

10-3-2014

Early Characteristics of Children Who Lose Their Autism Diagnosis Between Age Two and Four

Emily Moulton

University of Connecticut - Storrs, emily.moulton@uconn.edu

Recommended Citation

Moulton, Emily, "Early Characteristics of Children Who Lose Their Autism Diagnosis Between Age Two and Four" (2014). *Master's Theses*. 678.

http://digitalcommons.uconn.edu/gs_theses/678

This work is brought to you for free and open access by the University of Connecticut Graduate School at DigitalCommons@UConn. It has been accepted for inclusion in Master's Theses by an authorized administrator of DigitalCommons@UConn. For more information, please contact digitalcommons@uconn.edu.

Early Characteristics of Children Who Lose Their Autism Diagnosis
Between Age Two and Four

Emily Moulton
B.A., Hamilton College, 2012

A Thesis
Submitted in Partial Fulfillment of the
Requirements for the Degree of
Master of Arts at the
University of Connecticut
2014

APPROVAL PAGE

Master of Arts Thesis

Early Characteristics of Children Who Lose Their Autism Diagnosis
Between Age Two and Four

Presented by

Emily Moulton, B.A.

Major Advisor _____
Marianne L. Barton, Ph.D.

Associate Advisor _____
Chi-Ming Chen, Ph.D.

Associate Advisor _____
James Green, Ph.D.

University of Connecticut
2014

ACKNOWLEDGEMENTS

Committee

Marianne Barton, Ph.D., Chi-Ming Chen, Ph.D., James Green, Ph.D.

Early Detection Team

Deborah Fein, Ph.D., Thyde Dumont-Mathieu, MD, Sarah Hodgson, Ph.D., Jamie Kleinman, Ph.D., Molly Helt, Ph.D., Katelin Carr, Lauren Herlihy, Kelley Knoch, Eva Troyb, Alex Hinnebusch, Lauren Haisley, Dasal Jashar, Kathryn Bradbury, Cara Cordeaux, Lauren Miller and Julia Chen

Undergraduate Research Assistants

Participating Families, Pediatricians, and Early Intervention Providers

TABLE OF CONTENTS

Acknowledgements -----	3
Table of Contents -----	4
Abstract -----	5
Introduction -----	6
Section 1: Diagnostic Stability of ASDs Across the Lifespan -----	7
Section 2: Diagnostic Stability in Toddlerhood -----	8
Section 3: Predicting Diagnostic Stability and Outcome -----	11
Section 4: Outcomes Following the Loss of an ASD Diagnosis ----	16
Section 5: Predicting Optimal Progress -----	18
Section 6: The Present Study -----	19
Methods -----	21
Participants -----	21
Procedures -----	24
Measures -----	25
Results -----	32
Discussion -----	42
Limitations and Future Directions -----	52
Conclusions -----	54
References -----	56
Appendix A: Tables -----	63
Appendix B: Figures -----	76
Appendix C: Questionnaires -----	96

Abstract

Emerging literature indicates that a subset of children with a documented ASD lose their diagnosis and demonstrate cognitive and adaptive abilities within the average range. Multiple factors including symptom severity, cognitive and language abilities, adaptive skills and early intervention may help to predict these highly positive outcomes. Participants in the present study include 207 children diagnosed with an ASD by clinical best estimate at approximately age two (T1) and subsequently re-evaluated at approximately age four (T2). 171 (82.6%) children retained an ASD diagnosis (ASD-ASD) at re-evaluation and 19 children (9.2%) were determined to meet the following criteria for an “Optimal Progress” (OP): met criteria for an ASD using gold standard diagnostic procedures at T1, no longer meet criteria for any ASD at T2, and demonstrated functioning in the average range on standardized measures of cognition, language, communication and social skills. Results indicate that a number of early (T1) child-level factors help to predict OP: a diagnosis of PDD-NOS, fewer restricted, repetitive behaviors (RRBs), less severe autism symptomatology and stronger adaptive skills (but not cognitive or language abilities). These early traits may reflect more intact central nervous system functioning, and/or may provide these children with a greater likelihood of benefiting from interventions and everyday interactions, and in turn, with a greater likelihood of demonstrating OP. In combination with the findings of the current study, future studies should attempt characterize the mechanisms at work in producing these outcomes, including the role of early intervention. In doing so, we can begin to promote an increase in the percentage of children attaining highly positive outcomes from ASD.

Autism Spectrum Disorders (ASDs) are a group of neurodevelopmental disorders characterized by deficits in communication and socialization accompanied by repetitive behaviors or restricted interests. In addition to these core deficits, individuals with ASD often experience a number of comorbid deficits including cognitive delays/intellectual disabilities, adaptive skill deficits and motor delays (Charman et al., 2011; Levy, Mandell, & Schultz, 2009; Lloyd, MacDonald, & Lord, 2013; Macdonald, Lord, & Ulrich, 2013; Volkmar, Lord, Bailey, Schultz, & Klin, 2004). The diagnostic criteria for ASD were revised in the recently published Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5, American Psychiatric Association (APA), 2013); however, some current research in the field utilizes diagnostic criteria from the DSM, Fourth Edition (DSM-IV, APA, 2000). According to the DSM-IV, ASDs include Autistic Disorder (AD), Pervasive Developmental Disorder – Not Otherwise Specified (PDD-NOS) and Asperger’s Disorder (APA, 2000).

Autistic Disorder is characterized by onset before age three and the presence of six total symptoms across three symptom domains: social interaction, communication and repetitive behaviors or restricted interests. Specifically, to meet criteria for Autistic Disorder, an individual must demonstrate at least two symptoms of qualitative impairments in social interaction (e.g., impairment in the use of eye gaze, decreased seeking of shared enjoyment), at least one symptom of significant impairment in communication (e.g., delay or lack of spoken language) and at least one restricted, repetitive and stereotyped behavior or interest (e.g., preoccupation with parts of objects, repetitive motor movements) (APA, 2000). A diagnosis of Asperger’s Disorder is given when impairments are present in the aforementioned areas of social interaction and restricted, repetitive behaviors or interests, with an absence of significant impairment in the areas of communication, cognitive abilities and self-help skills. A diagnosis of PDD-NOS is given when

an individual demonstrates significant impairment in social interaction accompanied by deficits in verbal or non-verbal communication or the presence of restricted, repetitive behaviors, but does not meet criteria for Autistic Disorder (APA, 2000). The Center for Disease Control (CDC, 2012) reports an overall prevalence rate for ASDs of one in 88, with boys affected at greater rates than girls (4.6:1). ASDs occur within all racial/ethnic and socioeconomic groups (CDC, 2012; Fombonne, 2003); however, evidence indicates that there may be disparities in access to health care that contribute to decreased reported prevalence and later age of diagnosis for minority children and children of families with lower socioeconomic status (Fombonne, 2003; Herlihy et al., 2014; Mandell, Listerud, & Levy, 2001).

Diagnostic Stability of ASDs Across the Lifespan

ASDs have long been considered lifelong disorders by clinicians and parents (Levy & Perry, 2011; Matson & Horovitz, 2010; Seltzer, Shattuck, Abbeduto, & Greenberg, 2004). Follow-up studies of individuals diagnosed in childhood indicate that between 80 and 90% of individuals continue to meet diagnostic criteria in adolescence or adulthood (Charman et al., 2005; Seltzer et al., 2004; Woolfenden, Sarkozy, Ridley, & Williams, 2012). While diagnoses remain largely stable, the stability of particular ASD symptoms appears to be highly variable across time, with course varying by symptom domain. In general, symptoms in the communication domain appear to improve over time, but remain impaired (Charman et al., 2005; Helt et al., 2008; Seltzer et al., 2004). Symptoms in the social interaction domain also appear to remain impaired for the large majority of individuals (Charman et al., 2005; Seltzer et al., 2004); however, some studies indicate improvement in this domain, particularly in adolescence (Matson & Horovitz, 2010a). In the domain of restricted interests and repetitive behaviors, evidence indicates that the severity of symptoms is quite stable, with modest improvements and possible

changes in the qualitative nature of symptoms over time (Charman et al., 2005; Matson & Horovitz, 2010; Seltzer et al., 2004).

Diagnostic Stability in Toddlerhood

Symptoms of ASDs are thought to emerge in early childhood, with recent studies indicating that behavioral symptoms may be present as early as two to six months of age in some children (Jones & Klin, 2013). Increases in the understanding of the early behavioral profiles of individuals with ASD have allowed reliable diagnoses to be given in early toddlerhood, often around age 24 months (Chawarska, Klin, Paul, Macari, & Volkmar, 2009; Eaves & Ho, 2004; Kleinman, Robins, et al., 2008; Turner & Stone, 2007). Given the increase in early diagnosis, it is of great importance that we understand the diagnostic stability of ASDs during the early years of a child's life. A number of studies have investigated diagnostic stability in toddlerhood, and broadly, evidence indicates that diagnostic stability is high following diagnoses given as early as 18 to 24 months. In some samples, 100% of children diagnosed with an ASD at approximately age two retain an ASD diagnosis later in their toddler years (Chawarska et al., 2009; Lord, 1995). Other studies have reported between 68 and 93% stability of diagnoses made at age two to follow up at approximately age four (Eaves & Ho, 2004; Kleinman et al., 2008a; Sutera et al., 2007; Turner & Stone, 2007). The stability of an ASD diagnosis is higher following a diagnosis of Autistic Disorder than a diagnosis of PDD-NOS. Across studies, 68 to 100% of children initially diagnosed with AD retain an ASD diagnosis in toddlerhood or early childhood, compared to 40 to 90% of children initially diagnosed with PDD-NOS (Chawarska et al., 2009; Eaves & Ho, 2004; Kleinman, et al., 2008a; Sutera et al., 2007; Turner & Stone, 2007).

Movement Within the ASD Spectrum. While broadly defined diagnostic stability of ASDs is high, movement within the autism spectrum is common. A number of children show an

improvement in symptoms over time, changing their diagnostic status from AD to PDD-NOS. Across studies, this occurs for 6 to 26% of children initially diagnosed with AD (Chawarska et al., 2009; Eaves & Ho, 2004; Kleinman, Ventola, et al., 2008; Lord et al., 2006; Turner & Stone, 2007). Another subset of children demonstrates a worsening of symptoms overtime, changing their diagnostic status from PDD-NOS to AD. Across samples, this occurs for 13-59% of children (Chawarska et al., 2009; Eaves & Ho, 2004; Kleinman, Ventola, et al., 2008; Lord et al., 2006; Turner & Stone, 2007). Children initially diagnosed with Asperger's Disorder also may change their diagnostic status. In a comparative longitudinal follow-up study, Cederlund and colleagues (2008) found that 10% of individuals diagnosed with Asperger's Disorder in childhood showed a worsening of symptoms, causing them to meet criteria for AD later in adolescence or adulthood (with the exception of the criteria of an early language delay).

Movement Off the ASD Spectrum. A subset of children initially diagnosed with an ASD at approximately age two appear to lose their ASD diagnosis by age four. Across studies, this occurs for 0 (Chawarska et al., 2009) to 37.5% of children (Turner & Stone, 2007). Notably, the majority of studies investigating diagnostic stability in toddlers found that between 7 and 18% of their sample lost their diagnosis (Eaves & Ho, 2004; Kleinman et al., 2008b; Sutera et al., 2007), indicating that more extreme findings (0%, 37.5%) may be the result of specific sample characteristics of those studies. For example, Chawarska et al. (2009), who reported that no children lost their diagnosis, had a slightly earlier age at initial diagnosis and younger age at follow-up than studies that found evidence for loss of diagnosis over time. Additionally, Turner and Stone (2007), who reported the highest percentage of these outcomes, reported that 100% of their sample received some form of early intervention between their two diagnostic evaluations. Unfortunately, studies reporting lower percentages of positive outcomes did not specifically

report on the proportion of their samples that received intervention, and therefore, it is unclear whether this factor is unique to Turner and Stone (2007).

In studies finding evidence for the loss of an ASD diagnosis over time, it appears that children initially diagnosed with PDD-NOS are much more likely to demonstrate this outcome than children initially diagnosed with AD. Kleinman et al. (2008) found that of 15 children initially diagnosed with PDD-NOS at approximately age two, 33.3% (5), no longer met diagnostic criteria for any ASD at age four, compared to 9.7% (4/46) children initially diagnosed with AD. In a partially overlapping sample, Sutera and colleagues (2007) found that 39% (7/18) of children initially diagnosed with PDD-NOS, and 11% (6/55) of children initially diagnosed with AD no longer met criteria for any ASD diagnosis. Eaves and Ho (2004) found that 56% (5/9) of children initially diagnosed with PDD-NOS, and 6% (2/34) of children initially diagnosed with AD no longer met criteria for any ASD diagnosis. In the study finding the highest percentage of these outcomes (i.e. 37.5%), Turner and Stone (2007) found that 60% of children (6/10) initially diagnosed with PDD-NOS and 31.5% (12/38) of children initially diagnosed with AD no longer met diagnostic criteria for any ASD. In sum, it appears that among studies with similar sample sizes, diagnostic procedures, and follow-up, between 7 and 37.5% of children lose their ASD diagnosis in toddlerhood, with this outcome consistently found to be more common in children initially diagnosed with PDD-NOS.

In studies with longer follow-ups, similar rates of movement off the spectrum are reported. In two separate studies investigating diagnostic stability following diagnosis at approximately age two to follow up at approximately age nine, between 4.6 and 12% of children were found to move off the spectrum (Lord et al., 2006; Turner, Stone, Pozdol, & Coonrod, 2006). Importantly, both studies note that movement off the spectrum most commonly occurred

either by age three, or between the years of two and five, rather than later in childhood (Lord et al., 2006; Turner et al., 2006). In a comparative longitudinal follow-up study, Cederlund and colleagues (2008) found that 10.7% of individuals diagnosed with Asperger's Disorder, Autistic Disorder, or Atypical Autism between the ages of 5.5 and 24.5 years no longer met criteria for any ASD at follow-up (follow-up was at least five years after initial diagnosis). Individuals initially diagnosed with Asperger's Disorder were more likely to move off the spectrum (11%) than their peers who were initially diagnosed with Autistic Disorder or Atypical Autism (1%) (Cederlund, Hagberg, Billstedt, Gillberg, & Gillberg, 2008). Thus, as we may expect, it appears that children with relatively less severe diagnoses (e.g., PDD-NOS or Asperger's Disorder vs. AD) are the most likely to no longer meet criteria for any ASD sometime during development.

Predicting Diagnostic Stability and Outcome: A Brief Overview

Age at Diagnosis. In addition to a diagnosis of PDD-NOS (discussed above), a number of factors have been found to be related to diagnostic stability in the toddler and early childhood years. Firstly, children diagnosed at younger ages appear to have less stable diagnoses over time as well as more positive outcomes. In a systematic review of the literature that included 23 studies of diagnostic stability, Woolfenden and colleagues (2012) found that diagnoses were least stable when made before age three years. Similarly, in two separate studies, Turner, Stone and colleagues (2006, 2007) found that children with the least stable diagnoses and the most positive outcomes were diagnosed before age three. Importantly, Turner and Stone (2006) note that this does not indicate that early diagnoses are inaccurate or that clinicians should wait to diagnosis children until later in toddlerhood. Rather, they explain that children diagnosed early appear to have the greatest likelihood of benefiting from early intervention, and thus, exhibit less stable diagnoses in a positive sense.

Early Cognitive and Language Abilities. Early cognitive and language abilities are associated with both diagnostic stability and later functioning more broadly. In terms of diagnostic stability, Lord and colleagues (2006) found that children with high verbal and nonverbal IQ at age two were most likely to change diagnostic status from AD to PDD-NOS or PDD-NOS to non-spectrum. Turner and Stone (2007) found that children who moved off the ASD spectrum had higher visual reception abilities and receptive language abilities (as assessed on the Mullen Scales of Early Learning) than children who remained on the spectrum. In terms of later functioning, Eaves and Ho (2004) found that children with uneven cognitive performance and higher non-verbal IQ at age 2.5 made the greatest gains in verbal abilities by age 4.5 years. In a review of longitudinal studies investigating outcomes in adolescence and adulthood, Levy and Perry (2011) found that individuals with higher cognitive functioning had more positive outcomes in the areas of independent living, education level and work.

Language abilities also appear to be predictive of diagnostic stability and outcome. In a treatment outcome study following 23 children matched on pre-treatment IQ, Sallows and Graupner (2005) found that children with the most positive post-treatment outcomes demonstrated stronger early language abilities than peers with less positive outcomes. Luyster and colleagues (2007) found that age three receptive and expressive language abilities predicted nonverbal IQ as well as overall autism symptoms as measured by the Autism Diagnostic Interview – Revised and the Autism Diagnostic Observation Schedule composite scores at age nine years. Similarly, in a longitudinal study investigating change in symptomatology between ages five and 15 years, Baghdadli et al. (2012) found that children with stronger early expressive language abilities showed steeper growth in social skills over time.

In terms of the relationship between language abilities and diagnostic stability, Turner and Stone (2007) found that children who moved off the spectrum had stronger receptive (but not expressive) language abilities at age two than their peers who remained on the spectrum. It is important to note that while some studies (e.g., Lord et al., 2006; Turner & Stone, 2007) found that cognitive and language abilities helped to predict unstable versus stable diagnoses, other studies did not find such differences (e.g., Chawarska et al., 2009). Therefore, while there exists substantial support for higher cognitive and language abilities predicting more positive outcomes broadly, evidence for the predictive utility of cognitive and language abilities in terms of diagnostic stability is mixed.

Symptom Severity. Symptom severity has also been found to be related to diagnostic stability and outcome. Turner and Stone (2007) found that children who moved off the autism spectrum between ages two and four had lesser overall symptom severity (particularly in the social interaction domain) at age two than their peers who remained on the spectrum. Lord et al. (2006) found that children with fewer or less severe restricted, repetitive behaviors were more likely to show diagnostic improvement (e.g., AD to PDD-NOS or PDD-NOS to non-spectrum). In terms of outcomes more broadly, Eaves and Ho (2004) found that children with the greatest gains in verbal skills had lesser early symptom severity. Further, Bopp and colleagues (2009) found that children with stronger social interaction skills showed greater gains over time in language comprehension and production. In addition, Baghdadli et al. (2012) found that children with lesser early symptom severity showed steeper growth in social skills over time. Therefore, it appears that across studies, as we may expect, lesser early symptom severity is predictive of more positive outcomes later in development, particularly in the domains of language abilities and social skills. Additional investigations of symptom severity in each separate domain (e.g.,

social interaction, communication, restricted repetitive behaviors) will be necessary to gain a more nuanced understanding of the role of symptom severity in diagnostic stability and outcome.

Additional Possible Predictors: Adaptive and Motor Skills, Imitation and Play.

Findings regarding other possible predictors of diagnostic stability and outcome are more mixed. Sutter and colleagues (2007) found that children with stronger early daily living and motor skills were the most likely to move off the ASD spectrum between ages two and four. Additionally, Turner and Stone (2007) found that fine motor abilities were predictive of movement off the autism spectrum, and Sallows and Graupner (2005) found that daily living skills predicted more positive outcomes. A number of other studies investigating diagnostic stability did not investigate adaptive skills or motor skills as possible predictors (e.g., Eaves & Ho, 2004; Lord et al., 2006), and therefore, these findings need replicating in additional samples.

