DEVELOPMENTAL TRAJECTORIES OF AUTISM SPECTRUM DISORDER SYMPTOMS IN AT-RISK TODDLERS

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ABSTRACT

Allison T. Meyer: Developmental Trajectories of Autism Spectrum Disorder Symptoms in At-Risk Toddlers
(Under the direction of Laura G. Klinger)

Symptoms of Autism Spectrum Disorder (ASD) including impairments in social communication and the presence of restricted and repetitive behavior (RRBs) are present in the first 2 years of life, prior to diagnosis. This study compared two developmental models describing the early emergence of ASD symptoms, Social Motivation and Attention theories, in a community-based sample of infants who were followed from 12 months of age into early childhood. Participants included 43 children identified at high-risk for a later diagnosis of ASD based on a positive screen on the First Year Inventory (FYI) at 12 months of age. Toddlers were evaluated at 13 and 22 months, followed by a diagnostic evaluation at 3-5 years. Video coding for social motivation (looking at people) and difficulty disengaging attention (shifting of attention and RRBs) was completed at 13 and 22 months. Path analyses were conducted to evaluate the direct and indirect effects of measures of looking at people, attention shifting, and RRBs on ASD symptom severity. Results indicated a significant indirect effect from decreased looking at people at 13 months to decreased attention shifting at 22 months to increased ASD symptom severity at age 3-5 years. Further analyses indicated that this indirect effect remained when only examining social-specific shifting (i.e., attention shifting including a person) but was not present when only including non-social shifting (i.e., shifting attention between objects). Results from this study better support the Social Motivation theory of the early emergence of ASD symptoms than the Attention theory. While impairments in attention clearly play an integral role in later diagnosis of ASD,

attention shifting that included social information appears to play a larger role than attention shifting to non-social information. Further, results suggest that RRBs appear to develop separately and in parallel to symptoms of decreased looking at people and attention shifting involving people. Further examination of how these early symptoms predict later school-aged outcomes (e.g., symptom severity, comorbid attention difficulties, etc.) is needed to gain a further view of the developmental processes in ASD.

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LIST OF ABBREVIATIONS

ADOS	Autism Diagnostic Observation Schedule
ASD	Autism Spectrum Disorder
CSBS	Communication and Symbolic Behavior Scale
DAS-2	Differential Ability Scales-Second Edition
DD	Developmental Delay
EDP2	Early Development Project-2
FYI	First Year Inventory
ID	Intellectual Disability
MSEL	Mullen Scales of Early Learning
RBS-R	Restricted and Repetitive Behavior Scale-Revised
RRB	Restricted and Repetitive Behavior
RSM	Repetitive and Sensory Movement Scale

CHAPTER 1: Developmental Trajectories of Autism Spectrum Disorder Symptoms in At-Risk Toddlers

Autism Spectrum Disorder (ASD) is characterized by impairments in social communication and interaction and the presence of restricted and repetitive behaviors and interests (RRBs). These broader symptom categories are present throughout life, however the specific symptoms change throughout development. ASD can be diagnosed accurately by between 2-3 years of age with symptoms both in social communication and interaction and repetitive behaviors developing during the first two years of life (Ozonoff et al., 2015; Rondeau et al., 2011; Woolfenden, Sarkozy, Ridley & Williams, 2012). For example, studies have suggested that individuals with ASD exhibit impairments in social attention in the first 6-12 months of life (Dawson et al., 2004). Individuals with ASD also exhibit atypical repetitive motor behaviors by 12 months (Wolff et al., 2014). Although we know that these atypical social communication and repetitive behaviors unfold across time, little is known about how these two symptom areas interact and influence one another during early development. Understanding these developmental processes can help inform intervention programs that target these impairments early in life. The broad overall goal of this study is to better understand the developmental processes of social attention, attention shifting, and RRBs and their relationship to one another during early development in children with and without ASD from age 1-5 years.

To better understand the co-occurring development and relationship between symptoms, a review of the literature on atypical development of social communication and RRBs in

ASD is needed. Specifically, these symptom categories will be discussed as they relate to infants and toddlers later diagnosed with ASD and be compared to early typical development and developmental delay to better understand when differences in development begin to arise. Last, two theoretical models will be discussed in their relation to ASD symptom development. These models focus on how early impairments related to ASD symptoms influence one another and the cascading effects of early impairments in specific developmental areas.

Social Communication and Reciprocity

Individuals with ASD exhibit impairments in social communication and interaction throughout their life. The DSM-IV-TR (American Psychiatric Association, 2000) separated social and communication impairments into two separate categories. However, given the overlap in these symptoms and the difficulty in differentiating social specific and communication specific impairments, the most recent diagnostic criteria in the DSM-5 (American Psychiatric Association, 2013) for ASD focuses on impairments in social communication and interaction as a single symptom category.

Core impairments in social communication and interaction are maintained throughout the lifespan; however, the presentation of these symptoms changes over time as the individual develops and gains new skills. Impairments in social emotional reciprocity include difficulties with back-and-forth conversations, reduced sharing of interests, emotions, or affect, or failure to initiate or respond to social interactions. For young children with ASD, this symptom may appear as difficulty with back-and-forth social games such as *peek-a-boo* or infrequent showing of preferred toys or other objects. Nonverbal communication difficulties include poorly integrated verbal and nonverbal communication and difficulties

using and understanding nonverbal communication such as eye contact or body language. Infants and toddlers later diagnosed with ASD may use less eye contact or fewer gestures such as pointing or reaching to objects to indicate a preference. Deficits in developing, maintaining, and understanding relationships include difficulties understanding changes in behavior in different contexts, difficulty making friends and engaging in shared play and enjoyment with others. In young children with ASD, this may be exhibited in their decreased interest in other people or children in their environment and/or a difficulty in developing early play skills.

When more specifically examining the development social communication and interaction in infants and toddlers later diagnosed with ASD, the literature has focused on two types of social impairments: dyadic interactions and triadic interactions (e.g., joint attention).

Dyadic Interactions

In children with typical development, dyadic social engagement develops during the first few months of life (Bakeman & Adamson, 1984). That is, young children engage their attention with one other object or person at time (e.g., just a toy or just a person). Dyadic interactions typically involves another person as infants show a preference for and response to social stimuli within the first few months of life (Haith, Bergman, & Moore, 1977). Early dyadic interactions with other people include reciprocal social interactions with a single person such as during peek-a-boo or responding to their name being called.

Studies with children with ASD have examined dyadic interactions through measuring social engagement and social orienting (Dawson et al., 1998). In general, research suggests that infants later diagnosed with ASD show atypical patterns of attention to social stimuli

without clear evidence of a preference for social information by 12 months. Atypical social engagement has been found in studies documenting decreased dyadic orienting, attention to faces (e.g., eye contact), and social reciprocity in infants who are later diagnosed with ASD (Osterling & Dawson, 1994; Wetherby, Woods, Allen, Cleary, Dickinson, & Lord,. 2004). One of the hallmark "red flags" for ASD is failure to respond to one's name being called or other bids for social attention during early development (Baranek, 1999; Clifford & Dissanayake, 2008; Leekam & Ramsden, 2006; Osterling & Dawson, 1994). Clifford and Dissanayake (2008) used both retrospective parent interview and old home videos of children from birth to 24 months of age to examine dyadic interactions in children with ASD compared to those with typical development and developmental delay. Compared to the other groups, parent's reported that children with ASD exhibited impairments in dyadic interactions by 6-12 months including infrequent eye contact and decreased responsive social smiles. Video coding of these same dyadic behaviors during the first two years of life confirmed that children with ASD had impairments in eye contact both in frequency and quality compared to those with typical development and developmental delay (DD).

Impairments in social engagement during in-person and naturalistic play assessments persists throughout early development (Dawson et al., 2004; Leekam & Ramsden, 2006). Leekam and Ramsden (2006) suggested that 4-year-old children with ASD responded to significantly fewer verbal bids for attention from adults compared to those with DD and typical development during a naturalistic play-based assessment as evidenced by reduced social orienting. Dawson and colleagues (2004) measured how frequently children with ASD, DD, or typical development oriented to social (e.g., name call) stimuli compared to non-social stimuli (e.g., timer beeping). Results suggest that 3-year-old children with ASD

oriented to social stimuli less frequently than non-social stimuli. Similar to Leekam and Ramsden (2006), those with ASD oriented less to all types of social stimuli compared to those with DD. These findings suggest that social orienting is specifically impaired in children with ASD rather than non-social orienting.

Social engagement is also frequently measured through eye-tracking of human faces. These computer-based measures can help to identify impairments in dyadic interactions in very young children using a controlled experimental design. These tasks measure face recognition or preferential gaze toward social versus nonsocial stimuli. Studies examining how infants and toddlers later diagnosed with ASD attend to social stimuli such as faces have found mixed results. Chawarska and Volkmar (2007) examined face recognition of people versus monkeys in 2 and 4 year-old children with ASD and DD. After being familiarized with the people and monkeys, 2-year-olds with ASD were no better at recognizing human faces compared to monkeys while those with DD did perform better with human faces versus monkey faces, although this difference did not present in 4-year-old participants. This research suggests that very young children with ASD appear to be processing and attending to faces in a fundamentally different way that is separate from symptoms of developmental delay. Prospective eye-tracking studies indicate that as early as 2-6 months of age, infants later diagnosed with ASD look at faces differently, initially spending more time looking at the eyes compared to those with typical development and later (i.e. at 6 months) spending significantly less time looking at eyes (Jones & Klin, 2013). Similarly, during a simple videotaped social scene with an actress and 4 toys on the screen, 6-month-old infants with ASD spent significantly less time looking at the actress' face compared to infants with typical development (Chawarska, Macari, & Shic, 2013). Infants later diagnosed with ASD

appear to have decreased or atypical spontaneous attention to social information, particularly faces, which may interfere with their ability to process social information.

Across this body of literature, there is evidence that children later diagnosed with ASD exhibit impairments in social orienting and dyadic interactions at a young age that is not related to developmental delay and may influence subsequent social skill development as evidenced by parent report, behavior observation, and eye-tracking studies.

Triadic Interactions/Joint Attention

Following the development of dyadic interactions, 9 to 10-month-old infants with typical development engage in triadic interactions with others by incorporating objects and other information into social interactions (Striano & Rochat, 1999). During these interactions, infants shift their gaze between an object and a person to communicate with a person about the object; to share their interest in an object, request an object, express emotional distress, or express other needs. This shared or joint attention is an integral part of development that helps children better understand social relationships, interactions, and their general environment (Tomasello, 1995). In typical development, infants are learning that by making eye contact and interacting socially with others, they can see, learn, and experience more things in their environment (Carpenter, Nagell, Tomasello, Butterworth, & Moore, 1998). This skill is imperative in developing social relationships and understanding the importance and power of social engagement (Mundy & Newell, 2007; Tomasello, Carpenter, Call, Behne, & Moll, 2005). Infants experience positive interactions and feedback when looking and smiling at other people by seeing that the person will smile back at them or react positively.

Studies suggest that infants later diagnosed with ASD are impaired in triadic interactions including a lack of or impairment in both their response and initiation of joint attention (Dawson et al., 2004). A lack of joint attention skills and lack of appropriate gaze during the first 2 years of life is considered one of the "red flags" for parents of children with ASD (Wetherby et al., 2004). Specifically, infants and toddlers later diagnosed with ASD have difficulty following another person's gaze or point in order to share information or attend to relevant social information (Charman, 2003; Charman, Swenttenham, Baron-Cohen, Cox, Baird, & Drew, 1997). For example, infants later diagnosed with ASD may not look up when their mom points to a plane in the sky and says "Look! A plane!" Based on retrospective parent report and detailed video coding, infants later diagnosed with ASD between birth and 24 months initiate and respond to joint attention less frequently than children with typical development or DD (Clifford & Dissanayake, 2008). Similarly, during naturalistic play assessment, three-year-old children with ASD make fewer attempts to initiate joint attention and are less likely to respond to joint attention compared to children with DD or typical development (Dawson et al., 2004).

To further understand triadic engagement, studies have specifically examined how infants and toddlers later diagnosed with ASD shift their attention between social and nonsocial stimuli. Chawarska, Volkmkar, and Klin (2010) suggest that infants with typical development and those with DD had *more* difficulty disengaging from social stimuli, such as faces, compared to those with ASD. Toddlers with ASD were less interested in social stimuli suggesting differences or impairments in attentional focus at an early age towards social and nonsocial stimuli. Impairments in dyadic interactions in children later diagnosed with ASD may interfere with later development of joint attention and triadic interactions.

Summary

Infants and toddlers with ASD exhibit difficulties both in general dyadic social engagement and triadic interactions in which enjoyment is shared with others. The literature is compelling in suggesting that infants with ASD interact with people and their surrounding environment differently compared to infants with typical development (Osterling & Dawson, 1994; Wetherby et al., 2004). Infants with ASD are clearly searching, scanning, focusing on, and engaging with people, objects, and their environment differently very early in life. Understanding how infants and toddlers with ASD engage with their social environment can provide insight into how infants engage with nonsocial stimuli.

One hypothesis regarding impairments in social engagement is that individuals with ASD have an underlying impairment in social motivation (for review see, Dawson, Webb, & McPartland, 2005) that leads to decreased social interaction with others. Conversely, this hallmark impairment in social motivation suggests that individuals with ASD may be more motivated to engage in nonsocial interactions and experiences. For infants and toddlers with ASD, these nonsocial interests likely include RRBs. That is, infants and toddlers with ASD may experience more positive reinforcement from engaging in repetitive behaviors with objects than from engaging in social interactions with others.

Restricted and Repetitive Behaviors and Interests

Repetitive behaviors have generally been defined as "broad and often disparate classes of behavior linked by repetition, rigidity, invariance, and inappropriateness (Turner, 1999, p. 839)." However, some repetitive behaviors are considered to be developmentally normative, particularly in infants and toddlers (Thelen, 1979, 1981). Until more recently, researchers hypothesized that repetitive behaviors in infants and toddlers did not differentiate

between those with ASD and those without as infants and toddlers with ASD engage in repetitive kicking or banging of objects similar to those with typical development (Charman & Baird, 2002; Cox, 1999; Ventola et al., 2006) or those with developmental delay (Stone et al., 1999). Instead, social and communication impairments associated with ASD were thought to be present during the first two years of life while repetitive behaviors and restricted interests were believed to not appear until early childhood. More recent research, though, has suggested that young children with ASD *do* in fact appear engage in behaviors that are representative of the RRB symptoms described in the DSM-5 (e.g., Wolff et al., 2014; Elison et al., 2014).

There are 4 areas of RRBs described in the DSM-5 that are considered when providing a diagnosis of ASD. The first area includes stereotyped repetitive motor movements, use of objects, or speech as core symptoms of ASD. These atypical behaviors are present early in life for infants later diagnosed with ASD, continue throughout early childhood, and interfere with a child's ability to appropriately and effectively interact with their world. Insistence on sameness and the presence of circumscribed interests are the last 2 types of RRBs. This includes behaviors related to rigidity, ritualized patterns of behavior, and overall difficulties with changes in routine and/or environment. These behaviors increase during early childhood and persist throughout development in individuals with ASD (Chowdhury, Benson, & Hillier, 2010; Hattier, Matson, Tureck, & Horovitz, 2011). RRBs also include the presence of circumscribed or restricted interests that may be atypical in their intensity and/or focus. For example, a young child may have a strong attachment to a particular object or only want to play with very specific toys. The last area of RRBs includes sensory sensitivities, which have long been an associated characteristic of ASD (e.g., Baranek et al., 2006; Ben-Sasson et al.,

2009) although it was not included in the diagnostic criteria until the publication of the DSM-5. This symptom category includes "hyper- or hypo-reactivity to sensory input or unusual interest in sensory aspects of the environment (American Psychiatric Association, 2013)." These symptoms may be evident in ASD as early as 9-12 months of age evidenced by sensitivity to touch and other sensitivities (Baranek, 1999). While RRBs are a core symptoms category within ASD, it is imperative to understand the development of RRBs in the context of repetitive behaviors during early typical development to compare to atypical development in infants and toddlers later diagnosed with ASD.

