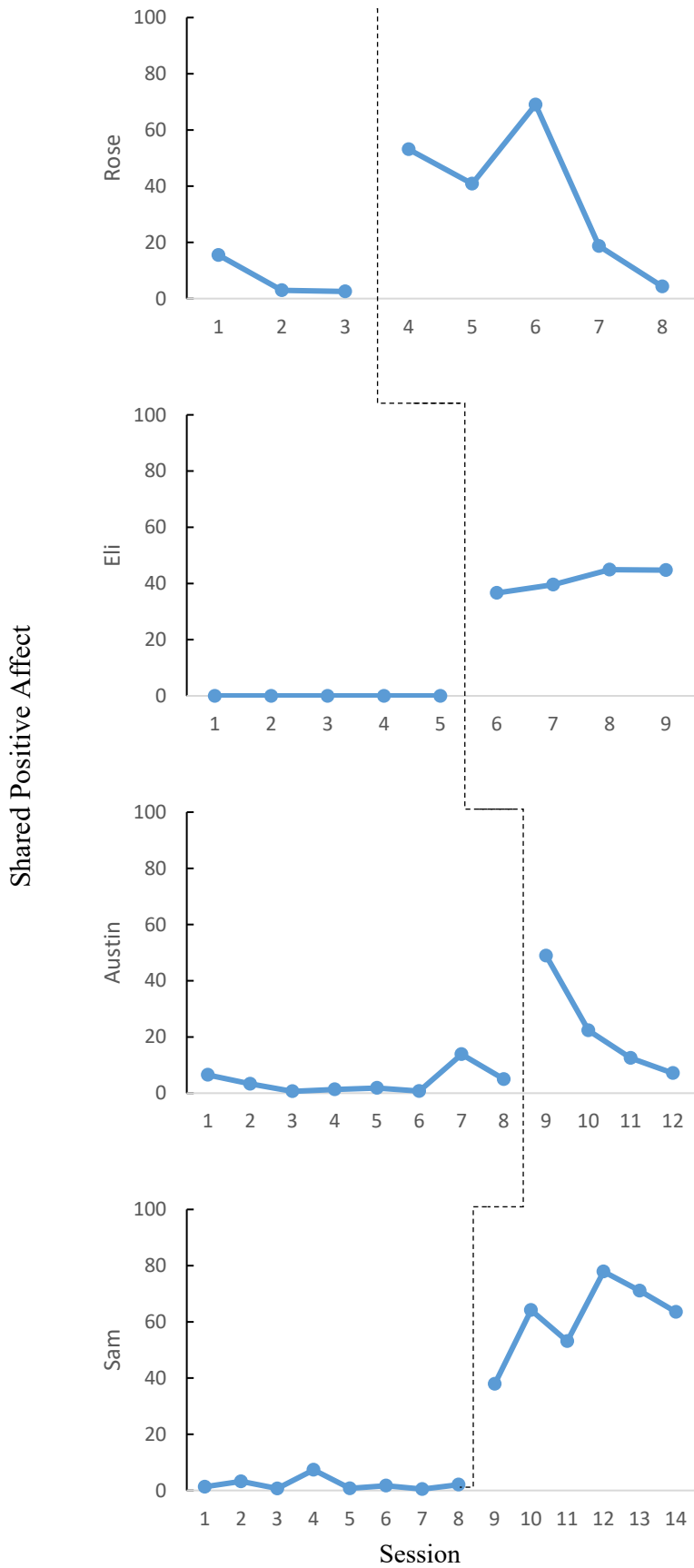


Figure 4. Percentage of session intervals during which the child and parent shared positive affect.

Functional Relations

The determination of whether a functional relation exists for any intervention target involves ascertaining whether data, across phases, document experimental control. The data presented in the current study provide a convincing demonstration of control by the independent variable with regard to changes in level, trend, variability, immediacy of effect, non-overlapping data between phases, as well as effect size for two of three measures of toy play: a) variety of functional play actions and b) variety of functional play objects (see Tables 4 and 5, respectively, for an overview of the elements of visual analysis used to determine the presence/absence of an effect for each outcome variable). However, a functional relation was *not* observed for frequency of functional play (see Table 6 for overview of visual analysis elements). The current data also provide compelling evidence of experimental control and a functional relation between baseline and the introduction of parent-responsiveness training in order to boost children's and parents' shared positive affect (see Table 7 for overview of visual analysis elements). Importantly for the context of using a SCED, the demonstrated functional relations between the independent variables and dependent variables represent socially valid impacts on the target behaviors of concern, i.e., on behaviors that hold high clinical significance for the population of preschoolers diagnosed with ASD. A more detailed overview of basic effects observed, by participant and variable, is provided, below.

A basic effect for frequency of functional play was not observed for any child. For variety of functional play actions, a basic effect was observed for Rose, Eli, and Austin. However, there was a change in level for Sam, in the therapeutic direction, and immediacy of effect was observed [with a very large effect size (26.10), 53% of Sam's

data in the intervention phase overlapped with that of his baseline phase]. For variety of functional play objects, a basic effect was observed for all four participants. For shared positive affect, a basic effect was observed for Rose, Eli, and Sam. Although Rose's overall trend line was calculated as "deteriorating" in both phases, the variability within her data was very high. Her trend is less meaningful in the presence of a level change of high magnitude between her baseline and intervention phases. Austin's data showed an impressive change in level between his baseline and intervention phases (increase in time spent engaged in sharing positive affect with parent from a mean of 4% of a session to 23%); however, data overlapped by 50% between both phases. It is important to note, though, that his intervention phase comprised only four measurement occasions.

Evaluation of Whether Current Evidence Meets SCED Standards

The total intervention, as packaged (i.e., RIT and parent-responsiveness training), served to improve behaviors in *both* intervention target areas, i.e., toy play and shared positive affect, in three of the four participants: Rose, Eli, and Sam. Austin did not experience an intervention effect for shared positive affect. Breaking down the intervention components, RIT successfully increased variety of functional play objects for all participants. Thus, there is moderate evidence that the PRIMeR module improves variety of functional play objects (if it were not for Rose's baseline phase comprising three data points, this intervention would have met strong evidence standards; see Byiers et al., 2012, and Kratchowill et al., 2013, for a summary of standards). Reciprocal imitation training successfully increased variety of functional play actions for three of four participants. For the fourth participant (Sam), all indicators were met, aside from percentage of non-overlapping data. As such, there is moderate evidence that the

PRIMeR module improves variety of functional play actions. Parent- and therapist-responsiveness training served to convincingly boost shared positive affect for three of four participants: Rose, Eli, and Sam, thus providing moderate evidence for the intervention. For Austin, all indicators were met aside from percentage of non-overlapping data.

Table 4

Summary of Basic Effects Observed: Variety of Functional Play Actions

	Level	Trend	Immediacy of effect	PND	Variability	Cohen's <i>d</i> effect size
*Rose	Improving between phases	Improving in both phases (in therapeutic direction)	Absent (but presence of a transition state)	84%	High	4.50 Very large
*Eli	Improving between phases	Zero-accelerating to improving (in therapeutic direction)	Present	100%	High	5.50 Very large
*Austin	Improving between phases	Deteriorating in both phases (in contra-therapeutic direction)	Present	72%	High	2.20 Very large
Sam	Improving between phases	Improving in both phases (in therapeutic direction)	Present	53%	High	26.10 Very large

Note. (*) denotes the presence of a basic effect for that participant. PND = percentage of non-overlapping data (between phases).

Table 5

Summary of Basic Effects Observed: Variety of Functional Play Objects

	Level	Trend	Immediacy of effect	PND	Variability	Cohen's <i>d</i> effect size
*Rose	Improving between phases	Zero-celerating to improving (in therapeutic direction)	Present	84%	High in intervention phase	5.30 Very large
*Eli	Improving between phases	Improving in both phases (in therapeutic direction)	Present	77%	High	3.70 Very large
*Austin	Improving between phases	Deteriorating to improving (in therapeutic direction)	Present	94%	High	3.50 Very large
*Sam	Improving between phases	Improving in both phases (in therapeutic direction)	Absent (but presence of a transition state)	77%	High	2.50 Very large

Note. (*) denotes the presence of a basic effect for that participant. PND = percentage of non-overlapping data (between phases).

Table 6

Summary of Basic Effects Observed: Frequency of Functional Play

	Level	Trend	Immediacy of effect	PND	Variability	Cohen's <i>d</i> effect size
Rose	Deteriorating between phases	Zero-celerating to improving	Effect was contra-therapeutic to baseline	100%, but in the contra-therapeutic direction	High	-3.64 Large, but in the wrong direction
Eli	Improving between phases	Improving to deteriorating	Present	53%	High	0.81 Large
Austin	Improving between phases	Improving to deteriorating	Effect was contra-therapeutic to baseline	17%	High	-0.82 Large, but in the wrong direction
Sam	Deteriorating between phases	Improving to deteriorating	Present	100%, but in the contra-therapeutic direction	High	1.10 Large

Note. (*) denotes the presence of a basic effect for that participant. PND = percentage of non-overlapping data (between phases).

