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## THE BABY AND INFANT SCREEN FOR CHILDREN WITH AUTISM TRAITS: A PROPOSED CRITICAL ITEM ALGORITHM

#### A Dissertation

Submitted to the Graduate Faculty of the Louisiana State University and Agricultural and Mechanical College in partial fulfillment of the requirements for the degree of Doctor of Philosophy

in

The Department of Psychology

by
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August 2011

### **Dedication**

I dedicate the completion of my Ph.D. to......

my family, especially my Mom, Dad, and Brother for providing me with the love and support needed to make it through the past five years;

my adviser, Dr. Matson, for his guidance and teaching me how to get through difficult situations by putting one foot in front of the other;

my friends for believing in me, for the laughs, and for the encouragement;

Tim Dempsey for taking this ride with me through the ups and downs, nearly missed deadlines, hurricanes, and back surgeries;

and all the kids I've worked with who have inspired me to lead a meaningful life.

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#### **Abstract**

Since its first description, the definition of autism has varied as a function of emphases on particular defining features, changes to the age of onset, and confusion with other disorders. However, a recurring theme has been the importance of social impairments with evidence that specific social symptoms, such as joint attention deficits, are predictive of autism within the first or second year of life. In addition to the core domains of impairment, autism is associated with various medical conditions, intellectual disability, comorbid psychopathology, and problem behavior. This is alarming considering that there may be a true increase in the disorder's prevalence and that it is associated with poor long-term outcomes. Fortunately, effective treatments exist that may alter the course of the disorder if administered early in a child's life. A method to facilitate early intervention is through the early screening of autism with instruments such as the Baby and Infant Screen for Children with aUtIsm Traits (BISCUIT). The BISCUIT is a comprehensive assessment battery designed to measure autism symptoms as well as associated comorbid psychopathology and problem behavior. The primary purpose of the current investigation was to further develop the BISCUIT by creating an abbreviated scoring algorithm to enhance the measure's diagnostic utility. Participants included 2,168 children ages 17 to 37 months enrolled in an early intervention program who were classified as having atypical development (n=1526) or an autism spectrum disorder (n=642). Discriminant function analysis (DFA) and receiver operating characteristic (ROC) analysis were conducted, resulting in a 5 item scoring algorithm with comparable diagnostic accuracy to the existing scoring procedure. Implications and directions for further research are discussed.

#### Introduction

Since its first clinical description in 1943 (Kanner), the definition of autism has taken many forms; however, a prevailing theme has been the presence of social abnormalities, or a social disconnect, evident in the first years of life. Our current definition, as described in the Diagnostic and Statistical Manual Fourth Edition, Text Revision (DSM-IV-TR; American Psychiatric Association, 2000), includes social and communication impairments with the presence of stereotyped behaviors. The symptoms are pervasive and are unfortunately associated with other medical disorders, psychiatric disorders, and problem behaviors (Gillberg & Billstedt, 2000). Based upon the totality of difficulties persons with autism are likely to face in their lives, it is not surprising that long-term outcomes have historically been poor (Howlin, 2005).

Fortunately, there is accumulating evidence that intensive behavioral treatments may lead to improvements in many facets of a child with autism's life and potentially alter the trajectory of his or her development, especially when the intervention is initiated at a young age (Harris & Handelman, 2000). However, the opportunity to provide these essential services early is dependent on first identifying the disorder in a timely manner. Fortunately, research has shown that an effective means to accomplish this goal is through the early screening of a large number of children to identify those at risk for autism who would benefit from a more thorough diagnostic evaluation. While several screeners specific to autism exist, the selection of an appropriate measure is dependent not only on psychometric properties and diagnostic accuracy, but also based upon comprehensiveness and the populations in which the measure is intended to be used.

A recently developed scale, The Baby and Infant Screen for Children with aUtIsm Traits (BISCUIT; Matson, Boisjoli, & Wilkins, 2007), is a comprehensive screening instrument that

measures the core symptoms of autism, in addition to symptoms potentially indicative of comorbid psychopathology and problem behavior. The primary purpose of the present study was to further increase the utility of the BISCUIT by identifying critical items that are most discriminative of the disorder and that can be used to compose an abbreviated scoring algorithm. A secondary purpose was to determine if the identified critical items are consistent with prior research indicating that specific social behaviors are distinguishing features of infants with autism. A description of the study as well as the implications of the results are preceded by an overview of autism as well as research that will set the foundation for the current investigation.

#### **History of Autism**

A thorough understanding of the definition of autism is best accomplished in relation to the history of the disorder (Matson & Minshawi, 2006). From Leo Kanner's (1943) initial account to the present depiction in the DSM-IV-TR, the conceptualization of the disorder has varied as the result of emphases on particular defining characteristics, confusion with other disorders, and in reaction to social and political pressures. Despite the changing face of the clinical entity we refer to as autism, a testament to Kanner's work is that his original account closely resembles our present interpretation.

In the seminal article, "Autistic Disturbances of Affective Contact," Kanner (1943) portrays the development of 11 children who, despite individual differences in development and symptoms, encompassed a unique disorder whose primary feature was a social disconnect evident from birth. Kanner described this social disconnect as "autism", a word that was unfortunately borrowed from the Swiss Psychiatrist Eugene Blueler. Blueler used it to describe a symptom of schizophrenia where the individual mentally departs into a private fantasy world characterized by delusional thinking and an inability to interact with others. Unfortunately the shared use of the word "autism" would contribute to confusion between the disorders for years to come.

Kanner elaborated on the social impairments characteristic of autism. The parents of the children he observed described their children as being happier when alone, acting like they were in a shell, and appearing hypnotized. Kanner pronounced that this tendency to shut out the social world was evident from birth, expressed by a lack of anticipatory posture in preparation of being picked up. As the children outgrew infancy, the salient features of their social aloneness took other forms. For instance, Kanner described children who appeared to have as much interest in

people as they did furniture. Upon entering a room, people were ignored at the expense of preferred toys or objects. As Kanner described, if an adult attempted to intrude upon the child's activity, the child would scorn the adult's offending appendage without acknowledging the appendage's owner (Kanner, 1943).

In addition to the predominant social deficits, Kanner described difficulties with communication and abstraction. Three of the 11 children were nonverbal and the remaining 8 lacked functional speech. However, communication difficulties were partially masked for some, as they possessed excellent articulation skills allowing them to pronounce complex words and recite such things as nursery rhymes, lists of presidents, and foreign phrases. This tendency to recite useless information was thought to be a consequence of excellent rote memories, an inability to use speech functionally, and the encouragement they received from their parents who were eager to hear any form of communication (LoVullo, 2009). Simple concepts such as the word "yes" took years for them to understand and even then were highly situation specific. For instance, one of the children, Donald, consistently said "yes" when he was asked to be picked up by his father, but did not use the word appropriately in other situations. Other abnormal language features included echolalia and difficulties with pronoun use (Kanner, 1943).

Kanner depicted the children's stereotypical behaviors, need for sameness, and aversion to sensory input. He described their strong desire to block out intrusions from the outside world, a feature that was first evident in their refusal of foods. Loud sounds were also disruptive and resulted in severe fear reactions to the extent that one of the children avoided the closet where the vacuum cleaner was housed. Kanner elaborated on their repetitive motor movements and what appeared to be an obsessive need for sameness in their environment. This was evident by behavioral outbursts in reaction to disruptions in the configuration of objects in their

environment, or when the sequence of events in a routine had been changed. Kanner commented on their unusual sensory preferences and the joy they expressed when taking part in rhythmic body movements.

Kanner described what he believed were associated features. Although the majority of the children resided in settings for the intellectually disabled, Kanner believed that they all had good intellectual potential with traces of marked intelligence. For the most part, all were described as physically normal; however, five had large heads, one had right sided convulsions, and several others displayed gross motor impairments contrasted with fine motor strengths. Especially noteworthy were the familial commonalities and parent child relationships. Kanner indicated that all of the children came from intelligent families, comprised of medical doctors, lawyers, scientists, and other college graduates. He believed that the parents were overly involved in academic and professional pursuits and possessed minimal ability to display warmth or affection towards others. He believed that this contributed to the development of autism, and unfortunately was a belief that served as a breeding ground for parent's guilt for years to come (LoVullo, 2009).

One year after Kanner described what would eventually be referred to as autism, Hans Asperger (1944), an Austrian physician, described four children with similar characteristics except that they had normal language and cognitive abilities. While Kanner's clinical description was written in English, Asperger's was written in German and did not receive widespread dissemination until 1991 when the German psychologist Uta Frith translated his work and included it into a book chapter dedicated to the disorder for which he became associated (Asperger, 1991). Similar to Kanner, the most salient features Asperger portrayed were difficulties engaging in social interactions and understanding the rules that underlie such

interactions. Social behaviors that most individuals exhibit with minimal effort were drastically impaired in these children as evident by their fleeting eye contact, disregard for rules of physical proximity, and difficulties using gestures to aid in conversational exchanges. Similar to Kanner's description, Asperger's children also displayed unusual stereotypical behaviors. For instance, they had intense interests in particular topics which became apparent in conversations that would regularly turn into awkward, one-sided diatribes. In contrast to Kanner, Asperger described children who seemed less socially impaired with less intense stereotypical behaviors. While autism became associated with nonverbal individuals and cognitive impairment, Asperger's Syndrome did not (LoVullo, 2009).

#### **Definition of Autism**

From Kanner (1943) until present, several iterations of autism have been proposed. Even Kanner himself strayed to an extent from his original conceptualization. For instance, Kanner and Eisenberg (1956) restated the basic tenants of the 1943 study and proposed a new set of guidelines. They concluded that autism could be differentiated from mental retardation and schizophrenia by an obsessive adherence to rituals and social isolation beginning early in life. They stated that the genesis of the disorder was due to unknown, heterogeneous biological mechanisms and parental coldness. The latter assertion was harmful and led to a generation of so called "refrigerator parents," who were burdened with the false belief that their child's affliction was the result of obsessive and cold rearing practices. Eisenberg (1956) presented the results of an outcome study involving 63 adolescents with autism. He reiterated that autism could be differentiated from seemingly associated disorders by social aloneness and need for sameness in the environment. He concluded that, although autism and schizophrenia share abnormalities in language, the former lacks evidence of delusions and hallucinations.

Furthermore, while autism is often characterized by severe intellectual disability (ID), children with only ID lack the affective disconnect that is central to autism.

In a departure from Kanner and Eisenberg, Creak (1961) developed nine criteria which she believed represented childhood schizophrenia, when in fact it became apparent she was describing autism. Her criteria included abnormalities in social relationships, resistance to change, captivation by parts of objects, unusual body movements and postures, abnormal reactions to tactile input, intellectual disability, lack of speech or loss of the use of speech, and oddities in the use of speech. Because these criteria were not well operationalized nor was it specified how they were unique to autism, they were incorporated into future definitions of the disorder as well as associated diagnostic instruments (Matson & Minshawi, 2006).

An influential researcher who helped clarify the boundaries between autism and schizophrenia was Michael Rutter (1968). He described three groups of supposed psychotic disorders that could be differentiated by age of onset: onset during adolescence corresponding to the schizophrenia exhibited in adults; onset between the ages of 3 and 5 preceded by a period of nondescript illness; and onset during early infancy corresponding to Kanner's autism. He stressed the unfortunate use of the term "autism" to both describe a disorder (Kanner, 1943) and a symptom (Bleuler, 1911), referring to Bleuler's use of the term to describe the social withdrawal in persons with schizophrenia. Additionally, autism could be differentiated from schizophrenia by autism's higher male to female ratio, the fact that parents of children with autism are generally of a higher social class, and the fact that a family history of autism does not increase one's chances of producing offspring with schizophrenia.