Comparison of RRBs to Typical Development and Developmental Delay

Studies examining RRBs in individuals with ASD compared to those with typical development have primarily looked at RRBs as one broad symptom category rather than each area of RRBs as a separate symptom. However, given the importance of repetitive motor behavior in early typical infant development, some research has examined repetitive motor mannerisms and repetitive objects use more specifically in typical development compared to infants later diagnosed with ASD.

Studies that examine RRBs as a single, broad category have found mixed results. These studies primarily use parent-report measures and interviews rather than video coding or experimental measures. For example, Cox et al. (1999) examined RRBs in children at 20 months and 42 months using the Autism Diagnostic Interview- Revised (ADI-R), a comprehensive parent interview related to ASD symptoms. There were no significant differences in RRBs between toddlers with ASD and typical development at 20 months when using the ADI-R parent interview. Significant differences were present by 42 months based

on this parent interview such that those with ASD exhibited more RRBs than children with typical development.

Other studies have suggested that while children with typical development exhibit some RRBs, children with ASD have behaviors that are above and beyond that of typical development (Harrop, McConachie, Emsley, Leadbitter, & Green, 2014; Ray-Subramanian & Ellis Weismer, 2012; Wolff et al., 2014). For example, Harrop et al. (2014) found that while children with typical development had some RRBs from age 3-5, the RRBs of children with ASD were consistently higher in frequency and/or intensity.

Repetitive motor behaviors have been thoroughly studied in infants with typical development as the movements are an important part of early development. Early work by Thelen (1979, 1981) found that infants with typical development from 6-9 months of age engage in more than 40 different types of motor mannerisms including repetitive kicking movements. She found that this repetitive motor behavior was similar to the movement of legs during walking. This precursor to walking suggests that these types of repetitive motor behaviors are developmentally appropriate and serve an important function for developing motor skills. In a study using a community sample of 15-month-old infants with typical development, parents reported increased motor and sensory behaviors compared to previous studies of 2-year-olds with typical development (Arnott et al., 2010). This indicates that some motor behaviors that appear similar to those in ASD are present in typical development, particularly during early development. However, Wolff et al. (2014) reported that infants who later met criteria for ASD had more parent-reported repetitive motor mannerisms at 12 months compared to those that did not go on to develop ASD. These same behaviors persisted across development or increased in frequency for those later diagnosed with ASD

and continued to be present at 24 months. In those with typical development, these RRBs were present at lower frequency at 12 months and either decreased or stayed significantly lower compared to those later diagnosed with ASD (Wolff et al., 2014). Repetitive motor mannerisms in infants with ASD do not appear to have a clear function or assist in the learning and development of everyday skills as it does in typical development (e.g. transition from crawling to walking).

Studies examining repetitive use of objects in infants later diagnosed with ASD have been mixed in identifying whether this is an early developing symptom of ASD (Bruckner & Yoder, 2007; Elison et al., 2014; Ozonoff et al., 2008; Watt, Wetherby, Barber, & Morgan, 2008). For example, Ozonoff and colleagues (2008) found that 12-month-old infants that were later diagnosed with ASD explored objects differently, such that they spent more time rotating, spinning, and engaging in unusual visual exploration compared to infants with delayed or typical development. In contrast, Elison et al. (2014) suggest that while 12-month olds later diagnosed with ASD engaged in repetitive object manipulation, high risk infants (i.e., infants with a sibling diagnosed with ASD) who were not later diagnosed with ASD showed the same amount of repetitive object manipulation. Thus, this symptom did not differentiate between diagnostic groups. Together, this suggests that repetitive object use may be developmentally representative of subclinical ASD characteristics in infants rather than specific to ASD at this young age.

Overall, this literature is compelling in providing evidence that infants and toddlers later diagnosed with ASD exhibit RRBs more than their typical counterparts. However, it is necessary to assess these behaviors in ASD as compared to those with DD in order to understand whether differences in RRBs in ASD is due to general developmental delay or

whether it is specific to ASD. Studies have suggested that while children with DD also exhibit RRBs similar to those with ASD, they are lower in frequency and intensity compared to those with ASD and these group differences increase with age and development. For example, At 12 months, retrospective parent report suggests that children with ASD showed significantly more ASD symptoms than those with DD (Watson et al., 2007). This report from the First Year Inventory (FYI) examined specific items that targeted RRBs indicating that those with ASD were getting "stuck" on an activity or part of toy more often than those with DD at 12 months suggesting an "insistence on sameness" in activities as well as a "repetitive use of objects" in ASD at a very early age. At 18-24 months, differences in RRBs continue to be present, particularly with regards to repetitive and stereotyped behaviors with body and objects in toddlers with ASD compared to those with DD (Morgan, Wetherby, & Barber, 2008). When children with ASD and DD are compared at 28 months, those with ASD continue to exhibit more RRBs than those with other delays. For example, Kim and Lord (2010) examined RRBs in ASD and non-ASD delays (e.g. language delay, mild intellectual disability, global developmental delay) based on scores from the Autism Diagnostic Observation Schedule (ADOS). Results suggest that while the ASD group showed the higher rates of RRBs, the non-spectrum delays group also exhibited some RRBs.

Joseph, Thurm, Farmer, and Shumway (2013) found that young children (mean age of 4 years) with ASD exhibited significantly more RRBs including compulsive behaviors, restricted interests and stereotyped behavior than those with DD. As children with ASD and DD get older, the differences in their symptoms and behavior become more and more apparent indicating that the presence of RRBs early in childhood are not only a function of a

developmental delay. In this same study, those with ASD continued to show higher levels of RRBs on all subscales of the RBS-R compared to those with typical development.

Summary

Overall, this literature shows that infants and toddlers with ASD show higher rates of RRBs compared to those with typical development suggesting that RRBs are a defining characteristic of ASD in the first few years of life and can be a useful tool to assist in ASD diagnosis during toddlerhood. The differences in symptomatology between ASD, DD, and typical development throughout infancy and toddlerhood suggest a different developmental pattern of RRBs in ASD.

Developmental Theories of ASD

For decades, researchers have tried to identify an underlying theory behind ASD symptom development during infancy and toddlerhood. Two primarily symptom categories have been clearly identified characterized: (1) impairments in social communication and interaction and (2) the presence of RRBs. Studies suggest that both of these symptom categories are present early in life in infants and toddlers later diagnosed with ASD. Further, these symptom categories persist into early childhood and beyond. However, it is not clear how the development of these two symptom categories relate to one another in early development. Is there a core underlying impairment that precedes, and possibly, links these symptom categories? Or do both symptom categories develop in parallel to one another? Or, does one symptom area clearly precede the other and in turn, effects the development of the second symptom category? Two primary theoretical models have been proposed in the ASD literature. These theories suggest an alternative explanation for a core impairment in ASD and alternative theories about how these broad symptom categories interact with each other.

Social Motivation Theory

Broadly, the Social Motivation Theory of ASD suggests that individuals with ASD have less motivation to attend to social information in their environment from a very early age. As a result, young children with ASD spend less time engaged in social interactions and their social communication and interaction skills develop atypically during the first years of life. Because of this decreased social motivation, engagement is instead directed towards objects, which, in turn, leads to atypical use of objects or repetitive use of objects (see Chevalier, Kohls, Troiani, Brodkin, & Schultz, 2012; Dawson et al., 2004).

Social Motivation theory has primarily focused on individual's attention to social information, particularly faces and people during early childhood. One of the hallmark features of ASD is impaired or atypical use of eye contact (Mundy, Sigman, Ungerer, & Sherman, 1986; Senju & Johnson, 2009) and difficulties interpreting others facial expressions (for reviews see Dawson, Web, & McPartland, 2005; Gremiel et al., 2014). In infancy, researchers hypothesize that this begins as decreased motivation to look at faces and social information in the environment.

Several studies have suggested impairments in attending to social information at 6 months of age in infants later diagnosed with ASD (Jones et al., 2016; Chawarska et al., 2012). Jones and colleagues (2016) examine the possible underlying cognitive and neural impairments in ASD starting at 6 months by examining infants' engagement and attention to social stimuli. Results indicated that at 6 months, infants later diagnosed with ASD had delayed sensitization to social stimuli and spent less time looking at social stimuli compared to infants that were not later diagnosed with ASD.

To date, though, there is not a clear understanding of how this decreased attention and interest in social information is related to behavioral symptoms occurring later in development. It is hypothesized that this early social impairment has a cascading effect that leads to the core social impairments observed in ASD, although there is no clear evidence on the progression of this effect. Based on this theory, reduced dyadic engagement with faces in the first few months of life leads to impairments in triadic interactions and increased focused on objects during the first two years of life.

This increased focused on objects may show a cascading effect with regards to the development of RRBs. It has been suggested that infants with ASD have a *heightened* interest in objects rather than what may otherwise be perceived as a *disinterest* in people and social interactions (e.g., Bruckner & Yoder, 2007; Ozonoff et al., 2008). For example, twelve-month old infants later diagnosed with ASD spend significantly more time exploring objects (i.e. visually inspecting) in an unusual way compared to infants with typical and delayed development (Ozonoff et al., 2008). This suggests that at a young age, individuals with ASD are paying attention to and visually exploring objects in a fundamentally different way. Their attention to objects is different such that it may impair their ability to appropriately explore their environment to understand the true function of object and focus their attention on meaningful, functional stimuli. In addition, Ozonoff et al. (2008) found that these infants also spent more time rotating and spinning objects, an atypical use of the presented objects, compared to the other groups. This suggests that in addition to the presence of restricted object use early in life, there is also an atypical focus on objects that may, in turn, influence an infant with ASD's further exploration of their environment, including social information. However, research has yet to determine whether the lack of

engagement with people or social stimuli corresponds directly with heightened focus on objects. While many studies examine behavior during the first year of life, there is then not a clear intermediary assessment of behavior prior to diagnosis at 2-3 years of age. It is necessary to examine the cascading effects of decreased social engagement through longitudinal studies with repeated measures of social engagement and core impairments related to ASD.

Attention theory

In contrast, attention theory proposes that individuals with ASD have an underlying impairment in general attention focus and engagement across both social and non-social information. This atypical development of attention interferes with an individual's ability to appropriately engage with both social and nonsocial information. Specifically, it is hypothesized that infants with ASD have "sticky attention" such that they have difficulty disengaging and shifting their attention (Landry & Bryson, 2004; Sacrey et al., 2013).

From 6-12 months, children with typical development begin to spontaneously disengage and shift their attention between objects and people. That is, after first engaging their attention with an object or person (i.e., they look at it), they then *disengage* their attention from that object or person and then *shift* to something new to attend do (e.g., Carpenter et al., 1998). Researchers have proposed that this process of disengagement of attention is impaired in ASD, although results have been mixed. Some studies suggest that disengagement and shifting of attention is impaired in children diagnosed with ASD as well as infants and toddlers later diagnosed with ASD (Elsabbagh et al., 2009, 2013; Sacrey et al., 2013) while others suggest no impairments in attention (Fischer et al., 2015).

This mixed literature may be, in part, due to the age of participants studied. Few studies have examined attention shifting in infants who are later diagnosed with ASD. Studies with older children and adults with ASD typically do not find significant differences in saccadic reaction time and latency to disengage and shift attention during visual disengagement tasks (see Sacrey et al., 2014 for review). For example, adults with ASD show similar reaction time to adults with typical development on Gap and Overlap trials in a Gap-Overlap task (Kawakubo et al., 2004). Similarly, Fischer and colleagues (2013) suggest that children with ASD ages 5-12 perform similarly to their peers with typical development during a disengagement task. Studies with preschool-aged children tend to show longer reaction times in disengaging attention from a central stimulus and shifting to a new stimulus in children with ASD compared to those with typical development and those with Down syndrome (Landry & Bryson, 2004). This suggests that impaired ability to disengage and shift attention may not be a byproduct of developmental delay but rather a true attention difficulty in young children.

Evidence for impaired disengagement of attention in infants at-risk for developing ASD continues to be mixed, although with more studies suggesting impairments in attention and disengagement. Elsabbagh et al (2009) found that at 9-10 month old infant siblings of children with ASD took longer to disengage their attention compared to infants without siblings with ASD. At 14 months, this effect differentiated those later diagnosed with ASD in that they were significantly slower to disengage their attention in a standard gap-overlap task (i.e., nonsocial task) compared to those at-risk for ASD that were not later diagnosed with (Elsabbagh et al., 2013). In slightly older toddlers, this effect was not as clearly present (Fischer et al., 2015). Their results suggested that newly diagnosed children with ASD

between age 21 and 37 months performed similar to children with typical development. The authors suggest that sticky attention is not an underlying impairment in ASD nor is it a causal factor in the development of ASD in early childhood.

Sacrey, Bryson, and Zwaigenbaum (2013) examine the process of disengagement of attention when grasping objects in a naturalistic play setting from 6-36 months in infants at high-risk for ASD. By 12 months of age, children later diagnosed with ASD took longer to disengage their attention compared to those that were low-risk and/or did not receive a diagnosis of ASD. This effect persisted through the second year of life suggesting that the longer latency of disengagement of attention is present through the second year of life. While attention disengagement is a critical component of understanding attention impairments, the next step in the process is to examine what happens *after* a child disengages their attention from an object. Are they shifting their attention to another object or person in the environment? Are they disengaging their attention and then their gaze wanders for a period of time before then re-engaging their attention with something else?

Differences in attention to social information, specifically faces, are clear from an early age, although the underlying function continues to be uncertain. Jones and Klin (2013) suggest atypical visual attention to faces by 6 months in infants later diagnosed with ASD and varied trajectories of attention to faces in a longitudinal study of infants at-risk for ASD. It is possible that this type of atypical attention may be present in attention to both objects and social information in early development of infants and toddlers with ASD as there is evidence of atypical attention to both people and objects in infants and toddlers with ASD.

In a naturalistic setting, Swettenham and colleagues (1998) explicitly examined attention shifting in toddlers with ASD (age 20 months), noting that they had fewer shifts of

attention between two people and between people and objects compared to those with developmental delay and children with typical development. Attention shifting is decreased in ASD within the context of social information. Another study examined attention shifting during a play interaction where parents were asked to engage face-to-face with their child, then to hold their face still for 2 minutes, and then re-engage in a face-to-face interaction (Ibanez, Messinger, Newell, Lambert, & Sheskin, 2008). Infant siblings of children with ASD shifted less frequently and spent longer looking away from parents' faces compared to infant siblings of those with typical development. Together, this highlights the importance of both sticky attention *and* attention to social information including people and faces in the environment when investigating long-term developmental trajectories.

Thus, some evidence points to broad impairments in attention, rather than attention differences that are specific to social information (i.e., social motivation theory); specifically, attention shifting may be impaired in both social and non-social settings. These impairments in attention to non-social information may be a precursor to the development and presence of RRBs in children with ASD. That is, repetitive behaviors, particularly repetitive motor behaviors and repetitive use of objects may be a manifestation of sticky attention. Indeed, spending more time looking at objects compared to people is related to increased repetitive behaviors in children and adolescents with ASD (Sasson, Turner-Brown, Holtzclaw, Lam, & Bodfish, 2008). Young children later diagnosed with ASD get their attention "stuck" on a particular object or motor movement and have difficulty disengaging their attention from that specific behavior (Stronach & Wetherby, 2014). In turn, there is a reinforcement of that behavior and a lack of integration of other parts of the environment while they are overly focused on a movement or object. A child not later diagnosed with ASD may engage in the

same behavior one or two times before shifting their attention to something else or looking at a person whereas a child later diagnosed with ASD may continue to engage in a repetitive behavior several times before shifting their attention to something else in their environment. The Attention theory suggests that there is a cascading effect of ASD symptomatology due to an underlying impairment in attention more broadly.

Impairments in attention and the presence of sticky attention appear to be present early in development for children later diagnosed with ASD, particularly when examining social information (i.e., faces). These impairments may have cascading effects on the development of social communication as well the increased presence of RRBs in ASD. However, literature is mixed as to whether this impairment persists across development through childhood and adulthood. A longitudinal study is needed to look at the developmental unfolding of attention shifting and sticky attention. Further, attention needs to be examined in relation to both social and nonsocial information as much of the research focuses more exclusively on social information.