Table 7

Summary of Basic Effects Observed: Shared Positive Affect

	Level	Trend	Immediacy of effect	PND	Variability	Cohen's <i>d</i> effect size
*Rose	Improving between phases	Deteriorating in both phases (in contra-therapeutic direction)	Present	80%	High in intervention phase	4.10 Very large
*Eli	Improving between phases	No slope to slightly improving (in therapeutic direction)	Present	100%	Stable	Large to very large, but could not be calculated because denominator was zero
Austin	Improving between phases	Slightly improving to deteriorating (in contra-therapeutic direction)	Present	50%	High in Intervention phase	4.18 Very large
*Sam	Improving between phases	No slope to improving (in therapeutic direction)	Present	100%	High in Intervention phase	26.1 Very large

Note. (*) denotes the presence of a basic effect for that participant. PND = percentage of non-overlapping data (between phases).

Inter-rater Reliability

Toy play variables. For each dependent variable, 21% of all video-recordings across baseline and intervention phases were coded by a second rater, blind to study phase. Videos were randomly selected from each phase of each participant's data set. The intervention phase was divided in half prior to random video selection to ensure

approximately equivalent representation from early versus later intervention (across the 12 weeks). Prior to inter-rater coding, the blind rater and the researcher coded two practice videos together. The blind rater then coded the dataset selected for reliability statistics, intermixing videos across participants and phases so that videos were not coded in any particular order.

Intra-class correlation coefficients (ICC) and their 95% confidence intervals (CI) were calculated for each of the toy play variables using SPSS statistical package (version 25; IBM Corp., 2017) based on a two-way mixed effects model (Koo & Li, 2016). The ICC and 95% CI for frequency of functional play was 0.83 (0.54-0.93). According to a guideline of selecting and reporting ICCs for reliability research (Koo & Li, 2016), this correlation is considered good, with the CI spanning a wide range, from moderate to excellent. For variety of functional play actions, the ICC was 0.97 (CI: 0.92-0.99), falling in the excellent range. The ICC and 95% CI for variety of functional play objects was 0.94 (0.86-0.97), also falling in the excellent range.

Shared positive affect. Twenty-seven percent of participants' videos were coded for inter-rater reliability. The average index of concordance across videos scored for all participants was 0.87 (i.e., 87% agreement; standard deviation = 0.10). Indices of concordance ranged from 0.71 to 1.00. Important to note is that coding for inter-rater reliability yielded three indices of concordance that fell below three standard deviations of the mean (out of 30 inter-rater reliability scores). These three scores were considered to be outliers and were removed from the dataset. Kappa (Cohen, 1960) was also calculated; mean $\kappa = 0.64$ indicating "substantial agreement". Kappa values for every video were statistically significant ($p < 0.0001$). It is acknowledged that guidelines in the

literature for the magnitude of kappa are not universally accepted, therefore, indices of concordance, i.e., inter-observer agreement (Jansen, Wiertz, Meyer, & Noldus, 2003) are also reported.

Fidelity of Implementation of RIT

The researcher-therapist's fidelity of RIT implementation was measured by a naïve trained rater for 20% of the 10-minute video-recorded intervention sessions targeted at boosting children's toy play. Kappa coefficients were calculated for each of the five core components included for measurement of fidelity of implementation. Calculations yielded kappa coefficients of .94 for contingent imitation, 1.00 for linguistic mapping, .99 for modelled action, 1.00 for prompting, and 1.00 for contingent reinforcement.

Social Validity

Social validity was assessed on the basis of parents' satisfaction with the intervention, the perceived relevance and importance of the objectives, the adequacy of the procedures, and recognition of clinical significance (i.e., perceived effects).

Family quality of life. Each child's parent(s) reported their family's quality of life using the Beach Centre's Family Quality of Life Scale. This scale uses satisfaction as the primary response format; items are rated on a 5-point scale where 1 = *very dissatisfied*, 3 = *neither satisfied nor dissatisfied* and 5 = *very satisfied*.

Rose's mother rated their family quality of life, prior to intervention. Maternal ratings yielded a mean of 3.48 [standard deviation (SD) = 0.65] which remained identical following intervention (SD = 0.77). Rose's father did not complete the survey before intervention began, but rated his family's quality of life after intervention similarly to

Rose's mother: 3.64 (SD = 0.64). Generally, Rose's parents felt neither satisfied nor dissatisfied across time points.

Eli's parents' self-reported family quality of life scores at baseline were similar to one another and indicated satisfaction (father: mean = 4.52; SD = 0.51 and mother: mean = 4.80; SD = 0.41). Both parents' scores remained similar following intervention [father: 4.64 (0.49) and mother: 4.80 (0.41)].

Austin's mother's ratings of her family's quality of life, pre-intervention, yielded a mean of 4.52 (SD = 0.51). These ratings remained consistent following intervention [4.16 (0.69)], indicative of satisfaction across time points.

Family quality of life was reported similarly, at baseline, by both of Sam's parents: father = 3.68 (1.07) and mother = 4.92 (0.28). Both parents' scores remained the same following intervention: father = 3.92 (0.95) and mother = 4.28 (0.89) and indicated satisfaction across time points.

Self-perceived parenting efficacy. Before and after intervention, children's parents rated to what extent they felt competent and confident using strategies to help their children. This scale uses frequency with which a parent feels efficacious in various parenting areas as the primary response format; items are rated on a 5-point scale where 1 = *never*, 3 = *sometimes* and 5 = *very often*.

For Rose, paternal self-perceived parenting efficacy at baseline was higher than maternal ratings [father: 4.60 (0.99); mother: 3.33 (0.58)] and reflected feeling efficacious some of the time to often. Both parents' ratings decreased somewhat after intervention, with both parents' mean scores at 3.19 (0.81).

Eli's father rated his self-efficacy as slightly higher than did Eli's mother at baseline: 4.05 (0.67) versus 3.40 (0.60). Eli's mother reported an increase in self-perceived parenting efficacy following intervention [to 4.60 (0.99)], reflecting feeling efficacious often to very often, whereas Eli's father remained similar to his pre-intervention ratings, with a mean of 3.71 (0.56).

For Austin, maternal self-reported ratings of parenting efficacy were similar before and after intervention [4.19 (0.93) to 4.05 (1.36)], reflecting often feeling self-efficacious in parenting her child.

Sam's father rated his self-perceived parenting efficacy as slightly lower than did Sam's mother pre-intervention [3.32 (0.67) versus 4.19 (0.93), respectively]. Following intervention, Sam's mother rated her parenting self-efficacy similarly to pre-intervention, 4.05 (SD = 1.36), reflecting feeling self-efficacious often. Sam's father did not complete this questionnaire post-intervention.

Parent satisfaction with the intervention. Following intervention, each child's parent(s) completed a survey that captured how satisfied or dissatisfied they felt regarding the intervention on a scale of 1 to 5 (*not helpful at all to extremely helpful*). Data were missing from Austin's mother.

All respondents (both of Eli's parents; both of Rose's parents, and Sam's mother) rated the interventionist's responsiveness to their questions and concerns as "extremely helpful". Eli's parents rated the training's capacity to increase their child's toy play skills as "extremely helpful"; Rose's parents described this intervention as "somewhat helpful" and Sam's mother rated it as "helpful". Eli's parents also rated the training's capacity to increase their child's shared smiling with the parent as "extremely helpful"; Rose's

parents and Sam's parents rated this aspect as "helpful". Both of Eli's and both of Rose's parents rated the "live coaching" they received at the beginning of the intervention as "extremely helpful"; Sam's mother rated it as "helpful". Eli's mother, both of Rose's parents, and Sam's mother all described the length of the training period as "just right"; Eli's father indicated that it was "too short". All parents rated the number of home visits and the length of visits to be "just right". In terms of overall experience, Eli's parents, Rose's mother, and Sam's mother rated their experience as "extremely helpful"; Rose's father rated it as "helpful" and reported that "given the amount of training already participated in", this served as "an enhancement in the quality of time spent with Rose." When asked about what each parent found to be most helpful about the training sessions and/or the intervention, Eli's dad offered that he "learned how to interact better with [his son]". Eli's mother noted that the connection she witnessed between her son and the interventionist was a highlight, as well as "learning new tricks". Rose's father noted the interventionist's "positive connection with [his daughter]", in addition to "consistent and frequent sessions" as highlights. Rose's mother offered that she found the "one-on-one time with [her daughter] was helpful" during coaching and implementation of strategies outside of intervention, in addition to "the parent coaching, good feedback, and support". When asked for suggestions about how else the investigator could make the training and/or intervention experience more helpful, the only parent to offer a suggestion was Rose's mother; she suggested "videotape review and progress reports of intervention sessions".

Acceptability, feasibility, and social validity. Following intervention, each child's parent(s) completed a survey that captured how acceptable, feasible, and socially

valid they felt the intervention was, on a 7-point Likert scale (1 = *strongly disagree*; 4 = *neutral*; and 7 = *strongly agree*). Overall, parents' ratings from the social validity questionnaire indicated that parents found the intervention to be feasible and acceptable (see Table 8).

