Sensory processing function and early intervention programs for toddlers with early signs of autism

DISSERTATION

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By

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Abstract

The prevalence of sensory processing deficits in children with Autism Spectrum Disorders (ASD) ranges from 69 to 95%. Sensory processing deficits are thought to contribute to the clinical presentation and functional difficulties experienced by individuals with ASD. Detection of sensory processing deficits during the second year of life has the potential to significantly impact the development of the disorder. Although sensory processing dysfunction in ASD is widely documented in the literature, the majority of the research in this area is limited to retrospective reviews and examination of dysfunction occurring during preschool years or later. Additionally, the majority of research in this area utilizes parent report and/or videotaped analysis and very few studies utilize neurophysiological assessments. No research study to date has employed a multi-modal assessment protocol utilizing both neurophysiological and clinical (parent report and direct observation) assessment tools to measure sensory processing function.

Early identification of sensory processing deficits has the potential to lead to early intervention in individuals who may go on to develop ASD. Early intervention can significantly impact sensory processing deficits and ASD characteristics. In order to move toward addressing sensory processing in an early intervention program, sensory
processing function during the first few critical years of life must be better understood.
The goals of this project are to: (1) describe sensory processing function and dysfunction using a multi-modal assessment protocol during the second year of life (2) examine differences in sensory processing between toddlers with and without early signs of ASD (3) determine the relative relationship between developmental skills and sensory processing abilities with the presence of early signs of ASD and (4) to examine the effectiveness of early intervention on ameliorating the clinical symptoms of autism for individuals with or at risk for a diagnosis of autism between 0-24 months of age.
Dedication

To my husband, Steve, and my son, Tyler. This would not have been possible without their love and support throughout this process.

To my parents who gave me continued encouragement to set high goals in life and that anything was possible.
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Chapter 1: General Introduction

1.1 Autism Spectrum Disorder Overview

Autism Spectrum Disorder (ASD) has progressed from a rare diagnosis to a common disorder affecting approximately 1 in 91 children (Kogan et al., 2009). The exponential rise in ASD prevalence makes it more common than childhood cancer, juvenile diabetes, spina bifida, and Down Syndrome combined (Filipek et al., 1999). According to the U.S. Department of Education, autism is growing at an exorbitant rate of 10-17 percent per year (NIDRR Long Range Plan, 2009). These facts, along with the life-long impact of the disorder, make it an urgent public health concern that warrants extensive research.
Autism Spectrum Disorder is a neurodevelopmental disorder characterized by core deficits in communication and socialization along with the presence of restricted, repetitive, and stereotyped behaviors (Fig. 1) (American Psychiatric Association, 1994; American Psychiatric Association, 2000; Baker, Lane, Angley, & Young, 2008; Filipek et al., 1999; Filipek et al., 2000). In addition to core deficits, individuals with ASD present with a variety of associated clinical features including difficulties with attention, challenges with orienting behaviors, and sensory processing deficits (Kern et al., 2006). There are five disorders included within the “autism spectrum”: (1) Autism (2) Pervasive Developmental Disorder-Not Otherwise Specified (PDD-NOS) (3) Asperger’s Syndrome (4) Childhood Disintegrative Disorder (CDD) and (5) Rett Syndrome. The first three mentioned are the most prevalent (Matson, González, & Wilkins, 2009). For the
remainder of this dissertation, the term ASD will refer to individuals with autism, Asperger’s Syndrome, and PDD-NOS.

The cost of ASD is a huge financial burden on individual families and society as a whole. Specifically, recent reports estimate that the average family with an individual diagnosed with ASD pays an additional 3 to 5 million dollars over a lifetime more than a family without a member with ASD. In the United States, it costs an average of 90 billion dollars a year to support ASD (Ganz, 2006; Ganz, 2007; Knapp, Romeo, & Beecham, 2009). Research indicates that even minor improvements made in functional independence and/or adaptive behavior can reduce this cost significantly (Kogan et al., 2008).

1.2 Early Detection and Diagnosis

There are inconsistencies in the literature regarding initial age of onset and detectability of ASD. Although it has been detected in children as early as 18 months, typically, it is not diagnosed this early (Filipek et al., 1999; Filipek et al., 2000). The average age for diagnosis of ASD ranges between 2 to 7 years (Filipek et al., 2000; Howlin & Moorf, 1997; Osterling & Dawson, 1994; Trillingsgaard, Ulsted Sørensen, Němec, & Jørgensen, 2005). More than fifty percent of parents of children with ASD have suspicion of a developmental issue during their child’s first year of life (Baranek, 1999; Osterling & Dawson, 1994; Osterling, Dawson, & Munson, 2002; Werner, Dawson, Osterling, & Dinno, 2000; Young, Brewer, & Pattison, 2003). Unfortunately, parental
concerns are often ignored secondary to individual variability inherent with typical development. (Filipek et al., 2000; Osterling & Dawson, 1994). Pediatricians and other health professionals are hesitant to diagnose ASD under the age of two for the following reasons:

1. A diagnosis of ASD is highly reliant on language and communication skills, which often develop later in infancy (Lord et al., 2000).

2. There is a wide variation in typical child development (Filipek et al., 2000)

3. Uncertainty that a child’s diagnosis at two years old will be the same at a later age (Kasari & Wong, 2002)

However, research indicates a firm ASD diagnosis at age two has been generally accepted as both reliable and stable over time (Cox et al., 1999; Lord, 1995; Lord et al., 2006; Moore & Goodson, 2003). Utilizing retrospective home video analysis, several studies found consistencies in behavioral characteristics evident in children under 2 years of age that were later diagnosed with ASD. These include variability in social engagement (Cox et al., 1999; Kasari & Wong, 2002; Mars, Mauk, & Dowrick, 1998; Werner et al., 2000), less frequent joint attention attempts (Moore & Goodson, 2003), limited imitation skills (Kasari & Wong, 2002), poor eye contact or visual avoidance, absence of response to familiar voices (Kasari & Wong, 2002), and infrequent orienting to their name (Osterling & Dawson, 1994).
Although the majority of the studies detecting of the early signs of autism are retrospective, there is a move towards studying high-risk infant siblings prospectively. Siblings are at approximately 5-10% higher risk of developing autism than the general population (Szatmari, Jones, Zwaigenbaum, & MacLean, 1998). Although high-risk infant sibling studies are in their preliminary phases, infants as young as 4 months of age have been included in research projects. Prospective research demonstrates that children later diagnosed with ASD exhibited differences in the areas of communication (Chawarska, Klin, & Volkmar, 2008; Landa & Garrett-Mayer, 2006; Volkmar, Chawarska, & Klin, 2008), socialization (Yirmiya et al., 2006; Zwaigenbaum, 2005), visual fixations and eye contact (Klin & Jones, 2008; Wetherby et al., 2004; Zwaigenbaum, 2005) at an early age. As technology advances and continued research defines the many facets of ASD, we have the unique opportunity to provide innovative insights into the neurophysiology and behavioral mechanisms contributing to this disorder.

An accurate and reliable diagnosis of ASD at age two is possible with comprehensive clinical assessments (Moore & Goodson, 2003). Comprehensive assessments typically include parent interview and child observation in concert with cognitive, developmental, and language testing. Hence, a diagnosis based upon a single diagnostic assessment is less reliable. Consensus in the literature calls for making ASD research a priority with a focus on the development and implementation of infant screening and diagnostic tools allowing for intervention to begin at an early age (Bristol-Power & Spinella, 1999; Bryson, Zwaigenbaum, McDermott, Rombough, & Brian, 2008).
Additionally, the American Academy of Pediatrics has published two recent reports recommending the implementation of more aggressive behavioral and educational interventions, earlier identification and increased routine surveillance for ASD (Johnson & Myers, 2007; Myers & Johnson, 2007). The benefits of early identification of ASD include:

1. Allowing intervention to begin sooner therefore possibly reducing the severity of the disorder (Mars et al., 1998; Moore & Goodson, 2003; Rogers & Vismara, 2008; Sigman, Dijamco, Gratier, & Rozga, 2004).

2. Allowing for examination of infant development in infants with or at risk for ASD, broadening our knowledge of how this disorder develops (Sigman et al., 2004).

Early intervention significantly impacts the developmental outcome in individuals with ASD (Dawson, 2008; Rogers, 1996; Rogers & Vismara, 2008; Smith & Dillenbeck, 2006). Improvements in communication, language, IQ, and a decrease in ASD severity have been shown following early intervention (Rogers & Vismara, 2008). Accurate screening and early assessment tools allow specific interventions to be developed (Moore & Goodson, 2003). Furthermore, successfully identifying ASD characteristics at a younger age would encourage appropriate daycare and/or school placements with appropriate support staff specifically trained to address core deficits of ASD (Moore & Goodson, 2003). Researchers are beginning to focus on earlier diagnosis and treatment to capitalize on the evidence suggesting vast improvements in
developmental outcomes following early, intensive interventions (Cohen, Amerine-Dickens, & Smith, 2006; Dawson, 2008; Rogers, 1998; Smith, Groen, & Wynn, 2000).

1.3 Sensory Processing

In addition to social and communication deficits, the majority of individuals with ASD also present with difficulties in processing sensory information. Sensory processing (SP) refers to receiving, modulating, and integrating sensory information and in turn, producing an adaptive response (Schaaf & Miller, 2005). To function adequately in the environment and participate in activities of daily living, one must have adequate sensory processing abilities (Johnson-Ecker & Parham, 1999). SP deficits affect individuals with ASD in many aspects of daily functioning in addition to social, cognitive and sensorimotor development (Dunn, 1997). These deficits affect the way individuals respond to sensory input, communicate, and participate in daily activities. The prevalence of SP deficits in individuals with ASD range from 69-95% (Baker et al., 2008; Baranek, 2002; Baranek, Boyd, Poe, David, & Watson, 2007; Dawson & Watling, 2000; Harrison & Hare, 2004; Kientz & Dunn, 1997; Tomchek & Dunn, 2007). SP deficits are common within this population. They include sensory fascinations, sensitivities, and aversions, as well as difficulty integrating and understanding simultaneous sensory information, involving multiple sensory systems (Grandin, 2000; Grandin, 2004; Rodier, Ingram, Tisdale, & Croog, 1997). Despite the high prevalence rate, disturbances in SP are not universally recognized in individuals with autism nor is there consensus that SP
deficits are specific to ASD diagnosis (Ben-Sasson et al., 2009; Harrison & Hare, 2004; Rogers, Hepburn, & Wehner, 2003). Currently, SP dysfunction is not clearly understood; however, we know that it is not attributable to peripheral sensory deficits (i.e., vision or hearing loss) (Baranek, 2002; Scharre & Creedon, 1992).

Sensory abnormalities have been well documented throughout the lifespan in individuals with autism (Adrien et al., 1991; Adrien et al., 1993; Crane, Goddard, & Pring, 2009; Greenspan & Wieder, 1997; Kern et al., 2006; Kern et al., 2007; Talay-Ongan & Wood, 2000). There is a significant amount of evidence demonstrating that SP deficits emerge early in life and contribute to clinical symptoms associated with ASD (Baranek, Parham, & Bodfish, 2005; Dawson & Watling, 2000; Lane, Young, Baker, & Angley, 2010).

SP deficits identified prior to diagnosis are most often documented using retrospective home videotaped analysis and case studies.

1. Home video analysis: Studies utilizing retrospective videotaped analyses report that infants with ASD mouth objects more frequently, exhibit social touch aversion, respond to their name less frequently, and display decreased orientation to visual stimuli (Baranek, 1999). Osterling and colleagues had similar findings in terms of decreased response to own name, less eye contact (Osterling et al., 2002), covering their ears and engaging in self stimulation (Osterling & Dawson, 1994). Additional studies have identified sensory
processing differences between infants who were later diagnosed with ASD, infants diagnosed with developmental disabilities (Baranek et al., 2006; Baranek, David, Poe, Stone, & Watson, 2006; Rogers et al., 2003), and typical controls (Baranek, Boyd, Poe, David, & Watson, 2007; Kern et al., 2006; Mottron, Dawson, Soulieres, Hubert, & Burack, 2006; Rogers et al., 2003; Talay-Ongan & Wood, 2000; Watling, Deitz, & White, 2001). Kientz and Dunn (1997) found that behaviors in young children with autism (ages 3-10) differed on 85% of the Sensory Profile compared to children with typical development. Similarly, Gillberg and colleagues (1990), examined children with ASD under 3 years (N=12) and found that 83% had sensory processing difficulties.

2. A case study examining sensory processing of an infant with ASD reported over-sensitivity to loud environments and over-sensitivity to tactile stimulation at 2.5 months of age followed by over-sensitivity to visual and auditory stimuli at 9 months of age (Dawson, Osterling, Meltzoff, & Kuhl, 2000).

Additionally, SP deficits are commonly documented in individuals with a diagnosis of ASD in studies utilizing parent report and personal accounts.

1. Parent report: Parent report can be used retrospectively or prospectively to measure sensory processing in research and clinical settings. Utilizing parent report, caregivers document the frequency of their child’s behaviors in response to sensory stimuli within their typical environment. For example, using the
Sensory Experience Questionnaire, parents of children with ASD (preschool age) indicated that 69% of the sample exhibited sensory processing deficits. Specifically, 63% of these children were under-responsive to sensory stimuli and 56% were over-responsive (Baranek et al., 2005). Additionally, Rogers and colleagues found that parents of children with ASD (ages 26-41 months) most often indicated that their children exhibited taste/smell and tactile sensitivities according to the Short Sensory Profile (Rogers et al., 2003).

2. Personal account: Several firsthand accounts have been published with a recurring theme regarding unusual sensory experiences occurring throughout their lifetime. Abnormal sensory processing has been reported in all sensory systems including auditory, visual, tactile, gustatory, olfactory, and proprioception. These individuals report a combination of over- and under-responsivity to sensory stimuli and often report a fluctuation between the two (Cesaroni & Garber, 1991; Grandin, 1984; Grandin, 1992; Grandin, 2000; Grandin, 2004; O'Neill & Jones, 1997; Volkmar & Cohen, 1985).

Sensory processing deficits present differently in children with and without ASD, affect each sensory modality, and can affect each sensory modality in varying degrees (Adamson, O'Hare, & Graham, 2006; Leekam, Nieto, Libby, Wing, & Gould, 2007). These findings impacted the push to develop screening tools for detecting SP deficits at an earlier age (Stone, Coonrod, & Ousley, 2000; Wetherby et al., 2004).
1.3.1 Auditory Processing

The auditory modality is one of the most commonly affected sensory systems in individuals with ASD (Tomchek & Dunn, 2007). Auditory processing difficulties were identified in 100% of children with ASD prior to 24 months of age (Dahlgren & Gillberg, 1989). Similarly, Baranek and colleagues identified auditory sensory processing disorder in 30% of children with ASD and 11% of adults with ASD (Baranek, Foster, & Berkson, 1997). Further, individuals with ASD between the ages 3 to 56 were reported to have more severe deficits in auditory processing as compared to typical controls (Kern et al., 2007).

Individuals with ASD exhibit a variety of under- and over-responsive behavioral patterns to auditory stimuli. Specifically, individuals with ASD exhibit difficulty with auditory processing and modulating contextually relevant auditory stimuli (Jones, Quigney, & Huws, 2003; Paul, Chawarska, Fowler, Cicchetti, & Volkmar, 2007; Siegal & Blades, 2003). Children with ASD also exhibited greater difficulty with auditory filtering than children with other developmental disabilities (Rogers et al., 2003; Tomchek & Dunn, 2007; Wiggins, Robins, Bakeman, & Adamson, 2009). Additionally, individuals with ASD demonstrate the following behavioral patterns: decreased orienting responses to sounds (Baranek, Boyd, Poe, David, & Watson, 2007), increased sensitivity to auditory stimuli (Dahlgren & Gillberg, 1989), and under-responsiveness to sounds (Baranek, 1999; Osterling & Dawson, 1994).
1.3.2 Impact of Sensory Processing Deficits

Sensory processing difficulties are apparent during infancy in individuals at-risk for autism and continue to negatively impact daily functioning as time progresses (Baranek et al., 2008). These difficulties affect each individual in a unique way. Individuals displaying over-responsive behavioral patterns to sensory stimuli might avoid unwanted or uncomfortable sensory experiences, limiting his or her ability to explore and learn from the environment (Baranek, 2002; Baranek, Boyd, Poe, David, & Watson, 2007). Other individuals may exhibit difficulties with dietary restrictions (Smith, Roux, Naidoo, & Venter, 2005), sound/noise avoidance or increased sensitivity (Grandin, 1984), texture avoidance during play (Talay-Ongan & Wood, 2000), excessive seeking of sensory stimuli, unusual preoccupations with smells, and fearful responses to typical activities involving touch, movement and/or sound (Kientz & Dunn, 1997). SP deficits can cause impaired occupational engagement and decreased social participation in some individuals which often decreases their overall quality of life (Baranek et al., 2008). Additionally, individuals with sensory processing deficits can exhibit unreliable motor responses, have difficulty coordinating their bodies, have decreased motor imitation and signs of dyspraxia secondary to sensory processing deficits (Koenig & Kinnealey, 2008). Furthermore, SP deficits not only affect the individual but often negatively impact the family unit. Family members often make adaptations to
accommodate a child’s sensory response patterns and/or reactions to sensory experiences that influence the quality, type, and amount of shared social experiences or family events.

Deficits in sensory processing have been linked to core symptoms of ASD including repetitive behaviors, communication, and socialization deficits (Ashburner, Ziviani, & Rodger, 2008; Baker et al., 2008; Just, Cherkassky, Keller, & Minshew, 2004). First, abnormal sensory processing abilities correlate with the presence of stereotypic, rigid, and repetitive behaviors (Boyd, McBee, Holtzclaw, Baranek, & Bodfish, 2009; Watt, Wetherby, Barber, & Morgan, 2008). Additionally, impairments in the auditory system are thought to negatively affect social development and have an impact on language and communication impairments present in individuals with ASD. SP deficits could eventually be a valuable tool used to screen individuals for early signs of ASD, help tailor individualized intervention programs, and determine the most effective intervention strategies to implement (Baranek, 1999).

1.3.3 Measurement of Sensory Processing

Researchers have used a variety of assessment tools to document the occurrence of SP deficits. As discussed, the vast majority of clinical research studies assessing SP deficits are retrospective and utilize behavioral assessment tools that are primarily based upon parent report (Dunn, 1997; Dunn & Daniels, 2002). Few studies have utilized direct observation of SP (Baranek, Boyd, Poe, David, & Watson, 2007;
DeGangi, Berk, & Greenspan, 1988; DeGangi & Greenspan, 1989) and fewer studies utilize neurophysiological assessments. Although the majority of research involving SP and ASD are reliant on retrospective methodology, studies utilizing prospective designs are becoming more prevalent. Prospective high-risk infant studies are becoming more common allowing investigators to examine the presence or absence of behaviors at a particular point in time rather than relying on retrospective report. Further, implementing high-risk, infant sibling studies, researchers are able to focus on ASD recurrence risks and familial contributors to ASD (Bryson et al., 2007). Very few studies examine SP concurrently with parent report, direct observation and neurophysiologic measures. McIntosh and colleagues reported that children aged 3 to 9 years with sensory modulation disorder exhibited atypical responses on electrodermal responses (EDR) to sensory stimuli along with increased atypical behavioral responses as documented on parent report (McIntosh, Miller, Shyu, & Hagerman, 1999). Similar findings were reported for a group of children, aged 5-13 years, with Attention Deficit Hyperactivity Disorder (ADHD). Children with ADHD exhibited greater atypical responses to EDR and parent report measures as compared to typical controls (Mangeot et al., 2001). To our knowledge, there are no studies measuring clinical and neurophysiological assessments concurrently with very young children with early signs of ASD.
1.3.4 Limitations with Current Behavioral Sensory Processing Research

Current research involving sensory processing and individuals with ASD has a number of limitations:

1. The vast majority of research is retrospective.

2. Studies using a prospective design and implementing direct observation measures are sparse (Baranek, Boyd, Poe, David, & Watson, 2007; Chawarska et al., 2008).

3. The outcome measures and methodological designs are often highly variable.

4. Videotaped studies have methodology issues including lack of tester blindness, variability in the context and settings recorded, difficulty approximating the age of the individual, and lack of standardization (Iarocci & McDonald, 2006).

5. There are subjective biases inherent with parent report measures (Chawarska et al., 2008).

6. The generalizability of findings is limited due to small sample sizes (Ben-Sasson et al., 2009; Ermer & Dunn, 1998; Kientz & Dunn, 1997), heterogeneity within the groups of children with ASD (Eide, 2003; Ermer & Dunn, 1998; Kientz & Dunn, 1997; Talay-Ongan & Wood, 2000), no control group (Kientz & Dunn, 1997; Talay-Ongan & Wood, 2000; Watling et al., 2001).
7. Few studies compare SP deficits in children with ASD to children with other developmental delays. This leaves the question of whether SP deficits are unique to ASD or are common among all developmental disabilities.

8. The developmental progression of SP function has been rarely studied. Few studies are available exhibiting mixed results (Baranek et al., 2007).

Research studies with higher methodological rigor, larger sample sizes, more homogeneous samples, and similar assessment tools are needed to allow for comparisons across studies.

1.8 Neurophysiologic Assessment of Sensory Processing

Neurophysiologic research of SP function in individuals with ASD is in its infancy. Event Related Potentials (ERP), Magnetoencephalography (MEG), and electrodermal responses (EDR) are some of the neurophysiological tools newly utilized to evaluate sensory processing. MEG recordings of mismatch field (MMF) have been used to indicate disturbances in the ability of individuals with ASD to discriminate the physical properties of two consecutively presented auditory stimuli. Specifically, participants (aged 8-32 years) with ASD exhibited either a significant reduction in amplitude or an absent MMF that differed significantly from the control group (Tecchio et al., 2003). Miller et al. used electrodermal responses (EDR) showing increased sensory sensitivity to auditory stimuli in young adults with Fragile X Syndrome (Miller et al., 1999).
ERP is a physiological measure of sensory processing and attention. It assesses the association between brain structure, function, and behavior related to processing sensory stimuli as it is actually occurring (Davies & Gavin, 2007). The brain produces electrical activity in response to an internal or external event. ERP provides insight into how the brain processes and integrates particular sensory stimuli. ERP waveforms produced by the brain can be broken down into specific components. The sequences of the components measured reflect the flow of information through the brain and the different stages of processing (Schmidt & Segalowitz, 2008). In our study, we examined the mismatch negativity (MMN) component of ERP which typically occurs between 100-300 ms after the stimulus onset (Lepistö et al., 2005).

The mismatched negativity (MMN) component is elicited by an oddball paradigm, involving standard and deviant stimuli (Cheour et al., 1998; Schmidt & Segalowitz, 2008). Although the amplitude of MMN is relatively similar in infants and adults, the mean latency decreases with age (Cheour et al., 1998). The auditory system appears to mature very early in life (Cheour et al., 1998) and remains relatively stable over time (Cheour et al., 1998; Kushnerenko, Čeponiene, Balan, Fellman, & Naatanen, 2002). MMN has been recorded in previous research studies using changes in pure tones and speech sounds (Cheour et al., 1998; Čeponienė, Lepistö, Shestakova, Vanhala, Alku, Näätänen, & Yaguchi, 2003b). The main source that generates MMN is in the bilateral auditory cortex (Cheour et al., 1998) (Fig. 1.2). MMN is a valuable tool for studying auditory discrimination in individuals of all ages as it is elicited by unattended
stimuli and can be recorded in a variety of states, including sleep (Naatanen, Paavilainen, Alho, & Reinikainen, 1993). Auditory discrimination has been successfully studied in clinical groups including Alzheimer’s, schizophrenia, and Parkinson’s disease (Cheour et al., 1998). Recent studies suggest that MMN has the potential to be utilized for diagnostic and clinical purposes (Schmidt & Segalowitz, 2008).

Figure 1.2: Bilateral auditory cortex: main source for generation of MMN (www.brainconnection.positscience.com)

MMN was examined longitudinally in the first year of life by several investigators. Evidence from these studies suggests that infants exhibit stability in regards to amplitude and latency of the components during the first year of life (Cheour et al., 1998; Kushnerenko et al., 2002). Additionally, infant grand averages exhibited a MMN component approximately 75% to 100% (Kushnerenko et al., 2002) of the time
during the first year of life (Cheour et al., 1998; Kushnerenko et al., 2002). However, there was significant variability in individual MMN components within the same subjects over time (Kushnerenko et al., 2002).

1.8.1 Mismatch Negativity and ASD

As discussed previously, many individuals with ASD exhibit difficulty with auditory processing. Research has shown that individuals with ASD and Receptive Developmental Language Disorder (RDLD) demonstrate typical functioning with the components leading up to the MMN. Specifically, they exhibited normal early (0-10 ms) and midlatency (10-80 ms) auditory evoked responses (Courchesne, Lincoln, Yeung-Courchesne, Elmasian, & Grillon, 1989). MMN is an excellent pre-attentive measure of sensory processing, allowing researchers to exclude the possibility of an effect relating to impaired attention, motor reaction, or task related responses (Schmidt & Segalowitz, 2008). The MMN component is elicited with the detection of a novel stimulus. Individuals with ASD prefer sameness which could play a role in the difficulty these individuals have in detecting and attending to novel stimuli (Jeste & Nelson, 2009). Individuals with ASD could be either under-responsive or over-responsive to changes in stimuli. Dunn and colleagues found a smaller amplitude in the MMN component of children with autism (ages 6-12 years) as compared to typical controls using non-speech stimuli during unattended conditions but similar MMN during attended stimuli (Dunn, Gomes & Gavel, 2008). Similarly, Kuhl and colleagues reported that individuals with
autism displayed smaller MMN amplitude to unattended speech stimuli (Kuhl, Coffey-Corina, Padden, & Dawson, 2005). Reduced MMN amplitude in individuals with ASD for unattended stimuli but not attended stimuli supports the theory that individuals with ASD have difficulty automatically processing auditory stimuli (Dunn et al., 2008). Impairments in automatic processing of auditory stimuli could partly explain why individuals with ASD have difficulty with language development (Dunn et al., 2008).