Issues on Research in Developmental Trajectories in ASD

One of the most difficult aspects of identifying early developmental trajectories in ASD is that the average age of ASD diagnosis is 4.5 years of age (Baio, 2014). While ASD can be diagnosed as early as 24 months, it is rare that children are identified within the first two years of life and thus research on the early emergence of ASD symptoms is difficult to conduct. Therefore, the majority of studies attempt to determine infants that are "at-risk" for a later ASD diagnosis and track their development during infancy and early childhood before being able to identify their diagnosis.

One way to identify infants at-risk for a later diagnosis of ASD is through infant siblings of children with ASD. These infants are identified to be at high-risk for ASD based on the strong underlying genetic component in ASD as evidenced by the higher rates of ASD in siblings of those with ASD compared to those with siblings without a diagnosis of ASD (Ozonoff et al., 2011). In these prospective studies, infant siblings of children with ASD and infant siblings of children with typical development have been evaluated several times during the first 3-5 years of their life (Elison et al., 2014; Landa & Garrett-Mayer, 2006; Landa, Gross, Stuart, & Faherty, 2013; Ozonoff et al., 2010, 2011; Wolff et al., 2014; Zwaigenbaum, Bryson, Rogers, Roberts, Brian, & Szatmari, 2005). Patterns and signs of development are identified and then tracked across time to determine which are predictive of a later diagnosis of ASD or other developmental delays during toddlerhood and early childhood. These studies provide insight into very early developmental processes that are present prior to an official, behavioral diagnosis of ASD.

While using infant-sibling of children with ASD is very helpful in being able to capture individuals that have a higher likelihood of being diagnosed with ASD, the results are indicative of early signs of ASD in families with a genetic risk for the disorder. Whether these same early symptoms are present in infants without a genetic risk is not yet clear. Zwaigenbaum and colleagues (2007) discuss several additional drawbacks to prospective studies of infant siblings. For example, once a parent has one child already diagnosed with ASD, their behavior may change or be fundamentally different from children who do not have a sibling or do not have a sibling with ASD. There is also an increase in parent stress among parents with one child with ASD and again may not be representative of population of children without a sibling with ASD.

While prospective infant sibling studies of ASD have been integral in building our understanding of the development of ASD symptoms; it is necessary to evaluate community-based samples of children at-risk for ASD in order to maintain a truly representative sample of individuals with ASD. Community-based samples of infants at high-risk for ASD are able to identify a larger group of children at-risk for ASD that may or may not have a clear genetic and family history of ASD. These types of samples provide a broader perspective on a representative ASD population as only a portion of individuals with ASD have an older sibling with ASD. However, utilizing community-based samples of children at-risk for a later diagnosis of ASD presents its own challenges. First, many of the early behavioral signs of ASD are not present until 9-12 months of age (Ozonoff et al., 2008; Dawson et al., 2004) or later in the second year of life (Wetherby et al., 2004, 2007). This makes it difficult to determine developmental trajectories starting closer to birth. Further, given the rate of ASD is 1 in 68 individuals, community-based samples require high volume of screening within the larger community in order to identify a large sample of children at-risk for ASD. Even with the need for an extensive and high-volume screening process, research utilizing community-based samples provides exceptional opportunities to understand the symptoms of ASD and their associated developmental trajectories within the broader population.

Present Study

The present study aims to compare two developmental theoretical models about the emergence of ASD across the first two years of life (e.g., social motivation and attention). Specifically, the study utilized a community-based sample of children identified at 12-months as being at high-risk for ASD who participated in comprehensive evaluations at 13 months, 22 months, and 3-5 years of age to track early developmental change and establish

any appropriate diagnosis during the preschool years. Relevant social engagement, attention shifting, and RRBs were coded from video-recorded assessments at 13 and 22 months to track behavioral symptoms in a naturalistic environment. The ability to measure social engagement, attention shifting, and RRBs as they unfolded across a two to three year developmental period allowed for a direct comparison of these two theories of early development in ASD.

Social Motivation Theory Aims:

Social motivation theory suggests that during the first year of life, infants with ASD have a decreased preference for social information that leads to the development of ASD symptomatology across the first few years of life. If the early unfolding of ASD symptoms is related to decreased social motivation, it is hypothesized that decreased looking at people (i.e., decreased social engagement) at 13 months will predict the later development of RRBs and decreased attention shifting at 22 months, which together will predict later ASD diagnosis at, age 3-5 years. See Figure 1.

Attention Theory Aims:

Atypical development of attention suggests that infants with ASD get "stuck" on objects and people and have difficulty disengaging and shifting between objects and people. Thus, it is predicted that infants with ASD will show fewer shifts in attention and that this over focused attention will be associated with increased RRBs. This over-focused attention will result in decreased engagement with social information and triadic interactions (e.g., decreased sharing of attention) and the eventual diagnosis of ASD. Based on this theory, I hypothesize that at 13 months decreased attention shifting and the presence of RRBs will predict later impairments in social

engagement (i.e., looking at people) at 22 months, which together will predict later ASD severity at 3-5 years. See Figure 1.

Exploratory Aim:

This sample of high-risk infants was randomized to receive 1 hour per week of in-home treatment from 13-22 months of age with half receiving the treatment and the other half being in a "services as usual" group. Early data analyses have suggested that the treatment did not significantly change the children's cognitive ability, ASD symptoms, or adaptive behavior at 22 months. However, it is not yet clear whether treatment had long-term effects on ASD symptoms or diagnosis. Before evaluating the developmental theories of ASD as described above, the present study will examine whether participation in treatment during the second year of life significantly predicted ASD symptoms at 3-5 years of age.

CHAPTER 2: Methods

Study Design and Participants

Participants from this study were from the Early Development Project-2 (EDP2), a longitudinal study conducted at the University of North Carolina-Chapel Hill (PIs: Linda Watson, Elizabeth Crais, Lauren Turner-Brown, Grace Baranek, Steve Reznick). This study examined ASD symptoms in a community sample of infants though the use of a caregiver survey, the First Year Inventory (FYI; Watson et al., 2007). The FYI is a 63-item parent report screener used to identify infants at risk for ASD or atypical development at 12 months of age. Using birth records, EDP2 conducted a mass mailing of the FYI to 60,237 families with 12-month old infants in the Triad region of North Carolina (e.g., Wake, Orange, Chatham, Durham, Alamance, and Guilford Counties in the Chapel Hill/Raleigh and Greensboro area were included in these mailings). In total, 14.5% or 8,717 packets were returned with a completed FYI form. Three percent (i.e., 280 families) of the returned forms indicated high scores on the FYI suggesting atypical development or higher risk for ASD. All of the 280 families were called and invited to participate in a research study just after the child's first birthday. Ninety-six of the 280 toddlers participated in an in-person evaluation at 13-16 months (M=13.78 months, SD=.77; Time 1). Eighty-six of the children were again evaluated 9 months later, prior to their 2nd birthday (M=22.55 months, SD=.90; Time 2). Group differences at Time 1 and Time 2 based on diagnosis at Time 3 are presented in Table 1.

Forty-six children were evaluated again at Time 3 to determine diagnostic outcome (M=54.04 months, SD=10.93). Within this final sample, 68.1% are male, 78.7% were White, 12.8% were African-American, 2.1% were Asian American, and 8.5% were mixed race. Some previous studies utilizing community-based samples of children at-risk for ASD or other communication delays had higher rates of male participants (83-89% males) and similar rates of Caucasian vs. non-Caucasian participants (e.g., Wetherby et al., 2004). However, participants from Wetherby and colleagues (2004) were recruited from childcare and healthcare agencies rather than through birth records.

Diagnostic Outcome

Diagnoses and clinical feedback were provided to participants at the Time 3 evaluations. Diagnoses included ASD, Intellectual Disability (ID) or developmental delay (DD), Language Disorder or Language Delay (LD), Attention Deficit Hyperactivity Disorder (ADHD), Anxiety disorder, and/or Social Pragmatic Communication Disorder (SPCD). Diagnostic categories were grouped into 3 categories including "no concerns," "some concerns," and "significant concerns" or meets criteria for full diagnoses. Of the 46 children evaluated at time 3, 16 received a diagnosis of ASD while 18 had no concerns about ASD symptoms. Twelve children presented with some symptoms and concerns regarding ASD, but they did not meet clinical criteria for an ASD diagnosis. In the analyses described below, ASD diagnosis is discussed as a binary variable in that those with some concerns but no diagnosis are grouped with those with "no concerns."

Three of the children with an ASD diagnosis demonstrated significant cognitive and developmental impairments and were also diagnosed with ID. These three children also all had suspected and/or diagnosed genetic disorders. Given the difficulties in separating the

effects of genetic disorders and ID compared to ASD, these 3 children were excluded from analyses. Recent population studies indicate that approximately 38% of children with ASD also have comorbid diagnoses of ID (Baio, 2014). Given the small number of children with ID in the present study, the ASD sample is not representative of the larger ASD population variation in intellectual ability. In addition, no children without ASD were diagnosed with ID and thus there was not an appropriate comparison sample in the non-ASD group. As a result, 13 participants with ASD were included in the final sample and are considered to have high-functioning ASD as evidenced by their average to above-average cognitive abilities at Time 3.

Eight children were diagnosed with a language delay or disorder separate from a speech articulation concern and 34 did not have any language delay. Four children had concerns for language delay although these delays did not significantly interfere with their language and communication. With regards to ADHD, one child was diagnosed with ADHD while 6 children had some symptoms of ADHD and 39 children had no concerns. Given that many children were not yet in a school setting and were under age 5, a complete clinical diagnosis could not be provided. Two children were exhibited significant clinical symptoms of anxiety while nine children had some concerns or symptoms and 35 children had no concerns. Last, one child had significant concerns and symptoms of SPCD, three children had some concerns, and 39 children had no concerns. Overall, 43 children were included in the final sample for this study - 16 children had significant symptoms or met clinical criteria for at least one of the previously mentioned diagnoses (with 13 of the 16 having ASD) and 27 children and no concerns or some concerns. Group differences between those diagnosed

with ASD without comorbid ID and those without ASD or ID are presented in Tables 1 and 2.

Measures

Communication and Symbolic Behavior Scale (CSBS; Wetherby & Prizant, 2002). The CSBS is a standardized 20-30 minute assessment to assess children on their social communication skills between 6 and 24 months. This assessment includes 6 different sampling opportunities to elicit a variety of social and communication skills with different objects. The objects used during the sampling opportunities are a balloon, a windup toy, bubbles, jar with cheerios, books, and pretend play toys (e.g., bowl, spoon, cup, doll). In each sampling opportunity, the examiner presents a toy to the child by making the toy work as it is intended (e.g., making the windup toy work). The examiner then waits for the child to respond by requesting or refusing another turn with the toy. During the CSBS, the child is placed in a seat that attaches to a table and the examiner and a caregiver are on either side. As a result, the child generally stays in one spot and their behavior can be consistently monitored throughout the assessment. This assessment was completed by trained clinicians at Time 1 and Time 2 and includes a broad, global score to determine risk status for communication and social delays. Average standard scores for the CSBS are 100 with a standard deviation of 15 suggesting appropriate development and skills in the areas of communication and symbolic play and behavior for their chronological age. Lower scores indicate delays related to social communication and play skills. The average standard score for the present sample was 88.55 (SD=13.03) at Time 1 and 91.76 (SD=17.61) at Time 2 suggesting that participant scores on the CSBS were in the low average range at Time 1 and average range at Time 2. In order to better understand the developmental trajectories of social engagement, attention

shifting, and RRBs proposed in this study, more detailed video coding is needed. Coding activities for measuring social engagement, attention shifting, and RRBs are discussed below.

Social Motivation. Social motivation was defined as social engagement through looking at people during the CSBS evaluation. Social motivation was measured at Time 1 and Time 2 through video coding of the CSBS. Social motivation was measured as the duration of eye gaze towards people (e.g., the examiner and caregiver). Specifically, the child's eye gaze patterns were coded to measure where they spent time looking and for how long they fixated on any part of that person (i.e., face, body or hand).

The onset and offset of the child's gaze for each person was coded throughout the entire assessment. From this data, several composite variables were computed for analyses. First, the total amount gaze time for looking at people was calculated. Total time calculated did not include the time of the assessment where the child was not visible on the screen and therefore their gaze behavior could not be coded. The social engagement variable of Looking at People was calculated as a proportion of total time in order to account for variability in total assessment time.

Attention Shifting. Instances of attention shifting were coded when a child was fixated on one object or person and then shifted directly to another object or person. A shift was only coded when there was a clear disengagement from one thing or person followed immediately by a clear fixation of their attention on something or someone else. A shift was *not* coded when the child disengaged their attention from an object or person but did not fixate their gaze on another object or person. That is, a shift was *not* coded if they were looking at an object and then their gaze wandered around the room for several seconds. Shifting was calculated as an overall proportion with composite variables measuring number

of shifts per minute in order to account for differences in total assessment time. Three shifting composite variables were calculated with one including all shifts and then one variable measuring Social-only Shifting (i.e., a shift that included another person) and Non-social Shifting (i.e., a shift between objects only).

Video Coding. The social engagement and attention shifting video coding was conducted using ELAN 4.6.1 (Sloetjes & Wittenberg, 2008; http://tla.mpi.nl/tools/tla-tools/elan/). This coding software allows for detailed time-based coding that accurately measures duration of gaze and can mark instances of other relevant behaviors. See attached appendix for coding scheme. While the CSBS is a standardized assessment measure, the length of the assessment varied from 20-30 minutes. In order to account for this variability, composite variables for looking time were converted into proportions of the total assessment time.

Videos were not coded if the CSBS was not recorded during the assessment session or if less than half of the sampling opportunities were not recorded or if the child was not visible during more than half of the activities. In total, 42/43 (98%) CSBS videos from time 1 and 40/43 (93%) videos from time 2 were coded. A second coder, blind to participant diagnosis and treatment group and did not participate in any of the diagnostic evaluations, coded 10 videos (15%) in order to establish reliability and consistency. Five videos coded for reliability were from time 1 and 5 were from time 2. Videos for reliability were chosen randomly with some exceptions. Because the author (and primary coder) completed several diagnostic evaluations at time 3 and was not blind to diagnosis, 4 videos were chosen from participants that the author was more familiar with (i.e., the author completed their diagnostic evaluations) in order to account for the author's potential bias during coding. Reliability was

calculated for all composite variables using Intraclass Correlations Coefficients (ICC) and exhibited excellent interrater reliability for Looking at People (.99), and Total Attention Shifting (.94).

Repetitive and Sensory Movement Scale (RSM; Wetherby & Morgan, 2007). RRBs were measured at Time 1 and Time 2 using a coding protocol developed for use with infants and toddlers participating in the CSBS (Wetherby & Morgan, 2007). This scale has been used in several different studies with young children and has differentiated between RRBs in young children with ASD compared to those with typical development (Damiano et al., 2013; Elison et al., 2014; Morgan et al., 2008; Watt et al., 2008).

This coding scheme examined 2 separate areas of RRBs: repetitive movements with body and repetitive or stereotyped movements with objects. In each sampling opportunity (e.g., each set of toys presented), the coder recorded the frequency of each type of repetitive behavior with body and/or objects. In addition, the coder recorded what type of behavior occurs within each category. For repetitive movements with body, behaviors included flapping arms or hands; patting, tapping, or pressing body part; rubbing body part; and stiffening fingers, hands or arms. For repetitive or stereotyped movements with objects, behaviors were categorized into preoccupation in intensity or focus with a particular object and insistence on sameness or difficulty with change in an activity. For the preoccupation category, behaviors included swiping object away; rubbing or squeezing an object; rolling or knocking over an object; rocking, flipping, or turning over an object; and spinning or wobbling an object. For the insistence on sameness category, behaviors included collecting objects; moving or placing objects to one location; lining up or stacking objects; and clutching objects.

Variables were calculated based on the total frequency of behaviors with 3 variables being considered. There are separate total frequencies for RSM with Body, RSM with Objects, and Total RSM. Total RSM was used as the primary variable for overall RRBs in this sample.