Early imitation and play abilities have also been proposed as possible predictors of diagnostic stability and outcome. In a treatment outcome study following 23 children matched on pre-treatment IQ, Sallows and Graupner (2005) found that early verbal and nonverbal imitation abilities (as measured by the Early Learning Measure; Smith, Buch and Gamby, 2002) correlated with outcome measures of IQ, language and social skills. Turner and Stone (2006) found that all three children who moved off the spectrum between ages two and three demonstrated stronger motor imitation abilities than the average in their sample. Toth and colleagues (2006) found that immediate imitation abilities at approximately age 4 predicted group differences in communication abilities (as measured by the Vineland Adaptive Behavior Scales) at approximately age 6.5 years. Further, they found that deferred imitation and play abilities at age four predicted rates of growth in communication abilities between age four and 6.5 years. Specifically, they found that children with play and deferred imitation abilities 1 SD above the

mean of their peers demonstrated a growth rate in the communication domain of 13.5 months per one year of chronological age (Toth, Munson, Meltzoff, & Dawson, 2006). This rate of growth was similar to that seen in typically developing peers. Toth and colleagues (2006) discuss the important relationships between imitation and play skills, motor abilities and attention, such that imitation and play skills require a certain degree of social attention and motor planning. In turn, it is likely that rather than being isolated abilities, separate child-level predictors (e.g., language skills, imitation, play, motor skills) are strongly related and likely work together to promote more positive outcomes.

Intervention. In addition to child-level factors, intervention characteristics also play a role in diagnostic stability and outcome in the toddler years. Broadly, evidence indicates that early intervention can produce positive changes in cognitive abilities, adaptive skills, and autism symptoms, however, effects vary widely by study (Eldevik et al., 2009; Jónsdóttir et al., 2007; Lovaas, 1987; Rogers & Vismara, 2008; Sallows & Graupner, 2005). There is evidence that earlier (Harris & Handleman, 2000) and more intensive (Bryson, Rogers, & Fombonne, 2003) intervention is associated with more positive outcomes. Some studies do not find significant relationships between age at intake (Eldevik et al., 2009) and outcome; however, this may be a result of the restricted range of intake ages (i.e., all under seven years). Additionally, some studies (Jónsdóttir et al., 2007) do not find significant relationships between number of hours of intervention and outcome. Critically, however, as discussed in Helt et al. (2008) and others, the relationship between hours and outcome is unlikely to be linear given that lower functioning children (who may be less likely to make substantial progress) often receive more hours of intervention. (See Lord et al. (2005) for a review of issues regarding the evaluation of early intervention for ASD).

Outcomes Following the Loss of an ASD Diagnosis

Meeting Criteria for Another Diagnosis. A number of outcomes are possible for children who lose their ASD diagnosis in the toddler years or later in childhood. The majority of children who lose their ASD diagnosis appear to be diagnosed with another developmental disorder (e.g., Developmental Delay, Developmental Language Disorder). Kleinman et al. (2008) found that of the children who moved off the autism spectrum by age four, 60% moved to a non-ASD developmental disorder or other condition. Eaves and Ho (2004) found that 100% of children who moved off the spectrum by age 4.5 years had other developmental concerns (e.g., borderline cognitive functioning, dyspraxia, Attention-Deficit Hyperactivity Disorder, Language Disorder). Similarly, Turner and Stone (2007) found that 94.4% (17/18) of children who moved off the spectrum by age four years continued to have developmental concerns (e.g., Developmental Delay, Language Disorder).

Functioning in the Average Range. Of particular interest to the present study are the remaining children who lose their ASD diagnosis and appear to demonstrate more or less typical functioning. In the first documented report of average functioning following an ASD diagnosis, O. Ivar Lovaas (1987) found that 47% of his sample was functioning in the average range following intensive behavioral therapy. Importantly, however, Lovaas did not report whether individuals in his sample continued to meet criteria for an ASD following intervention. Relatively few studies have attempted to thoroughly characterize children who move off the spectrum (e.g., in terms of both cognitive abilities as well as remaining ASD symptoms), and therefore, it difficult to estimate the percentage of children who move off the spectrum and are functioning in the average range. In an extensive review of literature reporting on outcomes, Helt and colleagues (2008) determined that between 3 and 25% of children appear to lose their ASD

diagnosis sometime in development and demonstrate functioning in the average range cognitively, adaptively and socially.

A few studies have attempted to characterize these children who appear to demonstrate an “Optimal Outcome” from an early ASD diagnosis. “Optimal Outcome” has been defined as follows: the child must have met diagnostic criteria for an ASD following a gold standard diagnostic assessment, must no longer meet criteria for any ASD based on gold standard diagnostic assessment, must be participating in mainstream classrooms without the help of an aid, and must demonstrate a full scale IQ greater than 70 (Kelley, Naigles, & Fein, 2010). Kelley and colleagues (2010) compared 13 children who attained Optimal Outcome (OO) to 14 children who demonstrated typical functioning and to 14 children who were classified as having High Functioning Autism (HFA). At a mean age of 10.5 years, children who attained Optimal Outcomes demonstrated similar functioning to typically developing children in their adaptive skills and broad language abilities (Kelley et al., 2010).

Fein and colleagues (2013) compared a larger sample of 34 OO children, 44 children with HFA, and 34 children with typical development at a mean age of 13 years. Criteria for OO remained largely the same as that described in Kelley et al. (2010), with the exception of stricter criteria for average social and communication functioning (i.e., scores within 1.5 SD of the mean on the Vineland Adaptive Behavior Scales Socialization and Communication domains). In a thorough assessment of language abilities, facial recognition abilities, socialization, communication and ASD symptoms, they found average range functioning across measures for the OO group and very few differences between the OO and TD groups. In a more in depth analysis of language functioning in a subset of this sample, Kelley and colleagues (2006) found that the OO group demonstrated subtle deficits in pragmatic and semantic language when

compared to typically developing peers. Overall, it appears that children with Optimal Outcomes are functioning very similarly to their typically developing peers across domains, with very subtle deficits detectable on only the most fine-grained measures.

Predicting Optimal Progress

While a number of studies have investigated diagnostic stability in toddlerhood, and a few studies have attempted to characterize the most optimal outcomes (discussed above), relatively fewer studies have attempted to *predict* highly positive outcomes in the toddler years. Turner and Stone (2007) followed 48 children diagnosed with an ASD (38 AD, 10 PDD-NOS) at approximately age two years to follow-up at approximately age four years. As discussed above, they found that 62.5% of children remained on the spectrum at age four (stable group) and 37.5% moved off (change group). Turner and Stone (2007) found that at age two, the change group showed less severe overall autism symptoms and less severe symptoms in the social interaction domain, but that there were no group differences in communication or RRBs. Further, they found that the change group had stronger early cognitive abilities (visual reception), language abilities (receptive language) and motor skills (fine motor) as measured by the Mullen Scales of Early Learning (Turner & Stone, 2007). Additionally, they found that diagnoses made before 30 months predicted diagnostic change at age four. In terms of intervention characteristics, Turner and Stone (2007) found that the stable and change groups did not differ in the amount of services received (as per parent report). In sum, they found that earlier age at diagnosis, stronger cognitive abilities, and lesser early symptom severity predicted movement off the autism spectrum. Importantly, Turner and Stone (2007) note that all but one child in the change group continued to exhibit significant cognitive and/or language delays at age four, and therefore, these children cannot be considered to be functioning in the average range across domains.

Sutera and colleagues (2007) appear to be the first study to attempt to predict movement off the autism spectrum accompanied by cognitive and language functioning in the average range. Utilizing gold standard diagnostic procedures, 73 children were diagnosed with an ASD (55 AD, 18 PDD-NOS) at approximately age two and were followed-up at approximately age four. Sutera and colleagues (2007) found that 17.8% of children moved off the spectrum by age four and did not exhibit cognitive impairment. An additional four children had moved off the spectrum but demonstrated significant cognitive impairment, and therefore, were excluded from the study because of its focus on optimal functioning. Sutera and colleagues (2007) found that the group who moved off the spectrum was more likely to have been diagnosed with PDD-NOS and demonstrated stronger early fine motor and daily living skills. In contrast to Turner and Stone (2007), they found no group differences in early cognitive or language abilities. Based on their findings of relatively few group differences (e.g., fine motor, daily living skills), Sutera and colleagues (2007) conclude that movement off the autism spectrum is challenging to predict.

The Present Study

Given the state of the research on highly positive outcomes from ASDs in the toddler years, the current study seeks to address four critical questions. Firstly, given the high degree of variability in the rates of reported movement off the ASD spectrum (accompanied by average range functioning), the current study seeks to identify the percentage of children demonstrating optimal progress in a large prospective study of individuals initially diagnosed with an ASD at approximately age two and re-evaluated at approximately age four. The criteria for “Optimal Progress” used in the current study stems from established criteria for Optimal Outcome (see Helt et al., 2008) with some adjustments to reflect the developmental level of toddlers. Criteria for Optimal Outcome include requirements for close peer relationships as well as participation in

mainstream classrooms, both of which are criteria that are developmentally appropriate for school-age children, but not yet toddlers. In the current study, “Optimal Progress” is defined as follows: a child must have met criteria for an ASD using gold standard diagnostic procedures, must no longer meet criteria for any ASD at follow-up, and must demonstrate functioning in the average range (within 1.5 SD of the mean) on standardized measures of cognition, language, communication and social skills.

The second aim of the current study is to characterize early cognitive and behavioral differences between children who demonstrate Optimal Progress (OP) and those who remain on the spectrum (ASD-ASD). Specifically, we will investigate possible group differences in initial diagnosis (AD vs. PDD-NOS), cognitive abilities, language abilities, motor skills, adaptive skills and severity of ASD symptoms. Based on previous research, we hypothesize that children who demonstrate Optimal Progress will be more likely to have an initial diagnosis of PDD-NOS and will show stronger early cognitive, language, and motor skills than their peers who remain on the spectrum. Additionally, we hypothesize that children who demonstrate Optimal Progress will exhibit less severe ASD symptomatology at age two. The third aim of the current study is to characterize the pattern of growth between ages two and four in cognitive, language and adaptive skills for children who demonstrate Optimal Progress and those who remain on the spectrum. We hypothesize that in addition to early group differences in a number of domains, children who demonstrate Optimal Progress will show steeper growth across all domains than their peers who remain on the spectrum.

The fourth aim of the current study is to characterize possible group differences in the intervention received by children who demonstrate Optimal Progress and children who remain on the spectrum. This includes the age at which services were initiated, amount of weekly hours

of services received and the type of services received. Based on a retrospective study of older children who attain Optimal Outcomes (Orinstein et al., 2014), we hypothesize that an earlier age at service initiation and a greater number of hours of targeted early intervention will be seen in the Optimal Progress group when compared to their peers who remain on the spectrum. Further, we hypothesize that a greater percentage of children in the OP group will have received Applied Behavior Analysis (ABA) during their toddler years than their peers who remain on the spectrum. Through addressing these critical research questions, we hope to gain a more thorough understanding of Optimal Progress in the toddler years and the early characteristics of children who demonstrate this highly positive outcome.

Methods

Participants

Participants include a subset of individuals participating in an ongoing study to evaluate the psychometric properties of an autism-specific screening questionnaire, the Modified Checklist for Autism in Toddlers (M-CHAT, Robins, Fein, Barton, & Green, 2001) and a second-generation questionnaire, the M-CHAT-Revised/Follow-up (M-CHAT-R; Robins et al., 2014). Children were recruited for the study through four sources; receiving the screener at their 18 or 24 month pediatric well-child visit, receiving the screener from an early intervention provider, receiving the screener following referral from a psychologist, or receiving the screener following caregiver self-referral. Children who received the screener from their early intervention provider or following referral from a psychologist are considered to be at “High Risk” for ASD based on the presence of existing developmental concerns. Additionally, children who are younger siblings of children with a confirmed diagnosis of an ASD are considered to be at “High Risk” for ASD. Children who received the screener at their 18 or 24 month pediatric

well-child visit or following caregiver self-referral who had no prior developmental concerns are considered to be at “Low Risk” for ASD.

Following positive screening on the MCHAT or MCHAT-R/F, 311 children (see Figure 1) were evaluated at approximately 26 months (Time 1), and subsequently re-evaluated at an average age of 52 months (Time 2). These 311 children represent approximately 70% of all children who were evaluated at Time 1 following positive screening. Approximately 30% were lost to attrition before re-evaluation and therefore, will not be included in the current study.

Within the broader study seeking to validate the MCHAT or MCHAT-R/F, there is evidence that individuals who did not return for re-evaluation (e.g., who were lost to attrition) were more likely to be of non-White ethnicity and were less likely to have an advanced degree (e.g., Associate’s, Bachelor’s, Master’s etc.).

Of the 311 children evaluated at both time points, 209 children were diagnosed with an ASD at their initial evaluation and were considered for inclusion in the current study. Of these 209 children, 2 were excluded due to missing data regarding diagnostic status at re-evaluation. Of the 207 children diagnosed with an ASD at Time 1, 171 (82.6%) children retained an ASD diagnosis at re-evaluation (ASD-ASD). Nineteen children (9.2%) were determined to meet the previously discussed criteria for “Optimal Progress” (OP): at Time 1 the child met criteria for an ASD, and at Time 2 the child no longer met criteria for any ASD and were functioning in the average range (within 1.5 SD of the mean) on standardized measures of cognition, language, social and communication skills. The remaining 17 children moved from an ASD diagnosis at Time 1 to a different diagnosis (e.g., Developmental Delay) or had other developmental concerns at Time 2 and will not be considered in the majority of the subsequent analyses.

A total of 190 children, including 19 children demonstrating OP and 171 children who retained their ASD diagnosis (ASD-ASD), will be the focus of the current analyses. The majority of the current sample (62%) enrolled in the study after receiving the screener from an early intervention provider. 24% of the current sample enrolled in the study after receiving the screener at their 18 or 24 month pediatric well-child visits, 8% enrolled following caregivers self-referral and 6% were referred by a psychologist. 11.6% of the sample were younger siblings of children with confirmed ASD diagnoses (this includes children enrolling in the study through all of the aforementioned routes). In total, 72.6% ($n = 138$) children were at “High Risk” for ASD (based on existing developmental concerns or status as a younger sibling of a child with an ASD) and 27.4% ($n = 52$) were at “Low Risk” for ASD at the time of their referral. The two groups (OP, ASD-ASD) did not differ significantly in referral source as indicated by Fisher’s Exact Test, $p = .594$, nor did they differ significantly in risk status, $X^2(1) = 0.67$, $p = .754$.

The overall sample was 82% male ($n = 156$) and 18% female ($n = 34$) (See Table 1). This ratio (4.6:1) reflects the currently estimated gender ratio in the wider population of children with ASD (4.6:1) (CDC, 2012). The percentage of males and females did not differ based on group (OP vs. ASD-ASD), $X^2(1) = 2.69$, $p = .101$. The majority of children in the sample were White ($n = 155$, 81.5%), as indicated by their caregivers. 6.3% ($n = 12$) of children were Hispanic/Latino, 3.6% ($n = 5$) of the sample was Asian or Pacific Islander, and 2.1% ($n = 4$) were biracial. One caregiver indicated that their child’s race/ethnicity did not fall into any of the aforementioned categories (i.e., “other”). Race/ethnicity information was not available for the remaining 6 children. The two groups (OP, ASD-ASD) did not differ significantly in race/ethnicity as indicated by Fisher’s Exact Test, $p = .764$ (See Table 1). The two groups also did not differ significantly in maternal education (Fisher’s Exact Test, $p = .719$); however,

information regarding maternal education was missing for a large number of participants (See Table 2).

At the initial evaluation (Time 1), the OP group was on average 26.21 months ($SD = 4.81$) and the ASD-ASD group was 26.32 months of age ($SD = 4.37$) (See Table 1). The two groups did not significantly differ in age at initial evaluation, $t(187) = .100$, $p = .921$, $d = .02$. At re-evaluation (Time 2), on average, the OP group was 51.47 months of age ($SD = 7.23$) and the ASD-ASD group was 52.30 ($SD = 9.52$) months of age. The two groups did not significantly differ in age at re-evaluation, $t(188) = .361$, $p = .718$, $d = .11$.

Procedure

Children's caregivers were provided the M-CHAT ($n = 176$) or M-CHAT-R/F ($n = 14$) autism-specific screening measures to complete at their pediatrician's office during their child's 18 or 24-month well-child visit, at an early intervention site, or in their home. Once the questionnaire was completed, it was sent to the University of Connecticut Early Detection laboratory to be scored. If a caregiver's responses indicated that a child failed the screener, they were contacted via telephone to confirm failed items. If a child was confirmed to have failed the screener during the follow-up phone interview, he or she was invited to attend a free developmental and diagnostic evaluation conducted at the University of Connecticut. If a family was unable to attend an evaluation due to transportation difficulties, a number of options were available including providing transportation (i.e., cab service), or conducting the evaluation at the family's pediatrician's office.

A licensed clinical psychologist or a developmental pediatrician and a graduate student in the Clinical Psychology program at the University conducted the evaluations, which consisted of measures of cognitive skills, adaptive skills, language abilities and ASD-specific measures. At

the conclusion of the evaluation, caregivers were provided with feedback regarding the assessment, which included any diagnoses the child may qualify for as well as primary recommendations. Six to eight weeks after the evaluation, caregivers received a written report detailing the results of the assessment.

A diagnosis of an ASD was assigned based on clinical judgment of experienced clinicians (licensed psychologists or developmental pediatricians), utilizing scores from the Toddler Autism Symptom Interview (TASI) or Autism Diagnostic Interview (ADI(-R)), Autism Diagnostic Observation Schedule (ADOS), Childhood Autism Rating Scale (CARS), Mullen Scales of Early Learning (Mullen) and Vineland Adaptive Behavior Scales (VABS), and in accordance with DSM-IV diagnostic criteria. ASD diagnoses included a diagnosis of AD, PDD-NOS or Asperger's Disorder. An additional diagnostic category, ASD – Low Mental Age (ASD-Low MA) was given to children who met DSM-IV diagnostic criteria for AD or PDD-NOS and were functioning below the 12 month level across all domains on the Mullen. Clinical judgment in the assignment of ASDs has been shown to have high inter-rater reliability and is considered best practice in the field of ASDs (Klin, Lang, Cicchetti, & Volkmar, 2000).

All children who were evaluated at approximately 24 months (Time 1) were invited for a second evaluation around their 4th birthday (approximately age 48 months, Time 2). For children who were screened using the M-CHAT, Time 2 procedures replicated those performed at Time 1. For children who were screened using the M-CHAT-R/F, Time 1 evaluations included the TASI, whereas Time 2 evaluations included the ADI(-R). This is due to the development of the TASI as a novel measure of autism symptomatology in toddlers during the same period as the development of the M-CHAT-R/F.

Measures

The following measures were utilized in the ongoing study: M-CHAT, M-CHAT-R/F, ADI(-R), TASI, Mullen, VABS, and CARS. These measures have been determined to have excellent psychometric properties and are widely used in the field of ASDs, with the exception of the TASI, which is currently being validated. Additionally, the current study utilized a questionnaire to determine characteristics of the interventions received by children between ages one and five years. Three versions of the questionnaire were utilized (parent report, early intervention provider report, preschool provider report). The current study analyzes data from the measures described below.