Rutter (1978) provided an influential definition of autism consisting of an age of onset prior to 30 months, impairments in social relationships and language, and the presence of

stereotypical behaviors. The 30 month cutoff was established to differentiate autism from other psychotic disorders with later onsets. In addition, similar to the multiaxial system that would be adopted in the DSM, Rutter believed it was important to interpret the expression of autistic symptoms through the prism of a child's developmental level and medical and neurological status. A competing definition of autism at the time was developed by Edward Ritvo and the National Association for Autistic Children (Ritvo, 1978). It also specified an age of onset by 30 months, but included four categories: (1) disturbances in social or motor development, (2) hyperor hypo-sensitivity of the sensory systems, (3) disruption in the use of speech, understanding of language, and use of nonverbal communication and (4) disturbance in the formation of appropriate social relationships, insistence on sameness, and inappropriate use of objects or difficulty with the symbolic use of objects. Other associated features included mood liability and the presence of psychotic symptoms.

Although Rutter and Ritvo's definitions were similar in many regards, their differences were more apparent. Both acknowledged the heterogeneity of symptom expression, emphasized social and language impairments, and specified symptom onset by the age of 30 months.

However, Rutter underscored the importance of stereotypical behaviors, while Ritvo highlighted abnormal sensory behaviors. The NSAC definition included developmental disturbances while Rutter believed that autistic symptoms should be interpreted as a function of development. It has been suggested that differences in the definitions were the result of Rutter developing his in light of the history of the disorder in combination with an extensive analysis of research, while Ritvo's was intended to help the NSAC create political leverage to increase research and treatment services (Matson & Minshawi, 2006; Rutter & Schopler, 1988).

Autism was not included in the DSM as a separate clinical entity until 1980 with the release of the DSM-III from the APA (1980) and its inclusion in a group of childhood disorders referred to as Pervasive Developmental Disorders (PDD). In prior versions of the DSM, children who exhibited autistic like symptoms were classified under childhood schizophrenia. The DSM-III was considered a significant development in that it rejected the relationship (excluded a dual diagnosis) between autism and schizophrenia and incorporated a multiaxial approach to classification. However, the DSM-III was not without problems. One problem was the insertion of Childhood Onset Pervasive Developmental Disorder (COPDD) to include children who exhibit autistic features after 30 months of age, but who are not synonymous with the disintegrative disorder described by Heller. As Volkmar, Stier, and Cohen (1985, p. 1450) noted, children diagnosed with COPDD are indistinguishable from those diagnosed with infantile autism and "age of onset" should more accurately be labeled "age of recognition." Another problem was the inclusion of the diagnosis of Residual Autism in reference to children who, at one point, met criteria for autism but later outgrow the symptoms, implying the disorder may be impermanent (Volkmar, Klin, & Cohen, 1997).

The definition of autism underwent a major overhaul with the introduction of the DSM-III-R. Not only was the diagnostic concept of autism broadened, but specific criteria were included for each domain (social interaction, communication, and stereotypical behaviors). Sixteen criteria were developed and a total of eight criteria were required with a specific number per domain, for a diagnosis. Other changes included removing the diagnosis of COPDD, removing the exclusion of a dual diagnosis of autism and schizophrenia, and removing the age of onset criterion. Despite improvements over the DSM-III, a major drawback of the revision was

the broadened definition of autism, which resulted in an increase in the number of people diagnosed.

Partially in reaction to changes in the International Statistical Classification of Diseases and Related Health Problems, 10<sup>th</sup> Edition (ICD-10, World Health Organization, 1992) and shortly after the DSM-III-R was released, the DSM-IV was developed along with changes to the Pervasive Developmental Disorders. The development process consisted of extensive literature reviews for each of the diagnostic categories and field trials examining the diagnostic accuracy of prior and existing classification systems (i.e., DSM-III, DSM-III-R, and ICD-10). The research resulted in decisions to reinstate an age of onset and to narrow the diagnostic boundaries of autism. Goals of the DSM-IV definition included providing user friendly definitions of autistic disorders that were compatible with the ICD-10 definitions and which also balanced research and clinical needs (Volkmar et al., 1997). A major change in the DSM-IV was the addition of three new PDD: Rett's Disorder, Childhood Disintegrative Disorder (CDD), and Asperger's Disorder. Although the addition of all three disorders was controversial, none was as controversial as the inclusion of Asperger's Disorder due to disparate definitions among the three major classification systems.

The concept of autism spectrum disorders continues to evolve with the next iteration of definitions set to be released in 2013 with the DSM-V. Clues to the final product have been released by the APA (2011) with the following proposed changes. The category of Pervasive Developmental Disorders will be changed to Autism Spectrum Disorders (ASD). The diagnostic category will be conceptualized as a spectrum varying by verbal skills, severity, and associated genetic and medical factors. Social and communicative abnormalities are considered indivisible and as a result the two domains will be combined into one. Asperger's Disorder will be removed

because it is viewed as an unnecessary category that is unreliably diagnosed and does not represent a unique subgroup consisting of distinct etiology, cognitive profile, developmental course, or treatment requirements. Rett's Disorder will be removed because it contains autistic-like symptoms that tend to be less permanent, and because, currently, the DSM defines disorders behaviorally instead of by a specific etiology. In regards to age of onset, it is unclear if the DSM-V will institute changes; however, the proposed definition describes that, although symptoms must be present early in life, they may not fully develop until social demands exceeds the individual's capabilities (APA, 2011).

#### **Current Diagnostic Criteria**

Currently there are two classification systems used by clinicians and researchers to identify mental health disorders: the DSM-IV-TR (2000) and the International Statistical Classification of Diseases and Related Health Problems, 10<sup>th</sup> Edition (ICD-10; World Health Organization, 1992). The ICD-10 is a classification system of disease, associated signs, and symptoms that is used across the world as a common language to track mortality and morbidity statistics. Chapter 5 of the ICD-10 is a section that classifies mental health disorders and was developed in conjunction with the DSM. For the purposes of the present paper, and because the diagnostic criteria of the DSM-IV-TR and the ICD-10 are very similar, the following description will be limited to the DSM-IV-TR.

The DSM-IV-TR is a current manual published by the APA and employs a categorical approach to the classification of mental health conditions. The manual consists of 16 primary diagnostic categories, of which the first section, Disorders Usually First Diagnosed in Infancy, Childhood, or Adolescence, contains the Pervasive Developmental Disorders. Use of the DSM-IV-TR results in clinical information presented in accordance with a multiaxial system that provides a framework for describing mental health conditions in relation to relevant medical, environmental, and psychosocial variables. There are five axes. Axis I is for reporting mental health disorders. Axis II is devoted to personality disorders, pronounced maladaptive personality features, and intellectual disabilities. Axis III is for reporting medical conditions that are pertinent to the conceptualization or management of an individual's mental disorder. Axis IV is reserved for psychosocial or environmental factors that may affect the treatment or course of a mental disorder. Such factors include problems related to an individual's social circumstances, primary support group, educational status, occupational status, economic status, access to health

services, and legal status. Finally, Axis V is used to provide an indication of the patient's overall occupational, psychological, and social functioning along a 0 to 100 scale (APA, 2000; LoVullo, 2009).

Pervasive Developmental Disorders is the umbrella category within the DSM-IV-TR that contains Autistic Disorder, Childhood Disintegrative Disorder, Asperger's Disorder, Rett's Disorder, and Pervasive Developmental Disorder Not Otherwise Specified (PDD-NOS). In general, these disorders are characterized by pervasive abnormalities in social skills, language and communication, with the presence of stereotypical behaviors. Symptoms are typically noted within the first few years of life, with a presentation that can be heterogeneous and associated with intellectual disability and a host of other conditions and features. Because a detailed description of all the PDD is beyond the scope of this paper, only Autistic Disorder and PDD-NOS will be discussed in detail.

#### **Autistic Disorder**

According to the DSM-IV-TR (APA, 2000) the primary characteristics of Autistic Disorder include abnormal development in social skills and communication with the presence of stereotyped behaviors and restricted interests. A diagnosis requires six total criteria met with two from the domain of social impairment and one each from communication, and restricted and stereotyped behaviors. In addition, onset must occur before the age of 3 years and symptoms cannot be better accounted for by Childhood Disintegrative Disorder or Rett's Disorder. Criteria include: 1) impairment in the use of nonverbal behaviors for use in social interactions, 2) failure in forming social relationships appropriate for one's developmental level, 3) lack of spontaneously seeking to share things of mutual interest with others, and 4) lack of reciprocity socially or emotionally (e.g., playing games with others, preferring to be alone, etc.).

Impairments in communication cut across verbal and nonverbal skills. Criteria include: 1) lack of speech to communicate; 2) difficulty in sustaining communicative exchanges with others; 3) use of language that is stereotypical and/or repetitive; and 4) lack of play that is make believe or lack of developmentally appropriate play that is social and involves imitation. The third area of dysfunction embodies behaviors that are stereotyped and contains the following criteria: 1) preoccupation with interests that are either considered overly narrow and specific or abnormally intense; 2) obedience to routines that are repetitive and nonfunctional; 3) repetitive and stereotypical motor behaviors; and 4) captivation or fascination with parts of objects.

#### Pervasive Developmental Disorder Not Otherwise Specified

PDD-NOS is designated for individuals who have a severe disturbance in reciprocal social skills that occur in conjunction with impairments in verbal and nonverbal communication skills and/or with stereotyped behaviors. In addition, criteria cannot be concurrently met for Schizophrenia, Schizotypal Personality Disorder, Avoidant Personality Disorder, or a better defined Pervasive Developmental Disorder. PDD-NOS includes atypical autism which represents individuals who do not meet criteria for Autistic Disorder because of symptomotology that is not typical, that is below threshold, or with an onset after 3 years of age (Towbin, 2005).

Towbin (2005) suggested four clinical uses for PDD-NOS. First, it can be used under less than ideal circumstances when a person's diagnostic history is unclear and a temporary diagnosis is needed until more accurate information can be obtained. Second, it can be used for higher functioning individuals who have subthreshold symptoms or abnormal language use and stereotyped behaviors, but for whom social abnormalities are too severe to disregard a diagnosis on the autism spectrum. Third, the definition can be designated for rare instances where autistic symptoms emerge after three years of age but the person does not meet criteria for CDD.

Finally, Towbin (2005) suggested that PDD-NOS may be used for a heterogeneous group of conditions that share an early onset and abnormalities in social relatedness but also present with unrelated symptoms that may be indicative of a disorder not within the autism spectrum (e.g., schizoid type disorder).

#### Features of Autism in Early Childhood

Although the DSM states that autistic symptoms should be apparent within the first few years of life, research suggests that precursor symptoms are apparent as early as the first or second years of life. However, determining what these symptoms are is complicated by the fact that infancy is characterized by rapid change. For instance, a behavior that represents typical development during one segment of life may indicate abnormal development if it persists. While play that is exploratory is typical for children younger than 12 months of age, abnormality may be noted if this behavior persists in the absence of more advanced or symbolic forms of play. Another factor that may interfere with the detection of autism at an early age is the fact that regression commonly occurs between 18 and 20 months of age. This consists of apparently normal development followed by a disruption in skill development or deterioration in social or language skills. Studies are unclear whether regressive autism represents a unique subtype, a varied expression of a similar genetic underpinning, or a phenomenon similar to that displayed in CDD (Chawarska & Volkmar, 2005).

Regression occurs in approximately 15-40% of children with autism (Baird et al., 2008). While it's common for parents to report dramatic changes in their child's behavior and skill level, some report a more gradual regression. Functioning is often perceived as normal prior to regression; however, retrospective methods suggest subtle preexisting abnormalities (Baird et al., 2008). Regression is typically succeeded by a regaining of skills to some extent; however, researchers have reported various outcomes. For instance, Kobayashi and Murata (1998) examined the long-term prognosis of children with regressive autism and found the majority to have higher rates of epilepsy and a lower language attainment. However, Lord (2004) found that the time between word loss and the reemergence of words was only 4-5 months in children who

would go on to speak in phrases at 5 years of age. Regression is also displayed in children with CDD who experience several years of seemingly normal development, followed by a broad deterioration that occurs between 3 and 5 years of age (Volkmar, Koenig, & State, 2005). Although the onset of regression can be gradual or sudden, its occurrence is striking because of its severity and global impact.