Videos were included in RSM coding if all activities were completed and were recorded on video. Some videos included only partial recordings of the CSBS and were not included because the composite RSM variables were total frequency rather than a proportion. In total, 80 tapes were coded with 40 videos (93%) at Time 1 and 40 videos (93%) at Time 2). The videos not coded from Time 1 and Time 2 were not from the same participants. Each participant had a CSBS tape at Time 1 or Time 2 or both. All coding was completed by the author after consulting with Dr. Lindee Morgan, one of the authors of the coding scheme (Wetherby & Morgan, 2007). Coding was completed by hand while watching videos using Windows Media Player. A second coder, blind to diagnostic outcome and treatment group, coded 20 videos (25%) for reliability. Intraclass correlation coefficients indicated good to excellent reliability for all measures (RSM Total=.89; RSM Body=.96; RSM Object=.86). See attached appendix for RSM coding scheme.

ASD Diagnosis. The Autism Diagnostic Observation Schedule-Second Edition (ADOS-2; Lord et al., 2012) was used as a behavioral measure of ASD symptoms at Time 2 and Time 3. The ADOS-2 consists for 5 modules and is a semi-structured play-based assessment that is used to evaluate social communication or social affect, overall play behaviors, and the presence of RRBs. Scores are calculated in 2 broad areas of Social Affect and Restricted and Repetitive Behaviors and Interests (RRB) based on the observations during the ADOS. A total sum of these two categories accounts for the total ADOS score.

Modules are selected based on the child's language ability and age. Module 1 is for children who are pre-verbal or use single words and was administered to all participants at Time 2. While formal diagnoses were not provided at Time 2, a total ADOS score was calculated as a measure of ASD symptom severity. A total score of 7 or higher on a Module 1 indicates significant symptoms of ASD.

A licensed psychologist or speech pathologist provided ASD diagnoses using behavioral diagnostic measures (i.e., ADOS-2), parent interview, as well as clinical judgment. Participants were administered either a Module 1, 2, or 3 based on their language ability and age. Module 1 (pre-verbal or single words) was administered to 2 children; Module 2 (phrase speech) was administered to 20 children, and Module 3 (fluent speech for children above 48 months) was administered to 20 children. From the total ADOS-2 score, an overall severity or comparison score for ASD symptoms was computed on a 1-10 scale with higher scores indicating more severe ASD symptoms. This severity score allows for appropriate comparison across all modules for all participants at Time 3. This score was used as a continuous measure of ASD symptom severity across all children at Time 3.

Mullen Scales of Early Learning (MSEL; Mullen, 1995). The MSEL is a measure of cognitive ability for children from birth to 68 months and was administered at Time 1 and Time 2. A Developmental Quotient (DQ) Standard Score provides and overall score for cognitive ability in young children. The DQ consists of the Visual Reception, Fine Motor, Expressive Language, and Receptive Language subscales. These subscales are calculated as T-scores with a mean of 50 and standard deviation of 10. An overall Early Learning Composite Standard Score represents overall developmental ability with a mean of 100 and

standard deviation of 15. Group differences for the ASD compared to the non-ASD group at Time 1 and Time 2 can be found in Table 1.

Differential Ability Scales-2nd Edition (DAS-2; Elliot, 2007). The DAS-2 is a measure of IQ for children ages 2 years 6 months through 17 years 11 months. This study used subtests for the DAS-2 Early Years as a measure of intellectual ability at Time 3. While Time 1 and Time 2 used the MSEL as a measure of developmental and cognitive ability, it was not appropriate to use at Time 3 given that participants were older and may hit the ceiling of the MSEL without providing a true representation of their abilities. The DAS-2 has a wider age-range for administration and provides a IQ score rather than a developmental quotient (DQ) and thus may be more representative of the child's true abilities. The DAS-2 provides a Verbal IQ, Nonverbal IQ, Spatial IQ, and General Conceptual Ability (GCA). Overall, the children seen at Time 3 and included in the present study were in the average range for intellectual ability (GCA M=101.91, SD=15.99) indicating that across all participants, the majority were in the average range of functioning suggesting that this sample of children with ASD is considered "high-functioning."

Repetitive Behavior Scale-Revised (RBS-R; Bodfish et al., 2000). The RBS-R is a 43-item caregiver report measure on the occurrence and interference of repetitive behaviors and restricted interests. At time 3, the RBS-R provides information about the child's current repetitive behaviors using parent report. The RBS-R is divided into 5 subcategories providing separate scores for stereotypies, compulsions, self-injurious behavior, restricted interests, and ritualistic and sameness. In addition, a total score is calculated. For all subscales and the overall scale, higher scores indicate more frequent RRBs and a higher level of interference in the child's day-to-day life. The RBS-R provides an additional measure of RRBs at Time 3

above and beyond scores from the ADOS-2. Because the ADOS-2 is only 45-60 minutes of a child's behavior, it may not truly capture many RRBs that occur in other environments. The RBS-R allows for further assessment of RRBs across environments based on parent report.

Treatment

After Time 1, participants were randomized to participate in a specific treatment called Adapted Responsive Teaching (ART) or participated in community Services as Usual (SAU) for 6 months (see Baranek et al., 2015 for description of ART from a pilot study). ART is a home-based intervention that works with parents and their young children using a responsive teaching curriculum. The 2 primary goals of ART were related to social-communication and sensory regulatory behaviors. For each goal, different behaviors were targeted using strategies across 5 domains (reciprocity, contingency, control, affect, and match). Of the children participating in the current study, seen at time 3, 61.7% (n=29) participated in ART and 38.3% (n=18) received SAU. There were no main effects of ART for social communication, sensory-regulatory, or adaptive behaviors (Watson et al., in preparation) in the larger study (e.g., 29 receiving ART; 19 receiving SAU). However, given that treatment was provided during the ages in which the present study examined developmental trajectories, exploratory analyses were conducted to determine whether treatment is a significant predictor of outcome variables.

CHAPTER 3: Results

Data Analysis Plan

Descriptive and preliminary analyses were conducted using Statistical Package for Social Science Version 23.0 (SPSS; IBM, 2015). To more thoroughly examine the competing theories on the role of social motivation and attention in predicting ASD symptom severity, two path analyses were conducted using MPlus 7 (Muthén & Muthén, 2012). The models used a maximum likelihood estimation. Model fit was determined using several specifiers including Chi Square Test of Model Fit, the Root Mean Square Error of Approximation (RMSEA), the Comparative Fit Index (CFI), and Tucker Lewis Index (TLI). RMSEA values less than .05 and CFI and TLI values greater than 0.9 are considered to be excellent fit (Kelloway 2015; Tabachnick & Fidell 2013).

To assess the Social Motivation Theory, we examined whether low levels of looking at people at Time 1 predicted Sticky Attention (i.e., as measured by decreased attention shifting or increased repetitive motor behaviors with body and objects) at Time 2, which in turn predicted higher ASD symptom severity at Time 3 (see Figure 1). To assess the Attention Theory, we examined whether higher levels of Sticky Attention (i.e., decreased attention shifting and increased repetitive motor behaviors with body and objects) at Time 1 predicted less time Looking at People at Time 2 and then predicted increased ASD symptom severity at Time 3 (see Figure 1). The relationships between the same variables at Time 1 and Time 2 (e.g., Time 1 Looking at People and Time 2 Looking at People) as well as the relationship between the variables occurring at the same time (e.g., Time 1 Attention Shifting

and Time 1 Looking at People) was included in the model (solid arrows in Figure 1). Preliminary analyses were conducted to determine the relationship between RRBs and attention shifting as the Attention Theory assumes they are related to one another. If these two variables are not related to one another, then it is unlikely they are measuring the same construct and will be included in larger path analysis models simultaneously. Path analyses were conducted initially using the overall composite measure of overall attention shifting (e.g., combined social and non-social shifts). Next, models were conducted using the separate social and non-social shifting measures in order to determine whether shifting involving social information differed in its role compared to shifts that did not include social information. See Figure 1 for a schematic of the proposed analyses.

Analysis of CSBS Measures of Social Motivation and Sticky Attention Variables

Several variables were calculated to measure and assess Social Motivation and Sticky Attention. For Social Motivation, Looking at People was a measure of social engagement calculated from the proportion of total time the child spent looking at a person (face, hand, body). Attention Shifting was calculated based on total time and represents the average number of attention shifts per minute. This variable includes both shifting between social and non-social information (i.e., objects and people). Composite variables were also calculated that separated social and non-social attention shifting. That is, Social Shifting includes any attention shift with a person and Non-social Shifting includes attention shifts only between two objects. Last, the total frequency of RSM behavior was used as a measure of overall repetitive behaviors within the CSBS. As previously stated, the total RSM was only calculated if the video included all 6 opportunities thus there are fewer children with RSM

data across both time points. The Total RSM consists of both RSM with Body and RSM with Objects.

Overall means at Time 1 and Time 2 for measures of Social Motivation and Sticky Attention can be seen in Table 3. Means based on diagnostic group identified at Time 3 can be seen in Table 4. Paired-samples t-tests were conducted to examine significant changes in overall group behavior from Time 1 to Time 2. Proportion of time Looking at People at Time 2 (M=9.7%, SD=4.5%) was significantly lower than at Time 1 (M=11.2%, SD=5.1%; p=.02) indicating that children spent less time looking at people at 22 months compared to 13 months of age. There was no significant change in Total Shifting and Social Shifting behavior between Time 1 and Time 2. However, non-social shifting at Time 1 (M=1.19 per min, SD=.48) was significantly lower than non-social shifting at Time 2 (M=1.43 per min, SD=.50, p=.04) suggesting that while overall shifting remained the same, children had higher rates of attention shifting between objects at 22 months compared to 13 months of age. Both RSM with Body and Total RSM were significantly higher at Time 1 compared to Time 2 (RSM Body Time 1: M=2.78, SD=2.44; Time 2: M=1.23, SD=1.50; p<.001; RSM Total Time 1: M=5.15, SD=3.48; Time 2: M=2.92, SD=2.80; p<.001). There was no significant difference in RSM with Objects at Time 1 (M=2.38, SD=2.42) and Time 2 (M=1.69, SD=1.87; p=.24).

Between-group differences. Independent samples t-tests were conducted to understand differences between diagnostic groups as determined at Time 3 for Looking at People, Attention Shifting, and RSM behavior identified at Time 1 and Time 2. Results for these t-tests can be seen in Table 4. Overall, there were no significant group differences between these variables at Time 1 (all ps >.40) suggesting that at-risk children later

diagnosed with ASD did not exhibit significantly different behavior in gaze behavior, shifting, and RSM at 13 months of age (i.e., Time 1) compared to at-risk children who did not receive a later diagnosis of ASD. However, at Time 2 children later diagnosed with ASD had lower rates of total attention shifting *(M*= 2.67 per min, *SD*=.89) compared to children not receiving an ASD diagnosis (*M*=3.96, *SD*=1.04; *t*(37)=3.63, *p*=.001) and lower rates of social-only attention shifting (ASD: *M*= 1.43 per min, *SD=.91;* No ASD: *M*=2.46, *SD=1.08*; *t*(37)=2.95, *p*=.006). There were no significant group differences in non-social attention shifting (ASD: *M*= 1.30 per min, *SD*=.34; No ASD: *M*=1.50, *SD*=.58; *t*(37)=1.39, *p*=.17) at Time 2. At Time 2, there were no significant differences in any measures of RSM behavior (all *p's*>.15).

Within-group differences. Paired-samples t-tests were conducted to identify significant change in Social Motivation and Sticky Attention within each diagnostic group from Time 1 to Time 2. Results of these analyses are presented in Table 4. Notably, the ASD group showed a significant decrease in Looking at People (Time 1 *M*=11.1%, *SD*=5.8%; Time 2 *M*=8.3%, *SD*=4.7%; *t(*11)=2.52, *p*=.03) and Social Shifting across time (Time 1 *M*=2.31 shifts per minute, *SD*=1.07; Time 2 *M*=1.43 shifts per minute, *SD*=.91; *t(*11)=3.09, *p*=.01*)*. The No ASD group showed a significant *increase* in Non-Social Shifting across time (Time 1 *M*=1.20, *SD*=.35; Time 2 *M*=1.50, *SD*=.58; *t(*25)=2.61, *p*=.02). Both diagnostic groups showed a significant decrease in RSM behavior with Body (ASD Group: *t(*11)=3.53, *p*=.005; No ASD Group: *t(*21)=5.38, *p*<.001). The ASD Group did not show a significant change in RSM with Objects (Time 1 *M*=2.42, *SD*=2.78; Time 2 *M*=2.23, *SD*=2.68; *t(*11)=.22, *p*=.83) but the No ASD Group had a marginally significant decrease in RSM with Objects (Time 1 *M*=2.44, *SD*=2.29; Time 2 *M*=1.36, *SD*=1.25; *t(*21)=1.90, *p*=.07).

Correlation Analyses. The primary variables used for the path analyses included Looking at People, Shifting including Social and Non-social Shifting, and RSM Total. Preliminary correlation analyses were conducted with only these experimental variables across both Time 1 and Time 2 in order to better understand their relationships. While several composite variables were calculated, these experimental variables extrapolated from the video coding of the CSBS were utilized in the path analyses discussed below.

Pearson correlations between variables used for later path analyses were conducted to describe initial relationships. Correlations between experimental variables at Time 1 and Time 2 are included in Table 5. Pearson correlations indicate strong positive relationships between Looking at People and Total Shifting. Lower levels of Time 1 Looking at People was associated with lower rates of Time 1 Total Shifting, $r(43)=.69$, $p<.001$, and Time 2 Total Shifting ,$r(39)=.39$, $p=.01$. That is, 13-month old infants who spent less time looking at people showed lower rates of attention shifting at both 13 and 22 months. There was a significant *negative* correlation between Time 1 Non-social Shifting and Time 2 Social Shifting, $r(39)=-.34$, $p=.04$, and a marginally significant *negative* correlation between Time 1 Non-social Shifting and Time 2 Total Shifting, $r(39)=-.30$, $p=.06$. That is, higher rates of Non-social Shifting at Time 1 were related to *lower* rates of Social Shifting and Total Shifting at Time 2. There is also a marginally significant *negative* relationship between concurrent measure of social and non-social shifting at Time 2, $r(39)=-.27$, $p=.06$, indicating that at Time 2, lower rates of social shifting were related to higher rates of non-social shifting.

With regards to RSM behavior, Time 1 Total RSM was significantly positively correlated with Time 2 Total RSM, $r(35)=.53$, $p=.001$. Time 1 total shift was significantly

negatively correlated with Time 2 Total RSM, $r(38)=-.32$, $p=.05$. Contrary to predictions that Attention Shifting and RRBs are related constructs and both measure sticky attention, Time 1 measures of Total Shifting were *not* significantly correlated with Time 1 measures of Total RSM, $r(40)=-.12$, $p=.47$, nor was Time 2 Total Shifting correlated with Time 2 Total RSM, $r(39)=-.17$, $p=.31$. These effects were maintained when examining the relationship between Social and Non-social Shifting with Total RSM. See Table 5 for all correlations. Attention shifting and RSM behavior are likely not measuring the same construct of sticky attention. As a result, both measures were included in comprehensive path analyses as separate constructs.

Cognitive Ability & ADOS Symptom Severity

Independent samples t-tests indicated that at 13 months, children later diagnosed with ASD did not differ in their developmental abilities (i.e., MSEL scores) from those that did not get diagnosed with ASD. See Table 1 for all MSEL scores. At Time 2, those later diagnosed with ASD had significantly lower Fine Motor Skills, $t(41)=3.02$, $p=.004$, and Expressive Language Skills, $t(41)=3.11$, $p=.03$, compared to those that were not later diagnosed with ASD. At Time 2, children later diagnosed with ASD also exhibited significantly more symptoms related to ASD as measured by the ADOS compared to those not later diagnosed with ASD (all $ps<.01$).

At Time 3, children diagnosed with ASD were not significantly different from those without ASD on measures of cognitive ability measured by the DAS-2. As expected, children with ASD at Time 3 exhibited significantly more ASD symptoms as measured by the symptom severity comparison score on the ADOS-2 (ASD=6.50; No ASD=2.63; $t(41)=-5.47$, $p<.001$). A comparison score of 4 or great indicates the child met criteria for ASD

based on the ADOS-2. It should be noted that ADOS-2 comparison scores were not normally distributed given that the majority of participants did not meet criteria for ASD. As a result, 60% of participants had ADOS-2 severity scores of 3 or less while 40% had scores of 4 or above. Total scores from the RBS-R indicate that children with ASD also exhibited more RRBs based on parent report compared to those without ASD (ASD=30.31; No ASD=10.47; $t(41)=-3.74$, $p=.001$). Children diagnosed with ASD presented with significantly more ASD symptoms in a clinical setting based on behavioral measures as well as at home and in the community based on parent report.