Eli's parents both indicated that they *agreed* or *strongly agreed* with all statements about the intervention's value and related benefits (see items in Table 8). Austin's mother *strongly agreed* to all statements, except for one (having been able to teach other family members to use the learned strategies, to which she *agreed*). For Sam, maternal ratings indicated that his mother *agreed* to all statements except for three statements related to having noticed meaningful changes in her son, which she deemed *neutral*. Rose's parents' ratings were more variable. Rose's mother *agreed* with every statement with four exceptions: she *somewhat agreed* that she noticed meaningful increases in her Rose's toy play and level of shared positive affect and also that other people noticed a significant positive change in her child. In terms of noticing meaningful increases in her child's imitation of people in her environment, she rated this statement as *neutral*. Paternal responses were even more variable across items, ranging from *disagree* to *strongly agree*. He *disagreed* that he noticed meaningful increases in his daughter's imitation of people in her environment and felt *neutral* about the extent to which he was confident that the learned skills would make a meaningful difference in his daughter's development, whether or not his family would continue to use this intervention model, and whether he noticed meaningful increases in Rose's toy play and shared positive affect with others. Rose's father's ratings fell in the *somewhat agree* to *strongly agree* range for the rest of his responses.

Table 8

Parents' Mean Ratings of Acceptability, Feasibility, and Social Validity of Intervention

Survey item	Mean rating (SD; range)
This intervention model was:	
Valuable to my family	6.12 (0.75; 5 to 7)
A positive experience for my child and me	6.50 (0.55; 6 to 7)
Easy to incorporate into my family life	6.33 (0.82; 5 to 7)
Not complicated to learn	6.33 (0.82; 5 to 7)
Easy to use	6.33 (0.82; 5 to 7)
Easy to teach other family members	6.33 (0.52; 6 to 7)
Provided a significant positive change for my family	5.83 (1.17; 4 to 7)
An intervention I used at home on a regular basis	6.12 (0.75; 5 to 7)
An intervention I will continue to use	6.17 (1.17; 4 to 7)
An intervention I would recommend to other parents	6.33 (0.82; 5 to 7)
I noticed:	
Meaningful increases in my child's toy play	5.67 (1.51; 4 to 7)
Meaningful increases in my child's imitation of people in his/her environment	5.12 (2.14; 2 to 7)
Meaningful increases in my child's level of positive affect / the extent to which s/he shares emotion	5.50 (1.38; 4 to 7)
Other people noticed a significant positive change in my child	6.00 (1.10; 5 to 7)
I feel confident that skills I have learned will help me make a meaningful difference in my child's development	6.17 (1.17; 4 to 7)

Note. SD = standard deviation.

Chapter 6: Discussion

In this dissertation, I sought to develop and evaluate the efficacy of an intervention designed to remediate behavioral deficits associated with poor progress during a PRT-based intervention program, based on empirically determined profiles, in preschoolers diagnosed with ASD. Four children, aged between 3 and 5 years, participated in a multiple baseline across participants SCED study. Children represented those predicted to make minimal gains in the PRT-based program; that is, children exhibited low intellectual functioning and were minimally verbal, and also expressed low levels of positive affect and displayed low levels of toy contact (Fossum et al., 2018). The aim of the intervention was to shift low responders' skills in toy play and the frequency with which these children shared positive affect with their parent(s). These changes were intended ultimately to increase children's responsivity to PRT strategies. Each child received 12 weeks of the PRIMeR intervention package, which involved 2–3 hours per week of therapist-implemented RIT and responsiveness training in order to boost toy play and shared positive affect, respectively. Parents were coached in responsiveness strategies.

The goal of the dissertation was to generate efficacy data that may support adaptations to individual programming within PRT-based intervention programs, yielding positive outcomes for a wider range of children. This study provides initial support for the efficacy of this partially parent-mediated naturalistic and developmental treatment package, PRIMeR, in shifting preschoolers' low responder profiles by boosting skills in areas that are theoretically important for optimal response to PRT-based programs.

Impact of the PRIMeR Module on Functional Toy Play

It was hypothesized that this brief parent- and therapist-mediated intervention would increase levels of appropriate toy play in children diagnosed with ASD who matched the low responder profile. An intervention effect (functional relation between baseline and the introduction of treatment in at least three cases) was obtained for two of three aspects of children's toy play: 1) variety of functional play actions (Rose, Eli, and Austin) and 2) variety of functional play objects (all four participants). At one-month follow-up, Eli and Austin (but not Rose) exhibited higher than baseline rates of number of functional play actions. With respect to number of functional play objects, Eli, Austin, and Sam showed higher than baseline rates at the follow-up time-point. An effect of intervention was not observed for frequency of functional play. It could have been that children's engagement in restricted and repetitive behavior and thus inattention/disengagement during RIT sessions precluded participation in toy play activities across intervals of a session. That is, while children learned to interact with a variety of toys in a variety of ways, the amount of time spent engaged in toy play (i.e., frequency of functional play) did not improve as a function of the intervention.

In this study, the PRIMeR module was effective in boosting variety of functional play and extends research showing that brief, low-intensity NDBIs have an impact on play skills, even for lower functioning children with ASD. For example, children who participated in a JASPER pilot RCT (Goods et al., 2013) demonstrated greater play diversity compared to TAU (ABA-based therapy) in a sample of similar age and language ability to the current participants (minimally verbal 3- to 5-year-old children with ASD), while also using similar treatment intensity. JASPER showed promise in

improving play and engagement outcomes for children who were making limited progress despite receiving intensive behavioral interventions. Progress was made in a short period (12 weeks) with a low dose of intervention (two 30-minute weekly sessions). The current study's findings are therefore consistent with previous studies' findings using similar parameters in terms of design (brief and low-intensity treatment) as well as baseline characteristics of participating children (i.e., lower functioning). As another example, baseline characteristics of children included in a SCED using RIT to boost imitation skills (Ingersoll & Schreibman, 2006) were quite similar to the characteristics of children included in the current study. The children in Ingersoll and Schreibman's (2006) study ranged in age from 29 to 45 months at intake (2.40 to 3.75 years) with mental ages ranging from 15 to 29 months (on the Bayley Scales of Infant Development, 2nd edition; Bayley, 1993). Language ages ranged from less than 8 months (non-verbal) to 25 months; four of the five children required ADOS Module 1 (i.e., up to single words only), and two of the five were characterized as severely autistic by the Childhood Autism Rating Scale (Schopler, Reichler, DeVellis, & Daly, 1980). Ingersoll and Schreibman (2006) showed that RIT with this relatively low functioning group of preschoolers was effective in boosting children's imitation and pretend play skills and that gains were maintained at 1-month follow up.

Rationale for use of the PRIMeR module is consistent with research suggesting that toy play may need to be explicitly targeted within NDBI programs for some preschoolers with ASD. Contaldo et al. (2020) examined initial individual characteristics that may have contributed to progress in skills acquisition following one year of ESDM. They examined each ESDM developmental domain as reflected on the ESDM

Curriculum Checklist (i.e., subscale scores for Receptive and Expressive Communication, Social Skills, Imitation and Play) and by a parent-report MB-CDI subscale score, Actions with Objects. They found that progress for the fast responding group (versus the slow responding group) was significantly associated with a) increased baseline imitation and social skills (fast responders imitated 8–11 one-step actions on objects), and b) increased baseline actions with objects and play abilities (fast responders played independently and appropriately with 10 one-step toys, pushed a toy car or truck, threw a ball, or mixed a cup or a bowl with a spoon). Slow responders would have thus imitated fewer one-step actions and played appropriately with fewer toys; these levels of abilities align very closely with findings from the NS EIBI low responder profile (Fossum et al., 2018). The documentation of deficits in children’s imitation and play abilities as they relate to slower/poorer progress in NDBIs highlights the potential value of targeting such skills prior to or concurrently with participation in some NDBIs.

Interventions that target play, such as the PRIMEr module, can have far-reaching impacts. Following a meta-analysis of NDBIs for young children with ASD, Tiede and Walton (2019) discussed the idea that even small progress in key domains such as functional play is clinically meaningful. Children’s initiation of behavior in key domains could create a cascade of collateral benefits. That is, the child may increasingly create their own learning opportunities and thus increase the frequency with which adults may capitalize on such instances to promote children’s learning (Koegel, Bradshaw, Ashbaugh, & Koegel, 2014; Koegel, Carter, & Koegel, 2003).

Possible contributory factors for current findings. A possible explanatory factor for findings specifically related to Sam’s toy play involves pre-treatment level of

restricted and repetitive behavior (RRB). Sam's level of RRB interfered with RIT activities on an ongoing basis. This may explain, in part, why an intervention effect was not obtained for Sam with respect to variety of functional play actions. Often, when Sam performed a functional play action with a toy, he would begin (or return to) using the toy in a repetitive non-functional manner. Keeping a particular toy in hand long enough to perform multiple functional actions with it rarely occurred, thereby limiting Sam's total number of functional play actions during any one session. Variety of functional play objects improved during intervention, perhaps due to the diligence of the researcher-therapist in introducing new objects on a continual basis, when he was engaged.