The early identification of autism requires that there be reliable symptoms of the disorder during infancy. Potential symptoms have been identified through case studies, retrospective parent reports, videotape analyses, and prospective studies. A description of this research follows with the exception of prospective studies, which will be integrated into a later section on autism screening. It is important to note that many of the following studies lack appropriate control groups, making it unclear if the symptoms listed are specific to autism or if they are features common to a broader range of developmental disabilities (Gray & Tonge, 2001)

#### **Case Studies**

While case studies provide detailed information pertaining to a single child with autism, generalizability of findings are often limited by small sample sizes and absence of control children. Nevertheless, a common theme is the description of children who are profoundly socially disconnected, who lack appropriate eye contact, and appear more interested in objects than in people. For instance, Kanner (1943) noted that, although many of the abnormal features of autism become pronounced as the child develops the requisite cognitive and motor skills, the underling social aloneness is evident from birth.

Dawson, Osterling, Meltzoff, and Kuhl (2000) described the development of an infant with autism who was extensively examined from birth. Until the age of six months, the child was described as hypersensitive to touch and easily started, with muscle tone that alternated

between hypertonia and hypotonia. After the age of six months, social abnormalities began to appear in the form of poor eye contact, lack of vocal imitation, and absence of imitative play. Klin et al. (2004) reported on a 15-month-old female who was administered a battery of assessments covering adaptive behavior, cognition, and symptoms of autism. At 15 months, she appeared disinterested in social exchanges and displayed an impaired use of eye contact and nonverbal gestures. Rutherford (2005) examined the medical records and detailed journal of a mother with fraternal twins. One child had autism while the other did not. During the first 6 months of life, both children appeared to develop typically; however, prior to their first birthdays, the child who would later be diagnosed with autism developed abnormalities in language, social skills, sleep patterns, and sensitivity to pain. Additionally, his use of vocabulary regressed as did his social engagement (eye contact, affection, and interactive play), which were especially pronounced in comparison to his twin.

#### **Retrospective Parent Report**

Studies utilizing retrospective parent report suggest that the early symptoms of autism involve deficits in social interaction and problems with the regulation of arousal (Charwarska & Volkmar, 2005). Although informative, this type of research has potential limitations. For one, it is based upon the recall of prior information, which is likely to contain error. Second, a diagnosis of autism later in life may bias a parent's recollection of behavior at an earlier time. Furthermore, the accuracy of observations may be dependent of the parent's age, educational attainment, intelligence, and overall awareness of their child's symptoms as an infant (Gray & Tonge, 2001).

Consistent with most research related to the early symptoms of autism, retrospective parent report studies often fail to include appropriate comparison groups consisting of children

with developmental disabilities who do not have autism. Consequently, symptoms reported in these studies may not be specific to autism. For instance, Young, Brewer, and Pattison (2003) interviewed parents of 153 children with autism. The mean age at which developmental abnormalities were first noted was 15 months. Areas of concern included social awareness, eye contact, shared enjoyment, social interactions, and gross motor skills. De Giacomo and Fombonne (1998) used the introductory section of the Autism Diagnostic Interview (ADI; Le Couteur, Rutter, Lord, & Rios, 1989) and found that the early developmental concerns for children later diagnosed with autism included language and speech development, socioemotional response, medical difficulties, and non-specific behavior difficulties. Ozonoff, Williams, and Landa (2005) used the Early Development Questionnaire to compare the early symptoms of children with regressive autism, unclear regressive autism, and early onset autism. The early onset group had the greatest number of symptoms; however, eight children in the regressive group and six in the unclear regressive group showed early social abnormalities. Symptoms included problems with joint attention, social games, pretend play, and showing objects of interest.

Fortunately, several retrospective parent report studies include children with non-autistic developmental disabilities. For instance, Vostanis et al. (1998) compared the early symptoms of children with autism, Asperger's Syndrome, atypical autism, learning disability, and semantic pragmatic disorder, and found that items most predictive of autism included repetitive play with toys, lack of appropriate pointing, and appearing to be deaf. Using the Vineland Adaptive Behavior Scales (VABS; Sparrow, Balla, & Cicchetti, 1984), Klin, Volkmar, and Sparrow (1992) found that compared to children with only developmental disabilities, children with autism were less likely to play simple interaction games, reach for familiar people, demonstrate a

readiness to be picked up, and show an interest in other children. Wimpory, Hobson, Williams, and Nash (2000), administered a semi-structured interview of social engagement, the Detection of Autism by Infant Sociability Interview (DAISI), to compare the responses of 10 children with autism to 10 children with non-autistic developmental disabilities. Results indicated that the children with autism had significantly impaired skills related to eye contact, raising their arms in anticipation of being lifted, turn taking during conversation, sharing objects with others, and using and responding to pointing.

#### **Videotape Analysis**

Another method utilized to identify early symptoms of autism is through retrospective videotape analysis of children later diagnosed with autism. An advantage of this method is that symptoms can be directly observed rather than relying on a parent's biased recollections of their child's development (Goin & Myers, 2004). However, studies based upon this methodology commonly lack appropriate control groups.

Lösche (1990) compared children with autism to those with typical development using Piaget's developmental sequence as the outcome variable and found that marked differences emerged at 13 months with the autism group continuing to engage in sensorimotor behaviors. However, a major flaw of this study was its reliance on the questionable validity of Piaget's developmental construct. Adrien, Lenoir, Martineau, and Perrot (1993) compared the home movies of 12 infants later diagnosed with autism to 12 infants who were typically developing. Videos were coded using the Behavioral Summarized Evaluation Scale (BSE, Barthelemy, Adrien, Tanguay, & Garreau, 1990). During the first year of life, five behaviors were indicative of autism: lack of a social smile, impaired social interaction, impairments in the use of facial

expressions, hypotonia, and inconsistent attention. Beyond 12 months, differences remained and became more pronounced.

A significant step in the analysis of home videotapes was the use of coding systems for behavior. Osterling and Dawson (1994) rated the social, communicative, and repetitive behaviors of 11 children with autism to 11 with typical development. The researchers attempted to minimize some of the problems with prior videotape studies by comparing children at the same age under similar conditions (i.e., first birthday parties). Differences were apparent between groups in the areas of social skills, joint attention, and autistic behavior. Frequency of looking at other children was the best predictor of diagnosis, and when combined with "failing to orient to name", "showing", and "pointing," over 90% of children were classified accurately. However, the identified symptoms were not specific to autism and were not useful in predicting whether the child would go on to develop an intellectual disability.

Werner, Dawson, Osterling, and Dinno (2000) reexamined the videotapes from Osterling and Dawson (1994) in addition to 8 new children between the ages 8 to 10 months. An elaborate coding system was used to ensure comparability between video segments. Group differences in regards to the core domains of autism were apparent when children with regressive autism were removed. Items that best differentiated groups involved: orienting to one's name spoken and looking towards others (Receveur et al., 2005). Using a similar coding system, Mars, Mauk, and Dowrick (1998) investigated whether the intensity of early symptoms of autism corresponded to specific diagnoses (i.e., autism, PDD-NOS, typical development). Behaviors related to joint attention were the strongest indicators of ASD; however, data supporting a continuum of intensity across diagnoses were less convincing.

Saint-Georges et al. (2010), conducted an extensive review of studies employing retrospective videotape analyses, and found that the symptoms indicative of a risk of autism during the first year of life included deficits in social interest (lack of eye contact, pointing), communication (use of speech and gestures to communicate, responsiveness to name), and the expression of affect. In addition, symptoms become more salient during the second year of life as deficiencies in language become more pronounced and behaviors related to emotionally reciprocity and joint attention endured.

#### **Associated Features Relevant to Early Identification**

In addition to the early emergence of social skills impairments, communication difficulties, and repetitive behaviors that are part of an ASD diagnosis, the disorder is associated with a host of medical and psychiatric disorders, as well as problem behaviors. However, the condition that is most commonly associated with autism is intellectual disability (Edelson, 2006).

#### **Intellectual Disability**

Previously referred to as mental retardation, ID is presently defined by substantial deficits in adaptive and intellectual functioning, which is a conceptualization derived from Heber (1959) and the American Association of Mental Retardation (AAMR). Founded in 1876, the AAMR, now the American Association of Intellectual and Developmental Disabilities (AAIDD), has played a major role in developing definitions of ID (LoVullo, 2009).

Heber's (1959) definition required deficits in intellectual functioning and impairments in social skills, learning, and maturity, all of which were later referred to as adaptive behavior. This was followed by a new yet controversial definition of ID as part of the ninth edition of the AAMR (Luckasson et al., 1992). It specified that ID consists of considerable limitations in present functioning with significant deficits in intellectual functioning in at least two of ten adaptive skill areas: home living, communication, social skills, self-care, self-direction, functional academics, health and safety, work, and leisure. Other changes included an age of onset of 18 years and a reference to present functioning, implying that intellectual disability was potentially temporary (LoVullo, 2009). Probably the most controversial changes were raising the intelligence quotient cutoff from 70 to 75 and removing severity of impairment as a means to grade levels of ID. The standard score was likely increased to account for the standard error of measurement (Greenspan, 1999); however, it resulted in a doubling of the prevalence of ID.

Severity of impairment (mild, moderate, severe, profound) was replaced with a focus on the intensity of supports needed, in an effort to shift the emphasis on providing supports to help individuals with ID reach their fullest potentials (AAIDD, 2011; LoVullo, 2009).

In stark contrast to the controversial AAMR definition (Luckasson et al., 1992), the 1996 APA definition (Cuskelly, 2004) consisted of: (a) significant deficits in intellectual functioning; (b) significant deficits in adaptive functioning; and (c) intellectual and adaptive deficits prior to the age of 22 years. Significant deficits were operationally defined as scores two standard deviations below the mean. The current DSM-IV-TR definition of intellectual disability includes substantial deficits in general intellectual functioning and corresponding limitations in adaptive skill domains: social/interpersonal skills, self-care, home living, communication, work, safety, health, and leisure. Intellectual and adaptive functioning are measured using appropriately normed instruments (Sparrow et al., 1984) and the onset of adaptive and intellectual deficits must occur before the age of 18 years. In addition, intellectual deficits are defined as intelligence quotient (IQ) scores of 70 or below.

The DSM-IV-TR (APA, 2000) provides severity levels of intellectual disability. Mild ID, with IQ scores between 50-55 and 70, represents 85% of the ID population. These individuals typically obtain academic competency at the sixth-grade level and live as adults with minimal support. Moderate ID, with IQ scores between 35-40 and 50-55, represents 10% of ID population. This group commonly obtains academic skills up to the second grade level and is employed in semi-skilled work with moderate supervision. Severe ID, with IQ scores between 20 and 34, constitutes 3%-4% of the ID population. These individuals typically acquire functional academics, work performing simple tasks, and live with caregivers or in the community with extensive supervision. Profound ID, consisting of IQ scores below 20, constitutes 1%-2% of the

ID population. Individuals with profound ID require extensive support in most aspects of living. Finally, the DSM provides a catchall category of ID when IQ scores cannot be established, which is referred to as ID, severity unspecified (APA, 2000; LoVullo, 2009).

The prevalence of intellectual disability is dependent on how the construct is defined. Medically, it is conceptualized as the result of specific pathology, whereas statistically it is defined by subthreshold scores on normed intelligence and adaptive measures. Historically, the definition has been altered by such things as the deletion and re-addition of severity levels, emphasis on levels of support rather than severity of impairment, and changes to the required age of onset. There has been debate about the necessity of adaptive behavior in defining ID, and many studies have focused solely on IQ as a means to define the disorder. Another problem is the use of a binary classification of severity (mild and severe), instead of the existing four levels (mild, moderate, severe, profound). Nevertheless, in a comprehensive review of prevalence studies, Leonard and Wen (2002) found that, using a binary classification system, the average prevalence for severe ID was 3.8 per 1000 and the average prevalence for mild ID was 35.0 per 1000.