1.9 Project Significance

Examination of sensory processing function and dysfunction in young children during the second year of life with and without early signs of autism is imperative. Retrospective research indicates that SP deficits can be detected during the first two years of life. Identifying these deficits early in development can lead to early intervention focused on improving sensory processing function. Early intervention can significantly impact the developmental outcome in individuals.

The aims of this research project were to: (1) describe sensory processing function in toddlers aged 12-24 months (2) examine differences in SP function between toddlers with and without early signs of ASD using clinical and neurophysiological measures and (3) examine the relative contribution of sensory processing patterns, language abilities, motor skills, and cognition to the prediction of early signs of autism in young children 12-24 months of age. The aim of this systematic review was to
determine if early intervention services ameliorated ASD characteristics for children with or at risk for ASD.
Chapter 2: Sensory processing function in toddlers with and without early signs of autism

2.1 Abstract

The prevalence of sensory processing deficits in children with Autism Spectrum Disorders (ASD) ranges from 69 to 95%. Sensory processing (SP) deficits are thought to contribute to the clinical presentation and functional difficulties experienced by individuals with ASD. Detection of SP deficits during the second year of life prior to a diagnosis of ASD could significantly benefit the developmental course of the disorder. Prospective studies examining SP dysfunction in school age and preschool children with ASD are common; however, infant and toddler SP research is often limited to retrospective reviews or parent report.

Early identification of SP deficits leads to early intervention (EI) for individuals who may go on to develop ASD. EI has the potential to significantly improve SP deficits and minimize ASD characteristics. In order to move toward addressing SP in an EI program, SP function during the second year of life must be better understood. The goals of this project were to: (1) describe SP function in a group of toddlers aged 12-24 months (2) examine differences in SP between toddlers with and without early signs of
ASD and (3) examine the relative contribution of SP patterns, language abilities, motor skills, and cognition to the prediction of early signs of autism in young children 12-24 months of age.

2.2 Background and significance

Autism Spectrum Disorder (ASD) has progressed from a rare diagnosis to a common disorder, affecting approximately 1 in 91 children (Kogan et al., 2009). The exponential rise in ASD prevalence makes it more common than childhood cancer, juvenile diabetes, spina bifida, and Down Syndrome combined (Filipek et al., 1999). According to the U.S. Department of Education, autism is growing at an exorbitant rate of 10-17% per year (NIDRR Long Range Plan, 2009). The recent increase in prevalence and the severity of the impact of the disorder makes a priority for researcher.

Although there is no consensus in the literature as to the initial age of onset of ASD, early signs of ASD are often detected by the second year of life (Barbaro & Dissanayake, 2009; Chawarska, Klin, Paul, & Volkmar, 2007; Landa, Holman, & Garrett-Mayer, 2007; Mitchell et al., 2006; Ozonoff et al., 2010). Early signs of autism are most often evident in the areas of socialization, motor, language, communication, and sensory processing. To date, the majority of the research addressing early signs of autism has been retrospective home video tape analysis and parent report although more researchers are using prospective designs with at-risk groups. Home video studies have identified behavioral and sensorimotor characteristics evident in young children that were later diagnosed with ASD. These characteristics include variability in social
engagement, infrequent joint attention attempts, limited imitation skills, poor eye contact or visual avoidance, excessive mouthing of objects, social touch aversion, absence of response to familiar voices, and infrequent orienting to their name (Cox et al., 1999; Kasari & Wong, 2002; Mars et al., 1998; Moore & Goodson, 2003; Osterling & Dawson, 1994; Werner et al., 2000). Additionally, studies utilizing parent reports suggest that young children with ASD are under-responsive to certain sounds, under-responsive to pain, and are sensitive to certain tastes and textures more often than typically developing children (Tomchek & Dunn, 2007).

The prevalence of SP deficits in children with ASD ranges from 69% to 95% (Baranek, 2002; Baranek, Boyd, Poe, David, & Watson, 2007; Dawson & Watling, 2000; Harrison & Hare, 2004; Kientz & Dunn, 1997; Tomchek & Dunn, 2007). The majority of SP deficits in individuals with ASD manifest as auditory under-responsiveness and are thought to contribute to the functional limitations experienced by these children (Baranek, 1999; Rogers & Ozonoff, 2005). Behaviors related to SP deficits can be detected prior to the diagnosis of ASD (Baranek, 1999). Several retrospective studies report that infants later diagnosed with ASD can be distinguished from neurotypical and developmentally delayed infants in sensorimotor and social development (Adrien et al., 1992; Baranek, 1999). These authors suggest that certain characteristics of sensorimotor functioning could be used to enhance measures of social responsiveness for detection of ASD during infancy (Baranek, 1999). Further, early identification and treatment of SP deficits in young children with early signs of autism may reduce the
negative effects on a child’s opportunities to learn from social experiences thus promoting more typical developmental patterns and preventing secondary symptoms of autism (Zwaigenbaum, 2005).

Toddlers with typical SP abilities are able to take in sensory information from the environment, process it and make an adaptive response. Typically developing toddlers regulate their behavioral responses to sensory input whereas toddlers with SP deficits tend to exhibit under- or over-responsive behavioral patterns interfering with their sensorimotor and emotional development (DeGangi, Porges, Sickel, & Greenspan, 1993; Greenspan & Wieder, 1997; Greenspan & Wieder, 1993). Throughout infancy and toddlerhood, the nervous system continues to mature in typically developing children, therefore improving regulation of behavioral responses and sensory processing abilities (Covington, Cronenwett, & Loveland-Cherry, 1991; DeGangi et al., 1993).

Deficits in SP have been linked to the core symptoms of ASD including repetitive behaviors, communication, and socialization deficits (Ashburner et al., 2008; Just et al., 2004). Abnormal SP functioning has been highly correlated with the presence of stereotypic, rigid, and repetitive behaviors (Boyd et al., 2009; Watt et al., 2008). Additionally, impairments in the auditory system could negatively affect social development and compound language and communication impairments. Measures of SP deficits could eventually be used to screen individuals for early signs of ASD, influence individualized intervention programs, and determine the most effective
intervention strategies and at what age treatment should begin (Baranek, 1999).

Further, if started at a young age, early intervention has the potential to minimize the core symptoms of ASD and decrease the severity of the disorder (Rogers & Vismara, 2008).

Similar to the identification of early signs of autism, SP deficits in young children are reliant on imprecise, proxy-report measures. Specifically, retrospective reviews based on behavioral data using parent report and videotaped analysis are the most commonly utilized. Retrospective videotaped analysis suggests that young children later diagnosed with ASD exhibited auditory under- and over-responsiveness, delayed response to name, excessive mouthing of objects and unstable visual orientation/attention more often than typically developing children or children with developmental delays (Adrien et al., 1992; Baranek, 1999).

This study will address a gap in literature specific to SP function during the second year of life in children with and without early signs of autism. The aims of this study are to: (1) describe SP function during the second year of life (2) examine differences in SP between toddlers with and without early signs of autism, and (3) describe the relationship between early signs of autism, SP function, and developmental status.
2.3 Research Design and Methods

2.3.1 Participants and Recruitment

Forty-six toddlers (Mean age = 17.9 months, SD= 3.0; 28 males) participated in this study. Toddlers with and without risk factors for autism were recruited to the study from the following facilities and events: The Ohio State Child Care Center, Medical Center and Nisonger Center, local businesses/offices (pediatricians, therapy clinics, schools, etc), Nationwide Children’s Hospital, Research Match, Autism Speaks Walk, and Autism Society of America: Central Ohio Chapter (website and newsletter) (see appendix A & B for recruitment materials).

Inclusion criteria for the study were:

- Aged between 12-24 months
- Intact hearing (assessed by parent report of newborn hearing screening)

In order to maximize the number of participants showing early signs of autism, toddlers with high risk for autism including siblings, history of premature birth and/or developmental delay, were targeted in recruitment efforts. Toddlers were not eligible for study participation if they were diagnosed with a developmental disorder other than autism prior to entering this study (including cerebral palsy, seizure disorders, brain injury e.g. periventricular leucomalacia, intraventricular hemorrhage, Down syndrome).
Our final sample consisted of:

- 2 siblings of children with ASD, one of which was a half sibling
- 4 toddlers born prematurely (prior to 37 weeks gestation)
- 40 toddlers born full term

2.3.2 Study procedures

Parents interested in participating contacted the SenSA Lab at The Ohio State University. A telephone screening ensured inclusion criteria were met prior to scheduling an appointment (Appendix C). Parents were either mailed or emailed directions to the lab and consent form for review (Appendix D-F). Additionally, a reminder email/phone call was given the day prior to the appointment. Parental consent was obtained at the study visit along with a verbal explanation of the procedures and any questions were answered. All data were collected by the same researcher in a small treatment room with caregiver(s) present. Appointments were scheduled around the toddler’s meal times and nap schedules. Assessments were administered in the same order for all subjects. This study was approved by the Institutional Review Boards at The Ohio State University and Nationwide Children’s Hospital.

2.3.3 Instrumentation

Sensory Processing Assessment for Young Children (SPA): (Baranek, 1999) The SPA is a semi-structured assessment used to examine SP patterns in young children, specifically
those with ASD and other related disorders. It is designed to allow direct observation of an individual’s responses to tactile, auditory, and visual stimuli through play with novel sensory toys and unexpected sensory stimuli. Individual responsiveness to toys is rated on a 3-point scale according to level of engagement with the activity. A lower score indicates more typical functioning. Behavioral habituation to a repeated auditory stimulus is assessed through observation of orienting behavior over 10 trials. This assessment is designed for infants 9-62 months of age.

**Autism Detection in Early Childhood (ADEC):** (Young, 2007) Risk factor status was classified through administration of the ADEC which detects early signs of ASD in pre-verbal infants. The ADEC consists of 16 items and an adaptation period measured through direct observation. The adaptation period is used when the toddler needs a break and/or becomes fussy. The items presented in the ADEC fit into one of three domains: (1) disturbances in interacting with others and with objects (e.g. poor imitation, lack of orienting to name, rare use of gestures, etc.) (2) stereotyped, repetitive movements (arrange objects in a line, hand flapping, finger flicking, etc.) (3) bizarre responses to environmental stimuli (covering ears in response to sound, excessive mouthing of objects, and smelling objects or people). Children score between 0-2 on each item. A score of zero indicates an appropriate response within the first or second item administration. A task performed in a modified way, with a delayed response, or performed spontaneously during testing but not when originally administered is scored a 1. A score of 2 is given for either an atypical sensory reaction
or if a task was not attempted. A higher score indicates increased presence of early signs of autism.

**Bayley Scales of Infant and Toddler Development – Third Edition (BSID-III):** (Bayley, 2006) This observational scale is considered the gold standard for evaluation of general development in young children aged 1-42 months. The BSID-III consists of test items assessing fine and gross-motor, cognition, and receptive and expressive language skills. Scores are compared with a large normative dataset. All subscales of the BSID-III were administered.

**Infant/Toddler Sensory Profile (ITSP):** (Dunn, 2002) The ITSP is a parent report measure of SP describing behavioral patterns related to toddler’s sensory reactivity in their typical environments. Parents rate 48 items using a 5 point Likert scale, ranging from “almost never” to “almost always”. Total scores are obtained for each quadrant category (sensory sensitivity, sensory seeking, low registration, and sensation avoiding) and each sensory modality (auditory, visual, vestibular, touch, oral sensory processing). The total scores for each category fall into a descriptive category: typical performance, probable difference (at least 1 standard deviation (SD) above or below the normative mean) and definite difference (at least 2 SDs above or below the normative mean). SP behaviors are scored along a continuum of under-responsive behavioral patterns, typical behavioral responses and over-responsive behavioral patterns.
2.4 Analyses

All statistical analyses were completed using SPSS, version 19 (Chicago: SPSS, 2011).

Aim 1: Descriptive statistics were calculated for the entire sample on all sensory processing, developmental, and early signs of autism assessments. A TwoStep cluster analysis was completed to identify SP patterns occurring within our sample. Two clusters were identified and independent t-tests were used determining if there were statistically significant differences on all sensory and developmental measures between the clusters (except for SPA). Chi-Square analysis was used to test for significance on the SPA based on typical or atypical SP function. On the SPA, SP function was identified as typical if the score was a 1. Scores of 2 or more were considered atypical.

Aim 2: Participants were classified into two groups: no early signs of autism group (N-ES) or early signs of autism group (ES) based on ADEC total score. Descriptive statistics were calculated and group comparisons were made using independent t-tests for (1) the demographic information by group and (2) SP function by group for the ITSP: auditory, visual, vestibular, tactile, and oral processing. A Chi-Square analysis was used to test for significance on the SPA based on typical or atypical SP function on the following subscales: avoidance score, mouthing, and sensory seeking.

Aim 3: Curve estimations were completed for each independent variable (IV) with the dependent variable (DV) to determine if the relationship between the two variables was
linear. The dependent variable (DV) for the curve estimations was the ADEC total score. Independent variables included: BSID-III standard scores (cognition, receptive language, expressive language, fine motor and gross motor), ITSP modality scores (auditory, visual, vestibular, tactile, and oral processing), and the SPA Avoidance category score. Next, a stepwise, linear regression analysis was completed evaluating the relationship between SP function and developmental skills with the presence of early signs of autism. The dependent variable (DV) for the stepwise regression analysis was the ADEC total score. Independent variables included: BSID-III composite scores (cognition, language, and motor), ITSP sensory processing category (typical and atypical), SPA sensory processing category (typical and atypical) and chronological age. On the ITSP, typical SP was defined as having all sensory modality scores in the typical range or one sensory modality score in the probable difference category. Atypical SP was defined as two or more sensory modality scores in the probable difference category or one or more score in the definite difference category. On the SPA, typical SP was defined as scoring a 1 on the avoidance rating. Atypical SP was defined as scoring 2 or above on the avoidance rating.
2.5 Results

Aim 1a: Descriptive statistics for sample as a whole

Means and SDs for the SPA, ITSP and BSID-III for the sample as a whole are presented in Table 2.1. Results indicate mean SP performance for our sample as a whole fell within the typical range. Specifically, the mean scores for all ITSP quadrant scores and all sensory modality scores fell within the ITSP typical performance category. The mean scores on the SPA for mouthing of objects and sensory sensitivities were very close to zero indicating that these behaviors were rarely observed. Therefore, these subscale scores were not included in any further analysis. The mean score on the SPA avoidance rating is close to 1 indicating that as a whole, the avoidance behaviors were within the typical functioning category. The mean scores for the developmental skills (all subscales on the BSID-III) of the toddlers in our sample were all within normal limits. The mean ADEC score was in the low risk category. Overall, when looking at group statistics, our sample demographics represent typical performance in developmental and sensory processing abilities and fell in the low risk category for early signs of autism.
Table 2.1: Descriptive statistics of entire sample (N=46) by assessment

<table>
<thead>
<tr>
<th>Assessment</th>
<th>Mean (SD)</th>
<th>Descriptive Category</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>ITSP: Quadrant Category</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Low Registration</td>
<td>49.4 (3.8)</td>
<td>Typical Performance</td>
</tr>
<tr>
<td>Sensory Seeking</td>
<td>29.9 (8.2)</td>
<td>Typical Performance</td>
</tr>
<tr>
<td>Sensory Sensitivity</td>
<td>46.9 (5.2)</td>
<td>Typical Performance</td>
</tr>
<tr>
<td>Sensory Avoiding</td>
<td>51.4 (4.4)</td>
<td>Typical Performance</td>
</tr>
<tr>
<td><strong>ITSP: Sensory Modality</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Auditory</td>
<td>38.4 (4.4)</td>
<td>Typical Performance</td>
</tr>
<tr>
<td>Visual</td>
<td>23.4 (3.5)</td>
<td>Typical Performance</td>
</tr>
<tr>
<td>Tactile</td>
<td>55.7 (5.8)</td>
<td>Typical Performance</td>
</tr>
<tr>
<td>Vestibular</td>
<td>19.2 (2.7)</td>
<td>Typical Performance</td>
</tr>
<tr>
<td>Oral Processing</td>
<td>27.7 (3.6)</td>
<td>Typical Performance</td>
</tr>
<tr>
<td><strong>SPA</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Avoidance Rating</td>
<td>1.17 (1.4)</td>
<td>Typical Performance</td>
</tr>
<tr>
<td>Sensory Seeking</td>
<td>0.04 (0.3)</td>
<td>Typical Performance</td>
</tr>
<tr>
<td>Mouthing</td>
<td>0.28 (0.6)</td>
<td>Typical Performance</td>
</tr>
<tr>
<td><strong>BSID-III (Scaled Scores)</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Cognitive</td>
<td>12.02 (2.4)</td>
<td>Within normal limits</td>
</tr>
<tr>
<td>Receptive Language</td>
<td>11.02 (3.2)</td>
<td>Within normal limits</td>
</tr>
<tr>
<td>Expressive Language</td>
<td>10.13 (3.0)</td>
<td>Within normal limits</td>
</tr>
<tr>
<td>Fine Motor</td>
<td>11.87 (2.4)</td>
<td>Within normal limits</td>
</tr>
<tr>
<td>Gross Motor</td>
<td>10.2 (2.7)</td>
<td>Within normal limits</td>
</tr>
<tr>
<td><strong>ADEC</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Total Score</td>
<td>5.85 (3.3)</td>
<td>Low Risk</td>
</tr>
</tbody>
</table>

*See Appendix G for cut off scores for clinical categories for scoring
A finer analysis of SP function was completed using the ITSP. For the first analysis, probable difference and definite difference categories for the sensory modality and quadrant scores were combined and labeled “atypical SP”. Graphical displays of the subject distribution within each quadrant category and sensory modality are presented in Figures 2.1 and 2.2. Specifically, sensory seeking was the most prominent atypical sensory processing behavior observed in this sample, with 30% of the sample exhibiting these behaviors. The sensory processing modality scores indicated that toddlers had the most difficulty processing sensory information in the auditory and vestibular domains. Thirty-three percent of this sample scored as having atypical SP in the auditory domain (44% under-responsive and 56% over-responsive) and 30% scored as atypical SP in the vestibular domain (14% under-responsive and 86% over-responsive).
Figure 2.1: Distribution of subjects scoring in each quadrant category

Figure 2.2: Distribution of subjects scoring in each sensory modality category
Additional analysis examined the sample distribution of scores across the functional categories (typical, probable, and definite difference) for the quadrant categories and the sensory modalities (Figure 2.3). Specifically, the distribution across quadrant categories shows that 20 subjects (43%) had all scores in the typical processing category, 19 subjects (41%) had at least one score in the probable difference category and 7 subjects (16%) had at least one score in the definite difference category. Considering sensory modalities, 18 subjects (39%) had all scores in the typical processing category, 25 (53%) had at least one score in the probable difference category and 4 (8%) had at least one score in the definite difference category. Figure 2.4 shows the number of typical and atypical (probable or definite difference) scores for each subject across the quadrant categories and sensory modalities. Quadrant score distribution was as follows: 20 subjects (43%) had all scores in the typical range, 16 subjects (35%) had one score in an atypical category and 10 subjects (22%) had more than one score in the atypical range. Sensory modality score distribution was as follows: 18 subjects (40%) had all scores in the typical range, 15 subjects (32%) had one score in the atypical range, and 13 subjects (28%) had more than one score in the atypical range. This analysis provides an additional way to examine the distribution of sensory processing function scores among our sample of toddlers.
Figure 2.3: Distribution of sensory processing scores by functional category on the ITSP. Subjects in typical category had all scores in typical range. Subjects in probable and definite difference categories had at least one score in this category.
Aim 1b: Patterns of sensory processing in toddlers

A TwoStep cluster analysis was completed to identify patterns of SP function within our sample. The following variables were entered into the cluster analysis: ITSP sensory modality scores and SPA avoidance rating. The ITSP quadrant categories and sensory modality scores are derived from the same set of questions. In order to prevent using the same information twice, only the sensory modality scores were entered into the cluster analysis.

Two sensory processing clusters emerged. There was no difference in chronological age between the clusters (p=0.895) (Descriptive statistics and p-values by cluster are presented in Table 2.2). The mean sensory modality scores on the ITSP for both clusters fell within the typical range. However, further analysis of the sensory
modality mean scores revealed Cluster 1 scores trended toward the under-responsive end of the continuum on all sensory modalities. Mean scores for Cluster 2 were closer to the over-responsive end of the continuum on all sensory modalities (Table 2.2 and Figure 2.5). Independent t-tests were conducted to test for differences in SP functioning between cluster groups for the ITSP. Significant differences were found between clusters for all sensory modalities (p=0.001) on the ITSP except for oral processing (p=0.413). A Chi Square analysis was conducted to test for differences on the SPA subscales. The SPA avoidance rating was different between clusters (p=0.001).

Additional analysis was completed to look at sensory processing function in more detail on the ITSP. Results indicate that cluster 2 exhibits more atypical sensory processing scores than cluster 1 (Figure 2.6). Specifically, on the ITSP, 9 subjects (35%) had all typical scores, 7 subjects (27%) had 1 score in the atypical range, and 10 subjects (38%) had more than 1 score in the atypical range. On the SPA, 10 subjects (38%) scored in the typical range whereas 16 subjects (62%) scored in the atypical range. On the ITSP, the scores for cluster 1 are as follows: 9 subjects (45%) all had scores in the typical range, 8 subjects (40%) had 1 score in the atypical range, and 3 subjects (15%) had more than one score in the atypical range. On the SPA, 95% (19 subjects) in cluster 1 scored in the typical range and 1 subject (5%) scored in the atypical range.
Table 2.2: Descriptive statistics by cluster

<table>
<thead>
<tr>
<th>Assessment</th>
<th>Cluster 1: Mean (SD) n=20</th>
<th>Cluster 2: Mean (SD) n=26</th>
<th>p value</th>
</tr>
</thead>
<tbody>
<tr>
<td>ITSP</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Auditory</td>
<td>41.5 (2.7)</td>
<td>36.1 (4)</td>
<td>0.001**</td>
</tr>
<tr>
<td>Visual</td>
<td>26.1 (3.2)</td>
<td>21.3 (2.1)</td>
<td>0.001**</td>
</tr>
<tr>
<td>Tactile</td>
<td>60.1 (3.3)</td>
<td>52.3 (5)</td>
<td>0.001**</td>
</tr>
<tr>
<td>Vestibular</td>
<td>20.8 (2.6)</td>
<td>18.04 (2.2)</td>
<td>0.001**</td>
</tr>
<tr>
<td>Oral Processing</td>
<td>28.15 (3)</td>
<td>27.27 (4)</td>
<td>0.413</td>
</tr>
<tr>
<td>SPA</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Avoidance Rating</td>
<td>.25 (0.55)</td>
<td>1.88 (1.4)</td>
<td>0.001**</td>
</tr>
</tbody>
</table>

**statistically significant differences
Figure 2.6: Distribution of subjects according to number of scores they had in the typical/atypical functioning categories.
Variables in the cluster analysis were ranked on level of importance in predicting cluster membership. The scale ranges from 0-1 with 1 indicating most important and 0 indicating least important. Results indicate that the ITSP visual (1.0), ITSP tactile (0.98), ITSP auditory (0.79), and SPA avoidance rating (0.75) were the best predictors of cluster membership.

**Aim 2: Sensory processing in toddlers with and without early signs of autism**

Participants were grouped based on total ADEC score: those with a score between 0-5 were classified as N-ES (n=24) and those achieving a score of 6 and above were classified as ES (n=22) (see Table 2.3 for demographic information by group). The cut-off score for the ES group used for this study deviated from the cut-off scores
indicated in the assessment manual. The manual indicates individuals scoring between 0-10 are at low risk for ASD, 11-13 are at moderate risk, 14-19 are at high risk and >19 are at very high risk. In this study, clinical differences were apparent for toddlers scoring 6 or more on the ADEC. These clinical differences were discussed with the author of the assessment and permission was given to use a score of 6 or above as the cut-off score for the presence of early signs of ASD in this study.

Table 2.3: Descriptive statistics and p values for demographic information for N-ES vs ES groups

<table>
<thead>
<tr>
<th>Demographic Information</th>
<th>N-ES: Mean (SD) n=24</th>
<th>ES: Mean (SD) n=22</th>
<th>p value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Male/Female</td>
<td>12/12</td>
<td>16/6</td>
<td></td>
</tr>
<tr>
<td>Chronological Age</td>
<td>19.21 (3.5)</td>
<td>16.5 (3.8)</td>
<td>0.018**</td>
</tr>
<tr>
<td>ADEC</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Total Score</td>
<td>3.4 (1.4)</td>
<td>8.5 (2.5)</td>
<td>0.01**</td>
</tr>
<tr>
<td>BSID-III (Scaled Scores)</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Cognitive</td>
<td>13.1 (2.2)</td>
<td>10.9 (2.2)</td>
<td>0.001**</td>
</tr>
<tr>
<td>Receptive Language</td>
<td>12.7 (2.9)</td>
<td>9.1 (2.4)</td>
<td>0.001**</td>
</tr>
<tr>
<td>Expressive Language</td>
<td>11.5 (2.6)</td>
<td>8.7 (2.8)</td>
<td>0.001**</td>
</tr>
<tr>
<td>Fine Motor</td>
<td>12.8 (2.4)</td>
<td>10.9 (1.9)</td>
<td>0.005**</td>
</tr>
<tr>
<td>Gross Motor</td>
<td>11.3 (2.5)</td>
<td>9.0 (2.4)</td>
<td>0.003**</td>
</tr>
</tbody>
</table>

**statistically significant differences
Demographic information indicated that the ES group was significantly younger (p=0.018) than the N-ES group. An attempt was made to control for this age difference in analysis. The BSID-III is normalized for age in scoring therefore no adjustment was necessary. Curve estimations were completed to determine if there was a relationship between age and all other IV’s (SPA avoidance rating and ITSP modality scores).