Modified Checklist for Autism in Toddlers (M-CHAT)/ M-CHAT-Revised (M-CHAT-R/F). The Modified Checklist for Autism in Toddlers (M-CHAT; Robins et al., 2001) is a brief, autism-specific, parent-report screening measure. The checklist includes 23, yes/no items, six of which are considered “critical items” because they were found to be the best discriminators of children diagnosed with an ASD (Robins et al., 2001). Items include presence of pointing, interest in other children, imitation and play. Internal consistency reliability for the entire screener ($\alpha = .85$) and the six critical items ($\alpha = .83-.84$) was found to be adequate (Robins et al., 2001; Kleinman et al., 2008). To fail the screener (screen positive), children must fail any three questions or any two of the six critical items. A follow-up telephone interview is conducted for all children who fail the screener to verify failed items. Studies have indicated that the screener, in combination with the follow-up phone interview, has an estimated sensitivity of .91 to .97, an estimated specificity of .99, and a positive predictive value (PPV) of .68 to .74 (Robins et al., 2001; Kleinman et al., 2008).

The M-CHAT-R/F is a revised version of the M-CHAT created in 2009 that includes 20 yes/no items, seven of which are considered critical items. A follow-up phone interview is conducted if a child fails three of the 20 items or any two critical items. Children are determined to screen positive if they fail any two items after the follow-up phone interview. Internal consistency reliability for the screener with follow-up was found to be adequate ($\alpha = .79$) (Robins et al., 2014). The revised screener with follow-up was found to have an estimated sensitivity of .94 to .97, an estimated specificity of .83, and a positive predictive value (PPV) of .95 for any developmental delay or concern (Robins et al., 2014).

Autism Diagnostic Observation Schedule - Generic (ADOS). The Autism Diagnostic Observation Schedule (ADOS; Lord et al., 2000) is a semi-structured, standardized, play-based assessment of four areas: Reciprocal Social Interaction, Communication, Stereotyped Behaviors and Restricted Interests and Play, which is intended for use with children who are suspected to have an ASD. Importantly, the ADOS is not meant as a stand-alone measure, but rather, as a component of a battery of measures to assess for ASD. Children are administered one of four possible modules, which are selected based on a child's language level. Module 1 is administered to children with single words or simple phrases, Module 2 is administered to children with flexible phrase speech, Module 3 is administered to children/adolescents with fluent speech, and Module 4 is administered to adolescents/adults with fluent speech. Modules 1 and 2 were used in the current study. Each module consists of a series of unstructured and structured situations or activities that provide a hierarchy of presses for the behaviors of interest.

The ADOS is scored for individual domains (listed above), with higher scores indicating greater severity. Internal consistency reliability was found to be adequate or high for all domains in all modules (Social domain, $\alpha = .86-.91$; Communication domain, $\alpha = .74-.84$; Stereotyped

Behaviors and Restricted Interests, $\alpha = .47-.65$, Play, unreported) (Lord et al., 2000). Scores on the Social and Communication domains are combined, and a cutoff score is applied to determine whether a child falls into the following classifications: *no autism spectrum disorder*, *autism spectrum disorder*, or *autistic disorder*. Internal consistency reliability for the Social-Communication totals was found to be high ($\alpha = .91 - .94$) (Lord et al., 2000).

The inter-rater reliability (mean weighted kappas, $M\kappa_W$) of Modules 1 and 2 was found to be high, $M\kappa_W = .78$ and $M\kappa_W = .70$, respectively (Lord et al., 2000). Further, inter-rater agreement of classification (*autistic disorder* versus *non-spectrum*) was found to be 100% for Module 1 and 91% for Module 2. Sensitivity (when classifying AD and PDD-NOS versus non-spectrum) was found to be .97 for Module 1 and .95 for Module 2. Specificity (when classifying AD and PDD-NOS versus non-spectrum) was found to be .94 for Module 1 and .87 for Module 2 (Lord, 2000).

In 2009, Gotham and colleagues developed the ADOS Calibrated Severity Score (CSS) in order to assess symptom severity based on ADOS scores. The CSS is a measure of autism severity that takes into account a child's age and language abilities, allowing for a measure of symptom severity that is less influenced by age or verbal abilities (Gotham et al., 2009). As such, the CSS can be compared across ADOS modules. The CSS is computed from ADOS "raw total scores," which include children's scores on two domains devised by Gotham et al. (2007), *Social Affect* (SA, includes items from the Reciprocal Social Interaction, and Communication domains) and *Restricted, Repetitive Behaviors* (RRB, includes items from the Stereotyped Behaviors and Restricted Interests domain). The ADOS was administered at Time 1 and Time 2.

Vineland Adaptive Behavior Scales – Interview Edition (Versions I and II). The Vineland Adaptive Behavior Scales (VABS; Sparrow, Balla, & Cicchetti, 1984) is a structured,

parent-report interview measure of adaptive functioning across four domains: Communication, Daily Living Skills, Socialization and Motor Skills. Scores are determined for each domain individually, and are combined to form a total score, the Adaptive Behavior Composite (ABC). In the current study, children's caregivers were administered the VABS (Sparrow, Balla, & Cicchetti, 1984) or the Vineland Adaptive Behavior Scales – Second Edition (VABS-II), an updated version which was released in 2005 (Sparrow, Cicchetti, & Balla, 2005). For children aged two to five, the VABS has been found to have high internal consistency (split-half reliability) across all domains (.74 – .94), with the lowest split-half reliability for Motor Skills (Sparrow, Balla, & Cicchetti, 1984). The VABS has also been shown to have strong criterion-related validity when compared to other measures of adaptive functioning (Sparrow, Balla, & Cicchetti, 1984).

Changes between the VABS and the VABS-II (Sparrow, Cicchetti, & Balla, 2005) include an expansion of the age range, and the addition of new items to each domain, with a specific focus on increasing item density in the zero to three age range. For children aged two to five, the VABS-II has been shown to have similar internal consistency to the VABS across all domains (.79 to .95), and similarly strong criterion-related validity (Sparrow, Cicchetti, & Balla, 2005). For children with probable ASD, the VABS is considered to be a critical component of a developmental and diagnostic assessment and to be useful in providing additional information for treatment planning and family support (Perry et al., 2009). The current study collected data on this measure for children at Time 1 and Time 2, and, by convention, analyzed VABS and VABS-II scores collectively.

Mullen Scales of Early Learning. The Mullen Scales of Early Learning (Mullen; Mullen, 1995) reflects five domains of cognitive development. These include Visual Reception

(problem solving abilities), Gross Motor, Fine Motor, Expressive Language and Receptive Language. In addition to T-scores, percentile ranks and age-equivalents for each domain, the measure provides a summative “Early Learning Composite” (ELC) score, which is computed from the Visual Reception, Fine Motor, Expressive Language and Receptive Language domains. In the current study, the Gross Motor domain was not administered given that it is only available for children under 33 months (Mullen, 1995). Median internal consistency reliability (modified split-half reliability using Rasch item response theory model) was found to be .75 to .83 for all domains and the ELC (Mullen, 1995). Test-retest reliability over a period of approximately one to two weeks was found to be .84 for children one to 24 months and .76 for children 25 to 56 months for the cognitive scales (Visual Reception, Fine Motor, Expressive Language and Receptive Language) (Mullen, 1995). In terms of concurrent validity, the Mullen has been found to be highly correlated with other measures such as the Bayley Scales of Infant Development (BSID; Bayley, 1969). Specifically, the Mullen cognitive scales and the Bayley Mental Development Index showed correlations ranging from .53 to .59 (Mullen, 1995). The Mullen was administered at Time 1 and Time 2.

Childhood Autism Rating Scale (CARS). The CARS (Schopler, 1980) is a 15-item observation-based rating scale designed to accurately differentiate children with autism from those with developmental delays without features of autism. Each item reflects a sub-domain, examples of which include “Relating to people,” “Imitation,” “Emotional response,” “Adaptation to change,” “Verbal communication,” and “Nonverbal communication.” Each item/sub-domain is rated on a seven-point scale (1, 1.5, 2...4) ranging from “within normal limits for that age” to “severely abnormal for that age” (Schopler et al., 1995). To interpret the CARS, a total score is determined by summing the ratings on all 15 items, with total CARS scores range from a low of

15 to a high of 60. Children can be classified being *non-autistic*, having *mild autism* or having *severe autism* based on established cutoff scores (Schopler et al., 1995). In order to better reflect our more current understanding of autism as a spectrum, Chlebowski et al. (2010) recommend a cutoff of 25.5 be used to distinguish an ASD from a non-ASD for two year olds and four year olds.

Internal consistency reliability for the CARS total score has been found to be high ($\alpha = .90$ to $.94$) (Schopler et al., 1995; Chlebowski et al., 2010). In the current sample, the internal consistency reliability was found to be $.84$ at Time 1 and $.90$ at Time 2. Interrater reliability on the CARS ranges from $.71$ to $.94$ (Schopler et al., 1995; Chlebowski et al., 2010). The validity of the CARS has been assessed by comparing its classification of cases to the classifications made by other frequently used measures. Saemundsen et al. (2003) found a correlation of $.67$ between the CARS and the ADI-R. The sensitivity and specificity of the CARS have been found to be high ($.94$ and $.85$, respectively) (Perry et al., 2005). In order to better understand domains within the CARS total score, Magyar and Pandolfi (2007) conducted a factor structure evaluation of the CARS using Principal Axis Factor Analysis (PAF) and found four factors, which accounted for 41.67% of the variance. These include *Social Communication*, *Social Interaction*, *Stereotypies and Sensory Abnormalities*, and *Emotional Regulation*. The CARS was administered at Time 1 and Time 2.

Early Intervention Questionnaires. A questionnaire was developed in three forms (parent-report, early intervention provider-report, preschool provider-report) to characterize the early intervention services received by children between the ages of one and five years. This age span was selected to ensure that the period between the Time 1 and Time 2 evaluations was covered by the questionnaire. The parent-report version of the questionnaire includes questions

regarding parents' experiences with their child's early intervention and preschool providers (e.g., "What did you find most helpful about your child's preschool services?" "What would you have liked to change about your child's preschool services?"). The early intervention provider-report version includes questions regarding the amount, type and dates of services received in 6-month increments between ages one year and three years. The preschool provider-report version includes questions about the amount, type and dates of services received in 6-month increments between ages three and five years. Please see Appendix C for copies of these questionnaires.

Results

Time 1 and Time 2 Diagnoses: Entire Sample (n = 207)

The following analyses include all 207 children diagnosed with an ASD at Time 1 and re-evaluated at Time 2. Please see Table 3 for sample diagnoses at Time 1 and Time 2. In summary, we found an overall stability of ASD diagnosis between Time 1 and Time 2 of 82.6% (n = 171). These children form our ASD-ASD group. The remaining 17.4% of children no longer met criteria for an ASD (n = 36) at Time 2. These children either met criteria for other diagnoses (e.g., Developmental Delay, Developmental Language Disorder, Motor, Regulatory Issues), were given no diagnosis (indicating that they did not meet symptom criteria for any DSM-IV diagnosis, but were not fully typically developing) or were determined to be typically developing (See Table 4). Of the 36 children who no longer met criteria for an ASD, 19 met the aforementioned criteria for Optimal Progress and form our Optimal Progress (OP) group. This represents 9.2% of children diagnosed with an ASD at Time 1 in our sample.

Diagnostic Stability: AD, PDD-NOS and ASD-Low MA

Of the 108 children initially diagnosed with AD, 72 (66.7%) retained a specific diagnosis of AD at Time 2, 22 (20.3%) showed diagnostic improvement and received a diagnosis of PDD-

NOS at Time 2, and 14 (13%) no longer met criteria for any ASD (See Table 5). Of these 14 children, 8 met criteria for Optimal Progress. This represents 7.4% of children initially diagnosed with AD in our sample. Of the 79 children initially diagnosed with PDD-NOS, 31 (39.2%) retained a specific diagnosis of PDD-NOS at Time 2, 26 (32.9%) showed a worsening of symptomatology and received a diagnosis of AD, and 22 (27.8%) no longer met criteria for any ASD (See Table 6). Of these 22 children, 11 met criteria for Optimal Progress. This represents 13.9% of children initially diagnosed with PDD-NOS in our sample. Of the 20 children initially diagnosed with ASD - Low MA, 100% of children retained an ASD diagnosis at Time 2 (See Table 7). Notably, 80% showed improvement in cognitive abilities such that their mental age equivalents rose above 12 months.

Diagnostic Predictors of Optimal Progress: Time 1 Diagnoses of OP and ASD-ASD Groups

The remaining analyses will focus on the OP ($n = 19$) and ASD-ASD ($n = 171$) groups, and therefore, do not include the 17 children who lost their ASD diagnosis but did not meet OP criteria. There was a strong trend ($X^2(2) = 5.63, p = .06$) such that the OP and ASD-ASD groups differed in Time 1 diagnosis (see Table 8). Children initially diagnosed with PDD-NOS were the most likely to meet criteria for Optimal Progress at Time 2 (16.2%), followed by children initially diagnosed with AD (7.8%). As noted above, no children initially diagnosed with ASD-Low MA met criteria for an Optimal Progress at Time 2.

Diagnostic Predictors of Optimal Progress: Time 1 DSM-IV Symptomatology

To further understand potential diagnostic differences between the OP and ASD-ASD groups, Time 1 DSM-IV symptoms were analyzed. DSM-IV total scores include symptoms across three domains: Social Interaction, Communication, Restricted Interests and Repetitive Behaviors, and reflect the total number of symptoms out of a possible 12. DSM-IV diagnostic

information was available for 17 OP and 156 ASD-ASD children, and therefore, the subsequent analyses include 173 children. The OP group showed fewer total DSM-IV symptoms at Time 1 ($M=5.00$, $SD=1.87$) than the ASD-ASD group ($M=6.04$, $SD=1.75$), $t(171)=2.33$, $p = .021$, $d=.57$ (see Figure 2). In order to better understand group diagnostic differences, each domain of symptomatology was separately investigated. The OP group showed significantly fewer symptoms in the Restricted Interests and Repetitive Behaviors domain ($M=0.94$, $SD=0.66$) than the ASD-ASD group ($M=1.43$, $SD=1.00$), $t(24)=2.73$, $p =.011$, $d=.57$ (see Figure 3). The OP and ASD-ASD groups did not significantly differ in number of symptoms in the Social Interaction domain, $t(171) = .97$, $p = .333$, $d = .24$ (see Figure 4), nor did they differ in number of symptoms in the Communication domain, $t(17) = 1.57$, $p = .135$, $d = .45$ (see Figure 5).

Diagnostic Predictors of Optimal Progress: Time 1 Symptom Severity

Overall symptom severity at Time 1 was measured using the Childhood Autism Rating Scale (CARS) total score. CARS scores were available for all 19 OP children and 164 ASD-ASD children, and therefore, 183 children are included in this analysis. Higher scores indicate greater symptom severity. The OP group showed significantly lesser symptom severity ($M=28.68$, $SD=5.57$) at Time 1 than the ASD-ASD group ($M=33.02$, $SD=5.20$), $t(181)=3.41$, $p = .001$, $d=.80$ (see Figure 6).

In order to better understand in which specific domains OP children showed lesser symptom severity, analyses were conducted for the following factors: Social Communication, Social Interaction, Stereotypies and Sensory Abnormalities, and Emotional Regulation (Magyar and Pandolfi, 2007; See Figure 7). Independent groups t -tests indicate that in the Social Communication domain the OP group showed significantly lower Time 1 symptom severity ($M = 2.12$, $SD = 0.47$) than the ASD-ASD group ($M = 2.56$, $SD = 0.47$), $t(181) = 3.98$, $p <.001$,

$d=.98$. Additionally, the OP group showed significantly lesser Time 1 symptom severity ($M = 1.85$, $SD = 0.36$) than the ASD-ASD group ($M = 2.10$, $SD = 0.45$) in the domain of Stereotypies and Sensory Abnormalities, $t(181) = 2.29$, $p = .023$, $d = .61$. The two groups did not differ in symptom severity in the Social Interaction domain, $t(181) = 1.52$, $p = .131$, $d = .37$, nor did they differ in the Emotional Regulation domain, $t(181)=1.31$, $p = .191$, $d = .32$ (see Figure 7).

Severity of Time 1 autism symptomatology was also measured utilizing the ADOS calibrated severity score (CSS) computed from participant's scores on the ADOS, as per the procedure outlined by Gotham et al. (2009) (described above). Necessary scores were available for 15 OP and 143 ASD-ASD participants, and therefore, 158 children are included in the following analysis. Independent groups t -tests indicate that the OP and ASD-ASD groups did not differ on this measure of autism symptom severity at Time 1, $t(156) = .592$, $p = .55$, $d = .14$ (see Figure 8).

Predictors of Optimal Progress: Time 1 Cognitive Abilities

Cognitive abilities were assessed using the Mullen Scales of Early Learning. Mullen scores were available for 15-16 OP and 126-135 ASD-ASD children (depending on domain), and therefore, between 142 and 150 children were included in each of the subsequent analyses. Preliminary analyses indicated that the assumption of normality was violated in that Time 1 Mullen T-scores were not normally distributed in our sample. This appeared to be due to a large number of children receiving the lowest possible T-score (20). In order to address these "floor effects," estimated IQ scores were calculated for each domain of the Mullen for each participant. Ratio IQ scores were calculated using the following formula: mental age / chronological age x 100. In order to assure the appropriateness of using these estimated IQ scores in place of T-scores, ratio IQ scores were correlated with T-scores. These correlations were all found to be

significant at the .01 level, ranging from .52 to .86 (see Table 9), indicating that estimated IQ scores were highly representative of T-scores. There were no significant group differences in Time 1 estimated IQ scores for any domain of the Mullen; Visual Reception ($t(149) = 0.87, p = .386, d = .26$), Fine Motor ($t(149) = 1.30, p = .195, d = .40$), Expressive Language ($t(148) = 1.65, p = .101, d = .46$), Receptive Language ($t(150) = 1.60, p = .112, d = .49$) (see Figure 9).

Predictors of Optimal Progress: Time 1 Adaptive Skills

Adaptive skills were assessed using the Vineland Adaptive Behavior Scales (VABS), version I or II (described above). 171 children received the VABS and 16 children received the VABS-II. Based on the strong correlations seen between the VABS-I and VABS-II, as well as their similar overall psychometric properties (Sparrow, Cicchetti, & Balla, 2005), VABS-I and VABS-II scores were analyzed collectively. VABS scores were available for all 19 OP children and 164-167 ASD-ASD children depending on domain, and therefore, between 183 and 186 children are included in each of the subsequent analyses. The OP group showed significantly stronger overall adaptive abilities, as indicated by the VABS total score ($M = 72.00, SD = 7.39$) than the ASD-ASD group ($M = 66.21, SD = 7.67$), $t(181) = 3.12, p = .002, d = .79$ (see Figure 10).