Restricted and repetitive behavior (motor actions/movements as well as physical and/or sensory manipulation of objects; APA, 2013) constitutes a core symptom of ASD and is commonly manifested by younger children with developmental delays and language impairment (Harrop et al., 2014; Lam, Bodfish, & Piven, 2008). Through factor analysis (Honey, McConachie, Turner, & Rodgers, 2012; Szatmari et al., 2006), RRBs have been categorized into higher- and lower-order behaviors. Lower order RRBs consist of stereotyped motor behavior or sensorimotor rituals (Szatmari et al., 2006). These behaviors sometimes preoccupy children and interfere with developmentally appropriate/necessary activities (Anthony et al., 2013). Further, restricted interest in specific objects or toys has been associated with repetitive manipulation of toys, such as spinning or arranging them (Lydon, Healy, & Leader, 2011). Children who display RRB may behave as persistent because of interest; frustrated; driven instead of playful; upset if prevented from interest; and inattentive to other tasks (Smerbeck, 2019). Repetitive behaviors and routines are therefore likely to reduce engagement and play skills (Boyd,

Conroy, Mancil, Nakao, & Alter, 2007) and, indeed, have been associated with inattention to instruction to conventional EIBI (Lovaas, 1987). It has been hypothesized that a preference for/inclination towards such lower-order repetitive behaviors over social engagement may predict poorer response to EIBI (Klintwall & Eikeseth, 2012). Specific to RIT, repetitive stereotypic behaviors have been associated with poorer progress as well (quasi-RCT; Malik, 2016; dissertation).

Children's level of RRB has implications for future studies. Firstly, perhaps RRB should be measured as part of the set of characteristics that may predict progress during PRT; indeed, increased stereotyped and repetitive *vocalizations* was part of the low responder profile established by Fossum et al. (2018). Secondly, for low responders participating in PRIMeR who exhibit RRB that interferes with implementation of RIT, perhaps RRB could be targeted with treatment prior to or alongside participation in PRIMeR. Future research could also assess possible reductions in RRB through the PRIMeR intervention itself.

Impact of the PRIMeR Module on Affect

It was hypothesized that this brief parent- and therapist-mediated intervention would increase levels of shared positive affect in children diagnosed with ASD who matched the low responder profile. An intervention effect, i.e., a functional relation between baseline and the introduction of treatment in at least three cases, was obtained for shared positive affect (Rose, Eli, and Sam).

Results of the current study indicated that children's sharing of positive affect was amenable to improvement through this PRIMeR intervention. This is consistent with previous research showing that use of a relational strategy in the context of parent-

mediated intervention increases synchrony within parent-child interactions and children's attentiveness to parents (in infants at high risk for ASD), as well as reducing prodromal ASD symptoms (Green et al., 2015). The importance of improving sharing of positive affect between child and parent was highlighted in Wan, Green, and Scott's (2019) systematic review of parent-infant interaction in infants/toddlers at risk for and with emerging ASD. They suggested that intervention targeting parent-child interactions may be beneficial for optimizing social and communicative outcomes. Specifically, Wan and colleagues (2019) suggested that dyadic reciprocity may be an important intervention target for children with ASD and their parents. Parental sensitivity and parental synchrony have been shown to enhance the communication and social aspects of parent-child interactions when the young child has ASD (Aldred, Green, & Adams, 2004; Green et al., 2010). Research suggests that these two variables play a vital role in the effectiveness of various treatments in enhancing outcomes such as "reciprocity of social interaction toward others" [e.g., joint attention interventions (Kaale, Smith, & Sponheim, 2012; Kasari, Gulsrud, Wong, Kwon, & Locke, 2010); and parent-mediated interventions to increase responsive parental behaviors and children's communication (Siller, Hutman, & Sigman, 2013)]. Tachibana and colleagues (2017) suggested, following a meta-analysis of 14 RCTs of EIBIs and NDBIs for preschoolers with ASD, that "parental synchrony" should be considered an essential and promising target for early interventions for children with ASD. Targeting parents' shared positive affect through responsiveness teaching aligns with this conceptual target, different language notwithstanding. Gains in shared positive affect due to the PRIMeR module should increase readiness to benefit from PRT-based programs where the quality of parent-child relationships is important for

parents' effective use of strategies.

Possible contributory factors for current findings. Parenting styles and abilities, especially as they relate to diminished parental responsiveness, may have contributed to the null findings for shared positive affect for one parent-child participant dyad. Parent-driven differences in parent-child interactions may have partially accounted for the lack of a shared positive affect intervention effect for Austin. This was especially noteworthy, as Austin showed frequent and high levels of positive affect directed towards the researcher-therapist. The broader autism phenotype (BAP) may be a consideration for some families. The BAP refers to sub-clinical ASD-related characteristics observed in individuals without ASD, e.g., parents of children with an ASD diagnosis, and is more prevalent in fathers than mothers of children with ASD (Losh, Childress, Lam, & Piven, 2008). The BAP includes such characteristics as social pragmatic difficulties, poor communication skills, and impaired emotion recognition (Gerds & Bernier, 2011). Hartley, Hickey, DaWalt, and Rodriguez (2019) discussed the notion that high BAP in fathers may hinder emotional intimacy through an impaired ability to empathize and communicate emotions. Facial affect is considered to be a distinct endophenotype of BAP (Bolte & Poustka, 2003); diminished facial affect would have implications for a parent's effective use of responsiveness strategies. Variation in parents' own styles of interacting with their children may influence their ability or inclination to learn and/or use responsiveness strategies with their children. Taking parental profiles into consideration in research interventions that encompass a parent coaching element is an area in great need of empirical development.

Additional parental behaviors, aside from level of responsiveness towards children, may influence the reciprocal nature of the parent-child relationship. The style in which parents play with their children may vary across families. Anecdotally, parents in this study, who had been instructed to play as they naturally would with their children, often focused on demonstrating pre-academic skills such as identification of letters, colors, and shapes, or counting. Generally, the baseline phases for each participant were characterized by such parental style of interacting during free play as well as by low levels of positive affect directed towards their child. Some parents found it hard to shift from teaching these pre-academic concepts during their interactions, even after coaching to follow their child's lead and to use fun, social routines. Indeed, research on parenting styles suggests that as a group, parents of children with ASD may show a higher level of directiveness than those of typically developing children matched by developmental language age (Freeman & Kasari, 2013). Research also shows this pattern in parent-infant interactions; parents of high-risk ASD siblings are more directive than parents of low-risk infants at six months (Wan et al., 2012) and nine months (Harker, Ibanez, Nguyen, Messinger, & Stone, 2016). As Wan and colleagues (2012) suggested, it could be that since children with ASD are less likely to orient socially or initiate joint attention (Adamson, Bakeman, Suma, & Robins, 2019; Jones & Klin, 2013), parents may find it more difficult to follow their children's play (Adamson et al., 2017). From an early developmental period, interaction with a less emotionally responsive child may influence parental behavior. Indeed, Dawson, Hill, Spencer, Galpert, and Watson (1990) found that mothers of 2.5- to 6-year-old children with ASD smiled less frequently and were also less likely to show responsive smiling towards their children's smiles, compared to mothers

of typically developing children. Parenting style with infants at high risk for ASD has been shown to predict growth in their social smiling. In a study by Harker and colleagues (2016), parent directiveness predicted slower growth in children's social smiling between 9 and 18 months (Harker et al., 2016). However, when maternal directiveness was taken into account, high-risk infants exhibited greater growth in social smiling than their low-risk counterparts (Harker et al., 2016). These findings suggest that parenting style, especially when reflective of responsiveness and reciprocity, contributes to the development of social engagement in infants at high and low risk for ASD. Further, Yirmiya and colleagues (2006) found that parents of infants at high risk for ASD (with an older sibling with ASD diagnosis) exhibited less synchronous coordination of engagement states when interacting with their infants than did parents of low-risk infants, suggesting that parents change their behavior based on their children's social-emotional characteristics. It could be that responsiveness training is especially important for parents whose children are low in expressed positive affect, as in the current study. Its use may be particularly warranted in our low responders and their parents. Of course, we do not know to what extent parenting styles of interacting versus children's skills and abilities contributes to the disruption in reciprocal parent-child interactions. Examining the direction of effects within behavioral dyadic interactions requires a large sample size and a powerful research design such as structural equation modelling (path analysis).

Impact of the PRIMeR Module on Parents' Perceived Self-Efficacy, Family Quality of Life, and Satisfaction

I hypothesized that the intervention would increase parent-reported family quality of life and perceived parenting self-efficacy. Although I hypothesized that parents' sense

of family quality of life/well-being would increase following participation in this intervention, parents' ratings were consistent across pre- and post-intervention time points. With respect to parents' perceived self-efficacy, parents who completed the measure at both time-points also reported similar levels pre- and post-intervention with two exceptions. The first exception was Eli's mother who reported a post-intervention increase in parenting efficacy. The second exception was the decrease in parenting efficacy scores at post-intervention, compared to pre-intervention, for Rose's parents. Delayed completion of these surveys overlapped with receipt of NS EIBI parent coaching. Their decreased ratings may have reflected their new state of learning and the extent to which they felt that their implementation of newly learned PRT strategies was effective. For both family quality of life and perceived self-efficacy, parents' similar ratings between time-points may reflect sufficient levels of personal and ASD-related support and services received prior to participating in this study. An alternative explanation is that the measures were insufficiently sensitive to short-term change in family quality of life and perceived self-efficacy.

With respect to parent satisfaction with the treatment, all respondents rated qualities related to the therapist, the intervention, the coaching, and their overall experience as helpful to extremely helpful, except one parent who rated the intervention as somewhat helpful. In regard to the acceptability and feasibility of PRIMeR, all parents either agreed or strongly agreed with aspects related to the intervention a) being valuable to their family and being a positive and helpful experience for their child, and b) comprising strategies that they will continue to use and would recommend to others.

