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Maximizing the potential for infants at-risk for autism spectrum disorder through a parent-mediated verbal behavior intervention

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ABSTRACT
Over the past 10 years, very early detection of ASD has opened the way to establishing much-needed evidence-based interventions for infants under 18 months of age. A Behavior Skills Training package developed in a verbal behavior framework was the foundation for a parent-mediated service delivery model for 12–16-month infants presenting ASD symptoms. Training consisted of 12 weekly coaching sessions conducted with each parent-infant dyad in the family’s home with the aim to increase social, communication and play skills. A battery of assessment tools was used pre and post-training, while parent and infant target behaviors were scored through videos taken during weekly sessions. Results indicate that social communication behaviors increased in all infant-parent dyads, while severity and number of autism symptoms decreased. By beginning intervention during infancy, brain neuroplasticity is leveraged maximizing the child’s developmental trajectory. This is the first prospective study that provides a verbal behavior analytic framework to treat symptomatic infants.

Prospective research on Autism Spectrum Disorder (ASD) examines the developmental onset of symptoms by following high-risk infants from birth to diagnosis and marking the emergence of symptoms as they unfold during the first year of life. Pre-diagnostic markers have been found to emerge between 2 and 18 months of age; however, the reliability of these markers increases as the child approaches 12 months of age (Chawarska et al., 2014). One of the most common early markers is atypicality in social communication and eye gaze and decreased eye contact (Elsabbagh et al., 2015; Gangi et al., 2018; Osterling, Dawson, & Munson, 2002). Additional markers include variations in temperament, such as lower positive affect, joint attention and social smiling (Garon et al., 2016; Lambert-Brown et al., 2015; Macari, Koller, Campbell, & Chawarska, 2017; Nichols, Ibañez, Foss-feig, & Stone, 2014), reactive temperament (Clifford, Hudry, Elsabbagh, Charman, & Johnson, 2013), and decreased object exploration and anticipatory response (Landa, Haworth, & Nebel, 2016; Kaur, Srinivasan, & Bhat, 2015; Korterba, Leezenbaum, Iverson, 2014). One of the earliest observed indicators is atypical and delayed motor skills (Bhat, Galloway, & Landa, 2012; Estes et al., 2015; Flanagan, Landa, Bhat, & Bauman, 2012). However, the
emergence of these symptoms among infants may be highly variable with regards to the pattern, presentation, severity and age of onset (Rogers, 2009). Restrictive and repetitive behaviors have been less frequently observed in infants and more commonly observed in a later onset, being more likely to worsen as the child approaches 24 months of age (Kleinman et al., 2008; Wolff et al., 2014). Research in the emergence of ASD symptoms continues to improve the empirical validity of infant screening tools which can be predictive of a future diagnosis of ASD in early childhood. Early screening has become more commonplace in the US and Canada; this is essential as early screening leads to early detection, which allows for very early intervention.

Early Intensive Behavioral Intervention (EIBI) commencing prior to three years of age is significantly correlated with children reaching optimal treatment outcomes, measured by increases in IQ to same age neurotypical peers and significant decreases in core autism symptoms to a level where criteria for ASD are no longer met (Anderson, Liang, & Lord, 2014; Itzchak & Zachor, 2009; MacDonald, Parry-Cruwys, Dupere, & Adhern, 2014; Reichow & Wolery, 2009). Over the past five years, an increase in pre-diagnostic intervention, also known as pre-emptive intervention (prior to 18 months of age), has begun proactively with the aim to capitalize on the critical development period that occurs in the first two years of life. During this time, neuro-plasticity is at its peak, allowing for more rapid changes in development and ultimately more effective intervention (Bradshaw, Steiner, Gengoux, & Koegal, 2015). The importance of early detection and early intervention is predicated upon the knowledge of brain neuroplasticity during the first few years of life. From birth to 36 months, the human brain undergoes the most transformative development period in all post-natal life, during which neural circuitry and synaptic density go from simple and sparse connections to complex connections responsible for social behavior, communication and cognition (Courchesne & Pierce, 2005). In addition to exponential increases in neural connections during this time, the brain also begins refinement which is the strengthening of essential connections and the pruning of non-functional connections, enhanced through participation and exploration of an enriched environment (Pierce, Courchesne, & Bacon, 2016). Therefore, in order to capitalize on plasticity of the brain and to maximize the effects of intervention, it is essential to begin intervention while these connections are still being formed rather than trying to change maladaptive neural connections once they are established (Pierce et al., 2016).