Investigating each domain individually revealed that the OP group showed significantly stronger abilities across all domains of adaptive skills. The OP group showed significantly stronger Time 1 Communication skills ($M=72.00, SD=9.58$) than the ASD-ASD group ($M = 66.82, SD = 8.55$) ($t(184) = 2.47, p = .014, d = .57$) and stronger Time 1 Social Skills ($M = 73.42, SD = 8.22$ vs. $M = 68.88, SD = 8.47$) ($t(184) = 2.22, p = .028, d = .54$). Additionally, the OP group showed significantly stronger Daily Living skills ($M = 75.53, SD = 10.56$) than the ASD-ASD group ($M=69.52, SD=9.23$) ($t(184)=2.65, p=.009, d=.66$), as well as stronger Motor

skills ($M = 86.58$, $SD = 10.43$ vs. $M = 80.80$, $SD = 11.68$) ($t(184) = 2.07$, $p = .040$, $d = .52$). See Figure 11.

Summary of Child-Level Predictors of Optimal Progress

In summary, Optimal Progress appears to be predicted by a number of child-level characteristics at age two (Time 1). In regards to diagnostic predictors, we found a strong trend such that children initially diagnosed with PDD-NOS are more likely to demonstrate OP than children initially diagnosed with AD. Further, we found that children who later demonstrated OP showed fewer DSM-IV symptoms in the Restricted Interests, Repetitive Behaviors domain, but not in the Communication or Social Interactions domains. Additionally, we found lesser early symptom severity in the domains of Social Communication and Stereotypies and Sensory Abnormalities, but not in the Social Interaction or Emotional Regulation domains (as measured by the CARS). In regards to cognitive abilities, we found no significant group differences between the OP and ASD-ASD groups at Time 1. In regards to adaptive skills, we found stronger early abilities across domains (Communication, Socialization, Daily Living, Motor).

Pattern of Growth Between Time 1 and Time 2: Cognitive Abilities

A repeated measures ANOVA was conducted to analyze possible group differences in the pattern of change in cognitive abilities between Time 1 and Time 2. The number of months between evaluations was used as a covariate. In each of the described analyses, multivariate tests were utilized given that the assumption of sphericity was violated. Additionally, as noted above, estimated IQ scores were used in place of T-scores as a result of the non-normality of the T-score distribution. Mullen scores at both time points were available for 12-13 OP children and 122-127 ASD-ASD children, and therefore, 125-140 children were included in each of the subsequent

analyses. Analyses were conducted for each domain of the Mullen (Visual Reception, Expressive Language, Receptive Language, Fine Motor) separately.

In regards to Visual Reception, there was a significant interaction between Time (1, 2) and Group (OP, ASD-ASD), Wilks' Lambda = .862, $F(1,140) = 22.43$, $p < .001$, partial $\eta^2 = .138$. As Figure 12 indicates, the OP group showed a significantly steeper pattern of growth in Visual Reception abilities over time than did the ASD-ASD group. This significant interaction held when initial autism severity (CARS Time 1 total score) was used as an additional covariate, indicating that the OP group showed a steeper pattern of growth over time, even when controlling for initial differences in autism symptom severity (see Table 10).

A significant Time (1, 2) by Group (OO, ASD-ASD) interaction was also found for Expressive Language abilities, Wilks' Lambda = .895, $F(1,138) = 16.25$, $p < .001$, partial $\eta^2 = .105$, indicating steeper growth in the OP group (Figure 13). This significant interaction held when initial autism severity (CARS Time 1 total score) was covaried (see Table 10). The same pattern of results (significant Time by Group interactions with steeper growth in the OP group) was found for Receptive Language abilities as well as Fine Motor skills, with (Table 10) and without (see Figures 14 and 15) initial autism severity as a covariate.

In order to characterize the magnitude of growth, average increases in standard scores (using standard deviation units) were calculated for each group. In regards to Visual Reception abilities, the OP group demonstrated an average increase of 2.7 standard deviations in standard scores, in contrast to the ASD-ASD group who showed an average increase of 0.44 standard deviations. In terms of language abilities (i.e., Mullen Expressive Language, Receptive Language), the OP group demonstrated an average increase of 2.01 standard deviations, in comparison to an average increase of .52 standard deviations for the ASD-ASD group. In terms

of fine motor skills, the OP group demonstrated an average increase of 1.7 standard deviation units, while the ASD-ASD group who demonstrated an average increase of .22 standard deviations.

Pattern of Growth Between Time 1 and Time 2: Adaptive Skills

A repeated measures ANOVA was conducted to analyze possible group differences in the pattern of change in adaptive skills between Time 1 and Time 2. Again, the number of months between evaluations was used as a covariate. For all analyses described below, multivariate tests were utilized given that the assumption of sphericity was violated. VABS scores at both time points were available for 18-19 OP children and 155-159 ASD-ASD children, and therefore, 173-178 children were included in each of the subsequent analyses. In regards to overall adaptive abilities as measured by the VABS total score, there was a significant interaction between Time (1, 2) and Group (OP, ASD-ASD), Wilk's Lambda = .686, $F(1, 170) = 77.67$, $p < .001$, partial $\eta^2 = .314$. As Figure 16 reveals, the OP group showed a significantly steeper pattern of growth in adaptive skills over time than did the ASD-ASD group. This interaction remained significant when initial autism severity (as measured by the CARS Time 1 total score) was used as an additional covariate, indicating that the OO group showed a steeper pattern of growth over time, even when controlling for initial differences in autism symptom severity (see Table 11).

Additional analyses were conducted for each domain of the VABS. In regards to Communication skills, there was a significant interaction between Time (1, 2) and group (OO, ASD-ASD), Wilk's Lambda = .763, $F(1, 175) = 54.48$, $p < .001$, partial $\eta^2 = .237$. Similarly to above, the OP group showed a significantly steeper pattern of growth over time in Communication skills than did the ASD-ASD group (see Figure 17). This significant interaction held when initial autism severity (CARS Time 1 total score) was covaried (see Table 11).

Significant Time (1, 2) x group (OO, ASD-ASD) interactions were also found for Social skills, Daily Living Skills, and Motor Skills with (Table 11) and without (Figures 17 through 20) initial autism severity as a covariate.

In terms of magnitude of change in adaptive skills (VABS Communication, Socialization, Daily Living), the OP group demonstrated an average increase of 1.32 standard deviations in standard scores, in comparison to the ASD-ASD who showed an average overall decrease in these adaptive skills. In terms of motor skills, the OP group demonstrated an average increase of 0.50 standard deviation units, while the ASD-ASD group demonstrated an average decrease of .76 standard deviations.

Summary of Patterns of Growth in Cognitive and Adaptive Skills

In sum, we found that across all domains on the Mullen (Visual Reception, Expressive Language, Receptive Language, Fine Motor) and VABS (Communication, Socialization, Daily Living, Motor) the OP group showed a significantly steeper pattern of growth between Time 1 and Time 2 than did their ASD-ASD peers. Across all measured domains of cognitive and adaptive abilities, this steeper growth trajectory was found both when initial autism symptom severity was covaried and when it was not.

Predictors of Optimal Progress: Early Intervention Characteristics

Data collection for the fourth aim of this study, characterizing possible group differences in early intervention, is in progress and has not yet been completed. Logistical challenges in contacting families and locating previous early intervention and preschool providers have caused data collection to proceed past the expected timeframe. This fourth aim is considered to be of great importance to furthering our understanding of Optimal Progress, and therefore, will continue to be pursued and will be presented in a later project.

Summary of Functioning at Time 2

At Time 2, the OP and ASD-ASD groups differed across all measured domains of functioning. This is as we would expect given that, in many cases, this is how the groups were defined. The magnitude of group differences, however, is of interest to our understanding of the clinical significance of the outcome of “Optimal Progress.” In regards to DSM-IV symptoms, the OP group demonstrated significantly fewer total symptoms ($M = 1.44$, $SD = 1.21$) at Time 2 than did their ASD-ASD peers ($M = 6.35$, $SD = 1.86$), $t(176) = 10.35$, $p < .001$, $d = .312$ (see Figures 2-5). Further, the OP group showed significantly lesser ASD symptom severity (7 points below threshold for ASD) as measured by the CARS ($M = 18.67$, $SD = 1.92$) than their ASD-ASD peers ($M = 31.61$, $SD = 5.86$), $t(63) = 20.12$, $p < .001$, $d = 2.97$ (see Figure 6). They also showed lesser symptom severity as measured by the ADOS CSS, $t(41) = 18.76$, $p < .001$, $d = 3.17$ (see Figure 8).

In regards to cognitive, language and motor abilities as assessed by the Mullen, the OP group showed significantly stronger abilities in each domain (Visual Reception, Fine Motor, Expressive Language, Receptive Language) when compared to their peers (as measured by estimated IQ scores, see Table 12). The OP group demonstrated an average ELC standard score of 102.36 ($SD = 11.39$), representing performance 2.32 standard deviations above their ASD-ASD peers. The greatest magnitude of group difference was found in receptive language abilities ($d = 1.86$), with the smallest, but still significant, group difference in fine motor abilities ($d = 1.67$) (see Table 12). In regards to adaptive skills as measured by the VABS, the OP group showed stronger overall adaptive skills ($M = 91.76$, $SD = 11.43$) than their ASD-ASD peers ($M = 61.59$, $SD = 13.54$), $t(176) = 9.06$, $p < .001$, $d = 2.40$. The OP group demonstrated stronger

adaptive skills in each domain individually, with the greatest magnitude of group difference in social skills ($d = 2.53$) (see Table 13).

Discussion

Diagnostic Stability

The results of the current study support the findings of previous studies investigating diagnostic stability of ASDs in the toddler years. As in previous work, the current study found that, broadly, diagnostic stability is high, with 82.6% of children retaining an ASD diagnosis between ages two and four. This finding is within the range of previously reported stability rates of ASD in toddlers of between 68 and 100% (Chawarska et al., 2009; Eaves & Ho, 2004; Kleinman, Robins, et al., 2008; Lord, 1995; Sutera et al., 2007; Turner & Stone, 2007). Our findings support continued efforts to diagnosis ASDs early in development, as they appear to be stable following diagnoses made at approximately age two years.

The results of the current study also support previous studies reporting that the diagnostic stability of AD is higher than the diagnostic stability of PDD-NOS (Chawarska et al., 2009; Eaves & Ho, 2004; Kleinman, et al., 2008a; Sutera et al., 2007; Turner & Stone, 2007). As in previous studies, children diagnosed with AD were less likely than their peers with PDD-NOS to lose their ASD diagnosis in toddlerhood. Further, children diagnosed with PDD-NOS were more likely than children diagnosed with AD to change diagnostic classification within the ASD spectrum. Differences in diagnostic stability of AD and PDD-NOS are likely due to the greater number and severity of symptoms seen in children with an AD diagnosis in comparison to peers with PDD-NOS. Understanding these differences in diagnostic stability will help clinicians to best inform parents of expected outcomes following particular ASD diagnoses. Despite changes in DSM criteria that have eliminated specific diagnoses (e.g., of AD or PDD-NOS), the current

findings remain informative for understanding differences in diagnostic trajectories for children with more or less severe symptomatology.

The current study also investigated diagnostic stability of ASD diagnoses in children with severe cognitive delays (e.g., age equivalents below 12 months with chronological age of approximately 24 months). Results indicate that children with severe cognitive delays show highly stable diagnoses over time (100% in the current sample), despite cognitive improvement in the large majority of these children. This supports the early diagnosis of ASD in children who exhibit ASD symptoms accompanied by severe cognitive delays, rather than waiting to diagnosis these children until their cognitive abilities rise above the currently accepted age of diagnosis of between 18 and 24 months.

Diagnostic Predictors of Optimal Progress

Diagnosis at Age Two. The current study attempted to expand upon previous studies investigating predictors of highly positive outcomes from ASD in the toddler years. Specifically, the current study attempted to determined early (i.e., age two) child-level predictors of Optimal Progress (see “The Present Study” section for definition of Optimal Progress). Results of the current study indicate that, as hypothesized, children initially diagnosed with PDD-NOS are more likely to demonstrate Optimal Progress than children initially diagnosed with AD. This supports the work of Sutera and colleagues (2007) and is consistent with the aforementioned differences in the broad diagnostic stability of PDD-NOS verses AD. It appears that in addition to exhibiting a greater likelihood of losing their ASD diagnosis over time, children with PDD-NOS are more likely to demonstrate average range functioning in their toddler years. This, as above, is likely due to the less severe ASD symptomatology demonstrated by children with

PDD-NOS, which places them at a greater likelihood of making gains (both diagnostically and cognitively) that place them in the Optimal Progress group by age four.

DSM-IV Symptoms. In order to better understand differences in outcome between children with different initial diagnoses, age two DSM-IV symptoms were investigated. Our results indicate that children who later demonstrate Optimal Progress show fewer total DSM-IV symptoms at age two than their peers who remain on the ASD spectrum. In order to gain a more thorough understanding of early symptom differences, each domain of the DSM-IV (Social Interaction, Communication, Restricted Interests and Repetitive Behaviors) was separately investigated. Our results indicate that early symptoms of Restricted, Repetitive Behaviors and Interests help to predict Optimal Progress, but early symptoms in the Social Interaction and Communication domains do not. Specifically, children in the Optimal Progress group demonstrated fewer symptoms in the RRBs domain than their ASD-ASD peers, but similar numbers of symptoms in the other two domains. This is consistent with our finding that children initially diagnosed with PDD-NOS (e.g., children who may not exhibit RRBs) are the most likely to demonstrate Optimal Progress. Further, it is consistent with the work of Lord and colleagues (2006) who found that children with little or no repetitive behaviors during the ADOS and ADI-R were the most likely to change diagnosis from AD to PDD-NOS or from PDD-NOS to non-spectrum.

Given our finding that children with fewer RRBs (but not fewer symptoms across all domains) are more likely to demonstrate Optimal Progress, it is important to consider the role or significance of RRBs in development. It may be that restricted, repetitive behaviors or interests impede children from optimally engaging in their environment, and in turn, prevent them from fully benefitting from important learning experiences in both daily interactions and early intervention. Children who demonstrate fewer of these behaviors may be most likely to benefit

and learn from their interactions and surroundings, and in turn, to demonstrate Optimal Progress. It is also possible that the presence of RRBs reflects more severe overall ASD symptomatology, which will be discussed in the subsequent section.

Our finding that the presence of fewer RRBs helps to predict Optimal Progress is of particular importance given recent changes in DSM criteria, which now requires individuals to demonstrate at least two symptoms in the RRB domain (APA, 2013). Work by Worley and Matson (2012) indicates that when applying DSM-V criteria to children diagnosed with an ASD using the DSM-IV, 32% of children will lose their diagnosis despite showing significant levels of impairment. Importantly, it is largely children who would meet DSM-IV criteria for PDD-NOS who will no longer meet DSM-V criteria for ASD. In combination, this indicates that children who are the most likely to benefit from early intervention and to demonstrate Optimal Progress (i.e., children with fewer RRBs, children with diagnoses of PDD-NOS) are the same children who are most likely to no longer meet diagnostic criteria. Without a DSM diagnosis of ASD, these children will be unlikely to receive adequate services, and in turn, may not reach the highly positive outcomes of which they are capable. It is critical that we advocate for services for these children so that they can demonstrate their maximum potential.

Symptom Severity. Symptom severity at age two was also investigated as a possible predictor of Optimal Progress. Results of the current study indicate that, as hypothesized, children who later demonstrate Optimal Progress show lesser total symptom severity at age two (as measured by the CARS) than their peers who remain on the spectrum. This finding supports the work of Turner and Stone (2007) who found that children who lost their ASD diagnosis demonstrated lesser early symptom severity than children who retained their diagnoses. More

broadly, our results support previous findings that lesser early symptom severity is predictive of more positive outcomes later in development (Baghdadli et al., 2012; Eaves & Ho, 2004).

Notably, however, the current study found no group differences in age two ADOS CSS scores (Gotham, Risi, Pickles, & Lord, 2007), which are also a measure of ASD symptom severity. This difference is likely due to important differences in the CARS and ADOS CSS. Both measures are observation-based; however, the ADOS includes observations made during a standardized, play-based assessment, whereas the CARS includes observations made across a range of assessments (e.g., cognitive, ASD-specific) as well as clinician's impressions based on parent-report of a child's development and symptomatology. In terms of the types of items included, both measures assess similar symptoms and behaviors, with the CARS reflecting a slightly broader range of items than the ADOS (e.g., DSM-IV-like symptoms as well as "adaptation to change", "activity level", "listening response," etc.). Perhaps most importantly, the ADOS CSS consistently accounts for language level and age in its ratings, whereas the CARS does not. In sum, it is possible that group differences were found on the CARS (and not the ADOS CSS) because of its broader range of symptoms assessed, its inclusion of language level in severity ratings, and its inclusion of information gleaned from parent-report.

Analyses of individual factors within the CARS may help us to better understand the source of this discrepancy, as well as to better understand which specific symptom types may help us to predict Optimal Progress. Utilizing the factors determined by Magyar and Pandolfi (2007) the current study found that children who later demonstrate Optimal Progress show lesser early symptom severity in the domain of Social Communication, but not in the domain of Social Interaction. Items in the Social Communication domain include imitation, verbal communication

and nonverbal communication¹. Items in the Social Interaction domain include a child's general ability to relate to others and their visual response (e.g., eye contact). Therefore, it appears that children who later go on to demonstrate Optimal Progress show less impaired communication skills than their ASD-ASD peers at age two, but show similar levels of impairment in the ability to relate to others. Further, the current study found that children in the Optimal Progress group show similar levels of impairment to their peers in their emotional regulation abilities (e.g., emotional response, adaptation to change, and activity level). Our findings of similar levels of impairment in social interaction and emotional regulation should be interpreted cautiously, however, given the large effect sizes seen (.37 and .32 respectively) in these analyses.

The current study also found that children who later go on to demonstrate Optimal Progress show lesser early symptom severity in the domain of Stereotypies and Sensory Abnormalities (e.g., a child's body use, taste, smell and touch response and listening response). This finding is consistent with our finding that children in the OP group showed fewer symptoms in the RRB domain than their peers who remain on the spectrum. As discussed above, perhaps fewer symptoms or lesser severity of symptoms in this domain helps children to most optimally engage in and learn from their interactions and surroundings. This, in combination with stronger communication abilities, may help children to most fully benefit from important learning in both daily interactions and early intervention, and in turn, to demonstrate Optimal Progress.

Cognitive and Language Predictors of Optimal Progress

Based on previous studies of both diagnostic stability and outcomes more broadly, we hypothesized that children who later demonstrate Optimal Progress would show stronger early

¹ Note: This domain also includes a child's level and consistency of intellectual functioning and the clinician's general impressions of ASD symptomatology. Given the lack of theoretical relevance of these items to the Social Communication domain, analyses were run with and without these items. No differences in results were found.

cognitive and language abilities than their peers who remain on the spectrum. Contrary to our hypothesis, we found no significant group differences in any domain of cognitive or language ability as assessed by the Mullen (Visual Reception, Receptive Language, Expressive Language). This finding is in contrast with the findings of Turner and Stone (2007) who found that children who moved off the ASD spectrum had higher visual reception abilities and receptive language abilities (also as assessed on the Mullen Scales of Early Learning) than children who remained on the spectrum. More broadly, it is in contrast with the general conception that individuals with higher levels of cognitive and language functioning have outcomes that are more positive across a range of areas of functioning (A. Levy & Perry, 2011; Luyster, Lopez, & Lord, 2007; Sallows & Graupner, 2005).