**Differential Diagnosis.** Seventy to eighty percent of individuals with ASD also have ID, and 40% of individuals with ID also meet criteria for ASD (La Malfa et al., 2007; Matson & Shoemaker, 2009). Differential diagnosis can be formidable considering that autism is characterized by, and ID associated with, disturbances in communication and social skills with the presence of stereotypical behaviors. However, the distinction between the two conditions is important considering that IQ is often a prognostic indicator and outcome measure for early intervention programs (Matson & Shoemaker, 2009).

Despite a high rate of comorbidity (Matson & Nebel-Schwalm, 2007), our current classification system provides little guidance in regards to differential diagnosis. For instance, the DSM-IV-TR states that in individuals with ID, Autistic Disorder should only be considered when there are deficits in communication and social skills with other autistic behaviors present. Towbin (2005) suggests that impairments must be compared in relation to the child's general level of intellectual functioning; however, even when standardized instruments such as the VABS-II are used, it is unclear how low specific domain scores must be to qualify for diagnosis. For infants, differentiating ID from ASD is further complicated by the fact that instruments used to assess developmental functioning are unreliable in predicting later intellectual functioning. Fortunately, research has resulted in autism assessment measures that include items useful in differential diagnosis.

### **Comorbid Psychopathology**

There is sufficient research to conclude that autism co-occurs with other forms of psychopathology (LoVullo, 2009; Matson & Nebel-Schwalm, 2007). For instance, one condition frequently associated with ASD is Attention Deficit Hyperactivity Disorder (ADHD). Although the disorders are quite different, it is common to see symptoms of each in the other. In fact, hyperactivity, inattention, and impulsivity are often present in individuals with autism (APA, 2000; Christopher Gillberg & Billstedt, 2000). Anxiety is also estimated to occur at greater levels and at increased rates in individuals with autism than in the general population (Bellini, 2004; Gillott, Furniss, & Walter, 2001; Kim, Szatmari, Bryson, Streiner, & Wilson, 2000; White, Oswald, Ollendick, & Scahill, 2009). The value of early identification of anxiety is magnified by the fact that, in the general population, these problems persist into adulthood if not treated early (as cited in Davis III et al., 2011; Spence, Rapee, McDonald, & Ingram, 2001).

Other comorbid conditions include depression, tic disorders, and feeding disorders. Depression occurs in at least 2% of individuals with autism and greater in those with Asperger's Disorder (Ghaziuddin, Ghaziuddin, & Greden, 2002; Ghaziuddin, Weidmer-Mikhail, & Ghaziuddin, 1998; Lainhart, 1999), while Tourette Syndrome occurs in 6.5% of children with autism (Baron-Cohen, Scahill, Izaguirre, Hornsey, & Robertson, 1999). Feeding disorders are not only more common in children with autism and developmental disabilities (80%-90% prevalence), but they are more likely to refuse foods by texture, type, and quantity, which can lead to severe health consequences if left untreated (Marchand & Motil, 2006; Schreck, Williams, & Smith, 2004). Comparison prevalence rates of comorbid psychopathology in the general population include 3%-7% for ADHD, 2%-9% for Major Depression, .001%-.030% for Tourette's Syndrome, and 25% for feeding disorders (APA, 2001; Marchand & Motil, 2006).

#### **Problem Behavior**

Another area of concern relevant to early identification is problem behavior. Children with ASD are more susceptible to problem behavior than those with other developmental disabilities (Eisenhower, Baker, & Blacher, 2005; Matson, Boisjoli, Rojahn, & Hess, 2009). This is alarming considering that possessing a developmental disability already increases one's risk. For instance, Feldman, Hancock, Rielly, Minnes, and Cairns (2000) interviewed parents of children with developmental disabilities and 42% indicated that their child had problem behavior. Problem behavior that develops in childhood is likely to maintain or get worse if not addressed. Myrbakk and Von Tetzchner (2008) found that 20% of a sample of adolescents and adults with ID were rated as having at least on severe problem behavior. In addition, those with severe ID or autism were more likely to have problem behavior. In a research review, Murphy, Hall, Oliver, and Kissi-Debra (1999) reported problem behavior prevalence rates of 8-15% for

individuals in institutions and 2-4% for adults living in the community. They also found that self-injury is common in children with ID (4-12%), and the severity and prevalence appear to increase drastically between the ages of 5 and 25. Problem behaviors also negatively impact daily activities and are distressing to parents (Hartley, Sikora, & McCoy, 2008). In addition, they create obstacles for schools, interfere with treatment goals, and increase the risk of isolation from inclusive activities (National Research Council, 2001; Eisenhower et al., 2005).

Luckily, there is substantial evidence for effective treatment (Horner, Carr, Strain, Todd, & Reed, 2002; Matson & LoVullo, 2008). Horner, Carr, Strain, Todd, and Reid (2002) summarized research on the behavioral treatment of problem behavior in children with autism and found that interventions resulted in at least an 80% reduction in problem behavior in 66% of studies, and 90% reductions were reported for all types of problem behavior (e.g., aggression, self-injury, disruption, pica, food refusal, etc.). A diagnosis of autism was not related to the intervention selected or the effectiveness of the intervention; however, the use of functional assessment methodology increased the probability of a successful intervention. Furthermore, considering that children with ASD are at an increased risk for problem behavior that is likely to maintain or worsen over time without intervention, early screening and treatment for these behaviors is recommended (Horner et al., 2002; Kurtz et al., 2003).

#### **Prevalence**

The importance of the early identification and treatment of autism in children is magnified by the disorder's increased prevalence. For instance, the Centers for Disease Control (CDC; Rice, 2009) reported prevalence rates for autism of 6.7 per 1,000 in the year 2000 and 9.0 per 1,000 in the year 2006. Fombonne (2005) calculated rates of 6.0 per 1,000 for all ASD, 1.3 per 1,000 for Autistic Disorder, 2.1 per 1,000 for PDD-NOS, 0.26 per 1,000 for Asperger's Disorder, and 0.02 per 1,000 for Childhood Disintegrative Disorder. Our current rates are especially remarkable considering that the first prevalence estimate for autism, calculated by Lotter (1966), resulted in a figure of 0.41 per every 1,000.

This upward trend in the prevalence has created a frenzy among parents and healthcare professionals who believe they are in the midst of an outbreak of a condition that not only interferes with the central human quality of being social, but is also associated with poor long term outcomes (LoVullo, 2009). However, it is unclear to what extent the increase in diagnoses corresponds to an increase in people who actually have the disorder. Researchers argue that much of the change can be accounted for by other factors. For instance, Wing (2002) reviewed 39 studies investigating potential explanations, and found that the prevalence of ASD altered in concert with changes in the diagnostic criteria. As the definition of autism expanded, individuals who previously would have been on the cusp of meeting the diagnostic requirements now qualified. Wing also proposed that increase could be attributed to a greater awareness of autism and the availability of intensive services designed specifically for children with the disorder.

Another proposed explanation is diagnostic substitution, which is the notion that as one diagnosis becomes popular, it is given more frequently as an alternative to a previously diagnosed disorder (LoVullo, 2009; Matson & Minshawi, 2006). Several studies support this

explanation. Croen, Grether, Hoogstrate, and Selvin (2002) investigated changes in the prevalence of autism over a seven year period as reported by California's Department of Developmental Services and found that increases in ASD co-occurred with decreases in ID. Shattuck (2006) examined changes in the prevalence of autism in the United States special education system from 1984 to 2003 and found that ASD diagnoses increased by 2.5 per 1000 while ID and learning disability declined by 2.8 and 8.3 respectively. Similarly, Coo (2008) examined the administrative prevalence of autism in the District of Columbia between 1996 and 2004 and concluded that diagnostic substitution accounted for at least one third of the increase.

Arguments have been made against the proposed casual factors above. In a stern rebuke to diagnostic substitution, Blaxill, Baskin, and Spitzer (2003) claimed that Croen et al.'s (2002) conclusions were based upon statistical anomalies, diagnostic biases, and unreliable data; and that, in effect, the authors were blocking funding to an increasingly severe epidemic. Volkmar et al. (1997) argued against causation based upon a broadened diagnostic concept, and stated that although the definition of autism broadened from the DSM-III to the DSM-III-R, it narrowed with the creation of the DSM-IV (LoVullo, 2009). Hertz-Picciotto and Delwiche (2009) not only reported a 700%-800% increase in the incidence of autism in California since the 1990s, but based upon mathematical analyses, concluded that the proposed factors (i.e., diagnostic substitution, changing of diagnostic criteria, access to services, etc.) could only account for a fraction of the increase. The authors stated that other explanations have yet to be analyzed; therefore it is unclear if a true increase exists.

Along these lines, efforts to identify environmental causes for the increase have resulted in a myriad of suspects including gluten, heavy metals, pesticides, cable television, cleaning products, and most infamously, vaccines. Perpetuated by the efforts of Jenny McCarthy, the

Mumps, Measles, and Rubella vaccine (MMR) has received the most attention. The origin of this hypothesis was put forth by a British surgeon, Andrew Wakefield (1998), in his report of 12 children hospitalized with gastrointestinal symptoms. Wakefield proposed that the MMR vaccine caused an allergic-like reaction in the body creating an opioid effect, which resulted in neurological damage and autism. Since the release of his article in 1998, several investigators have published studies contrary to his findings (Honda, Shimizu, & Rutter, 2005; Smeeth et al., 2004), and just recently the *Lancet*, a high impact medical journal, retracted his article.

#### **Outcomes**

The importance of the early identification and treatment of autism is highlighted by evidence that suggests poor long-term outcomes. Although research on long-term outcomes is limited, especially compared to the abundance of research focusing on children with ASD, available studies depict poor social, educational, and occupational prognoses. Sources of information on outcomes are derived from autobiographical descriptions, clinical accounts, and studies that trace developmental trajectory. Although interesting, autobiographical descriptions are primarily limited to individuals with higher functioning forms of ASD such as Temple Grandin. There are also clinical accounts of younger adults with ASD; however, sources are often unsystematic and do not provide information on the developmental trajectory of the individual over time. Systematic studies that describe development from childhood to adulthood are most informative (Howlin, 2005).

Some of the first systematic outcome studies were conducted by Rutter and associates (Howlin, Goode, Hutton, & Rutter, 2004; Lockyer & Rutter, 1969, 1970; Rutter, Greenfeld, & Lockyer, 1967; Rutter & Lockyer, 1967) who examined 38 individuals, 16 years of age or older, who were diagnosed with autism between 1950 and 1960. During follow-up, more than 50% resided in hospital settings, 18% lived with their parents and were unemployed, 11% attended day programs, 8% were in residential settings, and 8% obtained paid employment (Howlin et al., 2004). Lotter (1974) reported on 32 children with autism initially investigated at 8-10 years of age and then again 8 yrs later. Of 22 who were out of school, only one was employed and 62% required extensive daily supports. Approximately 50% resided in a hospital setting, 2 lived at home, and 5 in day service facilities (Howlin, 2005).

Gillberg and Steffenberg (1987) presented data on 23 children from Sweden examined until the ages of 16-23. Initial IQ (>50) at diagnosis and communicative speech before the age of 6 were the best prognostic indicators. At follow-up, only one individual was considered independent, half were classified as functioning fairly well, and the other half were classified as functioning poorly. Additionally, 22% had increases in challenging behavior including self-injury and aggression. Eaves and Ho (2008) followed 48 young adults (mean age of 24), and evaluated their global outcomes in regards to work, independence, and friendships. In all, 46% were rated as having poor outcomes, 32% fair, and 17% good. Fifty-six percent resided with their parents, 35% in group homes, and 8% lived independently. Thirty-three percent had at least one friend and 10% a romantic relationship. Approximately half were employed at some point in their lives; however, jobs were mostly limited to part time and sheltered work.

Although evidence suggests that IQ is predictive of outcome, historically higher functioning individuals have also had compromised futures. For instance, Engstrom, Ekstrom, and Emilsson (2003) reported on 16 adults (18-49 years of age) with autism from Sweden.