Pearson correlations were conducted between measures of cognitive ability and ASD symptoms at Time 1, Time 2, and Time 3 to better understand their relationship across early development. For all correlations, see Table 6. Measures of developmental and cognitive ability were positively correlated between Time 1, Time 2, and Time 3. ADOS comparison scores at time 3 were significantly correlated with cognitive ability as measured by the MSEL at time 1 ,$r(40)=-.32$, $p=.04$, and time 2, $r(40)=-.61$, $p<.001$. That is, more impaired cognitive ability at 13 and 22 months was related to higher ASD symptoms in preschoolers. Similarly, Time 2 MSEL was also significantly negatively correlated with concurrent ADOS scores, $r(44)=-.43$, $p=.004$.

Simple linear regressions were then conducted to better examine the effects of cognitive ability on ASD symptoms with Time 3 ADOS symptom severity as the dependent variable and cognitive measures from Time 1, Time 2, and Time 3 as the predictor variables. Time 1 MSEL independently significantly predicted Time 3 ADOS symptom severity, $F(1,38)=4.40$, $p=.04$, with an R^2 of .10. Time 2 MSEL independently significantly predicted Time 3 ADOS symptom severity, $F(1,38)=22.99$, $p<.001$, with an R^2 of .38. That is, ADOS

symptom severity scores increased .83 points for every 10 point decrease in Time 2 MSEL standard score. Finally, DAS-2 GCA scores did not significantly predict Time 3 ADOS scores, $F(1,38)=.52$, $p=.48$. These findings suggest that early assessments of cognitive ability were significantly predictive of later ASD symptom severity while concurrent measures of intellectual ability during the preschool years were not predictive of ASD symptoms.

Treatment. Possible effects of ART treatment were examined as part of an exploratory analysis to determine whether treatment significantly predicated ASD symptoms or diagnosis. If so, ART treatment group should be used covariate in the larger path analyses to control for a possible confound between children who received and did not receive treatment throughout the course of this project.. In the No ASD group, 18 participants received ART treatment and 12 participants received no treatment (SAU group). In the ASD group, 7 children received ART treatment and 6 children did not (SAU group). Time 3 ADOS severity scores were not significantly predicted by treatment group, $F(1,38)=.05$, $p=.83$. Similarly, Time 3 categorical diagnosis of ASD vs. no ASD was not significantly different based on treatment group, $\chi^2(1)=.14$, $p=.71$. In addition, there were no significant differences by treatment group for any Time 2 variables related to Looking at People, Attention Shifting, or RSM Behavior (ts<1; all ps>.48). As a result, treatment group was not included as a covariate in all other analyses including path analyses.

Effects of ART treatment on measures of Social Motivation and Sticky Attention were evaluated using a hierarchical linear regression using a step-wise progression. For these analyses, Time 1 variables were entered first followed by treatment group participation. Because some individuals did not have both Time 1 and Time 2 data, missing data was accounted for by a replacement with the mean of the overall group. Results indicated that

ART treatment did not significantly predict Time 2 measures of Social Motivation and Sticky Attention after entering Time 1 variables. Beta coefficients for treatment group after Time 1 variables were entered were as follows: Looking at People β=.003, *t*=.03 *p*=.97; Total Shifting β=-.13, *t*=-.83 *p*=.41; Social Shifting β=-.12, *t*=-.87 *p*=.39; Non-social Shifting β=-.08, *t*=-.53 *p*=.599; Total RSM β=.03, *t*=.21 *p*=.83. In sum, participation in treatment did not influence the change in Social Motivation and/or Sticky Attention between 13 and 22 months of age.

Gender and Race. In addition to cognitive ability and treatment, both gender and race were assessed as possible covariates. Time 3 ASD symptom severity was not significantly different between gender, $F(1,38)=1.14$, $p=.29$; nor was Time 3 categorical diagnosis of ASD vs. no ASD significantly different for gender, $\chi^2(1)=.76$, $p=.38$. For race, there was a significant difference when all participants were included, $F(3, 36)=3.86$, $p=.02$. However, this is likely due to an outlier, wherein one race-group had only one participant with a very high comparison score (ADOS Symptom Severity=10). When this participant was excluded from the analysis, there was no difference in ASD symptoms severity or categorical diagnosis of ASD based on race, $F(2, 36)=2.03$, $p=.15$, $\chi^2(3)=2.43$, $p=.49$, respectively. It is unlikely that the child's race was related to their ASD symptom severity and, a more parsimonious interpretation is the small sample size. As a result, race was not included as a covariate in the following path analyses.

Analysis of CSBS Variables, Cognitive Ability, and ASD Symptoms

Given the significant relation between ASD diagnosis and developmental level on the MSEL at Time 1, Pearson correlations were conducted to examine the relationship between

MSEL and CSBS measures of Looking at People, Attention Shifting, and Total RSM prior to conducting path analyses. See Table 7 for all Pearson correlations.

Time 1 MSEL was only significantly negatively correlated with Time 1 RSM behavior. That is, lower MSEL scores at Time 1 were related to higher repetitive motor movements with body and objects at Time 1, $r(40)=-.35$, $p=.03$. Time 2 MSEL standard scores were positively correlated with Time 1 and Time 2 Looking at People (Time 1: $r(43)=.42$, $p=.005$; Time 2: $r(40)=.31$, $p=.05$), Time 1 and Time 2 Total Shifting (Time 1: $r(43)=.43$, $p=.004$; Time 2: $r(40)=.46$, $p=.003$) and Time 1 and Time 2 Social Shifting (Time 1: $r(43)=.49$, $p=.001$; Time 2: $r(40)=.45$, $p=.004$). That is, less time looking at people and lower rates of overall and social attention shifting at 13 months were related to lower MSEL scores at 22 months of age. This same relationship was maintained when examining the concurrent relationship at 22 months. Interestingly, Time 2 MSEL scores were not significantly correlated with non-social shifting at Time 1 or Time 2 (Time 1: $r(43)=-.05$, $p=.78$; Time 2: $r(40)=.05$ $p=.78$). Time 2 MSEL was negatively correlated with Time 1 and Time 2 RSM total indicating higher MSEL scores at Time 2 was related to lower frequency of RSM behavior at Time 1 and Time 2. Time 3 DAS-2 GCA scores were not significantly correlated with Looking at People, Attention Shifting, or RSM Total at Time 1 or Time 2. See Table 7 for all correlations.

Time 2 ADOS total scores were significantly *negatively* correlated with Time 1 and Time 2 Looking at People (Time 1: $r(43)=-.30$, $p=.05$; Time 2: $r(40)=-.35$, $p=.03$), and Time 2 Total Shifting, $r(40)=-.49$, $p=.001$, and Social shifting, $r(40)=-.53$, $p<.001$. In other words, spending less time looking at people at 13 months and 22 months was related to increased ASD symptoms at 22 months. In addition, lower rates of overall attention shifting and social

shifting was related to increased ASD symptoms. While Time 1 Total Shifting was not significantly correlated with Time 2 ADOS scores, $r(43)=-.18$, $p=.24$, Time 1 Social Shifting was marginally significantly correlated with Time 2 ADOS scores, $r(39)=-.29$, $p=.06$, suggesting that lower rates of attention shifting that included people at 13 months was related to increased ASD symptoms at 22 months. Time 3 ADOS comparison scores were significantly negatively correlated with Time 2 shifting, $r(36)=-.45$, $p=.006$, and social shifting, $r(36)=-.38$, $p=.02$. It was *not* significantly correlated with non-social attention shifting, $r(36)=-.18$, $p=.30$. That is, lower rates of overall shifting at social attention shifting at 22 months was related to increased ASD symptoms during the preschool years. Non-social attention shifting between objects only was not related to ASD symptoms during the preschool years.

Path Analysis Model with Overall Attention Shifting

Path analyses were completed as proposed to examine the social motivation theory compared to the attention theory. The model included Time 1 and Time 2 variables of Looking at People, Total Shifting, and RSM behaviors as the 3 predictor variables and ADOS comparison scores at Time 3 as the outcome variable. Preliminary analyses determined there was no significant relationship between concurrent measures of RSM behavior and attention shifting so both variables were included in the same model. Time 1 MSEL scores were included as a covariate to account for initial differences in developmental ability and the relation between developmental level at Time 1 and diagnosis at Time 3.

The model was initially conducted with all Time 1 variables predicting all Time 2 variables (See Figure 2 for initial model). Upon initial examination, non-significant predicting relationships were removed except for correlations between the Time 1 and Time

2 variables in order to account for relationships within each time point. The full model was appropriately trimmed to further test the two proposed theoretical constructs. The final model (see figure 3) demonstrated excellent fit, $\chi^2(9)=3.58$, $p=.94$; RMSEA<.001; CFI=1.00; TLI=1.25, and justified the use of the variables in the model. Significant direct pathways are presented in Figure 3. After accounting for the effects of Time 1 MSEL scores and the relationship with other Time 1 variables, Time 1 Looking at People significant positively predicted Time 2 Looking at People and Time 2 Total Shifting. In addition, Time 1 Total RSM only significantly predicted Time 2 Total RSM. Time 2 shifting significantly predicted Time 3 ADOS score, $\beta=-.51$, $p=.001$, after accounting for the relationship between other Time 2 variables and the above and beyond the effects of Time 1 MSEL scores. That is, lower rates of attention shifting at 22 months predicted increased ASD symptoms during the preschool years. Time 2 RSM total was marginally significant predicting Time 3 ADOS score, $\beta=.24$, $p=.06$, suggesting that higher frequency of RSM behavior at 22 months predicted increased ASD symptoms at 22 months. Time 2 looking at people did not significantly predict Time 3 ADOS score, $\beta=.27$, $p=.11$, suggesting that the amount of time spent looking at a person at 22 months was not predictive of ASD symptoms during the preschool years.

Indirect effects were assessed to determine significant pathways from Time 1 variables to Time 3 ADOS scores with the mediating effects of Time 2 variables. There was a significant indirect pathway from Time 1 looking at people to Time 2 total shifting to Time 3 ADOS Scores ($\beta=-.20$, $p=.04$) indicating that less time looking at people at 13 months predicted higher ASD symptoms during the preschool years as mediated by attention shifting at 22 months. No other indirect pathways significantly predicted Time 3 ADOS scores. The

indirect effect from Time 1 Total Shifting to Time 2 Looking at People to Time 3 ADOS symptoms was not significant, β=-.02, p=.61. The indirect effect from Time 1 RSM behavior to Time 2 RSM behavior to Time 3 ADOS symptoms was not significant, β=.12, p=.12. The indirect effect from Time 1 Looking at People to Time 2 Looking at People to Time 3 ADOS symptoms was not significant, β=.18, p=.12.

Path Analysis Model with Social vs. Non-Social Attention Shifting

The first path analysis model suggested that lower rates of attention shifting at 22 months played an important role in predicting increased ASD symptoms. In order to better assess the Social Motivation vs. Sticky Attention theories, 2 additional path analysis models were conducted with one using social shifting and the other using non-social shifting.

Analysis with Social Attention Shifting. This model was conducted using the same methodology as previously described for the path analysis model using total shifting with the exception being the use of Social Shifting at Time 1 and Time 2 replacing Total Shifting. Social Shifting was defined as attention shifts that included a person. The model demonstrated excellent fit, $\chi^2(9)=3.76$, p=.92; RMSEA<.001; CFI=1.00; TLI=1.17, and justified the use of the variables in the model. Significant direct pathways are presented in Figure 4. Similar to the previous model, Time 1 Looking at People significantly positively predicted Time 2 Looking at People and Time 2 Social Shifting above and beyond the effects of Time 1 MSEL and the relationships with other Time 1 variables. In addition, all Time 2 variables significantly predicted Time 3 ADOS scores after accounting for their relationship to one another and above and beyond Time 1 MSEL scores.

With regards to indirect pathways, there was a significant indirect effect from Time 1 looking at people to Time 2 social shifting to Time 3 ADOS score, β=-.36, p=.02, indicating

that less time looking at people at 13 months predicted increased ASD symptoms during the preschool years as mediated by social attention shifting at 22 months. The indirect pathway from Time 1 Looking at People to Time 3 ADOS scores as mediated by Time 2 Looking at People was not significant, β=.29, p=.10, although this is trending towards significance. Similarly, the indirect pathway from Time 1 RSM behavior to Time 3 ADOS scores mediated by Time 2 RSM behavior was not significant, β=.13, p=.10, but was close to reaching significance.

Analysis with Non-Social Attention Shifting. This model was conducted using the same methodology as previously described with the exception of using Non-social Shifting instead of Total Shifting. The model demonstrated excellent fit, $\chi^2(9)=2.08$, p=.97; RMSEA<.001; CFI=1.00; TLI=1.47, and justified the use of all variables in the model. Significant direct pathways are presented in Figure 5. Similar to previous models, Time 1 Looking at People significantly predicted Time 2 Looking at People. In contrast, though, Time 1 Looking at People *negatively* predicted Non-social Shifting suggesting that less time looking at people at 13 months of age predicted increased rates of non-social shifting at 22 months. Time 1 RSM behavior continued to positively predict Time 2 RSM behavior. After accounting for the relationship between Time 2 predictor variables and Time 1 MSEL, only Time 2 non-social shifting significantly predicted Time 3 ADOS score, β=-.31, p=.04, such that lower rates of non-social attention shifting at 22 months significantly predicted higher ASD symptoms during the preschool years. Neither Time 2 looking at people or RSM significantly predicted Time 3 ADOS scores in this model.

Indirect effects of Time 1 variables on Time 3 ADOS scores were evaluated. In contrast to previous models, there were no significant indirect effects from Time 1 variables

to Time 3. The indirect pathway from Time 1 looking at people to Time 2 non-social shifting to Time 3 ADOS scores was not significant, β=.11, p=.11, although it is trending towards significance. It is possible this indirect effect may be present in the data with a larger sample size. However, given that the effect is not statistically significant, it must be interpreted with caution. Time 1 Non-social Shifting did not have an indirect effect on Time 3 ADOS scores through Time 2 Looking at People, β=.03, p=.40, or Time 2 RSM behavior, β=-.05, p=.23. Time 1 RSM behavior did not have an indirect effect on Time 3 ADOS scores through Time 2 Non-social Shifting, β=.03, p=.51, or Time 2 RSM behavior, β=.11, p=.13.

CHAPTER 4: Discussion

The purpose of this study was to compare two different theoretical models relating to the early development of ASD in infants and toddlers. The Social Motivation theory of ASD suggests that individuals later diagnosed with ASD have decreased interest and motivation to engage in social information (i.e., people) indicated by decreased dyadic and triadic interactions during infancy and early childhood (see Chevallier et al., 2012; Dawson et al., 2004). This reduced engagement with social information follows a cascading pattern that leads to later impairments in social communication and reciprocal social interactions that are core symptoms of ASD (Klin, Jones, Schultz, Volkmar, & Cohen, 2002). This lack of engagement in social information also leads to a corresponding increased engagement with objects or non-social information. In turn, this increased focus on objects leads to overly focused and non-functional interests on specific objects and interests in non-social information (e.g., Bruckner & Yoder, 2007; Stronach & Wetherby, 2014). In contrast, the Attention theory of ASD posits that infants later diagnosed with ASD have difficulty disengaging and shifting their attention between information in their environment (i.e., "sticky attention"; Landry & Bryson, 2004; Sacrey et al., 2013). Because attention is overly focused on one specific object or person, it is predicted that these infants often do not attend to important information in their environment and have difficulty integrating information across multiple experiences in the environment. This atypical development of attention leads to difficulties in understanding and maintaining social interactions that require an integration

of social and nonsocial information in the environment (e.g., triadic interactions that link eye gaze, gesture, and objects in the environment).

To examine these two theoretical models, several path analyses were conducted to understand the development of social motivation/social engagement (i.e., looking at people), shifting attention between stimuli in the environment, and RRBs in children at high-risk for being diagnosed with ASD at 13 and 22 months of age and how these behaviors relate to ASD symptoms during the preschool years. Overall, results more strongly supported the Social Motivation model. Specifically, reduced looking at people at 13 months predicted a lower frequency of attention shifting at 22 months, which then predicted higher ASD symptoms and ASD diagnosis during the preschool years. In contrast, the alternate theory that the increased presence of "sticky attention" difficulties at 13 months would predict later social difficulties and more ASD symptoms was not supported. While attention difficulties at 22 months of age were predictive of later symptom severity and diagnosis, difficulties in shifting attention at 13 months of age was not predictive of this pattern.