This critical period of brain development is substantiated in a study by MacDonald et al. (2014) who examined the outcomes of children who began EIBI prior to the age of 24 months compared to after 30 months, finding that children who began intervention prior to two years were 60% more likely to make significant gains in their first year of intervention compared to those who began EIBI after 30 months of age, underscoring the notion that with regards to very early intervention, every month counts. Support for very early intervention is further established by a Randomized Control Trial (RCT) with a sample of 54 infants identified by 9 months of age and completing pre-diagnostic treatment by 15 months of age, with the latter proving successful at decreasing the overall severity of autism symptoms and altering the developmental trajectory by the time children reached 39 months of age (Green et al., 2017). These findings provide additional support to earlier literature (Dawson et al., 2012; Steiner, Gengoux, Klin, & Chawarska, 2013) that early intervention maximizes children’s potential and that early detection and intervention could mitigate and significantly decrease ASD symptoms prior to the full onset and
In the light of these recommendations and as best practices for pre-emptive interventions are being established, there is a logical tendency to use a parent-mediated service delivery model given the young age of the child (Brian, Smith, Zwaigenbaum, Roberts, & Bryson, 2015; Green et al., 2017; Rogers et al., 2014). In addition to being cost-effective and developmentally appropriate, parent-mediated intervention can also vastly improve relevant parental skills that are key to mitigating rather than amplifying the severity of ASD symptoms (Bakermans-Kranenburg, van Ijzendoorn, & Juffer, 2003). As a response to atypical neuro and social communication development in infancy, the quality of parent-infant interactions can develop along an impaired trajectory, creating an atypical interaction cycle that could maintain or intensify the severity of the symptoms in the child (Green et al., 2015). By incorporating the child’s parents into the intervention, the quality of the dyadic interaction cycle improves, thereby maximizing the child’s potential by removing barriers towards typical development (Wan et al., 2013). Many researchers have been quick to adapt this pragmatic trend of using parent-mediated intervention with at-risk infants or young toddlers with a confirmed diagnosis, effectively targeting behaviors such as joint attention, responsivity to parent and referencing faces (Green et al., 2015; Kasari, Gulsrud, Wong, Kwon, & Locke, 2010; Wetherby et al., 2014) and positive changes in parent responsiveness (Brian et al., 2015; Green et al., 2015). A pilot study known as “Infant Start” (Rogers et al., 2014) examined the effectiveness of a parent-coaching intervention model with at-risk infants between 7 and 15 months of age and effectively reduced autism symptoms in 5 out of 7 infants by 36 months of age. The Social ABCs, a manualized treatment package (Brian et al., 2015), effectively increased social smiling and social orienting along with receptive and expressive language through a 12-week parent-coaching model with at-risk and confirmed infants and toddlers with ASD. Similarly, the Intervention in the British Autism Study of Infant Siblings-Video Interaction for Promoting Positive Parenting (IBASIS-VIPP, Green et al., 2015) was also successful in increasing social communication behaviors while reducing the severity of prodromal autism symptoms in at-risk infants between 9 and 14 months of age in another 12-session parent-mediated intervention. What these early studies have consistently shown is that early developmental behavioral interventions delivered by parents in a natural setting produce significant positive outcomes that have the potential to alter an infants’ developmental trajectory. These and other notable parent-mediated interventions are demonstrating promising effects; however, the publications offer limited details regarding the specifics of how to select, sequence and teach early social communication skills to infants, therefore limiting the access of researchers, professionals and parents to the underlying processes.

In the present study, weekly training topics were developed to target social-communicative deficits which were previously established throughout early signs literature (Zwaigenbaum et al., 2015) along with typical developmental milestones established within the Verbal Behavior Milestones Assessment and Placement Program (VB-MAPP; Sundberg, 2008). Social communication and interaction such as attending to familiar voices (Decasper & Fifer, 1980), orienting to faces (Haith, Bergman, & Moore, 1977) and face-like stimuli (Johnson, 2005) and eye gaze (Grice et al., 2005) begin to emerge in the first weeks of life (Farroni, Csibra, Simion & Johnson, 2002). By 4 weeks of age, infants often initiate eye-contact during nursing
Zeifman, Delaney & Blass, 1996) and by 9 weeks infants direct eye gaze toward the adult when they are speaking (Haith, Bergman & Moore, 1977). Early eye gaze is foundational to facial recognition and understanding early communicative gestures. For infants at-risk of ASD, eye gaze and preferential attention to faces begin to decline between 2 and 6 months of age (Jones & Klin, 2013); therefore, increasing eye gaze and orienting towards familiar faces was the first topic trained in the 12-week program. Once a child’s preferences were established, eye gaze directed at an adult was conditioned as an early mand for continuation of access to the preferred item. Parents were taught to conduct informal preference assessments with their child and upon observing their child’s interest and engagement with an item they would withhold the item and wait for eye-contact from the child and then immediately deliver the preferred item. Eye contact was chosen as the first manding topography to be taught as participants had the necessary pre-requisite skills, it required a low-response effort, it has been described as a behavioral cusp for all other social-communicative behaviors, and it is a socially significant behaviour. Additionally, it is common for infants at risk to display low frequency or unreliable babbling or vocal production as well as demonstrate deficits in motor skills and coordination, therefore teaching eye contact over a vocal or motor response was more likely to lead to the participants contacting greater reinforcement thereby allowing both parents and infants to experience early success in the program.

Manding using gestures such as pointing or signing and echoic to mand transfers were used in subsequent sessions. Typically developing infants will often use a variety of gestures to communicate by 12 months of age and continue to use and respond to gestures prior to the onset of vocal language, whereas children who are later diagnosed with autism use and respond to less gestures during this developmental period (Wetherby & Prizant, 2002). Since gestures are a motor behavior which can be physically prompted whereas vocal responses cannot be physically prompted, manding with gestures was selected as the second requesting topography after eye contact. Through the use of antecedent manipulations (i.e., setting up the environment) and shaping procedures, parents taught their children to request an item through the use of an open-hand reach which was shaped to a single-finger point in subsequent sessions. Further topic development focused on following simple directions, completing an action such as clapping and selecting a preferred object from a small field of items. Additional topics focused on using well-established Naturalistic Environmental Teaching (NET) procedures such as the interspersal of verbal operants throughout a child’s preferred play activity. The progression of topic development is shown in Table 1, aiming to help clinicians develop and sequence topics in order to best support early learning development. The present 12 topic curriculum will be referred to as the Sequential Parent Curriculum (SPARC) (See Table 1).