Although not longitudinal, the study provided a useful snapshot of autistic functioning in adulthood. Most of the participants lived alone but required extensive support. One had regular employment, three sheltered employment, and one had state facilitated employment. None of the participants were married and only 5 had romantic relationships. Two individuals were rated as having good social adjustment, 12 fair, and 2 poor. Howlin et al. (2004) described 69 children with autism and performance IQs of 50 or greater who were followed up as adults (mean age of 29 years). Participants with IQs greater than 70 had better outcomes. Overall, 12 % were rated as having very good outcomes, 10% good, 19% fair, 46% poor, and 12% very poor. Only 7 percent were educated in mainstream schools and more than half left school without graduating.

Although 23 had some form of employment, only 8 worked independently. The majority did not have friendships, 26% had friendships consisting of shared interests, and 15% had acquaintances. Three individuals lived independently, 26 lived with parents, and more than half lived in residential facilities. Venter, Lord, and Schopler (1992) reported on 22 people with autism over the age of 18 with IQs over 60. Nonverbal IQ was predictive of academic achievement and overall outcome. Eight attained competitive employment; though, jobs were generally unskilled and took place in sheltered settings. In addition, only four lived independently.

# **Treatment and Importance of Early Identification**

#### **Treatment**

Although outcomes for individuals with ASD have been historically poor, the advent of effective treatments may improve long-term prognoses. By far the treatment modality with the most research and greatest support for efficacy is applied behavior analysis (ABA). In fact, in an evaluation of 19,000 journal articles, Palmieri, Valluripalli, Arnstein, and Romanczyk (1998) found that approximately 500 involved a combination of ABA and ASD.

The ABA instructional method most associated with early autism intervention is discrete trial teaching (DTT). DTT involves the use of: 1) task analysis, 2) teaching sub-skills until mastery, 3) repeated practice, 4) prompting and prompt fading, and 5) reinforcement. A discrete trial, the primary instructional unit that is repeated within a teaching session, consists of the following sequence: therapist instruction (e.g., "clap hands"), optional prompt, student response, and then reinforcement/corrective feedback. In traditional programs in order to promote success, early stages of instruction are highly structured, take place in environments with minimal distractions, and incorporate reinforcers that are systematically identified but are often unrelated to the target responses. However with success, instruction becomes less structured, takes place in more natural settings, and incorporates natural reinforcers. Comprehensive intervention programs that use DTT are associated with curriculums addressing a broad array of domains including communication, socialization, play, and daily living skills (Leaf, McEachin, & Harsh, 1999).

Naturalistic behavioral methods typically place a greater emphasis on natural reinforcers and incidental learning opportunities. This includes the use of toys and other stimuli from the child's natural environment as teaching items. The instructor systematically manipulates the

environment (e.g., blocks the door when the child attempts to open it) to motivate the child to elicit a target behavior (e.g., "Open the door please"), which is then reinforced (e.g., door is opened). A popular intervention package that uses such techniques is Pivotal Response Training (PRT). The main components of PRT include: child's choice of instructional materials to increase motivation, use of clear instructions, reinforcement of attempts at correct responding, reinforcement using items logically related to the instructional task, and use of a variety of stimuli from the outset of teaching to promote generalization (Koegel & Koegel, 1995).

Evidence for the effectiveness of ABA in the treatment of autism is abundant. Eikeseth (2009) evaluated the scientific merit and magnitude of results of treatment outcome studies corresponding to popular comprehensive treatment programs. Evaluations were based upon methods of diagnosis, research design, treatment fidelity, and the measurement of dependent variables. Overall, studies utilizing ABA had the strongest research methodology and the greatest magnitude of results. For instance, Smith, Groen and Wynn (2000) compared ABA treatment to a parent training program. At follow-up, children receiving ABA scored higher in regards to IQ, language, academics, and school placement with 25% successfully placed in general education environments. Eikeseth et al. (2007) compared ABA to an eclectic approach, and although group assignment was not random, at follow-up the ABA group showed greater improvement in language, IQ, adaptive functioning, maladaptive functioning, and social skills. Howard et al. (2005) evaluated ABA, one-to-one eclectic treatment, and public early intervention. Children receiving ABA had the highest scores on all dependent measures including communication, intellectual functioning, and adaptive behavior. Cohen et al. (2006) compared ABA to public special education. Results were partially confounded by the fact that

group assignment was based upon parental preference. Nevertheless, children receiving ABA scored higher on adaptive functioning, intelligence, and school placement.

# **Importance of Early Intervention**

The value of detecting autism early and providing timely treatment is supported by the neurosciences (Coonrod & Stone, 2005). Researchers propose that species have evolved to take advantage of predictable environmental stimulation during a sensitive period of brain development. Although synaptic connections are somewhat malleable throughout one's lifetime, their capacity to change is much greater during this sensitive period (Squire, 2008), which is the target for early intervention. Dawson (2008) hypothesized how the sensitive period is important for a specific aspect of social development in autism. Specifically, infants with autism have limited social interest and therefore do not gain the needed experience with faces and facial expressions leading to severe problems with facial processing later on in life. This is exacerbated by a lack of appropriate responding during a sensitive period when their brain is most ready to process facial information. A goal of early intervention is to improve the behavioral performance of facial processing during the sensitive period, therefore, improving the overall trajectory of this skill set. Dawson explains that similar logic can be applied to other areas of dysfunction related to autism.

Evidence supporting early intervention comes from data suggesting that children who receive intensive services at younger ages have better outcomes. In one of the first studies to investigate the impact of early autism intervention, Fenske (1985) compared the outcomes of 18 children with autism. Half received intensive behavioral intervention before 60 months of age and the other half after 60 months of age. Severity of autism was matched between groups. Comprehensive services included a school based treatment program, group home residential

intervention as needed, parent training, transition services, and follow-up services. In all, 67% of the earlier intervention group attained positive outcomes (i.e., lived at home and attended public school), while only 11% of the later intervention group attained positive outcomes. Harris and Handelman (2000) examined the relationship between age and IQ at admission with outcomes after an intensive ABA treatment program. Participants consisted of 27 children receiving services at the Douglas Developmental Disabilities center between 1990 and 1992. Treatment included DTT and naturalistic instruction. Children receiving treatment before 48 months of age and/or those with higher IQs were more likely to be placed in inclusive educational settings.

In addition to creating better long term outcomes for individuals with ASD, intervening early in a child's life may be more cost effective than providing extensive supports once he/she reaches adulthood. At the risk of coming across as insensitive by evaluating early intervention in regards to monetary costs, the harsh reality is that our public sector often lacks the necessary financial resources to fund public services. Therefore, it is crucial that we spend public money in a cost effective manner. Jacobsen, Mulick, and Green (1998) analyzed available public policy reviews to provide a cost benefit analysis of early intervention for young children with ASD. Educational costs for children and service costs for adults with developmental disabilities in the state of Pennsylvania were used in the analysis. It was estimated that per person cost savings from early intervention services would be \$1,686,061 to \$2,816,535 when calculating money that would be spent to support these individuals from 3 to 55 years of age.

# **Screening for Autism**

There is sufficient research to suggest that children with autism should receive intensive behavioral intervention at an early age (Matson, Wilkins, & González, 2008); however, this is of course limited by the extent to which autism can be detected at an early age. A method for facilitating early identification is through screening, which in this context refers to an assessment process that identifies a large portion of children who may be at risk for autism who would benefit from a more thorough diagnostic evaluation. A diagnostic evaluation leading to an ASD diagnosis is often a gateway for access to treatment services specific to autism.

Autism screening is widely recommended by health organizations including the

American Academy of Pediatrics, National Academy of Sciences, Pediatric Committee on

Children with Disabilities, American Academy of Neurology, and the Child Neurology Society

(Coonrod & Stone, 2005). These organizations developed autism screening guidelines.

Screening instruments can be classified by their intended populations. Level 1 instruments are
intended for pediatric settings and are implemented as part of well-child visits. Level 1

screening involves routine evaluation for developmental problems from infancy through the start
of school. Although a few level 1 screeners exist specific to autism, the majority assess a

broader range of developmental concerns. The guidelines recommend further evaluation in the
absence of specific developmental milestones such as pointing, waving, and multiple word
phrases and if there is regression in social or language skills. Further evaluation would involve
the use of level 2 screeners to help differentiate children with autism from those with general
developmental concerns. As such, these instruments require greater expertise and are typically
administered in clinical settings as part of early intervention and identification services (Coonrod

& Stone, 2005). Although several level 1 and level 2 screening measures exist, only measures that are commonly used and/or specific to the present study will be discussed.

#### **Checklist for Autism in Toddlers**

One of the first early autism screeners was the Checklist for Autism in Toddlers (CHAT; Baron-Cohen, Allen, & Gillberg, 1992; Baron-Cohen et al., 1996). The CHAT is a level one screener developed in the United Kingdom where home healthcare visits are common.

Consistent with this method of service delivery, the CHAT consists of two parts: the first to be completed through parent interview and the second by the home healthcare nurse based upon observations of the child. The instrument was designed to detect autism in children as young as 18 months and incorporates items measuring prodeclarative pointing, make believe play, and eye gaze monitoring (Baird et al., 2000; Coonrod & Stone, 2005). In the initial study, the CHAT was first administered to fifty 18-month-old children and the items that the majority of them passed were retained. These items were then administered to 41 children at risk for autism (siblings with the disorder), and based upon a threshold of two or more items failed, all four children eventually diagnosed with the disorder were identified, without false positives.

Baron-Cohen (1996) attempted to replicate the findings of the 1992 study to evaluate the effectiveness of the CHAT in differentiating autism from general developmental delays. The CHAT was administered to 1,600 eighteen-month-old children who, based upon items failed, were placed into one of three groups: autism risk, developmental delay, and typical development. Children from all three groups were then invited to receive a comprehensive diagnostic evaluation and were given actual diagnoses of autism, developmental delay, and no diagnosis. Of the 12 children with positive CHAT screens indicating autism, 10 received actual diagnoses

(positive predictive value of 83.3%). Of the 22 children with CHAT scores indicating developmental delay, 15 (68.2%) received corresponding diagnoses.

In a follow-up study, Baird et al., (2000) screened 16,000 children on two occasions, at 18 and 19 months of age. Based upon the number of key items endorsed, a level of autism risk (from high risk to no risk) was assigned. Actual diagnoses of autism at seven years of age were confirmed through several methods including assessment by the research team, referral to a clinical center, and review of records. Screening at 18 months of age resulted in a sensitivity (% with the disorder who screened positive) of 38% and a specificity (% without the disorder who screened negative) of 98% in identifying children with autism with the positive predictive value (% with a positive screen who were correctly identified) increased by using the high risk autism threshold. Additional screening at 19 months resulted in a sensitivity of 20%, specificity of 99.9%, and a positive predictive value of 75%.

#### **Modified Checklist for Autism in Toddlers**

The Modified Checklist for Autism in Toddlers (M-CHAT; Robins, Fein, Barton, & Green, 2001) is a modification and extension of the CHAT that was initially developed as a level 1 screening instrument to be self administered by parents during visits to their child's pediatrician's office. The M-CHAT was developed for several reasons. For one, the observational component of the CHAT was removed because the M-CHAT was intended to be used in the United States where there is no equivalent of the home healthcare nurse. The target population was raised from 18 months to 24 months of age in order to include children who exhibit autistic regression prior to their second birthday. Furthermore, the number of parent report items was increased and the content broadened to capture repetitive behaviors and some of the behaviors previously assessed by the home health nurse. Efforts were made to increase the

specificity of the M-CHAT in comparison to the CHAT by decreasing the cutoff score for a positive screen while incorporating a follow-up phone call after a positive screen to double check the accuracy of items indicative of autism (Coonrod & Stone, 2005; Robins et al., 2001).

Robins et al. (2001) describe the development and validation of the M-CHAT. The sample consisted of 1,293 children of which 87% were recruited as part of routine pediatrician visits (low risk), while the remaining 13% were recruited through referrals from early intervention programs (high risk). The latter group consisted of toddlers with atypical development without current Axis I diagnoses but who were receiving minimal intervention services such as occupational therapy. Initially the M-CHAT consisted of 30 items; however 8 were removed due to poor interpretability and poor differentiation between groups. A scoring algorithm was created based upon two cutoff scores: two or more critical items derived from a discriminate function analysis, or three or more total items. Of the 1,293 participants, 149 initially failed the screener and received follow-up phone calls to determine the accuracy of failed items. Failed screeners were confirmed for 58 children who were then referred for thorough diagnostic evaluations, of which 39 received ASD diagnoses. Because the study was published prior to all participants receiving follow-up evaluations, the authors conceded that true diagnostic accuracy could not yet be determined. Results provided were an estimate, which varied according to the whether the total score or critical item cutoff score was used. Values ranged from .95-.99 for specificity, .95-.97 for sensitivity, .36-.79 for positive predictive power, and .99 for negative predictive value. The internal consistency was good with a Cronbach's alpha of .85 for the total scale, and .83 for the critical items (Coonrod & Stone, 2005; Robins et al., 2001).

Other studies have examined the utility and psychometric properties of the M-CHAT. For instance, Eaves, Wingert, and Ho (2006) administered the M-CHAT to 84 children between the ages 2 and 4. Two-thirds met criteria for ASD and the other third had at least one other diagnosis (i.e., language disorder). Sensitivities for the critical item and total score cutoffs were fair to excellent with Cronbach alphas of 77% and 92%, respectively. However, specificities were poor with Cronbach alphas of 43% and 27%, respectively. Poor specificity may have been the result of the control group consisting of children with severe psychopathology, and because follow-up phone calls were not conducted to verify the accuracy of failed items. Ventola (2007) compared the characteristics of children who failed the M-CHAT and were later diagnosed with an ASD, Developmental Delay, or Developmental Language Disorder. In general, children with ASD differed in regards to joint attention, social skills, communication, sensory, and play skills. When language skill level was controlled for, joint attention behaviors (requesting by pointing, pointing to express interest, following pointing, and responding to name) were the most discriminative.

### **CHAT and M-CHAT Variations**

The M-Chat has been translated into 28 languages with more translations in development (Robins, 2011). Seif Eldin et al. (2008) validated the M-CHAT for use in Arabic Countries. Two hundred-twenty children between the ages of 18 and 124 months took part in the study. The accuracy of the measure in identifying groups was similar to Robins et al. (2001) with a specificity of .80, sensitivity of .86, and positive predictive power of .88. Wong et al. (2004) described the development of the CHAT-23 which is a composite of the M-CHAT and CHAT intended to detect autism in Chinese children between the ages of 18 and 24 months. The CHAT-23 consists of the 23 questions from the M-CHAT in addition to the 5 observational

items from the CHAT. Similar to the Robins et al. (2001), two criteria were used to indicate failure on the questionnaire portion. The first was by failure of 6 of the total 23 items, and the other was by failure of 2 of 7 critical items that were derived through discriminate function analysis. Critical items from the CHAT-23 were similar to the discriminating items from the CHAT and M-CHAT. Items were related to imitation, social interest, joint attention, pretend play, prodeclarative pointing, and social referencing. The authors proposed a multiple tier screening procedure consisting first of the questionnaire portion, followed by observations from a trained healthcare worker, and then referral for a more thorough diagnostic evaluation.

Depending on the criteria used to identify failure, the procedure resulted in a sensitivity of .74-.93, specificity of .77-.91, and positive predictive value of .74-.85.

In another iteration of the CHAT, Allison et al. (2008) developed the Quantitative CHecklist for Autism in Toddlers (Q-CHAT). The Q-CHAT was designed to improve upon the accuracy of the M-CHAT and CHAT, while providing a measure that could be used in both clinical and research settings. The authors stated that, while the M-CHAT has good sensitivity and specificity, a large portion of the original sample consisted of children referred for early intervention services and it is unclear how the measure would fare in a general population of toddlers. Likewise, the CHAT's sensitivity was limited by the wording of items, which contained absolute words such as "ever" (i.e., "Does your child ever use his/her index finger to point?"). The Q-CHAT was instead designed to be more flexible. Unlike the CHAT which consisted of a binary, yes/no rating system, the Q-CHAT allowed more response options in the form of a likert scale ranging from 0 to 5. Another modification of the Q-CHAT was the addition of items to assess stereotypical behavior and sensory problems not included in the M-CHAT.

Although the Q-CHAT appears to be a promising screener, further research is necessary to determine its clinical utility. Limited information was obtained from the initial study (Allison et al., 2008) that included 799 typically developing toddlers and 160 toddlers with diagnoses on the autism spectrum. Results of the study showed that the Q-CHAT had good test-retest reliability with an intraclass correlation of .82., and that there were significant group differences between the typical toddlers and those with ASD.

# Pervasive Developmental Disorders Screening Test, Second Edition

Another level one screener is Stage 1 of the Pervasive Developmental Disorders

Screening Test, Second Edition (PDDST-II; Siegel, 2004). In all, the PDDST-II contains 3 stages
for application in different settings. Stage 1 is intended for primary care environments for
children ages 12 to 48 months. Stage 2, is intended for clinics where children are assessed for
potential developmental disabilities. Stage 3 is designated for use in clinics as part of
comprehensive autism diagnostic batteries. The purposes of the PDDST-II are to screen for ASD
during the first 48 months of life while helping differentiate ASD from other developmental
disorders.

Stage 1 consists of 23 items with 3 answer options indicating the extent to which specific behaviors occur in relation to typically developing children. Scores of five or greater indicate the need for a more thorough evaluation. According to the authors, the PDDST-II yielded a sensitivity of .92 and specificity of .91 when tested on a sample of 1,000 children considered either at risk for an ASD or who had mild/moderate developmental disabilities (Siegel, 2004; Matheiw, 2005).

Stage 2 of the PDDST-II (Siegel, 2004) is a level 2 screener that was designed to be used in early childhood environments as a means to help differentiate children with ASD from those

with other developmental disabilities. An analysis of 260 children with Autistic Disorder/PDD-NOS and 120 children with other developmental disabilities was used to develop cutoff scores, resulting in a sensitivity of .69-.88 and a specificity of .25-.63 (Coonrod & Stone, 2005). Despite promising initial data, more information regarding the development of the PDDST-II and standardization procedures was recommended to determine its clinical utility (Coonrod & Stone, 2005).

### Baby and Infant Screen for Children with aUtIsm Traits

The Baby and Infant Screen for Children with aUtIsm Traits (BISCUIT; Matson, Boisjoli, & Wilkins, 2007) is an informant based assessment instrument designed to measure symptoms of autism and associated problems in young children (17-37 months) who present with developmental concerns. It is comprised of three parts. Part 1 is intended to help identify children with PDD-NOS and Autistic Disorder from those who present with general developmental problems. Part 2 is used to measure symptoms potentially indicative of comorbid psychopathology consistent with tic disorders, ADHD, Obsessive Compulsive Disorder (OCD), and Specific Phobia. Part 3 measures problem behaviors associated with autism such as self-injury, aggression, disruption, and repetitive behaviors (Matson, Boisjoli, & Wilkins, 2007).

The initial BISCUIT study (Matson, Wilkins, Sevin et al., 2009) described the generation and selection of items based upon the administration of the measure to parents of approximately 300 children enrolled in an early intervention program in the state of Louisiana. BISCUIT-Part 1 began with 71 items that were generated through an extensive literature review as well as input from experts in the field. Several items were removed as the result of low endorsement rates and inter-item and inter-scale correlations. In a follow-up study of 405 infants from the same early intervention program, factor analytic techniques were used to identify 3 primary factors for

BISCUIT-Part 1: Socialization/Nonverbal Communication, Communication, and Repetitive Behavior/Restricted Interests (Matson, Boisjoli, Hess, & Wilkins, 2010).

Matson, Wilkins, Sharp et al. (2009) developed cutoff scores for BISCUIT-Part 1 to help differentiate among children with Autistic Disorder, PDD-NOS, and atypical development. Diagnoses were made by a licensed psychologist with extensive expertise in the field of autism and developmental disabilities. Diagnoses were based upon M-CHAT scores, Battelle Developmental Inventory-Second Edition (BDI-2; Newborg, 2005) scores, and DSM-IV-TR criteria. Participants consisted of 1007 children ages 17 to 37 months enrolled in an early intervention program as described above. Cutoff scores were calculated using a standard deviation approach (Jacobson & Truax, 1991) and profile analyses to determine scores that would result in the largest spread between groups. This was followed by logistical regression procedures and receiver operating characteristic (ROC) analysis to calculate and optimize sensitivity and specificity. A cutoff score of 17 was selected to differentiate atypical development from PDD-NOS resulting in a sensitivity of 84.7 and a specificity of 86.4. A cutoff score of 39 was selected to differentiate PDD-NOS from Autistic Disorder resulting in a sensitivity of 84.4 and a specificity of 83.3.

As a precursor to the current investigation, Matson, Fodstad, and Dempsey (2009) identified items from the BISCUIT-Part 1 that best classified autism and PDD-NOS in toddlers with developmental delays or who are at risk for developmental delays. Two studies were conducted. In the first, logistic regression analyses were conducted to identify a subset of items that best distinguished Autistic Disorder/PDD-NOS from atypical developmental. Using thirteen items as predictors, 92% of children were correctly classified as having ASD and 98% of children were correctly classified as having atypical development (no ASD). Similar statistical

analyses were used to discriminate Autistic Disorder from PDD-NOS. Using 11 predictor items, 88.9% of children with Autistic Disorder were correctly classified as were 88.2% of children with PDD-NOS. Interestingly, in both studies the reduced models derived from the logistic regressions included items representing all three core areas of impairment (social, communication, and stereotypical behaviors). Unfortunately, the predictive weights of items were not listed in the study; therefore, the relative importance of particular items in classifying group membership was unclear.

The current investigation, as described below, looked to build upon the findings of Matson, Fodstad, and Dempsey (2009) by developing an abbreviated scoring algorithm for the BISCUIT-Part 1. While Matson, Fodstad, and Dempsey (2009) demonstrated that a collection of BISCUIT-Part 1 items could be used to correctly classify diagnoses, more comprehensive analyses were performed to identify and evaluate items to be included in the scoring algorithm and to develop cutoff scores that would optimize sensitivity and specificity.

# **Purpose**

Autism Spectrum Disorders are characterized by pervasive impairments in socialization, communication, and stereotypical behavior; and are associated with a myriad of other debilitating features including medical conditions (APA; 2000), intellectual disability (La Malfa et al., 2007; Matson & Shoemaker, 2009), other forms of psychopathology (Baron-Cohen et al., 1999; Bellini, 2004; Ghaziuddin et al., 2002; Schreck et al., 2004), and severe problem behavior (Matson et al., 2009).

It is not surprising then that a diagnosis of ASD is associated with poor social, occupational, and educational outcomes (Eaves & Ho, 2008; C. Gillberg & Steffenburg, 1987). Although evidence suggests that IQ is predictive of outcome, higher functioning individuals also tend to have compromised futures (Engstrom et al., 2003; Howlin et al., 2004). Compounding outcomes is data suggesting that the prevalence of the disorder has drastically increased over the years, with a recent prevalence rate of 6.7 per 1,000 (Rice, 2009). Although it has been proposed that systemic factors such as diagnostic substitution and broadened diagnostic criteria may account for a large portion of the increase (Coo et al., 2008; Croen et al., 2002; Shattuck, 2006), others argue that these explanations have been overstated (Hertz-Picciotto & Delwiche, 2009).

Fortunately, effective treatments exist that, when applied in an intensive manner, result in substantial gains in such areas as language, social skills, IQ, and school placement (Cohen et al., 2006; Eikeseth et al., 2007; Smith et al., 2000). These gains can be capitalized on by early intervention with evidence that children who receive intensive services at younger ages have better outcomes (Fenske et al., 1985; Harris & Handleman, 2000), In addition, early intervention is supported by research in the neurosciences (Coonrod & Stone, 2005; Squire, 2008), and may

be a more cost effective method of service delivery to individuals with ASD over their lifetimes (Jacobson et al., 1998).

However, providing early intervention to children with autism is dependent on the timely identification of the disorder. While several psychometrically sound diagnostic instruments exist, what are also needed are screening instruments that can be easily administered to a large number of children at an early age to identify those at risk for autism who would benefit from a more thorough diagnostic evaluation (Coonrod & Stone, 2005). Of the autism screeners that are available some are intended to be administered for children in the general population (level 1) while others are intended for children already at risk for a developmental disability (level 2; Coonrod & Stone, 2005). A recently developed level two measure is the BISCUIT, which is currently being used in tandem with the M-CHAT in the state of Louisiana as part of their state run early intervention program. A unique feature of the BISCUIT is that, in addition to measuring risk for autism (BISCUIT-Part 1), it includes a scale to identify symptoms potentially consistent with comorbid psychopathology (BISCUIT-Part 2), and a scale that measures problem behaviors that frequently co-occur with autism (BISCUIT-Part 3; Matson et al., 2007). Based upon the comprehensive nature of the BISCUIT, further refinement of the battery seemed prudent.

The focus of the current study was on the refinement of the diagnostic portion of the BISCUIT (Part 1). Research suggests that social abnormalities are central to ASD and that deficiencies in specific social behaviors (imitation, social interest, joint attention, prodeclarative pointing, and social referencing) are characteristic of autism in young children (Eaves et al., 2006; Ozonoff et al., 2005; Robins et al., 2001; Wimpory et al., 2000; Wong et al., 2004). The purpose of the present study was to create an abbreviated scoring algorithm for BISCUIT-Part 1

by identifying critical items that correctly classify autism and by calculating a cutoff score to optimize sensitivity and specificity.

A scoring algorithm for the BISCUIT Part-1 is important for several reasons. For one, scoring algorithms have been developed for other autism screening measures (Robins, Fein, Barton, & Green, 2001; Wong et al., 2004) and provide an alternative means to detect autism based upon a set of critical items that represent key behaviors predictive of the disorder in young children. Considering that autism screeners should be developed to maximize true positives, even at the expense of false positives, a two-pronged scoring procedure may prove beneficial. Furthermore, a scoring algorithm could be used independently to minimize assessment time, especially for children with more clear-cut symptomotology. Finally, the identification of critical items for the BISCUIT-Part 1 would add to the increasing body of knowledge regarding the early symptoms of autism and help professionals better understand the nature of autism in young children.

### Method

# **Participants**

The sample initially consisted of 2,253 participants; however, 85 were removed during data cleaning process (see results section). The final sample consisted of 2,168 children ages 17 to 37 months (see Table 1) enrolled in Louisiana's EarlyStep's early intervention program, which provides services to families with children ages birth to 36 months who either have a developmental delay or who have a physical condition likely to result in a developmental delay.

Table 1 Demographic Characteristics per Diagnostic Group

	Diagnostic  classifications		
Demographic characteristics	Atypical Development (n = 1526)	PDD-NOS (n = 287)	Autistic Disorder (n = 355)
Age (in months), mean (SD)	25.74 (4.89)	26 (4.73)	27 (4.73)
Gender			
Male, no. (%)	1045 (68.5)	203 (70.7)	267 (75.2)
Female, no. (%)	475 (31.1)	82 (28.6)	87 (24.5)
Unspecified, no. (%)	6 (0.39)	2 (0.70)	1 (0.28)
Race/ethnicity, no. (%)			
Caucasian	741 (48.6)	136 (47.4)	173 (48.7)
African-American	595 (39.0)	118 (41.1)	139 (39.2)
Hispanic	45 (2.9)	4 (1.4)	6 (1.7)
Other	68 (4.5)	15 (5.2)	18 (5.1)

Diagnostic classifications for participants, including atypical development (n=1526) and Autistic Disorder/PDD-NOS (n=642), were assigned by a licensed doctoral level psychologist with over thirty years in the felid of autism and developmental disabilities. Diagnostic classifications were

based upon DSM-IV-TR criteria, M-CHAT scores, and Battelle Developmental Inventory-2nd Edition (BDI-2; Newborg, 2005) scores. Similar diagnostic methodology has been described in prior research where ASD diagnoses were established (Fombonne et al., 2004). Diagnostic classifications included Autistic Disorder, PDD-NOS, and atypical development. The study involved the review of participant records and was approved by the institutional review boards of Louisiana State University and Louisiana's Office for Citizens with Developmental Disabilities.

### **Test Administration**

The BISCUIT was administered to parents by professionals with licensure or certification in disciplines (e.g., occupational therapy, speech-language pathology, psychology, physical therapy, social work, and education) that permitted them to provide services through EarlySteps, Louisiana's Early Intervention program for infants and toddlers with disabilities. Test administrators were required to attend an all day workshop that included information on ASD, scale development, and BISCUIT test procedures. Administration of the BISCUIT took place in the child's home or daycare setting, where the tester interviewed the child's primary caregiver (Matson, Wilkins, Sevin et al., 2009).

## **Measures**

The BISCUIT is a comprehensive assessment instrument used to identify symptoms of ASD, comorbid psychopathology, and problem behavior in infants. It is comprised of three components. The component under analysis for the present study is BISCUIT-Part 1, which was designed to assist in the identification of children at risk for Autistic Disorder and PDD-NOS. Part 1 has an internal reliability of .97 (Matson, Wilkins, Sevin et al., 2009), and contains 62 items that are rated along a 3-point scale indicating the extent to which the child under investigation compares to a typically developing child of the same age. Items are scored as 0 (not

different; no impairment), 1 (somewhat different; mild impairment), or 2 (very different; severe impairment).

Initial cutoff scores for BISCUIT-Part 1 were based upon the administration of the BISCUIT to caregivers of 1007 children between the ages of 17-37 months enrolled in an early intervention program in the State of Louisiana. Diagnostic classifications were based upon DSM-IV-TR criteria, scores on the M-CHAT, and scores on the Battelle Developmental Inventory-Second Edition (BDI-2; Newborg, 2005); and were made by a licensed psychologist with extensive expertise in the field of autism and developmental disabilities. Cutoff scores designed to differentiate atypical development from PDD-NOS resulted in a sensitivity of 84.7 and a specificity of 86.4, while cutoff score designed to differentiate atypical development from PDD-NOS resulted in a sensitivity of 84.7 and a specificity of 86.4.

Biscuit-Part 2, has an internal reliability of .96 and was designed to assess symptoms potentially consistent with comorbid psychopathology including: ADHD, Tic Disorder, OCD, Conduct Disorder, Specific Phobia, and eating problems. BISCUIT-Part 3 has an internal reliability of .91 and consists of 17 items used to measure problem behaviors associated with ASD, self-injury, aggression, disruption, and stereotypical behaviors.

### **Results**

Prior to the discriminant function analysis (DFA), data were examined for outliers, data entry errors, missing values, and adherence to the assumptions of DFA. Of the original 2,253 participants, 49 (2.2%) were removed for incomplete packets, missing ages, or ages outside the target range of 17-37 months. Thirty-six participants (1.6%) were identified as univariate or multivariate outliers and were deleted (Tabachnick & Fidell, 2007). An evaluation of the assumptions of DFA, including multivariate normality, homogeneity of variance-covariance matrices, linearity, and absence of multicollinearity were either satisfactory or protected by the large sample size.

Results of the one-way analyses of variance (ANOVA) indicated significant differences (p<.001) between groups on each of the 62 items that comprise the BISCUIT-Part 1, providing support for subsequent use of DFA. The DFA included all 62 items of the BISCUIT-Part 1 as independent variables and group membership (Autistic Disorder/PDD-NOS vs. Atypical Development) as dependent variables. An *a priori* power analysis was calculated using GPOWER (Erdfelder, Faul, & Buchner, 1996) to determine the necessary sample size for a DFA with an effect size of .40, alpha of .05, power of .95, 2 groups, and 62 response variables. The required sample size was 164.

# **Discriminant Function Analysis**

The DFA resulted in a significant discriminant function, F(62) = .297, p < .01, with a canonical correlation of .84 indicating that the model accounted for 70.40% of the variation between groups. The relative importance of BISCUIT Part-1 items in indentifying group membership are depicted in Table 2 in descending order according to the weights of the standardized canononical discriminant function coefficients. Items with the highest partial

Table 2 Standardized Canononical Discriminant Function Coefficients of BISCUIT-Part 1

		Items Failed				Items Failed	
Item	Discriminant Coefficients	% of Atypical Development	% of Autism Spectrum	Item	Discriminant Coefficients	% of Atypical Development	% of Autism Spectrum
59	.384	4.5	71.0	49	.054	10.4	40.3
53	.352	16.1	65.1	11	.053	9.2	36.9
4	.291	8.8	53.1	31	.050	10.2	63.9
17	.265	1.6	52.2	8	.050	9.3	54.1
19	.175	1.9	50.0	52	.048	10.9	72.7
3	.153	22.5	70.7	45	.048	7.0	55.1
27	.135	2.0	42.2	18	048	5.4	57.6
58	134	2.8	28.7	12	046	5.0	48.3
13	126	1.1	12.8	1	044	82.7	96.4
10	.115	15.5	75.8	28	.038	7.0	54.4
34	.108	3.7	40.2	20	.030	7.6	53.0
62	106	2.1	48.5	29	.029	5.5	44.5
50	.099	81.6	98.1	37	029	1.8	9.7
40	.094	7.2	25.5	54	.028	18.9	44.9
55	085	1.5	40.5	30	025	8.6	32.7
32	082	1.8	32.2	56	025	3.3	37.6
24	.074	56.8	87.5	41	023	2.2	30.5
21	073	5.2	44.4	43	023	1.1	22.9
46	.068	3.3	39.7	9	018	74.0	96.0
42	.064	5.1	36.5	14	.018	10.0	70.8
61	.064	5.8	48.9	51	.016	6.2	49.5
38	.063	7.2	40.2	2	.015	23.7	67.8
48	.062	16.0	49.8	39	.009	2.0	33.4
35	.061	10.9	72.9	33	.009	2.6	27.6
36	060	4.3	40.8	25	.008	2.7	23.7
16	.060	65.2	90.7	6	007	15.0	41.8
57	060	2.7	30.9	5	.006	82.3	96.3
47	058	2.7	37.0	23	005	3.2	28.2
60	.056	6.0	34.9	44	.004	3.5	17.8
15	.056	4.5	20.4	7	004	6.6	49.8
26	055	2.3	30.3	22	.003	3.0	37.5

contributions in descending order were 59, 53, 4, 17, 19, 3, 27, 58, 13, and 10. These items were selected as candidates for inclusion in the abbreviated scoring algorithm and subject to further analyses.

# **Receiver Operating Characteristic**

A receiver operating characteristic (ROC) analysis was then conducted to determine the number of items from the DFA that would be required to comprise an abbreviated scoring algorithm with adequate sensitivity and 1-specificity. Selection of the number of items to be included was based upon an inspection of the Area Under the Curve (AUC; see Table 3) values,

Table 3. AUC Values for Classifiers Based Upon the Number of DFA Items Included.

Items Included in Algorithm	Area	Asymptotic Sig.
59	.840	.000
59, 53	.909	.000
59, 53, 4	.942	.000
59, 53, 4, 17	.968	.000
59, 53, 4, 17, 19*	.980	.000
59, 53, 4, 17, 19, 3	.975	.000
59, 53, 4, 17, 19, 3, 27	.979	.000
59, 53, 4, 17, 19, 3, 27, 58	.981	.000
59, 53, 4, 17, 19, 3, 27, 58, 13	.982	.000
59, 53, 4, 17, 19, 3, 27, 58, 13, 10	.983	.000

<sup>\*</sup>Items selected for the scoring algorithm.

which give an overall indication of a classifier's predictive performance (Fawcett, 2006) and an inspection of sensitivity and 1-specificty payoffs for scoring thresholds within each classifier. The 5 item solution (59, 53, 4, 17, 19) with an AUC value of .980 was selected as the scoring algorithm; containing 3 items from the social domain, 1 from communication, and 1 from restricted and stereotyped behaviors. Item descriptions were as follows: (59) Development of

social relationships; (53) Use of nonverbal communication; (4) Engages in repetitive motor movements for no reason; (17) Shares enjoyment, interests, or achievement with others; and (19) Interest in participating in social games, sports, and activities. The ROC curve was then used to determine an optimal cutoff threshold based upon a total score derived from the 5 items selected. Sensitivities and specificities for specific cutoff values are listed in Table 4.

Table 4. Sensitivities and Specificities for Potential Cutoff Scores using the 5 Item Algorithm

Cutoff Score	Sensitivity	Specificity
1	.994	.688
2*	.941	.947
3	.723	.999
4	.542	1.000
5	.403	1.000
6	.270	1.000
7	.194	1.000
8	.111	1.000
9	.061	1.000
10	.031	1.000

<sup>\*</sup>Selected cutoff score.

A cutoff score of 2 was chosen as it corresponded to the optimal tradeoff between sensitivity and specificity. Figure 1 below, displays the ROC curve for the 5 item algorithm and the location of the selected cutoff score. Scores closest to the upper left corner of the graph represent an optimal balance between sensitivity and 1-specificity (Wong, 2004). The AUC statistic is defined as the area between the diagonal reference line and the curved line depicted by the algorithm. Overall, the 5 item solution with a cutoff score of 2 yielded a sensitivity of .941, specificity of .947, and positive predictive power of .883. This was similar to the accuracy of the

62 item scoring procedure with a cutoff score of 17, which yielded a sensitivity of .928, specificity of .865, and positive predictive power of .744.

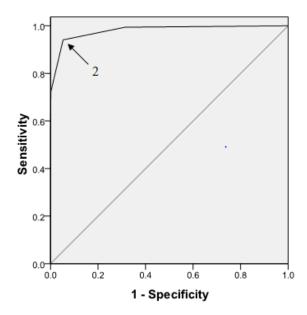


Figure 1. ROC curve representing the tradeoffs between sensitivity (true-positive rate) and 1-specificity (false-positive rate) for the 5 item scoring algorithm. The arrow identifies the location of the 2 point cutoff score.

### **Discussion**

The present study described the initial development of an abbreviated scoring algorithm for the BISCUIT-Part 1, using procedures similar to those employed in the development of scoring algorithms for other autism screening measures (Wong et al. 2004; Robins et al., 2001). Specifically, a discriminant function analysis was conducted to identify critical items that could effectively differentiate ASD from atypical development. The DFA generated a list of standardized canonical discriminant function coefficients representing the unique contribution of each item in discriminating groups. Sets of items with the highest weights were further evaluated based upon their overall discriminating performance using AUC values, and sensitivity and 1-specificity combinations for cutoff scores within each set. The ideal cutoff score was defined as one that would identify virtually all children with ASD with the fewest false positives (Robins et al., 2001). The selected 5 item algorithm with a cutoff score of 2 was as accurate as the 62 items in differentiating groups, confirming the hypothesis that the abbreviated scoring algorithm would produce a sensitivity and specificity within .10 of the existing scoring procedure.

These results are consistent with prior BISCUIT research and other autism screening measures that have developed abbreviated scoring algorithms. For instance, Robins et al. (2001) reported a sensitivity of .97, specificity of .95, and a positive predictive power of .36 using all items of the M-CHAT; and a sensitivity of .95, specificity of of.99, and a positive predictive value of .79 using 6 items derived through DFA. Likewise, Wong (2004) reported a sensitivity of .84, specificity of .85, and a positive predictive power of .79 for the entire CHAT-23; and a sensitivity of .93, specificity of of.77, and a positive predictive value of .74 for the 7-item score. Furthermore, Matson, Dempsey, and Fodstad (2009), used logistic regression procedures with an

earlier version of the Early Steps Database, and found a similar collection of items as those from the present study that best differentiated groups.

As unreasonable as it may seem for 5 items to accurately identify group membership, it is more compelling when you consider the effects of constructing total scores from items derived through DFA. When computing a total score based on the sum of all items from a scale, there is potential for a lot of variability across participants in the composition of items that make up the total score. For instance, it would be possible for a child without ASD to score high (1s and 2s) on items that are less predictive of an ASD, and derive a total score that exceeds the threshold for a positive test. However, using a scoring algorithm, the identification of group membership occurs through the prism of items with the most discriminating power.

Although the scoring algorithm has the potential to be an effective and efficient means of predicting risk for autism, more research is necessary. The study involved an a posteriori analysis of an existing database examining items that were administered as part of a 62 item scale. Cross validation on an additional sample of participants is recommended. Furthermore, the abbreviated scoring procedure should not be used as a replacement for the larger scale which provides a more comprehensive account of symptoms associated with ASD. Again, the algorithm is comprised of a collection of items that contribute the most in regards to unique variance in identifying group membership. Other items by themselves may be more "predictive"; however because they account for the same "slice of the pie" in regards to their predictive contribution, they were excluded from the algorithm. Although uniquely predictive, items from the algorithm are limited to the extent in which they capture all facets of the autism construct. While the BISCUIT-Part 1 is primarily a diagnostic tool, information obtained from the broad array of 62 items may prove useful for such things as progress monitoring, the creation

of treatment plans, and research related to the development of autistic symptomotology over time. Therefore it is suggested that the scoring algorithm be used in conjunction with the existing scoring procedure as a "safety net" in order to maximize sensitivity even at the expense of specificity.

As hypothesized, at least 3 of the 5 items with the greatest standardized canonical discriminant function coefficients represented the social abnormalities characteristics of autism. Items 59, 17, and 19 pertained to the development of social relationships, sharing interests and enjoyment with others, and participating in social activities, respectively. These finding are consistent with Robins et al. (2001), Wong et al. (2004), and other studies examining the predictive symptoms of autism in young children. For instance, Klin, Volkmar, and Sparrow (1992) found that compared to children with only developmental disabilities, children with autism were less likely to play simple interaction games, reach for familiar people, demonstrate a readiness to be picked up, and show an interest in other children. Likewise, Wimpory, Hobson, Williams, and Nash (2000) reported that children with autism had significantly impaired skills related to eye contact, raising their arms in anticipation of being lifted, turn taking during conversation, sharing objects with others, and using and responding to pointing.

There are several possibilities in regards to future directions. For instance, DFA and ROC analysis could be used to develop a cutoff score to differentiate PDD-NOS from Autistic Disorder, consistent with the original structure of the BISCUIT-Part 1. Conversely, scoring procedures could be adapted to meet the proposed diagnostic specifications of the DSM-V (APA, 2011). According to the APA development website, specific ASD will no longer be differentiated, and will be accounted for by a general diagnosis of Autism Spectrum Disorder with specifiers for level of severity. In a similar fashion, cutoff scores for the BISCUIT-Part 1

could be modified to reflect predicted severity. Consistent with the trend of earlier identification of autism, a next step may be to identify items that are predictive of even younger children who will go on to develop ASD. The BISCUIT-Part 1 is best suited to detect autism risk in children between the ages of 17-37 months with a mean age of 26 months; however, it is possible that specific items differ in their predictive abilities based upon ages within this range. While social abnormalities are the best indicators of autism in very young children, items representing other domains may become more important in older children.

The abbreviated scoring algorithm for the BISCUIT-Part 1 has the potential to be useful tool in the early identification of children with ASD. However, as recommended above, more research is necessary to determine if the 5 item solution holds up to cross validation using additional participants. Refinement of early autism screening measures such as the BISCUIT is critical considering the increased prevalence of autism, associated poor long-term outcomes, and evidence that early and intensive interventions may improve the trajectory of the disorder. While other scales focus solely on the identification of ASD, the BISCUIT incorporates the added benefits of measuring comorbid psychopathology and problem behavior associated with autism. Based upon the comprehensive nature of this instrument, continued development and analysis of its utility are recommended.

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## **Appendix**

## **Items of the BISCUIT-Part 1**

- 1. Communication skills.
- 2. Intellectual abilities (i.e., as smart as others his/her age).
- 3. Age appropriate self-help and adaptive skills (i.e., able to take care of self).
- 4. Engages in repetitive motor movements for no reason (e.g., hand waving, body rocking, head banging, hand flapping).
- 5. Verbal communication.
- 6. Prefers foods of a certain texture or smell.
- 7. Ability to recognize the emotions of others.
- 8. Maintains eye contact.
- 9. Use of language to communicate.
- 10. Social interactions with others his/her age.
- 11. Reactions to normal, everyday sounds (e.g., vacuum, coffee grinder).
- 12. Response to others' social cues.
- 13. Reaction to normal, everyday lights (e.g., streetlights, etc.).
- 14. Peer relationships.
- 15. Rhythm of speaking (e.g., sing-song; If nonverbal, rate "0").
- 16. Use of language in conversations with others.
- 17. Shares enjoyment, interests, or achievement with others (e.g., parents, friends, caregivers).
- 18. Ability to make and keep friends.
- 19. Interest in participating in social games, sports, and activities.

- 20. Interest in another person's side of the conversation (e.g., talks to people with intention of hearing what others have to say).
- 21. Able to understand the subtle cues or gestures of others (e.g., sarcasm, crossing arms to show anger).
- 22. Use of too few or too many social gestures.
- 23. Body posture and/or gestures.
- 24. Communicates effectively (e.g., using words, gestures or sign language).
- 25. Likes affection (e.g., praise, hugs).
- 26. Displays a range of socially appropriate facial expressions.
- 27. Restricted interests and activities.
- 28. Motivated to please others (e.g., peers, caregivers, parents).
- 29. Eye-to-eye gaze.
- 30. Reaction to normal, everyday sounds (e.g., vacuum, coffee grinder)
- 31. Awareness of the unwritten or unspoken rules of social play (e.g., turn taking, sharing).
- 32. Facial expression corresponds to environmental events.
- 33. Sticking to odd routines or rituals that don't have a purpose or make a difference.
- 34. Abnormal preoccupation with the parts of an object or objects.
- 35. Plays appropriately with others.
- 36. Reads nonverbal cues (body language) of other people. (If blind, rate "0")
- 37. Speaks in monotone (e.g., voice is flat, does not change in sound; If nonverbal, rate "0").
- 38. Expects others to know their thoughts, experiences, and opinions without communicating them (e.g., expects others to "read his/her mind").
- 39. Interest in a highly restricted set of activities.

- 40. Talking to others in a social context (If nonverbal, rate "0").
- 41. Use of facial expressions.
- 42. Abnormal fascination with the movement of spinning objects (e.g., closing doors, electric fan blades).
- 43. Curiosity with surroundings.
- 44. Saying words and phrases repetitively (If nonverbal, rate "0").
- 45. Make-believe or pretend play.
- 46. Understanding of age appropriate jokes, figures of speech, or sayings.
- 47. Gives subtle cues or gestures when communicating with others (e.g., hinting).
- 48. Becomes upset if there is a change in routine.
- 49. Needs reassurance, especially if events don't go as planned.
- 50. Language development.
- 51. Responds to others' distress.
- 52. Socializes with other children.
- 53. Use of nonverbal communication.
- 54. Clumsiness.
- 55. Limited number of interests.
- 56. Imitation of an adult or child model (e.g., caregiver waves "bye" then the child waves "bye").
- 57. Abnormal, repetitive hand or arm movements.
- 58. Abnormal, repetitive motor movements involving entire body.
- 59. Development of social relationships.
- 60. Respect for others' personal space (e.g., stands too close to others).

- 61. Isolates self (i.e., wants to be by him/her self).
- 62. Participation in games or other social activities.

## Vita

Santino LoVullo was born in Montebello, California, in 1975. He earned a Bachelor of Arts degree in psychology in 1997. In 1998, he began an eight year career as an elementary school teacher for children with ASD. To further his knowledge of ASD, he enrolled in a doctoral program in clinical psychology at Louisiana State University. Currently he is finishing his 5<sup>th</sup> year of studies and works as a predoctoral intern at the Kennedy Krieger Institute and Johns Hopkins School of Medicine. His research and clinical interests focus on the assessment and treatment of individuals with ASD and severe problem behaviors.