Given the small to moderate effect sizes found in the current study's analyses (ranging from .26 to .49) it is possible that group differences in cognitive and/or language abilities would be found in a larger sample. Further, it is possible that despite the transformation of standard scores into estimated IQ scores, remaining floor effects on the Mullen in our sample prevented us from finding significant group differences. It is also possible that the Mullen may not be a sensitive enough measure to detect subtle group differences in cognitive or language abilities in two year old children, and therefore, that a more sensitive measure would be needed to characterize possible differences between the OP and ASD-ASD children. Nonetheless, our findings have important implications for understanding the early factors necessary for producing the outcome of Optimal Progress. In a review, Helt et al. (2008) indicated that highly positive outcomes were unlikely for children with cognitive functioning below a standard score of 70, however, children in the current study who went on to demonstrate Optimal Progress exhibited

standard scores as low as 49. Therefore, it appears that cognitive functioning below 70 does not preclude Optimal Progress.

Adaptive Skill Predictors of Optimal Progress

The current study attempted to replicate and expand previous findings that stronger early adaptive skills (specifically, daily living skills) help to predict both the loss of an ASD diagnosis in toddlerhood (Sutera et al., 2007) and more positive outcomes broadly (Sallows & Graupner, 2005). Our results indicate that children who later demonstrate Optimal Progress show stronger early adaptive skills across domains (Communication, Socialization, Daily Living, Motor) as indicated by parent-report on the VABS. Given the relatively limited research and discussion of the role or significance of adaptive skills in producing highly positive outcomes, each domain will be discussed below.

Our results indicate that at age two, the Optimal Progress group demonstrated stronger communication abilities and social skills than their peers who remain on the spectrum. Stronger social skills, in combination with stronger communication abilities, may reflect greater early social motivation in children who go on to demonstrate Optimal Progress. Greater social motivation may increase the likelihood that these children would regularly engage with peers and adults, and in turn, would increase the number of social learning experiences in which these children could develop their social and communication abilities (Chevallier, Kohls, & Troiani, 2012). In support of this hypothesis, Bopp and colleagues (2009) found that children with stronger social interaction skills showed greater gains over time in language comprehension and production. Further, Lord and colleagues (2006) found that children who showed more prosocial behavior at age two were the most likely to show diagnostic improvement by age nine.

In addition to stronger communication abilities and social skills, the Optimal Progress group demonstrated stronger motor skills at age two than their peers who remain on the

spectrum. Motor skills are a prerequisite for much of the learning and play that toddlers engage in on a daily basis. Lloyd and colleagues (2013) explain that movement is a critical element of active play, which facilitates the development of social skills, understanding of the world, daily living skills and play skills. Therefore, as discussed by MacDonald and colleagues (2013), motor skills deficits may hinder improvements in social communication skills. The stronger early motor skills, in combination with stronger social and communication abilities, demonstrated by children in the Optimal Progress group may allow these children to engage more regularly, consistently and fully in active, social play. This, in turn, may facilitate the rapid improvements seen in social and communication abilities in these children by age four.

It is also possible that, as discussed by Mostofsky and colleagues (2007), motor skills deficits and social/communication deficits are related at a more basic neurological level. Specifically, Mostofsky and colleagues (2007) argue that global deficits in procedural learning mechanisms may underlie deficits in both motor skills and social/communicative skills. Stronger early motor skills may be reflective of more typical neurological functioning, specifically, more typical patterns of white matter in the precentral cortex, which plays a role in motor functioning (Mostofsky, Burgess, & Gidley Larson, 2007). More typical neurological functioning would likely predispose children to demonstrate outcomes that are more positive. Additional neuroimaging studies will be required to determine if children who show highly positive outcomes (i.e. Optimal Progress) demonstrate neurological differences when compared to their peers who remain on the spectrum.

Finally, the Optimal Progress group demonstrated stronger daily living skills than their peers who remained on the spectrum. As discussed by Suter et al. (2007), stronger daily living skills may reflect a number of unmeasured factors including greater independence or greater motivation to learn in these children, as well as more proactive parenting. Additionally, stronger

daily living skills may be reflective of stronger motor skills, which may be important for the many reasons discussed above. Importantly, it is likely the interaction of all of these factors (e.g., motor skills, social and communication abilities) that predispose or contribute to highly positive outcomes by age four.

Patterns of Growth between Ages Two and Four

Our results indicate that, as hypothesized, children in the Optimal Progress group demonstrated significantly steeper trajectories of growth between ages two and four across all measured domains of cognitive, language, adaptive and motor skills than their peers who remain on the spectrum. It is notable that the Optimal Progress group demonstrated significantly greater growth in cognitive and language abilities despite showing similar levels of functioning in these areas to their peers at age two. In combination with our previously discussed findings, this supports the belief that particular factors within the Optimal Progress and ASD-ASD groups place them on different developmental trajectories. Importantly, the Optimal Progress group demonstrated this steeper trajectory of growth even when age two ASD symptom severity was controlled for, indicating that differences in trajectory may be related to factors beyond initial symptom severity. These factors could include child-level factors in which we found group differences at age two (e.g., communication abilities, social skills, motor skills), subtle differences in cognitive abilities, unmeasured child-level factors, and/or intervention-level factors.

Clinical Significance: Functioning at Age Four

By definition, the Optimal Progress group no longer met symptom criteria for any ASD at age four, and were functioning within 1.5 standard deviations of the mean (i.e., standard scores of 77 or greater) on standardized measures of cognitive and language abilities, communication

and social skills. Beyond these criteria, results of the current study indicate that the Optimal Progress group showed a near absence of any ASD symptoms by age four (i.e., average of 1 symptom, symptom severity within the range of typically developing children) and demonstrated overall cognitive and adaptive abilities squarely in the average range (within .5 standard deviations of the mean). These findings indicate that at age four, these children are likely functioning quite similarly to their typically developing peers in the assessed domains. This replicates previous studies comparing Optimal Outcome and typically developing children which found only minimal or subtle differences between the two groups (Fein et al., 2013; Kelley et al., 2010; Kelley, Paul, Fein, & Naigles, 2006). Our results support the possibility of truly optimal outcomes for children diagnosed with ASD early in development.

Limitations and Future Directions

When considering the findings of the current study, several limitations should be considered. Firstly, while a sample size of 19 is adequate given the rarity of Optimal Progress, it remains a small sample with limited power. Future studies should attempt to ascertain a larger group of children who demonstrate this type of outcome in toddlerhood in order to increase the power of analyses. Secondly, it is notable that in the current study large group differences were detected in adaptive skills, but not in cognitive abilities, which is in conflict with previous research. This may indicate that at age two, parent-reported adaptive skills may serve as stronger predictors of outcome than a child's cognitive abilities. Given previous research supporting the predictive value of cognitive abilities, however, it is important to consider an alternative possibility that this is a result of the specific measure(s) utilized. The current study may be limited in its ability to detect early differences in cognitive and language abilities by the measure used, the Mullen Scales of Early Learning. Future studies should utilize a more fine-tuned

measure of early cognitive and language abilities in order to determine whether differences exist between children who demonstrate Optimal Progress and those who remain on the spectrum.

Thirdly, the age of follow up in the current study (age four) serves as both a strength and a possible limitation. Follow-up at age four allows us demonstrate that highly positive outcomes are possible very early in development when children are diagnosed at approximately 26 months. As discussed above, children who demonstrate Optimal Progress are functioning well within the average range across domains and are likely difficult to distinguish from their typically developing peers. Future studies should compare children with Optimal Progress directly to typically developing peers as has been done in studies of Optimal Outcome. While the children in our Optimal Progress group appear to be optimally functioning four year olds, our follow-up to age four limits our ability to assess these children's later peer relationships and school functioning. Future studies should include longer follow-up periods to determine the extent to which these children continue on this optimal trajectory, and whether residual, subtle deficits exist for these children later in childhood. Additionally, later follow-up will allow future studies to characterize children who may not yet show this outcome at age four, but meet criteria for Optimal Progress or Optimal Outcome later in development.

Fourth, a major limitation of the current study is the lack of information on the interventions received between the age two and age four evaluations. The large majority of children in our sample received early intervention, and based on previous research, it is likely that early intervention plays a large role in producing highly positive outcomes from ASD. The fourth aim of our study, to characterize the early intervention received by the Optimal Progress and ASD-ASD groups, is in progress and will be presented in a later project. Importantly, it is likely the interaction between child-level factors, such as those investigated in the current study,

and intervention-level characteristics, that produce highly positive outcomes such as Optimal Progress. Therefore, future studies should attempt to characterize these interactions so that we can best understand the mechanisms by which Optimal Progress occurs. Additional factors, such as parent (e.g., mental health) and family characteristics (e.g., socioeconomic status), should also be considered.

Conclusions

As evidenced by a great deal of previous research, a wide range of outcomes are possible for individuals diagnosed with ASD early in their development. For the majority of individuals with ASD, symptoms appear to be largely stable across the lifespan, with modest improvements over time (Charman et al., 2005; Matson & Horovitz, 2010b; Seltzer et al., 2004). The Optimal Progress group represents a distinct subset of individuals with ASD who demonstrate large, clinically significant changes in symptom presentation by age four such that they no longer met criteria for any ASD, and who are functioning within the average range on standardized measures of cognitive, language, social and communication abilities. The current study found that a number of early child-level factors held to predict this highly positive outcome including a diagnosis of PDD-NOS, lesser early symptom severity, fewer symptoms in the domain of RRBs, as well as stronger early communication, social, daily living and motor skills.

Through characterizing the Optimal Progress group, the current study advances our understanding of the multiple possibilities of developmental trajectories seen in children with early diagnoses of ASD. Further, by improving our understanding of the relationship between early child characteristics and highly positive outcomes, the current study hopes to inform parents and clinicians as to the outcomes that are likely or possible for their children. In combination with the findings of the current study, future studies should attempt to characterize

the mechanisms at work in producing these outcomes, including the role of early intervention. In doing so, we can begin to promote an increase in the percentage of children attaining highly positive outcomes from ASD.

References

American Psychiatric Association. (2000). *Diagnostic and statistical manual of mental disorders* (4th ed., text rev.). Washington, DC: American Psychiatric Publishing.

- American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders*. (5th ed.). Arlington, VA: American Psychiatric Publishing.
- Barton, M.L., Boorstein, H., Dumont-Mathieu, T., Herlihy, L.E., & Fein, D. (2012). *Toddler ASD Symptom Interview (TASI)*. Self-published.
- Baghdadli, A., Assouline, B., Sonié, S., Pernon, E., Darrou, C., Michelon, C., ... Pry, R. (2012). Developmental trajectories of adaptive behaviors from early childhood to adolescence in a cohort of 152 children with autism spectrum disorders. *Journal of Autism and Developmental Disorders*, 42(7), 1314–25. doi:10.1007/s10803-011-1357-z
- Bryson, S. E., Rogers, S. J., & Fombonne, E. (2003). Autism Spectrum Disorders: Early Detection, Intervention, Education and Psychopharmacological Management. *Canadian Journal of Psychiatry. Revue Canadienne de Psychiatrie*, 48(8), 506.
- Cederlund, M., Hagberg, B., Billstedt, E., Gillberg, I. C., & Gillberg, C. (2008). Asperger syndrome and autism: a comparative longitudinal follow-up study more than 5 years after original diagnosis. *Journal of Autism and Developmental Disorders*, 38(1), 72–85. doi:10.1007/s10803-007-0364-6
- Charman, T., Jones, C. R. G., Pickles, a, Simonoff, E., Baird, G., & Happé, F. (2011). Defining the cognitive phenotype of autism. *Brain Research*, 1380(1943), 10–21. doi:10.1016/j.brainres.2010.10.075
- Charman, T., Taylor, E., Drew, A., Cockerill, H., Brown, J.-A., & Baird, G. (2005). Outcome at 7 years of children diagnosed with autism at age 2: predictive validity of assessments conducted at 2 and 3 years of age and pattern of symptom change over time. *Journal of Child Psychology and Psychiatry, and Allied Disciplines*, 46(5), 500–13. doi:10.1111/j.1469-7610.2004.00377.x

- Chawarska, K., Klin, A., Paul, R., Macari, S., & Volkmar, F. (2009). A prospective study of toddlers with ASD: short-term diagnostic and cognitive outcomes. *Journal of Child Psychology and Psychiatry, and Allied Disciplines*, *50*(10), 1235–45. doi:10.1111/j.1469-7610.2009.02101.x
- Chevallier, C., Kohls, G., & Troiani, V. (2012). The social motivation theory of autism. *Trends in Cognitive Sciences*, *16*(4), 231–239. doi:10.1016/j.tics.2012.02.007
- Eaves, L. C., & Ho, H. H. (2004). The very early identification of autism: outcome to age 4 1/2-5. *Journal of Autism and Developmental Disorders*, *34*(4), 367–78. Retrieved from <http://www.ncbi.nlm.nih.gov/pubmed/15449513>
- Eldevik, S., Hastings, R. P., Hughes, J. C., Jahr, E., Eikeseth, S., & Cross, S. (2009). Meta-analysis of Early Intensive Behavioral Intervention for children with autism. *Journal of Clinical Child and Adolescent Psychology* □: *The Official Journal for the Society of Clinical Child and Adolescent Psychology, American Psychological Association, Division 53*, *38*(3), 439–50. doi:10.1080/15374410902851739
- Fein, D., Barton, M., Eigsti, I.-M., Kelley, E., Naigles, L., Schultz, R. T., ... Tyson, K. (2013). Optimal outcome in individuals with a history of autism. *Journal of Child Psychology and Psychiatry, and Allied Disciplines*, *54*(2), 195–205. doi:10.1111/jcpp.12037
- Fombonne, E. (2003). Epidemiological surveys of autism and other pervasive developmental disorders: an update. *Journal of Autism and Developmental Disorders*, *33*(4), 365–82. Retrieved from <http://www.ncbi.nlm.nih.gov/pubmed/12959416>
- Gotham, K., Risi, S., Pickles, A., & Lord, C. (2007). The Autism Diagnostic Observation Schedule: revised algorithms for improved diagnostic validity. *Journal of Autism and Developmental Disorders*, *37*(4), 613–27. doi:10.1007/s10803-006-0280-1

- Harris, S. L., & Handleman, J. S. (2000). Age and IQ at intake as predictors of placement for young children with autism: a four- to six-year follow-up. *Journal of Autism and Developmental Disorders*, *30*(2), 137–42. Retrieved from <http://www.ncbi.nlm.nih.gov/pubmed/10832778>
- Helt, M., Kelley, E., Kinsbourne, M., Pandey, J., Boorstein, H., Herbert, M., & Fein, D. (2008). Can children with autism recover? If so, how? *Neuropsychology Review*, *18*(4), 339–66. doi:10.1007/s11065-008-9075-9
- Herlihy, L. E., Brooks, B., Dumont-Mathieu, T., Barton, M. L., Fein, D., Chen, C.-M., & Robins, D. L. (2014). Standardized screening facilitates timely diagnosis of autism spectrum disorders in a diverse sample of low-risk toddlers. *Journal of Developmental and Behavioral Pediatrics* □: *JDBP*, *35*(2), 85–92. doi:10.1097/DBP.0000000000000014
- Jones, W., & Klin, A. (2013). Attention to eyes is present but in decline in 2-6-month-old infants later diagnosed with autism. *Nature, advance on*. Retrieved from <http://dx.doi.org/10.1038/nature12715>
- Jónsdóttir, S. L., Saemundsen, E., Asmundsdóttir, G., Hjartardóttir, S., Asgeirsdóttir, B. B., Smáradóttir, H. H., ... Smári, J. (2007). Follow-up of children diagnosed with pervasive developmental disorders: stability and change during the preschool years. *Journal of Autism and Developmental Disorders*, *37*(7), 1361–74. doi:10.1007/s10803-006-0282-z
- Kelley, E., Naigles, L., & Fein, D. (2010). An in-depth examination of optimal outcome children with a history of autism spectrum disorders. *Research in Autism Spectrum Disorders*, *4*(3), 526–538. doi:10.1016/j.rasd.2009.12.001
- Kelley, E., Paul, J. J., Fein, D., & Naigles, L. R. (2006). Residual language deficits in optimal outcome children with a history of autism. *Journal of Autism and Developmental Disorders*, *36*(6), 807–28. doi:10.1007/s10803-006-0111-4

- Kleinman, J. M., Robins, D. L., Ventola, P. E., Pandey, J., Boorstein, H. C., Esser, E. L., ... Fein, D. (2008). The modified checklist for autism in toddlers: a follow-up study investigating the early detection of autism spectrum disorders. *Journal of Autism and Developmental Disorders, 38*(5), 827–39. doi:10.1007/s10803-007-0450-9
- Kleinman, J. M., Ventola, P. E., Pandey, J., Verbalis, A. D., Barton, M., Hodgson, S., ... Fein, D. (2008). Diagnostic stability in very young children with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 38*(4), 606–15. doi:10.1007/s10803-007-0427-8
- Levy, A., & Perry, A. (2011). Outcomes in adolescents and adults with autism: A review of the literature. *Research in Autism Spectrum Disorders, 5*(4), 1271–1282.
doi:10.1016/j.rasd.2011.01.023
- Levy, S. E., Mandell, D. S., & Schultz, R. T. (2009). Autism. *Lancet, 374*(9701), 1627–38.
doi:10.1016/S0140-6736(09)61376-3
- Lloyd, M., MacDonald, M., & Lord, C. (2013). Motor skills of toddlers with autism spectrum disorders. *Autism: The International Journal of Research and Practice, 17*(2), 133–46.
doi:10.1177/1362361311402230
- Lord, C. (1995). Follow-Up of Two-Year-Olds Referred for Possible Autism. *Journal of Child Psychology and Psychiatry, 36*(8), 1365–1382.
- Lord, C., Risi, S., DiLavore, P. S., Shulman, C., Thurm, A., & Pickles, A. (2006). Autism from 2 to 9 years of age. *Archives of General Psychiatry, 63*(6), 694–701.
doi:10.1001/archpsyc.63.6.694
- Lovaas, O. I. (1987). Behavioral treatment and normal educational and intellectual functioning in young autistic children. *Journal of Consulting and Clinical Psychology, 55*(1), 3–9.
Retrieved from <http://www.ncbi.nlm.nih.gov/pubmed/3571656>

- Luyster, R., Lopez, K., & Lord, C. (2007). Predicting Outcomes of Children Using the MacArthur-Bates Communicative Development Inventory, *50*(June), 667–682.
- Macdonald, M., Lord, C., & Ulrich, D. (2013). Research in Autism Spectrum Disorders The relationship of motor skills and adaptive behavior skills in young children with autism spectrum disorders §, *7*, 1383–1390.
- Mandell, D. S., Listerud, J., & Levy, S. E. (2001). Race Differences in the Age at Diagnosis Among Medicaid-Eligible Children With Autism, 1447–1453.
doi:10.1097/01.CHI.0000024863.60748.53
- Matson, J. L., & Horovitz, M. (2010). Stability of Autism Spectrum Disorders Symptoms over Time. *Journal of Developmental and Physical Disabilities*, *22*(4), 331–342.
doi:10.1007/s10882-010-9188-y
- Mostofsky, S. H., Burgess, M. P., & Gidley Larson, J. C. (2007). Increased motor cortex white matter volume predicts motor impairment in autism. *Brain*: *A Journal of Neurology*, *130*(Pt 8), 2117–22. doi:10.1093/brain/awm129
- Orinstein, A. J., Helt, M., Troyb, E., Tyson, K. E., Barton, M. L., Eigsti, I.-M., ... Fein, D. a. (2014). Intervention for optimal outcome in children and adolescents with a history of autism. *Journal of Developmental and Behavioral Pediatrics*: *JDBP*, *35*(4), 247–56.
doi:10.1097/DBP.0000000000000037
- Robins, D. L., Casagrande, K., Barton, M., Chen, C.-M. a, Dumont-Mathieu, T., & Fein, D. (2014). Validation of the Modified Checklist for Autism in Toddlers, Revised With Follow-up (M-CHAT-R/F). *Pediatrics*, *133*(1), 37–45. doi:10.1542/peds.2013-1813
- Robins, D. L., Fein, D., Barton, M. L., & Green, J. a. (2001). The Modified Checklist for Autism in Toddlers: an initial study investigating the early detection of autism and pervasive

developmental disorders. *Journal of Autism and Developmental Disorders*, 31(2), 131–44.