Findings indicate that there was no relationship between these factors and age (see table 2.4)

**Table 2.4: Curve estimations for SPA and ITSP with Age**

<table>
<thead>
<tr>
<th></th>
<th>R²</th>
<th>p</th>
</tr>
</thead>
<tbody>
<tr>
<td>SPA Avoidance</td>
<td>0.003</td>
<td>0.725</td>
</tr>
<tr>
<td>ITSP Vestibular</td>
<td>0.067</td>
<td>0.083</td>
</tr>
<tr>
<td>ITSP Visual</td>
<td>0.007</td>
<td>0.593</td>
</tr>
<tr>
<td>ITSP Tactile</td>
<td>0.015</td>
<td>0.422</td>
</tr>
<tr>
<td>ITSP Auditory</td>
<td>0.004</td>
<td>0.685</td>
</tr>
<tr>
<td>ITSP Oral Processing</td>
<td>0.020</td>
<td>0.350</td>
</tr>
</tbody>
</table>

Demographic information also indicated that all BSID-III subscale scores were significantly different between groups (p<0.05) with the ES group scoring lower than the N-ES group on all scales.

Independent t-tests were conducted to determine if there were differences between the N-ES and ES groups on the ITSP sensory modality scores and Chi square was conducted on the SPA avoidance scale (Table 2.5). The ES group scored significantly lower on the ITSP vestibular processing modality (p=0.041) (90% over-responsive and
10% under-responsive). Additionally, trends were identified on the ITSP oral, tactile and visual SP modalities with the ES group scoring lower that the N-ES group on all three modalities and a trend on the SPA avoidance rating with the ES group scoring higher than the N-ES group.

Table 2.5: Descriptive statistics and p values for sensory processing for N-ES vs ES groups

<table>
<thead>
<tr>
<th>Assessment</th>
<th>N-ES: Mean (SD) n=24</th>
<th>ES: Mean (SD) n=22</th>
<th>p value</th>
</tr>
</thead>
<tbody>
<tr>
<td>ITSP Auditory</td>
<td>39.3 (5.1)</td>
<td>37.5 (3.2)</td>
<td>0.167</td>
</tr>
<tr>
<td>Visual</td>
<td>24.3 (3.8)</td>
<td>22.4 (2.9)</td>
<td>0.076*</td>
</tr>
<tr>
<td>Tactile</td>
<td>57.2 (5.8)</td>
<td>54.1 (5.5)</td>
<td>0.073*</td>
</tr>
<tr>
<td>Vestibular</td>
<td>20.0 (2.5)</td>
<td>18.4 (2.8)</td>
<td>0.041**</td>
</tr>
<tr>
<td>Oral Processing</td>
<td>28.6 (2.8)</td>
<td>26.6 (4.1)</td>
<td>0.064*</td>
</tr>
<tr>
<td>SPA Avoidance Rating</td>
<td>0.71 (1.04)</td>
<td>1.68 (1.52)</td>
<td>0.073*</td>
</tr>
</tbody>
</table>

**statistically significant differences  
*trend with differences

Additional analysis of ITSP by group was completed to examine the distribution of typical and atypical SP function across sensory modalities by group (Figure 2.7). Results indicate that the N-ES group had more atypical auditory SP function (n=10 with atypical SP; 6 were under-responsive, 4 were over-responsive) than the ES group (n=5 with atypical SP, 1 was under-responsive, 4 were over-responsive). Additionally, the ES group exhibited more atypical vestibular sensory processing function (n=10 with atypical
SP; 1 was under-responsive, 9 were over-responsive) than the N-ES group (n=4 with atypical SP; 1 was under-responsive, 3 were over-responsive). Visual, tactile, and oral sensory processing between the two groups was relatively similar. The groups were very similar when comparing SP function relative to the number of atypical and typical sensory responses on sensory modalities by group (Figure 2.8).

Figure 2.8: Distribution of subjects scoring in each sensory modality category
Aim 3: Prediction of early signs of autism in toddlers

The final aim was to examine the relative contribution of SP patterns, language abilities, motor skills, and cognition to the prediction of early signs of autism in young children under two years of age. First, curve estimations were completed using DV (ADEC) with each IV to check for linearity. R square values (linear, logarithmic, and exponential) for each independent variable are listed in Table 2.6. For each IV, R square values are similar among linear, logarithmic, and exponential so linear relationship was assumed. Additionally, figures 2.9-2.19 show the relationship between DV and IV.
<table>
<thead>
<tr>
<th>Assessment</th>
<th>R square-Linear</th>
<th>R square-Logarithmic</th>
<th>R square-Exponential</th>
</tr>
</thead>
<tbody>
<tr>
<td>BSID-III Cognitive</td>
<td>.266</td>
<td>.280</td>
<td>.186</td>
</tr>
<tr>
<td>BSID-III Receptive Lang.</td>
<td>.334</td>
<td>.333</td>
<td>.280</td>
</tr>
<tr>
<td>BSID-III Expressive Lang.</td>
<td>.308</td>
<td>.323</td>
<td>.270</td>
</tr>
<tr>
<td>BSID-III Fine Motor</td>
<td>.237</td>
<td>.262</td>
<td>.219</td>
</tr>
<tr>
<td>BSID-III Gross Motor</td>
<td>.158</td>
<td>.173</td>
<td>.114</td>
</tr>
<tr>
<td>SPA Avoidance Rating</td>
<td>.153</td>
<td></td>
<td>.159</td>
</tr>
<tr>
<td>ITSP Auditory</td>
<td>.034</td>
<td>.027</td>
<td>.038</td>
</tr>
<tr>
<td>ITSP Visual</td>
<td>.047</td>
<td>.047</td>
<td>.040</td>
</tr>
<tr>
<td>ITSP Tactile</td>
<td>.047</td>
<td>.044</td>
<td>.047</td>
</tr>
<tr>
<td>ITSP Vestibular</td>
<td>.052</td>
<td>.057</td>
<td>.049</td>
</tr>
<tr>
<td>ITSP Oral Processing</td>
<td>.129</td>
<td>.120</td>
<td>.087</td>
</tr>
</tbody>
</table>
Figure 2.10: Curve Estimation plot for DV (ADEC) and IV (BSID-III Cognition)
Figure 2.11: Curve Estimation plot for DV (ADEC) and IV (BSID-III Receptive Language)
Figure 2.12: Curve Estimation plot for DV (ADEC) and IV (BSID-III Expressive Language)
Figure 2.13: Curve Estimation plot for DV (ADEC) and IV (BSID-III Fine Motor)
Figure 2.14: Curve Estimation plot for DV (ADEC) and IV (BSID-III Gross Motor)
Figure 2.15: Curve Estimation plot for DV (ADEC) and IV (SPA Avoidance)
Figure 2.16: Curve Estimation plot for DV (ADEC) and IV (ITSP Auditory)
Figure 2.17: Curve Estimation plot for DV (ADEC) and IV (ITSP Tactile)
Figure 2.18: Curve Estimation plot for DV (ADEC) and IV (ITSP Visual)
Figure 2.19: Curve Estimation plot for DV (ADEC) and IV (ITSP Vestibular)
Next, a regression analysis was used to examine the relative contribution of SP patterns, language abilities, motor skills, and cognition to the prediction of early signs of autism in young children under two years of age. The independent variables included in the regression analysis were: BSID-III composite scores (cognition, language, and motor), SPA SP category (typical and atypical), ITSP SP category (typical and atypical), and chronological age. (Table 2.8 reports distribution of subjects in typical and atypical categories on SPA and ITSP). Subjects were categorized as having either typical or atypical sensory processing abilities on the ITSP and SPA to minimize multicollinearity.
On the ITSP, typical SP was defined as having all sensory modality scores in the typical range or one sensory modality score in the probable difference category. Atypical SP was defined as two or more sensory modality scores in the probable difference category or one or more score in the definite difference category. On the SPA, typical SP was defined as scoring a 1 on the avoidance rating. Atypical SP was defined as scoring 2 or above on the avoidance rating.

Pearson’s correlation between the dependent and independent variables is reported in Table 2.7. A stepwise linear regression analysis revealed two possible models of prediction (Table 2.9). The first model included the language composite only (adjusted $R^2 = 0.344$) and the second model included language composite and chronological age (adjusted $R^2 = 0.447$). The second model was a stronger model because it had a larger $R^2$ value and included age which is clinically relevant to early autism detection.
Table 2.7: Correlation analysis for SP function, developmental functioning and early signs of autism

<table>
<thead>
<tr>
<th>SP Measure</th>
<th>ADEC</th>
<th>Cognition</th>
<th>Language</th>
<th>Motor</th>
<th>SPA</th>
<th>ITSP</th>
<th>Age</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td><em>r</em></td>
<td><em>p</em></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>ADEC</td>
<td>1</td>
<td>-0.468</td>
<td>-0.599</td>
<td>-0.432</td>
<td>0.362</td>
<td>0.121</td>
<td>-0.312</td>
</tr>
<tr>
<td></td>
<td>0.001*</td>
<td>0.001*</td>
<td>0.002*</td>
<td>0.007*</td>
<td></td>
<td>0.215</td>
<td></td>
</tr>
<tr>
<td>BSID-III</td>
<td>-0.468</td>
<td>1</td>
<td>0.632</td>
<td>0.620</td>
<td>-0.405</td>
<td>-0.158</td>
<td>0.049</td>
</tr>
<tr>
<td>Cognitive</td>
<td>0.001*</td>
<td>0.001*</td>
<td>0.001*</td>
<td>0.003*</td>
<td></td>
<td>0.151</td>
<td></td>
</tr>
<tr>
<td>BSID-III</td>
<td>-0.599</td>
<td>0.632</td>
<td>1</td>
<td>0.536</td>
<td>-0.425</td>
<td>-0.273</td>
<td>-0.039</td>
</tr>
<tr>
<td>Language</td>
<td>0.001*</td>
<td>0.001*</td>
<td>0.001*</td>
<td>0.002*</td>
<td>0.035*</td>
<td></td>
<td></td>
</tr>
<tr>
<td>BSID-III</td>
<td>-0.432</td>
<td>0.620</td>
<td>0.536</td>
<td>1</td>
<td>-0.390</td>
<td>-0.201</td>
<td>0.163</td>
</tr>
<tr>
<td>Motor</td>
<td>0.002*</td>
<td>0.001*</td>
<td>0.001**</td>
<td>0.004*</td>
<td></td>
<td>0.093</td>
<td></td>
</tr>
<tr>
<td>SPA</td>
<td>0.362</td>
<td>-0.405</td>
<td>-0.425</td>
<td>-0.390</td>
<td>1</td>
<td>0.009</td>
<td>0.163</td>
</tr>
<tr>
<td></td>
<td>0.007*</td>
<td>0.003*</td>
<td>0.002*</td>
<td>0.004*</td>
<td></td>
<td>0.477</td>
<td></td>
</tr>
<tr>
<td>ITSP</td>
<td>0.121</td>
<td>-0.158</td>
<td>-0.275</td>
<td>-0.201</td>
<td>0.009</td>
<td></td>
<td>0.001</td>
</tr>
<tr>
<td></td>
<td>0.215</td>
<td>0.151</td>
<td>0.035*</td>
<td>0.093</td>
<td></td>
<td>0.477</td>
<td></td>
</tr>
<tr>
<td>Age in</td>
<td>-0.312</td>
<td>0.049</td>
<td>-0.039</td>
<td>0.163</td>
<td>0.163</td>
<td>0.001</td>
<td>1</td>
</tr>
<tr>
<td>Months</td>
<td>0.018*</td>
<td>0.375</td>
<td>0.400</td>
<td>0.143</td>
<td>0.142</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

*statistically significant differences

Table 2.8: Distribution of subjects in typical and atypical categories of SP based on SPA and ITSP

<table>
<thead>
<tr>
<th>SP Measure</th>
<th>Typical SP</th>
<th>Atypical SP</th>
</tr>
</thead>
<tbody>
<tr>
<td>SPA</td>
<td>28</td>
<td>17</td>
</tr>
<tr>
<td>ITSP</td>
<td>33</td>
<td>13</td>
</tr>
</tbody>
</table>
Table 2.9: Regression analysis; DV=ADEC score

<table>
<thead>
<tr>
<th></th>
<th>Coefficients</th>
<th>Standard Error</th>
<th>Significance</th>
</tr>
</thead>
<tbody>
<tr>
<td>Constant</td>
<td>22.323</td>
<td>2.771</td>
<td>p=0.001**</td>
</tr>
<tr>
<td>Language Composite</td>
<td>-0.113</td>
<td>0.021</td>
<td>p=0.001**</td>
</tr>
<tr>
<td>Age in Months</td>
<td>-0.277</td>
<td>0.092</td>
<td>p=0.005**</td>
</tr>
</tbody>
</table>

**statistically significant differences**

2.6 Discussion

The results of this study are consistent with the findings of previous literature indicating that differences in sensory processing can be detected during the second year of life in children with early signs of ASD. Our study is unique in that it examines SP function in toddlers aged 12-24 months using a direct observation measure in conjunction with parent report. As noted below, the direct observation measure and parent report identified similar patterns of sensory processing in cluster groupings and those with and without early signs of autism.

Sensory processing of the sample as a whole

While the group mean SP scores fell within the typical range of functioning on all ITSP quadrant scores and sensory modality scores and all subscales on the SPA. Finer analysis of individual differences revealed significant patterns of SP difference in our sample. Specifically, examining the distribution of scores within functional categories (typical, probable difference, and definite difference) showed the variability of sensory
processing function within the sample. Most toddlers in our sample exhibited a probable difference in at least one sensory modality (53%). Only 39% of toddlers scored within the typical range on all sensory modalities. Further, only 8% of the sample had at least one score in the definite difference range. Considering quadrant scores, the number of subjects scoring all in the typical range (43%) was similar to those scoring at least one score in the probable difference range (41%). These results suggest that there is subtle variability in sensory processing function in toddlers during the second year of life.

As our results show, it is fairly common with toddlers to exhibit variability in sensory processing function during the second year of life. Sensory processing function was studied in a group of typically developing 4-18 month olds finding similar levels of variability in sensory processing development. Specifically, children in this study exhibited under- and over-responsiveness patterns to tactile, vestibular and visual stimuli (Degangi, 1989). Additionally, these infants exhibited variability in their visual-tactile integration, ocular-motor control, and adaptive motor responses. The majority of the toddlers in our sample scored in the atypical range in one sensory modality and in the typical range for the rest of the sensory modalities. Our sample was recruited from a population of toddlers with increased risk factors for ASD. Specifically toddlers with (1) siblings diagnosed with ASD (2) history of premature birth and (3) developmental delay were specifically targeted to participate in this study. Our final sample included:
• 2 siblings of children with ASD, one of which was a half sibling
• 4 toddlers born prematurely (prior to 37 weeks gestation)
• 40 toddlers born full term

Moreover, we examined how many toddlers in our sample exhibited a delay in any developmental domain (on the BSID-III) and early signs of autism (ADEC) or a combination of the two. A delay in a developmental domain was based on a score of 1 or more standard deviations below the mean. The results indicate that:

• 2 toddlers present with a delay in at least one developmental domain and N-ES
  • 1 toddler with delay in development of 1 SD below mean and 1 toddler with delay in development of 2 SD’s below mean

• 10 toddlers present with no delay in any developmental domain but with ES

• 10 toddlers present with a delay in at least one developmental domain and ES
  • 7 toddlers with delay in development of 1 SD below mean and 3 toddlers with delay in development of 2 SD’s below mean

This information demonstrates that almost half of our sample exhibited either early signs, a delay in developmental skills or a combination of the two. Having children with increased risk for ASD or a delay in development could have contributed to our sample exhibiting increased variability of SP function and the presence of two distinct patterns of sensory responsiveness. Further, toddlers in our sample who do not receive a
diagnosis of ASD might be categorized as the broader autism phenotype. Finally, of the two toddlers who had older siblings diagnosed with ASD, one had ES and one did not have ES.

Sensory processing patterns of sample as whole

The results of our cluster analysis confirmed the existence of two distinct sensory processing patterns within our sample. Cluster 1 was characterized by an under-responsive SP behavioral pattern. Cluster 2 was characterized by an over-responsive behavioral pattern along with increased sensory avoiding behaviors on the SPA and increased atypical sensory processing behaviors on the ITSP sensory modalities. Identification of sensory responsiveness patterns early in development is crucial. During the second year of life, the nervous system is still developing and identifying a child’s sensory response patterns can inform parents and clinicians how to adapt the environment for optimal learning to occur. It should be noted that even in a group whose mean scores indicated typical functioning, distinct SP patterns were identified. These data support the current theoretical categories of under- and over-responsiveness patterns reported in the literature (Dunn & Daniels, 2002).

The sensory processing differences between the two clusters were statistically significant (p. < 0.05) on SPA avoidance rating and all sensory modalities except for oral processing. It is not surprising that the mean scores for both clusters fell within the typical processing range on this analysis because of the findings mentioned above
regarding using mean scores on the ITSP. Analyzing data using group mean scores on ITSP with a sample that exhibits subtle sensory processing differences tends to diminish any differences that might be present.

**Patterns of sensory processing among toddlers with and without early signs of autism**

Results indicated that SP responses of toddlers in ES group differ significantly from those in the N-ES group. Specifically, statistically significant differences were exhibited on ITSP vestibular processing (p=0.041). Trends for differences between the groups were identified with avoidance behaviors on the SPA (p=0.073) and on the ITSP visual, tactile and oral processing modalities. The ES group scored lower on all sensory modalities as compared to the N-ES group indicating that the ES group exhibited a more over-responsive behavioral pattern. Ben-Sasson reported similar results of over-responsiveness in a typically developing group. Specifically, Sensory Over-Responsivity (SOR) was identified in 16.5% of a sample of typically developing children aged 7-11 years recruited from a birth-cohort sample (Ben-Sasson, Carter, & Briggs-Gowan, 2009). Other studies have identified a co-existence of under- and over-responsiveness patterns within a sample of children with ASD. Ben-Sasson and colleagues found that toddlers with ASD exhibited three patterns of sensory processing (1) low frequency of sensory behaviors compared to other clusters (2) mixed cluster with high frequencies of under and over responsiveness with low seeking and (3) high frequency of all sensory behaviors (Ben-Sasson et al., 2007). Baranek and colleagues used the Sensory
Experiences Questionnaire, a parent report tool, to measure sensory processing function in children 5-80 months. Findings indicated that 56% of children exhibited over-responsive behaviors, 63% exhibited under-responsive behaviors and 38% exhibited a combination of under- and over-responsive behaviors (Baranek et al., 2006). Similarly, a group of children with Asperger Syndrome, aged 8-14 years, displayed a range of under- and over-responsive behavioral patterns (Dunn, Myles, Orr, 2002). Differences in the age ranges studied and a confirmed diagnosis could account for these conflicting results.

Further, toddlers with and without early signs of autism displayed either under- or over-responsiveness patterns rather than a mixed SP behavioral pattern. Autism studies report a coexistence of extreme under- and over-responsiveness patterns within individuals with ASD attributed to poor sensory modulation abilities (Ben-Sasson et al., 2007). As mentioned previously, more research is needed to determine if these responsivity patterns are a function of age for this sample.

Although these groups exhibit differences in SP on the SPA and ITSP, the scores of both groups still fall within the typical range. Scores in the typical range could be present for a number of reasons including: (1) the sensory processing function in this group is typical (2) as mentioned previously, SP behaviors may be very subtle at this young age, therefore using mean scores could diminish any differences that were present (3) the tools used might not be sensitive enough to classify SP patterns in the
atypical range at such a young age (4) although the ES group exhibits early signs, they do not have ASD or (5) our sample size was too small.

Research indicates that signs and symptoms of autism are present before 2 years of age and are often noted prior to a formal diagnosis. Although there is consensus in the literature that early signs of autism are present prior to two years of age, there is no consensus as to which behavioral symptoms are most predictive of early signs of autism. Our results indicated that language and chronological age were most predictive of early signs of autism in our sample. Various studies support the findings that language deficits are present and detectable at a young age. Landa and Garrett-Mayer found that language delays were apparent by 14 months of age (Landa & Garrett-Mayer, 2006). Another study indicated that parents reported less babbling and use of gestures in early development of their children with ASD as compared to typically developing children (Ornitz, Guthrie, & Farley, 1977). Osterling and Dawson (1994) used retrospective videotaped analysis finding that children later diagnosed with ASD showed fewer communication and social behaviors compared to typically developing peers at their first birthday.

The development of assessment tools examining early signs of autism is in its infancy. It wasn’t until recently that there was a push for early detection and early diagnosis of autism. The majority of the tools available for early detection of autism are newly developed and are undergoing sensitivity and specificity testing. Without having
much data regarding the sensitivity and specificity of the tools for later ASD diagnosis, we cannot confirm which early signs are most salient.

There are several limitations of our study that should be discussed. First, our sample size was small. Second, although the ITSP is one of the most utilized SP assessments available at the present time for toddlers during the second year of life, it is a parent report which has limitations in itself. Similarly, the SPA and ADEC are new assessment tools with limited information on their sensitivity, specificity, reliability and validity.

**Future studies**

There is a need for additional SP assessments for children under three years of age. Additionally, more research is needed with the current SP assessment tools to establish sensitivity and specificity for young children with or at-risk for ASD. Additional research is needed to determine if SP function is more subtle at this age explaining scores in the typical range or if the tools utilized were not sensitive enough to detect SP dysfunction at this age. Specifically, a longitudinal study following young children to diagnosis is needed to determine the developmental progression of SP function of children with and without early signs of autism. In doing so, one could determine if SP function remains stable, gets better or deteriorates over time.
2.7 Conclusion:

This study confirmed that SP deficits are present in the ES group and can be detected during the second year of life. Overall, our sample mean scores exhibited typical SP function. When divided into clusters based on SP function and groups based on presence of early signs of autism, clear patterns of SP were identified. Toddlers with early signs of autism exhibited an over-responsive behavioral pattern towards sensory processing. Identifying patterns of sensory processing function is important in choosing which specific interventions strategies to implement in order to improve SP function. Additionally, this study has identified that chronological age and communication skills were the best predictors of ADEC score. More research is needed in the area of SP with children during the second year of life.
Chapter 3: Examining auditory processing in toddlers with and without early signs of autism using event related potentials

3.1 Abstract:

Novelty speech discrimination was investigated in toddlers, aged 12-24 months, with and without early signs of autism. Event-related potentials were recorded using an EGI GES 300 system (128 electrodes) while speech sounds were presented within an oddball paradigm. The component of interest was mismatched negativity. Between group differences of standard versus deviant waveforms were analyzed through visual inspection and statistical comparison at 8 electrode sites. Significant differences in novelty speech discrimination between groups were found at two electrode sites (T3 and Pz). Additionally, the ES group exhibited more negative-going difference waves on the right side as compared to the N-ES with more negative-going difference waves on the left side. These findings suggest that toddlers with early signs of autism processed speech sounds differently than toddlers without early signs of autism. Specifically, toddlers with early signs of ASD exhibit attenuated responses to novel speech sounds.

3.2 Background

Autism Spectrum Disorder (ASD) is a neurodevelopmental disorder characterized by core deficits in communication and socialization along with the presence of
restricted, repetitive, and stereotyped behaviors (American Psychiatric Association, 2000). In addition to core deficits, individuals with ASD frequently exhibit difficulties in processing auditory information (Bomba & Pang, 2004; Fein et al., 1996; Tomchek & Dunn, 2007). More specifically, individuals with ASD can be under-responsive to auditory information, have difficulty modulating contextually relevant auditory stimuli, have poor auditory attention, have difficulty listening while attending to another task, and/or can be over-responsive to sounds (Downs, Schmidt, & Stephens, 2005; Gomot, Giard, Adrien, Barthelemy, & Bruneau, 2002; Jones et al., 2009; Jones et al., 2003; Siegal & Blades, 2003). Dahlgren and Gillberg (1989) examined children under 2 years finding 100% of children with ASD exhibited auditory processing difficulties as compared to none of the typical children. Another study showed that 30% of children with ASD and 11% of adults with ASD exhibited an auditory processing disorder (Baranek et al., 1997). Further, Kern and colleagues reported that individuals with ASD (ages 3 to 56) displayed more severe deficits in auditory processing as compared to typical controls (Kern et al., 2006).

Event Related Potential (ERP) studies are being implemented more frequently to assess auditory processing, especially in young children. ERP is a non-invasive tool, measured from the scalp, examining sensory and cognitive processing in real time (Davies & Gavin, 2007). The brain produces electrical activity in response to an internal or external event producing an ERP waveform. ERP waveforms can be broken down
into specific components. The sequences of the components measured reflect the flow of information through the brain and the different stages of processing (Schmidt & Segalowitz, 2008). ERPs are recorded by time-locking the EEG to a specific event or stimulus presentation. Segments are created around the stimulus and these segments of the repeated stimuli are averaged together to produce a grand average waveform for each stimulus type (Segalowitz & Davies, 2004).

Mismatch negativity (MMN) is the component of interest in this study. MMN occurs between 100-300 milliseconds (ms) following stimulus onset (Lepistö et al., 2005) and is an automatic neuronal response to a change in auditory information (Dunn et al., 2008). The greater the acoustic differences between the standard and deviant stimuli, the larger the MMN will be (Cheour-Luhtanen et al., 1995; Lepistö et al., 2005; Leppänen & Lyytinen, 1997; Näätänen, 1992). MMN is elicited during an oddball paradigm where a steady stream of repetitive or “standard” stimuli is interrupted by a novel or “deviant” stimuli (Cheour et al., 1998; Schmidt & Segalowitz, 2008). MMN is elicited when a novel stimulus is detected within a stream of repetitive stimuli. The ERPs produced by the standard and deviant stimuli are compared and a difference wave is computed by subtracting the grand average standard and deviant waveforms. In typically developing individuals, the deviant stimuli often produce a larger amplitude in the negative direction at the frontocentral and temporal sites (Gomot, Giard, Roux, Barthélémy, & Bruneau, 2000). Because MMN is pre-attentive, participants are often
engaged in a different task such as a silent video or a book while MMN is being recorded. (Courchesne et al., 1989). MMN is an excellent pre-attentive measure of sensory processing and novelty detection, allowing researchers to exclude the possibility of an effect relating to impaired attention, motor reaction, or task related responses (Schmidt & Segalowitz, 2008).

The majority of ERP studies involving the autism population focus on auditory processing and its potential link to language and communication impairments (Whitehouse & Bishop, 2008). While children with autism tend to experience typical function in peripheral sensory registration (e.g. hearing acuity is within normal limits) (Rogers & Ozonoff, 2005), they exhibit attenuated levels of responsiveness to auditory stimuli, particularly to speech sounds (Dunn, Gomes, & Gravel, 2008). Recent studies suggest that MMN has the potential to be utilized for diagnostic and clinical purposes (Schmidt & Segalowitz, 2008).