Importance of the PRIMeR Module

It is encouraging to note that social communication behaviors can be boosted in children with these levels of impairment even later in the preschool period. With respect to participants' pre-treatment characteristics, the preschoolers enrolled in the current study were minimally verbal, i.e., they had fewer than 10 words that they used spontaneously and functionally. Tager-Flusberg and Kasari (2013) referred to this group of children with ASD as “the neglected end of the spectrum”. Despite an estimated 30% of children with ASD who will remain minimally verbal (Pickles, Anderson, & Lord, 2014), relatively little is known about interventions that may be effective for this subset of the population (Brignell et al., 2018). Such sparse information is due, in part, to a lack of clear defining pattern of skills or deficits that characterizes this highly variable group (Bal, Katz, Bishop, & Krasileva, 2016). For example, minimally verbal children vary with respect to nonverbal IQ scores (Munson et al., 2008) as well as receptive language abilities (Rapin, Dunn, Allen, Stevens, & Fein, 2009). Child participants in the current study were chronologically older than those who have participated in studies of other interventions that utilize responsiveness strategies such as the Social ABCs (up to 3 years of age; Brian et al., 2017) and ESDM (up to 3.25 years of age; Contaldo et al., 2020). The present participants were also older than those included in the aforementioned RIT study with low-functioning children (up to 3.75 years of age; Ingersoll & Schreibman, 2006). However, the preschoolers in the current study were functionally similar to these at-risk toddlers, i.e., at similar early developmental levels.

Implications of the Current Research

Lack of appropriate services are especially devastating for children who are most severely affected, and thus most likely to fall further behind and to develop cumulative behavior problems. Importantly, these children are already at highest risk for poorer outcomes in PRT programs (Fossum et al., 2018). Acquisition of some spoken language by age 5 is important, as it is less likely that a child with ASD will acquire significant linguistic skills after this point (Tager-Flusberg, Paul, & Lord, 2005). The PRIMeR intervention is predicated on the idea that the greater the improvement of specific pre-verbal skills prior to PRT entry, the greater the expressive language gains that children can make during treatment. Development of these pre-verbal skills also has implications for children's social skills development with same aged-peers, as social-communication difficulties and lack of play skills limit a child's opportunity to participate in preschool classroom activities. Moreover, individuals with ASD typically experience poor social integration (Magiati, Tay, & Howlin, 2014); adolescents suffer from loneliness and lack of companionship (Locke, Ishijima, Kasari, & London, 2010); and almost half of affected individuals will have poor outcomes in adulthood (Steinhausen, Jensen, & Lauritsen, 2016) thereby highlighting the importance of early, effective intervention. Social communication and play skills represent pivotal domains that can have positive cascading effects on other areas of learning and adaptive functioning for children with ASD (Doctoroff, Greer, & Arnold, 2006; Kasari et al., 2005).

Generally, my findings pertaining to shared positive affect provide additional support for including parents in the treatment of their children via education and coaching in treatment strategies they can use within the family's natural environments. Disrupted

parent-child relationships are likely to have a cascade of effects on children's social and communicative development (Sameroff, 2009). The current intervention yielded increases in shared positive affect between parent and child, thereby enriching the parent-child relationship. Importantly, the PRIMeR module increased children's enjoyment in interaction with others. This inclination for social interaction should enhance the likelihood that a child will benefit from PRT-based intervention programs, as PRT is delivered in the context of interactions that are intended to be enjoyable and thus motivating for the child. The gains obtained in aspects of toy play also have potential implications for participation in PRT-based programs. Although short-term change was observed in the current study, whether these gains better position children and their families to make gains during PRT-based intervention remains an empirical question.

Active Ingredients/Mechanisms of Change

There is a consensus among researchers about the importance of identifying active ingredients within treatment programs. The current study sought to contribute to the growing literature focused on the empirical investigation of active treatment ingredients. Contingent imitation used as a strategy in the current study within the toy play context may explain observed gains in children's social-communicative outcomes. Contingent imitation is a treatment strategy represented in the ASD literature (e.g., Ingersoll & Dvortcsak, 2010; Ingersoll & Schreibman, 2006; Kasari et al., 2006). Research suggests that this responsive parental behavior is associated with increased social gaze and social vocalizations among minimally verbal children when implemented by researchers and mothers (Field, 2017; Sanefuji & Ohgami, 2011). Moreover, research shows that mothers' use of contingent imitation with their children who have ASD is

linked to increased responsiveness during play interactions (Dawson & Galpert, 1990). Its potential unique contribution to treatment response has not been empirically investigated. However, Ingersoll and Schreibman (2006), as well as Gulsrud and colleagues (2016), reported specific effects of contingent imitation and concluded that this strategy may be an active ingredient for improving social engagement outcomes. Studies that test the relation between isolated core components of NDBIs and primary outcomes, i.e., mediation, are needed to better understand how an intervention is affecting change.

Potential Interaction of Intervention Components/Rationale for Intervention Package

Together, targeting a) toy play via RIT and b) shared positive affect via the implementation of responsiveness strategies shifted these preschoolers' low responder profiles in a favorable direction. It is unknown, however, what extent each intervention component may have contributed to the observed intervention effects on each dependent variable. In other words, RIT could have facilitated increased shared positive affect in children and responsiveness training could have influenced toy play outcomes.

Reciprocal imitation training strategies inherently involve social reciprocity between two interaction partners when imitation and "turn-taking" are taking place (Coogle et al., 2013). Strategies such as imitation of a child's actions, vocalizations, and affect, as well as reinforcement of a child's imitative act, may have generally boosted social engagement and communication (Jung & Sainato, 2013; Killmeyer, Kaczmarek, Kostewicz, & Yelich, 2019) and thus facilitated effectiveness of responsiveness strategies implemented during the other 30-minute session taking place that day, targeting shared

positive affect. To mitigate possible order effects, the order of the majority of sessions was counterbalanced each week. Complementing these transactional processes, the intervention targeting shared positive affect could have produced collateral benefits such as progress in toy play skills. Learning to enjoy and derive reward value from social interactions (as in responsiveness training) may partially explain children's boost in toy play skills during RIT sessions—especially since imitation (the modality used to foster toy play development) has a social component.

Complementary strategies (i.e., in RIT and responsiveness training) aimed at both toy play and shared positive affect may have led jointly to the effects seen in the present study, thereby supporting the PRIMeR intervention's efficacy when used as a package. Within the NDBI framework, improvement in targeted developmental areas is theoretically contingent on establishing shared attention to objects and people. Building communication skills and diversifying object interactions hinges first on establishing reciprocal engagement (Schreibman et al., 2015), which could have been accomplished in the current study by the reciprocal nature of both RIT and parent responsiveness training. However, future research is needed to determine the contributions of each component to changes in the dependent variables.

Limitations of the Current Study

Measurement bias. As the researcher-therapist, I served as the implementer of the intervention as well as the primary coder of the outcome variables, which may have introduced measurement bias. This bias was mitigated by acceptable inter-observer agreement with a second coder, blind to study phase. A second limitation involves data collection from brief snapshots of children's and parents' behavior; data were collected

from 10-minute video-recorded behavioral samples of children with the interventionist, and children with parents. Time sampling runs the risk of under-estimating or over-estimating a child's or parent's skills. However, I attempted to mitigate this limitation by obtaining samples of behavior during every intervention session over the 12-week period.

Methodological limitations. Features of the current study design limit the conclusions that can be drawn in relation to intervention effects for functional toy play variables. Differences between the baseline and intervention phases weaken confidence in the observed associations between intervention onset and children's subsequent behavior. These differences relate to a) toys available in each child's environment at time of video-recorded behavior samples, and b) context and interaction partner during measurement of this toy play behavior. During the baseline phase, children's play skills were measured using children's own toys found within their natural environments, whereas during the intervention phase, toy play skills were measured using a standard set of toys. Moreover, during baseline, children's toy play behavior was measured in the context of a typical play routine with their parents, whereas play during the intervention phase was measured in the context of RIT sessions with the researcher-therapist. Such differences could have resulted in Type I or Type II errors with respect to intervention effects. In the current study, measurement consistency between phases was captured by the play assessment, pre- and post-intervention, that included administration of the PEAR (by the researcher-therapist at the local children's hospital). In future studies, children's baseline and intervention performance should be measured using consistent setting characteristics across phases.

Generalizability to other interaction partners and settings. It is always important to consider the degree to which specific skills taught in one context will generalize to other contexts. Shared positive affect was measured here in the context of parent-child interactions. This outcome may be context-bound to interactions with the child's parent and if so, children's gains would not generalize to other social partners, e.g., other family members, teachers, and peers who do not interact with them using the responsiveness strategies that parents learned. It is also unknown whether these children's gains in toy play would generalize to other settings, such as daycares or preschools, and other social partners. However, the PRIMeR module was designed to promote generalization by teaching skills in natural, everyday routines and including parents. Further, toy play skills were shown to generalize, in the current study, from the RIT teaching context to the context of a semi-structured play protocol (using the PEAR) in a novel setting (the Autism Research Centre at the local children's hospital) during children's post-intervention assessment.

Strengths of the Current Study

Study design. Using a SCED allowed for close examination of effects in a few well-characterized children as a first step toward establishing efficacy of the PRIMeR intervention. The SCED is a rigorous scientific methodology often employed in applied psychology to establish evidence-based practices by documenting functional relations between independent and dependent variables. Typically, they are used in the development and evaluation of interventions designed to alter a specific human behavior (Kazdin, 2011) and thus was appropriate to address my present research questions.