The present study examines a pre-emptive intervention with an early onset phenotype of high-risk infants and adds to the existing literature by providing a behavior analytic framework with an emphasis on verbal behavior which clinicians can readily apply to increase appropriate social communication and play behaviors in symptomatic infants and toddlers. It should be noted that the goal of pre-emptive intervention is not the
prevention of an ASD diagnosis but rather to alter the developmental trajectory and maximize the potential of each child.

Table 1. Sequential parent curriculum intervention topics.

<table>
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<tr>
<th>Session</th>
<th>Topics</th>
<th>Activities</th>
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| 1–2 Baseline | Unstructured play with primary caregiver in home and clinic | (a) Free operant preference assessment  
  a. Find 5 new activities or toys that make your child smile; 
  anticipation games (e.g., peek-a-boo, tickles, horsey rides) 
  or tangibles (e.g., bubbles, spinning tops, pop-up toys)  
  b. Create a list of all preferred activities that child seems to enjoy (old and new)  
  (b) Deliver reinforcing item or activity 2–3 times; once child is engaged, pause activity and wait for eye contact for continuation of activity |
| 3 Pairing and manding using eye contact for continuation of preferred activity | (a) Parent following child’s lead and move with child  
  (b) Optimal body position (at eye level with child, facing child)  
  (c) Narrating play with single words and sounds  
  (b) Placing preferred items in-sight but out of reach  
  (c) Manding with eye-contact to open/retrieve item and for continuation |
| 4 Parent tacting what child is playing and/or engaging with | (a) Controlling materials  
  (b) Placing preferred items in-sight but out of reach  
  (c) Manding with eye-contact to open/retrieve item and for continuation |
| 5 Antecedent interventions and motivating Operations | (a) Holding up preferred item and non-preferred item  
  (b) Child scans items and reaches for preferred item  
  (c) Fading items farther back to shape reach into open-handed point  
  (d) Placing item slightly out of reach and fading to open-hand reach or pointing to request item |
| 5 Making a choice between two items | (a) Controlling materials  
  (b) Placing preferred items in-sight but out of reach  
  (c) Manding with eye-contact to open/retrieve item and for continuation |
| 6 Manding for help using gestures | (a) Controlling materials  
  (b) Placing preferred items in-sight but out of reach  
  (c) Manding with eye-contact to open/retrieve item and for continuation |
| 7 Motor Imitation and Echoics | (a) Set up pairs of various objects  
  (b) Provide continuous opportunities for motor and vocal imitation across playsets by modelingmotor-imitation with 1-syllable vocal sound  
  (c) Say/Do 3 times and allow time for child to imitate  
  (d) If imitation is not emerging use prompting and prompt fading for motor imitation |
| 8 Listener Responding | (a) Practice 3–5 age-appropriate instructions with gestures such as give me, put in, turn page (book), roll ball |
| 9 Echoic to Mand Transfer or Motor Imitation to Mand Transfer | (a) Using any vocal approximations to request preferred item or baby sign language to request preferred items |
| 10 Interspersing learning opportunities across various activities | (a) Providing opportunities to request with eye contact, gestures, vocalizations  
  (b) Providing opportunities for imitation  
  (c) Providing opportunities for listener responding |
| 11 Individualized Session | Individualized session specific to parent and child’s needs |
| 12 Generalizing skills from play sessions to daily routines | (a) Providing learning opportunities across:  
  • Diaper changes  
  • Dressing  
  • Bath-time  
  • Feeding  
  • Bed Time |
| 13–14 1-month follow-Up | (a) Administration of Screening tools  
  (b) Maintenance and booster Training |
| 15 3-month follow-Up | (a) Maintenance and booster Training  
  (b) Program debrief  
  (c) Recommendations and referrals |
The first aim of the present study was to assess the effectiveness of a 12-week parent-mediated pre-diagnostic intervention program to reduce autism symptoms with an early onset phenotype cohort of high-risk infants. The second aim of the study was to provide a comprehensive procedural overview of the 12-week behavior analytic intervention framework, which clinicians can readily apply to increase appropriate social communication and play behaviors in symptomatic infants and toddlers. Treatment fidelity and social validity were also assessed.

**Method**

**Experimental design**

The present study used a multiple baseline design across five parent-infant dyads. The study consisted of extended baseline sessions that are consistent with a concurrent multiple baseline design, 12 weekly 1-hour sessions followed by a 1 and 3-month follow-up session.

**Participants**

Participants were between 12 and 16 months at the beginning of intervention (note that pseudonyms Sara, Ben, Leah, Ray and Ann are used). The infant sample consisted of three girls and two boys and the primary parent participant of each dyad was the child’s biological mother. A small selection of sessions included more than one primary caregiver, this being the other parent, nanny or grandparent, with the exception of Sara, whose nanny attended more than 80% of sessions in addition to her mother who attended 100% of sessions. Three of the five children (Sara, Ben & Ann) did not have siblings and are considered to be an “only-child”, while Ray and Ann were “high-risk siblings” in that they had older siblings who had a confirmed ASD diagnosis.