To date, the auditory ERP studies focusing on MMN in individuals with ASD have reported inconsistent findings. There seem to be differences in the way individuals with ASD process speech versus non speech sounds. Some researchers are finding that with speech sounds, individuals with ASD have absent or reduced MMN as compared to typically developing peers while others find no difference. Additionally, some researchers are finding enhanced MMN (to non-speech sounds) in individuals with ASD. Exhibiting an absent or reduced MMN could indicate difficulty with automatic
processing of auditory information. Dunn and colleagues found a smaller amplitude (using pure tones) in the MMN component for children with autism (ages 6-12 years) as compared to typical controls during unattended conditions but similar MMN during attended conditions (Dunn et al., 2008). Whitehouse and Bishop also reported that children with ASD (7-14 years) exhibited a reduced MMN to speech sound, but not complex tones, when not attending to the sounds (Whitehouse & Bishop, 2008). Kuhl and colleagues reported that individuals with ASD displayed smaller MMN amplitude to unattended speech stimuli (Kuhl et al., 2005). Reduced MMN amplitude in individuals with ASD for unattended stimuli but not for attended stimuli supports the theory that individuals with ASD have difficulty automatically processing auditory stimuli (Dunn et al., 2008). Impairments in automatic processing of auditory stimuli could partly explain why individuals with ASD have difficulty with language development (Dunn et al., 2008).

Kuhl and colleagues examined auditory processing in a group of preschool children with three interesting findings (1) as a whole, preschoolers with ASD did not produce an MMN to speech sounds whereas typically developing children did (2) as a group, preschoolers with ASD preferred non-speech sounds over speech sounds (3) preschoolers with ASD were separated into two groups: those who preferred speech and those who preferred non-speech. Those who preferred speech sounds exhibited a MMN similar to the typical group; those who preferred non-speech did not exhibit a MMN (Kuhl et al., 2005). Additionally, these authors reported an association between speech processing and more severe symptoms of ASD.
In contrast to these findings of attenuated responses to MMN, other researchers have found no difference in MMN in individuals with and without ASD. Cepioniene and colleagues measured MMN in high functioning children with ASD and typical controls (using simple tones, complex tones, and vowel sounds), finding no difference in MMN response between the two groups. Instead, they found an absent P3a for speech sounds only (Čeponienė, Lepistö, Shestakova, Vanhala, Alku, Näätänen, & Yaguchi, 2003). P3a involves involuntary orienting to changes in an auditory stimulus. This study was in agreement with Kemner and colleagues who found no differences in MMN elicited by speech sounds in children with ASD (Kemner, Verbaten, Cuperus, Camfferman, & van Engeland, 1995). Additionally, Gomot et al. (2002) found no significant amplitude differences in MMN to changes in tone frequency in children (5-9 years) with and without ASD. However, they did find shorter latency of MMN in the ASD group and the MMN was followed by a P3a component (Gomot et al., 2002). Ferri et al. (2003) found an enhanced MMN in a group of individuals with ASD (6-19) to changes in frequency of tones followed by a P3a component (Ferri et al., 2003). Reports of increased MMN to increased pitch sensitivity could at least partially explain the auditory hypersensitivity reported in some children with ASD (O’Neill & Jones, 1997).

Although MMN has not been studied in children with autism younger than preschool age, it has been studied early in life with typically developing infants. MMN has been examined longitudinally in the first year of life by several investigators and has
been detected in infants as young as 3 months of age (Kushnerenko et al., 2002).

Evidence from these studies suggests that infants (aged 0-12 months) exhibit stability in regards to amplitude and latency of the components. (Cheour et al., 1998; Kushnerenko et al., 2002) Additionally, infants exhibited a MMN component approximately 75% (Kushnerenko et al., 2002) to 100% of the time during the first year of life (Cheour et al., 1998; Kushnerenko et al., 2002). However, there was significant variability within individual MMN components within the same subjects over time (Kushnerenko et al., 2002). In infants, mean latency of MMN decreases with age, however, the amplitude is relatively similar in infants and adults, the (Cheour et al., 1998). The auditory system mature early in life (Cheour et al., 1998) and remains relatively stable over time (Cheour et al., 1998; Kushnerenko et al., 2002). MMN has been recorded using changes in pure tones and speech sounds in infants under 12 months of age (Cheour et al., 1998; Čeponienė, Lepistö, Shestakova, Vanhala, Alku, Näätänen, & Yaguchi, 2003). MMN is a valuable tool for studying auditory discrimination in young children as it is elicited by unattended stimuli and can be recorded in a variety of states, including sleep (Naatanen et al., 1993).

Although ERP studies are known more for their temporal resolution rather than their anatomical localization patterns, studies have reported abnormal activity at the frontal and temporal scalp electrode sites in children with ASD. Kleinhans and colleagues showed that adults with ASD exhibited greater activation patterns on the
right frontal and right superior temporal lobes during a letter fluency task than typical controls (Kleinhans, Müller, Cohen, & Courchesne, 2008). Another study used water positron emission tomography (PET) to examine brain organization for language and auditory functions in adults with and without ASD. They found that the ASD group exhibited reversed hemispheric dominance during a verbal auditory task and a trend in reduced activation of the auditory cortex during an auditory task (Müller et al., 1999). These studies support the theory that atypical functional neural organization could contribute to language difficulties reported in individuals with ASD (Kleinhans et al., 2008).

Auditory processing deficits are thought to negatively impact the development of speech and language. Language abilities vary dramatically amongst children with ASD. Approximately 25-50% of children with ASD never develop functional language skills and those who do develop functional language often do so at a slower rate or at a later age than typically developing children (Gillberg & Coleman, 2000; Rapin & Dunn, 2003; Tager-Flusberg & Joseph, 2003). Recently, there has been a strong emphasis on detection of autism at a younger age. Earlier detection leads to earlier intervention and a potentially better outcome. Taking into consideration that (1) a diagnosis of autism is strongly reliant on language and communication abilities and (2) there is a high prevalence of auditory processing and language disorders in older children with autism,
a logical next step would be to examine auditory processing abilities as they relate to language in very young children for possible early detection.

Research indicates that typically developing infants and toddlers can discriminate differences in speech sounds, show social interest in speech and language, and learn through increased exposure to language (Kuhl, 2000; Tsao, Liu, & Kuhl, 2004; Tsao, Liu, & Kuhl, 2004). An infant’s interest in speech and language enhances his or her ability to learn language (Fernald, 1985; Fernald & Kuhl, 1987; Kuhl, Tsao, & Liu, 2003) and a parent’s response to their child’s language attempts affects language development. Specifically, parents who immediately respond to their child’s vocalizations see improvements in the quality and quantity of their vocalizations (Goldstein, King, & West, 2003; Kuhl et al., 2003). Additionally, the ability to perceive speech sounds during infancy has been shown to reliably predict language skills at a later age. Kuhl and colleagues showed that infants’ speech discrimination skills at 6 months predicted their language abilities at 13, 16, and 24 months of age (Tsao, Liu, & Kuhl, 2004).

When comparing language development of typical infants as discussed above to language development of individuals with autism, there are several differences noted. Specifically, individuals with autism often lack social interest in communication and have difficulty processing emotional and social information during social situations (Cohen, Baron-Cohen, & Tager-Flusberg, 1993). Additionally, some children with ASD have
abnormal responses to speech sounds affecting their socialization abilities (Klin, 1991; Klin, 1992). Our knowledge relating to typical infant and toddler language development and the language impairments of toddlers with ASD solidifies that collecting ERP data to examine early auditory processing in toddler with early signs of autism could be advantageous. A young child's ability to learn through exposure to language could depend on both an early social interest in speech and language and the ability to discriminate speech sounds.

This is the first study to our knowledge to report auditory processing in toddlers with and without early signs of autism using ERP. The aims of this study were to (1) examine the differences in auditory processing between toddlers with and without early signs of autism; (2) examine brain activation patterns between 0-300 ms post stimulus onset in toddlers with and without early signs of autism; (3) determine the feasibility of collecting ERP data on this population. Our hypotheses were: (1) toddlers with early signs of autism will exhibit differences in detection of novel speech sounds compared to toddlers without early signs of autism, and (2) brain activation patterns to novel speech sounds will differ between toddlers with and without early signs of autism. Specifically, toddlers with no early signs of autism will have increased activation in the left temporal and left frontal lobes whereas the toddlers with early signs of autism will have increased activation patterns on the right sides or bilateral activation patterns. This study is the initial step in examining whether or not there are differences in how toddlers with and
without early signs of autism detect very subtle changes in speech sounds and testing
the feasibility of obtaining reliable ERP data from this population. The detection of
auditory processing deficits in toddlers with early signs of ASD has the potential to
significantly impact the development of language, communication, and socialization
skills of these children. Early identification of auditory processing deficits can lead to
access to early intervention services and provide insight as to how to focus intervention.

3.3 Research Design and Methods

3.3.1 Participants and Recruitment

Forty-six toddlers (Mean age =17.9 months, SD=+/- 3.0; 28 males) participated in this
study. Toddlers with and without risk factors for autism were recruited to the study
from the following facilities and events: The Ohio State Child Care Center, Medical
Center and Nisonger Center, local businesses/offices (pediatricians, therapy clinics,
schools, etc), Nationwide Children’s Hospital, Research Match, Autism Speaks Walk, and
Autism Society of America: Central Ohio Chapter (website and newsletter). Inclusion
criteria were:

- Aged between 12-24 months
- Intact hearing (assessed by parent report of newborn hearing screening)

In order to maximize the number of participants showing early signs of autism, toddlers
with high risk for autism including siblings, history of premature birth and/or
developmental delay, were targeted in recruitment efforts. Toddlers were not eligible
for study participation if they were diagnosed with a developmental disorder other than autism prior to entering this study (including cerebral palsy, seizure disorders, brain injury e.g. periventricular leucomalacia, intraventricular hemorrhage, Down syndrome).

For the purposes of analysis, participants were classified into either the no early signs of autism group (N-ES) or early signs of autism group (ES) using an early autism detection tool, the Autism Detection in Early Childhood (ADEC) (Young, 2007) (described below). Groups were significantly different based on ADEC score (p=0.001). Further, the ES group was younger than the N-ES group (p=0.018). (Participant demographics are described in Table 3.1.) One subject from each group was not able to tolerate wearing an ERP net.

Table 3.1: Sample demographics

<table>
<thead>
<tr>
<th>Sample Characteristics</th>
<th>No Early Signs</th>
<th>Early Signs</th>
</tr>
</thead>
<tbody>
<tr>
<td>Number recruited</td>
<td>24</td>
<td>22</td>
</tr>
<tr>
<td>Male/female</td>
<td>12/12</td>
<td>16/6</td>
</tr>
<tr>
<td>Mean age in months (SD)</td>
<td>19 (3.5)</td>
<td>16.5 (3.8)</td>
</tr>
<tr>
<td>ADEC Score (SD)</td>
<td>3.4 (1.4)</td>
<td>8.5 (2.5)</td>
</tr>
</tbody>
</table>

3.3.2 Study procedures

Parents interested in participating in the study contacted the SenSA Lab at The Ohio State University. An initial telephone screening ensured that toddlers met the inclusion criteria. Following the telephone screening, two study visits were scheduled. The ADEC was administered in the first visit along with other developmental and clinical
measures and the ERP conducted in the second. On average, the first and the second visit occurred 12 days apart. Parents were either mailed or emailed directions to the lab and consent form for review. Additionally, a reminder email/phone call was given the day prior to the appointment. This study was approved by the Institutional Review Board at The Ohio State University and Nationwide Children’s Hospital.

3.3.3 Instrumentation

Autism Detection in Early Childhood (ADEC): (Young, 2007) Risk factor status was classified through administration of the ADEC which detects ASD in pre-verbal infants. The ADEC consists of 16 items and an adaptation period measured through direct observation. The adaptation period is used when the toddler needs a break and/or becomes fussy. The items presented in the ADEC fit into one of three domains: (1) Disturbances in interacting with others and with object (e.g. poor imitation, lack of orienting to name, rare use of gestures, etc.), (2) Stereotyped, repetitive movements (arrange objects in a line, hand flapping, finger flicking, etc.) and (3) Bizarre responses to environmental stimuli (covering ears in response to sound, mouthing objects, and smelling objects or people). Grouping was based on the following criteria: toddlers scoring between 0-5 were classified as N-ES (n=24) and toddlers scoring a 6 and above were classified as ES (n=22). The cut-off score for the ES group used for this study deviated from the cut-off scores indicated in the assessment manual. The manual indicates individuals scoring between 0-10 are at low risk for ASD, 11-13 are at moderate risk, 14-19 are at high risk and >19 are at very high risk. In this study, clinical
differences were apparent for toddlers scoring 6 or more on the ADEC. These clinical differences were discussed with the author of the assessment and permission was given to use a score of 6 or above as the cut-off score for the presence of early signs of ASD in this study.

**Event Related Potential: Auditory Paradigm:** Speech sounds were presented using an oddball paradigm in blocks of 400 stimuli, each block lasting approximately 8.5 minutes. The standard (repetitive) stimuli were presented 85% of the time and deviant (novel) stimuli presented 15% of the time. The stimulus duration was 340 ms and the inter-stimulus interval (ISI) was 960 milliseconds. Synthesized speech was utilized, specifically phonemes (dae and daa). For each block, randomization occurred as to which phoneme was chosen as the standard/deviant stimuli. The order of each phoneme presentation was pseudo-randomized. Within each block, at least 4 standard stimuli were presented before and after each deviant stimulus. The deviant stimuli were pseudo-randomized so they were spread out among the standard stimuli within the block to eliminate them all occurring at the beginning or the end of the block.

### 3.3.4 Data collection and signal filtering procedure

At the first study visit, parental consent was obtained and a verbal explanation of the procedures was given. Before beginning data collection for the ERP experiment, parental questions were answered and an explanation of the importance of minimizing movement while collecting data was given.
Continuous EEG data was collected in a small, sound proof room with auditory stimuli presented binaurally via wall speakers. The ERP recording device was in a room adjacent to the data collection room. Participants sat on their caregivers’ lap (Figure 3.1) throughout the electrode net placement and testing session to increase the toddler’s comfort level and minimize movement. While the auditory stimuli were presented, one of the researchers (KH) remained in the room with the participant and their caregiver completing a variety of tasks to visually entertain the toddler, while attempting to minimize movement. These tasks included: silent movie, ipod touch, bubbles, puppets, books, stickers, putting foam blocks and playdoh balls into a
container, crayons, and making silly faces/facial expressions. Although the duration of activities varied among toddlers, all of the above mentioned activities typically occurred during each session. Activities were graded according to amount of movement they created. Tasks were terminated if they created too much movement causing excessive artifact and/or if it did not capture the toddler’s attention.

### 3.3.5 Signal Filtering

Data were collected using an EGI GES 300 system (Electrical Geodesics, Inc, Eugene, OR) utilizing a HydroCel 128 Channel Geodesic Sensor Net and a Net Amps 300 amplifier. The Cz electrode was used as the reference electrode. The signal was processed with Net Station software undergoing the following analysis: (1) filtering (high-pass=0.1 Hz and low-pass=30Hz), (2) segmentation, (3) artifact detection, (4) bad channel replacement, (5) referencing and (6) baseline correction. Continuous EEG data were segmented into epochs of 200 ms pre-stimulus (baseline) and 900 ms post-stimulus. Trials with eye blink artifact >140 mV, eye movement >55 mV, or artifact >100 mV were removed from the data set. Any subject who had less than 10% of the deviant stimuli remaining after signal processing were removed from the data set (ES, n=10 removed; N-ES, n=15 removed).

The final data set included the following deviant/standard ratios: N-ES standard: 81-853 (M=225, SD=239), deviant: 18-151 (M=41, SD=42); ES standard: 62-493 (M=248, SD=128), deviant: 18-83 (M=43, SD=21). Note that in the N-ES group there was one subject who sat very still and was considered an outlier in the number of stimuli
remaining in the data set following signal processing with 853 standard stimuli and 151 deviant stimuli. With this subject removed, standard stimuli: M=147, SD=44 and deviant stimuli: M=27, SD=7.

3.3.6 Data analysis

The component of interest in this study was MMN. The MMN was defined as the largest negative deflection within 100-300 ms (Lepistö et al., 2005). Data analysis was completed within and between groups. For each subject, grand average waveforms were computed for each stimulus type (standard and deviant) at all electrode sites. Difference waveforms were calculated for each subject and averaged for each group. Grand average standard and deviant waveforms were calculated for each group across all electrode sites. Visual analysis compared groups for: (1) differences in amplitude and latency for standard compared to deviant stimuli for eight electrode sites, (2) differences in brain activation patterns for standard and deviant stimuli and (3) comparison of difference waves. Difference waves were calculated by subtracting grand average deviant and standard waveforms and compared between groups. Statistical analysis for between-group differences for the area under the curve between 100-300 ms were calculated for the deviant – standard waveform using independent t-tests. Statistical analysis was completed in Excel 2010.

3.4 Results
**Visual analysis**: Grand average waveforms for standard and deviant stimuli at 8 electrode sites for both groups are presented in figure 2. The electrodes used for analysis are located in the following scalp areas: T3 and T5- left temporal; T4 and T6- right temporal; Cz, C3 and Fz- frontocentral; Pz- parietal. Visual inspection of the data shows a more pronounced difference between standard and deviant waveforms between 0-300 ms, where we would expect to see a MMN, at four electrode sites for the N-ES group when compared to the ES group (See Figure 3.2). Specifically, there is a bigger difference between standard and deviant stimuli at approximately 150 ms for electrodes T3, T4, T5, and Pz, and at approximately 225 for electrode C3. Additionally, the difference waves were compared between groups across time points to see which group had the more negative-going wave at each electrode. A more negative difference wave suggests that the deviant stimuli were detected among a stream of standard stimuli. Each group had seven difference waves that were more negative at different time points (Table 3.2). Specifically, the difference waves for the N-ES group were more negative between 0-100 ms at the T3, T5, and Fz electrodes, between 100-200 ms at the T3 and T4 electrodes and between 200-300 ms at electrodes T5 and Fz. The ES group was more negative than the N-ES group between 0-100 ms at electrode sites Pz and T6, between 100-200 ms at electrode Pz and between 200-300 ms for electrodes Pz, Cz, T4, and T6 (Figure 3.3). Four of the seven electrodes for the ES group that had a more negative difference wave occurred between 200-300 ms post stimulus onset. Differences in activation patterns were also noted. Specifically, the N-ES had more
activation on the left hemisphere whereas the ES group had more activation on the right hemisphere.
Figure 3.2: Grand average of deviant and standard waveforms for NO-ES and ES groups

Continued
Figure 3.2 Continued

No Early Signs

Early Signs

Standards

Deviants
Table 3.2: Visual representation reporting which group had a more negative difference wave between 0-300 ms post stimulus onset (by electrode). The empty blocks represent time periods when the difference wave was similar between two groups.

<table>
<thead>
<tr>
<th>Time</th>
<th>T3</th>
<th>T5</th>
<th>Pz</th>
<th>Fz</th>
<th>Cz</th>
<th>C3</th>
<th>T4</th>
<th>T6</th>
</tr>
</thead>
<tbody>
<tr>
<td>0-100 ms</td>
<td>N-ES</td>
<td>N-ES</td>
<td>ES</td>
<td>N-ES</td>
<td></td>
<td></td>
<td></td>
<td>ES</td>
</tr>
<tr>
<td>100-200 ms</td>
<td>N-ES</td>
<td></td>
<td>ES</td>
<td></td>
<td></td>
<td></td>
<td>N-ES</td>
<td></td>
</tr>
<tr>
<td>200-300 ms</td>
<td>N-ES</td>
<td>ES</td>
<td>N-ES</td>
<td>ES</td>
<td></td>
<td></td>
<td>ES</td>
<td>ES</td>
</tr>
</tbody>
</table>
No Early Signs

Early Signs

![Waveforms](image)

Figure 3.3: Difference waveforms (deviant – standard) for N-ES and ES groups

Continued
Figure 3.3 Continued
Statistical analysis: Results exhibited statistically significant differences for the area under the curve between 100-300 ms for difference waveforms at electrode sites T3 and Pz. The area under the curve is more negative at T3 for the N-ES group and more negative at the Pz electrode for the ES group. Additionally, at electrodes T5, Fz, and T4, the area under the curve is more negative for the N-ES as compared to the ES group, but the differences are not significant.

Table 3.3: Area under the curve for groups by electrode

<table>
<thead>
<tr>
<th>Electrode</th>
<th>N-ES (n=9) Area under the curve (SD)</th>
<th>ES (n=12) Area under the curve (SD)</th>
<th>p value</th>
</tr>
</thead>
<tbody>
<tr>
<td>T3</td>
<td>-75.74 (201.91)</td>
<td>98.31 (93.81)</td>
<td>0.036**</td>
</tr>
<tr>
<td>T5</td>
<td>-116.27 (299.19)</td>
<td>49.33 (122.65)</td>
<td>0.097</td>
</tr>
<tr>
<td>Pz</td>
<td>130.79 (112.86)</td>
<td>-137.06 (135.04)</td>
<td>0.007**</td>
</tr>
<tr>
<td>Fz</td>
<td>-21.93 (131.52)</td>
<td>14.19 (102.12)</td>
<td>0.487</td>
</tr>
<tr>
<td>Cz</td>
<td>91.52 (217.77)</td>
<td>-7.14 (142.40)</td>
<td>0.258</td>
</tr>
<tr>
<td>C3</td>
<td>126.58 (164.88)</td>
<td>60.67 (124.44)</td>
<td>0.309</td>
</tr>
<tr>
<td>T4</td>
<td>-72.88 (153.41)</td>
<td>-7.99 (162.67)</td>
<td>0.366</td>
</tr>
<tr>
<td>T6</td>
<td>51.09 (197.48)</td>
<td>-36.18 (145.57)</td>
<td>0.257</td>
</tr>
</tbody>
</table>

**Significant difference

3.5 Discussion

Using ERP to examine automatic auditory processing in a younger population is becoming more common. ERP has the potential to examine sensory processing, detection, and discrimination of auditory stimuli without requiring attention or a motor response, therefore it is ideal for very young children, children with cognitive
impairments and/or children with language impairments. The aims of the present study were to: (1) determine if there were differences in discrimination of speech sounds in toddlers with and without early signs of autism, (2) compare lateralization/activation patterns in toddlers with and without early signs of autism and (3) assess the feasibility of collecting ERP data in this population. The results indicate that there are subtle differences in the way that toddlers with early signs of autism detect novel speech sounds compared to toddlers without early signs of autism. Specifically, visual inspection of the data shows that the N-ES group exhibited greater difference in the amplitude between standard and deviant waveforms for electrodes T3, T4, T5, and Pz at 150 ms post-stimulus onset and for C3 at 225 ms post-stimulus onset. Additionally, visual comparison of the difference waves between the groups indicates that both groups had seven difference waves that were more negative at some time point between 0-300 ms post stimulus onset. Additionally, statistical analysis indicated that there were differences (p=0.036 and p=0.007) at two electrode sites (T3 and Pz, respectively) when comparing difference waves between groups. Results indicated that there are subtle differences in the way that toddlers with early signs of autism process novel speech sounds as compared to toddlers without early signs of autism. Toddlers with early signs of ASD exhibited attenuated responses to novel speech stimuli. Factors that could have influenced our signal making differences between groups subtle are presented in the next sections.
As expected when collecting data with young subjects, our EEG signal contained artifact. There are several reasons why EEG data contains artifact therefore reducing the signal-to-noise ratio. Signal-to-noise ratio is poor in infants and toddlers as compared to older children and adults because: (1) infants and toddlers tend to have larger and more variable background EEG noise leading to a noisier signal, (2) more trials are eliminated with infants/toddlers due to excessive artifact and (3) ERP data of infants and toddlers is more variable from trial to trial than in older children due to frequent changes in arousal or attention (Picton & Taylor, 2007). Our data naturally fit into all of these categories and had increased noise due to the toddlers moving, talking/babbling, and so on. To account for increased noise within the signal, a second level of analysis was completed to eliminate more noise and be more conservative about our data (results in Appendix H). Moving forward, studies with more subjects should be implemented to increase the power of the signal and improve signal-to-noise ratio. In planning for future studies, knowing that 25-50% of the subjects could be eliminated due to high levels of artifacts, over recruitment of subjects should be considered.

The differences between the two stimuli used in this experiment were very subtle. The stimuli (daa and dae) were only different in the ending vowel sounds. As we know from previous research in MMN, the more subtle the changes in speech stimuli, the less effect it has on the amplitude whereas larger changes between the stimuli have a larger affect on the amplitude. This could be one reason there were only 2 statistically
significant differences among our electrodes of interest. Future studies should use speech sounds with less subtle differences and differences in the beginning sound rather than the end sound (for example, changing the beginning consonant sound, daa vs maa).

McGee and colleagues found that MMN magnitude is reduced significantly after the first 10 minutes of the experiment. In McGee’s experiment, only a small MMN response was exhibited after a long data collection session whereas a larger response was exhibited at the beginning of the experiment (McGee et al., 2001). The majority of the toddlers in this experiment sat through 3 blocks of speech sounds, each lasting approximately 8.5 minutes. Small breaks were taken between blocks if needed. In total, the toddlers sat for approximately 30 minutes for this task. It is questionable whether task length affected our results. This question should be addressed in future studies by dividing the ERP visit into more than one session or providing an extended break in between blocks.

The sample utilized in this study, included a group of toddlers with early signs of autism and a group without early signs of autism. It is very likely that in the early signs of autism group, some of the toddlers will go on to develop autism and some toddlers will not develop autism. This factor could have a strong influence on how the group as a whole processed speech sounds. If only 3 of the 12 toddlers with early signs of autism go on to develop autism, this could add to the explanation of why there were significant
differences in only 2 electrodes. Additionally, as shown in Kuhl et al., preschoolers with autism who preferred speech sounds exhibited typical MMNs whereas those who preferred non-speech sounds exhibited attenuated MMN to speech sounds (Kuhl, Coffey-Corina et al., 2005). Adding an auditory preference test to this study would strengthen the design and allow for a more thorough analysis.

Our second aim was to examine brain activation and lateralization patterns while processing speech sounds between the two groups. Current evidence suggests that children with autism have different activation patterns for speech sounds as compared to typically developing children. Specifically, children with autism have exhibited greater right hemisphere activation patterns to speech tasks in the frontal, insular, and temporal areas as compared to the typical controls (Kleinhans et al., 2008). Additionally, Kleinhans et al. showed that the majority of the typically developing subjects exhibited greater left asymmetry exclusively activating the left frontal lobe whereas the majority of the ASD group showed either right, bilateral or weak left side activation patterns. The statistical results for the area under the curve indicated that the ES group had more negative amplitudes for right side electrodes as compared to the left side. In looking at the area under the curve for the N-ES group, they exhibit negative results for both left side electrodes and one right side electrode. The toddlers in our sample were under 2 years of age. The left dominance for language is not fully developed at this time (Amunts, Schleicher, Ditterich, & Zilles, 2003), which would explain why we observed a more bilateral activation and/or stronger activation pattern.
in the left hemisphere for the N-ES group. However, lateralization to the right side for toddlers with early signs of ASD could also indicate a pathological pattern of development which should be examined further.

Finally, our last aim was to test the feasibility of collecting ERP data on toddlers with and without early signs of autism. The results of our study show that it is feasible to collect ERP data with this population. However, there are several factors to consider when planning future ERP studies with toddlers in the second year of life to improve the signal-to-noise ratio. First, the researcher should be prepared to eliminate 25-50% of the subjects due to excessive artifact as toddlers naturally tend to move a lot. The amount of movement made by toddlers can be controlled to a certain extent by grading the tasks presented during data collection. Over recruitment is needed to offset the elimination of subjects with excessive artifact. In this experiment, more than 50% of the subjects were eliminated from the final data set. Second, with careful experimental design, data with a high signal-to-noise ratio can be collected. For example, activities completed while the toddler’s are listening to speech sounds should be chosen with caution. Activities performed in the toddlers lap with minimal head turning and minimal gross arm movements are ideal. The activities that were most successful in creating the least amount of artifact and capturing the toddlers attention for the longest in this experiment were: iPod touch®, putting stickers on a piece of paper or in a puppet’s mouth, and putting small foam blocks and playdoh balls into different plastic containers. In future experiments, an iPad® would also be a good distracter and attention keeper.
There are many silent applications for this device that require little to no motor accuracy, and are visually stimulating. Finally, a balance needs to be found between the number of trials presented (length of sitting) and the stillness that can be expected from toddlers in order to get enough usable trials once artifacts are eliminated. The optimal situation for collecting artifact-free ERP data is with a subject who is calm and sitting silently with minimal movement including eye blinking. When collecting ERP data on older subjects, this can be explained and a level of compliance can be expected. Explaining this to a toddler is not possible. In this experiment, the stimuli were presented in blocks of 400, each block lasting approximately 8.5 minutes. The stimuli were split into blocks so that the caregivers and toddlers could take small breaks in between blocks getting some movement out before having to sit still for another block.

Researchers collecting ERP data with young children are often interested in early ERP components because of their pre-attentive nature. However, there are pros and cons for examining early ERP components. Early components do not require the toddler to attend to the task or elicit a behavioral response. Additionally, they provide information regarding sensory processing and sensory detection that we cannot see behaviorally or clinically at such a young age. However, the earlier the component of interest, the smaller the effect it has on the amplitude of the ERP; therefore, requiring a larger number of trials to enhance the signal-to-noise ratio. Keeping this in mind, several aspects of the experiment were carefully considered to keep toddlers sitting for longer periods of time. First, toddlers sat on their caregivers lap to increase their
comfort level and reduce their anxiety during the experiment. Second, one experimenter was present in the testing room at all times playing with the toddler, keeping him/her content and still (tasks explained above). Because the ERP component of interest in this experiment was MMN, which is pre-attentive, toddlers were able to attend to tasks other than the speech sounds, allowing us to capture their attention and keep them seated for longer periods of time. Last, all data collection appointments were scheduled around the toddler’s nap and mealtime schedule and breaks were taken as needed to ensure the toddler would perform their best during data collection.

3.6 Conclusion

This is the first ERP study to report novel speech detection with toddlers aged 12-24 months with and without early signs of autism. Preliminary findings suggest that there are subtle differences in the way toddlers with and without early signs of autism process novel speech sounds. Toddlers with early signs of autism exhibit attenuated responses to novel speech sounds at some electrode sites. Replication of this study with a larger sample size is recommended to optimize the signal-to-noise ratio.

3.7 Limitations

Sample size was small. Although a total of 46 children participated in this study, the final sample size included 21 subjects. EEG waveforms had a high amount of artifact due to several factors including toddler movement resulting in elimination of more than 50% of the subjects from the data set. Additionally, it was difficult to determine the
appropriate level of artifact detection; therefore two sets of data were obtained making data interpretation difficult.

3.8 Future studies

This study should be replicated with a larger sample size. Once this study is replicated and validated as a valuable technique to use on toddlers with and without early signs of autism, it would be a valuable tool to combine with other behavioral measures (auditory preference test and behavioral sensory processing measures) to examine phenotypes within the groups. Future studies testing the methodological changes described above (less subtle differences between speech sounds, data collection occurring on separate days or with long breaks) are needed. To validate the sample, additional studies should also use the Modified Checklist for Autism in Toddlers (M-CHAT) or an additional early autism screening tool to verify that the toddlers in the ES group have early signs of ASD. Additionally, ERP has the potential to be a direct measure of the effect of treatment addressing auditory processing. In the adult population, ERP has been used as a pre and post measure to assess treatment effects (Kraus et al., 1995). Eventually ERP has the potential to be utilized before and after treatment to assess whether or not brain activity changes following intervention. Additionally, the majority of studies examining auditory processing differences between two subject groups use either parent report or behavioral measures. These measures should be used in addition to assessments with a direct measure of neural processing.
Finally, future studies should examine the association between severity of language deficit and social impairment with brain functioning with auditory processing.
Chapter 4: Effectiveness of interventions targeted at children with autism, ages 0-24 months: A systematic review

4.1 Abstract

Early intervention (EI) is thought to optimize outcomes for children with an autism spectrum disorder (ASD). This systematic review examines the effectiveness of early interventions for children with or at-risk for ASD and applies these findings to occupational therapy practice. Seven databases were systematically searched, resulting in 16 articles. Inclusion criteria were: (1) intervention began prior to 24 months (2) participants were with/at-risk ASD and (3) outcome measures addressed core autism symptoms. Articles were critically appraised by two independent reviewers. Studies were primarily single group cohort designs. Interventions included early intensive behavioral interventions, Early Start Denver Model, relationship-based intervention, blended approach and imitation based training. Key outcomes were gains in communication, social interactions, joint attention, adaptive behavior, motor skills, autism severity, and imitation skills. This review suggests that early intervention for children under 2 years of age with/at-risk for autism ameliorates core ASD symptoms.
The Early Start Denver Model studies demonstrated the highest level of methodological rigor and positive changes in autism behaviors.

4.2 Background

The prevalence of autism spectrum disorders (ASD) has increased over the past several decades progressing from a rare disorder to one of the most commonly diagnosed developmental disabilities \cite{482,192}. Current estimates suggest ASD affects 1 in 91 children \cite{192}. The rise in ASD prevalence along with the life-long impact of the disorder makes it an urgent public health concern that requires extensive research.

ASD is a neurodevelopmental disorder characterized by core deficits in communication and socialization and the presence of restricted, repetitive, and stereotyped behaviors \cite{482,118,63,194}. Research findings identifying the initial ages that ASD symptoms can be detected are inconsistent. Although it has been detected in children as early as 12-18 months, children are rarely diagnosed this early \cite{114,19,123,67,211}. The average age for ASD diagnosis ranges from 2 to 7 years \cite{19,103,106,189}. Researchers estimate that more than fifty percent of parents of children with ASD suspect a developmental issue before their child’s first birthday \cite{106,67,211,123}.
Unfortunately, parental concerns are often ignored or discounted as individual variability is inherent with developmental milestones \{106 Osterling, J. 1994; 19 Filipek, PA 2000\}).

Early signs of autism are most commonly identified in the areas of socialization, motor, language, communication, and sensory processing. Behavioral and sensorimotor characteristics identified prior to 24 months of age in children later diagnosed with ASD include variability in social engagement, fewer joint attention attempts, limited imitation skills, poor eye contact or visual avoidance, excessive mouthing of objects, social touch aversion, absence of response to familiar voices, diminished response to pain, sensitivity to tastes and textures, and decreased orienting to name \{123 Werner, E. 2000; 106 Osterling, J. 1994; 97 Moore, V. 2003; 433 Kasari, C. 2002; 108 Mars, A.E. 1998; 102 Cox, A. 1999; 28 Tomchek, S.D. 2007\}).

As the prevalence of ASD increases and the age of detection and diagnosis decreases, early intervention (EI) for children with/at-risk for a diagnosis of autism is an important topic. Several decades ago, it was thought that an ASD diagnosis meant irreversible, lifelong impairments in communication, socialization and restricted interests \{50 Dawson, G. 2008\}). Advances in the field of autism have led researchers and clinicians to be more optimistic in terms of possible outcomes for individuals with ASD. Research indicates that the earlier intervention begins, i.e., prior to four years of age, the better the outcome \{479 Green, G. 2002; 402 Harris, S.L. 2000; 484 National
Research Council 2001; 504 Birkin, C. 2008}) and EI has the potential to change the developmental trajectory of the disorder {50 Dawson, G. 2008}. With improved understanding of early signs of autism and increased access to early assessment and screening tools, clinicians and pediatricians have the opportunity to identify ASD within the first two years of life {217 Zwaigenbaum, L. 2009}. Currently, the literature as to which intervention program is most effective for children under 2 years with/at-risk for ASD {132 Klin, A. 2008; 492 Vismara, L.A. 2008; 112 Rogers, S.J. 2008} is inconclusive. Therefore, identifying which intervention(s) is most beneficial for young children with/at-risk for autism is a priority.

Data from scientifically validated research and interventions programs are needed to drive changes in policy regarding which intervention should be implemented, for what duration and at what intensity for children under two years of age who are with/at-risk for ASD. Recommendations of the National Research Council state that early intervention services for individuals with autism should be implemented at a high intensity {484 National Research Council 2001}). However, most early intervention programs publicly funded through Part C implement 90 minutes of intervention per week {499 Hebbeler, K. 2007}). Researchers and clinicians need to examine what takes place during intervention hours and assess intervention characteristics such as: (1)the quality of intervention (2)intensity and duration of intervention (3)whether or not generalization of skills is occurring (4)what curricular domains and functional skills are being taught (5)how is change or progress being monitored (6)how the program is
changing as child progresses and (7) the effect size of the intervention. The data collected supporting most efficacious interventions needs to influence changes in policy relating to the delivery of early intervention.

4.3 Methods

4.3.1 Search Strategy

Research reports published through August, 2010 were included in the review. Seven databases (Medline, Cinahl, Web of Knowledge, Cochrane, PubMed, DARE, and ProQuest) were searched for papers meeting the inclusion criteria. Additionally, experts in the field were contacted to ask about ongoing research and/or unpublished projects related to our topic. Search term combinations included:

1. population terms: autism/ASD/Asperger/PDD NOS/pervasive developmental disorder/autistic/prem/bab/low birth weight/LBW

2. age group terms: child/young person/young child/infant/toddler/bab

3. intervention terms: early intervention/head start

4. outcome terms: autism

severity/outcome/communication/social/language/behavior /development /sensory/motor

4.3.2 Selection Criteria:

To be included in this systematic review, studies had to meet the following criteria:
1. intervention began prior to 24 months of age
2. participants diagnosed with or at-risk for ASD
3. outcome measures addressed core autism symptoms
4. findings were reported in English.

Of the 2023 abstracts reviewed, 325 papers were examined to determine if full inclusion criteria were met. Sixteen articles met full criteria and critical appraisals were completed. All aspects of the systematic review were completed by two independent raters with a third rater utilized for disagreement (figure 4.1 reports study selection flow chart).

4.3.3 Research Question:

The research question addressed was: “What is the effectiveness of early intervention on ameliorating the clinical symptoms of autism for those with or at risk for a diagnosis of ASD?”
Figure 4.1: Systematic review flow chart

Initial Database Search
2023 abstracts

Included 325 papers for full text review

Excluded abstracts 1698

Critical appraisals
16 papers

Not written in English
7

Not addressing ASD or outcome of interest
584

No outcome measure
41

No intervention
617

Participants too old
190

Issues in autism only
260
4.3.3 Data Extraction

Details of study methodology, sample, and PEDro/CASP scores were summarized for all studies in Table 4.1 by intervention type. Methodological quality of the systematic review and review article were appraised using the Critical Appraisal Skills Programme (CASP) systematic review tool (Public Health Resource Unit, England, 2006). All other studies were appraised using the Physiotherapy Evidence Database (PEDro) scale (Centre for Evidence-Based Physiotherapy, 1999). These scales rate evidence quality by assessing the internal and external validity of a study.
### Table 4.1: Intervention Details

**Applied Behavior Analysis (ABA)**

<table>
<thead>
<tr>
<th>Study</th>
<th>N</th>
<th>Age range in months</th>
<th>Length and intensity of intervention</th>
<th>Environment</th>
<th>Assessments</th>
<th>Outcomes of interest</th>
<th>Follow up assessment</th>
<th>PEDro Score</th>
</tr>
</thead>
<tbody>
<tr>
<td>Ben-Itzchak &amp; Feinfield, 2003</td>
<td>25</td>
<td>20-32</td>
<td>12 months; 35 hours/week</td>
<td>Center</td>
<td>Autism Diagnostic Observation Schedule (ADOS), Autism Diagnostic Interview-revised (ADI-R), Bayley Scales of Infant Development, Second Edition (BSID-II) (Mental Scale), Stanford-Binet Intelligence Scale, Fourth Edition (SB, FE), Developmental domains</td>
<td>Imitation, receptive/expresive language, non verbal communication, play skills, stereotyped behaviors, autism severity, cognitive functioning</td>
<td>None</td>
<td>5/10</td>
</tr>
</tbody>
</table>

*Continued*
<table>
<thead>
<tr>
<th>Study</th>
<th>Participants</th>
<th>Age (months)</th>
<th>Time (hours/week)</th>
<th>Setting</th>
<th>Measures</th>
<th>Outcomes</th>
<th>Compliance</th>
</tr>
</thead>
<tbody>
<tr>
<td>Weiss, 1999</td>
<td>20</td>
<td>20-65</td>
<td>24 months; 40 hours/week</td>
<td>Home</td>
<td>CARS, Vineland Adaptive Behavior Scale (VABS), Developmental domains</td>
<td>Autism severity, adaptive behavior, rate of skill acquisition, curricular goals</td>
<td>None</td>
</tr>
</tbody>
</table>

Continued
Table 4.1 Continued

| Green et al., 2002 | 1 | 14 | 39 months; 25-33 hours/week | Home/school/community | Weschsler Primary and Preschool Scale of Intelligence-Revised (WPPSI-R), Rosetti Infant-Toddler Language Scale (RITLS), Preschool Language Scale (PLS), Peabody Picture Vocabulary Test-Revised (PPVT-R), Expressive One-Word Picture Vocabulary Test (EOWPVT), Clinical Evaluation of Language Functions (CELF), VABS, Cognitive functioning, language, motor skills, adaptive behavior, eye contact, social skills, play, and joint attention | 1 year post-intervention | 3/10 |

Continued
Table 4.1 Continued

<table>
<thead>
<tr>
<th>Study</th>
<th>N</th>
<th>Age range</th>
<th>Length and intensity of intervention</th>
<th>Environment</th>
<th>Assessment tools</th>
<th>Outcomes of interest</th>
<th>Follow up assessment</th>
<th>Pedro Score</th>
</tr>
</thead>
<tbody>
<tr>
<td>Boulware et al., 2006</td>
<td>8</td>
<td>18-29</td>
<td>9-21 months; 1.5 hrs 2X/week integrated playgroup, 2 hrs 3X/week individual instruction, 2hrs/week staff support in community/home and 5 hrs/week support from family</td>
<td>Center</td>
<td>BSID-II (Mental Scale), Temperament and Atypical Behavior Scale (TABS), Communication and Symbolic Behavior Scales (CSBS), Functional developmental outcomes</td>
<td>Cognitive functioning, temperament, communication, social skills, functional outcomes (speech, instruction following, motor imitation, toilet training, play, peer interaction and imitation)</td>
<td>School placement assessment in first grade</td>
<td>3/10</td>
</tr>
</tbody>
</table>
### Table 4.1 Continued

**Early Start Denver Model**

<table>
<thead>
<tr>
<th>Study</th>
<th>N</th>
<th>Age range in months</th>
<th>Length and intensity of intervention</th>
<th>Environment</th>
<th>Assessments</th>
<th>Outcomes of interest</th>
<th>Follow up assessment</th>
<th>Pedro Score</th>
</tr>
</thead>
<tbody>
<tr>
<td>Dawson, et al., 2010</td>
<td>45 n= 24 (ESD M) n= 21(control)</td>
<td>18-30</td>
<td>24 months; 20 hours/week from clinician, 5 hours/week from parent</td>
<td>Community, Center and Home</td>
<td>ADI-R, ADOS, Mullen Scales of Early Learning (MSEL) (fine motor, visual reception, expressive and receptive language), VABS, Repetitive Behavior Scale (RBS)</td>
<td>Adaptive behavior, receptive/expersive language, motor, socialization, autism severity, and repetitive behaviors</td>
<td>2, 4, 8 and 12 weeks post-intervention</td>
<td>7/10</td>
</tr>
<tr>
<td>Vismara et al., 2009</td>
<td>6</td>
<td>10-36</td>
<td>12 weeks; 1 hour/week</td>
<td>Community, Center and Home</td>
<td>MSEL, ADOS, ESDM curriculum checklist, ESDM fidelity scale, frequency of spontaneous functional verbal utterances, number of imitative behaviors, Child Behavior Rating Scale (CBRS)</td>
<td>Level of parent mastery, child social communication and imitative behavior during play with parents, therapist, and unfamiliar adult</td>
<td>2, 4, 8, and 12 weeks post-intervention</td>
<td>3/10</td>
</tr>
</tbody>
</table>
### Table 4.1 Continued

<table>
<thead>
<tr>
<th>Study</th>
<th>N</th>
<th>Age range in months</th>
<th>Length and intensity of intervention</th>
<th>Environment</th>
<th>Assessments</th>
<th>Outcomes of interest</th>
<th>Follow up assessment</th>
<th>Pedro Score</th>
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</thead>
<tbody>
<tr>
<td>Pedro et al., 2008</td>
<td>19</td>
<td>9</td>
<td>9 months; 12 weekly sessions, 1.5 hours each</td>
<td>Center and home</td>
<td>MSEL, ADOS, ESDM fidelity scale, frequency of spontaneous functional verbal utterances, number of imitative behaviors, Child Behavior Rating Scale (CBRS)</td>
<td>Level of parent mastery, child cognitive functioning, language, social and communication, imitative behavior</td>
<td>6 months post intervention</td>
<td>2/10</td>
</tr>
<tr>
<td>Katagiri et al., 2010</td>
<td>16</td>
<td>22-41.4</td>
<td>1 session; 7 minutes</td>
<td>Center</td>
<td>Frequency of: gazing at experimenters face, giving positive social emotional signals to experimenter, offering toys to the experimenter and requesting the experimenter to imitate his/her own actions</td>
<td>Imitation</td>
<td>None</td>
<td>3/10</td>
</tr>
<tr>
<td>Study</td>
<td>N</td>
<td>Age range in months</td>
<td>Length and intensity of intervention</td>
<td>Environment</td>
<td>Assessments</td>
<td>Outcomes of interest</td>
<td>Follow up assessment</td>
<td>Pedro Score</td>
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<tr>
<td>Wong &amp; Kwan, 2010</td>
<td>17 n= 9 (intervention) n= 8 control</td>
<td>17-36</td>
<td>2 weeks; 5 days/week for 30 minutes</td>
<td>Home</td>
<td>ADOS, Ritvo-Freeman Real Life Rating Scale, Symbolic Play Test, Parenting Stress Index</td>
<td>Communication skills, social interaction, parent rating of child’s skills, symbolic play, parent stress</td>
<td>None</td>
<td>7/10</td>
</tr>
<tr>
<td>Study</td>
<td>Sample Size</td>
<td>Age at Assessment</td>
<td>Measures</td>
<td>Joint Attention and Joint Action Routines</td>
<td>Ratings</td>
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<tr>
<td>Drew et al, 2002</td>
<td>N=24, n=12 parent, n=12 control</td>
<td>Mean age =23 months</td>
<td>Condensed version of Checklist for Autism in Toddlers (CHAT), ADI-R, MacArthur Communicative Development Inventory (CDI), Griffiths Scale of Infant Development (Non-Verbal IQ), Parental Stress Inventory (PSI), activity checklist</td>
<td>None</td>
<td>5/10</td>
<td></td>
<td></td>
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<tr>
<td>Study</td>
<td>Duration</td>
<td>Setting</td>
<td>Pre Intervention</td>
<td>Post Intervention</td>
<td>Outcome Measures</td>
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<tr>
<td>Schertz &amp; Odom, 2007</td>
<td>3 months</td>
<td>Home</td>
<td>Modified Checklist for Autism in Toddlers (M-CHAT), Pervasive Developmental Disorders Screening Test-II (PDD-ST-II), Infant Social-Communication Questionnaire (ISCQ), CARS, Developmental Programming for Infants and Toddlers (DPIT), Hawaii Early Learning Profile (HELP)</td>
<td>Focusing on faces, turn taking, responding and initiating joint attention, and social validity (parent questionnaire)</td>
<td>None</td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

Continued
| Newcomer, 2010 | 3 | 20-27 | 6-10 sessions, 1 hour each | Home | Frequency of parent teaching episodes, communication attempts, facial orientation, coordinated joint attention attempts. | Parent: learning of techniques, responsiveness to intervention, vocalization expansions; Child: eye contact, facial orientation, communicative attending, joint attention, | None | 3/10 |
Table 4.1 Continued

<table>
<thead>
<tr>
<th>Study</th>
<th># articles</th>
<th>Period of review</th>
<th>Age range</th>
<th>Population</th>
<th>Functional Emotional Assessment Scale (FEAS) ratings – self regulation and interest in the world, forming relationships and attachment and engagement, communication, behavioral organization, problem solving, representational capacity and representational differentiation.</th>
<th>CASP systematic review tool</th>
</tr>
</thead>
<tbody>
<tr>
<td>Solomon et al., 2007</td>
<td>68</td>
<td>18-72</td>
<td>1 year; 15 hours/week</td>
<td>Home</td>
<td>Functional Emotional Assessment Scale (FEAS), Clinical rating on Greenspan’s functional developmental levels, Client satisfaction survey.</td>
<td>None</td>
</tr>
</tbody>
</table>

Review Articles

<table>
<thead>
<tr>
<th>Study</th>
<th># articles</th>
<th>Period of review</th>
<th>Age range</th>
<th>Population</th>
<th>CASP systematic review tool</th>
</tr>
</thead>
</table>
4.4 Results

4.4.1 Study Participants

Prospective data were collected from 16 studies targeting 241 children with/at-risk for ASD. The ages of the children differed across studies, ranging from 9 to 72 months.

4.4.2 Description of intervention

The early intervention programs fit into 5 categories: early intensive behavioral intervention (EIBI), Early Start Denver Model (ESDM), imitation intervention, relationship-based intervention, and blended approach (see Table 4.1).

EIBI is defined as intensive applied behavioral analysis techniques in a one-on-one setting with intervention usually lasting at least two years {{475 Anderson, S.R. 1996}}. The EIBI studies implemented a comprehensive intervention program with a detailed set of formal and informal developmental assessments. Methodological rigor ranged from low to moderate, with variation in sample sizes, program duration, age ranges, intervention environments, assessments and outcome measures.

EIBI studies reported improvements in cognition (IQ improved up to 20 points), adaptive behavior, language, communication, and socialization. All studies indicated a reduction in autism severity and/or improved diagnostic categorization, including elimination of an ASD diagnosis {{478 Weiss, M.J. 1999; 477 476 Ben-Itzchak, E. 2007; 479 Green, G. 2002}}. Additionally, cognitive level and social abilities influenced rate of
skill acquisition {{476 Ben-Itzchak, E. 2007}} and overall level of improvement {{477 Perry, A. 2008}}. At two year follow up, Weiss (1999) reported that 35% of the children were placed in full-time, regular education classrooms without support, 15% were in regular education classrooms with minimal support, 25% were placed in regular education classroom with 1:1 discrete trial training for part of the day and a full time aide, and 25% were placed in special education.

One study utilized a blended intervention approach combining applied behavior analysis (ABA), integrated playgroups, service coordination, and family education/training{{480 Boulware, G.L. 2006}}. Improvements were documented in cognition, self-regulatory behavior, communication skills, and functional outcomes, including spontaneous language, following simple and complex directions, imitation skills, toilet training, interaction with peers, and play skills. At the follow-up assessment when the students were in first grade, 57% were in general education classrooms and 29% of children were in segregated special education classrooms. Fifty percent of these parents reported that their children had meaningful and reciprocal peer relationships both at school and in the community.

The ESDM program combines relationship-based intervention with Pivotal Response Training (based on the principles of ABA) {{481 Vismara, L.A. 2009}}. These studies followed the progression of intervention development from a case-study to a
cohort design and finally a randomized controlled trial (RCT). All of these studies included follow-up assessments.

Following ESDM intervention, children improved in language, communication, socialization, adaptive behavior, cognition (IQ increase of 17.6 points) play, visual motor, and imitation. Frequency of aggressive behaviors decreased, severity of autism decreased and diagnostic status improved. Additionally, parents achieved a high level of mastery for the ESDM. Follow-up assessments indicated continued improvements in language, imitation, level of attentiveness and social interactions.

The two imitation-based intervention studies {{Katagiri, M. 2010; Sanefuji, W. 2009}} varied significantly in duration, sample size, and intervention techniques. Toddlers made improvements in social attention, social emotional behaviors, joint attention, levels of engagement, and mother-child interactions.

Relationship based interventions focus on improving socialization skills and developing reciprocal relationships that are internally motivating {{Wong, V.C.N. 2010; Drew, A. 2002; Newcomer, A.L. 2010; Schertz, H.H. 2007}}. The relationship based intervention studies varied dramatically in sample size, duration, and intervention settings. Children who participated in relationship based interventions improved in communication, joint attention, vocalizations directed toward others, social interactions, eye contact, requesting, pointing, symbolic play, functional developmental
outcomes, and parent-child interactions. Additionally, parent’s stress level was reduced.

4.4.3 Types of outcome measures

The studies used a variety of standardized and non-standardized assessments utilized across the studies. For purposes of this review, outcomes were grouped into categories of social participation, communication and language, cognition, adaptive behavior/developmental domains. Table 4.2 describes the specific skills within each outcome category in which the children exhibited improvement.
Table 4.2: The outcomes targeted in interventions and specific skills that improved within each outcome category

<table>
<thead>
<tr>
<th>Social participation</th>
<th>Communication and language</th>
<th>Cognition</th>
<th>Adaptive behavior/ developmental domain</th>
<th>Parent education</th>
</tr>
</thead>
<tbody>
<tr>
<td>• eye contact</td>
<td>• receptive language</td>
<td>• IQ scores</td>
<td>• FEAS scores</td>
<td>• setting up learning opportunities</td>
</tr>
<tr>
<td>• social awareness</td>
<td>• expressive language</td>
<td>• mental age</td>
<td>• all areas of adaptive behavior</td>
<td>• arranging the environment</td>
</tr>
<tr>
<td>• reciprocal interactions</td>
<td>• joint attention</td>
<td>• developmental levels</td>
<td>• communication</td>
<td>• promoting social engagement</td>
</tr>
<tr>
<td>• play skills</td>
<td></td>
<td></td>
<td>• daily living skills</td>
<td>• emotional sharing</td>
</tr>
<tr>
<td>• peer interactions</td>
<td></td>
<td></td>
<td>• socialization</td>
<td>• implementing positive feedback for socialization</td>
</tr>
<tr>
<td>• spontaneous social initiations</td>
<td></td>
<td></td>
<td>• motor skills</td>
<td></td>
</tr>
<tr>
<td>• orienting to faces</td>
<td></td>
<td></td>
<td>• curriculum based assessments (mainly EIBI)</td>
<td></td>
</tr>
<tr>
<td>• turn taking</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>• joint attention</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>• attention during social situations</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>• preference for social play over isolated play</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>• mother-child bonding</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

129
4.5 Research Implications

The methodological rigor of the included studies varied considerably and should be taken into consideration when interpreting study results. Recurring themes regarding study design included: small sample size and lack of control group, randomization, follow up assessment, measure of skill maintenance and/or assessment of skill generalization.

Because the majority of the studies in this review did not have a control group few, if any conclusions about whether the developmental improvements were due to the intervention or maturation can be made. In addition, the duration of the interventions varied from one session to several years of intervention, limiting comparison across interventions. Comparison of interventions with similar durations was rarely possible due to the inconsistent assessment measures within and between studies. The majority of the studies either did not provide sufficient information to calculate effect size (ES) or used different pre and post measures within a study. Calculating the ES is beneficial in deciding which intervention had the strongest effect and the cost-benefit ratio, particularly when consistent measures are used. For example, as stated previously, duration of intervention varied dramatically. If the ES is similar for a 2 week intervention and a 6 month intervention, it would be more beneficial to implement the shorter intervention. ES calculations would be beneficial to clinicians and family members deciding which intervention model would be most appropriate for a specific child’s needs. Third, sample size varied from 1 to 68 subjects.
and was most often less than 10, limiting generalizability of outcomes. Finally, studies rarely included a follow-up measure to assess skill maintenance and generalization to other environments. Only The ESDM studies consistently included follow-up assessments. Maintenance and generalization of skills in children with/at-risk for autism is an important issue that needs to be addressed {494 McConnell, S.R. 2002}. The studies included in this review need to be replicated using stronger, more rigorous methodological designs that include measures with known validity and follow-up assessments to demonstrate the long term effect of EI.

4.5 Clinical Implications

While the studies in this review implemented different intervention programs, several common themes emerged addressing topics relevant to the field of occupational therapy. First, the outcomes targeted in the interventions included social participation, communication and language, cognition, imitation, and adaptive behavior. These outcomes are commonly targeted in occupational therapy interventions, are addressed in the Occupational Therapy Practice Framework {AOTA, 2008} and effect performance in areas of occupation. Additionally, all interventions were individualized to the participant, were play-based, and had a strong focus on parent education similar to occupational therapy intervention.

Play is an important occupation of a child and occupational therapists often use play-based techniques in therapy. Play is As a motivating, purposeful activity, play is the
context for much of the child’s learning and development. Infants and toddlers use play to develop roles and problem solving skills, build relationships, and to learn about the environment and social rules. Research shows there are many benefit of play including language development, socialization, and cognition (Main, 1983, Borgen; ervtin-trip; kasari 2000, kasari 2006). The majority of the relationship-based interventions, the ESDM, and the blended intervention approach applied play-based strategies in their intervention program.

Family-centered occupational therapy interventions include {{497 Turnbull, A.P. 1981; 496 Hanna, K. 2002}}. Parent education was the primary focus of several of the intervention programs and a secondary component of others in this review. Teaching parents how to (1) respond to their child’s communication techniques (2) expand communication attempts (3) give the child time to respond (4) reinforce positive behaviors and (5) seek and sustain eye contact improves developmental outcomes (Anderson, et al 2005, Engwal & McPherson, 2003, shields 2001). Educating parents also benefits parents, who report education increases their confidence in interacting with their child {{485 Newcomer, A.L. 2010}}.

Family-centered interventions focusing on parent-child relationship are considered best practice and should be a priority {{509 Prelock, P.A. 2003; 510 Beatson, J.E. 2002}}. The relationship between a child and his/her parent has a huge impact on development {{511 Wehman, T. 1998}}. Teaching parents how to respond to their child
and to facilitate reciprocal interactions fosters the child’s development of foundational social skills.

**4.6 Conclusion**

Our review of the evidence available for early intervention programs beginning prior to two years of age found that the ESDM has the highest level of evidence is the ESDM. The ESDM combines (1) relationship based intervention focused on language development, social functioning, and development of non-verbal communication and imitation as the foundation for verbal language with (2) Pivotal Response Training (PRT) which is based on the principles of ABA. PRT incorporates motivation with systematic teaching approaches to increase play skills, language, and communication in a natural setting. The ESDM model is presented in three research studies showing the effectiveness of the intervention program in children under 2 years of age. The methodological rigor progresses from a case study to a RCT. Additionally, toddlers receiving the ESDM intervention showed improvements in core ASD deficits including language and communication, socialization, adaptive behavior, cognition, play, and imitation skills. There was also a reduction of aggressive behaviors as well as improved and/or elimination of diagnostic status following intervention. Finally, the ESDM documented follow-up assessments for each study reporting continued improvement after the intervention ended.
4.7 Limitations

Most systematic reviews include only studies with high methodological rigor; however, in this review we included all studies that implemented an intervention program for children under the age of two years who were with/at-risk for ASD. Due to the limited number of intervention studies for this population, we thought our review would provide a more comprehensive picture of the efficacy of interventions for this population if it was broader in nature. Therefore interpretation of results must consider the range of scientific rigor as rated by the PEDro scale varied from 2/10-7/10. In addition, few studies reported effect size and in most sample size was limited.

4.8 Recommendations for future research

The studies included in this review all report some level of improvement in core ASD deficits for young children with or at-risk for a diagnosis of ASD. Research studies with higher levels of scientific rigor are needed to help guide therapists and families as to which EI program has the highest efficacy. Consistent, standardized assessment tools to evaluate developmental outcomes are needed so direct comparison of interventions is possible.
Chapter 5: Conclusion and Future Research

The main objective of this research was to examine sensory processing function and dysfunction in toddlers with and without early signs of autism during the second year of life. Sensory processing was examined using neurophysiologic, direct observation and parent report measures. Additionally, a systematic review was completed to determine if early intervention ameliorates the core deficits of ASD in children with or at risk for autism under 2 years of age.

5.1 Summary of findings:

Sensory processing function of toddlers with and without early signs of autism during the second year of life was examined using a multi-modal assessment protocol. The multi-modal assessment protocol consisted of two clinical assessments (direct observation and parent report) and a neurophysiologic assessment (ERP). Chapter 2 summarizes the results of the clinical protocol. As a whole, toddlers had the most difficulty processing sensory information in the auditory and the vestibular domains. Specifically, of the toddlers with atypical functioning in the auditory domain, 47% exhibited under-responsive behavioral patterns and 53% exhibited over-responsive patterns. In the vestibular domain, 86% of the toddlers with atypical functioning exhibited over-responsive behavioral patterns and 14% exhibited under-responsive
patterns. Additionally, sensory seeking was the most prominent atypical sensory processing behavior exhibited by this group. Two distinct patterns of SP behavior were identified. Toddlers displayed behaviors associated with either under-responsivity or over-responsivity. Further, toddlers displaying over-responsivity behavioral patterns also had more overall atypical sensory processing scores.

Toddlers with early signs of autism exhibited an over-responsive behavioral pattern, had the most difficulty processing information in the vestibular system (90% over-responsive and 10% under-responsive) and exhibited a higher amount of avoidance behaviors. The N-ES group exhibited an under-responsive behavioral pattern. Further, ERP findings indicated that there were subtle differences in the way toddlers with and without early signs of autism processed novel speech sounds. Specifically, toddlers in the ES group exhibited attenuated responses to novel speech sounds. Additionally, the N-ES group showed a tendency for more left side hemispheric activation in response to speech sounds while the ES group showed more right side activation. Finally, a regression analysis indicated that chronological age and language development were the best predictors of early signs of autism in young children aged 12-24 months.

The results from the systematic review in chapter 4 indicated that the studies utilizing the ESDM had the highest level of methodological rigor and reported improvements in core ASD deficits, including language and communication, socialization, adaptive behavior, cognition, play, and imitation skills. The majority of the
studies in this review had a small sample size, lacked a control group, and used a wide variety of assessment tools, making comparison of the interventions difficult.

5.2 Limitations

The limitations in this study include small sample size, measurement issues, and assessment of SP function at one time point. Both the clinical and ERP studies were limited by small sample size. Our results indicated SP function exhibited trends in the expected direction but were lacking in statistical power on all of the assessments. Small sample size influences how subtle or robust the results could be. Future studies should include a larger sample size, in particular when using ERP. In this study, we had to eliminate more than half of our subjects due to poor signal-to-noise ratio. Moving forward, the result of the ERP portion of this study would influence the following changes: two ERP data collection sessions, use of the iPad® during data collection to minimize movement, speech stimuli with less subtle differences, and over recruitment of subjects.

The ADEC was used to detect early signs of autism. This tool is newly developed with limited information on specificity and sensitivity for ASD diagnosis at a later age. Additionally, we used different cut-off scores than described in the ADEC manual. Without following this sample through to a diagnosis of autism, we don’t know if using the new cut-off scores was appropriate in detecting ASD. Hence, we are unable to draw conclusions at this point from our study about how many of the toddlers in the ES group
will actually develop autism and we cannot conclude as to the sensitivity or specificity of the ADEC using our cut-off scores.

Finally, our study examined sensory processing of toddlers with and without early signs of autism at one time point. Having only one time point for assessment limits our knowledge of whether or not the toddlers with early signs of ASD will actually go on to develop ASD. Additionally, multiple follow-up assessments would allow for examination of developmental progression of SP function and dysfunction.

5.3 Future Research

Recent advances in the field of autism have made it possible to identify signs and symptoms of ASD during the first two years of life (Boyd, Odom, Humphreys, & Sam, 2010). Identifying reliable early signs of ASD could lead to earlier diagnosis and timely access to early intervention programs. The data presented in this study indicate that differences in SP function can be assessed in a sample of toddlers with and without early signs of autism. Our results are supported by several retrospective and parent report studies identifying SP differences during the first and second year of life in children who go on to be diagnosed with an ASD (Baranek, 1999; Dahlgren & Gillberg, 1989; Gillberg et al., 1990; Lord, 1995; Osterling & Dawson, 1994). Detection of sensory processing deficits between 0-24 months of age has the potential to significantly impact the development of the disorder. Prospective research is needed examining SP function and
dysfunction in high-risk infants and infants and toddlers with and without early signs of ASD.

Additional examination of SP patterns of young children with and without early signs of autism during the first two years of life is needed. Our research indicated there were two clusters of sensory processing function, over-and under-responsiveness patterns, within a sample scoring in the typically developing range when group data were examined. Additionally, different patterns of SP were identified in toddlers with and without early signs of autism. Several studies have documented that SP patterns can be identified in older children with ASD (Baker, Lane, Angley, & Young, 2008; Baranek, Boyd, Poe, David, & Watson, 2007; Cheung & Siu, 2009; Lane et al., 2010), but little is known about the first two years of life. Identifying distinctive SP patterns at a young age could have etiological implications (Baranek et al., 2006). Further research could replicate and confirm the existence of under- and over-responsiveness SP patterns in this population. Our results indicated that toddlers exhibiting over-responsiveness patterns also exhibited more atypical SP behaviors than the under-responsive group. Additional longitudinal studies could determine how these patterns develop over time and if the toddlers exhibiting an over-responsive behavioral pattern went on to develop ASD, another developmental disorder, or if SP function improved with age leading to typical development. Additionally, identifying precise neurological abnormalities occurring simultaneously with the presence of behavioral SP pattern could inform the
mechanisms underlying SP dysfunction and support the implementation of sensory-based interventions to ameliorate core ASD deficits.

Research studies utilizing a combination of clinical and neurophysiologic assessment tools are needed. This study was the first to our knowledge to utilize a multimodal SP assessment in young children with early signs of autism. Our study provides preliminary evidence that results from clinical SP assessments align with results from neurophysiologic assessments. Specifically, the ES group exhibited attenuated responses to speech sounds on ERP along with over-responsive behavioral patterns, increased display of atypical SP behaviors and increased avoidance behaviors on clinical assessments. As discussed previously, the majority of the sensory processing studies involving young children utilized a single assessment type (i.e. parent report, direct observation or neurophysiologic techniques). Using a multimodal assessment protocol provides an in-depth look at sensory processing function. Additionally, combining parent report, direct observation, and neurophysiologic measures, allows for us to link observed behaviors with the neurological processes occurring simultaneously. Without neurophysiologic measures, we can only speculate about what is happening on a neurological level.

Longitudinal research studies are needed to better understand the developmental progression of sensory processing function and dysfunction during the first two years of life. Longitudinal studies can help determine the relationship between the presence of sensory processing deficits during the first year of life and ASD
characteristics evident at three years of age and later. Additionally longitudinal studies can explain how SP deficits progress over time. There is no consensus as to whether SP deficits diminish (Baranek, Foster, & Berkson, 1997; Baranek et al., 1997; Cheung & Siu, 2009), increase (Talay-Ongan & Wood, 2000), or stay the same with age (Rogers et al., 2003). Finally, employing the same assessments at various points in development will identify how sensory processing changes as a function of age. Understanding this information could lead to implementation of more sensitive screening and/or diagnostic tools and individualized early intervention strategies for children with ASD.

Additionally, focused studies examining the relationship between early sensory processing dysfunction and the development of core autism features are needed. Research involving older children with ASD have linked SP dysfunction with the presence of stereotypic behaviors (South, Ozonoff, & McMahon, 2005; Turner, 1999). The research supporting the presence of stereotypic behaviors and sensory fascinations during the first two years of life is minimal (Bryson et al., 2007; Loh et al., 2007; Mooney, Gray, & Tonge, 2006). Additionally, there are few studies documenting early social and communication deficits involving aspects of SP function that link to a later diagnosis of ASD (Baranek, 1999; Osterling & Dawson, 1994; Wetherby et al., 2004; Wetherby & Woods, 2006).

Due to the increased interest in early identification of ASD, there have been several tools developed to identify early signs of autism including SP assessments (Baird et al., 2000; Baranek et al., 2008; DeGangi & Greenspan, 1989). For example, the
Checklist for Autism in Toddlers (CHAT) has been used in the past to identify ASD at an early age. Baird and colleagues found that although the specificity was high (98%), the sensitivity was low (38%) (Baird et al., 2000). More studies are needed to document sensitivity and specificity of early autism detection tools.

Additionally, SP deficits can be identified during the first two years of life. Assessments to identify SP function at an early age are available (Baranek, Boyd, Poe, David, & Watson, 2007; DeGangi et al., 1988; DeGangi & Greenspan, 1989). Two measures of SP function for infants and toddlers are mentioned in the literature: SPA and Test of Sensory Function in Infants (TSFI). The TSFI targets infants 0-18 months of age. It is an older test, developed in 1989. The SPA was utilized in this study. Our results indicate the avoidance subscale as being the most sensitive to SP deficits. This is a new test still under development with limited sensitivity and specificity information available. As SP function and dysfunction during the first two years of life is better understood, more sensitive SP assessment tools should be developed.

SP deficits could eventually be salient indicators of early signs of ASD, help tailor individualized intervention programs, and determine the most effective intervention strategies to implement and at what age to begin treatment (Baranek, 1999). Further, if started at a young age, early intervention has the potential to minimize the core symptoms of ASD and decrease the severity of the disorder (Rogers & Vismara, 2008).

The ultimate goal of research relating to early detection of SP deficits and early signs of autism is to inform intervention. Early intervention programs have the potential
to ameliorate core ASD symptoms, lessen the severity of the disorder, and/or improve overall quality of life. There is no consensus in the literature as to which early intervention program is the most effective for children under two years of age. There is consensus that EI improves core deficits of ASD (Birkin et al., 2008; Boyd et al., 2010; Dawson et al., 2010; Green et al., 2002) and has the potential to change the developmental trajectory of the disorder (Dawson, 2008). The majority of the research studies available involving children with or at risk for ASD have many limitations, including small sample size, lack of control group, lack of follow up assessments, lack of randomization, and lack of measure for skill maintenance and generalization. Additional intervention studies are needed for young children using higher levels of scientific rigor to help guide therapists and families as to which EI program has the highest efficacy. Further, consistent, standardized assessment tools should be utilized allowing for comparison of intervention.

5.4 Conclusion

Sensory processing function and dysfunction in young children with and without early signs of ASD needs to be better understood. Retrospective and parent report research has indicated that sensory processing deficits can be detected in the first two years of life in children who go on to be diagnosed with an ASD. Prospective studies utilizing a multi-modal assessment protocol are needed to identify the developmental progression of SP dysfunction. Additionally, the relationship between early SP dysfunction and later ASD diagnosis needs to be defined. Combining parent report,
direct observation and neurophysiologic measures of SP will provide a detailed understanding as to what is occurring in relation to SP function and dysfunction.

Identifying SP dysfunction during infancy can lead to early intervention. Early intervention has the potential to significantly improve sensory processing deficits and minimize ASD characteristics. In order to move toward addressing sensory processing in an early intervention program, sensory processing function during the first and second year of life must be better understood.

**5.5 Clinical Bottom Line**

In conclusion, our study provides preliminary evidence indicating toddlers with and without ES of autism process sensations differently. Specifically, the ES group exhibit over-responsive behavioral patterns of SP, increased atypical SP behaviors, increased avoiding behaviors and attenuated responses to speech sounds. Toddlers in the N-ES group exhibited under-responsive behavioral patterns. Our data supports the notion that SP deficits can be detected during the second year of life and have the potential to be useful in early detection of ASD.
References


Centre for Evidence-Based Physiotherapy. (1999). *PEDro scale*


Chicago: SPSS, I. (2011). *SPSS for windows*


Appendix A: Recruitment message for website or email attachment
Young Children Needed

To participate in a research study conducted by Ohio State University:
“How Do Children With and Without Early Signs of Autism Respond to Sensory Stimuli?”

What is this study about?
This study will evaluate how children with and without early signs of autism respond to sensory stimuli in their environment.

Who can be involved in this study?
Children between the ages of 12-24 months of age with typical hearing

What will happen during this research study?
We will observe and play with your child for two visits, each lasting approximately 1-2 hours.

During these visits we will:
1. Test your child’s play skills, social interaction, movement skills, and how your child responds to sensations within their environment (such as the sound and sight of a rattle or reaction to a light up toy). These assessments will be videotaped for scoring purposes only.

2. Record your child’s response to speech sounds using heart rate monitors and an EEG to measure brain activity. An EEG with children is a common, non-invasive tool that causes no pain. A hat made of electrodes will be placed on your child’s head, a process that only takes approximately 1-2 minutes. Your child could become fussy during the test. If he/she does, we can take a short break to help him/her calm down and/or stop the test early if needed.

Where will I have to go?
This study will be conducted at The Ohio State University as a collaborative effort between the divisions of Occupational Therapy and Developmental Psychology. Parking for these visits will be compensated and incentives will be rewarded for participation.

To learn more about this research project, please contact:
Karen Harpster, OTR/L at harpster.17@osu.edu or 614-292-0204. This research is being conducted under the Principal Investigator Dr. Alison Lane, PhD, OTR/L, lane.350@osu.edu
Appendix B: Recruitment message for newsletter
Research is being conducted as a collaborative effort between the divisions of Occupational Therapy and Developmental Psychology at the Ohio State University. The purpose of this study is to describe how young children with and without early signs of autism process sensory information within the second year of life. We are currently recruiting children between the ages of 12-24 months to participate. The children will be tested on general development (including play skills, social development, and movement skills) and sensory processing. In addition, sensory processing will be measured using heart rate and an electroencephalogram (EEG). Two separate sessions will be conducted at The Ohio State University and will take approximately 1-2 hours each. If you know of anyone who might be interested, please give them our contact information listed below so we can tell them more about the study. In addition, feel free to contact us if you are interested in hanging up a flier or distributing information regarding the study to your clients and we will forward you the materials. Thank you!

Contact information: email: Karen Harpster, OTR/L at harpster.17@osu.edu, Dr. Alison Lane, PhD at OTR/L, lane.350@osu.edu or telephone: 614-292-0204.
Appendix C: Telephone screening tool
Parent name: ____________________________________________________________

Child’s name: __________________________________________________________

Child’s Date of Birth __________________________________________________

Address: ______________________________________________________________

Phone number: __________________________________________________________

Email address: __________________________________________________________

Was baby born full term? How many weeks? ________________________________

Any complications? ______________________________________________________

Did your child get a newborn hearing screening? ____________________________

Did he/she pass the hearing screening? ___________________________________

Is your child diagnosed with any conditions? ______________________________

If so, what condition? ___________________________________________________

To your knowledge, did your child reach his/her milestones on time? __________

Do you have any other children? __________________________________________

Are they older or younger? ______________________________________________

Is your child diagnosed on the autism spectrum? ___________________________

If yes, what tool was used to diagnose autism? _____________________________

By whom? __________________________________________________________________

Do you have any family members diagnosed with autism? _____________________

If so, what how are they related? __________________________________________
Appendix D: Consent Form
The Ohio State University Parental Permission
For Child’s Participation in Research

Study Title: Examining Sensory Processing in Young Children With and Without Early Signs of Autism During the Second Year of Life

Researcher: Dr. Alison Lane

Sponsor: Alumni Grants for Graduate Research and Scholarship through the Graduate School at OSU

This is a parental permission form for research participation. It contains important information about this study and what to expect if you permit your child to participate. If you permit your child to participate, you and your child will be considered participants. Your child will be asked to participate in approximately 2-3 hours of testing and you will be asked to complete a parent survey taking approximately 15-20 minutes to complete. Please consider the information carefully. Feel free to discuss the study with your friends and family and to ask questions before making your decision whether or not to permit your child to participate. If you permit you and your child to participate, you will be asked to sign this form and will receive a copy of the form.

Your child’s participation is voluntary.

Please consider the information carefully. Feel free to discuss the study with your friends and family and to ask questions before making your decision whether or not to permit your child to participate. If you permit your child to participate, you will be asked to sign this form and will receive a copy of the form.

Purpose: The purpose of this study is to describe how young children process sensory information during the second year of life. In particular, we will focus on how children process information that they see and hear. To do this, we will be using three clinical assessments: the Infant/Toddler Sensory Profile (ITSP), the Test of Sensory Functions in Infants (TSFI) and the Sensory Processing Assessment (SPA) and two neurophysiological assessments: Event Related Potentials (ERP) and heart rate (HR). ERP is a form of an EEG that measures how the brain respond to pictures and sounds. ERP is a commonly used with young children. It is a non-invasive procedure, causing no pain. In addition, we will administer the Bayley Scales of Infant and Toddler Development-Third Edition to test your babies development and the Autism Detection in Early Childhood as a screening tool for early signs of autism.
**Procedures/Tasks:** If you and your child participate in this study, you will be asked to bring your child to two sessions at The Ohio State University.

- **Session 1:** will be held in Atwell Hall where we will complete four clinical assessments and you will be asked to fill out one parent survey taking approximately 15-20 minutes. The clinical assessments include 4 semi-structured, observational assessments where an occupational therapist will interact with your child in a playful manner. The clinical assessments will take approximately 1-2 hours to complete with breaks as needed. The clinical assessments will be videotaped for scoring purposes only.

- **Session 2:** will take place at the Center for Cognitive Development where two neurophysiological assessments will be administered (ERP and HR). ERP and HR are non-invasive assessments that will take approximately 1-1.5 hours to administer. During these assessments, we will observe how your child responds to sights and sounds by monitoring your child’s heart rate with heart rate monitors and brain activity using an electroencephalogram (EEG).
  - An EEG measures how the brain responds/detects different sights and sounds. Your child will be seeing various shapes on a screen and listening to different sounds (spoken words). Your child will be wearing a hat that has special sensors called electrodes that are connected to a computer by wires. The computer will record your child's brain activity as he/she looks at shapes or listens to sounds. The electrodes measure the brain’s electrical activity in response to the sights/sounds and allows us to examine how the brain processes visual and auditory stimuli.
  - Your child will also be wearing electrodes on his/her chest to monitor his/her heart rate. Heart rate measures a child’s sensory processing and attention. In our experiment, we will be able to determine how quickly your child hear/sees the shapes and/or words and how long he/she pays attention to them.
  - These are non invasive procedures that can be stopped at any time if you or your child feels uncomfortable or if your child becomes fussy. All research staff working with your child have received training for this project and will be supervised by Dr. Alison Lane and Dr. Vladimir Sloutsky as this is a collaborative effort between the divisions of Occupational Therapy and Developmental Psychology.

**Duration:** There will be two study visits, each visit will last approximately 1-2 hours. Your child can take breaks for naps, snack, etc. as needed. You or your child may leave the study at any time. If you or your child decides to stop participation in the study, there will be no penalty and neither you nor your child will lose any benefits to which you are otherwise entitled. Your decision will not affect your future relationship with The Ohio State University.
**Risks and Benefits:** There are only very minimal known risks associated with this procedure. For instance, some children may get bored, fussy and/or become restless. The procedure can be stopped at any point if you or your child does not want to participate. Some children may not want to wear the ERP hat or they may find the ERP hat to be uncomfortable or itchy. The ERP hat can be removed at any point if you have any concerns.

There is a small chance that the study might show your child has some trouble with his/her senses, or exhibit possible early signs of autism. If this does happen, we will contact you by phone to discuss this information with you. In addition, every parent/guardian who participates in this study will receive two copies of the results (one for you and one for your primary care physician) along with a letter explaining the study and assessments. Please keep in mind that these results are not meant to serve as a diagnosis. It is encouraged that you share this information with your primary care physician, but it is ultimately your decision to do so. If you choose to share this report with your primary care physician, it may be placed into your child’s pediatrician records. This report will reflect information specific to your child. In the case that your child did not complete all of the tests involved in this study, the tests that were completed will be scored.

You and your child’s participation in the study will allow us to analyze sensory processing function for young children with and without early signs of autism in the second year of life. In addition, it may help us to identify signs of autism at a younger age and therefore begin treatment earlier. The results of this study will be presented at conferences and submitted for publication to a peer reviewed journal. The name of you, your child or your family will never be reported or disclosed.

**Confidentiality:** Efforts will be made to keep your child’s study-related information confidential. However, there may be circumstances where this information must be released. For example, personal information regarding your child’s participation in this study may be disclosed if required by state law. Also, your child’s records may be reviewed by the following groups (as applicable to the research):

- Office for Human Research Protections or other federal, state, or international regulatory agencies;
- The Ohio State University Institutional Review Board or Office of Responsible Research Practices;
- The sponsor, if any, or agency (including the Food and Drug Administration for FDA-regulated research) supporting the study.
Incentives: At the completion of each visit, you will receive a gift card for participation in the study. Specifically, after the first visit is completed, you will receive a $10.00 gift card to Target and after the second visit, you will receive a $20.00 gift card to Target. The gift card will be paid at the time of the study visit regardless of whether or not you decide to complete the entire session. Additionally, you will be given a parking pass upon arrival to OSU on the day of the study to be used while the research takes place. The parking pass will be paid at the time of the study visit regardless of whether or not you decide to complete the entire session.

Participant Rights: You or your child may refuse to participate in this study without penalty or loss of benefits to which you are otherwise entitled. If you or your child is a student or employee at Ohio State, your decision will not affect your grades or employment status.

If you and your child choose to participate in the study, you may discontinue participation at any time without penalty or loss of benefits. By signing this form, you do not give up any personal legal rights your child may have as a participant in this study.

An Institutional Review Board responsible for human subjects research at The Ohio State University reviewed this research project and found it to be acceptable, according to applicable state and federal regulations and University policies designed to protect the rights and welfare of participants in research.

Contacts and Questions: For questions, concerns, or complaints about the study you may contact Dr. Alison Lane (Email: lane.350@osu.edu; Phone: 614-292-0204) or Karen Harpster (Email: harpster.17@osu.edu).

For questions about your child’s rights as a participant in this study or to discuss other study-related concerns or complaints with someone who is not part of the research team, you may contact Ms. Sandra Meadows in the Office of Responsible Research Practices at 1-800-678-6251.

If your child is injured as a result of participating in this study or for questions about a study-related injury, you may contact Dr. Alison Lane at 614-292-0204.

Signing the parental permission form
I have read (or someone has read to me) this form and I am aware that I am being asked to provide permission for my child to participate in a research study. I have had the opportunity to ask questions and have had them answered to my satisfaction. I voluntarily agree to permit my child to participate in this study.

I am not giving up any legal rights by signing this form. I will be given a copy of this form.

The research team would like to retain participant contact information in a data repository upon completion of this study for follow-up purposes. Specifically, we would keep your name, your child’s name, child’s date of birth, your address, telephone number, and email address to allow us to contact your for a potential follow up study or similar research opportunities.

☐ Yes, I give permission to add my information to the data repository for additional follow-up opportunities in the future.

☐ No, I do not wish to have my information added to the data repository for additional follow-up opportunities.

Printed name of subject

Printed name of person authorized to provide permission for subject

Signature of person authorized to provide permission for subject

Relationship to the subject

Date and time

Investigator/Research Staff

I have explained the research to the participant or his/her representative before requesting the signature(s) above. There are no blanks in this document. A copy of this form has been given to the participant or his/her representative.

Printed name of person obtaining consent

Signature of person obtaining consent

Date and time
Appendix E: Scoring guidelines for clinical assessments
### ITSP

<table>
<thead>
<tr>
<th></th>
<th>Less Than Others</th>
<th>Typical Performance</th>
<th>More Than Others</th>
<th>Definite Difference</th>
<th>Probable Difference</th>
<th>Typical Performance</th>
<th>Probable Difference</th>
<th>Definite Difference</th>
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<tr>
<td>Low Registration</td>
<td>N/A</td>
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<td>45-43</td>
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<td>Sensation Avoiding</td>
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<td>Auditory Processing</td>
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<td>47-44</td>
<td>43-35</td>
<td>34-31</td>
<td>30-10</td>
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<td>15-7</td>
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<td>Tactile Processing</td>
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<td>67-62</td>
<td>61-48</td>
<td>47-42</td>
<td>41-15</td>
<td></td>
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<tr>
<td>Vestibular Processing</td>
<td>30-27</td>
<td>26-24</td>
<td>23-18</td>
<td>17-15</td>
<td>14-6</td>
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<tr>
<td>Oral Sensory Processing</td>
<td>N/A</td>
<td>35-32</td>
<td>31-23</td>
<td>22-19</td>
<td>18-7</td>
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<td></td>
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<tr>
<td></td>
<td>N/A</td>
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<td>32-24</td>
<td>23-20</td>
<td>19-7</td>
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### BSID-III

<table>
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<tr>
<th></th>
<th>Above Average</th>
<th>Typical Range</th>
<th>At risk for delay</th>
<th>Delayed</th>
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<tr>
<td>Scaled Score</td>
<td>&gt;9</td>
<td>12-8</td>
<td>7-5</td>
<td>&lt;4</td>
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</table>

### ADEC

<table>
<thead>
<tr>
<th></th>
<th>Low Risk</th>
<th>Moderate Risk</th>
<th>High Risk</th>
<th>Very High Risk</th>
</tr>
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<tr>
<td>Total Score</td>
<td>0-5</td>
<td>6-13</td>
<td>14-19</td>
<td>&gt;19</td>
</tr>
</tbody>
</table>

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Appendix F: Mat Lab analysis
Following Net Station signal processing, additional signal processing was completed using Mat Lab software attempting to further improve the signal to noise ratio, particularly at the baseline. Specifically, pre-processed data from Net Station was further processed in MatLab. Since data was being re-analyzed under a different algorithm, all subjects were included in this analysis, not just the subset remaining after Net Station analysis. MatLab relabeled segments as bad (ignoring NetStations labeling of bad segments) using the new algorithm. Subjects were eliminated from the data set if they had less than 50% of the deviant stimuli remaining following signal processing. The final data set included: ES, n=15; N-ES, n=14. Statistical analysis for this data set was performed using Mat Lab software.

**Visual inspection:** Grand average waveforms for standard and deviant stimuli for 8 electrode sites for both groups are presented in figures 1 and 2. The difference between standard and deviant waveforms within 100-300 ms, where we would expect to see a MMN, are very similar between the groups. Additionally, the difference waves for the two groups are very similar and hover around 0 mV throughout the entire waveform indicating minimal to no difference between standard and deviant stimuli (see figure 3 and 4).
Figure 1: N-ES Deviant and Standard waveforms
Figure 2: ES Standard and Deviant
Figure 3: Difference wave for ES
Figure 4: Difference waves for N-ES
Additionally, between-group, visual analysis of activation patterns for each stimulus type (standard and deviant) were compared at 170 ms post stimulus onset. Visual inspection indicates that the N-ES group has a slightly stronger activation pattern than the ES group for the deviant stimuli in bilateral temporal areas of the brain (Figure 5). Additionally, individual activation patterns were analyzed at 170 ms post stimulus onset by stimulus type (Figures 6-9). When examining individual activation patterns for subjects within each group, there is a tremendous amount of variability between subjects within the groups.
Figure 5: Activation patterns of N-ES and ES for Standard and Deviant stimuli at 170 ms post
Figure 6: N-ES individual activation patterns for deviant stimuli at 160 ms
Figure 7: N-ES activation patterns for standard stimuli at 160 ms
Figure 8: ES activation patterns for deviant stimuli at 160 ms
Figure 9: ES activation patterns for standard stimuli at 160 ms
Statistical Analysis: The only electrode site that exhibited statistical significant difference between the two groups was Cz (p=0.02). The difference at all other electrode sites were non-significant. See Table 1

Table 4: Area under the curve for deviant – standard stimuli between 150-250 for N-ES compared to ES

<table>
<thead>
<tr>
<th>Electrode sites</th>
<th>N-ES</th>
<th>ES</th>
<th>p values</th>
</tr>
</thead>
<tbody>
<tr>
<td>T3</td>
<td>3.77</td>
<td>12.84</td>
<td>0.61</td>
</tr>
<tr>
<td>T4</td>
<td>4.62</td>
<td>16.24</td>
<td>0.19</td>
</tr>
<tr>
<td>T5</td>
<td>4.02</td>
<td>36.17</td>
<td>0.15</td>
</tr>
<tr>
<td>T6</td>
<td>18.51</td>
<td>29.84</td>
<td>0.63</td>
</tr>
<tr>
<td>FZ</td>
<td>15.79</td>
<td>11.48</td>
<td>0.73</td>
</tr>
<tr>
<td>CZ</td>
<td>8.43</td>
<td>16.79</td>
<td>0.02**</td>
</tr>
<tr>
<td>PZ</td>
<td>8.88</td>
<td>7.09</td>
<td>0.66</td>
</tr>
<tr>
<td>C3</td>
<td>18.38</td>
<td>7.60</td>
<td>0.51</td>
</tr>
</tbody>
</table>

** Statistical significant difference

Conclusion: Additional signal processing in Mat Lab was completed to further improve the signal to noise ratio. This second level of analysis was completed in an attempt to remove artifact remaining in the baseline ERP waveforms after signal filtering with Net Station. Mat Lab processing eliminated any effect that was observed in the Net Station data. According to the Mat Lab data, there are no differences in the way toddlers with and without early signs of autism detect changes in speech sounds.
Appendix G: Summaries of articles in the systematic review
Two research questions were addressed in this paper:

1. Are pre-intervention cognitive, social interaction, and communication characteristics related to post-intervention developmental-behavioral outcome in young children with autism?

2. What developmental domains (imitation, receptive language, expressive language, play, non-verbal communication skills and stereotyped behavior) improve following intervention?

A center based Applied Behavior Analysis (ABA) was implemented for 35 hours per week for 1 year. Initial data analysis examined the differences in pre/post intervention scores on six developmental-behavioral (DB) domains on the entire group (N=25). Findings indicated a significant time effect (p<0.001) for all six developmental-behavioral domains. Additionally, there was a significant change (p<0.001) in IQ scores, increasing by an average of 17.3 points.

Next, groups were divided on several pre-intervention factors

1. IQ [high (HIQ) vs. low (LIQ)]

2. Severity of autism measured by

   a) Communication [high (HC) vs. low (LC)]

   b) Social interaction [high (HS) vs. low (LS)]
Results indicated that both the HIQ and LIQ groups made significant improvements (p<0.001) between pre/post intervention scores on all DB domain. However, the effect was greater for the HIQ group in receptive language ($n^2=0.960$ vs 0.830) and play domains ($n^2=0.816$ vs. 0.668). The effect was greater for the LIQ group on the imitation domain ($n^2=0.920$ vs. 0.853). When evaluating the effect of pre-intervention autism severity, groups were divided first into HS vs. LS group, then into HC vs. LC group, according to scores on the Autism Diagnostic Observation Scale (ADOS). When comparing DB scores post-intervention for the LS and HS groups, receptive language was the only domain showing a significant effect, although expressive language was trending in the right direction. There were no differences when comparing LS and HS groups on post intervention IQ scores. There were no significant differences between HC and LC group on post intervention DB scores or IQ.

Overall, these researchers controlled variables including age at start of therapy and type and intensity of intervention. Their main questions focused on how much impact pre-intervention cognitive, social, and communication characteristics had on the developmental outcomes of young children with ASD. Children with higher initial cognitive levels and children with fewer social interaction deficits exhibited better acquisition of developmental skills, especially in the areas of receptive language, expressive language, and play skills. More specifically, pre-treatment cognitive and social abilities related closely with progress made in receptive language. Higher social abilities were closely related to progress in expressive language and higher cognitive
abilities was closely related to progress in play skills. Severity of communication deficit did not impact outcomes in any developmental domain.

Strengths of this article include:

1) Groups were similar at the start of the intervention on age of diagnosis, evaluation process, lack of co-morbidities

2) Intervention was consistent among participants

3) Compared to other studies in this review, number of subjects is higher

4) The raters of the DB domains were blind to the predictor variables

The limitations include:

1) There was no control group

2) Progress on DB domains was scored based on child’s daily recorded progress charts rather than a prospective assessment

3) There was no follow up assessment or measure of generalization.

**Green et al., 2002** –

This case study lasted three years and 3 months. The intervention program progressed from an intensive 1:1, home based intervention program to a less intensive intervention program occurring in a variety of natural settings. Formal, home-based intervention was implemented by an ABA therapist for 25-33 hours per week for the first year. The intensity of the intervention increased to 30-36 hours per week, occurring at home and in the early intervention classroom during the second year. Additionally, the subject’s mother and family members provided 3-8 hours of instruction
per week. In pre-kindergarten she was initially supported by her home ABA therapist for the entire day. Support slowly faded over time to eventually not needing support. At 5 years, 5 months, she enrolled in a regular, public kindergarten class with no diagnosis, no IEP, and no ongoing special instruction needed.

Year 1 Results: At the start of the intervention, the subject had no imitation skills. Sixty days into intervention, she imitated 48 different one-step actions with no prompting. She showed similar rates of development for receptive and expressive labeling of objects. After 6 months of treatment, performance on the Leiter International Performance Scales indicated that her nonverbal cognitive skills were age appropriate. Additionally, the Vineland Adaptive Behavior Scale (VABS) showed gains on age equivalents of over a year on the test of receptive and expressive language and communication domains; 8 months on socialization domain. Smaller gains were shown on the daily living and motor domains which were age appropriate at start of intervention and not targeted during the first 6 months of treatment. Furthermore, the subject’s eye contact, social awareness, reciprocal interactions, play skills and general emotional tone improved during this first 6 months.

After a couple weeks of attending an early intervention classroom, the teacher reported increase in imitation of other children, following classroom routines without prompting, quickly learning new routines, remaining in close proximity to other children and exhibiting appropriate independent and parallel play
Year 2: At 2 years, 1 month chronological age (CA), improvements in all domains in the VABS except for daily living skills (not targeted in early treatment) were noted. At 2 years, 9 months CA, improvements were documented in all skill areas. After 18 months of treatment, most of the subject’s skills were at or above age level. Specifically, her score on the Leiter showed an age equivalent of 3 years, 5 months, The Expressive One-Word Picture Vocabulary Test indicated performance at a 4 year old level. Receptive language, daily living, socialization and motor skills were assessed to be slightly below her CA level and her overall communication skills measured slightly above her age level on the VABS. Additional subjective data reported she had good eye contact, spontaneous and age appropriate speech, joint attention, and play skills.

Year 3: Program emphasis was on daily living skills, peer social interactions, and answering social questions. When the subject was 3 years old, she was tested using the McCarthy Scales of Children’s Abilities indicated an age equivalent of 3 years, 3 months and a cognitive index of 107 (M=100, SD=16). Additional subjective data reported that she had excellent attention to task, cooperative and compliant, showed interest in peers, age appropriate social behavior, and good imaginative play.

The subject was not eligible for services following this assessment, however, the treatment team viewed her social and communication skills as emerging and fairly fragile so services were funded for one more year. At the end of the 4th year, the subject had age equivalents that were well above her chronological age and a full-scale IQ of 119 on the Wechsler Primary and Preschool Scale of Intelligence-Revised. She
scored at a 5 year old level on the draw a person test and had excellent eye contact and attention during a 2 hour testing session. She did not exhibit any behavioral or developmental abnormalities. The subject enrolled in a regular kindergarten classroom with no special services, support or intervention. She did not meet the diagnosis of autism or any other disorder. Teachers did not notice anything different and were not informed of her history.

Strengths:

1) Positive outcomes in all areas including loosing the diagnosis of autism
2) Support faded over time and learning occurred in natural environments

Limitations:

1) Small sample size
2) There was no control group
3) There was no follow-up measure or measure of generalization

Perry et al., 2008

This study examined 2 main research questions:

1) What statistical and/or clinical changes were made after completion of the IBI intervention program on developmental and diagnostic measures (severity of autism, cognitive level, adaptive level, and rate of developmental change)?
2) What was the range of progress upon completion of the program and did any individuals achieved “best outcomes” as reported in the literature?
This retrospective study reviewed files of children participating in intensive ABA programs 20-40 hours per week for 4 to 47 months in duration. The intervention programs took place in a variety of settings including home, center, and community.

The group was divided into 3 categories based on Vineland Adaptive Behavior Composite (ABC) Standard Scores (Communication, Daily Living, and Socialization scales). They were split into “higher” functioning (score of 60 or above), intermediate functioning (score 50-59), and lower functioning (score 49 or lower). Group were considered significantly different (p<0.001 or better) on CARS, Rate of development, adaptive and cognitive functioning prior to intervention verifying that the groups were meaningfully different subgroups.

All groups improved (p<0.001) on their autism severity (CARS) from pre to post intervention. More specifically, 138 children (50%) improved significantly enough to change to a milder category of autism severity. Cognitive level was measured by IQ and Mental Age (MA). The higher functioning group made gains of approximately 20 IQ points (p<0.001), Intermediate group gained approximately 10 IQ points (p<0.001), and the lower group did not change in IQ points, however, they did have significant improvement on MA (increased from 19 to 25 month level). In terms of MA, change was considered clinically significant if there was an increase of 12 months or more. Clinically significant changes in MA occurred in 61% of the children and 32% of these children increased 24 months or more. All 3 groups had significant changes in MA (p<0.001).
Adaptive behavior was measured by VABS age equivalents for all domains. All groups reported statistically significant changes \((p<0.001)\) in adaptive behavior after intervention as measured by the VABS age equivalents. Upon examination of standard scores (corrected for age), the higher functioning group showed significant improvement on Communication, Socialization, and Adaptive Behavior Composite (ABC), but no change on Daily Living Skills. The intermediate group significantly improved on communication and showed no change in socialization or adaptive behavior and declined on daily living skills. Finally, the lower functioning group did not change on communication or socialization and declined on daily living skills and adaptive behavior.

The rate of development was also calculated prior to intervention \((0.32, SD=12)\) and post intervention \((0.77, SD=0.76)\), typical rate of development would be 1.0. As a whole, there were statistically significant changes and rate of development varied by group. The higher functioning group more than doubled their initial rate of development and were improving at a rate higher than typical development. More specifically, 53\% of this group had rates of development at or above the typical range. The intermediate group doubled their rate of development. Specifically, 30\% of this group was developing at or above typical range. The lower group showed an increase 2.5 times their initial rate of development and 15\% of these children were developing at or above typical range.
To answer question 2 (What was the range of progress upon completion of the program and did any individuals achieved “best outcomes” as reported in the literature?), seven categories of progress/outcomes were established. The results are as follows:

- 10.8% - Average functioning
- 14.5% - substantial improvement
- 30.4% - clinically significant improvement
- 10.5% - less autistic
- 8.4% - minimal improvement
- 18.6% - no change
- 6.8% - worse

Strengths:

1) Large sample size
2) Positive gains were seen in all groups regardless of severity of autism

Limitations:

1) Retrospective review
2) There was no control group
3) The range of age at start of the program was large (20 to 86 months) and the length of intervention varied greatly (4 to 47 months)
4) Assessors were not blinded and/or not always independent of the treatment team
5) Assessments were completed at start and end of treatment rather than at regular time periods

6) No follow-up measure or measure of generalization

Weiss, 1999

The goal of this study was to evaluate the predictive utility of early learning rate within an intensive applied behavioral intervention. This home-based, ABA intervention program was implemented 40 hours per week for 2 years.

Results: Prior to intervention, all subjects (N=20) scored in the severely autistic range on the CARS. Post-intervention, 9 subjects scored in the non-autistic range, 4 others scored in this range but showed signs of mild manifestation of autism, 4 subjects scored in the mild-moderate range, and 3 in the severe range.

Mean scores on the VABS improved from pre-intervention (M=49.85, SD=7.84) to post-intervention (M=83.6, range 41-125, SD=28.28). There was great variability in skill acquisition across participants. For example, the range of days it took to master 30 receptive commands was 30-548 days (M=110.37, SD=126.49) and variability in verbal imitation (for 30 sounds/words) ranged from 30-659 days.

Additionally, a multiple regression analysis showed that the scores on the second administration of the CARS were predicted by rate of progress on three initial programs: Verbal Imitation, Receptive Commands and Object Manipulation. Specifically, acquisition of the first 5 items on these programs accounted for more than 70% of the variance on CARS scores two years into treatment ($R^2 = 0.731$). Furthermore, scores on
the second administration of the VABS were predicted by early acquisition data on the Verbal Imitation, Non-verbal Imitation, and Receptive Commands and accounted for more than 70% of the variance ($R^2=0.714$).

Two years after intervention, 7/20 children were placed in full-time, regular education classrooms without support. Another 3 children were in regular education classrooms with minimal support (related services or part-time instructional assistant). Five children were placed in regular education classroom and had 1:1 discrete trial instruction for part of the day and a full time aide. The last 5 children were placed in special education. Finally, 85% of the subjects were classified into final outcome group based on initial acquisition rate.

Strengths:

1) Positive gains among subjects

Limitations:

1) No control group

2) Both the CARS and VABS (the two main outcome measures) rely on parent report

*Boulware et al., 2006*

This study had two main objectives:

1. To develop an intervention program for young children with ASD that was effective, acceptable to the consumers and blended ABA and early childhood special education and
2. To determine which measures would be most useful to document progress in young children with ASD.

This study utilized a blended intervention model including applied behavior analysis, integrated playgroup, service coordination, and family education and training.

The intervention program consisted of several components including:

1. Family-centered, Inclusive Early Childhood Program, utilizing transdisciplinary teaming, and based on empirically tested interventions

2. Inclusive playgroups promoting high levels of engagement and systematic teaching

3. Extended instructional time utilizing developmentally and age appropriate activities.
   Instructional techniques include: naturalistic teaching, embedded learning opportunities, positive behavior support strategies, discrete trial teaching, visual supports, and response prompting strategies.

4. Technical and social support for families involving home or community visit for 2 hours per week.

5. Additionally, families were asked to provide 5 additional hours of training per week to their child and were given instructional program ideas and guided on how to support their child.

6. Monthly “toddler topic” educational groups were offered to parents. The ultimate goal was to increase family member’s competence and confidence in their abilities to promote their child’s development and handle challenging behavior
7. Service coordinators were liaisons among the staff members in playgroup and extended instructional time activities, parents, and any other professionals working with the family.

8. Service coordinators also provided assistance with the child’s transition planning.

Results: The Mental Scale on the Bayley Scales of Infant Development-Second Edition (BSID-II) was administered indicated that 6/8 children showed an increase in a Mental Developmental Index (MDI) score. At intake, five children scored in the significantly delayed range and three were in the mildly delayed range. At completion, two remained significantly delayed range, three were classified in the mildly delayed range, and four were functioning in the normal range.

The proportional change index (PCI) was calculated to measure rate of development during intervention. Upon completion of the program, six of the children demonstrated a score greater than 1.0 demonstrating that the intervention had a positive effect on the child’s development.

The Temperament and Atypical Behavior Scale (TABS) was utilized as a measure of self-regulatory behavior. The TABS showed that three children who were rated as atypical regulatory behavior at the start of the intervention improved and scored in the typical regulatory behavior upon completion. One child shifted from atypical regulatory behavior to “at risk” and the other four children did not change status.

Communication and Symbolic Behavior Scales (CSBS) indicated improvements in communication. Specifically, at intake, five children were classified at the pre-linguistic
stage, one at the early one-word stage and one at the multi-word stage. Upon completion, two children remained pre-linguistic, one entered the early one-word stage, and four were in the multi-word stage. Additionally, upon completion, children used a wider variety of forms and functions to communicate.

Functional outcomes- Upon completion the following gains were made:

1. 5/8 children used more than 5 words spontaneously as compared to one at start
2. 6/8 children followed simple instructions (compared to none at start) and 4/8 followed complex instructions
3. The entire group could imitate as compared to one at start
4. 6/8 children used toilet appropriately during waking hours (compared to none at start)
5. Play abilities increased for 6/8 children.

Elementary school placement: 4/7 children had full-time placement in general education classrooms; one of these children no longer had an individualized education program (IEP). Three children still received varying levels of support in general education and one student was being homeschooled. Two students were in full time special education classrooms focused on children with ASD.

Overall, upon discharge from early intervention, at age 3, this study showed many positive outcomes. 5/7 children were verbal and 2 children who were non-verbal were proficient in Picture Exchange Communication System (PECS). On the BSID-II, six children scored in either the normal or mild delay range and on the TABS, only one child...
scored as atypical. Additionally, all of the children made improvements in functional outcomes including toilet training, imitation, verbal communication, and interaction with peers (derived from a curriculum-based measure). Finally, follow-up data show that in first grade, 57% of the children were in general education classrooms meeting grade level academic demands and 29% of children were in segregated special education classrooms. Parents of 50% of the children also report their children have meaningful and reciprocal peer relationships both at school and in the community.

Strengths:
1) Children were followed up until elementary school placement to see if improvements were maintained over time.
2) All children made positive outcomes in at least one area

Limitations:
1) Small sample size
2) There was no control group
3) Homogeneous sample in terms of racial and ethnical background

Vismara & Rogers, 2008

The purpose of this case study was to pilot a parent-education program: the Early Start Denver Model (ESDM) for a very young child who demonstrated a behavioral profile similar to a young child with ASD. The ESDM combines (1) relationship based intervention focused on language development, social functioning, and development of non-verbal communication and imitation as the foundation for verbal language with (2)
Pivotal Response Training (PRT) which is based on the principles of ABA. PRT incorporates motivation with systematic teaching approaches to increase play skills, language, and communication in a natural setting. The duration of this intervention was 9 months with follow-up assessments at 3 month post intervention and at 24 months of age.

The initial evaluation took place at 9 months CA indicating: limited interest/non-responsive to parents or examiner, minimal eye contact, often “looked through” adults, limited social anticipation, limited interest in face to face interactions, no anticipatory movements upon being picked up, inconsistent orientation to name but consistent orientation to environmental noises, extreme interest in toys with appropriate play. Language/communication: limited vocalizations, occasional lip sounds (raspberries) and one consonant-vowel combinations; gesture use was limited to reaching for toys but was not combined with eye contact or vocalizations. There was also presence of unusual sensory and repetitive behaviors (visual attracted to carpet pattern and rubbing index finger on carpet until redirected to toy, and squirmed away when being picked up), limited social facial expressions and during a social tickle game, there was no eye contact or interest in examiner, only starred at ceiling.

There were 12 intervention sessions, each session lasting 1.5 hours followed by four, 1.5 hour follow-up visits at 2 weeks, 4 weeks, 2 months and 3 months post-intervention. Intervention took place in a clinic setting and focused on the ten main principles of the ESDM intervention with an emphasis on teaching skills to promote
social engagement, emotional sharing, imitation, non-verbal and verbal communication skills. The ESDM promotes development in areas known to be problematic for individuals with autism and follows a developmental sequence exhibited by children with no developmental delays.

Initially, at 10-12 months CA, a rapid rate of skill development was exhibited (referencing others during play, orienting to social sounds, imitation, increased language skills, etc). During the next couple months (12-15 months CA) there was a steady growth in social communication behaviors including joint attention, use of open vowel sounds to make requests, and using signs to communicate. From 15-17 months CA, interest in parent’s actions increased and interest in playing with toys alone decreased. He started to seek out activities that involved adult interaction with a preference for social play as compared to solitary play, he began vocally imitating several consonants related to objects and action words during play, he combined words and gestures, and eye contact increased. At 18 months CA, there were frequent initiations of social activities/play, consistency with turn taking, imitation of adults, frequent initiations of single and multiple syllable utterance paired with gestures, and good eye contact.

According to the Mullen Scales for Early Learning (MSEL), gains were made in receptive language and continued to score well on the visual reception and fine motor scales. Some gains were made on the expressive language scale although performance remained below average. Administration of the ADOS suggested a diagnosis of ASD.
At the follow-up (24 months CA), parents reported implementation of an intensive intervention (20-25 hours/week home based ABA, 1 hour/week of speech therapy, and 2 hour/week of playgroup) outside of this program since 18 months CA. The MSEL indicated average to above average skills in visual reception and fine motor skills. He demonstrated above average skills in receptive language and showed gains in expressive language scale. Additionally, upon re-administration of the ADOS, a diagnosis of PDD-NOS was given.

Parent acquisition of ESDM techniques was assessed. The level of mastery of intervention techniques for the subject’s father increased from 71% in the first intervention session to 95% across the final 3 sessions. Frequency of spontaneous functional verbal utterances and number of spontaneous imitative behaviors were also assessed. Prior to intervention, the subject showed no verbal production and very limited use of imitative behaviors. Spontaneous utterances during intervention ranged from 1-14 during parent-child interaction and 0-11 for child-therapist interaction. Imitation skills ranged from 1-8 with parent-child interaction and 0-5 with child-therapist interaction. Additionally, steadily improvements were made in attention and initiation of social behaviors with father. This paper provides preliminary evidence that the ESDM parent training intervention may be an effective model for early intervention for infants and toddlers with ASD.
Strengths:

1) Evaluation for ASD was completed by an independent clinician, not on the treatment team

2) Improvements were made in several developmental areas

Limitation:

1) Small sample size

2) There was no control group

3) Additional intervention began between end of this intervention and follow up, so new intervention could have influenced gains made during this time.

Vismara et al., 2009

The main purpose of this study was to develop a short-term intervention addressing parent education using the ESDM to be implemented immediately following diagnosis of ASD. The two objectives were:

1) To assess the parents’ acquisition of the ESDM teaching procedures

2) To assess the changes in participant social communicative behaviors following this short-term educational intervention.

Intervention was implemented in a clinic playroom for 12 weeks, involving at least 1 parent, a therapist, and the child present at each session. Parents were taught the principles from ESDM. A manual was given to parents with emphasis on 10 therapy
strategies pertinent to the ESDM. Each strategy was targeted during one of the sessions. Each session lasted 1 hour.

Results: Parents showed a moderate level of correct implementation of the ESDM principles at baseline and all but 1 parent showed mastery of principles by the 5th-6th session. ESDM principles were maintained at follow-up assessment for those parents who met mastery.

Visual inspection of the data showed improvement in spontaneous functional verbal utterances and imitative behaviors throughout the intervention during parent/child and child/therapist interactions. Follow up assessments were completed on six children indicating continued improvement (from end of intervention) in spontaneous functional language and imitation skills. Additionally, children showed larger gains in language and imitation once their parents reached mastery level on the ESDM. Furthermore, levels of attentiveness and social initiation behaviors during parent-child and therapist-child interaction continued to increase throughout intervention (low-moderate levels at baseline to moderately high during intervention) and remained at positive levels at follow-up.

This feasibility study is a good stepping stone to show the efficacy of the ESDM intervention for young children with ASD. All parents exhibited improvement in use of ESDM strategies to promote increased attention, positive affect, imitation, and communication. The majority of the parents mastered the ESDM techniques by the 5th-6th week of intervention. Furthermore, outcome data showed that parents utilized these
interactive skills into their everyday lives with their children and sustained them for at least 3 months after treatment ended. Individual data showed that the children had almost no social communicative behaviors, initiation, imitation, or speech prior to intervention. All children showed a gradual improvement in all behaviors during treatment and those who completed follow-up assessments continued to exhibit gains in these behaviors. Specifically, no child had speech prior to treatment and they were all using speech as a way to communicate intentionally during the final intervention session. Speech production continued to improve was even higher at follow-up. The fact that speech continued to increase at follow-up is a good indicator that parent’s new skills are pertinent in child’s gains.

Finally, there were 2 children with extreme, aggressive behaviors (biting, knocking down furniture, head banging) during the intervention period. For these children, the behavior management lessons were taught early on and children showed immediate decreases in disruptive behavior in a variety of environments. These aggressive behaviors occurred at low rates for the remaining time in intervention.

Strengths:

1) All parents and children showed positive improvements

2) Parent were active members of the intervention team

3) Follow-up assessment with maintenance and improvement of skills

Limitations:

1) Small sample size
2) There was no comparison group

3) Convenience sample

**Dawson et al., 2009**

This randomized controlled trial examined cognitive development in children with autism who received either the ESDM or a community-based assess and monitor (A/M) program. The ESDM is a comprehensive behavioral intervention that combines ABA with developmental and relationship-based approaches. Intervention occurs in the natural environment and is delivered by trained therapist and parents. The children in the ESDM group received 20 hours/week of intervention (2 hour sessions twice/day for 5 days) from a therapist and at least 5 hours/week from parents for a 2 year time period. The A/M group received yearly assessments and intervention from community therapists as recommended. On average, this group reported 9.1 hours/week of individual therapy and 9.3 hours/week of group intervention.

This study utilized blind assessors (to the intervention status and assessments). Assessments were administered at baseline, after 1 year of intervention and after 2 years of intervention or at 48 months CA (whichever was longer).

Results: Year 1: Significant results (p=0.018) were reported at the end of year 1 for the ESDM group for cognitive ability on the Mullen Scales of Early Learning (MSEL) composite standard scores. This group improved average IQ scores 15.4 points (>1 SD) compared to an increase of 4.4 points in the A/M group. Additionally, the ESDM group showed significant difference (p=0.046) on the MSEL visual reception with an increase of
5.6 T-score points compared to the A/M group increase of 1.7 points. This was the only subscale score on the MSEL that had a significant difference. The ESDM group gained 17.8 points on the receptive language subscale as compared to 9.8 points in the A/M group, falling just below significance (p=.051).

Year 2: Significant improvement (p=0.044) was reported at the end of year 2 for the ESDM group for cognitive ability on the MSEL. The ESDM group average IQ score improved 17.6 points compared to an increase of 7.0 points in the A/M group. The majority of this change was due to improvement in receptive and expressive language. The ESDM exhibited improvements of 18.9 and 12.1 points, respectively, compared to the A/M group which showed improvements of 10.2 and 4.0 points, respectively. Additionally, the two groups differed in their VABS composite standard score. The ESDM group had similar scores at 1 year and 2 year outcomes indicating a steady rate of development. The A/M group exhibited an 11.2 point decline indicating their adaptive behavior deficit became larger when compared to the normative sample. There was no difference in the groups in relation to ADOS severity score or Repetitive Behavior Scale (RBS) total score after 2 years of intervention.

Additionally, diagnosis improved from baseline to year 2 from autistic disorder to Pervasive Developmental Disorder Not Otherwise Specified (PDD NOS) for 29.2% (7 subjects) in the ESDM group as compared to 4.8% (1 subject) in the A/M group. Furthermore, diagnosis changed from PDD NOS to autistic disorder for 8.3% (2 subjects) for the ESDM group compared to 23.8% (5 subjects) in the A/M group.
Following intervention, children in the ESDM group exhibited significant changes in IQ, adaptive behavior, and diagnostic status compared to the A/M group. Specifically, the ESDM showed significant changes in communication abilities (receptive and expressive language) on the MSEL subscales and the VABS communication subscale. Additionally, according to the VABS, the ESDM group showed improvements in overall adaptive behavior, communication, daily living skills, and motor skills.

Strengths:
1) Random assignment
2) Comparison group
3) Blinding of assessors
4) Intervention group displayed higher levels of improvement as compared to control group

Limitations:
1) No follow up assessment or generalization

Katagiri et al., 2010

The purpose of this study was to evaluate whether “mirroring” interaction (being imitated by an adult) can promote social responsiveness in toddlers with ASD. Subjects were divided into 2 groups according to age (2 years vs. 3 years). The intervention lasted one session. Specifically, the session consisted of a baseline period (2 minutes), mirroring phase (3 minutes), and a second baseline period (2 minutes). There were 2
sets of toys (one for child/therapist). During the first and last period, the experimenter played with different toys than the child and during the mirroring phase, the experimenter did everything exactly as the child did (including facial expressions and vocalizations).

Behaviors were categorized into social attention: (gazing at the experimenters face) and socio-emotional behaviors (smiling, verbalizing, vocalizing, approaching, touching, offering toys to experimenter, and requesting the experimenter imitate his/her action). Significant differences were found between groups (p<0.01), phases (p<0.001), and categories (p<0.01). Additionally, the frequency of social behaviors was greater in the 3 year old group than the 2 year old group in both phases. Social behaviors occurred more frequently in the mirroring phase than in baseline 1 (p<0.001). Social attention was exhibited more often in the mirroring phase than in baseline 1 in the 2 year old group (p<0.05) and social emotional behaviors occurred more often in the mirroring phase than in baseline 1 for 3 year olds (p<0.05). Overall, All subjects improved in social attention during and/or after the mirroring phase. Social emotional behaviors appeared during and/or after the mirroring phase in 4/6 subjects who did not display any such behaviors in the baseline 1 phase. Additionally, IQ was negatively correlated with mirroring effect for social attention (r=-0.63, p<0.01) and was positively correlated with the mirroring effect for social emotional behaviors (r=0.56, p<0.05).

In summary, 2 year olds exhibited less social behaviors than 3 year old children with ASD and the mirroring effect varied for different types of social behavior by age.
Specifically, social attention was significantly improved in 2 year olds and social emotional behaviors were significantly improved in 3 year olds. Additionally the lower IQ of the toddler, the greater the mirroring effect on social attention. Previous studies suggest that imitation is beneficial for children with lower developmental functioning. The clinical significance of these results imply that mirroring effect can beneficial in a therapeutic setting for toddlers with ASD.

Strengths:

1) Positive outcomes for both groups

Limitations:

1) Intervention was very short
2) No follow-up
3) Small sample
4) No control group

Sanefuji et al., 2009

This case study examined the impact of a 6 month, imitation-based intervention on social communication development, of a 21 month old child at risk for ASD. Prior to the intervention, the child made no eye contact, did not respond to his name, mouthed objects, wandered aimlessly, and did not show interest in others. His CA was 19.5, 15 months for locomotion, manual movements, self help skills and interpersonal relationships, and 10 months for production and comprehension of language. He had a pre-occupation with sensory motor stimulation and his mother had difficulty interacting
with him. She was apprehensive about engaging with her child due to the non-responsive reaction she got back. She had negative emotions towards interacting with him and a low level of engagement. This child was at high risk for PDD NOS or ASD.

The intervention focused on imitation and was implemented 60 minutes per month at a public health center. It was supplemented with parents doing imitation therapy for 5 minutes each day. For the first 2 months, the mother was encouraged to provide contingent engagement with her child, immediately responding to him while focusing on his interests. The following 2 months, she was asked to imitate her child’s behaviors including meaningful utterances and facial expressions. For the last 2 months, she went back to contingent engagement with no imitation. According to the mother’s data, she provided the intervention all but 5 days during the 6 months of intervention.

Results: Joint attention – prior to intervention, the subject exhibited almost no joint attention skills. After the first 2 months (contingent engagement) the child’s joint attention increased to an 8 month old level. Following the first month of imitation (month 3), the child’s joint attention skills increased to the 11 month old level. After the 4th month (2nd month of imitation), the child’s joint attention skills increased to a 15 month old level. Following the 5th month (contingent engagement) the child’s JA skills improved, but were still at a 15 month old level. After the 6th month (contingent engagement), the child’s JA behaviors decreased slightly, but were still at a 15 month level.
Mother’s negative emotion towards child: Prior to intervention, the mother’s negative emotion score (on the Infant Bonding Scale) was a 12. After the first month of intervention, this score decreased to a 6, after the third session it was a 5, after the fourth it dropped to a 4 and by the sixth session, it was a 3. Additionally, the mother-child interaction continued to improve throughout the intervention from difficult, serious, and unsatisfying prior to start of therapy to enjoyable, high levels of engagement and mutually satisfying after the last session.

Child’s Development: Post intervention, the child was 27 months of age with a developmental age of 25.5 months. He was 19.5 months for locomotion, 17 months for manual movements, 22.5 months for self help skills, 11.5 months for interpersonal skills, and 17 months for production and comprehension of language.

Strengths

1) slight improvement in social behaviors

Limitations:

1) Small sample

2) Convenience sample

3) No control group

4) Improvements are minimal and could be due to maturation.

Wong & Kwan, 2010

This randomized controlled trial was a pilot program for a short-term, 2 week intervention implemented immediately after diagnosis focusing on gesture use, eye
contact, and vocalizations. The goal was to examine the effectiveness of the
intervention on improving communication and social interactions of toddlers with
autism, in a 2 week period of time, while waiting for community early intervention
services to begin.

The experimenters used a single-blind cross over design and assessments were
administered at 3 time periods (baseline, time 1 and time 2). The intervention was
carried out over 10 sessions (2 weeks). The RCT started with baseline continued
through time 1 (intervention delivered to treatment group while control group was the
no-treatment arm). Upon completion of the intervention, time 1 assessment began and
data was collected within 2 weeks for all subjects. The RCT ended with time 1
assessment for both groups. The researchers implemented the intervention with the
control group for a 2 week period and re-administered the assessments (time 2). Both
groups were combined for a larger sample size to assess for intervention effects.

The intervention was delivered 5 days per week, approximately 30 minutes each
session, over a 2 week period. An autism therapist taught the toddlers to use eye
contact, gestures and vocalizations and taught the caregiver how to implement similar
techniques at home. The majority of these children were pre-verbal so initial training
focused use of simple words to request favorite toys. Parents were given simple
instructions on how to teach their child and were asked to focus on one skill repeatedly
until the child was familiar with it prior to moving to a different skill. Parents were
encouraged to spend 5-10 minutes every hour teaching their child a skill. Parent’s logged their training hours.

Results:

The intervention group scored significantly lower (p=0.007) at time 1 than at baseline on the ADOS Language and Communication subscale as compared to the control group who had no significant difference. Additionally, Vocalizations Directed to Others, Pointing, and Gestures were put through further analysis since these were the main targets of intervention. Results showed that the intervention group showed significant (p=0.005) improvement on Vocalization Directed to others whereas the control group did not show significant difference.

The intervention group, had a significant (p=0.011) reduction in Reciprocal Social Interaction score on the ADOS as compared to no significant change in the control group (p=0.122). Additionally, the intervention group showed significant improvements on Requesting (p=0.01) as compared to the control group who did not show a statistical difference.

The intervention group improved in symbolic play skills with an increase in standard scores that approached significance (p=0.026 with an adjusted alpha level at 0.025) where as the results for the control group were non-significant.

Groups were combined to examine intervention effect for the entire group. There was no difference (p=0.331) between the groups. Group analysis indicates that there is a significant difference in Communication and Language subscale (p=0.004),
Pointing (p=0.001), Vocalization (p=0.001). There was no significant effect found in items that were not targeted in intervention. There was a significant reduction in the Reciprocal Social Interaction subscale score (p=0.002) and no significant findings in items not targeted in intervention. Toddlers made significant improvements in symbolic play (p=0.005). Parents report indicated significant improvement in Language (p=0.010), social relationships (p=0.007). No improvement perceived on subtests not targeted in intervention. There was a significant difference in parent stress level (pre and post scores) (p=0.004).

Overall, results show effectiveness of intervention in the areas of communication and social interactions of children diagnosed with autism. Specifically, improvements in vocalizations, quality of social interactions, pointing and requesting were shown. Parents also reported the felt confident in interacting with their child after learning appropriate techniques. Parents perceived significant positive changes in their child’s behaviors and in their level of stress on a daily basis. Involving the parents in the intervention allowed for generalization of skills to their natural environment.

Strengths:

1) Positive gains in core deficits

2) Short duration of intervention

3) Involvement of parents to help generalize
Limitations:

1) Small sample size
2) No measure of generalization or follow-up.

**Solomon et al., 2007**

The purpose of this study was to examine the effectiveness of the Play and Language for Autistic Youngsters (PLAY) Project Home Consulting model. The PLAY project is based on Stanley Greenspan’s developmental, individualized and relationship-oriented (DIR) model. This model focuses on improving social reciprocity and functional and/or pragmatic communication of children with communication disorders. It is an intensive, comprehensive and multidisciplinary approach to therapy.

Home consultants work with the families one time per month for 3-4 hour sessions. Duration of intervention lasted from 8-12 months. Parents were asked to interact with their child 1:1 for 15 hours per week implementing the DIR/Floortime model. During the therapy session, the consultants taught the caregivers how to provide 1:1, intensive, play based intervention to their child. A training manual was provided to help guide the parents and parents participated in a 1 day workshop focusing on how to contingently and reciprocally engage their child. There was a 7 step skill sequence used to train parents. Monthly visits consisted of modeling, coaching, video assessment, and review of written objectives to help apply and refine the skill sequence. The Preschool Autism Rating Scale (PARS) was used to help write objectives.
A pre/post research design was implemented to measure progress before and after the first year of the PLAY project.

Results: There were significant improvement noted in the children’s total and scaled Functional Emotional Assessment Scale (FEAS) scores following intervention (p<0.0001) but no significant change in parents’ FEAS scores. Overall, 45.5% of children made good to very good functional developmental gains based on the FEAS scaled score during the study. No statistical relationship was found between severity of ASD at start of the study and FEAS scores. Additionally, there was a trend in the association between fewer hours per week spent providing intervention and lower scores on outcome measures (p=0.09).

There were changes (p<0.0001) in the children’s clinical Functional Developmental Levels (FDL) from start to end of intervention. Specifically, 52% of subjects made very good (1.5 FDLs or better) clinical gains during intervention and 14% made good gains (increase 1-1.5 FDL). No statistical relationship was found between ASD severity at start of intervention and clinical scores. Lastly, 70% of the families involved reported that they were very satisfied with the PLAY project, 10% were satisfied and 20% were somewhat satisfied.

Strengths:

1) Positive results in core deficits

2) Parent involvement in intervention implementation
Limitations

1) No control group

2) No follow-up measure or measure of generalization.

**Schertz & Odom, 2007**

The purpose of this study was to determine the effectiveness of an intervention focused on teaching non-verbal communication skills thought to be the developmental precursors for joint attention skills.

Intervention was implemented by the parents in their homes over 17-21 weeks. This intervention promoted parent-child interactions in an open-ended but deliberate fashion within the context of building a relationship rather than in a skill based approach utilizing external reinforcement. Parents played face to face games to promote interaction. They were given The Joint Attention Mediated Learning (JAML) manual to provide a framework for their interactions with their child.

Parents were given a written and oral summary of mediated learning principles as they relate to promotion of joint attention skills. Intervention occurred in four phases. As each child progressed, their parent was coached and given suggested tasks to implement during each phase of intervention. Phases were implemented in sequence with each phase having 2 levels: (1) relying on parent initiative and (2) seeking more initiative from the child. Activity ideas were given to parents to help them develop ideas of their own to promote child success. Parents were encouraged to devote at least one hour per day to doing face to face parent-child interaction during
their daily routines and planned interactions. Weekly sessions were provided by the researcher to review parent notes, videotape a 10 minute parent-child interaction for data analysis, teaching of new material, planning for the next week, and reviewing any questions/concerns. Intervention ended when the children progressed through the phases of intervention and demonstrated several instances of initiating joint attention or there was a plateau in progress.

This study utilized a mixed methods, multiple baseline intervention to compare the children’s improvements across 4 intervention phases: focusing on faces, turn taking, responding to joint attention and initiating joint attention. Additionally, qualitative data was analyzed from parent-researcher conversations and parent’s notes.

Quantitative Results:

Prior to intervention, toddlers showed varying rates of focusing on faces and turn-taking but none of the toddlers responded to or initiated joint attention behaviors. Overall, all 3 toddlers improved upon their baseline levels on all 4 targeted outcomes. Toddler 1 improved the most on focusing on faces, had moderate progress with turn taking and minimal progress with either joint attention skill. Toddler 2 steadily improved in all phases of intervention. Toddler 3 quickly progressed through all phases before they were actually introduced. This toddler was focusing on faces prior to intervention start. Follow-up assessment five week post intervention indicated that all toddlers continued making improvements with scores higher than their mean intervention scores. Social validity measures indicated that parents were supportive of
the intervention and weekly goals, their role in delivering the intervention, and the level of intensity.

Qualitative results:

Several themes evolved from the qualitative data including child progress in social-communication development facilitated reduced aggression, physical activity motivated children’s interactions, simplifying the presentation of parent’s face facilitated focusing on faces, and turn taking activities revolving around face to face play encouraged joint attention better than play with toys.

Overall, 2 out of 3 toddlers with early identified autism exhibited joint attention skills following intervention. The third toddler made progress in focusing on faces and turn taking. Caregivers implemented the intervention in the child’s natural environments through a play-based approach. This improved the child’s skills in addition to benefiting the parents by helping them realize their child’s potential and how much impact they can have on their child’s development.

Strengths:

1) Positive gains were made

2) A manual was utilized to provide a framework for parent-child interactions

Limitations:

1) Small sample size

2) Convenience sample
Newcomer, 2009

This study focused parent education strategies such as setting up learning opportunities, arranging the environment appropriately and implementing positive reinforcement strategies. Behavioral data was collected via videotaped parent-child sessions. Parent measures included: arrangement of learning opportunities, responsive event and model delivery, and vocal expansions. Child measures include: facial orientation, communicative attending and coordination of joint attention.

The first 5-6 sessions focused on baseline data collection, rapport building between the therapist and the parent/child, and parents were given the parent manual. Baseline assessments consisted of a 10 minute play session between the parent and child during normal play. Once the intervention began, the therapist taught the parents foundational skills based on the Family Centered Parent-Child Training Program, Teaching DANCE. These skills are based on naturalistic behavioral interventions such as incidental teaching, milieu teaching, and pivotal response training. The child’s intervention goal was to increase eye contact measured through facial orientation. First the therapist modeled the behavior for the parents and then coached the parents through the steps, giving them feedback. Duration of intervention ranged between 6-10 sessions.

Results: Throughout the intervention, the number of teaching episodes between parent and child increased on an average of 14.2 episodes per 10 minute assessment and the amount of successful communicative attending attempts by the child increased on
average 10.5 attempts over 10 minute assessment. Facial orientation also increased from baseline (average of 1.8) to post-intervention an average of 9.8. Prior to baseline, children exhibited minimal orientation to faces and when they did orient, they were not orienting as a method of communication. Finally the average number of coordinated joint attention attempts increased from baseline (6.8) to post-intervention (25.8).

Strengths:

1) Positive gains made in all outcome measures
2) Parent involvement in intervention implementation
3) A manual was utilized to provide a framework for parent-child interactions

Limitations:

1) Small sample size
2) There was no control group
3) Convenience sample
4) No standardized assessments utilized