Retrieved from <http://www.ncbi.nlm.nih.gov/pubmed/11450812>

Rogers, S. J., & Vismara, L. a. (2008). *Evidence-based comprehensive treatments for early autism. Journal of clinical child and adolescent psychology* □: the official journal for the Society of Clinical Child and Adolescent Psychology, American Psychological Association, Division 53 (Vol. 37, pp. 8–38). doi:10.1080/15374410701817808

Sallows, G. O., & Graupner, T. D. (2005). Intensive behavioral treatment for children with autism: four-year outcome and predictors. *American Journal of Mental Retardation* □: AJMR, 110(6), 417–38. doi:10.1352/0895-8017(2005)110[417:IBTFCW]2.0.CO;2

Seltzer, M. M., Shattuck, P., Abbeduto, L., & Greenberg, J. S. (2004). Trajectory of development in adolescents and adults with autism. *Mental Retardation and Developmental Disabilities Research Reviews*, 10(4), 234–47. doi:10.1002/mrdd.20038

Sutera, S., Pandey, J., Esser, E. L., Rosenthal, M. a, Wilson, L. B., Barton, M., ... Fein, D. (2007). Predictors of optimal outcome in toddlers diagnosed with autism spectrum disorders. *Journal of Autism and Developmental Disorders*, 37(1), 98–107. doi:10.1007/s10803-006-0340-6

Toth, K., Munson, J., Meltzoff, A. N., & Dawson, G. (2006). Early predictors of communication development in young children with autism spectrum disorder: joint attention, imitation, and toy play. *Journal of Autism and Developmental Disorders*, 36(8), 993–1005. doi:10.1007/s10803-006-0137-7

Turner, L. M., & Stone, W. L. (2007). Variability in outcome for children with an ASD diagnosis at age 2. *Journal of Child Psychology and Psychiatry, and Allied Disciplines*, 48(8), 793–802. doi:10.1111/j.1469-7610.2007.01744.x

- Turner, L. M., Stone, W. L., Pozdol, S. L., & Coonrod, E. E. (2006). Follow-up of children with autism spectrum disorders from age 2 to age 9. *Autism*: *The International Journal of Research and Practice*, *10*(3), 243–65. doi:10.1177/1362361306063296
- Volkmar, F. R., Lord, C., Bailey, A., Schultz, R. T., & Klin, A. (2004). Autism and pervasive developmental disorders. *Journal of Child Psychology and Psychiatry, and Allied Disciplines*, *45*(1), 135–70. Retrieved from <http://www.ncbi.nlm.nih.gov/pubmed/14959806>
- Woolfenden, S., Sarkozy, V., Ridley, G., & Williams, K. (2012). A systematic review of the diagnostic stability of Autism Spectrum Disorder. *Research in Autism Spectrum Disorders*, *6*(1), 345–354. doi:10.1016/j.rasd.2011.06.008
- Worley, J. A., & Matson, J. L. (2012). Comparing symptoms of autism spectrum disorders using the current DSM-IV-TR criteria and the proposed DSM V diagnostic criteria. *Research in Autism Spectrum Disorders*, *6*, 965–970.

Appendix A

Table 1

Sample Demographics

	Optimal Progress n = 19	ASD-ASD n = 171	Total Sample n = 190	<i>t or X², effect size</i>
Age in Months (M, SD)				
Average age at Time 1	26.21 (4.81)	26.32 (4.37)	26.31 (4.40)	$t(187) = 0.10,$ $p = .921, d = .02$
Average age at Time 2	51.47 (4.81)	52.30 (9.75)	52.22 (9.52)	$t(188) = .36,$ $p = .718, d = .11$
Gender (#, %)				$X^2(1) = 2.69,$

				$p = .101, \phi = .12$
Male	n = 13 (68.4)	n = 143 (83.6)	n = 156 (82.1)	
Female	n = 6 (31.6)	n = 28 (16.4)	n = 34 (17.9)	
Race/Ethnicity (#, %)				Fisher's Exact Test = 3.85, $p = .605$
White	n = 16 (84.2)	n = 139 (81.3)	n = 155 (81.6)	
Hispanic/Latino	n = 1 (5.3)	n = 11 (6.4)	n = 12 (6.3)	
Black of African American	n = 0 (0)	n = 7 (4.1)	n = 7 (3.7)	
Asian or Pacific Islander	n = 1 (5.3)	n = 4 (2.3)	n = 5 (2.6)	
Biracial	n = 1 (5.3)	n = 3 (1.8)	n = 4 (2.1)	
Other	n = 0 (0)	n = 1 (0.6)	n = 1 (0.5)	
Not Available	n = 0 (0)	n = 6 (3.5)	n = 6 (3.2)	

Table 2

Maternal Education

	Optimal Progress n = 19 (#, %)	ASD-ASD n = 171 (#, %)	Total Sample n = 190 (#, %)	X^2
				Fisher's Exact Test = 6.37, $p = .719$
No degree or diploma	0 (0)	3 (1.8)	3 (1.6)	
GED	0 (0)	2 (1.2)	2 (1.1)	
High School Diploma	3 (15.8)	36 (21.1)	39 (20.5)	
Vocational or	0 (0)	7 (4.1)	7 (3.7)	

Technical Degree			
Associate's Degree	0 (0)	12 (7.0)	12 (6.3)
Bachelor's Degree	2 (10.5)	38 (22.2)	40 (21.1)
Master's Degree	4 (21.1)	20 (11.7)	24 (12.7)
PhD, JD, MD, etc.	0 (0)	3 (1.8)	3 (1.6)
Not Available	10 (52.7)	50 (29.2)	59 (31.1)

Table 3

Diagnostic Stability: Entire Sample

	Time 1 (N, %)	Time 2 (N, %)
Diagnosis		
AD	108 (52.2)	113 (54.6)
PDD-NOS	79 (38.2)	54 (26.1)
ASD-Low MA	20 (9.7)	4 (1.9)
Non-ASD Diagnosis	0 (0)	36 (17.4)
<i>Total</i>	190	190

Table 4

Time 2 Diagnoses of Children who lose their ASD Diagnosis

Diagnosis at Time 2	N
Developmental Delay	8
Developmental Language Disorder	2
Other Diagnosis (e.g., Regulatory Issues)	3
No Diagnosis	16
Typical Development	7

<i>Total Losing ASD Diagnosis</i>	36
<i>Total Demonstrating OP</i>	19

Table 5

Diagnostic Stability of Autistic Disorder

Diagnosis	Time 1 N (%)	Time 2 N (%)
AD	108 (100)	72 (66.7)
PDD-NOS		22 (20.3)
ASD-Low MA		0 (0.0)
Non-ASD		14 (13.0)

Table 6

Diagnostic Stability of PDD-NOS

Diagnosis	Time 1 N (%)	Time 2 N (%)
PDD-NOS	79 (100)	31 (39.2)
AD		22 (32.9)
ASD-Low MA		0 (0.0)
Non-ASD		22 (27.8)

Table 7

Diagnostic Stability of ASD-Low Mental Age

Diagnosis	Time 1 N (%)	Time 2 N (%)
ASD-Low MA	20 (100)	4 (20.0)
AD		15 (75.0)
PDD-NOS		1 (5.0)
Non-ASD		0 (0.0)

Table 8

Predictors of Optimal Progress: Time 1 Diagnosis

	Optimal Progress n = 19	ASD-ASD n = 171	χ^2
Diagnosis at Time 1			$\chi^2 = 5.63,$ $p = .06$
AD	8 (7.8%)	94 (92.2%)	
PDD-NOS	11 (16.2%)	57 (83.8%)	
ASD-Low MA	0 (0 %)	20 (100 %)	

Table 9

Correlation of Mullen Standard Scores with Calculated Mullen Ratio IQ Scores

Standard Scores	Calculated Ratio IQ Scores			
	EXL IQ	FM IQ	RL IQ	VR IQ
EXL	.859**			
FM		.801**		
RL			.522**	
VR				.803**

Table 10

Cognitive Abilities: Interaction between Time and Group in Mullen Estimated IQ Scores

Domain (covariates)	Interaction between Time and Group
Visual Reception (Months Between = 24.38, CARS = 32.08)	Wilks' Lambda=.888, $F(1,136)= 17.24, p<.001, \eta^2=.112$
Expressive Language (Months Between = 24.24, CARS =32.12)	Wilks' Lambda=.905, $F(1,133)=13.93, p<.001, \eta^2=.095.$
Receptive Language (Months Between = 24.21, CARS = 32.05)	Wilks' Lambda=.891, $F(1,135)=16.47, p<.001, \eta^2=.109$
Fine Motor (Months Between = 24.30, CARS = 32.07)	Wilks' Lambda=.914, $F(1,134)=12.53, p=.001, \eta^2=.086$

* Note: covariates include the number of months between evaluations and CARS total scores.

Table 11

Adaptive Skills: Interaction between Time and Group in Vineland Total and Domain Scores

Domain (covariates)		Interaction between Time and Group
Total (Months Between =26.05, CARS =32.63)		Wilks' Lambda=.712, $F(1,163)=66.09$, $p<.001$, $\eta^2=.288$
Communication (Months Between =26.04, CARS =32.53)		Wilks' Lambda=.787, $F(1,168)=45.53$, $p<.001$, $\eta^2=.213$
Socialization (Months Between =26.04, CARS =32.53)		Wilks' Lambda=.749, $F(1,168)=56.39$, $p<.001$, $\eta^2=.251$
Motor (Months Between = 24.93, CARS =32.59)		Wilks' Lambda=.889, $F(1,163)=20.38$, $p<.001$, $\eta^2=.111$
Daily Living		Wilks' Lambda=.841, $F(1,166)=31.39$, $p<.001$, $\eta^2=.159$

(Months Between = 25.70, CARS = 32.56)

* Note: covariates include the number of months between evaluations and CARS total scores.

Table 12

Time 2 Cognitive Abilities (Estimated IQ Scores)

	Optimal Progress (M, SD)	ASD-ASD (M, SD)	<i>t-test</i>
Domain			
Visual Reception	108.96 (13.01)	68.92 (30.54)	$t(42) = 9.88, p < .001,$ $d = 1.71$
Fine Motor	97.60 (14.18)	64.63 (24.07)	$t(29) = 8.27, p < .001,$ $d = 1.67$
Expressive Language	94.06 (8.94)	55.89 (28.03)	$t(65) = 11.96, p < .001,$ $d = 1.73$

Receptive Language	102.62 (11.78)	58.79 (31.10)	$t(50) = 11.35, p < .001,$ $d = 1.86$
--------------------	----------------	---------------	--

Table 13

Time 2 Adaptive Skills

	Optimal Progress (M, SD)	ASD-ASD (M, SD)	<i>t</i>
Domain			
Total Score	91.67 (11.43)	61.59 (13.54)	$t(176) = 10.37, p < .001,$ $d = 2.40$
Communication	102.58 (14.38)	68.99 (18.80)	$t(79) = 7.52, p < .001,$ $d = 2.01$
Socialization	91.63 (8.71)	64.94 (12.08)	$t(179) = 6.01, p < .001,$

Daily Living	86.11 (16.44)	61.80 (12.51)	$t(177) = 7.56, p < .001,$ $d = 2.53$
Motor Skills	94.11 (12.21)	69.77 (16.66)	$t(25) = 7.68, p < .001,$ $d = 1.66$

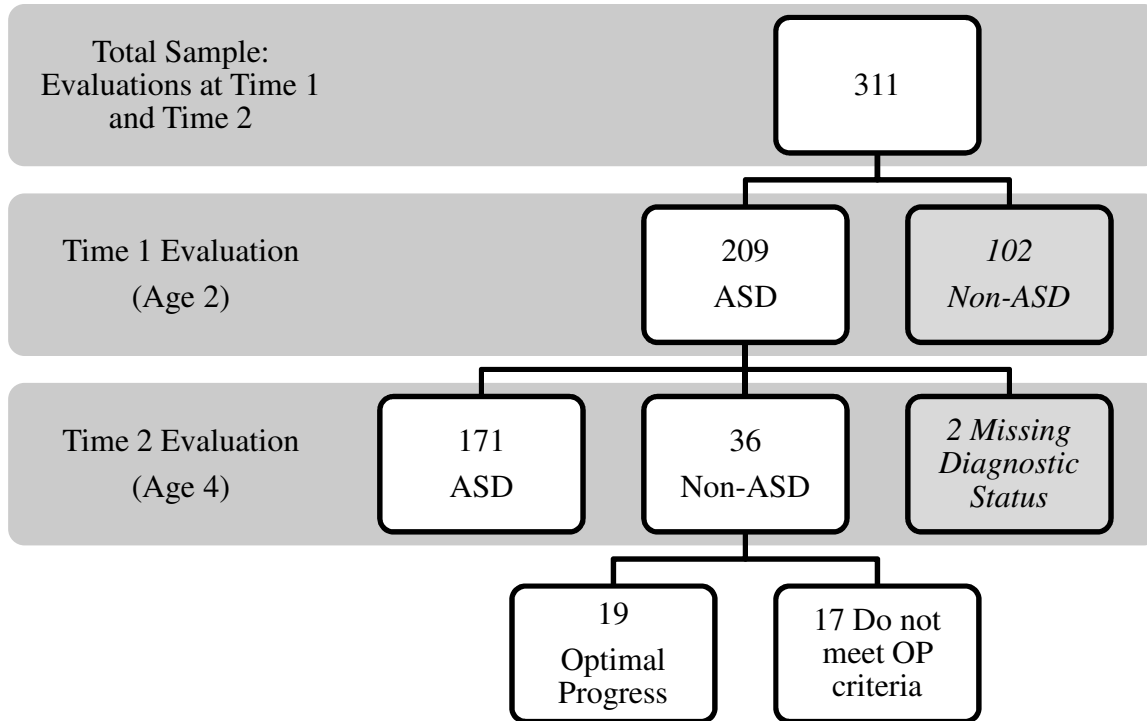


Figure 1. Flowchart indicating diagnostic results of Time 1 and Time 2 evaluations. The 102 children who received Non-ASD diagnoses at their Time 1 evaluation, and the 2 children who had missing information regarding diagnostic status at Time 2 were not included in the current analyses.

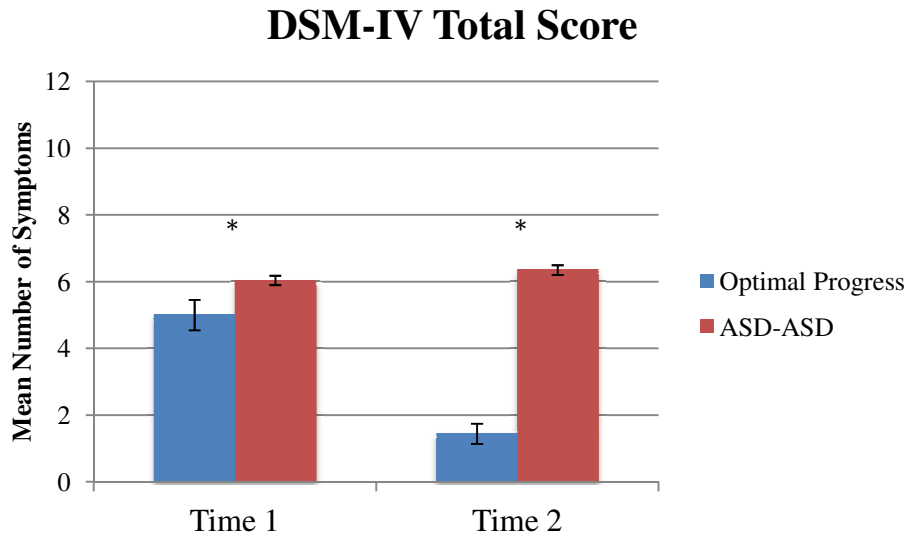


Figure 2. DSM-IV total score for the OP and ASD-ASD groups at Time 1 and Time 2.