To further examine the relationship between attention shifting and ASD symptom severity, analyses were conducted to examine differences between social shifting compared to non-social shifting. Results suggested that while both social and non-social shifting at 22 months predicted ASD symptoms at preschool age, only a statistically significant developmental pathway was seen with social shifting. That is, reduced looking at people at 13 months significantly predicted lower rates of social attention shifting which in turn predicted increased ASD symptoms during the preschool years. This same indirect effect was not maintained for non-social shifting in that looking at people at 13 months did not significantly predict ASD symptoms during the preschool years as mediated by non-social

shifting at 22 months. However, lower levels of non-social shifting at 22 months did *directly* predict increased ASD symptoms during the preschool years. Together, these results suggested that when examining developmental trajectories from 13 months to preschool-age, both social engagement and social attention shifting played a significant role in predicting ASD symptoms. While overall attention shifting and disengagement played an important role in predicting later ASD symptoms, these effects were not apparent until later in development, at 22 months. Thus, these results provided stronger support for the primacy of the Social Motivation theory over the Attention theory of ASD symptom development in the first two years of life. The implications of these results within each of these theoretical models will be discussed below.

Social Motivation Theory

Through all analyses, it was clear that lower social engagement (i.e., looking at people) at 13 months predicted lower rates of social attention shifting skills at 22 months. Interestingly, though, social engagement at 22 months was not significantly predictive of later ASD symptoms. That pathway suggests that, while social engagement is critical to the early emergence of ASD precursors, there may be an intermediary factor between social engagement and the development of ASD. Social engagement must be conceptualized in the context of the development of other symptoms or behaviors related to ASD. Social Motivation theory assumes that spending less time looking at and engaging with social information is indicative of a lack of motivation to engage with this social information (Dawson et al., 2004). This reduced motivation leads to both a decrease in engaging with faces but also a subsequent increase in objects. In addition, it leads to difficulties integrating social and non-social information within an environment.

Results from the present study were consistent with previous studies indicating early impairments in engagement and attention to faces and social information, particularly in the first year of life. For example, Jones and Klin (2013) noted differences in the eye-gaze towards faces from 2-6 months of age in high-risk infants later diagnosed with ASD compared to low-risk infants. Infants later diagnosed with ASD initially exhibited normative levels of attention to faces, particularly eyes at 2 months of age and then show a decline in this attention to faces by 6 months compared to infants not at-risk for developing ASD. Other studies suggest that this lack of engagement and interest in faces persists through early toddlerhood with 2-year-olds with ASD disengaging their attention from faces more frequently than children with DD and typical development (Chawarska et al., 2010). This research supports the present study suggesting that decreased looking at and engagement with people and social information may be a core underlying impairment in ASD during early development.

The integration of information, particularly social information, within an environment is imperative to development as evidenced by the importance of joint attention (Tomasello, 1995). These triadic interactions, joint attention, allow for increased communication and social interaction with others (Carpenter et al., 1998; Tomasello et al., 2005). Without the motivation to engage in social information, children later diagnosed with ASD miss vital opportunities for learning about their social world in the same way as their typical peers (Klin, 2000; Klin et al., 2002; Klin & Jones, 2006). Results from the present study suggested that a lack of engagement with social information (i.e., looking at faces) has cascading effects on developmental processes related to social interactions and ASD symptoms later in childhood. These results are consistent with several infant/toddler treatment programs

focused on increasing attention to social information through the use of child directed, naturalistic, behavioral strategies (Dawson et al., Kasari, Freeman, & Paparella, 2000; 2010; Rogers et al., 2012). Recent treatment studies have also focused on teaching parents of young children to increase social motivation leading to long-lasting positive effects on diagnostic outcome and ASD symptoms (Pickles et al., 2016). That is, 2-4 year old children with ASD showed improvement in symptoms after participating in a treatment program that focused on increased dyadic interactions with their parents compared to children with ASD that did not receive treatment. Future studies can examine the treatment effects of targeting these precursor symptoms earlier in development for children at-risk for and ASD but prior to a formal diagnosis.

When evaluating the differences in effects of social vs. non-social attention shifting, it appeared as though social shifting was a strong mediator compared to non-social shifting at 22 months of age. There was a significant indirect effect with social shifting in that reduced attention to people at 13 months predicted reduced shifting of attention between objects and people at 22 months which in turn predicted increased ASD symptom severity during the preschool years. This suggests that children later diagnosed with ASD are not integrating social information into their world by shifting their attention between people and objects in their environment. The mediating effects of social shifting, specifically, appeared to be partly a result of initial impairments in social engagement, which in turn had cascading effects on later development of triadic interactions and eventual ASD symptoms. The fact that this same type of indirect effect was not clearly present when examining non-social shifting suggest that triadic interactions with another person are an important part of the unfolding development of ASD symptoms. These results are consistent with previous research that

suggests that late in the second year of life (~18-24 months), children later diagnosed with ASD show fewer gaze shifts incorporating another person within their environment and reduced joint attention with another person (Wetherby et al., 2007). The present study, however, provides further specificity to the importance of attention shifting with social compared to non-social information in the environment.

When examining the trajectory of RRBs within this sample of children diagnosed with high-functioning ASD, they appeared to follow developmental trajectory independent of social engagement and attention. While previous studies suggest that RRBs are part of the cascading effects of Social Motivation theory such that there is an increase in motivation to engage with objects in a repetitive manner (Bruckner & Yoder, 2007; Ozonoff et al., 2008), results from this study indicated that RRBs may truly develop independently of other symptoms within children with high-functioning ASD. Further examination of the development of RRBs in this study will be discussed below.

Attention Theory

With regards to the theory of Attention in ASD symptom development, it was initially hypothesized that both attention shifting and RRBs were measures of difficulty disengaging or "sticky" attention. However, initial analyses indicated that RRBs and shifting were not significantly related and likely not measuring the same construct of difficulty with disengaging attention. Final path analyses were conducted with both RRBs and Attention Shifting in the model in order to better understand their developmental trajectories in relationship to one another.

Results indicated that early impairments in attention shifting at 13 months were not significantly predicting later attention shifting behavior nor was it indirectly predicting ASD

symptoms. This lack of relationship between the same variable at two separate time points highlights the complexity of these high-risk infants at 13 months. This is in direct contrast to theories of Attention that hypothesize impairments in attention shifting and disengagement in young children later diagnosed with ASD (e.g., Elsabbagh et al., 2013; Sacrey et al., 2014). Further examination of the means of attention shifting suggests that there was significant variability among the entire group of participants without any clear differences between later defined diagnostic groups at 13 months. However, the participants in the present study were all identified as "at-risk" for an ASD diagnosis at 13 months. By 22 months, children later diagnosed with ASD had significantly lower frequency of attention shifting compared to those not later diagnosed with ASD. Those later diagnosed with ASD showed a decrease in their shifting frequency while those not later diagnosed with ASD showed an increase, more clearly differentiating the diagnostic groups late in the 2nd year of life. Those that were identified at-risk for ASD at 13 months but did receive a diagnosis may have been delayed in the development of their attention shifting and then improved by 22 months. In contrast, those later diagnosed with ASD may have an atypical development of attention shifting at 13 months that is maintained at 22 months. The complexity of these varied developmental trajectories of attention shifting cannot be accurately assessed in this particular study, although future studies may examine more nuanced approaches to attention shifting and include a low-risk group of participants that represent neurotypical developmental behavior at 13 months. Thus, while this study did not support persistent difficulties in attention shifting in a high-risk sample, it is possible that a study including only children who develop the diagnosis may show more clear attention impairments (e.g., Swettenham et al., 1998; Elsabbagh et al., 2013; Klin et al., 2002).

It is important to note that the shifting measured in the present study was capturing a complex 3-part process of disengaging from a stimulus immediately followed by shifting to another stimulus and re-engaging attention in that new stimulus. Several studies have examined only a single aspect of this 3-part process, such as disengagement from attention rather than capturing the full process of attention shifting with some studies suggesting early impairments in this process around a child's first birthday (e.g., Elsabbagh et al., 2013; Sacrey et al., 2013). Other studies suggest that in 6-9 month-old infants later diagnosed with ASD attention may not be getting "stuck" but rather they are shifting too frequently or spending less time fixating on individual stimuli compared to those that are not later diagnosed with ASD (Wass et al., 2015). The atypical use of attention shifting and fixating on stimuli may represent difficulties integrating information within the environment. For example, initiation of joint attention occurs when a child can integrate both information they see (e.g., a picture) and a person with whom they want to share that information (Carpenter et al., 1998) and is present in typical development by 9-10 months of age (Striano & Rochat, 1999). In a sample of children at high-risk for developing ASD, joint attention is reduced in frequency compared to those at low-risk for developing ASD although this impairment is not consistently present until late in the 2nd year of life (Wetheryby et al., 2007). Further longitudinal research is needed to examine the individual trajectories and the role of these distinct attention mechanisms in infants and toddlers at risk for ASD.

Attention can also be evaluated through basic scientific experiments that examine individual components of attention (e.g., orienting, disengagement, shifting) in individuals with ASD. These individual attention processes provide key information about the impairments in attention in ASD. In simple orienting paradigms, individuals with ASD

exhibit deficits in the orienting to both social and non-social information (for review see Keehn, Muller, & Townsend, 2013). Some studies have further evaluated whether this impairment in orienting is related to endogenous versus exogenous attention with some studies showing intact endogenous but impaired exogenous orienting in ASD (Renner et al., 2006). In addition, disengagement of attention is frequently examined using a gap-overlap paradigm with several studies suggesting that individuals with ASD have longer latencies to disengage their attention from a stimulus (e.g., Kawakubo et al., 2007; Landry & Bryson, 2004; Zwaigenbaum et al., 2005). The individual processes of attention must continue to be examined within a cognitive neuroscience framework in order further understand the underlying impairments of attention in individuals with ASD.

While the Attention theory was not strongly supported in the present study given the lack of predictive power of early attention shifting on later ASD symptoms, this sticky attention clearly plays a key role in the development of ASD symptoms later in the 2nd year of life. It is possible that underlying mechanisms of both Social Motivation and Attention theories are present in early development in ASD. That is, in order to orient towards a person, one must have adequate attention, fixation, and disengagement skills to make that initial orientation towards social information. Jones and colleagues (2016) propose that reduced attention and engagement with faces in 6-month old infants later diagnosed with ASD may be a result of both impairments in social motivation *and* atypical patterns of attention. It is imperative to understand the shifting behavior in children with typical development and no clear risk of developing ASD at 13 months. This can inform our understanding of developmental trajectories within ASD, especially for early attention shifting and social orienting mechanisms.

Role of RRBs in Developmental Trajectories

Path analyses from the present study indicated that RRBs follow a developmental trajectory that is separate from the developmental processes of attention shifting and social engagement. Previous studies have suggested that the presence of RRBs may occur as a result of a co-occurring effect of a decreased interest in people and a heightened increase and over-focus on objects (Ozonoff et al., 2008; Bruckner & Yoder, 2007). However, results from path analyses suggest that differences in attention shifting and/or social engagement are not significantly predicting or influencing RRBs starting at 13 months of age. Overall, increased RRBs at 13 months significantly and consistently predicted increased RRBs at 22 months. Further, the presence of RRBs at 22 months was not a consistent predictor of ASD symptoms during the preschool years. There are several possible explanations for the relative independence of RRBs across development in this study.

One explanation is that RRBs in early development for at-risk toddlers may be more a function of developmental delay rather than truly atypical behaviors. The literature has been mixed with some studies suggesting that children with ASD do not have significantly more RRBs compared to those with DD during early development (Cox, 1999; Ventola et al., 2006). Although more recently, studies suggest there is a significant difference both in frequency and quality of RRBs in high-risk infants later diagnosed with ASD early in life compared to high-risk infants *not* diagnosed with ASD (Elison et al., 2014; Wolff et al., 2014). Results from the present study indicate that RRBs at 13 months are significantly *negatively* correlated with cognitive ability at 13 and 22 months such that those with lower cognitive abilities (i.e., with more developmental delays) have greater frequency of RRBs. This is consistent with previous studies finding that lower nonverbal cognitive ability is

related to more RRBs in children with ASD (Bishop, Richler, & Lord, 2006). Together, this indicates that cognitive ability may explain some of the variability in RRBs during early development in ASD.

Another explanation is that RRBs truly follow a separate trajectory compared to symptoms social communication and interactions. Factor analyses of parent-report measures of ASD symptom, such as the Social Responsiveness Scale-Second Edition (SRS-2; Constantino, 2013) identify 2 discrete symptom categories within ASD with one factor incorporating social communication and interaction and the other factor encompassing restricted and repetitive behaviors (Frazier, Ratliff, Gruber, Zhang, Law, & Constantino, 2013). An analysis of the Childhood Autism Rating Scale (CARS) in 2-year-olds similarly yielded separate factor structures for (1) social communication skills and (2) stereotyped behaviors and sensory sensitivities (Moulton, Bradbury, Barton, & Fein, 2016). Together with results from the present study, RRBs as a broad symptom category of ASD may develop independently of symptoms related to social communication and reciprocal social interactions.

Last, the ADOS-2 as a measure of symptom severity has historically been critiqued for its difficulty accurately capturing RRBs (Hus, Gotham, & Lord, 2012). The ADOS-2 evaluation captures a short time-period (i.e., less than 1 hour), and the manifestation of these atypical behaviors may not occur in such a short window of time. Indeed, the overall scores from the ADOS-G (Lord et al., 2000) did not incorporate the presence of RRBs into the algorithm, as it was believed RRBs were occurring at a low frequency during these short evaluations. The video coding of RRBs at 13 and 22 months in the present study may not capture the same frequency and presence of RRBs in the ADOS-2 conducted during the

preschool years. Additional measures of RRBs may be needed to accurately evaluate the developmental trajectory of RRBs in infants and toddlers at-risk for ASD.

Cognitive and Developmental Abilities

This study used the Mullen Scales of Early Learning (MSEL) at 13 and 22 months as a measure of cognitive abilities. Scores from the MSEL at 13 and 22 months were strongly correlated and predictive of later ASD symptom severity and diagnosis. Cognitive ability (IQ) as measured by the DAS-2 at age 3-5 years was not significantly correlated or related to diagnostic outcome and symptoms. This suggests that children later diagnosed with ASD score much lower on cognitive testing in early development, prior to diagnosis. Later in childhood, intellectual ability was not as relevant a factor for determining ASD diagnoses, as the concurrent IQ scores were not related. This supports previous literature suggesting that children's cognitive ability is strongly correlated with their later diagnostic symptom severity (Chawarska, Klin, Paul, Macari, & Volkmar, 2009). The developmental profile in young children later diagnosed with ASD is very fluid and volatile. These children can present a variety of behavioral challenges during testing that make it difficult to consistently know if the scores are truly valid and representative of their abilities. In addition, several items on the MSEL, particularly in the receptive language subscale, require children to orient to information, respond to their name, and understand simple requests and gestures. These skills are underlying impairments in ASD (Wetherby et al., 2004, 2007) and failing these items again may be more representative of their ASD symptoms rather than their impairments in receptive language.

Indeed, the children later diagnosed with ASD received receptive language scores from the MSEL approximately one standard deviation below the mean at 22 months. These

same children also had a significant decrease in their Fine Motor composite score from 13 and 22 months suggesting either a significant decrease in fine motor skills or lack of improvement in skills across early development. In contrast, those that did not later receive a diagnosis of ASD maintained average scores in areas of Visual Reception and Fine Motor while significantly increasing with Receptive and Expressive Language scores. Future research is needed on this instability in early cognitive and developmental measurement in ASD. Many research studies use MSEL scores as a measure of treatment outcome or improvement in skills during early childhood. If these skills such as receptive language and fine motor skills change drastically between 22 months and the preschool years, these earlier scores may not be accurately representing a child's skills.

Some studies suggest that infants later diagnosed with ASD show a slower rate of growth in their cognitive development compared to those that are not later diagnosed with ASD. For example, Landa, Gross, Stuart, and Faherty (2013) prospectively followed infant siblings of children with ASD and typical development from 6 to 36 months. Their results from the MSEL indicate that children later diagnosed with ASD show an increase in their raw score over time, but their rate of change is slowed compared to those that are not later diagnosed with ASD. The MSEL is a frequently used measure within ASD literature, although few studies have examined its validity within this atypical population. Bishop, Guthrie, Coffing, and Lord (2011) examined the convergent validity of the MSEL in 53 children with ASD. While they established convergent validity, the children participating in the study had an average of 4 years. Future studies must examine the convergent validity of the MSEL during the first two years of life in children at high-risk for ASD.

Implications for Early Intervention

Individuals participating in this study were randomized to receive ART treatment between 13-22 months of age. An exploratory aim for this study was to determine the effect of treatment on later ASD symptoms and diagnosis. Initial analyses indicated that the treatment provided did not have a significant effect on later ASD symptoms and diagnostic outcome nor did participation in treatment significantly affect measures of Social Motivation and Sticky Attention at 22 months of age. However, it may be beneficial to evaluate these measures of Social Motivation and Sticky Attention in direct relationship to change in parent responsiveness as a result of the intervention rather than just participation in treatment. It may also be worthwhile to assess whether treatment resulted in distal outcomes of increased social motivation and/or decreased difficulty with attention disengagement during the preschool year. Further analyses are needed to better understand the other possible effects of ART and other early intervention treatments on other developmental skills and behaviors.

Early intervention programs for young children with ASD incorporate significant parent involvement in order to improve outcomes and increase the intensity of intervention. In fact, for children without ASD, parent responsiveness during interactions with their children significantly influences their child's cognitive, language, and social-emotional development (Mahoney & Peralies, 2003; McDuffie & Yoder, 2010; Ruble, McDuffie, King, & Lorenz, 2008; Siller & Sigman, 2008). Due to the core social communication and interaction deficits in ASD, children with ASD may not maintain reciprocal social interactions with their parents in the same way as children with ASD. As a result, parent responsiveness and engagement with their child may change based on their child's individual level of engagement. This potentially can have lasting effects on both the parent-child relationship as well as the overall

development of the child. Siller and Sigmund (2002) found that mothers of children with ASD that were more responsive during interactions had children with higher levels of communication across childhood. Change and influence of parent responsiveness during parent-child interactions due to targeted interventions has been evaluated to better identify it's effects on child outcomes in ASD. Indeed, some studies have suggested that increased parent responsiveness improves social-emotional functioning in toddlers with ASD (Mahoney & Perales, 2003). Siller and colleages (2013, 2014) found that a parent-mediated intervention increased parent responsiveness to their children with ASD as well as improved attachments relationships between parent and child over the course of treatment. The effect of parent-child interactions including parent responsiveness to their child must be included when evaluating the effects of treatment in ASD as a positive and secure parent-child relationship is key to furthering cognitive, language, and social-emotional development in children.

Evaluation of Social Motivation and Attention theories provided important information about the development of early intervention programs for children at-risk for ASD. While it was not clear that attention shifting at 13 months is a significant predictor of later ASD symptoms, impairment at 22 months along with reduced early social engagement is related to increased ASD symptoms during the preschool years.

Randomized control trials (RCTs) with toddlers with ASD suggest that certain early intervention treatments improve the outcome for children showing improvement in important social interaction skills as well as improved communication (Dawson et al., 2010; Landa, Holman, O'Neil, & Stuart, 2011; Rogers et al., 2012; Warren et al., 2011). These early intervention programs can incorporate goals focused on improving social engagement and

the integration of information in the environment during the second year of life to provide direct treatment to a possible underlying cognitive mechanism present early in life in ASD. Many of these RCT incorporate naturalistic play-settings for toddlers in conjunction with behavioral strategies that focus on improving skills across domains (e.g., social interaction, communication, adaptive behavior). For example, the Early Start Denver Model (ESDM; Dawson et al., 2010; Rogers et al., 2012) uses naturalistic behavioral strategies to improve parent-child interactions, social engagement, communication, and developmental skills. Results indicate that toddlers with ASD that receive ESDM treatment show improvement in IQ, adaptive skills as well as a decrease in ASD symptoms.

A recent article by Brian, Bryson and Zwaigenbaum (2015) discusses four important developmental considerations when developing treatment targets and goals during early intervention. The authors suggest that targeting attentional control and social engagement during treatment will allow to optimize development for those that present with atypical behaviors and/or impairments. Attention as a larger construct and developmental process is important in how individuals learn about the world. Results from the present study support the importance of intervening around social engagement as well as the continued fluid integration of both social and non-social information into their interactions. Attention is the foundational skill and impairments in different forms of attention can have cascading effects on later development. Young children later diagnosed with ASD spend less time looking at people and faces as evidenced both by results from the present study as well as other studies (e.g., Chawarska et al., 2010, 2013; Chawarska & Volkmar, 2007; Dawson et al., 2004, 2005). Treatment programs must continue to consider these important skills related to social

engagement and attentional control when providing early intervention for children at high-risk for ASD.

Limitations and future directions

The present study has several limitations that point to areas of future research in understanding early developmental processes in ASD. First, the participants in this sample all exhibited atypical development and behaviors at 12 months based on the parent-report of the FYI at 12 months. In order to participate in this study, infants had to exhibit impairments in early social communication *and* repetitive behaviors or restricted interests. No typical comparison group was included. Thus, the results presented are representative of children with early atypical and/or delayed development. While a strength of this study is that it used a community-based sample at-risk for ASD, it is important for future studies to incorporate infants without concerns for ASD from the general population. For example, future studies can include children whose parents completed the FYI, but did not meet clinical cutoffs for atypical social communication *and* repetitive behaviors. This comparison group will improve our ability to construct developmental trajectories for infants with typical development, at-risk infants who are not later diagnosed with ASD, and at-risk infants that are later diagnosed with ASD within a community-based sample.

This study had a relatively small sample size and, thus, the types of analyses that could be completed and thus interpreted were limited. The final group of children with ASD consisted of 13 participants. Results should be interpreted with some caution and may not generalize to a larger group of participants. In addition, ADOS scores indicated a bimodal distribution of ASD symptom severity given that the cutoff for ASD is a symptom severity of 4 on the ADOS. All participants with ASD had symptom severity scores of 4 or greater while

those without ASD had scores less than 4. It is important to consider this variability in symptoms severity scores and strive for a normal distribution of scores within a clinical sample. However, despite a small sample size and bimodal distribution of ADOS scores, complex analyses identified significant and meaningful pathways based on theoretical models related to the development of ASD. It is also particularly important to note that participants included in this study all had average to above average intellectual abilities during the preschool years. The results presented here are specifically focused on those diagnosed with high-functioning ASD and the developmental trajectories may not be representative of those with more impaired cognitive abilities and ID. However, clear pathways were identified within this small sample with a narrowly defined set of parameters indicating that these results may be extrapolated to understand a larger population of individuals with high-functioning ASD.

As previously mentioned, attention shifting was measured as a 3-part process in which the child disengaged their attention from a particular object, person, or other stimulus followed immediately by a gaze-shift to another stimulus and then engaging attention to that stimulus. This measure of shifting captures a complex process that did not exhibit differences or predictive power until the participants were 22 months. Future studies can break down this process into separate parts to identify where the true impairment and/or atypical development occurs. Several studies have already examined the process of disengagement of attention, but research on the process of immediately shifting to re-engage attention is less thoroughly researched. Previous studies have primarily focused on attention shifting and disengagement during experimental tasks. It is imperative to understand how this translates to the naturalistic

setting. The present study begins to explore this area of research, although future studies are needed to identify the nuances related to attention development in naturalistic settings.

Alternatively, this study did not differentiate between child-directed and parent/examiner-directed attention shifting. That is, it is not clear whether a child shifted their attention based on their own volition or whether their parent or the examiner worked to get their attention. The CSBS is designed to engage a child in several different opportunities to engage with materials, request routines or behaviors to continue, and initiate joint attention. For example, in every evaluation the examiner points to a picture on the wall behind the child to try and shift and direct their attention to the picture. Other times, a child spontaneously looks up at the examiner, parent, or another object without any prompting. It is possible that early differences in shifting may be evident if the attention shifting is child-directed rather than examiner or parent-directed. Children that are more frequently and spontaneously shifting their attention may have more inherent flexibility in their attentional control. This level of specificity of evaluating attention shifting can allow for further examination of the Attention theory of ASD by identifying the key components involved in the attention shifting process.

Participants in this study were identified at-risk for developing ASD at 12 months and first seen in person at 13 months. Social motivation as measured by social engagement and attention shifting are present prior to 12 months and information about earlier development in these areas can provide further insight into the development of social motivation, attention shifting, and RRBs. One of the advantages of infant sibling studies is that they can track development in high-risk infants from very early in life because their "risk-factor" is based on the genetic relationship to a biological sibling with ASD. These evaluations in infancy

provide unique opportunities to understand the earliest developmental processes of social motivation and attention shifting. Incorporating these early developmental stages into larger developmental trajectories is necessary to identify the core underlying impairments in ASD.

The CSBS was the primary assessment used for video coding of social attention, attention shifting, and RSM behavior at 13 and 22 months. The CSBS was ideal for this type of video coding because the child was usually stationary in a chair that attached to a table and both a parent and the examiner were present on either side of the table. Unfortunately, this assessment is only normed for children ages 8-24 months of age and could not be administered and evaluated accurately for children when they were seen at ages 3-5. Thus, it was not possible to track social motivation and attention shifting behaviors across infancy to early childhood in this study. Future longitudinal studies can utilize and video record alternative behavioral measures that can be used with a larger age range may clarify the continued impact of social motivation and attention shifting as the symptoms of ASD become clearer in early childhood.

Summary and Conclusions

To date, no research studies have thoroughly evaluated and compared two proposed theoretical models for the development of ASD symptoms in early childhood. The present study aimed to build an understanding of the developmental trajectories occurring in early development in ASD and their alignment with these two theories. The Social Motivation theory proposes that individuals with ASD are less motivated by social interactions, engagement, and information early in life, which leads to later impairments in social communication and interaction. The Attention theory suggests that as infants, individuals with ASD have atypical attention shifting between stimuli in their environment and they are

getting “stuck” on different information and not appropriately integrating their social and non-social world. Results from the present study better support the Social Motivation theory in that decreased attention to people at 13 months predicted lower frequency of overall attention shifting particularly shifting that involves people at 22 months, which in turn predicted higher ASD symptom severity during the preschool years. This same trajectory was not present when examining non-social shifting suggesting that social-specific attention and attention shifting is key in the developmental unfolding of ASD symptoms, further supporting the Social Motivation theory. However, future studies are needed that include a larger sample size, a typically developing comparison group, as well as a better understanding of attention processes occurring prior to 13 months of age.

Table 1

Group Differences in Standardized Measures at Time 1 and Time 2: Mean (Standard Deviation)

	Time 1			Time 2		
	No ASD (n=30)	ASD (n=13)	*p*	No ASD (n=30)	ASD (n=13)	*p*
MSEL Standard Score [a]	84.53 (14.31)	83.15 (11.96)	.76	97.60 (15.80)	84.85 (24.55)	.10
MSEL Visual Reception	45.90 (10.24)	46.92 (10.68)	.77	48.97 (7.42)	50.15 (16.57)	.81
MSEL Fine Motor [b]	49.83 (8.20)	51.92 (5.87)	.41	47.47 (9.87)	36.85 (12.14)	.004**
MSEL Receptive Language [a]	33.97 (11.65)	34.31 (11.19)	.93	50.80 (14.96)	43.31 (16.58)	.15
MSEL Expressive Language [a]	37.70 (12.56)	31.31 (10.19)	.11	47.37 (8.35)	35.77 (16.23)	.03*
ADOS Social Affect				6.23 (3.88)	10.15 (3.63)	.003**
ADOS RRB				3.20 (1.61)	5.15 (1.73)	.001**
ADOS Total Score				9.43 (4.64)	15.31 (4.39)	<.001**

Note: Mullen Scales of Early Learning, Early Learning Composite Standard Score (MSEL Standard Score) has a mean of 100 and standard deviation of 10; MSEL Visual Reception, Fine Motor, Receptive Language, Expressive Language T-Scores have a mean of 50 and standard deviation of 10; Autism Diagnostic Observation Schedule Module 1 (ADOS) is for children with single words or no speech; ADOS RRB=ADOS Restricted and Repetitive Behavior.

*$p < .05$. **$p < .01$; a: $p<.05$ between Time 1 and Time 2 No ASD group.

b: $p<.05$ between Time 1 and Time 2 ASD group

Table 2

Demographics at Time 3: Mean (Standard Deviation)

	ASD (n=13)	No ASD (n=30)	p
Time 3 Chronological Age (months)	56.15 (8.13)	52.13 (11.85)	.20
Gender (% male)	76.9% (n=10)	63.3% (n=19)	.38
Race (% Caucasian)	76.9% (n=10)	86.7% (n=26)	.49
DAS-2 GCA	104.54 (16.17)	104.07 (10.12)	.91
ADOS Comparison Score	6.50 (1.90)	2.63 (2.04)	<.001**
RBS-R Total Score	30.31 (21.45)	10.47 (13.07)	.001**

Notes. Differential Ability Scales-2, General Conceptual Ability (DAS-2 GCA) has a mean of 100 and standard deviation of 10; Autism Diagnostic Observation Schedule (ADOS); Repetitive Behavior Scale-Revised (RBS-R) is a parent-report measure of RRBs in individuals with ASD; *p < .05. **p < .01

Table 3

CSBS Measures of Looking at People, Attention Shifting, RSM Behavior: Mean (standard deviation)

	Time 1	Time 2	*p*
Looking at People	11.2% (5.1)	9.7% (4.5)	.02*
Total Shifting	3.46 per min (1.03)	3.56 per min (1.13)	.89
Social Shifting	2.27 per min (.95)	2.12 per min (1.11)	22
Non-Social Shifting	1.19 per min (.48)	1.43 per min (.50)	.04*
RSM with Body	2.78 (2.44)	1.23 (1.50)	<.001**
RSM with Objects	2.38 (2.42)	1.69 (1.87)	.24
RSM Total	5.15 (3.48)	2.92 (2.80)	<.001**

Notes. RSM=Repetitive and Stereotyped Movements derived from the coding scheme from Wetherby & Morgan (2007).

*$p < .05$; **$p < .01$

Table 4

Participant Characteristics for CSBS Measures of Looking at People, Attention Shifting, and RSM Behavior: Mean (standard deviation) and significance levels of t-test comparing the diagnostic groups (p)

	Time 1			Time 2		
	No ASD	ASD	*p*	No ASD	ASD	*p*
Looking at People [b]	11.4% (5.0)	11.1% (5.8)	.88	10.5% (4.4)	8.3% (4.7)	.15
Total Shifting (per min)	3.47 (.96)	3.46 (1.24)	.79	3.96 (1.04)	2.67 (.89)	.001*
Social Shifting (per min) [b]	2.28 (.93)	2.31 (1.07)	.93	2.46 (1.08)	1.43 (.91)	.006**
Non-Social Shifting (per min) [a]	1.20 (.35)	1.18 (.76)	.95	1.50 (.58)	1.30 (.34)	.17
RSM with Body [ab]	2.59 (2.17)	3.33 (3.06)	.40	1.12 (1.26)	1.54 (1.90)	.42
RSM with Objects	2.44 (2.29)	2.42 (2.78)	.97	1.36 (1.25)	2.23 (2.68)	.18
RSM Total [a]	5.04 (3.43)	5.75 (3.62)	.56	2.48 (2.04)	3.77 (3.92)	.19

Notes.
RSM: Repetitive and Stereotyped Movements.

*$p < .05$. **$p < .01$.

a: p<.05 between Time 1 and Time 2 No ASD group.

b: p<.05 between Time 1 and Time 2 ASD group

Table 5

Pearson Correlations Between CSBS Measures of Looking at People, Attention Shifting, and RSM Behavior

Variables	T1 People	T2 People	T1 Shift	T2 Shift	T1 S-Shift	T2 S-Shift	T1 NS-Shift	T2 NS-Shift	T1 RSM	T2 RSM
T1 Looking at People	--									
T2 Looking at People	.57**	--								
T1 Shifting	.69**	.28	--							
T2 Shifting	.39**	.65**	.17	--						
T1 Social Shift	.91**	.48**	.88**	.40**	--					
T2 Social Shift	.57**	.84**	.28	.90**	.48**	--				
T1 Non-Social Shift	-.32*	-.33*	.39**	-.30^	-.08	-.34*	--			
T2 Non-Social Shift	-.37*	-.39*	-.23	.26	-.27^	-.18	.08	--		
T1 RSM Total	-.05	-.16	-.12	-.22	-.14	-.22	.03	.03	--	
T2 RSM Total	-.17	-.28	-.32*	-.17	-.25	-.22	-.19	.12	.53**	--

Notes. Time 1 (T1) occurred at 13 months of age; Time 2 occurred at 22 months of age; Social Shift (S-Shift) includes attention shifting with people; Non-social Shift (NS-Shift) includes attention shift with objects only.

*$p < .05$. **$p < .01$. ^$p<.07$.

Table 6

Correlation Between Cognitive Ability and ASD Symptoms

Variables	T1 MSEL SS	T2 MSEL SS	T3 DAS-2 GCA	T2 ADOS Score	T3 ADOS Score
T1 MSEL SS	--				
T2 MSEL SS	.46**	--			
T3 DAS-2 GCA	.52**	.52**	--		
T2 ADOS Score	-.04	-.43**	.09	--	
T3 ADOS Score	-.32*	-.61**	-.12	.53**	--

Notes. Time 1 (T1) occurred at 13 months of age; Time 2 (T2) occurred at 22 months of age; Time 3 occurred at 3-5 years of age; Mullen Scales of Early Learning, Early Learning Composite Standard Score (MSEL Standard Score) has a mean of 100 and standard deviation of 10; Differential Ability Scales-2, General Conceptual Ability (DAS-2 GCA) has a mean of 100 and standard deviation of 10; Time 2 Autism Diagnostic Observation Schedule-2 Module 1 (T2 ADOS Score) is utilizing the total algorithm score for ADOS at 22 months of age. Time 3 Autism Diagnostic Observation Schedule-2 Symptom Severity Score (T3 ADOS Score) utilizes the symptom severity score which allows for comparison of ASD symptom severity across different ADOS modules for children with varied language abilities.

*$p < .05$. **$p < .01$.

Table 7

Correlation Between CSBS Measures of Looking at People, Attention Shifting, and RSM Behavior with Cognitive Ability and ASD Symptoms

Variables	T1 MSEL SS	T2 MSEL SS	T3 DAS-2 GCA	T2 ADOS Score	T3 ADOS Score
T1 Looking at People	.19	.42**	.19	-.30*	.10
T2 Looking at People	.18	.31*	.19	-.35*	-.19
T1 Shifting	.23	.43**	.18	-.18	-.10
T2 Shifting	.24	.46**	.20	-.49**	-.45**
T1 Social Shift	.26	.49**	.21	-.29^	-.12
T2 Social Shift	.28	.45**	.28	-.53**	-.38*
T1 Non-Social Shift	-.01	-.05	-.04	.18	02
T2 Non-Social Shift	-.07	.05	-.17	.06	-.18
T1 RSM Total	-.35*	-.34*	-.13	.29^	-.19
T2 RSM Total	-.14	-.34*	-.17	.31^	.27

Notes. Time 1 (T1) occurred at 13 months of age; Time 2 (T2) occurred at 22 months of age; Time 3 occurred at 3-5 years of age; Mullen Scales of Early Learning, Early Learning Composite Standard Score (MSEL SS) and Differential Ability Scales-2, General Conceptual Ability (DAS-2 GCA) have a mean of 100 and standard deviation of 10; Time 2 Autism Diagnostic Observation Schedule-2 Module 1 (T2 ADOS Score) is from the total algorithm score for ADOS at 22 months of age. Time 3 Autism Diagnostic Observation Schedule-2 Symptom Severity Score (T3 ADOS Score) utilizes the symptom severity score which allows for comparison of ASD symptom severity across different ADOS modules for children with varied language abilities. RSM=Repetitive and Stereotyped Movements Scale; *$p < .05$; **$p < .01$; ^$p<.07$

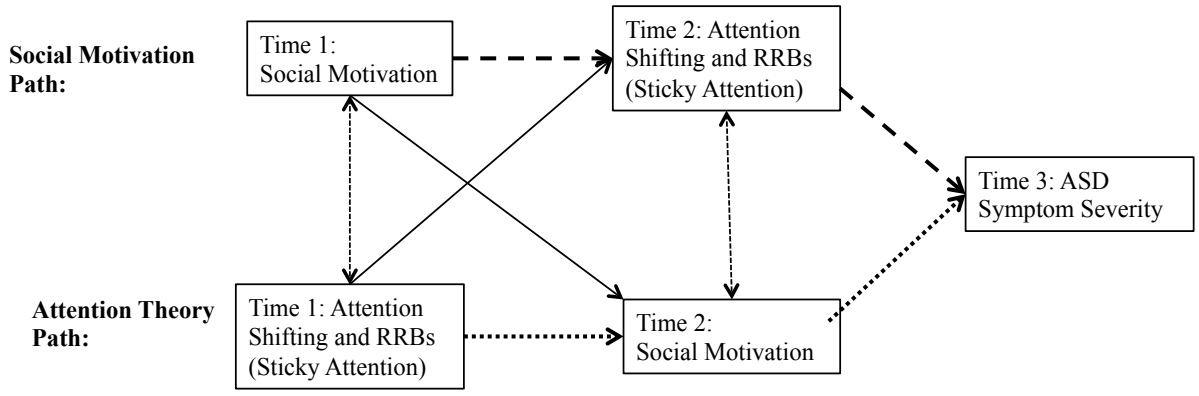

Figure 1. Proposed path analysis model to assess Social Motivation and Attention Theories. The dashed line represents the proposed trajectory of decreased social motivation at Time 1 predicting impaired sticky attention (i.e., attention shifting at RRBs) at Time 2 followed by increased ASD symptom severity at Time 3. The dotted line represents the proposed Attention theory trajectory with initial impairments in sticky attention (i.e., attention shifting at RRBs) at Time 1 predicting decreased social motivation at Time 2 followed by increased ASD symptom severity at Time 3.

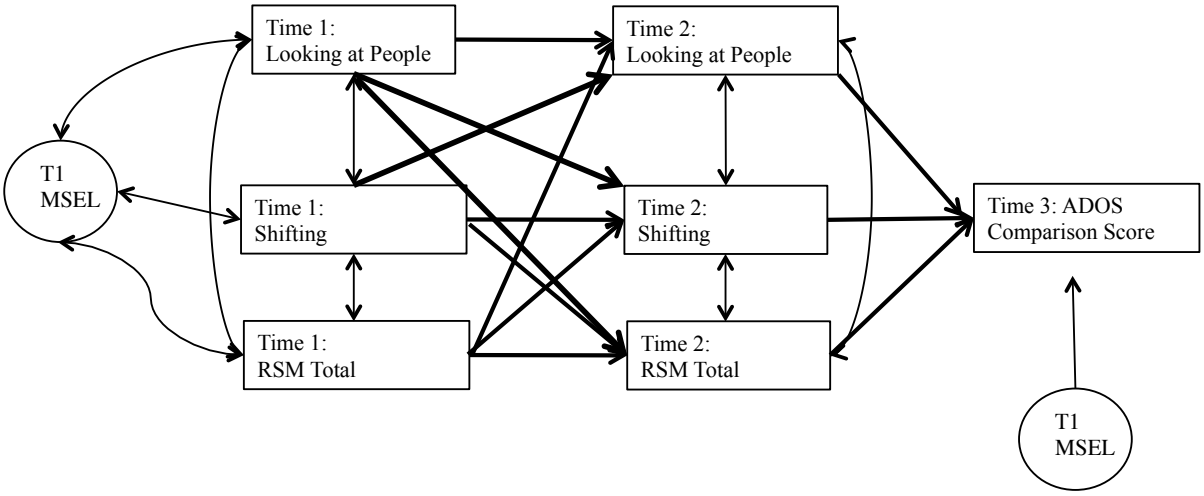

Figure 2. Proposed full model of path analysis with Looking at People, Attention Shifting, and Repetitive and Stereotyped Movements (RSM) Total as separate predictors at Time 1 and Time 2. Time 1 Mullen Scales of Early Learning, Early Learning Composite Standard Score (T1 MSEL) is included as a covariate to account for initial variability in cognitive skills.

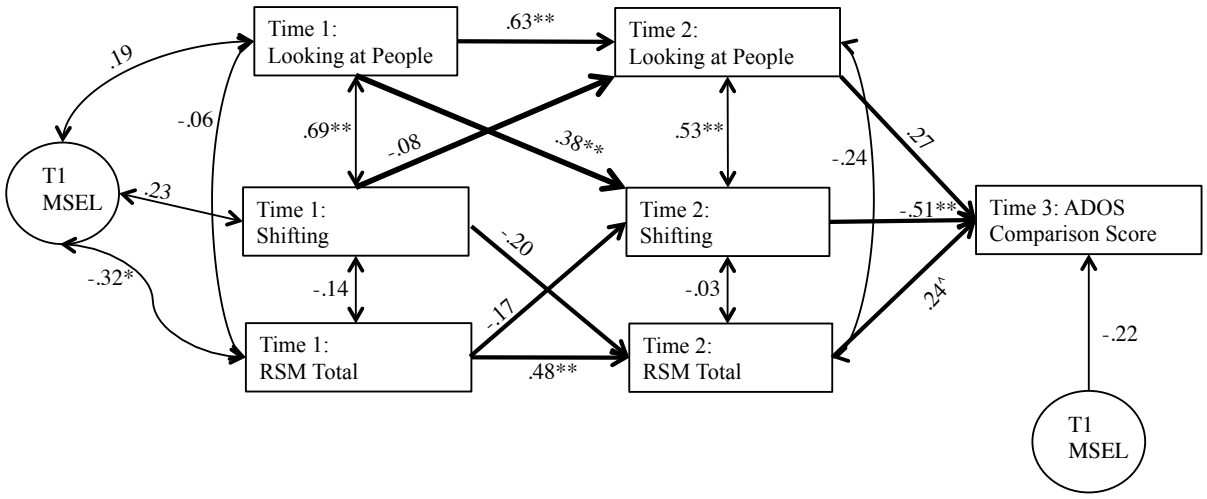

Figure 3. Results of path analysis examining the direct and indirect effects of Time 1 and Time 2 Variables of Looking at People, Attention Shifting, and Repetitive and Stereotyped Movements (RSM) predicting Time 3 ADOS symptom severity scores. Mullen Scales of Early Learning Standard Score at Time 1 (T1 MSEL) is included as a covariate. The full model was trimmed to better identify significant direct pathways, while maintaining correlations between variables within Time 1 and Time 2. Standardized estimates are displayed for direct pathways T3 ADOS Comparison Score: R^2=.32 $^{**}p<.01$; $^{*}p<.05$; $^{\wedge}p<.07$

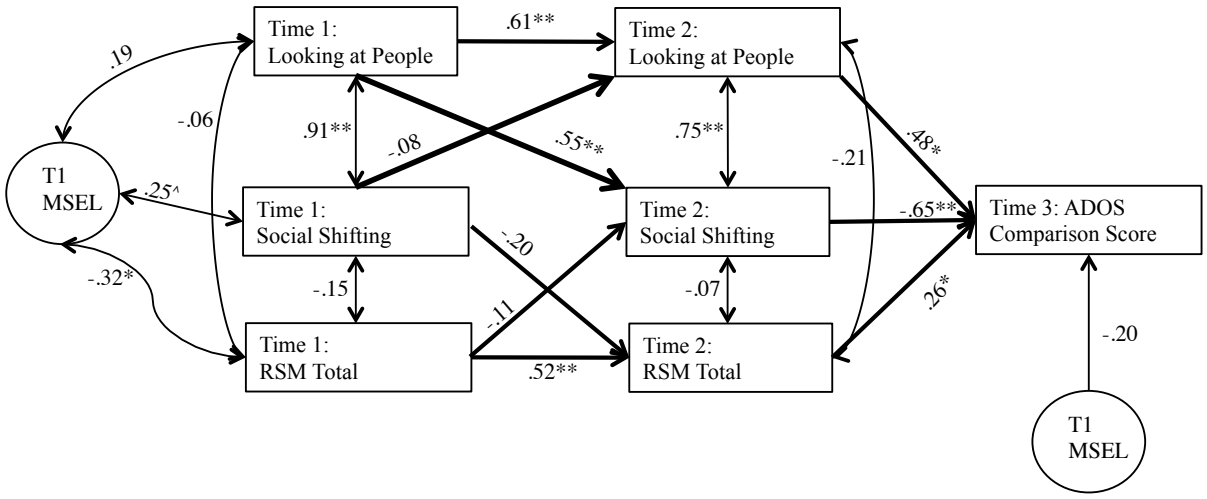

Figure 4. Results of path analysis examining the direct and indirect effects of Time 1 and Time 2 Variables of Looking at People, Social Attention Shifting (i.e., attention shifting that includes a person), and Repetitive and Stereotyped Movements (RSM) predicting Time 3 ADOS symptom severity scores. Mullen Scales of Early Learning Standard Score at Time 1 (T1 MSEL) is included as a covariate. The full model was trimmed to better identify significant direct pathways, while maintaining correlations between variables within Time 1 and Time 2. Standardized estimates are displayed for direct pathways T3 ADOS Comparison Score: R^2=.288; $^{**}p<.01$; $^{*}p<.05$.

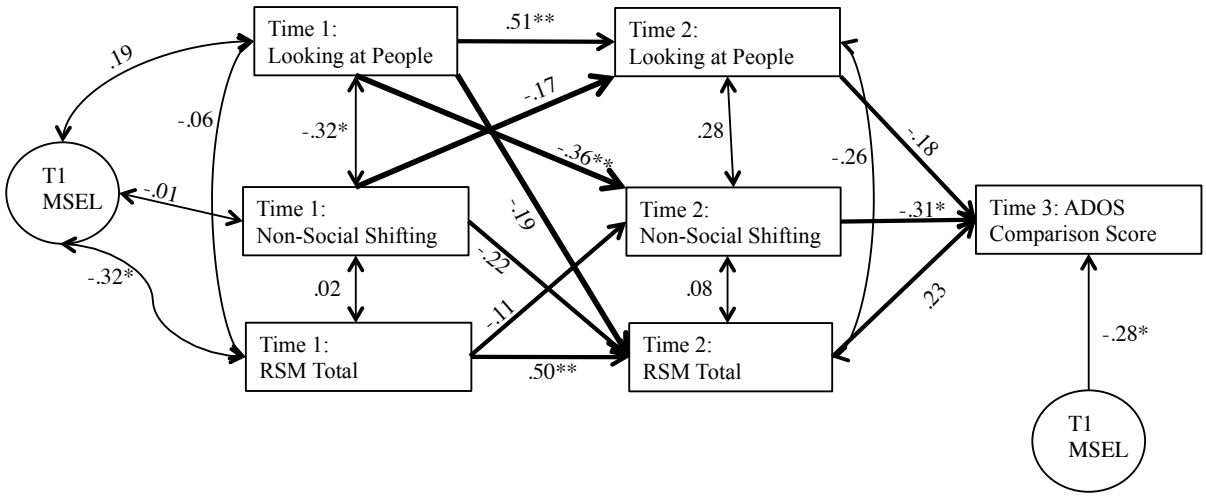

Figure 5. Results of path analysis examining the direct and indirect effects of Time 1 and Time 2 Variables of Looking at People, Non-social Attention Shifting (i.e., attention shifting with objects only), and Repetitive and Stereotyped Movements (RSM) predicting Time 3 ADOS symptom severity scores. Mullen Scales of Early Learning Standard Score at Time 1 (T1 MSEL) is included as a covariate. The full model was trimmed to better identify significant direct pathways, while maintaining correlations between variables within Time 1 and Time 2. Standardized estimates are displayed for direct pathways T3 ADOS Comparison Score: R^2=.252; $^{**}p<.01$; $^{*}p<.05$.

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