Approach to measuring behavior. One strength of this dissertation was the micro-analytic approach used to measure children's and their parents' shared positive affect during dyadic play. Some research approaches involve assessing global parent behaviors during a parent-child interaction by using a parent or clinician rating scale for broad categories (as in Wan et al., 2012), or by assessing parental cognitions about interactions (e.g., Ohr, Vidair, Gunlicks-Stoessel, Grove, & Lima, 2010). I did not rely on parent-report or subjective measures of children's skills; intervention targets were directly measured from children's behavior thereby providing objective evidence of behavior change. It is hoped that this approach limited rater bias/interpretation while capturing elements of an interaction that may have been lost by more global judgements. This measurement approach was especially important as judgements about quality of social behavior may be more difficult when children are minimally verbal and their behaviour is constrained by intellectual and adaptive functioning deficits.

Another strength of the measurement approach I used was recording duration of shared positive affect from video-recorded samples as opposed to documenting the frequency of such behavior (i.e., presence/absence per interval of an interaction session; e.g., Cyr, Pasalich, & McMahon, 2014). Instead I assessed interactions by capturing the duration of behaviors, which I believe provided a more accurate representation of the proportion of time spent engaged in targeted behavior within a session. Additionally, I measured when parent and child synchronously shared positive affect, not only the extent to which children directed positive affect towards their parents and vice versa. Synchronous sharing is important to capture since parent-child interactions are dynamic and each member contributes to the flow of the interaction. Conceptually, we measured

what we sought to improve—that is, the extent to which parent and child continuously responded to each other’s bids/responses for social communication through enjoyment directed towards the other.

Lastly, with respect to toy play, I extended measurement of functional / appropriate toy play (as part of the low responder profile; Fossum et al., 2018) to include other important aspects of toy play. Measuring variety of functional play actions and variety of functional play objects—and examining an intervention effect for these dependent variables—allowed me to contribute to the ASD play literature as per recommendations to include measures of play diversity in research (Barton et al., 2019).

Participation of fathers. In this dissertation, three of four fathers were included, two of whom were heavily involved and participated in parent coaching. Fathers of children with ASD are under-represented in research (Bogossian et al., 2019; Flippin & Crais, 2011; Potter, 2017), despite increasing interest in the father-child relationship within research and findings that show a positive association between greater paternal involvement in early intervention and more positive outcomes (Lundahl, Tollefson, Risser, & Lovejoy, 2008; Panter-Brick, et al., 2014). Mothers and fathers play unique important roles in their children’s development (Rankin, Paisley, Tomeny, & Eldred, 2019). For example, whereas mother-child play tends to be more verbal and didactic, fathers tend to be more physical in their play, incorporating such actions/games as tickling, wrestling, chasing, and throwing their children in the air (based on studies with typically developing children, e.g., Paquette & Dumont, 2013). Associations between fathers’ play and children’s social communication skills such as higher-level language skill and more complex play have been documented in the ASD literature (Flippin &

Watson, 2011, 2015). In a systematic review of parent-implemented communication and play intervention studies with children with ASD aged 2–5 years (Flippin & Crais, 2011), only three of 27 studies reported fathers' involvement in parent training. Rankin and colleagues (2019) sought to understand through a systematic review how often fathers of children with ASD were targeted directly in any capacity, in the context of various evidence-based intervention efforts (e.g., psychological, speech, occupational, pharmacological; majority of reviewed studies were of children younger than 8 years). They concluded that fathers of children with ASD were not often included in such research. The authors suggested, although not based on empirical evidence reviewed, that fathers may be equally as effective as mothers in implementing intervention strategies and that their inclusion in treatment may improve the overall family system. Soliciting fathers' involvement in the current study was aligned with current recommendations to include fathers in intervention research (Rankin et al., 2019).

Directions for Future Research

Given the known heterogeneity of ASD, no single intervention approach or method will be sufficient or optimal for every child (Delmolino & Harris, 2012; Sherer & Schreibman, 2005; Stahmer, Schreibman, & Cunningham, 2011). This understanding has led to a wealth of research with overarching aims to better understand how to tailor children's treatment to improve treatment uptake and outcome.

The current study's findings demonstrate that children's profiles can be shifted by targeting and improving particular characteristics predictive of poor progress in PRT. Hence these findings provide a rationale for ongoing inquiry into individualizing

children's treatment pathways. Demonstrating that children's profiles could be favorably changed was the first step toward studies of the potential to boost progress during PRT.

Having shown that PRIMeR is efficacious in improving skills in areas that predict greater ability to benefit from PRT, the next step towards individualizing treatment is to test whether children who receive the PRIMeR module do, in fact, benefit more from PRT-based intervention programs such as NS EIBI. It would be important to test the effectiveness of the PRIMeR module using an RCT to investigate differences in communication gains (targeted outcome of NS EIBI) between two groups of preschoolers with ASD: 1) those exhibiting the low responder profile receiving RIT and whose parent(s) receive parent-responsiveness coaching prior to NS EIBI, and 2) those exhibiting the low responder profile receiving NS EIBI alone. Ideally, this investigation would be accompanied by an assessment of the acceptability and feasibility of such an approach within the context of this community-implemented EIBI program (i.e., from the perspectives of NS EIBI staff as well as participating families). Using a combination of approaches is common within community-based interventions for children with ASD (Love, Carr, Almason, & Petursdottir, 2009; Stahmer, Collings, & Palinkas, 2005).

Clinical decisions about what interventions to incorporate into a child's treatment plan as well as the timing of the introduction of such components must be based on empirical evidence. My dissertation contributes to the empirical foundation on which such decisions about tailoring treatment to meet children's individual needs can be made. This line of research should also examine what level of (ongoing) support for children and consultation for parents, after intervention, will lend to maintenance of gains of varying periods of follow-up after this modular intervention is terminated.

Another area for possible future research includes exploration of PRIMeR's effects on dependent variables when PRIMeR's intervention components are implemented in isolation. PRIMeR was tailored to a behavioral profile of children predicted to make minimal gains during PRT. It is an open question, however, whether using PRIMeR's components independently will lead to increases in both target variables. Again, methodological limitations related to inconsistency in setting characteristics of RIT sessions between baseline and intervention phases would need to be addressed. Overall, an investigation of outcomes following delivery of PRIMeR's components is valuable when considering the relative efficiency of care pathways designed for identifiable groups of children. It is also unknown whether implementation of PRIMeR as it is currently packaged, or implementation of its independent components, would benefit children who are high on one dimension, e.g., functional toy play, but low on another, e.g., shared positive affect. Jobin (2020) highlighted that it may be beneficial to choose a treatment approach by learning domain for a particular child. Recall that she found that children responded uniquely to either PRT or DTT depending on what skill was targeted. There is precedence for the refinement of behavioral profiles through examining children's progress during intervention (SCED designs) by matching children with low responder profiles on all but one of the profile behaviors and assessing their response to different interventions (e.g., PRT and DTT; Schreibman et al., 2009). Tailoring treatment to meet unique and individual needs of children by integrating multiple treatment methods within a comprehensive program is hypothesized to increase treatment effectiveness for some preschoolers. However, research is needed to determine intervention factor(s) underlying PRIMeR's efficacy and to examine whether the low

responder profile (Fossum et al., 2018) can be further refined as it relates to gains made during the PRIMeR intervention.

Conclusion

This dissertation builds on previous work (Fossum et al., 2018; Schreibman et al., 2009; Sherer & Schreibman, 2005) by using knowledge of an empirically established behavioral profile predictive of less progress during PRT-based treatment. This knowledge informed the design and delivery of an intervention for children with characteristics predictive of less progress, in an effort to boost development in those specific domains. This study demonstrated the preliminary efficacy of this package of developmental treatment strategies in targeting areas of functioning that are potentially important for enhancing response to PRT.

Publicly funded, community-based EIBI programs represent a significant investment, and provide a key opportunity for children to make important, long-lasting gains. Such intervention programs' effectiveness may be enhanced by determining whether measurable, pre-treatment characteristics can be used to predict treatment uptake, and whether characteristics predictive of low response are modifiable through specific treatment pathways. By demonstrating PRIMeR's preliminary efficacy, this research helps to indicate a potential route by which more children can benefit from PRT programs.

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Appendix A

Play, Engagement, and Affect Ratings (PEAR) Form

Child: _____ Completed by: _____ Date: _____ Time: _____ Location: _____

- The assessment should take place in an environment where there are few distractions and at a time when the child is at his/her best (e.g., alert, feeling well)
- Materials should be in place on the floor before recording begins. Prevent access to other toys (e.g., block access; remove other toys if at all feasible).
- **Present each set of toys for 5 minutes (may be longer in some instances, to allow sufficient time to present 5 models, see below).** There should be at least a short break (e.g., 1-2 minutes) between sets. Longer breaks may be appropriate for some children.

Materials: PAIRS of the following:

- SET 1: Baby with clothes on; Blanket, folded in four; Play food (2 pieces on plate), Plate, Fork, Cup; Small Grip n' Roll ball; 8-piece inset puzzle, assembled
- SET 2: Little People bus (no batteries); 2 Little People in bus; Gas pump; Screw-driver; 4 nesting cups, nested and upright; 4 beads on cord

The goal is to engage the child in toy play with you.

- Sit with the child on the floor, within easy reach of toys. Say *"It's time to play with the toys!"*. Encourage the child to select a toy. This can include handing the child a toy or placing a toy in front of the child.
- For each toy that the child selects, allow a brief period of time for spontaneous play before presenting models. Join in with the child's play by playing with the paired item that is the same as the child's (imitate the child, make comments).
- Model 5 play actions that the child has not yet demonstrated during the 5-minute set. It may be helpful to use check-boxes to keep track of the number of models you have given. *If 5 minutes elapse and you have not yet presented 5 models, continue taping.*
 - Models should involve the toy that the child is holding/has in front of them.
 - Model only functional play acts (vs. dropping toys, spinning bus wheel, etc). See "PEAR functional play examples"
 - While modeling, say "Let's _____ (label of action you are doing)"
 - Model each action twice (action + "Let's"), unless the child imitates the first model. Prior to the second model, hand the child any relevant object(s) they are not/no longer holding (e.g., if child put down object after first model; if child is holding one of the two objects needed for play act). If the child does not take an object offered, place it in front of them.
 - A range of actions can be modeled. Make decisions based on the child's interest and play level.

- Examples for a child with fewer play skills who is playing with a nesting cup:
 - “*Let’s drink*” (pretend to drink); “*Let’s make music*” (*bang cup*); “*Let’s build*” (put two cups on top of each other); “*Let’s put in*” (put bead in cup); “*Let’s take out*” (unstuck cups)
 - Examples for a child with more advanced play skills who is playing with the beads and then the baby:
 - “*Let’s put a bead on*” (thread bead); “*Let’s be snakes!*” (pretend thread is a snake); “*Let’s feed the baby*” (give food to baby); “*Let’s put the baby to bed*” (lie baby down); “*let’s wake up!*” (sit baby up)
- Once or twice per 5-minute block, draw the child’s attention to other toys in the play set. If the child perseverates with a toy (e.g., very repetitive actions with toy, focus on toy appears to negatively affect engagement with adult), attempt to draw the child’s attention away. If the child continues to perseverate, remove the toy. Do not present models while a child is actively engaged in perseverative play.
- Respond to the child’s initiations (e.g., answer questions, join in with the child’s play ideas). Do not try to explicitly elicit language from the child, but rather use language as you naturally would (i.e., no model prompts or time delays, no focus on contingent responding to questions).
- *Coding Key (see below)*: occurrence at any point in interval (+); non-occurrence (-).

	Functional Play		Functional Play
:00-:15		:00-:15	
:15-:30		:15-:30	
:30-:45		:30-:45	
:45-1:00		:45-1:00	
1:00-1:15		1:00-1:15	
1:15-1:30		1:15-1:30	
1:30-1:45		1:30-1:45	
1:45-2:00		1:45-2:00	
2:00-2:15		2:00-2:15	
2:15-2:30		2:15-2:30	
2:30-2:45		2:30-2:45	
2:45-3:00		2:45-3:00	
3:00-3:15		3:00-3:15	
3:15-3:30		3:15-3:30	
3:30-3:45		3:30-3:45	
3:45-4:00		3:45-4:00	
4:00-4:15		4:00-4:15	
4:15-4:30		4:15-4:30	
4:30-4:45		4:30-4:45	
4:45-5:00		4:45-5:00	
	/20		/20
	%		%

Functional/Appropriate play – During the interval, the child is engaged in at least one functional play act. See “PEAR functional play examples” for a definition and examples of functional play acts.

Set 1

Set 2

Response to play models (demonstration + “Let’s”)

Action	1	2	3	4	5	6	7	8	9	10
---------------	----------	----------	----------	----------	----------	----------	----------	----------	----------	-----------

Engages in play

similar to model after

first model (✓ or X)

Engages in play

similar to model after

second model (✓ or X)

Appendix B

Child Affect* Coding form (for eligibility screening)

Child Code: Tape Code: Observer: Date Coded:

Sum of interval scores (i.e., interval 1 + interval 2 + ... + interval 10): _____

Average of interval scores (sum of interval scores/total number of scorable intervals): _____

Operational Definitions:

Highly negative affect (1): Child does not appear to be enjoying him/herself. There are clear signs of distress, anger, fear, sadness or frustration.

Mildly negative affect (2): No clear signs of negative affect, but some indication of irritation, impatience, boredom, apprehension. An impression that “he or she would rather be elsewhere.”

Neutral (3): Child does not display overall signs of positive or negative affect, displays an overall neutral aura.

Mildly positive (4): No clear “full-blown” joy, but the mood is nevertheless pleasant.

Highly positive (5): Child enjoys him/herself—may smile, laugh happily out loud, or jump with joy. Note: Must be jumping with the purpose of expressing joy, and not to display repetitive behavior, or to express discontent.

* Definition from Fossum (2014); adapted from Baker, Koegel & Koegel (1998); Brookman-Frazee (2004); Kochanska & Aksan, (1995).

Appendix C

CBQ temperament sub-scale descriptions*

Activity Level: Gross motor activity, including rate and extent of locomotion; e.g., “Seems always in a big hurry to get from one place to another.”

Anger and frustration: Negative affectivity related to interruption of ongoing tasks or goal blocking; e.g., “Has temper tantrums when s(he) doesn’t get what s(he) wants.”

Approach and anticipation: Amount of excitement and anticipation for expected pleasurable activities; e.g., “Gets so worked up before an exciting event that s(he) has trouble sitting still.”

Falling reactivity and soothability: Rate of recovery from peak distress, excitement, or general arousal; e.g., “Has a hard time settling down for a nap.”

Smiling and laughter: Positive affect in response to changes in stimulus intensity, rate, complexity, and incongruity; e.g., “Laughs a lot at jokes and silly happenings.”

High intensity pleasure: Pleasure or enjoyment related to situations involving high stimulus intensity, rate, complexity, novelty, and incongruity; e.g., “Likes going down high slides or other adventurous activities.”

Low intensity pleasure: Pleasure or enjoyment related to situations involving low stimulus intensity, rate, complexity, novelty, and incongruity; e.g., “Enjoys just being talked to.”

Inhibitory control: Capacity to plan and to suppress inappropriate approach responses under instructions or in novel or uncertain situations; e.g., “Can lower his/her voice when asked to do so.”

Shyness (versus Social Approach): Slow or inhibited (versus rapid) speed of approach and discomfort (versus comfort) in social situations; e.g., “Often prefers to watch rather than join other children playing.”

* Rothbart, Ahadi, Hershey, & Fisher (2001)

Appendix D

Toy Play Coding Form

Child Code: _____ Tape Code: _____ Observer: _____ Date Coded: _____

Interval (seconds)	Absent or present	Interval (seconds)	Absent or present
00:00 - 00:30		05:01 - 05:30	
00:31 - 01:00		05:31 - 06:00	
01:01 - 01:30		06:01 - 06:30	
01:31 - 02:00		06:31 - 07:00	
02:01 - 02:30		07:01 - 07:30	
02:31 - 03:00		07:31 - 08:00	
03:01 - 03:30		08:01 - 08:30	
03:31 - 04:00		08:31 - 09:00	
04:01 - 04:30		09:01 - 09:30	
04:31 - 05:00		09:31 - 10:00	

Operational Definition of Toy Contact:

Child interacts with a toy according to its function (e.g., roll train along the floor), or uses it to represent another object in play (e.g., toy banana as a phone).

The proportion of total intervals in which appropriate behavior occurred is calculated to capture frequency of functional toy play.

Coding will track the number of different (new) toys the child plays with, in addition to the number of unique actions performed using a toy.

* Definitions from Fossum (2014); adapted from Sherer & Schreibman (2005); Schreibman, Stahmer, Barlett, & Dufek (2009); Stahmer (1999).

Appendix E

Shared Positive Affect Coding

Operational definition of shared positive affect – considered to be present when child:

- | | | | | |
|-------------|-----|---------------------------|-------|------------------------------------|
| - Smiles | TO: | - Express happiness/joy | WHILE | - Making eye contact |
| - Laughs | | - Show interest | ALSO: | OR |
| - Jumps | | - Request continuation of | | - In response to an adult's action |
| - Gestures | | the dyadic interaction | | (reciprocity/intention to |
| - Vocalizes | | | | communicate, i.e., that they |
| | | | | would like the activity to |
| | | | | continue; must be clear) |
- And parent exhibits and directs positive affect towards his/her child, in this same fashion.

* Definition extended from Fossum (2014); adapted from Baker, Koegel & Koegel (1998); Brookman-Frazee (2004); Kochanska & Aksan, (1995).

Appendix F

FIDELITY OF IMPLEMENTATION

(Ingersoll & Lalonde, 2010)

Child: _____

Therapist: _____

Observer: _____

Date: _____

Session: _____

Contingent imitation: ___ Prompt: ___ Linguistic mapping: ___ Praise: ___ Model: ___

Avg. Fidelity: _____

RIT COMPONENT	LOW FIDELITY 1	2	3	4	HIGH FIDELITY 5
CONTINGENT IMITATION Imitate child's toy play, gestures, and vocalizations	Therapist does not imitate the child's toy play	Therapist imitates a few of the child's toy play, but misses the majority of opportunities	Therapist imitates the child's toy play up to 50% of the time, but misses many opportunities	Therapist imitates more than 50% of the child's toy play, but misses opportunities	Therapist imitates almost all of the child's toy play throughout the session
LINGUISTIC MAPPING Use simplified, repetitive language around child's attentional focus	Therapist does not use simplified language around the child's attentional focus; language is too complex, or therapist does not use any language	Therapist uses simplified language around the child's attentional focus during some of the session, but misses the majority of opportunities or majority of language is too complex	Therapist uses simplified language around the child's attentional focus up to 50% of the time, but misses many opportunities	Therapist uses simplified language around the child's attentional focus for more than 50% of the session, but misses opportunities or language is not sufficiently simple, stressed, or repetitive	Therapist uses simplified language around the child's attentional focus throughout the session. Almost all of the therapist's language is in this form and important words are stressed and repeated

<p>MODEL</p> <p>Model actions around child's focus of interest</p>	<p>Therapist models actions that are inappropriate for child's level/interest or does not recruit child's attention. Very low rates of modeling also scored here</p>	<p>Therapist models some actions that are appropriate for child's level/interest but also many that are not or often fails to recruit the child's attention. Low rates of modeling also scored here</p>	<p>Therapist models some actions that are appropriate for child's level/interest and recruits child's attention some of the time</p>	<p>Therapist models actions that are appropriate for child's level/interest more than 50% of the time and recruits child's attention the majority of the time</p>	<p>Therapist models actions that are very appropriate for child's level/interest and recruits child's attention</p>
<p>PROMPT</p> <p>Physically prompt child to imitate after 3 presentations of action</p>	<p>Therapist does not physically prompt child to imitate action after presenting the action 3 times</p>	<p>Therapist prompts child to complete action after the third trial a minority of the time, but misses many opportunities, or prompting often does not result in imitation (e.g., child switches activities without imitating)</p>	<p>Therapist prompts child to complete action after third trial up to 50% of the time, but misses many opportunities or prompting does not result in imitation (e.g., child switches activities without imitating)</p>	<p>Therapist prompts child to complete action after third trial the majority of the time, but misses opportunities or prompting occasionally does not result in imitation (e.g., child switches activities without imitating)</p>	<p>Therapist consistently prompts child to complete action after third trial if child has not spontaneously imitated. Once therapist begins a trial, therapist follows through such that the trial ends in imitation</p>
<p>PRAISE</p> <p>Animatedly praise child's spontaneous or prompted imitation</p>	<p>Therapist does not praise child's spontaneous or prompted imitation or consistently praises incorrect responses</p>	<p>Therapist praises minority of the child's spontaneous and prompted imitations, but misses the majority of opportunities or praises multiple responses</p>	<p>Therapist praises some of the child's spontaneous and prompted imitations, but misses many opportunities or praises incorrect responses</p>	<p>Therapist praises the majority of the child's spontaneous and prompted imitations, but misses some opportunities or praise is provided for an incorrect response</p>	<p>Therapist praises all of the child's spontaneous imitation throughout the session. Praise is withheld for incorrect responding</p>

Appendix G

RIT protocol (based on Ingersoll, 2010)

The researcher-therapist began each session by engaging the child in free play with sets of identical play materials. Toys were chosen based on the child's interest, to keep the child's motivation high. During this play, the researcher-therapist imitated all of the child's actions with toys (contingent imitation) to encourage responsivity to being imitated (i.e., reciprocity). At the same time, the researcher-therapist provided a running commentary of the child's actions using simplified language (linguistic mapping) to enhance the correspondence between the child and the therapist's actions (i.e., reciprocity, and to provide appropriate language models). To teach imitation, the researcher-therapist modelled an action with an object (the duplicate toy to which the child was attending, if child was attending to any particular toy) once per minute, on average. Actions were modelled up to three times and paired with a verbal marker describing the action. For example, the researcher-therapist modelled rolling a car on the floor and said, "Drive the car". The verbal marker was kept consistent across the three presentations of the action within a trial but varied across trials (e.g., "Roll the car"; "Car goes, *Wee*") so that the word(s)/phrase did not become associated with a specific toy or action. When the child imitated, the researcher-therapist praised the child and allowed him or her to engage with toys as he or she preferred (contingent reinforcement) and returned to imitating the child (contingent imitation). If the child did not imitate the action within 10 seconds of the third model, the researcher-therapist physically prompted (i.e., guided, hand-over-hand) the child to complete the action and then provided praise and continued access to toys. Since the goal is for the child to attempt to imitate a play

partner's actions, rather than to accurately produce specific actions in response to a model, contingent reinforcement included providing praise for physically prompted acts, i.e., attempts at imitation. An action was considered an attempt when a child tried to perform the same action modelled by the researcher-therapist within 10 seconds, but did not complete it, identically, due to, e.g., a fine motor difficulty or accidental drop of the toy(s). Other social behaviors (e.g., joint attention, verbal imitation) were not prompted or systematically reinforced. As the treatment progressed, actions were modelled using toys with which the child was not already engaged to encourage more flexible responding.

Appendix H

Responsiveness training protocol

The current intervention, aimed at boosting shared positive affect, used responsiveness strategies common to NDBIs such as ESDM, Social ABCs, and PRT: the presentation of various activities and social routines in an attempt to a) elicit a smile, and b) learn what the child liked, enjoyed, or was interested in so that a repertoire of activities could be accumulated that would serve to enhance the child's motivation to respond to the adult, e.g., with a smile that shared/communicated emotion with the adult. The reader is referred to the ESDM manual for strategies used. It was important for the researcher-therapist to note when a child smiled and to join the child when they were smiling. Joining the child included, first, getting the child's attention (e.g., by looking at them/orienting them) and, secondly, smiling back at the child. The researcher followed the child's lead where possible, i.e., by focusing on and joining the child when they were engaged in a preferred activity/routine. Following a child's lead is an established social communication strategy used in early intervention (Coogle et al., 2013).

Another aspect of ESDM adopted for use in the current intervention included sensory social routines, where the focus was on the affective and relationship-based aspects of a dyadic social experience between two individuals. Sensory social routines are believed to have the capacity to increase the salience of social rewards while enhancing a child's social attention and motivation for social interaction (Dawson et al., 2010). Once the researcher-therapist learned which routines motivated the child to interact with the researcher-therapist, the researcher used these very pleasurable sensory social routines (i.e., joint activity routines) to facilitate the child's growth in shared

positive affect. Within these routines, it was ideal for each partner's attention to be focused on the other person, rather than on objects, and that mutual pleasure and engagement dominated the play (as prescribed by the researcher-therapist). Typical sensory social routines introduced to the child involved "lap games" like "Peekaboo", song routines with motions such as "Itsy Bitsy Spider", floor songs, e.g., "Ring Around the Rosy", and movement routines such as "Airplane" and "Chase." First and foremost, the researcher-therapist followed the child's lead in perpetuating/expanding interactions that the child appeared to enjoy in humor, anticipatory excitement, laughter, etc. Following a child's lead or introducing a child to highly motivating routines increased natural opportunities for the child and communication partner (i.e., researcher-therapist/parent) to interact in a way that was characterized by shared positive affect. Brian and colleagues (2016) have acknowledged, though, that interventions aimed at fostering the development of shared positive affect may need to focus, first, on increasing the frequency of smiling (in both adult and child). Therefore, in the beginning, fostering reward value sometimes necessitated pairing a social experience with a non-social reward (i.e., access to a preferred object). Enhancing the reward value of the social experience, in this way, was thought to lay the foundation for motivation for purely social experiences characterized by shared positive affect. The expectation, then, was that increased smiling would lead to increased rates of shared positive affect (e.g., reciprocal adult-child smiling with mutual gaze).

There were four main goals that the adult attempted to attain with social routines (Dawson et al., 2010): 1) draw the child's *attention to others'* social-communicative cues, especially eye contact and the face, but also to physical gestures, postures, anticipatory

movements, and facial expressions; 2) develop the child's *awareness of* facial expressions and their ability to share emotional expressions face-to-face with another (e.g., the adult shared smiles and made silly faces and sound effects during games, and drew the child's attention to their face); 3) increase the child's *communications* to initiate, respond to, and continue social interactions (through eye contact, facial expression, gestures, sounds, and words that conveyed excitement/positive emotion); and 4) optimize the child's arousal, state, and attention (e.g., a social routine that could enliven a passive, "tired" child and calm an overactive or over-aroused child).

In trying to modulate and optimize the child's affect, arousal, and attentional state, the researcher-therapist 1) displayed clear, genuine, and natural positive affect through, e.g., facial expressions and tone of voice (matched by child positive affect); 2) took turns with the child and engaged in a dyadic interaction characterized by reciprocity; 3) stayed attuned to each child's states, motives, and feelings and then responded sensitively, responsively, and empathically to each child's communicative cues (by mirroring their emotion and communicating an understanding of that emotion, encouraging activities suited to the child's evolving emotional state); and 4) effectively managed transitions. Managing transitions included scaffolding the child's shift of interest by closing down one activity and introducing others, while being sensitive to the child's attention and motivation with regards to the timing of the transition. The combination of these techniques was designed to engage the child in positive emotional experiences with the researcher-therapist and parent and to foster their motivation to continue to engage with a social partner.