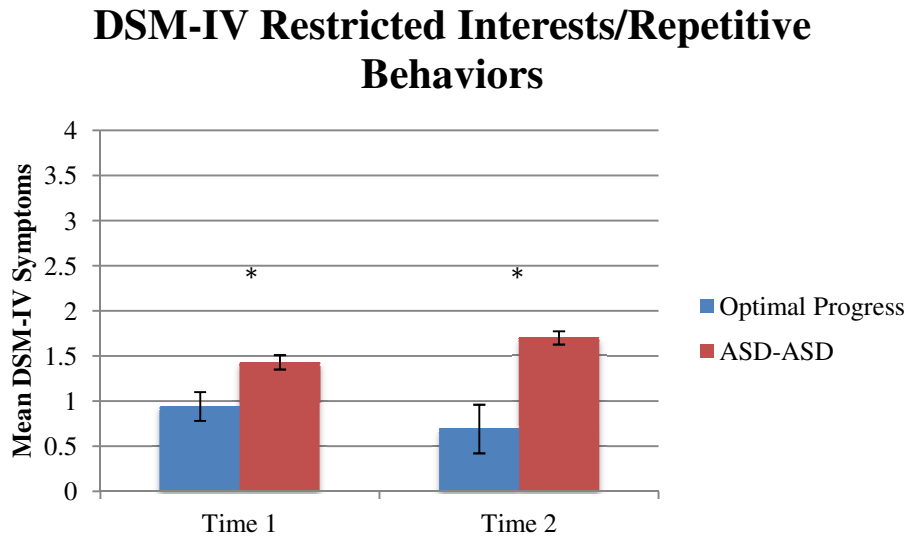


Figure 3. DSM-IV Restricted, Repetitive Behavior Symptoms for the OP and ASD-ASD groups at Time 1 and Time 2

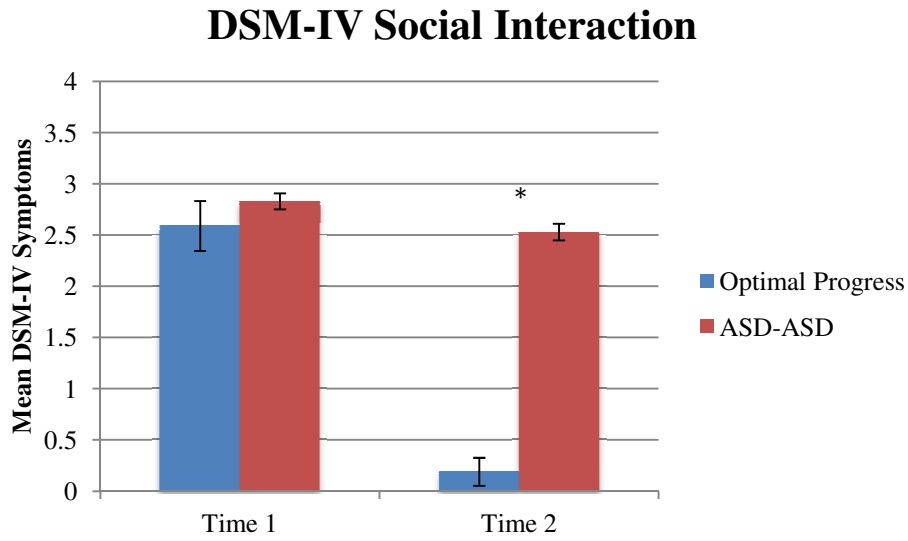


Figure 4. DSM-IV Social Interaction Symptoms for the OP and ASD-ASD groups at Time 1 and Time 2

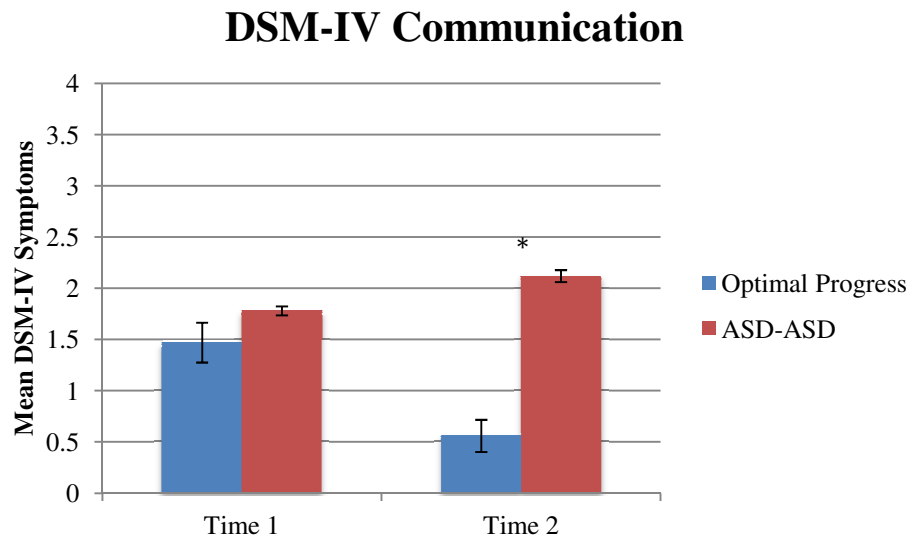


Figure 5. DSM-IV Communication Symptoms for the OP and ASD-ASD groups at Time 1 and Time 2

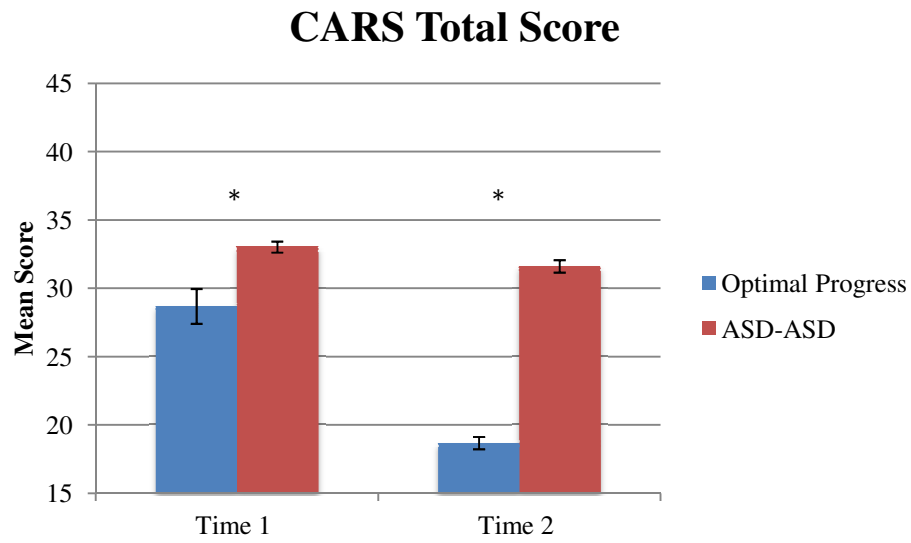


Figure 6. CARS total score for the OP and ASD-ASD groups at Time 1 and Time 2.

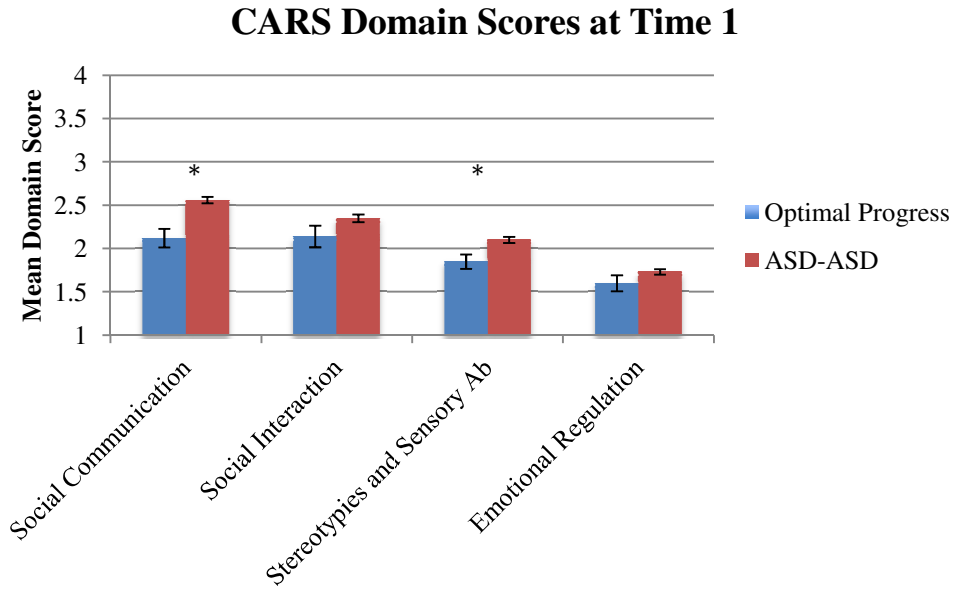


Figure 7. CARS domain scores for the OP and ASD-ASD groups at Time 1.

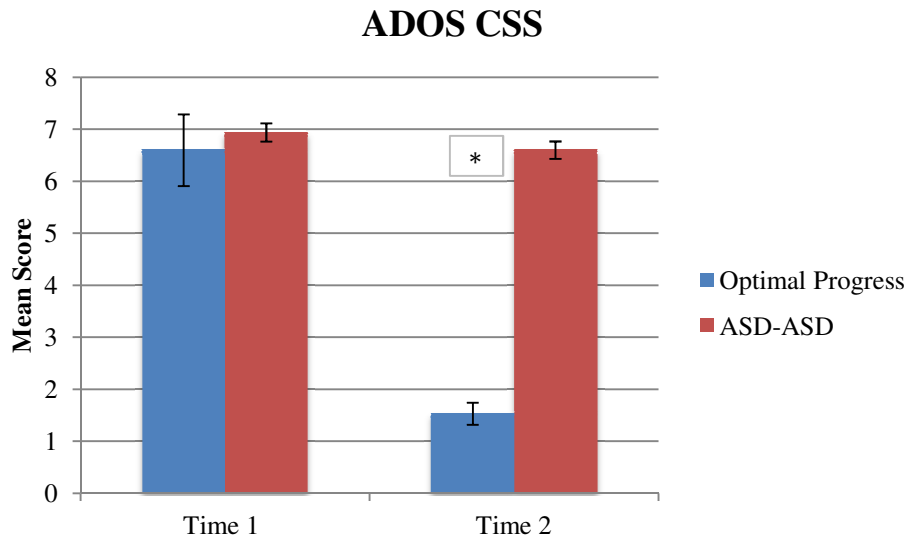


Figure 8. Mean ADOS CSS for the OP and ASD-ASD groups at Times 1 and 2.

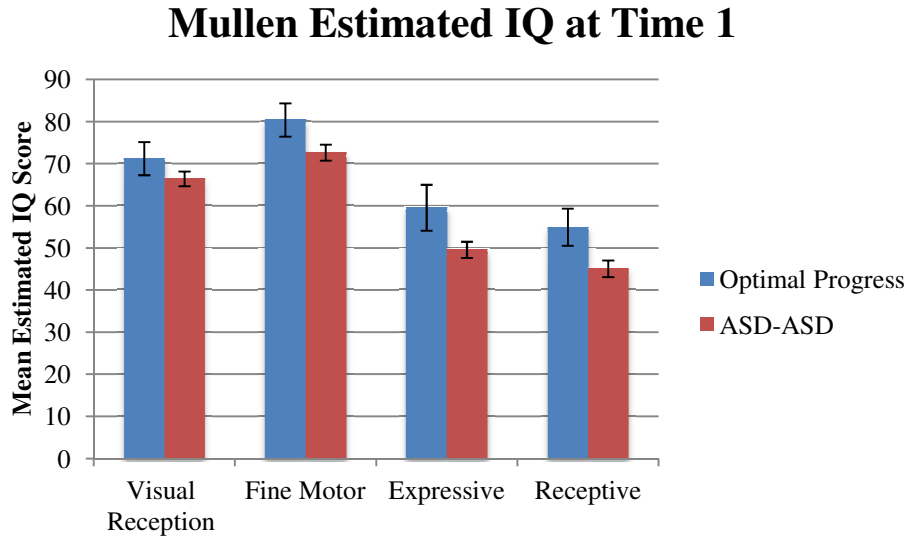


Figure 9. Mean Mullen estimated IQ scores at for the OP and ASD-ASD groups at Time 1. Estimated IQ scores were computed as follows: mental age / chronological age x 100.

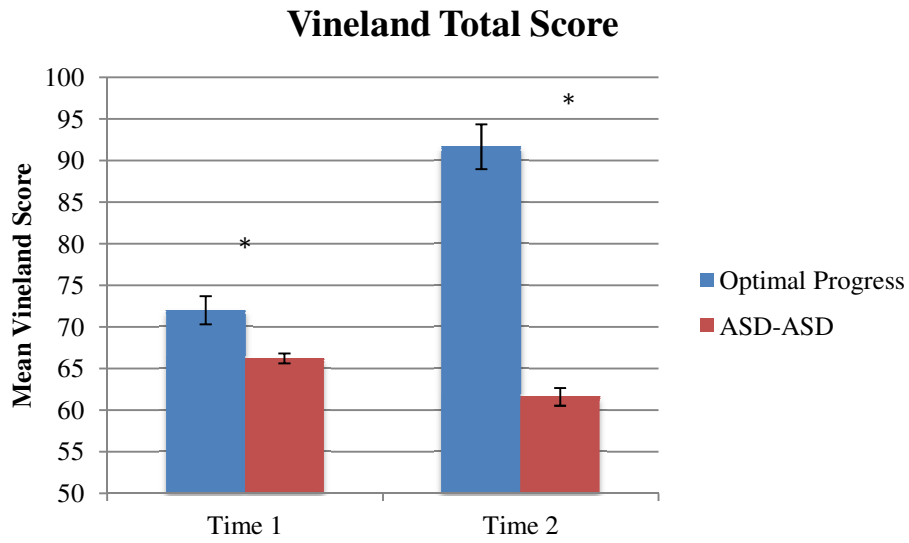


Figure 10. Mean Vineland total score for the OP and ASD-ASD groups at Times 1 and 2.

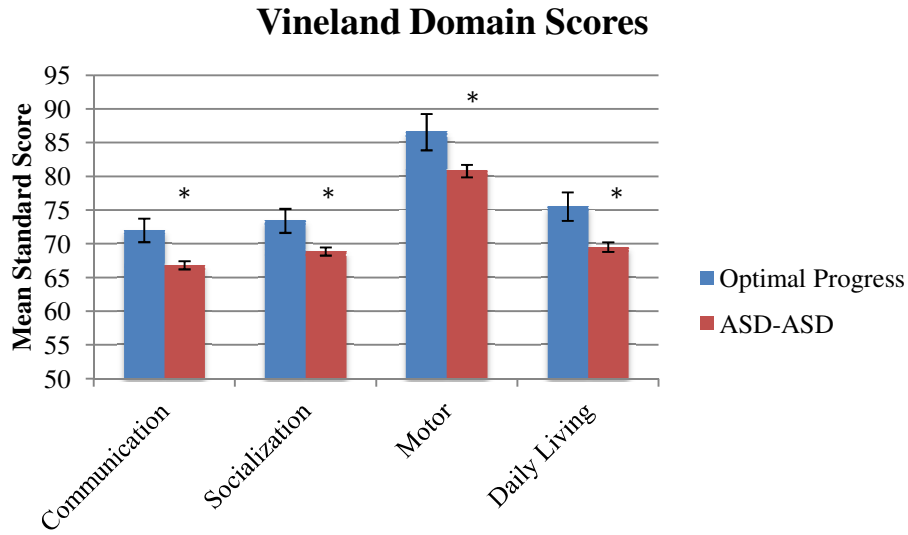


Figure 11. Mean Vineland domain scores for the OP and ASD-ASD groups at Time 1.

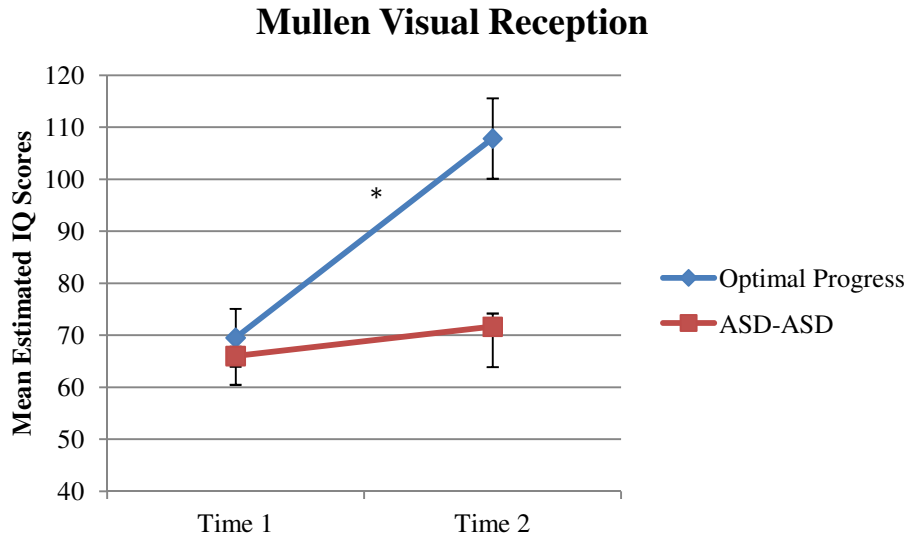


Figure 12. Pattern of changes between Time 1 and Time 2 for the OP and ASD-ASD groups in Mullen Visual Reception estimated IQ scores with months between evaluations covaried (24.51).

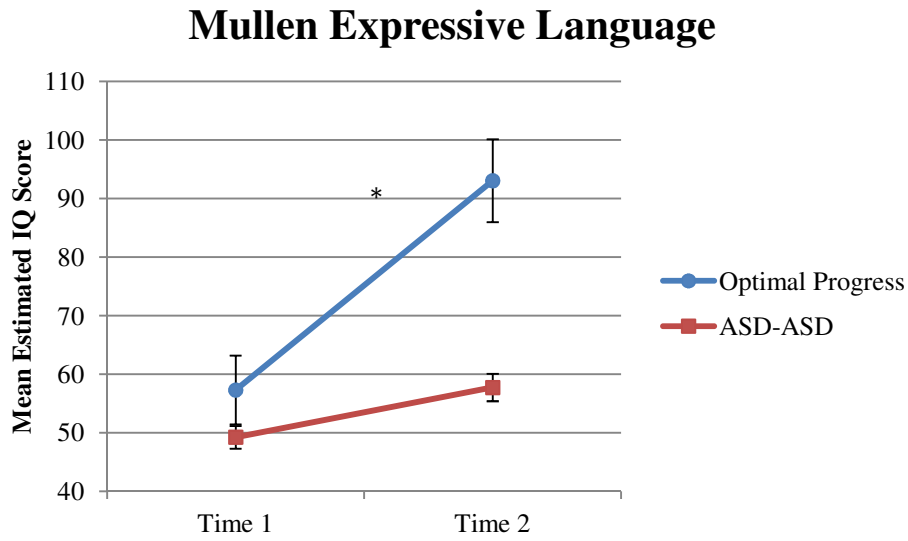


Figure 13. Pattern of changes between Time 1 and Time 2 for the OP and ASD-ASD groups in Mullen Expressive Language estimated IQ scores with months between evaluations covaried (24.40).

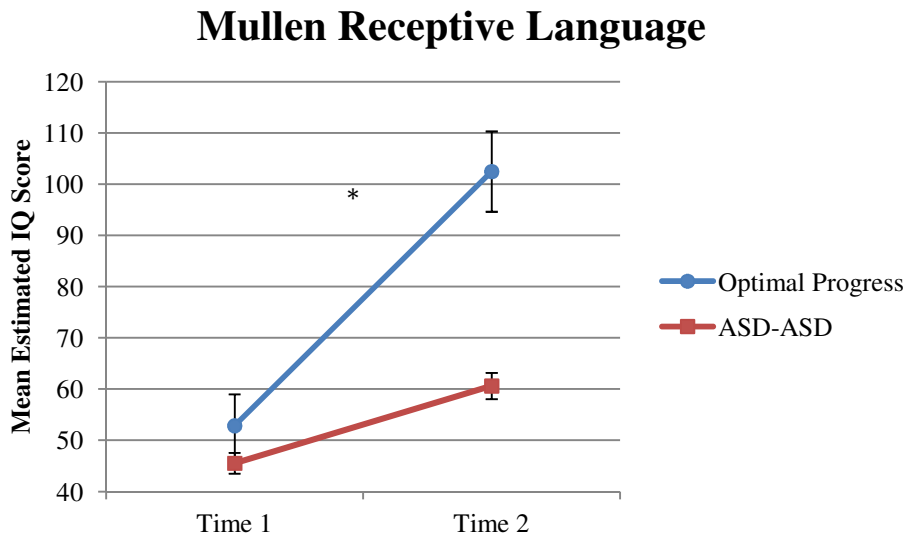


Figure 14. Pattern of changes between Time 1 and Time 2 for the OP and ASD-ASD groups in Mullen Receptive Language estimated IQ scores with months between evaluations covaried (24.37).

