EARLY AUTISM SCREENING USING A GENERAL SOCIAL-EMOTIONAL MEASURE: PRELIMINARY STUDY OF THE AGES AND STAGES QUESTIONNAIRES: SOCIAL EMOTIONAL- 2ND EDITION

by

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DISSERTATION ABSTRACT

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Title: Early Autism Screening Using a General Social-Emotional Measure: Preliminary Study of the Ages and Stages Questionnaires- Social Emotional – 2nd Edition

The Ages and Stages Questionnaires (ASQ) (Squires & Bricker, 2009) is used by pediatricians, educators, and parents to screen for developmental delays and identify children at risk for delay. The purpose of the ASQ: Social Emotional- Second Edition (ASQ:SE-2) is to screen for a child's self-regulation, compliance, communication, adaptive behaviors, autonomy, affect, and interaction with people. A recent revision of the ASQ:SE-2 included items that were intended to identify children at risk for autism spectrum disorder (ASD). The present study provides an examination of the validity of the ASQ:SE-2 in identifying children at risk for ASD.

In the present study, total scores on a subset of items from the ASQ:SE-2 that specifically addressed ASD were compared with results from a team clinical diagnosis of ASD for 60 children, between 18-48 months of age. When the ASD-related items were considered alone, the scores of children with ASD and those without ASD were significantly different, and there were no differences in ASQ:SE-2 total scores for children with and without ASD. These findings indicate that parents of children with ASD reported significantly more concerns on ASD-related items on the ASQ:SE-2 than parents of children without ASD. Moreover, the results suggest that an increasing ASD

total item score on the broadband instrument resulted in an increased likelihood of a child receiving an ASD diagnosis.

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CHAPTER I

INTRODUCTION

Autism Spectrum Disorder (ASD) is a neurodevelopmental disability that affects social interaction, communication, and behavior. While ASD is a heterogeneous condition, effects are often serious and may impact the individual as well as the family and community. Prevalence rates have risen dramatically in recent years, and the most recent data from the Center for Disease Control (CDC) suggest that ASD occurs in one in 68 children (CDC, 2014). Rising rates may be due to increased public awareness of the condition, changes in diagnostic practices, and/or an actual rise in prevalence. The effect of each is an even greater public awareness of the condition. With increased public awareness comes a great need to offer appropriate and effective screening tools that can respond to parental concern. The push for appropriate ASD screening presumes that ASD can be diagnosed at a young age. Indeed, a team of professionals can reliably make a stable medical diagnosis of ASD by two years of age (Guthrie, Swineford, Nottke, & Wetherby, 2013); Lord et al., 2006). Team diagnoses typically occur at centers with long waitlists, and accurate ASD screening can facilitate appropriate referrals.

In 2007, the American Academy of Pediatrics recommended routine screening for autism risk at the 18- and 24-month well-child appointments (Johnson & Myers, 2007). The Interagency Autism Coordinating Committee (IACC), established with federal support by the Combating Autism Act of 2006, has made the early detection of ASD a priority (IACC, 2012) to encourage detection efforts; however, a recent United States Task Force report concluded that there was insufficient evidence to recommend universal screening for ASD in young children for whom there are no concerns (Siu, 2016).

Additionally, some pediatricians question the rationale for ASD screening when there is no prevention, no cure, and limited evidence of effective treatments to recommend (Al-Qabandi, Gorter, & Rosenbaum, 2011). Recent research, however, has found early intervention to be effective at improving developmental outcomes for children with ASD (Dawson et al., 2010; Odom, Boyd, Hall, & Hume, 2010, Reichow, 2012). Clearly, more research is needed to provide an empirical base for specific treatments and to determine appropriate screening practices.

Statement of Problem

The increasing public awareness of ASD and the growing body of research supporting early treatment highlight a need for accurate and efficient early identification. There are myriad general screening instruments available that can be completed by a variety of caregivers or professionals in several settings. Fewer tools specific for ASD-related screening are available, and these tools are generally reserved for use in a medical setting. The Ages and Stages Questionnaires (ASQ; Squires & Bricker, 2009) is a general developmental screening tool that is useful for assessing a child's overall development, and the ASQ: Social Emotional, Second Edition (ASQ:SE-2) is a companion measure that is useful for assessing a child's social-emotional development (Squires, Bricker, & Twombly, 2015). It is possible that information resulting from the use of a broadband social-emotional screening tool could inform decisions regarding a child's risk for ASD.

The American Academy of Pediatrics (AAP) currently recommends screening a child's social and emotional development with ASD-specific tools early in life to promote early diagnosis of ASD and access to services (Briggs et al., 2012; Weitzman &

Wegner, 2015). While the ASQ:SE-2 was not created to be used as a screening instrument specific to ASD, the behaviors associated with ASD are consistent with social-emotional differences and delays that are included on the ASQ:SE-2 (Volkmar, Chawarska, & Klin, 2005). During the revision of the ASQ:SE-2 (Squires, Bricker, & Twombly, 2015), an effort was made to include red flag items that may identify children at risk for ASD. These items were included in questionnaire intervals for children between 15 and 48 months, based on both research-supported early indicators of ASD (Wetherby et al., 2004, Zwaigenbaum, Bryson & Garon, 2013) and the age at which the targeted skills might be considered atypical or missing (Ozonoff et al., 2010). Little is known, however, about how well these items function for children who have ASD and additionally how well the ASQ:SE-2 identifies children at risk for ASD.

Hypotheses

The purpose of this study was to investigate whether the ASQ:SE-2 identifies children at risk for ASD. This screening tool is administered by parents and caregivers, and is known to identify children with social-emotional differences. Little is known, however, about its ability to specifically identify characteristics of ASD. The study had two primary questions:

1. *What are the psychometric properties of the ASQ:SE-2 related to identification of ASD in children ages 18-48 months?*
    a. What are the sensitivity and specificity of the ASQ:SE-2 related to team diagnosis of ASD?
        i. The study described the validity of the ASQ:SE-2, with team diagnostic results acting as the criterion. The hypothesis was that

the ASQ:SE-2 would have high sensitivity and low specificity for ASD.

2. *Do parents of children referred for autism evaluations report concerns on specific ASQ:SE-2 red-flag items intended to identify children at risk for ASD?*
    i. The study examined ASQ:SE-2 response patterns related to ASD red flag items. Several items were added to the ASQ:SE-2 revision that were intended to help improve the tool's ability to identify children at risk for autism. These items were used to calculate an ASD item total score. It was hypothesized that parents of children with autism would answer that their child displays these behaviors, helping to differentiate children with and without autism. The ASD item total score was entered into a binary logistic regression to attempt to predict ASD diagnosis.

Significance of Study

This study provides a preliminary analysis of the validity of the ASQ:SE-2 as an ASD screener for young children. Outcomes will be useful to the parents, professionals, and caregivers with interest in early identification of ASD. Additionally, results will support subsequent research regarding screening instruments that can help identify children who may have ASD. Early and accurate screening can lead to earlier referrals for diagnostic assessment, which in turn can result in earlier access to intervention, and ultimately improved developmental outcomes.

An efficient, easy-to-administer screening tool that yields accurate results in a minimum amount of time is the ideal tool for most providers in medical and early

childhood settings. The most frequently cited barriers to ASD specific screening by pediatricians include their lack of time, lack of familiarity with tools, and a preference to refer to specialists rather than complete screening themselves (which may relate to lack of tool familiarity and time) (Dosreis, Weiner, Johnson, & Newschaffer, 2006). The ASQ:SE-2 is a tool that many pediatricians use and understand, given the popularity and widespread use of the Ages and Stages Questionnaires- Third Edition (ASQ-3; Squires & Bricker, 2009; Radecki, Sand-Loud, O'Connor, Sharp, & Olson, 2011). Using the ASQ:SE-2, which is a low cost and parent-friendly instrument, could provide valuable information related to early identification of autism-specific behaviors in young children. This may help providers place referrals more appropriately thereby using specialty developmental clinics more effectively.

CHAPTER II

REVIEW OF THE LITERATURE

Autism Spectrum Disorders

The most recent version of the Diagnostic and Statistical Manual (DSM-5) provided an updated description and critical changes to the diagnostic criteria for ASD (American Psychiatric Association, 2013). In the previous fourth edition of the manual (American Psychiatric Association, 1994), autism spectrum was divided into four separate disorders: autistic disorder, Asperger's disorder, childhood disintegrative disorder, and pervasive developmental disorder, not otherwise specified. The DSM-5 criteria were created to alleviate problems caused by differing clinical applications of these diagnostic labels. The current diagnostic label of ASD represents *a disorder characterized by persistent deficits in social communication and social interactions as well as restricted and repetitive patterns of behavior* (American Psychiatric Association, 2014). In addition, the impairments must be functional and have been present since the early developmental period.

Prevalence

Current prevalence rates for ASD suggest that the disorder affects 1 in 68 children (1 in 42 boys and 1 in 189 girls) (CDC, 2014). Due to a variety of factors, including long waitlists at diagnostic centers, the average age at diagnosis for children with ASD is about four years (Mandell, Novak, & Zubritsky, 2005). The diagnosis of ASD, however, can reliably be applied as early as two years of age (Lord et al., 2006). There has been a recent push for public awareness to support early identification with efforts to educate parents, pediatricians, and childcare providers. In 2013, the Centers for Disease Control

launched a campaign of social marketing to increase awareness and identification of ASD and developmental delay (Daniel, Prue, Taylor, Thomas, & Scales, 2009). The campaign's goal was to expand the common understanding of developmental progression to include cognitive and social-emotional milestones. Further, a recent report in Pediatrics urges physicians to screen for behavioral and emotional problems, in addition to traditional developmental milestones (Weitzman & Wegner, 2015).

Difficulties in Early Identification

While ASD diagnosis is possible by 24 months of age, the average age of a child at time of diagnosis continues to be above four years (Wingate et al., 2014). A recent systematic review identified various factors associated with age at ASD diagnosis (Daniels & Mandell, 2013). The factors ranged across child, family, and community levels. Factors associated with earlier ASD diagnosis included greater symptom severity, high socioeconomic status, and greater parental concern about initial symptoms.

The act of proving intervention effectiveness is increasingly difficult among a group of heterogeneous children with autism (Camarata, 2014). Consequently, even among pediatricians there are mixed feelings about the rationale for early screening when current interventions have limited empirical basis (Al-Qabandi, Gorter, & Rosenbaum, 2011). Webb, Jones, Kelly, and Dawson (2014) highlighted the need for continued research on treatment efficacy and to support specific recommendations on how and when to intervene. These arguments over early treatments in ASD affect the issue of early identification; however, even without robust empirical support, there is a general consensus that child outcomes improve with earlier access to intense interventions addressing the core components of ASD (Reichow, 2012).

Screening Practices

Current recommendations. At present, the American Academy of Pediatrics (AAP) recommends that pediatricians provide developmental surveillance at all well child visits and administer assessments at the 18-and 24-month check-ups to screen for ASD specifically (Johnson & Myers, 2007). The purpose of the screening is not to diagnose ASD in a child but simply to identify children who may be at risk for ASD and may need further evaluation. The AAP and Pinto-Martin and colleagues (2008) have recommended the use of autism-specific screening tools to better identify children at risk for ASD. Additionally, a recent study examined the efficacy of surveillance versus standardized screening practices in pediatric offices and found that use of tools was necessary for accurate identification of social delay and to detect autism risk (Gabrielson et al., 2015).

This year, the AAP recommendations have been challenged, however, by a recent United States Preventative Task Force recommendation on the accuracy, benefits, and potential harms of brief, formal screening instruments administered during well child checks (Siu, 2016). The report concluded that while there is sufficient evidence that such screening tools can detect ASD between 18 and 30 months of age, there is insufficient evidence to formally assess the benefits and harms of early detection. This recommendation is uniquely related to ASD-specific screening instruments and to children whose doctors and parents have no specific concern for ASD or other developmental difference. This equivocation of ASD screening has prompted ASD researchers and practitioners to protest the recommendation, fearing that years of work to lower the age of ASD diagnosis will be reversed and that children will miss out on early

intervention (Coury, D, 2015; Messinger et al., 2016). Both groups support ongoing research into both screening efficacy and developmental interventions for ASD.

ASD screening tools. Most of the screening tools for ASD are parent-completed, with solid empirical support. Parents have been found to be accurate reporters of developmental concerns (Glascoe, 2000; Glascoe & Dworkin, 1995), and after about 12 months of age, parent concern is correlated with later diagnostic labeling (Ozonoff et al., 2009). The Modified Checklist for Autism in Toddlers, Revised with Follow-Up (M-CHAT-R/F; Robins, Fein, & Barton, 2009) is one parent-report tool designed to evaluate risk for ASD. It is intended for children 16-30 months of age. Parents complete 20 yes/no questions, and children are assigned a risk level (low, medium, high) for ASD. The checklist is intended to be administered by a primary health provider (e.g., pediatrician), although parents can complete independently. The authors report a relatively high false positive rate and have created a follow-up interview for pediatricians to determine if there is a need for a referral; hence the two-stage assessment.

Another tool that offers information on young children's social communication is the Communication and Symbolic Behavior Scales Developmental Profile: Infant-Toddler Checklist (Wetherby & Prizant, 2002). It has been validated as an ASD screener for use with 9-24 month old children (Wetherby, Brosnan-Maddox, Peace, & Newton, 2008). This checklist consists of 24 questions about social communication milestones and a question about parent concern, resulting in a social composite, symbolic composite, and speech composite. Following the validation study (Wetherby et al., 2008), it was highlighted that a positive screen on the Infant Toddler Checklist does not distinguish children with ASD from children with other communication delays. A closer look at

composite scores within the Infant Toddler Checklist may be beneficial (i.e., the social composite) when screening specifically for ASD (Wetherby et al., 2008).

ASQ and ASD. ASQ (Squires & Bricker, 2009) has long been used by pediatricians and parents to screen for developmental delays and identify children at risk for delay. In 2002, the ASQ team added a companion measure to screen children's social and emotional development: the ASQ:SE (Squires, Bricker, & Twombly, 2002). The tool was designed to identify children at risk for social and emotional differences or delays and is often used in conjunction with the ASQ-3 by parents, pediatricians and childcare providers. A recent revision of the ASQ:SE was published in 2015 as the ASQ:SE-2 (Squires, Twombly, and Bricker, 2015). During the revision and data collection process, an effort was made to include items that may identify children at risk for ASD. These items were included in the questionnaire intervals between 15 and 48 months, based on both research-supported early indicators of ASD (Wetherby et al., 2004; Zweigenbaum, Bryson, & Garon, 2013) and the age at which the targeted skills might be considered atypical or missing (Ozonoff et al., 2010; Zweigenbaum, Bryson, & Garon, 2013). The ASQ:SE-2 is easy to administer, parent friendly, and already used in a variety of early childhood settings (medical, educational, and home-based). If the ASQ:SE-2 can accurately identify children at risk for ASD, more children may be accurately referred for evaluations, improving early identification efforts. This could reduce the need for specific ASD screens in addition to AAP recommended broadband screenings.

A recent study explored the utility of using the ASQ-3, a broadband developmental screener, to detect ASD in a sample of 2848 children across 20 sites (Hardy, Haisley, Manning, & Fein, 2015). Eighteen to 30 month old children were given

the ASQ-3 and the M-CHAT-R. Twenty-one children in the group received an ASD diagnoses and the ASQ-3 identified 95% of cases (20/21), using a monitor and/or fail category on the ASQ-3. The communication domain of the ASQ-3 seemed particularly sensitive to ASD, identifying 20/21 cases independent of the other ASQ-3 domains. In this study 412 children fell in the monitor or fail range for the communication domain, while only 20 of those children ended up with ASD diagnoses. The authors could not calculate specificity in the sample because children who failed the ASQ-3 but not the M-CHAT-R were not further evaluated. The authors note, however, that by combining the "monitor" and "fail" categories, the measure's sensitivity to ASD increased while at the same time decreasing the presumed specificity.

Alkherainej and Squires (2015) compared the ASQ-3 and the ASQ-SE (Squires, Bricker, & Twombly, 2002) with the Social Communication Questionnaire (Rutter, Bailey, & Lord, 2003) in 208 preschool aged children. Agreement among the three questionnaires was moderate to strong, although the ASQ-3 had slightly higher sensitivity and specificity for ASD. This study used the first edition of the ASQ:SE and utilized parent-report of diagnosis or school-label as ASD criterion.

Diagnosis

After children are identified as at risk for ASD, they are generally referred for a diagnostic evaluation. A team of specially trained providers, usually including a pediatrician, a psychologist, a speech-language pathologist, and an occupational therapist, traditionally work together to consider and determine a diagnosis of ASD in young children. Critical components of the diagnostic process include a parental interview and administration of a gold-standard assessment for social interaction and communication.

The Autism Diagnostic Observation Schedule, Second Edition (ADOS-2; Lord et al., 2012) is a semi-structured standardized assessment for observing and recording behavioral characteristics associated with ASD and is widely used for ASD identification. The ADOS-2 uses developmentally appropriate toys and activities to elicit social behaviors, communication, and play during a 30 to 45-minute session. Five modules make up this assessment, which are dependent on language ability (Toddler Module and Modules 1 – 4). Domains include social interaction, communication, play, and repetitive, restrictive behaviors. Following administration, the examiner evaluates the child's behavior and participation using codes, which translate into scores on an algorithm. A child's score describes the extent to which a child's behaviors are consistent with autism (Toddler Module) or provide a cut-off score for autism and autism spectrum classifications (Modules 1 - 4).

In addition to ADOS-2 test results, a member of the diagnostic team, typically the developmental pediatrician or developmental psychologist, conducts a structured parent interview to determine if DSM-5 criteria are met for ASD. Children in ASD evaluation clinics typically also receive a battery of additional testing, including cognitive, speech-language, and occupational therapy. These tests are done to support a diagnosis or identify other conditions that may be responsible for some ASD-like symptomatology.

ASD Treatment

There are a variety of reasons for the paucity of solid evidence to support treatment efficacy for ASD as well as a good empirical base to support continued intervention research. ASD is extremely heterogeneous; mildly affected individuals may be successful in society and fully independent, while severely affected individuals may

be nonverbal, self-injurious, and dependent on others for activities of daily living. All children with autism will certainly not benefit from the same type of treatment, and the DSM-5 has only recently established a categorization system for ASD severity level (American Psychiatric Association, 2013). These levels do not yet correspond with particular treatment methods.

Additional issues affect scientific inquiry into autism intervention. A limiting factor for building the evidence base for autism interventions is the nature of the core deficits characteristic of ASD. Scientists have different operational definitions for impairment in social interaction, differences in communication, and restricted interests and repetitive behaviors. Varying operational definitions result in variation in how progress is measured. To prove that a treatment works, there is a need to show a desired outcome is achieved; yet in autism treatment, there is no agreement on what the desired outcome is, or how to measure it.

Currently, intervention efficacy for ASD is generally reliant upon intensity and age at which children begin treatment (Corsello, 2005). In a review of treatment programs, Dawson and Osterling (1997) report that successful programs offer 15-25 hours of intervention per week. Current recommendations suggest that families strive to obtain 25 hours per week of appropriate intervention (Odom, Hume, Hall & Hume, 2010). Further, the age at which intervention begins seems to be important in providing the best long-term outcomes. Studies find that children who begin intervention at younger ages make greater gains than children who enter programs at older ages (i.e., prior to age 4) (Harris & Handleman, 2000; Sheinkopf & Siegel, 1998).

Dawson and colleagues (2010) conducted the first randomized, controlled trial,

which demonstrated improvements in cognitive and adaptive behavior as well as a reduction in ASD severity in children. The toddlers in the study completed a course of comprehensive developmental, behavioral intervention (i.e., the Early Start Denver Model [ESDM]). This treatment combines behavioral (applied behavioral analysis, or ABA) methods with a developmental approach and was administered with high dosage, longitudinally over two years. The 48 children in the study received two-hour sessions with an interventionist, twice per day, five days per week for two years. Their skills and progress were measured prior to interventions, one year into intervention, and at the completion of the intervention, two years after the initial evaluation. The intervention group was compared to a group of young children with autism enrolled in community-based services (non-ESDM therapies). Outcome measures included subscale and composite test scores on the Mullen Scales of Early Learning (MSEL), composite scores on the Vineland Adaptive Behavior Scales (VABS), and autism diagnostic category from the university clinic.

Results of the Dawson et al. (2010) study supported ESDM's positive effect on cognition (i.e., intelligence quotient), adaptive skills, and autism diagnosis. These outcome measures are commonly considered at diagnostic clinics; however, none of the measures represent direct improvements in the core deficits of autism: social interaction, communication, and repetitive/restricted interests. The category of "autism diagnosis" may indirectly represent core deficits, and there was a significantly greater likelihood of improvement in diagnostic category (though no children moved out of an ASD diagnosis). There are additional concerns with using cognition as an outcome measure for autism treatments. First, the CDC's latest community report on autism found that a

62% majority of children with autism did not have intellectual disability (Baio, 2012). This key finding calls into question the practice of selecting cognitive improvements as desired developmental outcomes. Additionally, the MSEL has not been validated for use among children with autism spectrum disorders, though there is some recent preliminary data suggesting convergent validity (Bishop, Guthrie, Coffing, & Lord, 2011).

Kasari, Paparella, Freeman, and Jahromi (2008) developed an intervention designed to specifically target two areas of difficulty for children with autism: joint attention and symbolic play. The sample included 58 three- to four-year old boys who were participants in a 30-hour per week behavioral intervention treatment program. Participants were randomly assigned to a control group (the regular 30-hour per week intervention), a symbolic play intervention, or a joint attention intervention. Experimental interventions were behavioral-developmental in nature and led by interventionists for 30 minutes per week. (The participants continued to receive 29.5 hours each week of behavioral intervention.) Developmental outcomes included joint attention and play skills demonstrated with interventionist, joint attention skills demonstrated with mother, and language (receptive and expressive). Joint attention was defined as pointing, showing, giving, and coordinated joint looks, and play was coded as either functional or symbolic. MSEL language subtests and the Reynell Developmental Language Scales were used to assess language growth. Results showed that interventionists were able to teach joint attention and play skills. Children also generalized joint attention and play skills to interactions with their mothers. Both groups showed more growth in language than the control group, and the authors hypothesized that the symbolic play intervention (including an adult and a child playing with objects)

indirectly focused on and taught joint attention as well as play (i.e., treatment diffusion).

Another important finding was that individual skills prior to intervention mattered: children with the lowest developmental skills at pretest benefited most from the joint attention intervention. The study has limited external validity, however, due to the participants already engaging in 30 hours per week of intensive behavioral interventions. It is unclear whether interventions focused on joint attention or symbolic play would have similar effects in the absence of intensive behavioral interventions.

Yoder and Stone (2006) conducted an evaluation of two treatments for autism and focused on social interaction as a primary outcome measure. Participants' ages ranged from 18 to 60 months and children were all nonverbal at start of interventions. The 36 children with autism were randomly assigned to either a Responsive Prelinguistic Milieu Teaching (RPMT) intervention or a Picture Exchange Communication System (PECS) intervention. Clinicians administered the interventions for 20 minutes, three times per week, for six months. Social interaction outcomes included frequencies of turn taking, joint attention, requests, vocalization, and intentional communication.

As in the Kasari et al. (2008) study, the findings of Yoder and Stone (2006) indicated that child-specific variables mattered and that there was a differential response to treatment. Specifically, children with some joint attention at pretest benefited more from RPMT than from PECS (i.e., by increasing their frequency of joint attention initiations), and children with little joint attention initiation at pretest benefited more from PECS than from RPMT (i.e., by increasing their generalized requests, not in joint attention initiation). The natural conclusion could be that certain interventions may be better suited for certain subgroups of children with autism, even in a relatively

homogenous population, at least with respect to language development; however, the study has external validity threats that limit the application of its findings to a broader context. The sample size was small, and the parents were highly educated (with an average education level of 3-4 years of college). Participants in the study had additional community based treatments that could have accounted for some of the changes in outcome, and parents in the RPMT group attended significantly more sessions than did the PECS group parents.

Carter et al. (2011) conducted an efficacy trial of the Hanen Program: More Than Words parent-training program with 62 toddlers (mean age of 20 months) and their families. The families randomly assigned to the treatment group participated in 8 weekly group treatment sessions with a speech-language pathologist and three individual family sessions. This study included child social interaction outcomes (initiating joint attention, requesting, non-verbal communication) and also included a parent responsiveness outcome. Participants were video recorded three times: prior to randomization, 5 months post-enrollment, and 9-months post enrollment. Results did not show any treatment effects from the parent-training program on the group as a whole, but when individual subgroups were examined, certain groups responded differentially. Children with less functional play prior to the study benefitted most from this type of intervention.

Kasari, Gulsrud, Wong, Kwon, and Locke (2010) evaluated a parent-mediated joint attention intervention for directly teaching joint attention to toddlers with autism. Thirty-eight toddlers were randomly assigned to a waitlist control group or to an intervention group. Treatment group participants received interventionist-led sessions (three per week for eight weeks) and were then expected to carry on with treatment

activities throughout the week. Outcomes were measured pre- and post-interventions and also at one year following intervention (only for the intervention group, due to the waitlist control design). Outcomes included frequency measures of joint attention initiations, responses to joint attention, type of functional play act, type of symbolic play act, percentage of time spent in object-only play, and percentage of time spent in joint engagement with people. The study indicated that joint attention intervention resulted in less object-focused play, more joint engagement, and more responsiveness to joint attention. There was no increase in initiation of joint attention. The authors point out that with caregivers leading the intervention, it is difficult to assess fidelity of implementation, which affects intervention density. The small sample size also poses a threat to its population validity, which relates to its generalizability.

Landa, Holman, O'Neill, and Stuart (2011) also conducted a randomized controlled trial on a supplemental curriculum for young children with ASD that targeted socially synchronous engagement. Fifty toddlers with ASD were randomly assigned to two types of treatment programs. This study evaluated outcome differences between children who received interventions related to "interpersonal synchrony" (i.e., joint attention, imitation, affect sharing) and those who did not. In all other ways, the treatment programs for the two groups of children were identical. This study used social outcomes as primary measures, which is important since social deficits make up many of the core features of ASD. Results indicated that the group that received socially synchronous engagement training demonstrated significantly more socially engaged imitation. This finding was generalized across contexts and maintained at follow-up. There were no significant between group differences for joint attention or shared affect.

This study suggested that social engagement targets are important components in early treatment for ASD.

Another body of research that provides support for efficacy in ASD interventions comes from studies utilizing single-subject research design (SSRD). Because of the heterogeneity of ASD and current efforts to characterize various ASD phenotypes, projects using SSRD have become increasingly popular. These studies allow for specific interventions to be evaluated for specific individuals, and give the experimenter direct control over the intervention. SSRD supports the goal of evaluating which interventions work and for whom. Odom and colleagues (2003) described SSRD interventions specific to young children with ASD, and Horner and colleagues (2005) established the rigorous criteria to determine how SSRD supported interventions should be considered evidence-based. Criteria specifies that multiple SSRD studies support a practice before it is considered evidence-based. Horner et al. (2005) proposed a minimum of five, methodologically sound, published studies demonstrating desired effects of the intervention. In addition, these studies must be conducted by at least three different research groups, and they must include at least 20 participants. These criteria have allowed interventions supported by SSRD to become increasingly comparable to interventions with empirical support from RCT studies.

In summary, several rigorous, randomized-controlled trials of treatments have been conducted on interventions for young children with autism. Initial promising findings suggest that a combination of developmental and behavioral approaches have positive effects on developmental trajectories. Additionally, single subject designs show that children with ASD are responsive to treatments aimed at promoting interaction and

joint attention. Together, these findings provide support for early identification and diagnosis. Though more studies are needed, these studies demonstrate effectiveness of early intervention for ASD and encourage future research in this domain.

The present study seeks to provide a preliminary examination of the validity of a social-emotional developmental screening questionnaire, the ASQ:SE-2 in identifying children at risk for ASD. The purpose of the ASQ:SE-2 is to screen for a child's self-regulation, compliance, communication, adaptive behaviors, autonomy, affect, and interaction with people. Since children with ASD typically have difficulties in these areas, it is hypothesized that children with ASD will score above the cut-off on the ASQ:SE-2. Additionally, it is hypothesized that higher scores on items specifically related to ASD will predict ASD diagnosis.

CHAPTER III

METHODS

The study addressed two primary research questions using a combination of procedures and analyses. The following chapter describes the procedures and analyses; questions, participants, measures, and analyses are summarized in Table 1.

1. *What are the psychometric properties of the ASQ:SE-2 related to identification of ASD in children ages 18-48 months?*
    a. What are the sensitivity and specificity of the ASQ:SE-2 related to clinical team diagnosis of ASD?
    b. Do ASQ:SE-2 total scores differentiate children with and without ASD?
2. *Do parents of children referred for autism evaluations report concerns on specific ASQ:SE-2 red-flag items intended to identify children at risk for ASD?*
    a. Do ASQ:SE-2 autism item scores differentiate children with and without ASD?
    b. Do autism items on ASQ:SE-2 predict ASD diagnosis?

Design

The design was a non-experimental, case-control, measurement study. Psychometric properties of a screening instrument were investigated. Young children with suspected ASD were evaluated and their parents completed screening assessments to determine psychometric properties related to ASD.

Table 1. *Research Questions with Description of Participants, Measures, and Analyses*

Question	Participants	Measures	Analyses
1. What are the sensitivity and specificity of the ASQ:SE-2 related to team diagnosis of ASD?	Children with suspected autism, 18-48 months and their parents	• ASQ:SE-2 • Clinical Diagnoses	Contingency table displaying sensitivity and specificity of ASQ:SE-2 using diagnosis as criterion.
2. Do parents of children referred for autism evaluations report concerns on specific ASQ:SE-2 red-flag items intended to identify children at risk for ASD?	Children with suspected autism, 18-48 months and their parents	• ASD item total score on ASQ:SE-2 •	Independent Samples T-Tests Binary Logistic Regression

Subjects

Sixty children with suspected ASD between 18 and 48 months of age and their parents were invited to participate. All participants' families spoke English as a primary home language. These children had been referred for an ASD evaluation at a regional ASD diagnostic center, Oregon Health and Science University (OHSU). Children who met inclusion criteria but had previously been diagnosed with a medical condition that affected their development (e.g., cerebral palsy, vision loss, hearing loss, genetic syndromes) were excluded from participation.

Procedures

Families attending the ASD clinic were invited to participate. OHSU's electronic medical health record system schedules were prescreened for possible participants

(reviewing age and primary language). Children who participated were referred for an ASD evaluation due to concerns about their social, behavioral, and/or communicative development (i.e., someone had concerns about possible autism: a parent, educator, caregiver, pediatrician).

Children who met the inclusion criteria received a packet of information upon check-in to the autism clinic that included an introductory letter, the "Information Sheet", and an ASQ:SE-2. If parents agreed to complete the questionnaires, they turned them in to the front desk staff in a closed envelope. The child then participated in regular clinical activities related to the diagnostic process. In addition to language and social interaction testing, the children received evaluations from audiology, psychology, occupational therapy, and/or developmental pediatrics. Following the visit, results from the social interaction assessment (ADOS-2) as well as primary diagnoses, if any, were recorded.

Outcome Measures

Social-emotional screening. The ASQ:SE-2 was completed by participants' family members. The ASQ:SE-2 is a broadband social-emotional screening instrument for children between one month and 6 years of age. The first edition was published in 2002 (Squires, Bricker, & Twombly), and the revision was recently published in 2015 (Squires, Bricker, & Twombly, 2015). ASQ:SE-2 areas include self-regulation, compliance, social communication, adaptive functioning, autonomy, affect, and interaction with people. The standardization sample included over 14,000 diverse children.

Studies on the ASQ:SE-2 reflected robust psychometric properties, including an overall sensitivity of 81% across age intervals (with a range from 77% to 84%) and an

overall specificity of 84% across age intervals (with a range of 76% to 98%). The second edition includes new behavior and communication items to improve sensitivity to autism and early social communication differences (Squires, Bricker, & Twombly, 2015). Studies on the ASQ:SE-2 reflected high reliability (89% test-retest reliability) and high internal consistency (84%). Convergent validity was established for the ASQ:SE-2 using the Devereux Early Childhood Assessment for Infants and Toddlers (DECA-IT; Mackrain, LeBuffe, & Powell, 2007), the Infant Toddler Social Emotional Assessment (ITSEA; Carter & Briggs-Gowan, 2006), and the Child Behavior Checklist (CBCL; Achenbach & Rescorla, 2000). The Technical Report on the ASQ:SE-2 contains complete results (Squires, Bricker, & Twombly, 2015).

Sample items from the ASQ:SE-2 (18-month interval) include: *Does your child look at you when you talk to him?, When you point at something, does your child look in the direction you are pointing?,* and *Does your child make sounds, or use words or gestures, to let you know she wants something (for example, by reaching)?* (An 18-month ASQ:SE-2 questionnaire can be found in Appendix A.) Parents have the option to choose "often or always," "sometimes," or "rarely or never." They are also given a place to mark if they have a concern about each particular item. Items are scored according to parent responses and result in 0-15 points per item. Each interval (e.g., 6-month, 18-month, 48-month) has its own empirically derived cut-off score. High scores are indicative of difficulties and children are typically referred for further evaluation. There is also a newly defined monitoring zone in the second edition, which identifies children who are close to the cut-off and should be monitored and rescreened (Squires, Bricker, & Twombly, 2015). In this study, the "monitor" and "fail" categories on the ASQ:SE-2

were combined, as suggested by Hardy, Haisley, Manning, and Fein (2015) in a study related to ASQ:3.

ASD item total scores. Within each of the five ASQ:SE-2 intervals between 18 and 48 months, nine items were selected as potentially representative of ASD at the corresponding ages. The nine ASD items per interval are presented in Table 2; these items were confirmed by ASQ:SE-2 authors and by an ADOS-2 trained professional as having a likely relationship with ASD. Examples of ASD items included questions regarding joint attention, eye contact, peer interaction, and conversational abilities. ASD item total scores were calculated by summing the parent response (zero to 15) for these items, making the possible ASD item total score range between 0 and 135.

Parental concerns. As noted above, the ASQ:SE-2 allows for parents to select if a behavior is present (and to what degree, resulting in 0, 5, or 10 points for the item). Parents also have the opportunity to note if this particular developmental skill is a concern for them or not. If parents mark that an item is a concern for them, the item receives an additional 5 points. Because the "concern" item adds a level of subjectivity, data for this study were analyzed in two ways: 1) without concerns added (i.e., each item worth 0-10 points) and 2) with concerns included (i.e., each item worth 0-15 points).

Table 2. *Questions for ASD item total scores*

Item Content	Interval(s)
Does your child respond to her name when you call her?	18, 24
When you point at something, does your child look in the direction you are pointing?	18, 24, 30
Does your child try to show you things (with point and check-in at later intervals)	18, 24, 30, 36
Does your child play with objects by pretending (symbolic at later intervals)?	18, 24, 30, 36
Does your child look at you when you talk to him?	18, 24, 30, 36, 48
Does your child do things over and over and get upset when you try to stop her?	18, 24, 30, 36, 48
Does your child let you know how she is feeling with gestures or words?	18, 24, 30, 36, 48
Does your child like to be around other children? (Also *family members and friends* for 18 month interval)	18, 24
Does your child greet or say hello to familiar adults?	24, 30
Does your child do what you ask him to do?	30
Does your child move from one activity to the next with little difficulty?	30, 36, 48
Can your child name a friend?	36, 48
Do other children like to play with your child?	36, 48
Does your child like to play with other children?	36, 48
Does your child show concern for other people's feelings?	48
Does your child have simple conversations with you?	48

Social interaction testing. The Autism Diagnostic Observation Schedule, Second Edition (ADOS-2, Lord et al., 2012) was administered to assess the participants' social interaction and functional communication skills. This test is considered a gold-standard for observational assessment for autism (though diagnoses are not made in isolation based on the results of any single measure). Examples of ADOS-2 activities include: a construction task, functional and symbolic imitation, joint interactive play, and a sequenced pretend play scenario (e.g., bath time, birthday party). The Toddler Module for the ADOS-2 is a relatively new addition (Lord et al., 2012) for children 12-30 months, allowing a more complete assessment for non-verbal children and children with limited communication. The ADOS-2 has sensitivity in the upper 90% range across all modules and specificity in the upper 80% to lower 90% range for differentiating ASD from non-ASD (Lord et al., 2008); psychometric data were not updated for the ADOS-2 from the original sample due to item similarity.

The training for ADOS-2 administration is rigorous with an even higher level of training required for research reliability. These advanced trainings are conducted with a goal of ensuring good reliability and consistency in administrations. Providers who were specifically trained and clinically reliable for ADOS-2 administration administered the ADOS-2 as a part of the clinical visit. ADOS-2 training typically includes a multi-day course followed by coaching and reliability training until providers are accurate with administration and algorithm scoring with at least 80% agreement. Providers at OHSU who administered these assessments included master's and doctoral level practitioners in speech-language pathology, psychology, and occupational therapy and had attained administration reliability (i.e., 80%) as part of ADOS-2 training. The approximate

average number of years of experience for these providers in using ADOS-2 was eight years.

Human Subjects Protection

The research described in this study was approved by the OHSU Institutional Review Board (IRB) prior to participant enrollment. Additionally, the University of Oregon's IRB provided a waiver of oversight to OHSU's IRB prior to interaction with subjects or data collection. All research actions were governed by IRB policies, including ethical considerations for human subjects.

Consent for this minimal-risk study was obtained from at least one parent prior to the clinical participant's entry into the study. Because the research presented no more than minimal risk and involved no procedures for which written documentation of consent is normally required outside of the research context, consent was obtained using an Information Sheet for the clinical participants (i.e., no signature was required). Participants received a $10 gift card for Starbucks, which was located in the hospital lobby.

Data Analyses

The analyses for this study, outlined in Table 1, were selected as best to answer the proposed research questions, given the study's sample size parameters. A classification matrix was developed to measure the sensitivity and specificity of the ASQ:SE-2, as well as to calculate the negative and positive predictive values. This information was used to describe how well the ASQ:SE-2 accurately identified children at risk for ASD.

Independent samples t-tests were conducted to determine significant differences between groups of children with and without ASD diagnoses. The two groups were compared based on total ASQ:SE-2 scores and ASD item total scores.

Binary logistic regression analysis was conducted to evaluate the likelihood of an ASD based on the autism-specific item total scores. Results from these analyses are discussed next in detail.

CHAPTER IV

RESULTS

Population

Sixty children were evaluated for ASD by a multi-disciplinary team at a regional center for ASD diagnostics. Their average age was 38 months, and 46 of the 60 participates were male. Children were 12% Hispanic and 75% White (see Table 3 for full participant demographics). As per study inclusion criteria, 100% of the children's families spoke English as a primary home language. While a precise measure of socio-economic status was unavailable, a child's insurance status (i.e., public or private) was used as a broad-level proxy for a family's socio-economic status; 63% of the children received publicly funded health insurance.

Screening and Diagnostic Results

ASQ:SE-2 screening. Eighty-five percent of pre-visit paperwork was completed by mothers; 12% was completed by fathers and 3% was completed by "other" caregiver (e.g., grandmother). Forty-five percent of the ASQ:SE-2 forms completed were the 48-month interval, while only one child was in the 18-month age interval. See Table 4 for ASQ:SE-2 intervals included in the study. Of the 60 children referred for ASD assessment, only one child passed the ASQ:SE-2. Fifty-three failed the ASQ:SE-2, and 6 had scores in the "monitoring zone". As per Hardy (2015), the monitor group and the fail group were combined for analyses. See Table 5 for score results and Figure 1 for a distribution of ASQ:SE-2 total scores. Total scores across intervals are reported; however, it is important to note that because the number of items differs across

Table 3. *Participant Information*

	n	*M* / Count	Minimum	Maximum	*SD*
Gender (male)	60	46 (77%)			
Age (months)	60	37.65	19	48	8.54
Ethnicity (Hispanic)	60	7 (12%)			
Race	60				
White	45	75%			
Black	3	5%			
Multiracial	6	10%			
Asian	2	3%			
American Indian	1	2%			
Declined	3	5%			
Insurance status (public)	60	38 (63%)			

Note: Count data are presented as *n* (%).

intervals, the total number of points possible also differs across intervals. The range of total points possible, with concerns included, for the selected intervals is 465 to 540 (e.g., 31 scored items on the 18-month interval; 36 scored items on the 48-month interval).

The scores followed a normal distribution, with one potential outlier. This participant's parent indicated a very high level of concern, scoring the maximum point value on many items. Because the answers appear to be an accurate representation of this parent's concern, the score remained in the analyses. The cut-off scores for each interval vary, but the group average ASQ:SE-2 total score (153.44, indicating social-emotional risk) was above the cut-off for every interval. See Table 5 for screening and diagnostic results.

Table 4. *ASQ:SE-2 Intervals*

	Frequency	Percent
18 month	1	1.7%
24 month	5	8.3%
30 month	11	18.3%
36 month	16	26.7%
48 month	27	45.0%
Total	60	100.0%

Table 5. *Screening and Diagnostic Results*

	n	*M* / Count	Minimum	Maximum	*SD*
ASQ:SE-2 total score	60	153.44	50	370	55.42
ASQ:SE-2 total score (without concerns)	60	134.77	50	255	42.20
ASQ:SE-2 (pass)	60	1 (2%)			
ASD item total score	60	51.33	15	115	23.54
ASD item total score (without concerns)	60	46.08	15	90	18.28
ASD diagnosis given	60	37 (62%)			

Note: Count data are presented as *n* (%). Also note that total possible scores differ across intervals.

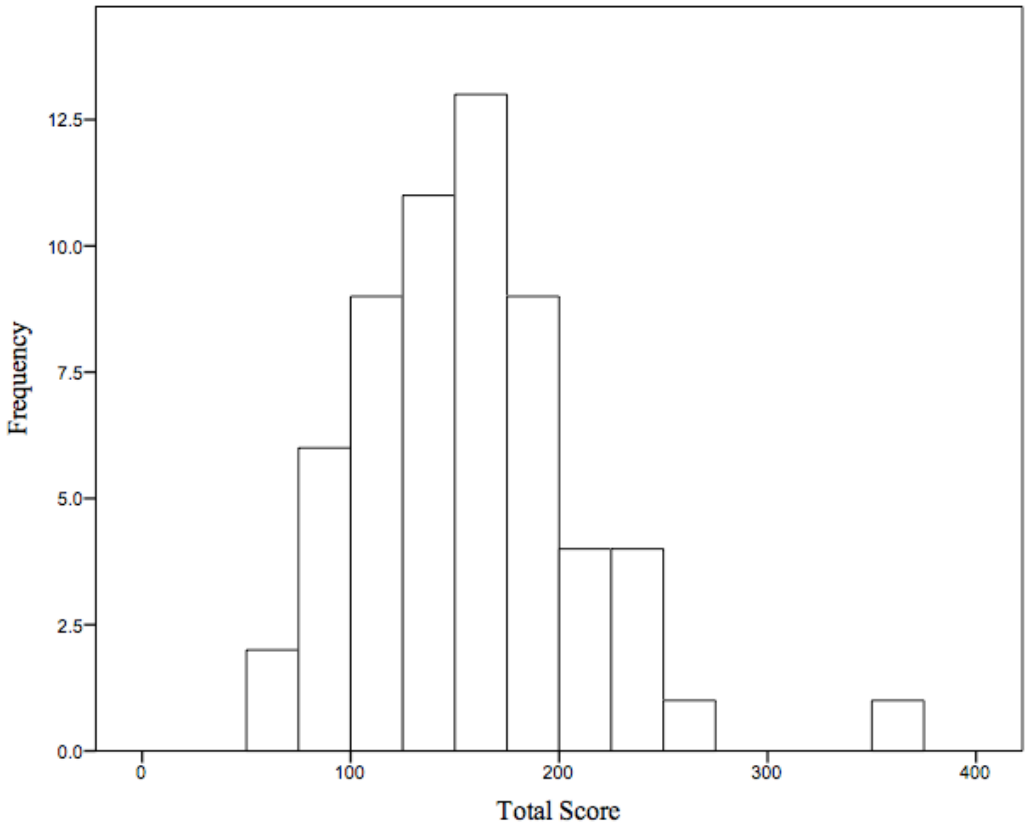

Figure 1. ASQ:SE-2 Total Score Distribution.

ASQ:SE-2 ASD items. The average ASD item total score across intervals was 51.33, with a standard deviation of 23.54 (also presented in Table 5). See Figure 2 for score distributions, which followed a normal curve. ASD item total scores were entered into the regression analyses described later in this chapter.

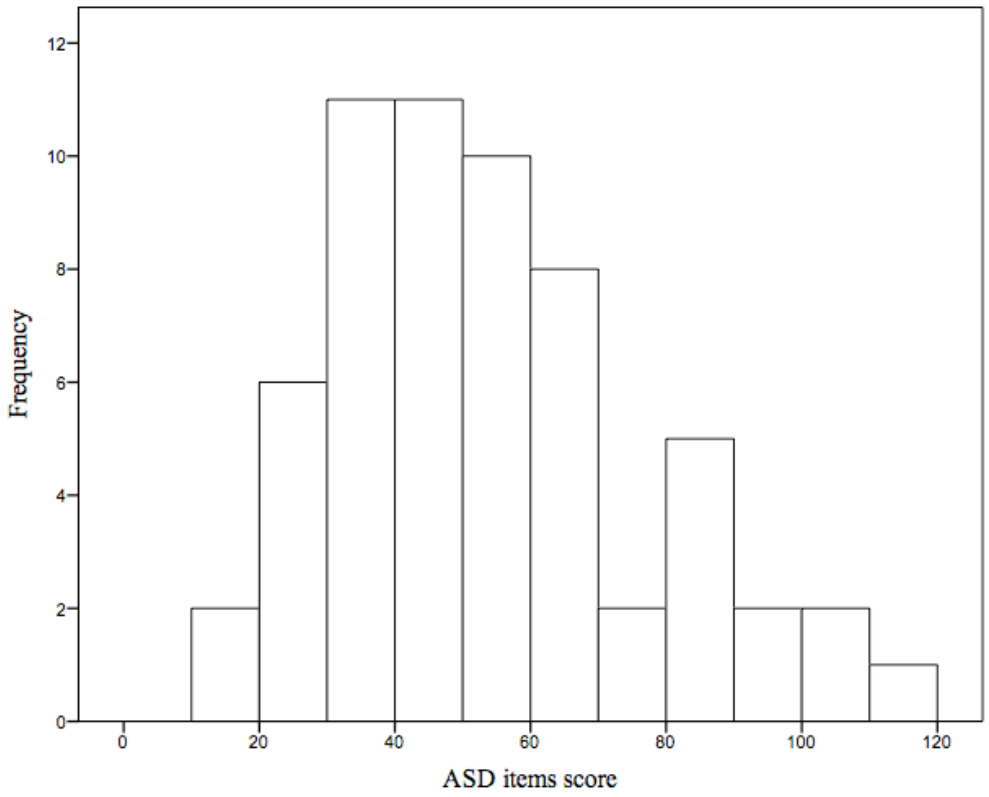

Figure 2. ASD Item Total Score Distribution

ASQ:SE-2 scores without concerns. As noted in Chapter 3, ASQ:SE-2 scores were also calculated without including the parent concern option, which adds an extra five points to the item score (i.e., "Check if this is a concern"). Thirty-eight percent of parents did not check any boxes in the concerns section (23 of 60 parents). Total points for the concerns item ranged from 0 to 115, with an average of 18.67 and a standard deviation of 23.83. Refer to Table 5 for ASQ:SE-2 Total Score without concerns, and ASQ:SE-2 ASD Item Total Score without concerns.

ASD Assessment. Of the 60 children referred for ASD evaluation, 37 (62%) received a diagnosis of ASD (Table 5). The two groups (ASD diagnosis or no ASD diagnosis) were compared using ASQ:SE-2 total score and ASD item total score (see

Figure 3). For girls, 64% of those referred were given a diagnosis of ASD, and 61% of referred boys were given a diagnosis of ASD. Table 6 displays ASD diagnoses by age using ASQ:SE-2 interval to group children by age.

Table 6. *ASD Diagnosis by ASQ:SE-2 Age-interval*

		ASD Diagnosis			
		No	Yes	Total	% ASD by age
ASQ:SE-2 Interval	18	1	0	1	0
	24	4	1	5	20
	30	4	7	11	62
	36	3	13	16	81
	48	11	16	27	59
	Total	23	37	60	62

Classification

A 2 x 2 contingency table was created to evaluate the classification agreement of the ASQ:SE-2 (pass or fail) and the diagnostic label (clinical team diagnosis of ASD or not). Table 7 displays the ASQ:SE-2's sensitivity, specificity, positive predictive value, and negative predictive value.

Comparing Groups

ASQ:SE-2 total score T-tests. There was no significant difference in total ASQ:SE-2 scores for participants with and without ASD diagnoses (see Figure 3). There

was also no significant difference in total ASQ:SE-2 scores for these two groups, when the concerns item was omitted from the total.

Table 7. *Classification Agreement Between ASQ:SE-2 and ASD Diagnostic Label*

		ASD Diagnosis		
		ASD	Non-Spectrum	Total
ASQ:SE-2 Result	Fail	37	22	59
	Pass	0	1	1
	Total	37	23	60

Sensitivity	100.00%
Specificity	4.35%
False Positive	95.65%
False Negative	0.00%
% Agreement	63.33%
Under-Identified	0.00%
Over-Identified	36.67%

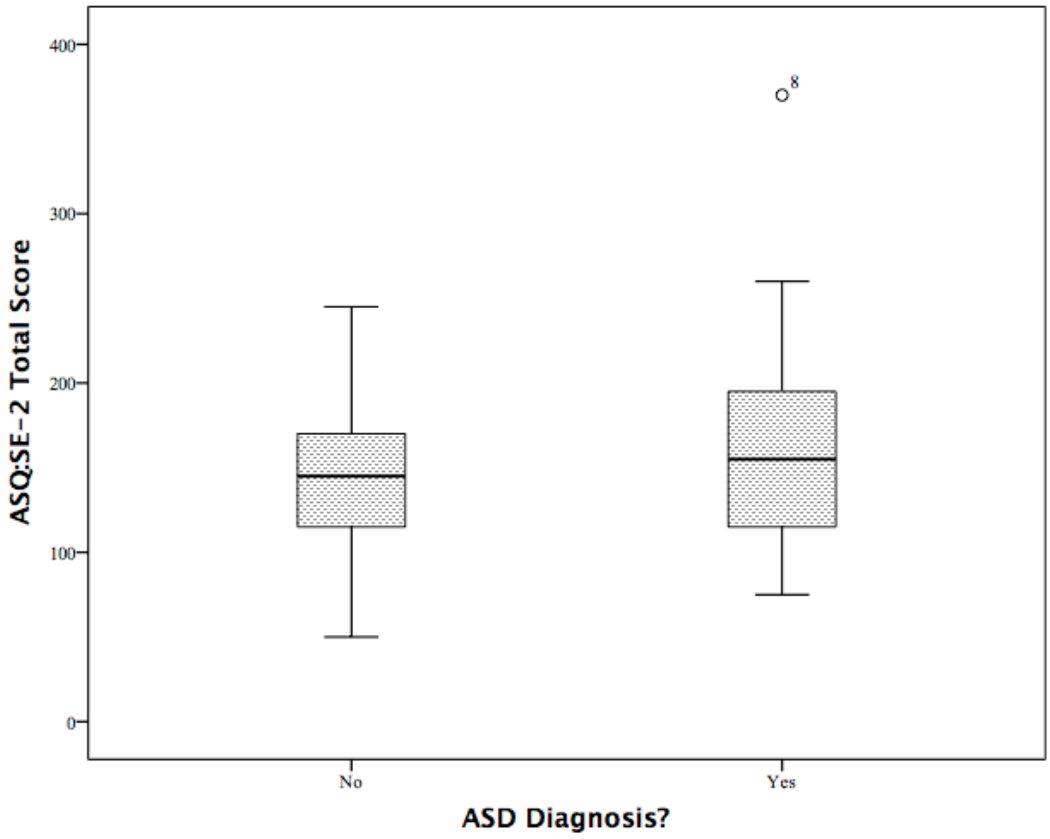

Figure 3. ASQ:SE Total Scores by ASD Diagnosis.

ASQ:SE-2 ASD item total score T-tests. There were 37 participants with an ASD diagnosis and 23 participants with no ASD diagnosis. An independent-samples t-test was run to determine if there were differences in ASD item total scores between participants with and without ASD. There was one outlier in the data, as assessed by inspection of a boxplot. It was left in the analysis as its presence was not expected to adversely affect the results; indeed, running the analysis without the outlier provided the same significance level. There was homogeneity of variances in ASD item total scores for each level of ASD diagnosis, as assessed by Levene's test for equality of variances (p = .08). Participants with ASD received higher scores on ASD items (60.00 ± 23.45) than

participants without ASD (37.39 ± 16.02), a statistically significant difference of 22.61 (95% CI, 11.48 to 33.74), $t(58) = -4.065$, $p < .001$.

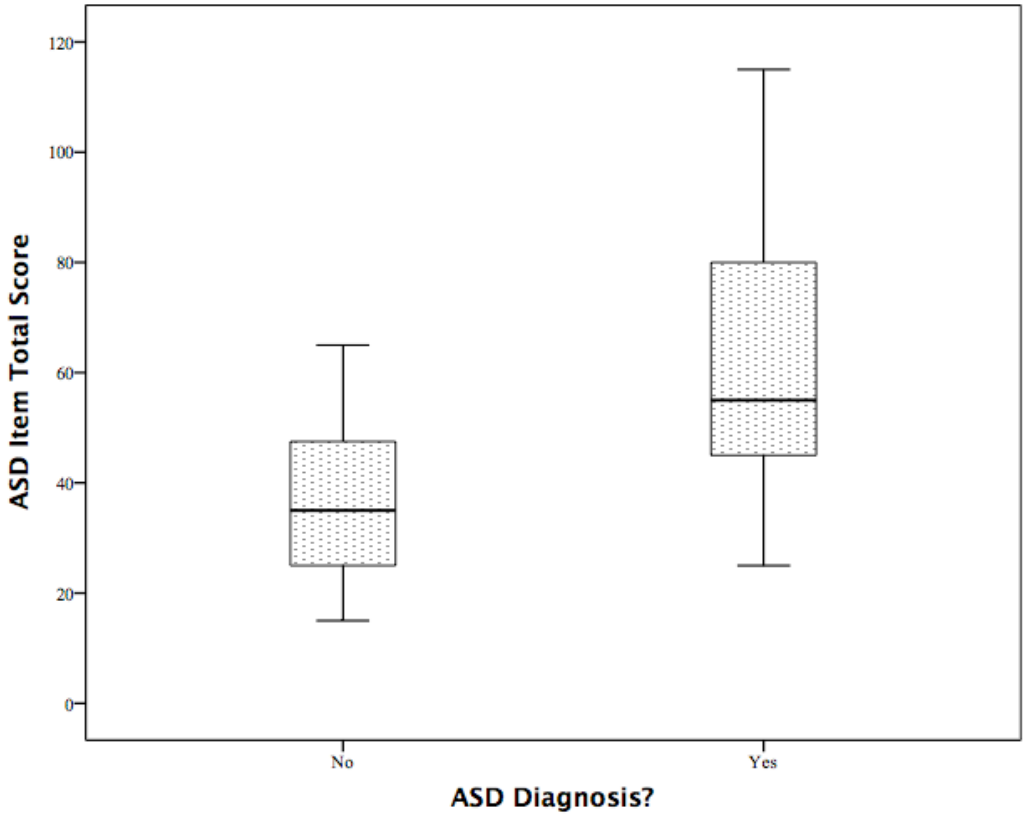

Figure 4. ASD Item Total Scores by ASD Diagnosis.

ASQ:SE-2 ASD item total scores without concerns. There were 37 participants with an ASD diagnosis and 23 participants with no ASD diagnosis. An independent-samples t-test was run to determine if there were differences in ASD item total scores between participants with and without ASD, when concerns were omitted from the totals. There were no outliers in the data, as assessed by inspection of a boxplot. There was homogeneity of variances in ASD item total scores for each level of ASD diagnosis, as assessed by Levene's test for equality of variances ($p = .09$). Participants with ASD received higher scores on ASD items (53.38 ± 17.48) than participants without ASD

(34.35 ± 12.73), a statistically significant difference of 19.03 (95% CI, 10.61 to 27.45), $t(58) = -4.522$, $p < .0001$ (see Figure 4).

Binomial Logistic Regression

ASD item total scores. A binomial logistic regression was performed to ascertain the effects of the ASD item total scores on the likelihood that participants had ASD. Linearity of the continuous variables with respect to the logit of the dependent variable was assessed via the Box-Tidwell (1962) procedure. Based on this assessment, the single continuous independent variable was found to be linearly related to the logit of the dependent variable. The logistic regression model was statistically significant, $X^2(1) = 16.67$, $p < .0001$. The model explained 33.0% (Nagelkerke R^2) of the variance in ASD diagnosis and correctly classified 70.0% of cases. Sensitivity was 81.1%; specificity was 52.2%. The positive predictive value was 73.2%, and the negative predictive value was 63.2%, though these values refer to the current clinical sample and not the population as a whole. The odds ratio was 1.06 (with 95% confidence interval between 1.03 and 1.10), indicating that for every one-point increase in ASD item total score, a diagnosis of ASD became 1.06 times as likely. Increasing ASD item total score was associated with an increased likelihood of exhibiting ASD.

ASD item total scores without concerns. A binomial logistic regression was performed to ascertain the effects of the ASD item total scores, when the concern item was omitted, on the likelihood that participants had ASD. Linearity of the continuous variables with respect to the logit of the dependent variable was assessed via the Box-Tidwell (1962) procedure. Based on this assessment, the single continuous independent variable was found to be linearly related to the logit of the dependent variable, and there

were no significant outliers. The logistic regression model was statistically significant, $X^2(1) = 18.62$, $p < .0001$. The model explained 36.3% (Nagelkerke R^2) of the variance in ASD diagnosis and correctly classified 70.0% of cases. Sensitivity was 75.7%; specificity was 60.9%. Positive predictive value was 75.7%, and negative predictive value was 60.9%, though these values refer to the current clinical sample and not the population as a whole. The odds ratio was 1.09 (with 95% confidence interval between 1.04 and 1.14), indicating that for every one-point increase in ASD item total score, a diagnosis of ASD became 1.09 times as likely. Increasing ASD item total score was associated with an increased likelihood of exhibiting ASD, both with the concerns item included and omitted.

Creation of Cut-off Score for ASD Screen on ASQ:SE-2

A receiver operating characteristic (ROC) curve was produced to interpret sensitivity and specificity levels for the ASD Item Total Scores related to ASD diagnosis. The resulting area under the curve (AUC) for this analyses was .78, representing moderate accuracy and reliability, which may be appropriate given its use as a screening measure (see Figure 5). This was a statistically significant finding. The 95% confidence of the AUC for this measure was between .67 and .90. Utilizing the coordinates of the curve, a cut score of 40 was selected indicating that a score of 40 or above on the Autism Item Total Scores would indicate a positive Autism Screen on the ASQ:SE-2. Using this cut score, a new contingency table was created (see Table 8). The cut score of 40 resulted in sensitivity and specificity levels that were equal to those created by the binomial logistic regression model: 81% sensitivity and 52% specificity.

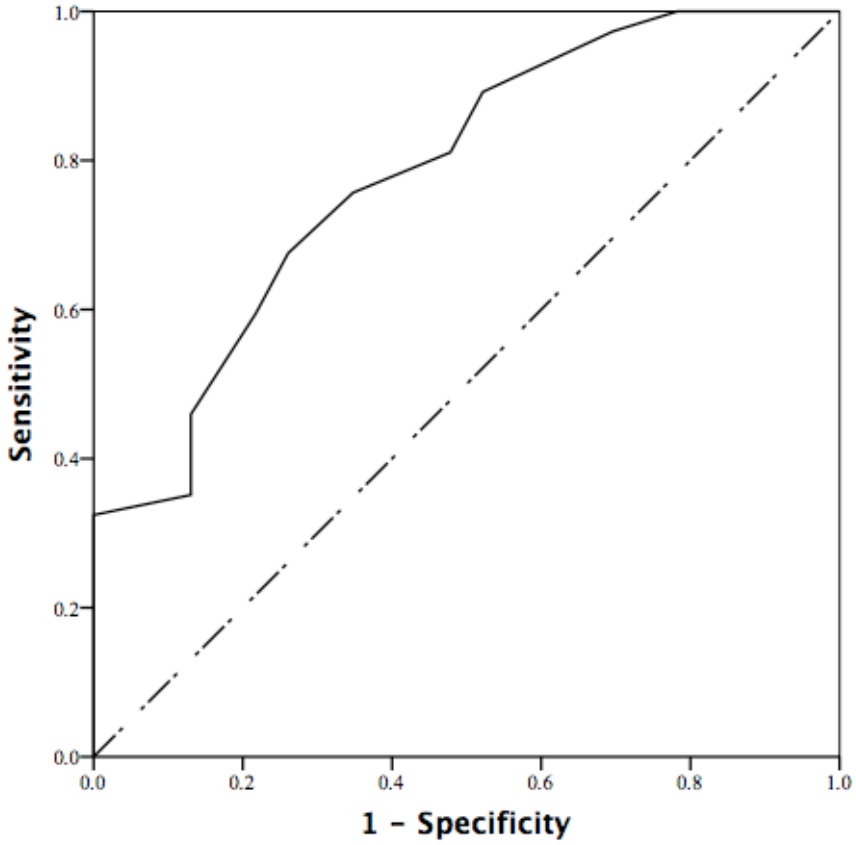

Figure 5. Receiver Operating Characteristic Curve for ASD Item Score by Diagnosis

Table 8. *Classification Agreement Between ASQ:SE-2 Autism Item Total and ASD Diagnostic Label*

		ASD Diagnosis		
		ASD	Non-Spectrum	Total
Autism Item Total	Fail	30	11	41
	Pass	7	12	1
	Total	37	23	60

Note. Autism Item Total cut-off = 40.

CHAPTER V

DISCUSSION

Early and accurate identification of children at risk for ASD remains a critically important component of pediatric healthcare. Recently the United States Preventative Services Task Force (2016) issued a statement on ASD screening, concluding that there was insufficient evidence to assess the balance of benefits and harms of developmental screening for children not at risk for ASD (i.e., "universal screening"). Some researchers worried that this statement may undermine recent efforts to increase screening for ASD (Dawson, 2016), but for many, including Dawson, the statement served as an impetus to increase research in the areas of screening and ASD treatment efficacy. Indeed, research has clearly shown benefits from early intervention for children with ASD, both in general developmental areas (Dawson et al., 2010; Corsello, 2005; Harris & Handleman, 2000; Sheinkopf & Siegel, 1998) and with regard to core ASD symptomatology (Landa et al., 2011; Kasari, Paparella, Freeman, and Jahromi (2008). There is a current need for more research to address the efficacy of screening tools and the efficacy of available interventions to support developmental growth.

This study explored the use of a general social-emotional screening tool as an ASD specific screener. It is expected that children with ASD will not pass a social-emotional screening tool for young children, but certainly not all children who fail such a screening will be diagnosed with ASD or will even require an evaluation to rule out ASD. Children with social-emotional differences may have language delays or disorders, behavior regulation problems, attention problems, developmental delays, or mental health disorders. In this study, it was hypothesized that certain items on a general screener may

be specifically related to ASD. By creating a subset of ASD related items, it was hypothesized that a general social-emotional screener, the ASQ:SE-2, may be able to assist in predicting ASD diagnosis, pointing to the need for further ASD evaluation.

As predicted, the vast majority of children (59 of 60) referred for an ASD evaluation failed the ASQ:SE-2, reflecting a parental concern in the area of social emotional development. Importantly, however, only 37 of these children received a diagnosis of ASD. All of the children with ASD failed the ASQ:SE-2, as well as 22 children who did not have ASD. This finding supports the hypothesis that a general social-emotional screener is insufficient for differentiating ASD from other social-emotional delays or differences.

Total score findings. While a simple pass or fail on the ASQ:SE-2 did not differentiate between ASD or No ASD groups, it was hypothesized that the two groups might differ on total scores on the ASQ:SE-2 (i.e., perhaps children with ASD would have overall higher total scores). This was not found to be the case, as there were no significant differences between the groups when considering their total scores on the ASQ:SE-2.

ASD item total score findings. This study included the creation of a subset of ASD related items (9) from each interval. Using scores from only these items, the two groups were again compared (ASD or no ASD). Children who were diagnosed with ASD scored significantly higher on ASD-related items (i.e., the ASD item total score) than children who did not have a diagnosis of ASD (though they may have received other diagnoses). Additionally, when the ASD item total scores were entered into a logistic regression, the ASD item scores significantly predicted a diagnosis of ASD, in that

increasing ASD item total scores were associated with an increased likelihood of a child having an ASD diagnosis.

The specific social-emotional screening tool used in this study included an option for parents to select if certain items were "a concern" or not. Because parents may utilize this aspect of the screening tool differently (e.g., 38% of parents never used these boxes at all), the analyses were conducted both with and without the concerns points added in. Results were the same for all analyses; there were significant differences in ASD item total scores for children with and without ASD regardless of whether the parent concerns option was included. Similarly, ASD item total scores without the parent concerns item included continued to significantly predict ASD diagnoses in the binomial logistic regression analysis.

Implications

This study provides preliminary support for the use of a subset of ASD related items from a broadband social-emotional screener to assist in identifying a child's risk for ASD. Findings suggest that total scores alone are inadequate for differentiating between ASD and other disorders that may result in social-emotional differences or delays. Analyses were conducted both with and without the use of a specific parent concern item, and results provided significant findings in both cases. This provides additional support for the consideration and validation of parent concerns.

As described above, the United States Preventative Services Task Force (Siu, 2016) issued an ambivalent statement on the recommendation to provide universal ASD screenings. While the American Academy of Pediatrics continues to recommend universal ASD related screenings (Dreyer, 2016), it may be useful to some practitioners if

similar information could be gained through the use of a broadband social-emotional screener. The Task Force statement is specific to universal ASD screening (i.e., administering a screening tool to all children regardless of parent or practitioner concern) and does not affect screening for children at risk for ASD (as in the present study). Importantly, the statement also does not apply to the use of general broadband screening tools, which continue to be recommended for use, universally. Providers may continue to use broadband screening tools and may appreciate the ability to screen for ASD at the same time.

One goal for the creation of the subset of ASD-related items was to improve the specificity of the ASQ:SE-2 (or a broadband social-emotional screener in general) for detecting ASD. Prior to any modification, and when using the ASQ:SE-2 Total Scores or pass/fail categorical result, the specificity for ASD was poor (see Table 6). Using the ASD subset of items, and with logistic regression, 70% of cases were correctly classified (as ASD or not), and the specificity improved from 4% to between 52% and 61% (depending on use of parental concerns or not). Because this is a screening tool, and not a diagnostic tool, a 70% classification rate is likely acceptable, particularly because this screening result would be paired with provider judgment and parental report of concern related to possible ASD.

Clinical relevance. While this is a preliminary study, there is potential for an Autism Item Total on the ASQ:SE-2 to be clinically useful. As described above, there are barriers (Dosreis, Weiner, Johnson, & Newschaffer, 2006) to providers completing the AAP recommended screenings (Dreyer, 2016; Johnson & Meyers, 2007. There is also a current uncertainty about differing recommendations from the United States

Preventative Services Task Force recommendations (Siu et al., 2016) and the American Academy of Pediatrics (Dreyer, 2016). Providers may appreciate the ability to gather some information related to a child's risk for ASD from the use of a broadband social-emotional screener, particularly one that is reliable, valid, and already in use across the country (ASQ:SE-2; Squires, Bricker, & Twombly, 2015). Many pediatric offices are stretched for time during well-child visits, and it may be helpful to utilize one developmental screening tool for multiple purposes. It is important to reiterate that the intended purpose for utilizing an ASD subset of items would be to support the referral process, not diagnose ASD. If a provider completes the ASQ:SE-2 and the score is in the "fail" range, use of the Autism Screen (i.e., the ASD Item Total Score) may help a provider determine if that child demonstrates a heightened risk specific to ASD and point to further ASD specific evaluation.

Use of an Autism Item Total indicator within a broadband social-emotional screener may also address the United States Preventative Task Force's concerns regarding the possible "harm" of conducting universal ASD screenings (Siu et al., 2016). Conducting an embedded ASD screening within the context of a broadband screening improves the universal ASD screening process in two ways. Use of the ASD Screen on the ASQ:SE-2 increases the specificity of the broadband screener for ASD, thereby reducing the false positive results. This reduces unnecessary parental concern for ASD and also reduces the amount of children referred for specialty developmental clinics that often have long waitlists. Use of the ASD Item Total may reduce the ASQ:SE-2's overall sensitivity to ASD, but this is to be expected with a rise in specificity, and the tool is

intended to be used in conjunction with clinical judgment, developmental surveillance, and discussion with family.

Limitations

One limitation of the study is its sample size of 60 participants. While the sample size is large enough for the statistical analyses used, it was not large enough to evaluate other demographic variables, including the race, gender, and socio-economic status of the participants. Recent studies have shown racial and ethnic disparities in identification of children with ASD; children who were Black, Hispanic, or of other race/ethnicity were less likely than White children to have documented ASD, and this finding has been consistent across various studies (Mandell et al., 2009; Zuckerman et al., 2013; Liptak et al., 2008). The present study included a diverse group of children, but the sample was limited to English-speaking families, and the numbers in each group were not adequate to allow for comparisons between groups of children.

Additionally, the current study did not consider the possibility that various subgroups within the group of participants may display ASD symptomatology differently. Tek and Landa (2012) reported differences in ASD symptoms between minority and non-minority toddlers. Recent research has also explored the idea of how children of different genders may display ASD characteristics differently (Lai, Lombardo, Auyeung, Chakrabarti, & Baron-Cohen, 2015). With regard to general social-emotional development (as measured by the ASQ:SE-2), Chen et al. (2015) described both gender and cultural differences on some social-emotional competencies.

Finally, the sample is a clinical sample, because all children were referred for an ASD evaluation. This sampling affects the interpretation of the positive and negative

predictive values because the prevalence of ASD in the current study (62%) does not reflect the current known prevalence of ASD, which is one in 68 children (CDC, 2014). As described in Parikh, Mathai, Sekhar, and Thomas (2008), using a clinical sample inflates the positive predictive value and deflates the negative predictive value. The sampling method for this study was chosen intentionally to be able to evaluate the ability of a broadband social-emotional screener to *differentiate* between children with potential behavior or language problems and children with ASD. The ASQ:SE-2 has established reliability and validity, so its ability to differentiate between the general population and those with ASD is presumed. A more difficult task is to differentiate between children with a variety of social-emotional differences (e.g., ASD, Attention Deficit Hyperactivity Disorder, Language Disorder, etc.). A larger sample size would allow for a more thorough examination into the various phenotypical presentations of ASD.

Future Directions

The current application of an ASD subset of scores to a broadband social-emotional screener will benefit from a larger scale study to further examine the psychometric properties. With a large sample size, subgroups can be compared to determine if demographic variables are related to the screening tool's efficacy. Variables of interest include race, ethnicity, language use, gender, maternal education and/or socio-economic status.

A larger scale study will also allow for population sampling to include both children at risk for ASD as well as children who are not at risk for ASD. This will allow for a more thorough examination of the sensitivity and specificity of the tool for ASD.

With more data, a cut-score could be created for the ASD item total score. This could be done using a receiver operating characteristic curve, or through the use of semi-interquartile ranges. With a cut-score, a provider would have a simplified method of interpreting ASD item scores; for example, an ASD item total score above a certain number may suggest an ASD discussion with the family or a referral for an ASD evaluation.

Conclusion

In the present study, a broadband social-emotional screening tool was used as an ASD specific screening tool by creating a subset of items that specifically related to ASD. This was a necessary step, as the broadband social-emotional screening tool does not differentiate well between children with ASD and children with other social-emotional differences or delays. Indeed, in the present study that included children in a clinical or referred sample, there was no difference in total scores between the group of children who eventually received a diagnosis of ASD and the group of children who did not. When the ASD-related items were considered alone, the two groups (i.e., ASD or no ASD) did have significantly different scores, indicating that parents of children with ASD report significantly more concerns on ASD-related symptoms and that these concerns can differentiate them from children without ASD. Moreover, the study results suggest that an increasing ASD item total score results in an increased likelihood of a child receiving an ASD diagnosis.

The findings from the present study are important for several reasons. First, early ASD identification appears to be related to prognosis, since some of the best empirically supported ASD treatments are geared towards very young children (Fernell, Eriksson, &

Gillberg, 2013; Dawson et al., 2010). If children at risk for ASD are identified early and referred for comprehensive evaluation, they may have earlier access to treatments and family support. The AAP recommends ASD specific screening, using ASD specific tools, at 18- and 24-months in addition to general developmental surveillance in the office (Dreyer, 2016). The AAP continues to recommend this process, even though a recent statement from the United States Preventative Services Task Force (2016) calls into question the benefit of universal screening for ASD. Clearly more research is needed to support the usefulness of universal screening, but in the meantime, it may be useful to be able to utilize a broadband screening tool to evaluate a child's risk for ASD.

Pediatricians and family practice physicians are pressed for time and have an obligation to observe a child's physical, mental, and developmental growth in a short amount of time. Screening tools can support practitioners' ability to reliably provide information in these areas. If some tools can provide information on multiple developmental areas at once, it may ameliorate some of the difficulty providers have in completing all necessary screenings and may support earlier access to comprehensive assessment and services. A broadband social-emotional screener, when used as recommended by Briggs et al., 2012, can provide information about a child's social-emotional development as well as specific information related to the child's risk for ASD. In the present study, increased parental report of difficulty on ASD-related items resulted in an increased likelihood of an ASD diagnosis. Findings from the present study support the potential use of a broadband social-emotional screening tool as an ASD-specific screener.

With continued research, improved screening instruments and procedures can be developed for early autism detection. Effective screening relies on easy-to-administer tests and family-friendly procedures such as those highlighted in this study. Early detection will result in improved outcomes for children and families, and cost savings for families, schools, and communities.

APPENDIX A

SAMPLE ASQ:SE-2 QUESTIONNAIRE

18 Month Questionnaire

15 months 0 days through 20 months 30 days

ASQ:SE-2 Ages & Stages Questionnaires® Social-Emotional SECOND EDITION

Date ASQ:SE-2 completed: _____

Child's information

Child's first name: | Child's middle initial: | Child's last name:

Child's date of birth: | If child was born 3 or more weeks premature, please enter the number of weeks:

Child's gender: ◯ Male ◯ Female

Person filling out questionnaire

First name: | Middle initial: | Last name:

Street address:

City: | State/ province: | ZIP/postal code:

Country: | Home telephone number: | Other telephone number:

E-mail address:

Relationship to child: ◯ Parent ◯ Guardian ◯ Teacher ◯ Other: _____
◯ Grandparent/ other relative ◯ Foster parent ◯ Child care provider

People assisting in questionnaire completion:

Program information (For program use only.)

Child's ID #: | Age at administration in months and days:

Program ID #: | If premature, adjusted age in months and days:

Program name:

P201180000

18 Month Questionnaire *15 months 0 days through 20 months 30 days*

ASQ:SE-2

Questions about behaviors children may have are listed on the following pages. Please read each question carefully and check the box ☑ that best describes your child's behavior. Also, check the circle ⊘ if the behavior is a concern.

Important Points to Remember:

- ❑ Answer questions based on what you know about your child's behavior.
- ❑ Answer questions based on your child's *usual* behavior, not behavior when your child is sick, very tired, or hungry.
- ❑ Caregivers who know the child well and spend more than 15–20 hours per week with the child should complete ASQ:SE-2.
- ❑ Please return this questionnaire by: _____
- ❑ If you have any questions or concerns about your child or about this questionnaire, contact: _____
- ❑ Thank you and please look forward to filling out another ASQ:SE-2 in _____ months.

	OFTEN OR ALWAYS	SOME-TIMES	RARELY OR NEVER	CHECK IF THIS IS A CONCERN	
1. Does your child look at you when you talk to him?	☐ z	☐ v	☐ x	◯ v	____
2. When you leave, does your child stay upset and cry for more than an hour?	☐ x	☐ v	☐ z	◯ v	____
3. Does your child laugh or smile when you play with her? 	☐ z	☐ v	☐ x	◯ v	____
4. Does your child look for you when a stranger comes near?	☐ z	☐ v	☐ x	◯ v	____
5. Is your child's body relaxed?	☐ z	☐ v	☐ x	◯ v	____
6. Does your child like to be hugged or cuddled?	☐ z	☐ v	☐ x	◯ v	____
7. When upset, can your child calm down within 15 minutes?	☐ z	☐ v	☐ x	◯ v	____

TOTAL POINTS ON PAGE _____

P201180100

18 Month Questionnaire

ASQ:SE-2

Check the box ☑ that best describes your child's behavior.
Also, check the circle ◉ if the behavior is a concern.

	OFTEN OR ALWAYS	SOME-TIMES	RARELY OR NEVER	CHECK IF THIS IS A CONCERN	
8. Does your child stiffen and arch his back when picked up?	☐ x	☐ v	☐ z	◯ v	____
9. Does your child cry, scream, or have tantrums for long periods of time?	☐ x	☐ v	☐ z	◯ v	____
10. Is your child interested in things around her, such as people, toys, and foods?	☐ z	☐ v	☐ x	◯ v	____
11. Does your child do things over and over and get upset when you try to stop him? For example, does he rock, flap his hands, spin, or _____? (Please describe.) _____ _____	☐ x	☐ v	☐ z	◯ v	____
12. Does your child have eating problems? For example, does she stuff food, vomit, eat things that are not food, or _____? (Please describe.) _____ _____	☐ x	☐ v	☐ z	◯ v	____
13. Does your child have trouble falling asleep at naptime or at night?	☐ x	☐ v	☐ z	◯ v	____
14. Do you and your child enjoy mealtimes together?	☐ z	☐ v	☐ x	◯ v	____
15. Does your child sleep at least 10 hours in a 24-hour period?	☐ z	☐ v	☐ x	◯ v	____
16. When you point at something, does your child look in the direction you are pointing?	☐ z	☐ v	☐ x	◯ v	____

TOTAL POINTS ON PAGE ____

P201180200

18 Month Questionnaire

ASQ:SE-2

Check the box ☑ that best describes your child's behavior.
Also, check the circle ◉ if the behavior is a concern.

	OFTEN OR ALWAYS	SOME-TIMES	RARELY OR NEVER	CHECK IF THIS IS A CONCERN	
17. Does your child get constipated or have diarrhea?	☐ x	☐ v	☐ z	◯ v	____
18. Does your child let you know how he is feeling with gestures or words? For example, does he let you know when he is hungry, hurt, or tired?	☐ z	☐ v	☐ x	◯ v	____
19. Does your child follow simple directions? For example, does she sit down when asked?	☐ z	☐ v	☐ x	◯ v	____
20. Does your child like to play near or be with family and friends?	☐ z	☐ v	☐ x	◯ v	____
21. Does your child check to make sure you are near when exploring new places, such as a park or a friend's home?	☐ z	☐ v	☐ x	◯ v	____
22. Does your child like to hear stories or sing songs? 	☐ z	☐ v	☐ x	◯ v	____
23. Does your child hurt himself on purpose?	☐ x	☐ v	☐ z	◯ v	____
24. Does your child like to be around other children? For example, does she move close to or look at other children? 	☐ z	☐ v	☐ x	◯ v	____
25. Does your child try to hurt other children, adults, or animals (for example, by kicking or biting)?	☐ x	☐ v	☐ z	◯ v	____

TOTAL POINTS ON PAGE ____

18 Month Questionnaire

ASQ:SE-2

Check the box ☑ that best describes your child's behavior.
Also, check the circle ◉ if the behavior is a concern.

	OFTEN OR ALWAYS	SOME-TIMES	RARELY OR NEVER	CHECK IF THIS IS A CONCERN	
26. Does your child try to show you things by pointing at them and looking back at you?	☐ z	☐ v	☐ x	◯ v	____
27. Does your child make sounds or use words or gestures to let you know he wants something (for example, by reaching)?	☐ z	☐ v	☐ x	◯ v	____
28. Does your child play with objects by pretending? For example, does your child pretend to talk on the phone, feed a doll, or fly a toy airplane?	☐ z	☐ v	☐ x	◯ v	____
29. Does your child wake three or more times during the night?	☐ x	☐ v	☐ z	◯ v	____
30. Does your child respond to her name when you call her? For example, does she turn her head and look at you?	☐ z	☐ v	☐ x	◯ v	____
31. Has anyone shared concerns about your child's behaviors? If "sometimes" or "often or always," please explain: _____ _____ _____	☐ x	☐ v	☐ z	◯ v	____

TOTAL POINTS ON PAGE ____

OVERALL Use the space below for additional comments.

32. Do you have concerns about your child's eating or sleeping behaviors? If yes, please explain: ◯ YES ◯ NO

33. Does anything about your child worry you? If yes, please explain: ◯ YES ◯ NO

34. What do you enjoy about your child?

18 Month Information Summary *15 months 0 days through 20 months 30 days*

ASQ:SE-2

Child's name: _____ Date ASQ:SE-2 completed: _____

Child's ID #: _____ Child's date of birth: _____

Person who completed ASQ:SE-2: _____ Child's age/adjusted age in months and days: _____

Administering program/provider: _____ Child's gender: ◯ Male ◯ Female

1. ASQ:SE-2 SCORING CHART:

- Score items (*Z* = 0, *V* = 5, *X* = 10, *Concern* = 5).
- Transfer the page totals and add them for the total score.
- Record the child's total score next to the cutoff.

TOTAL POINTS ON PAGE 1	
TOTAL POINTS ON PAGE 2	
TOTAL POINTS ON PAGE 3	
TOTAL POINTS ON PAGE 4	
Total score	

Cutoff	**Total score**
65	

2. ASQ:SE-2 SCORE INTERPRETATION: Review the approximate location of the child's total score on the scoring graphic. Then, check off the area for the score results below.

____ The child's total score is in the ▭ area. It is below the cutoff. Social-emotional development appears to be on schedule.

____ The child's total score is in the ▭ area. It is close to the cutoff. Review behaviors of concern and monitor.

____ The child's total score is in the ▬ area. It is above the cutoff. Further assessment with a professional may be needed.

3. OVERALL RESPONSES AND CONCERNS: Record responses and transfer parent/caregiver comments. YES responses require follow-up.

1–31. Any Concerns marked on scored items? **YES** no Comments:

32. Eating/sleeping concerns? **YES** no Comments:

33. Other worries? **YES** no Comments:

4. FOLLOW-UP REFERRAL CONSIDERATIONS: Mark all as Yes, No, or Unsure (Y, N, U). See pages 98–103 in the *ASQ:SE-2 User's Guide*.

____ **Setting/time factors** (e.g., Is the child's behavior the same at home as at school?)

____ **Developmental factors** (e.g., Is the child's behavior related to a developmental stage or delay?)

____ **Health factors** (e.g., Is the child's behavior related to health or biological factors?)

____ **Family/cultural factors** (e.g., Is the child's behavior acceptable given the child's cultural or family context? Have there been any stressful events in the child's life recently?)

____ **Parent concerns** (e.g., Did the parent/caregiver express any concerns about the child's behavior?)

5. FOLLOW-UP ACTION: Check all that apply.

____ Provide activities and rescreen in ____ months.

____ Share results with primary health care provider.

____ Provide parent education materials.

____ Provide information about available parenting classes or support groups.

____ Have another caregiver complete ASQ:SE-2. List caregiver here (e.g., grandparent, teacher): _____

____ Administer developmental screening (e.g., ASQ-3).

____ Refer to early intervention/early childhood special education.

____ Refer for social-emotional, behavioral, or mental health evaluation.

____ Other: _____

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