Three of the five participants were referred to the study through a developmental pediatrician specialized in pediatric ASD diagnosis, who had documented concerns with early manifestations of ASD symptoms in a 1-hour informal screening session. The remaining two participants were referred to the study through “Infant Development Consultants” who were assigned to them as they were considered a high-risk infant sibling.

Eligibility requirements included: (1) 1-hour informal screening with a developmental pediatrician; (2) “at risk” scores on the Infant-Toddler Checklist (ITC; Wetherby & Prizant, 2002); (3) “at-risk scores” on the Modified Checklist for Autism in Toddlers (M-CHAT-R; Robins, Fein, & Barton, 2009); (4) an elevated total score above 7 on the Autism Observation Scale for Infants (AOSI; Bryson et al., 2008); (5) a 3-month age-equivalent delay on the Mullen Scales of Early Learning (MSEL; Mullen, 1995); (6) 3-month age-equivalent delay on the Mullen Scales of Early Learning (MSEL; Mullen, 1995); (6)

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primary caregiver attending the intervention training is with the infant for at least 50% of
the infant’s waking hours; and (7) participant is 18 months or younger at initial intake;
and (8) not currently involved in any other early intervention treatments as reported by
the parents.

**Intervention**

All training sessions were conducted by a Board Certified Behavior Analyst (first author)
with a decade of clinical experience working with children and families in early intervention
programs who was also an active doctoral student studying this topic, all completed under
the supervision of a Board Certified Behavior Analyst-Doctoral (second author).

The intervention consisted of twelve 1-hour parent-coaching sessions, scheduled
weekly across 12 weeks. Each intervention session was approximately 60 minutes in
length and followed a Behavior Skills Training (BST) protocol for teaching, incorporating
instruction, modeling, rehearsal and feedback into each session. Every session allowed
time for parental questions and concerns, followed by recording a video of the previous
week’s topic, then an introduction to the new topic, modeling the new topic, a chance for
the parents to practice the new topic and receive performance feedback and coaching and
the session would finish with setting specific topic goals for the week. It was recom-
mended that parents attempt two 20-minute play sessions per day throughout the week
to focus on the topic for the week; however, fidelity of implementation was only collected
during weekly coaching session. Each child followed the same sequence of topics which
targeted social skills, communication and play across all sessions. Topics were organized
using Skinner’s analysis of verbal behavior (Skinner, 1957) targeting verbal operants such
as mands, tacts and echoics, as well as imitation and play skills (See Table 1 for sequential
framework of topics). Furthermore, the 12-sessions were play-based while maintaining
foundations in the science of Applied Behavior Analysis (ABA) with a focus on general-
ization techniques, such as the use of naturally occurring reinforcing contingencies,
training sufficient exemplars, using indiscriminable contingencies, programming com-
mon stimuli and mediated generalization (Stokes & Baer, 1977). At the start of every
intervention session, 5-minute videos were taken; follow-up sessions were conducted 1
and 3 months after the completion of the intervention to evaluate maintenance of
treatment gains, with post-testing instruments being administered at the 1-month fol-
low-up and a parent satisfaction questionnaire administered at 3-month follow-up. No
other known autism-specific intervention services were sought throughout the duration
of the 12-sessions and participant attrition was null with every dyad completing all
sessions.

**Data collection**

Three primary measures and four secondary measures were collected over the course of
the study to evaluate the reduction of autism symptoms and the acquisition of alternative
behaviors. Primary measures included three target behaviors across the parent/infant
dyad which were scored at baseline and from the weekly intervention sessions: one
parental target behavior and two infant target behaviors. The parental target behavior
(1) recorded the number of learning opportunities the parent presented towards their
child during a play session, defined as providing a discriminate stimulus for infant to respond. Positive exemplars included but are not limited to providing an opportunity for the child to mand for an item or activity, providing an opportunity for the child to imitate a motor or vocal response, and providing a clear instruction for the child to respond to. The infant target behaviors included (2) the frequency of eye contact directed towards the parent participant in the room and (3) the frequency of responding to learning opportunity presented by the parent, which can include but is not limited to establishing eye contact to request continuation, making eye-contact with adult in response to adult vocalizations, taking items when offered, following instructions, and imitation of motor or vocal behavior. Non-exemplars include defective operants such as pushing or swiping offered item away, biting, hitting, kicking or slapping the adult or eloping. Primary outcomes were scored from 5-minute session videos using partial interval recording (PIR) with 10-s intervals. Instructions for baseline videos were to “play with your child” while developmentally appropriate toys were provided in addition to the child’s own toys.

Effect sizes

Hedges g nonparametric effect size estimators were computed with 95% confidence intervals for children responsivity, children eye contact and parent behaviour. The SPSS macro DHPS was used for calculating effect sizes (Marso & Shadish, 2015). Interpretations of effect sizes mirror those employed by Taylor, Purdy, Jackson, Phillips, and Virues-Ortega (2019) and are in line with guidelines identified by Shadish, Hedges, Horner, and Odom (2015).

All screening tools (secondary outcome measures) were administered during the initial intake of the participant and were re-administered at the 1-month follow-up post intervention and are comprised of:

**The Infant-Toddler Checklist (ITC; Wetherby & Prizant, 2002).** The ITC is a 24-question inventory that produces cutoffs and standardized scores at monthly intervals from 6 to 24 months to determine the risk for ASD and communication disorders compared to a normative sample, initially comprised of 2188 infants. Raw scores below 1.25 standard deviation of the mean are considered to be “of concern”.

**The Modified Checklist for Autism in Toddlers Revised (M-CHAT-R; Robins et al., 2009).** The M-CHAT-R is a 20-item parent-questionnaire validated with infants between 16 and 30 months of age. Although validated for infants over 16 months of age, some research has looked at its use as a screener for infants as young as 8 months indicating potential acceptable reliability (Inada, Kamio, & Koyama, 2010). Scores between 3 and 8 require a follow-up, scores above 8 are considered high-risk and should by-pass the follow-up questionnaire and be referred for in-depth diagnostic assessments.

**The Autism Observation Scale for Infants (AOSI; Bryson et al., 2008).** The AOSI is an 18-item semi-structured assessment of autism symptoms in infants between the ages of 6–18 months. The AOSI produces two sets of scores: the number of markers (items which received a score) and a total score consisting of the total number of markers plus the severity of each marker. Scores equal to or above 7 for number of markers and equal to or above 9 for total score are considered positive screens.

**Mullen Scales of Early Learning (MSEL; Mullen, 1995).** The MSEL is a standardized assessment that is designed and tested for children from birth to 68 to measure language, cognitive and motor abilities. The MSEL produces scores across five domains (Fine Motor,
Gross Motor, Visual Reception, Receptive Language and Expressive Language) as well as provides total scores, t-scores, percentile rankings and age-equivalent measures.

**Treatment fidelity**

Parent target behaviors mastery fidelity was scored from 5-minute videos taken each session and rated on a scale from 0 (shows zero instances of previously taught topic) to 3 (shows three or more instances of previous topic in a 5-minute period). A score of 3 is considered the minimum required to demonstrate the intervention topic at mastery level. Parents were required to obtain a score of 3 prior to moving to the next topic (See Table 1 for training topics).

Fidelity of intervention was scored through a self-report checklist completed by the trainer and attached to the session notes and provided to parents as a copy at the end of each session. The checklist comprised of eight items: (1) allows opportunity for initial questions and update from previous week, (2) collects 5-minute video sample of parent-infant dyad, (3) confirms three demonstrations of previous topic in 5-minute video and provides feedback, (4) provides instructions for new topic, (5) models new topic, (6) allows opportunity for parents to practise new topic, (7) provides feedback and recommendations for the upcoming week in the form of three to five goals, and (8) allows time for further questions. Fidelity of intervention data was collected across all sessions and conditions, as the checklist was part of the session handouts. Parental fidelity of implementation “three demonstrations of previous topic in 5-min” was assessed during initial video scoring and 30% of videos were scored by a second observer.

**Inter-observer agreement**

Two additional observers, who were blind to the condition and session number, independently scored approximately 30% of all videos (26 out of 89 sessions) across experimental conditions. The additional observers were provided with operational definitions of target behaviors and trained to identify exemplars and non-exemplars from videoed sessions. IOA was calculated interval-by-interval, by dividing the number of agreed intervals by the number of agreements and disagreements and multiplying by 100 across all three target behaviors. The mean IOA across 26 sessions was 88% (range 78–96%). More specifically, for the parental target behavior of providing learning opportunities, the mean IOA was 86% with a range of 70–97%. For infant eye-gaze, the IOA ranged from 83% to 100% with a mean of 93%. For infant responsivity, IOA ranged from 73% to 97% with a mean of 86%.

**Results**

**Primary outcomes**

Learning opportunities provided by parents saw the most modest increases ranging from a change of −4% to 41% and a mean of 23.4% across all five participants by the end of the 12-week intervention. Changes in learning opportunities maintained at the 3-month follow-up averaging 24% change over baseline. Infants responding to learning opportunities showed significant changes in responsivity averaging an increase of 41% increase
across all children with a range of 33–54% change over baseline. At baseline, Sara’s mother provided a learning opportunity during 33% of the intervals, which increased to an average of 86% of the intervals during the last three intervention sessions (Figures 1 & 2). Ben’s mother was trained as a behavior interventionist prior to having Ben; therefore, she presented consistently high levels of learning opportunities across baseline, intervention and follow-up phases of the study. During baseline, Ben’s mother presented learning opportunities in 70% of the intervals, which slightly dropped to 66% at the end of the 12-week intervention and 3-month follow-up, likely due to ceiling effects during baseline. Leah’s mother significantly increased her presentation of learning opportunities from approximately 30% during baseline to approximately 70% by the final three sessions of the intervention (Figures 1 & 2) and to 80% by the 3-month follow-up, showing a 50% increase over baseline. Ray and Ann are considered to be high-risk sibling and with their older siblings both receiving at-home ABA services for approximately two years. Therefore, both parent participants had a foundational understanding of incorporating ABA into play. Ray’s mother increased learning opportunities from 42% at baseline to 83% by the end of the 12-week intervention with a slight decrease to 67% by the 3-month follow-up. Ann’s mother had the highest presentation of learning opportunities during baseline at 81% thereby making only a 2% increase by the end of the 12-week intervention which maintained through the 3-month follow-up.

Infant eye contact demonstrated the most significant gains out of all three target behaviors with a mean increase of 40% at the end of the intervention over baseline with results maintaining through the 3-month follow-up. Sara’s eye contact increased from approximately 25% of intervals during baseline to 75% of intervals during the final two sessions of the intervention phase. Ben’s largest gains were seen with the frequency of his eye contact increasing from only 21% of the intervals during baseline to 76% by the final two intervention sessions. Leah’s eye contact increased from 10% of intervals at baseline to 40% of intervals by the end of the intervention. Ray’s eye contact increased from 12% at baseline to 63% by the 3-month follow-up, while Ann made the most modest gains with regards to eye contact, increasing from 16% at baseline to 33% by the end of the 12-week intervention.

All five infants also showed notable improvements with responding to learning opportunities, beginning at an average of 19% during baseline and finishing the intervention with an average of 53%. Sara demonstrated the largest gains with an increase of 58% over baseline which maintained at the 3-month follow-up. Ben presented with atypical and delayed gross and fine motor strength and coordination which appeared to limit his ability to respond to various learning opportunities that required motor imitation and toy manipulation, however still made notable gains with regards to responding to learning opportunities, increasing his baseline scores by 47% (Figures 1 & 2). Leah, Ray and Ann demonstrated changes of 43%, 34% and 32%, respectively, from baseline to the end of intervention with results maintaining or improving at follow-up sessions.

The Shadish g effect size estimators for children responsivity, children eye contact and parent behaviour were all within the large or very large effect size range, with over one standard deviation improvement for children eye contact and parent behaviour and over two standard deviations improvement for children responsivity (Table 2).
Figure 1. Percentage of intervals with responsivity and eye contact across participants.
Figure 2. Percentage of intervals containing parent target behavior.
Secondary outcomes

All five infants in the sample showed a decrease in autism symptoms and an increase in appropriate acquisition behaviors within the 12-week intervention period (Figures 1 & 2 and Table 3). The ITC, M-CHAT-R AOSI and MSEL were re-administered at the 1-month follow-up (Table 3) however all four of the five infants did receive a positive diagnosis of ASD between 18 and 22 months of age.

Sara

Sara no longer obtained “concern” on the ITC, moved from “high risk” to “moderate risk” score on the M-CHAT-R, her AOSI total score decreased by 5 points and total marker score decreased by 3 markers, and her MSEL expressive and receptive scores went from an age-equivalent of approximately 3 months at intake to an age equivalent of approximately 25 months post intervention, which was 5 months ahead of her current age (Table 3). Sara made significant gains within the 12-week intervention; nevertheless, she was diagnosed with ASD prior to the 3-month follow-up at 21 months of age.

Ben

Ben showed large decreases in autism symptoms on both the ITC and M-CHAT-R, however, remained in the “concern” and “high risk” categories on these screeners. Ben’s total score and number of markers on the AOSI significantly decreased during the 12-week intervention, by 4 and 3 points, respectively, as well as and his MSEL-ELC age-equivalent score went from 1 month of age at intake to 15 months of age (only 1 month behind his current age) post intervention (Table 3). Ben was diagnosed with ASD at 18 months of age, between the 1 and 3-month follow-up.

Leah

Leah’s M-CHAT-R score decreased from the “high-risk” group to “no risk” group and her ITC score changed from “concern” to “no concern”. Her AOSI scores saw a decrease from a total score of 10 to a total score of 4, no longer showing a concerning score. Lastly,

Table 3. Summary of pre (baseline) and post-intervention scores (1-month follow-up) screening instruments.

<table>
<thead>
<tr>
<th>Age (months)</th>
<th>ITC</th>
<th>M-CHAT-R</th>
<th>AOSI (Total score/ number of markers)</th>
<th>MSEL-RLC (Raw Scores/Age equivalent mos.)</th>
<th>MSEL-ELC (Raw Scores/Age equivalent)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Pre</td>
<td>Post</td>
<td>Pre</td>
<td>Post</td>
<td>Pre</td>
<td>Post</td>
</tr>
<tr>
<td>Sara</td>
<td>16</td>
<td>20</td>
<td>18*</td>
<td>52</td>
<td>15</td>
</tr>
<tr>
<td>Ben</td>
<td>12</td>
<td>16</td>
<td>8*</td>
<td>20*</td>
<td>16</td>
</tr>
<tr>
<td>Leah</td>
<td>12</td>
<td>16</td>
<td>22*</td>
<td>40</td>
<td>11</td>
</tr>
<tr>
<td>Ray</td>
<td>13</td>
<td>17</td>
<td>9*</td>
<td>26*</td>
<td>13</td>
</tr>
<tr>
<td>Ann</td>
<td>15</td>
<td>19</td>
<td>21*</td>
<td>42</td>
<td>7</td>
</tr>
</tbody>
</table>

ITC uses standardized cut-off scores that vary by chronological age; * indicates “of concern” scores. Scores for M-CHAT-R are divided into three categories “no risk” (0–3), “moderate risk” (4–8) and “high risk” (9–20). A higher score in AOSI total score denotes increased severity of ASD symptoms with a maximum score of 44. MSEL raw scores for expressive and receptive language and the age-equivalent scores in months.
her MSEL receptive and expressive scores reached an age-equivalent score of 18 months, 2 months older than her current age at the time of testing (Table 3). With regards to secondary outcomes, she has not received a diagnosis of ASD, and her parents no longer have concerns regarding her development.

**Ray and Ann**

Ray made large gains across all of his scores, most notably his MSEL-ELR score went from an age-equivalent score of 5 months of age to 19 months of age, 2 months older than his current age at the time of the post-test. Ann had relatively higher scores at intake compared to the rest of the sample; however, she was still flagged “at risk” across all of the screeners and assessment tools. Post intervention, her M-CHAT-R and ITC screeners moved to “no concern” and on the cusp between “no risk” and “moderate” risk. She had made significant progress with receptive and expressive language during this time and had begun stringing two words together, which attributed to less concerning scores at the 1-month follow-up. Both Ray and Ann received a positive diagnosis for ASD between the 1 and 3-month follow-up.

**Social validity**

Furthermore, social validity was measured at the end of the 3-month follow-up session through a parent satisfaction questionnaire (Charlop-Christy & Carpenter, 2000) containing four questions: 1) How easy was it to implement the intervention each week? 2) Has the intervention helped your child improve their social communication? 3) Has the intervention helped you to better communicate with your child? 4) Would you recommend this intervention to another family similar to yours? (Table 4). All parents completed the questionnaire via pen and paper during the 3-month follow-up visit and 96% (24/25) responded to the questions as either positive or very positive.

**Discussion**

The present study provides strong evidence for the efficacy of a novel sequential curriculum (SPARC) delivered through a 12-week parent-mediated intervention. Outcomes of treatment effect on social communication and number and severity of autism symptoms are very positive, showcasing the potential of a low-intensity parent-mediated intervention for infants at risk to alter children’s developmental pathway.

All parent-child dyads presented significant improvements across primary outcomes, notably in child responsivity to learning opportunities, child eye contact and parent rate

<table>
<thead>
<tr>
<th>Table 4. Results of parent satisfaction questionnaire – social validity.</th>
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<tbody>
<tr>
<td>Question</td>
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<tr>
<td>1. How easy was it to implement the intervention each week?</td>
</tr>
<tr>
<td>2. Has the intervention helped your child improve their social communication?</td>
</tr>
<tr>
<td>3. Has the intervention helped you to better communicate with your child?</td>
</tr>
<tr>
<td>4. Would you recommend this intervention to another family similar to yours?</td>
</tr>
</tbody>
</table>

Likert Scale: 1 = not easy/not much, 5 = very easy/very much.
of presentation of learning opportunities. Overall, treatment effects on children responsiveness were very large. Treatment effects on children eye contact and parent behaviour were within the large range. These results provide strong evidence for the excellent potential that early behavioural treatment has for altering the developmental trajectory of infants at risk of receiving a diagnosis of ASD. These findings are in line with other studies examining the effect of a behavioural intervention on child outcomes (Taylor et al., 2019), adding to the vast literature around evidence-based practice for ASD that pinpoints Applied Behaviour Analysis as the scientific basis of treatments of choice.

With neuroplasticity maximized between 0 and 3 years of age typically resulting in rapid development and with the use of a multiple baseline design instead of an RCT, it is important to take into consideration to what extent maturation can be ruled out as responsible for the acquisition of new behaviors (Dounavi & Dillenburger, 2013). More specifically, we need to determine the degree to which behavior change would have been achieved with maturation alone versus the degree to which positive behavior change is a result that can be attributed only or majorly to the intervention assessed in this study. An RCT of a similar parent-mediated intervention found that while the control group of high-risk infants who did not receive intervention still saw a decrease in total AOSI score by an average of 1.77 (maturation effect) the 12-week treatment group saw a decrease of approximately 4.15 in total AOSI score (the treatment effect) (Green et al., 2017). In the present study, total AOSI scores decreased by an average of 5.2 per participant, slightly greater than the treatment group effect registered in Green et al. (2017) and significantly more than what would be expected as an effect of maturation alone over a similar period of time with similar-aged participants. Additionally, the MSEL and ITC calculate age-equivalent standardized scores and therefore changes in the participants’ pre and post-test age-equivalent scores can also be used to observe expected maturation effects versus treatment effects. With regards to the MSEL, typical development (i.e., maturation effects) would assume that children would show 1-month increases on the standard assessment for each month in-between assessments. For the present study, approximately 4 months elapsed between baseline and assessments completed at the 1-month follow-up; therefore, a change of 4 months to the age-equivalent score would be expected. However, the five infants saw mean changes of 14-month increases for receptive scores and 15-month increases for expressive scores over this four-month period, showing approximately a 10-month gain of skills that would not be accounted for by maturation alone.

The 12-sessions of parent-mediated intervention were successful at decreasing the severity of autism symptoms while increasing all of the social communication target behaviors in parents and infants using ABA principles and strategies, primarily; (1) Verbal Behavior (2) Natural Environment Teaching (3) Behavior Skills Training and (4) teaching for generalization. The present study is unique and adds to the previous body of literature in that it provides a sequential curriculum (SPARC) rooted in ABA to treat autism symptoms in infants, that can be implemented by any parent or entry-level professional, under the guidance of an adequately qualified professional (e.g., Board Certified Behavior Analyst®) with experience in very early intervention, without requiring additional credentialing. Nearly all published 12-week parent training programs require specialized certifications thereby limiting accessibility to those who cannot physically and financially attend the training. The present study aims to provide a framework (Table 1) for adequately qualified professionals (e.g., Board Certified Behavior Analyst®) to use as a guide when designing and implementing parent-mediated intervention with very
young children who are showing signs of ASD. Additionally, the present study provides support to existing literature in that using low-intensity parent-mediated intervention (12 hours) based on the SPARC is an effective way to reduce autism symptoms and increase appropriate social communication behaviors such as eye contact and responding to learning opportunities (Beaudoin, Sébire, & Couture, 2014; Brian et al., 2015; Green et al., 2017; Rogers et al., 2014).

The present study further adds to the validity of the various ASD screeners and assessment tools used (M-CHAT-R, ITC, AOSI, MSEL), as 4 of the 5 participants did meet criteria for an ASD diagnosis by approximately 19 months of age and had “at risk” scores across all four tools during intake. Scores across all four screening instruments in the present study were more concerning than the average scores in similar research using high-risk infant samples. This is likely explained from the fact that many “at-risk” infant samples are considered “at-risk” because they have a sibling with ASD. In the present study, three out of five participants were referred to the study because their parents noticed “red flags” very early in their development; therefore, the severity of the symptoms may have been greater than that of a larger sample of infant-siblings who are considered “at-risk” even when they do not present concerning behavioral indicators.

Two of the five participants were symptomatic infant siblings (Ray and Ann) and both have school-aged siblings with ASD and single mothers as their primary caregivers, who availed of minimal family support. This resulted in occasionally having the older sibling being present during the coaching session. Although both of these mothers had a greater foundation of ABA knowledge prior to beginning of the intervention, it was acknowledged that a parent-mediated intervention model was difficult to execute weekly based on the high needs of their school-aged child with more severe needs. Future directions for parent-mediated research should explore if certain parent or family profiles or characteristics tend to be correlated with more successful outcomes for this type of service delivery model. For example, for parents with limited resources due to having many children, additional children with special needs, being a single parent, having limited family support or a demanding job, etc., perhaps a hybrid service delivery model of parent training plus direct training by professionals may be more beneficial. It should be reiterated that the goal of pre-diagnostic intervention is not to eliminate the future diagnosis of ASD but rather maximizing the child’s developmental trajectory and potential.

Given the nature of applied research, there are a few limitations to consider. Firstly, the present study was comprised of a small sample of only five parent-infant dyads using a multiple baseline across participants design; however, outcome data are similar to the treatment effects found in recent RCTs (Gammer et al., 2015; Green et al., 2017). Secondly, the M-CHAT-R is validated for children between 16 and 30 months of age; however, it was administered as one of four screening tools prior to its designed cut-off age of 16 months with four out of the five participants. Many of the items on the M-CHAT-R, such as using simple gestures, following a point, referencing faces, social smiling and joint attention, are understood to develop before 12 months of age (Zwaigenbaum et al., 2005). In fact, as many as 75% of the items on the screener emerge in typically developing infants between 6 and 12 months (Inada et al., 2010). Thereby, significant deficits in these milestones even at 12 months of age should be a reasonable indicator of a developmental delay or an ASD trajectory (Brooks & Meltzoff, 2005; Wright & Poulin-Dubois, 2012). As documented in Table 3, scores from the ITC, AOSI and MSEL have indicated that all participants were “at
risk” and showed “of concern” delays in their development at the time of intake and had substantially improved scores by the 1-month follow-up, above typical maturation results, which supports that the observed changes were not due to extraneous variables but could rather be attributed to the intervention. Additionally, the fidelity of implementation checklist was completed by the trainer during each session however a copy of the checklist was provided to the parents at the end of each session. Lastly, as the intervention is designed to be completed throughout daily activities, parents did not record the amount of time they dedicated each week to practicing the weekly topic; future research could benefit from including a parent-reported measure of the number of minutes allocated per week to the intervention.

As early screening and detection continue to become part of common medical practice, it is imperative that as a field of autism service providers and researchers we continue to find solutions to build capacity for pre-emptive autism intervention. Using a parent-mediated service delivery model with at-risk infants allows service providers to have a greater reach as fewer direct clinician hours are initially required given the low-intensity clinical time commitment of the coaching model (1 hour per family per week), allowing more families to access pre-diagnostic services. Behavior analysts have the necessary education and tools to teach complex behaviors such as social communication, joint-attention, imitation, echoing, manding, tacting, following directions and play while reducing challenging or non-functional behaviors. With multi-year-long waitlists for ASD diagnosis, the time-sensitivity of the intervention and screening tools that are readily available to detect ASD symptoms in children by their first birthday, professionals should not feel compelled to wait for a confirmation of ASD prior to beginning treatment. Behavior analysts are trained to identify socially significant target behaviors (in this case, age-appropriate developmental milestones), assess current level of performance and then determine the steps and teaching method required to acquire target behaviors, thus the process is the same regardless of the presence or absence of an ASD diagnosis. This study aims to mobilize more behavior analyst towards the use of pre-emptive interventions in an attempt to build treatment capacity while simultaneously alleviating some of the strain that life-long care produces by maximizing the developmental trajectory potential of infants who are showing early signs of ASD.

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Compliance with Ethical Standards

The present study was conducted within the parameters of clinical practice and was not funded. Ethical approval: All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional research ethics committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed consent: Informed consent was obtained for all individual participants included in the study.
This article does not contain any studies with animals.

**Disclosure statement**

No potential conflict of interest was reported by the authors.

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