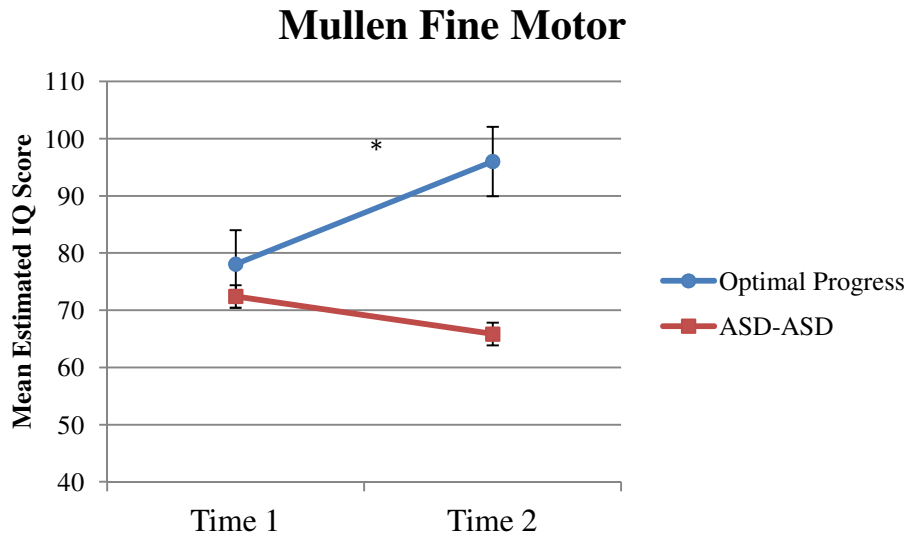


Figure 15. Pattern of changes between Time 1 and Time 2 for the OP and ASD-ASD groups in Mullen Fine Motor estimated IQ scores with months between evaluations covaried (24.45).

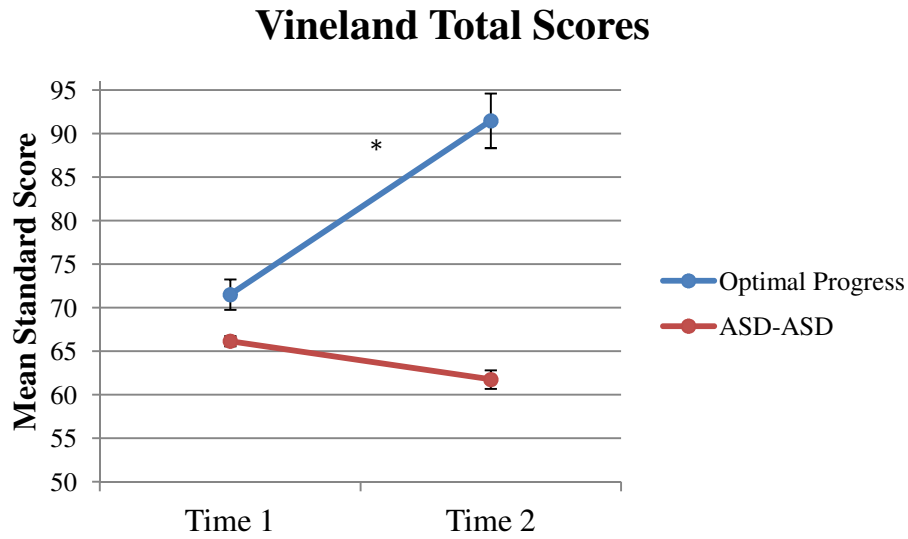


Figure 16. Pattern of changes between Time 1 and Time 2 for the OP and ASD-ASD groups in Vineland Total scores with months between evaluations covaried (26.15).

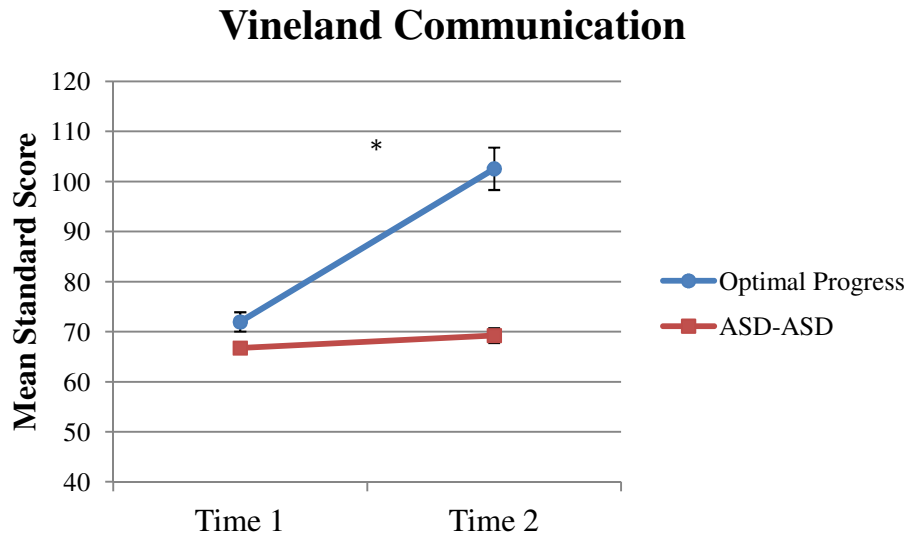


Figure 17. Pattern of changes between Time 1 and Time 2 for the OP and ASD-ASD groups in Vineland Communication scores with months between evaluations covaried (26.13).

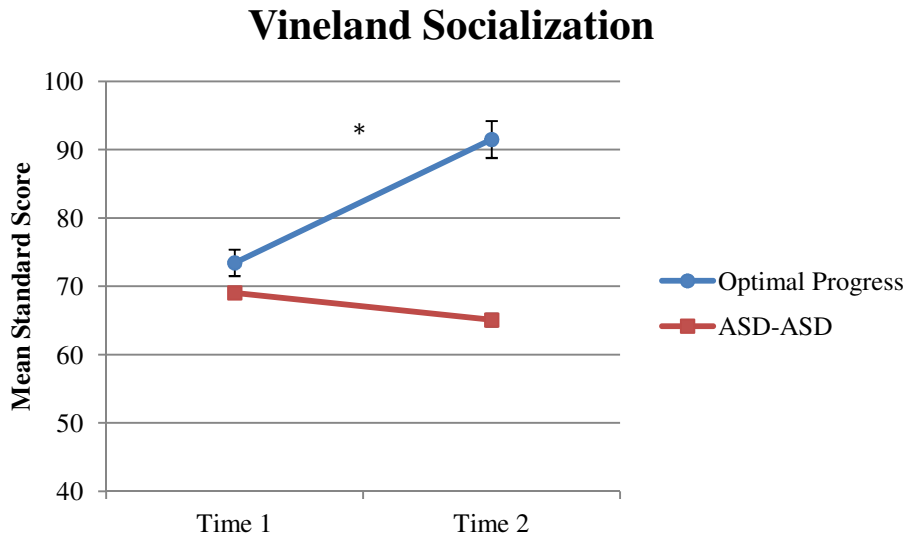


Figure 18. Pattern of changes between Time 1 and Time 2 for the OP and ASD-ASD groups in Vineland Socialization scores with months between evaluations covaried (26.13).

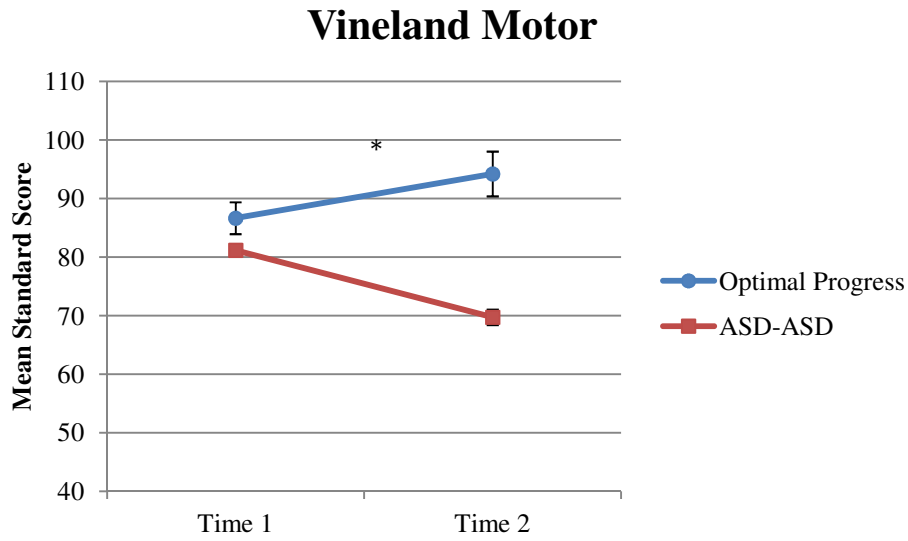


Figure 19. Pattern of changes between Time 1 and Time 2 for the OP and ASD-ASD groups in Vineland Motor scores with months between evaluations covaried (25.06).

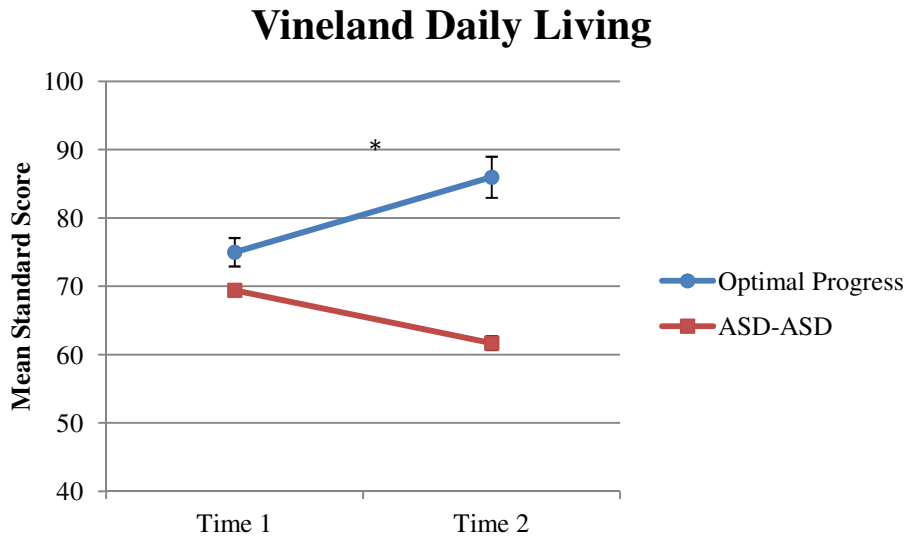


Figure 20. Pattern of changes between Time 1 and Time 2 for the OP and ASD-ASD groups in Vineland Daily Living scores with months between evaluations covaried (25.80).

Appendix C

Questionnaire for Parents

Treatment of Autism Spectrum Disorders in Toddlerhood

Date Form Completed: _____

Child's Name: _____ Date of Birth: _____

Highest level of education completed by child's mother (e.g. GED, high school diploma, Bachelor's degree): _____

A. Please complete the following questions regarding your experiences with _____'s Early Intervention services (ages 1 to 3):

1. What did you find most helpful about your child's early intervention services?

2. What would you have liked to change about your child's early intervention services?

B. Please complete the following questions regarding your experiences with _____'s Preschool services (ages 3 to 5):

1. What did you find most helpful about your child's preschool services?

2. What would you have liked to change about your child's preschool services?

C. Please provide an estimate of the number of additional hours (per week) that you or another caregiver spent working with your child at home on goals established by you and/or your intervention providers: _____.

D. Is there anything else you would like us to know about your child's intervention services?

Questionnaire for Early Intervention Providers

Treatment of Autism Spectrum Disorders in Toddlerhood

Date Form Completed: _____

Child's Name: _____ Date of Birth: _____

Intervention History

In six-month intervals, please indicate which of the following services the child has received, the start and end dates of each service type, the hours per week each service was provided, the location each service was provided, the format in which each service was provided, as well as any additional relevant information pertaining to each intervention.

- Birth to Three Services
- Speech-Language Services
- Occupational Therapy
- Special Education
- PECs
- Physical Therapy
- Sensory Integration Therapy
- Dietary Intervention (please specify)
- Vitamin Supplements (please specify)
- Work with an Early Intervention Associate
- Other (please specify)

A. Age 1.5 - 2 years:

I. Type (select from list above): _____

- i.** Start Date: _____
- ii.** End Date: _____
- iii.** Hours per week: _____
- iv.** Location (e.g. home, school): _____
- v.** Format (e.g. group, individual): _____
- vi.** Additional Information:

II. Type (select from list above): _____

- i.** Start Date: _____
- ii.** End Date: _____
- iii.** Hours per week: _____
- iv.** Location (e.g. home, school): _____
- v.** Format (e.g. group, individual): _____
- vi.** Additional Information:

III. Type (select from list above): _____

- i. Start Date: _____
- ii. End Date: _____
- iii. Hours per week: _____
- iv. Location (e.g. home, school): _____
- v. Format (e.g. group, individual): _____
- vi. Additional Information:

B. Age 2 – 2.5 years:

I. Type (select from list above): _____

- i. Start Date: _____
- ii. End Date: _____
- iii. Hours per week: _____
- iv. Location (e.g. home, school): _____
- v. Format (e.g. group, individual): _____
- vi. Additional Information:

II. Type (select from list above): _____

- i. Start Date: _____
- ii. End Date: _____
- iii. Hours per week: _____
- iv. Location (e.g. home, school): _____
- v. Format (e.g. group, individual): _____
- vi. Additional Information:

III. Type (select from list above): _____

- i. Start Date: _____
- ii. End Date: _____
- iii. Hours per week: _____
- iv. Location (e.g. home, school): _____
- v. Format (e.g. group, individual): _____

vi. Additional Information:

C. Age 2.5 – 3 years:

I. Type (select from list above): _____

i. Start Date: _____

ii. End Date: _____

iii. Hours per week: _____

iv. Location (e.g. home, school): _____

v. Format (e.g. group, individual): _____

vi. Additional Information:

II. Type (select from list above): _____

i. Start Date: _____

ii. End Date: _____

iii. Hours per week: _____

iv. Location (e.g. home, school): _____

v. Format (e.g. group, individual): _____

vi. Additional Information:

III. Type (select from list above): _____

i. Start Date: _____

ii. End Date: _____

iii. Hours per week: _____

iv. Location (e.g. home, school): _____

v. Format (e.g. group, individual): _____

vi. Additional Information:

D. Please select from the following list the *primary orientation* of the services provided (select one):

- Applied Behavior Analysis (ABA)
- Developmental Therapy (including floor time)
- Relationship Development Intervention (RDI)
- The Denver Model
- TEACCH
- Other (please specify): _____

F. Please indicate if any alternative orientations were also utilized (select any that are applicable):

- Applied Behavior Analysis (ABA)
- Developmental Therapy (including floor time)
- Relationship Development Intervention (RDI)
- The Denver Model
- TEACCH
- Other (please specify): _____

G. To your knowledge, has the family pursued any additional interventions (please specify)?:

Questionnaire for Preschool Providers

Treatment of Autism Spectrum Disorders in Toddlerhood

Date Form Completed: _____

Child's Name: _____ Date of Birth: _____

Please circle one of the following to indicate the structure of your preschool:

INTEGRATED

SELF-CONTAINED

Intervention History

In six-month intervals, please indicate which of the following services the child has received, the start and end dates of each service type, the hours per week each service was provided, the location each service was provided, the format in which each service was provided, as well as any additional relevant information pertaining to each intervention.

- Physical Therapy
- Speech-Language Services
- Occupational Therapy
- Special Education
- Social Skills Training
- Sensory Integration Therapy
- Dietary Intervention (please specify)
- Vitamin Supplements (please specify)
- Other (please specify)

E. Age 3 – 3.5 years:

I. Type (select from list above): _____

- i. Start Date: _____
- ii. End Date: _____
- iii. Hours per week: _____
- iv. Location (e.g. home, school): _____
- v. Format (e.g. group, individual): _____
- vi. Additional Information:

II. Type (select from list above): _____

- i. Start Date: _____
- ii. End Date: _____
- iii. Hours per week: _____
- iv. Location (e.g. home, school): _____
- v. Format (e.g. group, individual): _____
- i. Additional Information:

II. Type (select from list above): _____

- i. Start Date: _____
- ii. End Date: _____
- iii. Hours per week: _____
- iv. Location (e.g. home, school): _____
- v. Format (e.g. group, individual): _____
- vi. Additional Information:

B. Age 3.5 - 4 years:

I. Type (select from list above): _____

- i. Start Date: _____
- ii. End Date: _____
- iii. Hours per week: _____
- iv. Location (e.g. home, school): _____
- v. Format (e.g. group, individual): _____
- vi. Additional Information:

II. Type (select from list above): _____

- i. Start Date: _____
- ii. End Date: _____
- iii. Hours per week: _____
- iv. Location (e.g. home, school): _____
- v. Format (e.g. group, individual): _____
- vi. Additional Information:

III. Type (select from list above): _____

- i. Start Date: _____
- ii. End Date: _____
- iii. Hours per week: _____

iv. Location (e.g. home, school): _____

v. Format (e.g. group, individual): _____

vi. Additional Information:

C. Age 4 to 4.5 years:

I. Type (select from list above): _____

i. Start Date: _____

ii. End Date: _____

iii. Hours per week: _____

iv. Location (e.g. home, school): _____

v. Format (e.g. group, individual): _____

vi. Additional Information:

II. Type (select from list above): _____

i. Start Date: _____

ii. End Date: _____

iii. Hours per week: _____

iv. Location (e.g. home, school): _____

v. Format (e.g. group, individual): _____

vi. Additional Information:

III. Type (select from list above): _____

i. Start Date: _____

ii. End Date: _____

iii. Hours per week: _____

iv. Location (e.g. home, school): _____

v. Format (e.g. group, individual): _____

vi. Additional Information:

D. Age 4.5 to 5 years:

I. Type (select from list above): _____

- i. Start Date: _____
- ii. End Date: _____
- iii. Hours per week: _____
- iv. Location (e.g. home, school): _____
- v. Format (e.g. group, individual): _____
- vi. Additional Information:

II. Type (select from list above): _____

- i. Start Date: _____
- ii. End Date: _____
- iii. Hours per week: _____
- iv. Location (e.g. home, school): _____
- v. Format (e.g. group, individual): _____
- vi. Additional Information:

III. Type (select from list above): _____

- i. Start Date: _____
- ii. End Date: _____
- iii. Hours per week: _____
- iv. Location (e.g. home, school): _____
- v. Format (e.g. group, individual): _____
- vi. Additional Information:

E. Please select from the following list the *primary orientation* of the services provided (select one):

- Applied Behavior Analysis (ABA)
- Developmental Therapy (including floor time)
- Relationship Development Intervention (RDI)
- The Denver Model
- TEACCH
- Other (please specify): _____

H. Please indicate if any alternative orientations were also utilized (select any that are applicable):

- Applied Behavior Analysis (ABA)
- Developmental Therapy (including floor time)
- Relationship Development Intervention (RDI)
- The Denver Model
- TEACCH
- Other (please specify): _____

I. To your knowledge, has the family pursued any additional interventions (please specify)?:
