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THE PREDICTIVE INFLUENCE OF CHALLENGING BEHAVIOR ON PARENT STRESS IN YOUNG CHILDREN WITH AUTISM SPECTRUM DISORDER

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Abstract

Autism spectrum disorder (ASD) is a neurodevelopmental disorder characterized by deficits in social communication, restricted interest, and repetitive patterns of behavior. Individuals with ASD also exhibit challenging behaviors that affect parent and caregiver stress directly. However, researchers have not yet examined the predictive influence of specific challenging behaviors on parent stress, particularly in young children (i.e., infants and toddlers) with ASD. Therefore, the current study expands existing literature by a) investigating the influence that challenging behaviors of young children with ASD have on parent stress and b) examining the unique contribution that each behavior (i.e., aggressive/disruptive behavior, stereotypy, and self-injurious behavior) adds to the relationship. A standard multiple linear regression was conducted to examine the impact of challenging behavior severity scores on parent distress in 54 parents of young children with ASD. Results revealed that the overall severity of challenging behaviors significantly predicted parent distress. However, aggressive/disruptive behavior and stereotypy did not add statistical significance to the model. Interestingly, SIB was the only significant predicting factor that added unique variance to predicting parent distress.

Introduction

Autism Spectrum Disorder

History

As early as the 1800s, ASD symptomology has been documented by several physicians mentioning symptoms consistent with the current diagnostic criteria of the disorder (Deisinger, 2011; Feinstein, 2011; Vaillant, 1962). In 1908, the term 'autism' was first mentioned by a Swiss psychiatrist named Eugene Bleuler. Bleuler used the word to describe patients who were considered to have schizophrenia (Deisinger, 2011; Feinstein, 2011; Goldstein et al., 2008; van der Gaag, 2017). Before the DSM-III, ASD was not recognized as a distinct disorder and was often referred to and diagnosed as childhood schizophrenia.

In 1943, a child psychiatrist named Leo Kanner published a groundbreaking paper describing children with an "inborn disturbance of affective contact." Kanner's study was the first to separate ASD symptomology from schizophrenia and recognize ASD as a distinct disorder (Deisinger, 2011; Neumärker, 2003; Sanders, 2009). This study's participants included 11 children (i.e., eight boys and three girls) ranging from two to eight years of age, who demonstrated atypical development. The children reportedly preferred to socially isolate since infancy, displayed repetitive behavior, insisted on sameness, and exhibited atypical verbal behavior such as echolalia and pronoun reversal (Deisinger, 2011; Kanner, 1943). The most crucial observation that differentiated ASD from schizophrenia was atypical development before symptom onset (Adler et al., 2014; Gallo, 2010; Kanner, 1943). Kanner noted crucial characteristics that have contributed to the current diagnostic criteria of ASD (Volkmar & Reichow, 2014).

In addition to behavioral observations central to ASD, Kanner identified additional comorbid characteristics, including aggression, anxiety, and deficits in adaptive functioning (Kanner, 1943). Thirty years later, a follow-up study was conducted, and results indicated

significant intellectual impairment in four of the 11 participants (Adler et al., 2014; Kanner, 1943; Volkmar & Reichow, 2014). Kanner's work was formative in defining symptoms of ASD and provided observations of comorbid conditions such as intellectual impairment and challenging behavior.

In 1944, Hans Asperger published a paper describing 'autistic psychopathy' and used 'autistic' when referring to social isolation (Klin et al., 2005; Roth et al., 2010). The study included observations of four children ages six to 11. The participants reportedly displayed deficits in social and emotional functioning, exhibited unusual interests and repetitive behaviors, but did not demonstrate impairment in cognitive functioning. While the sample group did not demonstrate deficits in language acquisition, their speech was atypical for their age (Sanders, 2009). For instance, the participants spoke extensively about a narrow range of topics and experienced difficulty reading social cues to stop talking or allow reciprocation from the listener (Adler et al., 2014; Volkmar & Lord, 1998). A male predominance and social immaturity within the sample were also noted. Like Kanner, ASD symptomology was reported to be present in early development and recognized as a distinct condition he referred to as "Autistic Personality Disorder." However, Asperger's work went unnoticed by English-speaking countries until his findings were reviewed in 1981 and were later incorporated into the DSM-IV diagnostic criteria (Adler et al., 2014; Attwood, 2007).

Research differentiating ASD symptoms continued in the 1970s. In 1971, Israel Klovin documented differences between ASD characteristics and childhood schizophrenia (Feinstein, 2011; Goldstein et al., 2008; Goodyer, 2002; Volkmar et al., 2008). In 1972, Michael Rutter notably declared the term 'childhood schizophrenia' to be inappropriate when referring to ASD and several developmental disorders. In both studies, several differentiations between ASD and childhood schizophrenia were recognized. For instance, both schizophrenia and ASD exhibited

symptoms of social withdrawal; however, ASD's onset began in infancy. Additionally, schizophrenia symptoms were reported to wax and wane, while ASD symptoms were consistent over time. Lastly, positive symptoms of hallucinations and delusions were considered markers specific to schizophrenia. This distinction between ASD and schizophrenia symptomatology was a turning point in recognizing ASD as a separate disorder (Rutter, 1972).

Parent characteristics were a common theme discussed by several formative researchers. Kanner's studies described parent characteristics as having several traits similar to ASD symptoms, such as lack of interest in others, excessive attention to detail, and lack of affection and warmth towards others (Kanner, 1943, 1954). However, in 1967, Bruno Bettelheim introduced a theory discussing "the refrigerator mother." "The refrigerator mother" concept explicitly describes "attitudes of the mother," indicating the parents wish that their child "should not exist" as a causal factor of infantile autism (Bettelheim, 1967).

Unfortunately, this theory attributed blame to mothers of children diagnosed with ASD, further alienating parents. Due to the evolution of research, the etiology of ASD has been extensively studied, resulting in substantial criticism of Bettelheim's concept, a theory now considered obsolete, but unfortunately, it has not been forgotten (Joseph, 2018; Klin et al., 2005).

Diagnostic Criteria

Contributions of seminal research have led to the recognition of autism as a distinct disorder with specific diagnostic criteria (Deisinger, 2011). Although the diagnostic criteria of ASD have changed over the years, ASD was first introduced as a distinct disorder in the American Psychiatric Association's (APA) Diagnostic and Statistical Manual of Mental Disorders, Third Edition (American Psychiatric Association, 1980). The DSM-III introduced a new set of pervasive developmental disorders (Filipek et al., 1999; Volkmar, 1991; Volkmar et al., 2008; Volkmar et al., 1988). The DSM-III included three different diagnoses: infantile autism, childhood-onset

pervasive developmental disorder (PDD), and atypical PDD. All three categorizations required significant deficits in social relationships with impairment in communication or language, with the age of onset as the differentiating factor between infantile autism and childhood-onset PDD. Infantile autism's onset required symptomology before 30 months of age, and childhood-onset PDD presented between 30 months of age and 12 years of age. The diagnosis of atypical PDD was used for presentations that did not meet the criteria for infantile autism or childhood-onset but exhibited atypical development in several domains. Additionally, diagnostic criteria specified that hallucinations or delusions must not be present if the diagnoses were assigned (Volkmar et al., 1988).

In 1987, the Diagnostic and Statistical Manual of Mental Disorders, Third Edition-Revised, included revisions to diagnostic criteria and terminology (American Psychiatric Association, 1987). This edition collapsed infantile autism, childhood-onset PDD, and atypical PDD into the two diagnoses of Autistic Disorder and Pervasive Developmental Disorder- Not Otherwise Specified (PDD-NOS). PDD-NOS criteria were similar to atypical PDD in the DSM-III. Individuals met the criteria for PDD-NOS if they exhibited impairments in development but did not meet the criteria for Autistic Disorder. However, the required age range of onset for Autistic Disorder was marked by impairments in three domains of functioning. The three domains included functional impairment in social interaction, verbal or nonverbal communication, and the presence of restricted and repetitive behavior/interests (Volkmar, 1991; Volkmar et al., 1988). A presentation during infancy or childhood was also required, recognizing the pervasiveness of the disorder through the lifespan (Deisinger, 2011; Goldstein et al., 2008; Hincha-Ownby, 2008; Volkmar et al., 1988). The DSM-III-R stated that an additional diagnosis of schizophrenia could later be assigned, but only if the individual was experiencing hallucinations and/or delusions (Volkmar, 1991).

Significant changes to diagnostic criteria occurred when the Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition (DSM-IV) was published in 1994 (American Psychiatric Association, 1994). ASD diagnostic criteria were reclassified, creating four diagnoses, including Autistic Disorder, Asperger's disorder, Rett's Disorder, Childhood Disintegrative Disorder, and PDD-NOS. The criteria for Autistic Disorder and PDD-NOS remained relatively unchanged. However, a mistake of using the word "or" instead of "and" between diagnostic criteria caused an unintended effect that allowed individuals to be diagnosed if they exhibited impairment in either social impairment, communication impairment, or unusual interests and activities (Deisinger, 2011; Feinstein, 2011; Volkmar & Klin, 2005).

The classification of Asperger's Disorder, Rett's Disorder, and Childhood Disintegrative Disorder (CDD) introduced criteria based on the severity and presentation of symptoms. Diagnostic criteria for Autistic Disorder required onset before age three and at least six presenting symptoms. Six of the presenting symptoms were required to include two of social impairment (i.e., one related to deficits in communication, and one related to behavior and interests), excluding that the symptoms are not better explained by CDD or Rett's Disorder (Deisinger, 2011; Volkmar & Klin, 2005). Asperger's Disorder was described as impairments in social interactions accompanied by restricted interests and/or repetitive, stereotyped behavior; however, this diagnosis did not include deficits in communication or language. Both Rett's disorder and CDD criteria are both marked by a presence of regression. Rett's Disorder emphasized impairments in at least two of the three areas of impairment necessary for an Autistic Disorder and a loss of psychomotor development that happens earlier (i.e., between 5 and 48 months). In contrast, CDD also emphasized impairments in at least two of the three areas of Autistic Disorder, and included regression of language, social skills, adaptive behavior, bowel, or bladder control, play skills, or motor skills after 24 months of typical development (American Psychiatric Association, 1994).

In 2000, minor diagnostic changes were made to the DSM-IV, resulting in the publication of the Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition, Text Revision (DSM-IV-TR). This edition did not include changes to diagnostic criteria. Instead, it expanded the descriptive features, which aided in clinical utility when differentiating separate diagnoses. The DSM-IV-TR clarified confusion amongst researchers and clinicians regarding the differences in onset, exclusion criteria, and presentations of the disorders, particularly Asperger's Disorder and Autistic Disorder (American Psychiatric Association, 2000).

The most significant changes to diagnostic criteria were made when The Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5; American Psychiatric Association, 2013) no longer included sub-diagnoses. Instead, one umbrella diagnostic category called autism spectrum disorder (ASD) was created. Autistic Disorder, CDD, and Asperger's Disorder were dissolved within one single ASD diagnosis. Rett's Disorder was removed and determined to be a distinct neurological disorder called Rett Syndrome, though individuals with Rett syndrome may also have ASD (Volkmar & Reichow, 2014). The three symptom domains of the DSM-IV and DSM-IV-TR (i.e., social reciprocity, communication, restricted and repetitive behaviors) were re-categorized into two symptom domains. The new domains included A) impairments in social communication/interaction and B) restricted interests and/or repetitive, stereotyped behaviors. Within this umbrella diagnostic category are three designated levels of symptom severity. Each level reflects the amount of support necessary, with Level 1 requiring support, Level 2 requiring substantial support, and Level 3 requiring very substantial support. The DSM-5 changes also included specifiers of comorbid conditions such as accompanying intellectual impairment, accompanying language impairment, and an association with a known medical or genetic condition or environmental factor (American Psychiatric Association, 2013). Asperger's and Autistic Disorder diagnoses were no longer recognized as sub-diagnoses. Almost all individuals with these

diagnoses would meet under DSM-5 ASD criteria; therefore, re-assessment was unnecessary or unrequired unless it was encouraged based on clinical judgment. However, individuals who received a diagnosis of PDD-NOS were not as easily subsumed as the criteria require fewer DSM-IV presenting symptoms. Additionally, while Asperger's was no longer a discrete sub-diagnosis of autism, individuals who received the sub-category diagnosis were encouraged to identify as the diagnosis that satisfied their preference in terminology (Hyman, 2013).

The current criteria for a diagnosis of ASD follow DSM-5 criteria. DSM-5 criteria for ASD are broken down into two main symptom domains. The first is manifested by persistent deficits in social communication and interactions across settings, which are not accounted for by developmental delays. To meet the criteria for this domain, an individual's symptoms must characterize all three subdomains, including A1) deficits in social-emotional reciprocity, A2) deficits in nonverbal communication used for social interaction, and A3) deficits in developing maintaining relationships appropriate to their developmental level. The second domain refers to B) restricted, repetitive patterns of behavior, interests, or activities. Symptoms must be demonstrated by two of the four following subdomains: B1) stereotyped and/or repetitive speech, motor movements, and/or use of objects, B2) excessive adherence to routine, ritualized patterns of verbal or nonverbal behavior, and/or excessive resistance to change, B3) highly restricted, fixated interests that are atypical in intensity or focus, and B4) atypical reactivity to sensory input or unusual interest in sensory aspects of the environment. Lastly, the criteria require a third domain, stating that symptoms must be present in early childhood (approximately eight years old or younger). Last, the fourth domain, D), requires that individuals' symptoms impair their everyday functioning (American Psychiatric Association, 2013; Hyman, 2013).

Prevalence

The diagnosis of ASD was initially perceived to occur in less than 10 in 10,000 children

and was considered a rare disorder (Howlin, 2006; Matson & Kozlowski, 2011). However, prevalence rates of ASD have risen significantly over the decades (Matson & Kozlowski, 2011). At present, one in 54 children in the United States is estimated to have ASD (Maenner et al., 2020). Due to a significant increase in prevalence, researchers have identified several possible factors contributing to this increase. Contributing factors include increased awareness of ASD, expanding diagnostic criteria, early assessment and service availability, cultural and environmental factors, and improved research techniques (Fombonne, 2009; Matson & Kozlowski, 2011).

Screening and Early Assessment

ASD can be reliably diagnosed as early as 18-24 months of age (Charman & Baird, 2002; Daniels & Mandell, 2014); however, less than half of children in the United States receive a diagnosis before age five (Mandell et al., 2002; Yeargin-Allsopp et al., 2003). Several factors, such as gender, minority status, socioeconomic status, and symptom severity, have been associated with a delay in diagnosis (Daniels & Mandell, 2014; Durkin et al., 2010; Duvekot et al., 2017; Rosenberg et al., 2011).

Unfortunately, adverse effects result from children being assessed at older ages. For instance, delayed diagnosis prevents access to early intensive behavioral intervention (EIBI) and delays skill acquisition during a crucial developmental period (Zwaigenbaum et al., 2015; Zwaigenbaum & Penner, 2018). Therefore, leading to the hindrance of skill development and outcomes (Daniels & Mandell, 2014; Reichow, 2012; Zwaigenbaum & Penner, 2018). Adverse effects can also include a lack of or delayed understanding of the disorder and fewer familial supports. These effects often leave parents and caregivers feeling stressed and lacking quality support (Baron-Cohen & Bolton, 1993; Shattuck et al., 2009; Zuckerman et al., 2015). Therefore, early intervention programs have been urged to implement routine ASD screenings to aid early identification (Twyman et al., 2009). The implementation of regular screenings has been

associated with an earlier age of diagnosis, an effect that has been found across several demographic factors (Begeer et al., 2009; Fountain et al., 2011; Miller et al., 2011).

Challenging Behavior in ASD

While the definition can vary across studies (Darrow et al., 2011), challenging behaviors are typically defined as socially inappropriate behaviors negatively impacting education and have the potential to be dangerous depending on the intensity, frequency, and duration of the behavior(s) (Emerson, 2001; Jang et al., 2011; Matson, Mahan, et al., 2010). Challenging behaviors common in ASD include tantrums, aggression, property destruction, stereotypy, and self-injury (Horner et al., 2002; Jang et al., 2011; Matson, Wilkins, et al., 2009). Although challenging behavior occurs in both typically developing and other developmental disability populations, children with ASD are particularly at risk for developing challenging behaviors (Jang et al., 2011; McClintock et al., 2003). As mentioned previously, ASD is characterized by deficits in expressive communication; maladaptive, or challenging behaviors are likely used in replacement for the individual's difficulty expressing their needs and wants (Belva et al., 2013; Durand & Carr, 1991).

It was initially estimated that 13-30% of children with ASD exhibit challenging behaviors (Horner et al., 2002); however, further research suggests that up to 94.3% of children with ASD exhibit at least one type of challenging behavior (Matson et al., 2008). With current research demonstrating estimates of prevalence rates range from 35.8% to 94.3%, most studies identify at least 50% of individuals with ASD to engage in one or more challenging behavior (Baghdadli et al., 2003; Bodfish et al., 2000; Holden & Gitlesen, 2006; O. Murphy et al., 2009). The rate at which individuals engage in challenging behaviors is influenced by additional factors, including ASD symptom severity and the severity level of a comorbid ID diagnosis. A comorbid diagnosis of ID has been found to increase the risk of a child engaging in challenging behaviors (Holden & Gitlesen, 2006; Jang et al., 2011; McClintock et al., 2003; Myrbakk & von Tetzchner, 2008). Additionally, approximately one-third of participants who engage in challenging behaviors have comorbid diagnoses of both ASD and intellectual disability (Myrbakk & von Tetzchner, 2008).

Existing literature on challenging behaviors in ASD has primarily focused on children, adolescents, or adult age groups (Baghdadli et al., 2003; J. C. Fodstad et al., 2012; Holden & Gitlesen, 2006; Matson, Wilkins, et al., 2009). Therefore, research focusing on challenging behaviors in young children (i.e., infants and toddlers) with ASD is limited. While there are few prevalence rates identified exclusively within this age group, existing data suggest similar findings to older groups that challenging behaviors of young children with ASD exist at a rate above and beyond other typically developing or developmental delay populations (Cunningham & Schreibman, 2008; Dominick et al., 2007; MacDonald et al., 2007).

One of the first studies to examine the early emergence and trend of severe challenging behavior in infants and toddlers (i.e., ages 12 to 39 months of age) with ASD compared to other general delays identified infants and toddlers with ASD to have more severe behaviors than toddlers with other developmental delays. Additionally, younger children (ASD and non-ASD delays) appeared to engage in less severe challenging behaviors. In contrast, the severity of challenging behaviors tended to increase across age groups, particularly for aggressive and destructive behaviors. Therefore, challenging behaviors can emerge early in life, typically within the first 12 months, and can continue to progress as age increases, with problematic levels beginning at 25–39 months of age (Fodstad et al., 2012).

Several adverse effects of challenging behavior have been identified. For instance, challenging behaviors have been found to interfere with an individual's functioning (i.e., social, occupational, or academic) and usually require behavioral intervention. Although early behavioral interventions can reduce challenging behaviors by 80–90 % (Horner et al., 2002), several negative consequences can occur if the behaviors are left untreated (Matson & Kozlowski, 2011). Bodily injury to the individual or others (Emerson, 2001; Holden & Gitlesen, 2006), interference with learning new skills or adaptive behavior, negatively impacted or limited social relationships

(Matson, Mahan, et al., 2010; Matson & Wilkins, 2008; Myrbakk & von Tetzchner, 2008), poor academic performance, and overall lower quality of life has been associated with delayed or untreated challenging behavior (Herzinger & Campbell, 2007). Additionally, poor outcomes are also associated with individuals who engage in challenging behaviors. They are more likely to be placed in a residential care facility (Mansell et al., 2002) and be prescribed psychotropic medications for behavior management (Holden & Gitlesen, 2006).

Particularly in young children with ASD, interference with learning can be attributed to challenging behavior disrupting the progress of evidence-based treatment programs (e.g., EIBI; Jang et al., 2011). Notably, the frequency and intensity of a young child's challenging behavior(s) may account for some variance identified in the individual outcomes of those receiving EIBI, as children with the most deficits exhibit a higher frequency and severity of challenging behaviors. Consequently, more time is needed to address challenging behaviors; thus, taking time away from time allocated to skill acquisition during a critical developmental period (Jang et al., 2011). Therefore, there is a need to prioritize a better understanding of factors contributing to challenging behavior at young ages. Improved understanding can help researchers and practitioners develop more efficacious interventions for challenging behaviors, particularly in young children, and limit interference with crucial skill acquisition and positive outcomes.

Self-Injurious Behavior

Self-Injurious Behavior (SIB) is the most examined challenging behavior in ASD (Minshawi et al., 2014). SIB presents as various topographies, etiologies and can range in severity (Rojahn et al., 2007). SIB is typically defined as a class of behaviors in which an individual inflicts upon themselves and has the potential to result in a physical injury (Iwata et al., 1994; Minshawi et al., 2014). The most frequent SIBs include head banging, biting, scratching, or hair pulling (Baghdadli et al., 2003; Schroeder et al., 1980). More specifically, SIBs that occur the most in

children with ASD are self-biting, self-scratching, skin picking or pinching, self-punching, and headbanging (Minshawi et al., 2014; Rojahn et al., 2007).

Although Children can exhibit SIBs without and with developmental disabilities, the rate at which SIB occurs in individuals with ASD is significantly higher than the general population (Dominick et al., 2007). Individuals with ASD are six times more likely to engage in self-injury than those without ASD (McClintock et al., 2003), with an estimated prevalence of engaging in SIB from 33% to 71% (Baghdadli et al., 2003; Cooper, 2009; Dominick et al., 2007; O. Murphy et al., 2009; Oliver et al., 1987; Richards et al., 2012; Shattuck et al., 2009). However, more recent literature has indicated that over fifty percent of individuals with ASD have at least one occurrence of SIB in their lifetime (Minshawi et al., 2014). Higher rates of SIB are potentially more common in ASD with comorbid ID than in any other population (Matson & LoVullo, 2008)

Self-injurious behavior can range in severity, from 'mild,' indicating no bodily injury, to 'severe,' indicating functional or life-threatening bodily injury. Therefore, SIBs are maladaptive behaviors that can present significant harm to the individual themselves, their family members, and/or practitioners (Baghdadli et al., 2003; Matson & LoVullo, 2008; Rojahn et al., 2007). Additional consequences include limited educational and vocational opportunities, social isolation, limited access to community-based activities, increased medical and residential care costs, and risky restrictive treatment practices (e.g., protective equipment, physical holds; (Iwata et al., 1994; Minshawi et al., 2014).

Risk factors most associated with SIB in children with ASD include ASD severity, the presence of comorbid ID or other psychopathology, lower age, delays in daily living skills, and comorbid medical conditions (i.e., epilepsy, perinatal conditions; Ando & Yoshimura, 1979; Baghdadli et al., 2003; Minshawi et al., 2014; Saloviita, 2000; Schroeder et al., 1980). As SIB also occurs in individuals with ID and other psychopathology, the risk of children and adults with ASD

engaging in SIB increases when comorbid conditions are present; this relationship is also influenced by the presence and severity of these comorbid conditions (McTiernan et al., 2011; Murphy et al., 2009). Studies have identified speech delay as a high-risk factor for SIB in ASD. Researchers have hypothesized that SIB occurs more frequently in young children with ASD and decreases with age as adaptive and expressive communication skills increase over time (Ando & Yoshimura, 1979; Baghdadli et al., 2003; Saloviita, 2000).

While onset can occur within the first year of life, older children, or adults with ASD typically exhibit SIB that is more intense or severe than in younger children. Therefore, literature regarding the presence of SIB in young children is overshadowed by studies including participants of older age (Matson & LoVullo, 2008), resulting in limited research regarding the prevalence of SIBs, risk factors, outcomes, and treatments in young children with ASD. However, a study examining risk factors of SIB in 222 young children (i.e., 2 to 7 years of age) with ASD found at least half of the participants to display SIB (Baghdadli et al., 2003), similar to previous research (Chadwick et al., 2000; Matson et al., 2008; Poustka & Lisch, 1993). The following characteristics were also identified as high-risk factors for SIB in younger children with ASD and corroborated previous research: lower age, delays in daily living skills, and a higher degree of ASD symptomology and comorbid medical conditions (i.e.., epilepsy, perinatal conditions; Ando & Yoshimura, 1979; Saloviita, 2000; Schroeder et al., 1980).

Although there was not a statistically significant trend, another study of infants and toddlers with ASD demonstrated an increase in challenging behavior around 26 to 32 months of age (Fodstad et al., 2012). Behavioral intervention strategies commonly used to treat SIB include functional behavior assessments, antecedent manipulations, and a combination of reinforcement, extinction, and punishment procedures (Minshawi et al., 2014). In more severe cases, pharmacotherapy is also used (Davies et al., 1998; Falcomata et al., 2007; Matson & LoVullo,

2008). If left untreated, SIB is more likely to persist into adulthood and continue to increase in severity as the child matures physically (Matson et al., 2008; G. H. Murphy et al., 2005). Therefore, it is imperative to intervene at an early age, and additional research regarding factors that contribute to the presence of SIB in young children with ASD is needed.

Aggressive/Disruptive Behavior

The presence of physical aggression and destructive behavior has been associated with adverse outcomes not only in the general population (Card & Little, 2006; Lochman & Wayland, 1994; Zahn-Waxler et al., 2005) but within individuals with ASD and other developmental disabilities (Lecavalier et al., 2006; McIntyre et al., 2002; Richards et al., 2012; Tomanik et al., 2004). In particular, the presence of aggressive behavior is the primary cause of residential placement for individuals with ASD. The consequences of aggressive behavior exhibited by individuals with ASD include interference with learning (e.g., classroom removal), exclusion from services, and fewer opportunities for independence and interpersonal relationships (Benson & Aman, 1999; Farmer & Aman, 2011; Matson et al., 2008). Aggression is associated with adverse outcomes for children with ASD and their caregivers, including decreased quality of life, increased stress levels, and reduced educational and social support (Fitzpatrick et al., 2016).

Although there have been high reports of aggressive and destructive behaviors demonstrated by individuals with ASD, and their clinical influence is unquestionable, research evaluating the prevalence of aggressive and destructive behavior has yielded inconsistent results. For instance, the prevalence of aggressive and destructive behavior in individuals with ASD has ranged from 15% to 70% among studies (Farmer & Aman, 2011; Lecavalier, 2006; Matson & Rivet, 2008; Totsika et al., 2011). Discrepancies in prevalence rates have been attributed to differences in measurement, population characteristics, and the length of time to study behaviors. Studies with a larger sample size and robust methodology are needed to better establish the

prevalence of aggression and ASD severity, particularly across age groups (Mazurek et al., 2013). However, a more recent study examining prevalence rates demonstrated a considerably higher presence of aggressive and destructive behavior among a well-characterized sample of 1380 children with ASD. Parents reported 56% percent of the sample to demonstrate some degree of current physical aggression toward caregivers and 32% toward non-caregivers. Additionally, 68% reported a history of physical aggression toward caregivers and 49% toward non-caregivers (Kanne & Mazurek, 2010). A similar yet study evaluating the prevalence of aggression in ASD yielded consistent results with a prevalence rate of 53.7% of 1584 children and adolescents (i.e., ages 2-17 years) with ASD demonstrating aggressive behavior (Mazurek et al., 2013). The results further supported previous findings that aggression is more prevalent among children with ASD than in the general population.

Similar to trends identified in the general population, aggression decreased as participants age increased, and the prevalence of aggression in the current study was highest among young children with ASD (NICHD Early Child Care Research Network, 2004; Tremblay et al., 2004) with 54.6% prevalence within the 2–4-year-old group. It is also important to note that while aggression decreased as the age group increased, 46.9 % of the 14 to 17-year-old age group still exhibited aggressive behavior (Mazurek et al., 2013), which is significantly different compared to the chronic aggressive behavior displayed by 5% of typically developing adolescents (Tremblay, 2010). Lastly, a study of children (i.e., ages 12 months to 21 years) reported approximately 50% of the ASD group to have engaged in at least one physically aggressive behavior; findings that were corroborated by the previously mentioned studies (Kanne & Mazurek, 2010; Mazurek et al., 2013).

Several risk factors of aggressive behavior in typically developing children have been identified (e.g., younger age, low IQ, lower verbal abilities, socioeconomic status, and harsh

parenting practices; Dionne et al., 2003; Nagin & Tremblay, 2001; NICHD Early Child Care Research Network, 2004; Tremblay et al., 2004). However, data regarding risk factors of aggression in children with ASD is limited, particularly in younger age samples.

Only a few factors within the general population have been associated with an increased risk of aggression in children and adolescents with ASD (Kanne & Mazurek, 2010). Of the limited research available, data suggest adaptive behavior, cognitive functioning, and language ability are inversely related to aggressive or destructive behavior in children with ASD (Dominick et al., 2007; Estes et al., 2007; Hartley et al., 2008; Lecavalier, 2006). Higher incidences of aggressive and destructive behavior have been associated with ASD, particularly with a comorbid diagnosis of ID (Ando & Yoshimura, 1979; Mazurek et al., 2013; McClintock et al., 2003). Additionally, ASD symptom severity, the presence of SIB, ritualistic behavior, and resistance to change were predictors of physical aggression in children and adolescents with ASD (Kanne & Mazurek, 2010). Sensory problems have also been significantly associated with aggression (Mazurek et al., 2013). Therefore, more symptoms of ASD may be related to an increased incidence of aggression, and the ASD characteristic factors cause researchers to infer those difficulties specific to the symptomology of ASD may influence aggression (Dominick et al., 2007; Kanne & Mazurek, 2011; Mazurek et al., 2013).

Additionally, aggression levels have not previously correlated with age (Dominick et al., 2007; Farmer & Aman, 2011; Lecavalier, 2006); however, two studies demonstrated a decrease in physical aggression as the age of children with ASD increased (Kanne & Mazurek, 2011; Mazurek et al., 2013). In young children with ASD (i.e., infants and toddlers), aggressive/disruptive behavior significantly increased around 26–32 months of age (Fodstad et al., 2012). Prior studies of aggression among individuals with ID have also found a predominance of aggression in males (Archer, 2004; Card et al., 2008; McClintock et al., 2003). However, larger, and more recent

studies did not demonstrate sex differences of aggression in the ASD population (Kanne & Mazurek, 2011; Mazurek et al., 2013). Lastly, aggression has been linked to the presence of SIB. Aggression and SIB have been significantly associated among individuals with severe ID (Matson et al., 2008). SIB is a significant precursor of later aggression among individuals with ID (Nøttestad & Linaker, 2002). Although this relationship suggests replication in ASD is possible, studies have yet to investigate the relationship in children with ASD.

Although the literature is inconsistent regarding prevalence rates, the presence of aggression and destructive behaviors found in ASD is substantial. Variables such as assessment measures, operational definitions, and sample design have made aggression prevalence rates in ASD challenging to study. However, these factors are crucial in assessing correlates and responding with effective treatment (Connor et al., 1998). Additionally, multiple domains of functioning are potential risk factors for aggression in ASD; therefore, it is essential to consider these variables when assessing and treating aggression among children with ASD (Mazurek et al., 2013). Treatment strategies for aggressive behavior often include therapeutic behavior interventions, including functional behavioral assessment, reinforcement strategies, and functional communication training; however, psychotropic medication and institutionalization are used (Fitzpatrick et al., 2016; Tsakanikos et al., 2007).

Stereotypy

Stereotypic behavior is commonly defined as repetitive motor responses and/or vocal responses that do not serve an adaptive function (LaGrow & Repp, 1984; Matson et al., 1997; Smith & Van Houten, 1996). Although stereotypic behaviors occur in individuals with ID and other developmental disabilities, stereotypy has been found to occur at a higher intensity and frequency in individuals with ASD (Bodfish et al., 2000; MacDonald et al., 2007). Interestingly, children carrying a diagnosis of ASD, with or without a comorbid ID, demonstrate stereotypy more

frequently than individuals with only ID (Carcani-Rathwell et al., 2006; Matson & Dempsey, 2008). Also, children with an ASD exhibit more motor stereotypy than children with only atypical development (Goldman et al., 2009). Therefore, the extant literature confirms stereotypy as a core feature of ASD (Lewis & Bodfish, 1998; MacDonald et al., 2007).

Stereotypic behaviors can be challenging to manage and occur over the lifespan (Matson & Dempsey, 2008; Matson, Dempsey, et al., 2009). Examples of motor stereotypy include but are not limited to hand flapping, body rocking, spinning objects, abnormal positioning of body parts, and restricted play/leisure behavior. Vocal stereotypy usually consists of the following: echolalia, non-functional sounds, laughing or giggling out of context, and repetition of words or phrases (MacDonald et al., 2007). Risk factors associated with stereotypic behavior in ASD include ASD symptom severity (Bodfish et al., 2000), lower adaptive functioning (Matson et al., 1997), and low IQ (Lewis & Bodfish, 1998; MacDonald et al., 2007; Rojahn et al., 2007).

Stereotypic behavior can be identified at an early age (Fodstad et al., 2012; Matson, Wilkins, et al., 2009). Furthermore, a study examining the challenging behavior of infants and toddlers with ASD found stereotypic behavior to significantly increase at about 26–32 months of age (Fodstad et al., 2012). Also, an age-matched sample of toddlers and children (ages 2, 3, and 4 years) with ASD and typical development showed those with ASD to demonstrate higher levels of stereotypy. This difference significantly increased by ages 3 and 4. Indicating that while challenging stereotypic behavior may have an early onset, the severity and intensity increase with age (MacDonald et al., 2007; Matson et al., 1997).

Several negative consequences can result from stereotypic behavior, including interference with the skill acquisition (Dunlap et al., 1983; Koegel & Covert, 1972; Morrison & Rosales-Ruiz, 1997), impaired social interactions (Wolery et al., 1985), and the risk of stigmatization from peers (Jones et al., 1990). These impairments in overall function have led stereotypy to be considered a

challenging behavior and have been the center of many behavioral interventions (MacDonald et al., 2007). Additionally, stereotypies put the child and individuals with ASD at risk for invasive treatments such as psychotropic drugs (Matson, Dempsey, et al., 2009; Matson & Dempsey, 2008). These behaviors can increase in frequency and severity without effective intervention, persisting into adulthood (Eikeseth, 2009; Hartley et al., 2008). EIBI routinely targets stereotypy for treatment (Eikeseth, 2009; Matson, 2007; Matson, Dempsey, et al., 2009; Matson & Smith, 2008). Therefore, improved understanding of these behaviors, particularly in young children, helps inform treatments and improve overall outcomes. Challenging behaviors such as aggressive/disruptive behavior, SIB, and stereotypy are particularly common in individuals with ASD (Machalicek et al., 2007; Matson & Nebel-Schwalm, 2007; Smith & Matson, 2010). Although challenging behaviors are not distinct characteristics of ASD, these aberrant behaviors often occur in ASD at higher rates than typically developing populations and other developmental disabilities (Holden & Gitlesen, 2006; Murphy et al., 2005; Nicholas et al., 2008). While existing literature supports those challenging behaviors occurring in individuals with ASD, research has primarily focused on children and adolescents. Of the limited studies that have evaluated young children (i.e., infants and toddlers) with ASD who demonstrate challenging behaviors, the relationship between these specific challenging behaviors in young children with ASD and their differential impact on parent stress has not been studied.

Parent Stress in ASD

Parent Stress of Young Children with ASD

The most studied familial factor of ASD is parent stress (Davis & Carter, 2008; Hayes & Watson, 2013; Pisula, 2003). The study of parent stress is a framework for identifying key contributing factors that may affect the overall experience of stress. Particularly for families of children with ASD who experience the highest levels of parenting stress, the study of parent stress improves understanding of contributing factors. The study of parent stress can improve interventions targeting family support, overall family well-being, and individual improvement (Davis & Carter, 2008). Parent stress is defined as the 'experience of distress or discomfort that results from demands associated with the role of parenting' (Deater-Deckard, 1998; Hayes & Watson, 2013). However, this definition may be an oversimplification (Webster-Stratton, 1990), as distress or discomfort can result from several environmental, parent, and child factors.

Parents of children with ASD exhibit significantly more stress than parents of typically developing children or children with other developmental disabilities (e.g., down syndrome, cerebral palsy, intellectual disability; Lai et al., 2015) and demonstrate poorer psychological outcomes. For instance, significantly more depression and anxiety symptoms (Davis & Carter, 2008; Hayes & Watson, 2013; Lai et al., 2015), less overall well-being (Blacher & McIntyre, 2006), and more overall life and daily stress have been found in parents of children with ASD compared to parents of typically developing groups (Davis & Carter, 2008; Quintero & McIntyre, 2010).

Specific factors that may contribute to significant stress levels in parents of children with ASD have been identified (Davis & Carter, 2008). The severity level of ASD (Konstantareas & Papageorgiou, 2006), adaptive skills (Hall & Graff, 2011), age (Smith et al., 2008), gender (Mandell & Salzer, 2007), and challenging behaviors (Hall & Graff, 2012; Lecavalier, 2006) are

all examples of child factors that influence parent well-being leading to parent stress (Lai et al., 2015). However, seldom research has been conducted regarding parenting stress in toddlers and newly diagnosed children with ASD. Due to an improved understanding of ASD characteristics and etiology, ASD diagnoses occur at young ages (Charman & Baird, 2002; Cox et al., 1999). To help families cope and adapt to the challenges of caring for a young child with ASD, more research on parenting stress in young children with ASD is needed.

While little research has focused on young children with ASD and the effects on parent stress, the nature of both parents' stress, the difference between parent stress of mothers versus fathers, and the relationship to their child's behavior (54 toddlers with ASD) has been explored (Davis & Carter, 2008). Previous studies have demonstrated significantly higher parent stress levels in mothers compared to fathers (Beckman, 1991; Moes et al., 1992; Sharpley et al., 1997); however, this study identified high-stress levels in both mothers and fathers of young children with ASD (Davis & Carter, 2008). Recent reports have continued to corroborate shared levels of parent stress in both mothers and fathers (Hastings et al., 2005; Rimmerman et al., 2003), with increased stress levels identified in younger mothers and fathers (Lau, Gau, Chiu, & Wu, 2014). This shared effect on stress may be due to recent changes in gender roles and primary caregiver status. Additionally, when the stress of mothers and fathers were examined independently, regulatory problems and externalizing behaviors were significantly correlated with stress (Davis & Carter, 2008).

Parent Stress and Challenging Behaviors

Links between parent stress and challenging behaviors of children with ASD have been well documented in the literature (Baker et al., 2002; Davis & Carter, 2008; Lecavalier, 2006). Challenging behaviors are the most consistent child-related source of parental distress (Baker et al., 2002; Beck et al., 2004; Estes et al., 2009). Challenging behaviors are highest among children

diagnosed with ASD than other developmental disabilities or typically developing children and contribute to the higher stress levels in parents of children with ASD (Brereton et al., 2006; Estes et al., 2009; Singh et al., 2014). Parents of children with ASD who demonstrate challenging behaviors experience higher stress levels than children with ASD who exhibit any other comorbid characteristics. Moreover, these higher parent stress levels significantly increase over time without effective treatment targeting these behaviors (Lecavalier, 2006).

While parents of children with ASD typically have higher levels of parenting stress, challenging behaviors in children with ASD characteristics have a unique and additive effect on this relationship (Kasari & Sigman, 1997; Wolf et al., 1989). For instance, deficits in social communication (Bebko et al., 1987; Davis & Carter, 2008) and restricted interests or repetitive behaviors (Gabriels et al., 2008) are two factors identified to be particularly stressful for parents, and the severity of the symptoms is positively related to higher levels of parenting stress (Brobst et al., 2009; Ekas & Whitman, 2010; A. Estes et al., 2009; Lecavalier, 2006). Notably, core symptoms of ASD have been associated with higher levels of parenting stress independent of the child's level of intellectual functioning (Davis & Carter, 2008).

The core symptoms of ASD are strongly tied to parents' level of stress. However, challenging behaviors within these children often cause even more distress to caregivers than the core ASD symptoms themselves (Hastings et al., 2005; Lecavalier, 2006), demonstrating an additive effect to the overall relationship. For example, when other stress-related factors are controlled for, challenging aggressive behavior of children with ASD is independently associated with parent stress (Gupta & Singhal, 2005). Additionally, challenging behaviors in children with ASD are significantly related to increased maternal parenting stress and psychological distress for both mothers of children with ASD and children with developmental delay without ASD (Estes et al., 2009). Other studies have highlighted externalizing behaviors' association with increased

parental stress for both mothers and fathers independently (Davis & Carter, 2008). Due to the cross-sectional nature of previous literature, a causal relationship between challenging behaviors and parent stress cannot be assumed. However, the idea that challenging behaviors in children influence caregivers' stress levels is consistent with a transactional stress model. This is a model in which child problem behavior is the driving factor of parenting stress; therefore, disrupting parenting behavior and increasing existing challenging behaviors (Estes et al., 2009; Hastings & Brown, 2002). Several studies have further demonstrated this model by evidence that supports a bi-directional relationship between parenting stress and children with ASD who exhibit challenging behavior (Lecavalier, 2006; McGrath, 2013; Neece et al., 2012), and reinforce a cycle of parent stress and challenging behavior (Singh et al., 2014). While researchers have explored the effects of the presence and severity of challenging behavior on parent stress, research has yet to further identify differential effects of specific challenging behaviors on parents' stress, particularly of young children.

Purpose

Parents of children with ASD exhibit significantly more stress than parents of typically developing children or children with other developmental disabilities (Lai et al., 2015). As literature regarding the etiology and presentation of ASD advances, children are diagnosed at younger ages (Charman & Baird, 2002; Cox et al., 1999). During this critical diagnostic period, increased stress levels occur when parents adapt to a new diagnosis and cope with the effects of untreated ASD symptoms. Unfortunately, seldom has research been conducted on parenting stress in toddlers and newly diagnosed children with ASD. More research is needed on factors that influence parent stress to help families cope and adapt to caring for a young child with ASD.

Challenging behaviors have been the most consistent child-related source of parental distress (Baker et al., 2002; Beck et al., 2004; Estes et al., 2009). While challenging behaviors are not a core feature of ASD, these behaviors occur at significantly higher rates in ASD than in typically developing or other developmental disability populations (Holden & Gitlesen, 2006; Murphy et al., 2005; Nicholas et al., 2008). In turn, parents of children with ASD who engage in challenging behaviors exhibit higher stress levels than children with any other comorbid characteristics. Furthermore, when challenging behaviors occur in young children (i.e., infants and toddlers) with ASD, the behaviors can interfere with skill acquisition during critical developmental periods (e.g., EIBI; Fodstad et al., 2012; Itzchak et al., 2008; Itzchak & Zachor, 2011; Matson, 2007). Therefore, a better understanding of challenging behavior in young children with ASD is important and timely.

While researchers have explored the collective impact of challenging behaviors on parent stress, differential effects of specific challenging behaviors on parents' stress of young children with ASD have yet to be identified. Therefore, the purpose of this study was to investigate the predictive influence of specific challenging behaviors (i.e., aggressive/disruptive behavior,

stereotypy, and SIB) on parent stress in young children (i.e., infants and toddlers) with ASD. This study aimed to answer the following research questions: 1) Does parent-reported severity of challenging behaviors in young children with ASD (i.e., aggression/disruption, stereotypy, SIBs) predict parent stress? 2) Of these challenging behaviors (i.e., aggression/disruption, stereotypy, or SIB), do any uniquely predict parent stress of young children with ASD? When considering the extant literature, it was hypothesized that the *BISCUIT-Part 3* total factor scores of all challenging behaviors (i.e., SIB, aggressive/disruptive behavior, and stereotypy) would significantly predict *PSI-4-SF* Parental Distress. Researchers have yet to identify the differential effects of specific challenging behaviors on parent stress of young children with ASD. Therefore, it was hypothesized from clinical experience that the *BISCUIT-Part 3* total factor score of SIB would significantly add the most variance to the predictive model, followed by stereotypy and aggressive/disruptive behaviors. This study's findings may help inform treatment planning of early behavioral interventions when targeting challenging behaviors of newly diagnosed children with ASD.

Method

Participants

All 54 participants were referred to Louisiana State University's Psychological Services Clinic for further assessment by EarlySteps for scoring "at-risk" for ASD according to an ASD screener, Baby and Infant Screen for Children with aUtIsm Traits, Part 1 (BISCUIT- Part 1; Matson et al., 2007). EarlySteps is Louisiana's statewide early intervention program under the Individuals with Disabilities Act, Part C. Children under three years of age qualify for EarlySteps services if they have or are at risk for a developmental delay; therefore, all participants in the dataset exhibited some form of atypical development. Additionally, all participants received a formal assessment consisting of a parent/caregiver interview of the child's developmental history, administration of standardized measures, and direct observation. A licensed clinical psychologist then assigned diagnoses to each child based on the formal assessment results.

For the current study, participants had to meet the following inclusion criteria: (a) at least 17 to 37 months of age at the time of assessment, (b) have a DSM-5 diagnosis of ASD, (c) completion of the *BISCUIT-Part 3* by a parent informant and (d) completion of the *PSI-4-SF* by a parent informant. These criteria were applied to 345 young children, resulting in the final sample size of 54 parent informants of young children with ASD. The majority of young children with ASD were male (83.3%, n = 45), and ranged from 19 to 37 months of age (M = 29.943, SD = 4.829). The reported race/ethnicities of the young children were 66.7% White/Caucasian (n = 36), 20.3% Black/African American (n = 11), 3.7% Hispanic (n = 2), and 9.3% identified as another race/ethnicity (n = 5). The parents' ages ranged from 19 to 46 years (M = 27.630, SD = 5.535) and a majority of informants were mothers (94.4%, n = 51).

Measures

Baby and Infant Screen for Children with aUtIsm Traits - Part 3 (BISCUIT-Part 3)

The *BISCUIT-Part 3* is part of a larger comprehensive assessment battery, the Baby and Infant Screen for Children with aUtIsm Traits (Matson et al., 2007). The *BISCUIT* was developed to assist with identifying and measuring symptoms of ASD and comorbid conditions in young children (i.e., 17–37 months of age). The larger battery also includes a section designed to assess symptoms of ASD *(BISCUIT-Part 1*) and symptoms of comorbid psychopathology *(BISCUIT-Part 2)*. Specifically, the *BISCUIT-Part 3* aids in assessing and identifying challenging behaviors in young children with ASD or general developmental delays.

The *BISCUIT-Part 3* is an informant-based scale and is composed of 15 items. The scale measures challenging behaviors (i.e., aggressive/disruptive behavior, stereotypies, and SIB) in young children (i.e., 17-37 months of age) exhibiting ASD characteristics. The *BISCUIT-Part 3* uses a three-point Likert scale for scoring with 0 = "not a problem or impairment, not at all"; 1 = "mild problem or impairment"; and 2 = "severe problem or impairment." Example items include "poking him/herself in the eye," "unusual play with objects (e.g., twirling string, staring at a toy)," and "pulling others' hair" (Rojahn et al., 2009). Each item is scored considering the young child's present behavior(s) compared to same-age children with typical development. The informant-based measure can be administered to a parent, caregiver, or legal guardian in approximately 20 to 30 minutes (Matson et al., 2007; Rojahn et al., 2009). A factor analysis yielded three factors for *BISCUIT-Part 3*, including aggressive/disruptive behavior, stereotypy, and SIB (Matson, Wilkins, et al., 2009). Psychometric analyses of the measure indicated excellent internal reliability (α=0.91). Although no validity information is currently available on the *BISCUIT-Part 3* (Belva et al., 2013), separate severity cutoff scores have been established for young children with ASD (Rojahn et al., 2009) and non-ASD developmental delay (Matson, Mahan, et al., 2010). Therefore,

the *BISCUIT-Part 3* is an appropriate scale to measure parent-reported presence and severity of challenging behaviors in young children with ASD. Additionally, the *BISCUIT-Part 3* is the only known scale measuring challenging behaviors, particularly for young children demonstrating traits of ASD. For this study, the factor scores were used to measure parent-reported severity levels of Aggressive/Destructive behavior, Stereotypy, and SIB. A complete list of the scales' items, separated by factors, can be referenced in Table 1 (below), with descriptive statistics of the participants' challenging behaviors listed by factor and severity in Table 2 (below).

Table 1. *BISCUIT-Part 3* factors and items.

BISCUIT-Part 3 subscale factors and item numbers with description
Aggressive/ Disruptive Behavior
8. Throwing objects at others.
15. Property destruction (e.g., ripping, breaking, tearing, crushing, etc.).
12. Aggression towards others.
14. Yelling or shouting at others.
3. Kicking objects (e.g., doors, walls).
13. Pulling others' hair.
5. Removal of clothing at inappropriate times.
9. Banging on objects (e.g., doors, walls, windows) with hand.
11. Leaving the supervision of caregiver without permission (i.e., elopement).
7. Playing with own saliva.
Stereotypic Behavior
17. Repeated and unusual body movements (e.g., hand flapping, waving arms, etc.).
16. Repeated and unusual vocalizations (e.g., yelling, humming, etc.).
6. Unusual play with objects (e.g., twirling string, staring at a toy, etc.).
Self-injurious Behavior
1. Poking him/herself in the eye.
2. Harming self by hitting, pinching, scratching, etc.

Table 2. Descriptive statistics of *BISCUIT-Part 3* factors by severity and cutoff scores.

	M (SD)	n (%)	Cutoff Scores
Aggression/Destruction	6.074 (5.154)		
No/minimal impairment		40 (74.2)	0-9

(table cont'd)

	M (SD)	n (%)	Cutoff Scores
Moderate Impairment		7 (12.9)	10-13
Severe Impairment		7 (12.9)	14 and up
Stereotypy	2.611 (1.847)		
No/minimal impairment		38 (70.4)	0-3
Moderate Impairment		-	-
Severe Impairment		16 (29.6)	4 and up
Self-Injury	0.722 (1.017)		
No/minimal impairment		42 (77.8)	0-1
Moderate Impairment		10 (18.5)	2
Severe Impairment		2 (3.70)	3 and up
Total	9.407 (7.002)		
No/minimal impairment		38 (70.4)	0-12
Moderate Impairment		7 (12.9)	13-18
Severe Impairment		9 (16.7)	19 and up

The Parenting Stress Index, Fourth Edition, Short Form

Parent stress levels of children with ASD are affected by the degree to which behaviors are perceived as stressful; thus, parent stress measures are often self-reported and administered in questionnaire format (Davis & Carter, 2008). One of the most common parenting stress measures of children with ASD is the *Parenting Stress Index, Fourth Edition* (*PSI-4),* including its respective short form (Richard R. Abidin, 1983; Hayes & Watson, 2013). The *PSI-4* consists of 101 items and has a fifth grade reading level, and all forms can be administered by paper-and-pencil or computer software. The scale is used to elicit information about parent/child systems to identify stress while screening for systems that may lead to or indicate behavioral and emotional difficulties for the parent and/or the child.

The Parenting Stress Index–Short Form (*PSI-4-SF*) is an abbreviated measure of the *PSI-4*, a psychometrically and clinically grounded measure that, for over 30 years, has been reliable, valid, and continually revised (Abidin, 2012). The short form consists of 36-items and has an administration time of approximately 10 minutes. The use of the *PSI-4-SF* in samples of parents of children with ASD is practical when measuring the severity of parents' distress (Dardas &

Ahmad, 2014; Zaidman-Zait et al., 2010), and its short administration time is appealing for researchers or practitioners who have limited time with clients. Items on the *PSI-4-SF* prompt parents to report on their parent-child system, with items mapping on to one of three domains (i.e., Parental Distress, Parent-Child Dysfunctional Interaction, and Difficult Child), and a short-form Total Stress scale. Scores at or above the 90th percentile are clinically significant, indicating that the parent may require counseling (Abidin, 1995, 2012). The raw score of the *PSI-4-SF* Parental Distress domain was selected to measure parental stress in this study. The Parental Distress domain measures personal factors that directly relate to parenting by measuring the extent to which they feel competent, restricted, conflicted, supported, and/or depressed in their role as a parent (Abidin, 2012). Raw scores at or above 40 (90th. percentile) are considered clinically significant. In contrast, raw scores of 38 or 39 (86th or 88th. percentile) are considered subclinical, and raw scores of 37 (84th percentile) or below are considered typical levels of parent distress.

Additionally, normative psychometric analyses of the measure indicated excellent internal reliability (α=0.90; Abidin, 2012). The *PSI-4-SF* Total Stress score was not used to avoid the risk of a Type I error due to the potential overlapping nature of Difficult Child domain items measured on the *BISCUIT-Part 3*. A complete list of the PSI-PD domain items is referenced in Table 3 (below), and descriptive statistics regarding the current study are found in Table 4 (below).

Table 3. *PSI-4-SF* Parental Distress domain items with description.

PSI-4-SF Parental Distress
1. I often have the feeling that I cannot handle things very well.
2. I find myself giving up more of my life to meet my children's needs than I ever expected,
3. I feel trapped by responsibilities as a parent.
4. Since having a child, I have been unable to do new and different things.
5. Since having a child, I feel that I am almost never able to do the things that I like to do.
6. I am unhappy with the last purchase of clothing I made for myself.

(table cont'd)

PSI-4-SF Parental Distress
7. There are quite a few things that bother me about my life.
8. Having a child has caused more problems than I expected in my relationship with my spouse/parenting partner.
9. I feel alone and without friends.
10. When I got to a party, I usually expect not to enjoy myself.
11. I am not as interested in people as I used to be.
12. I don't enjoy things as I used to.

Table 4. Descriptive statistics of *PSI-4-SF* Parental Distress domain with cutoff scores.

PSI-4-SF Parent Distress	M (SD)	n (%)	Cutoff Scores (%ile)
Total	25.982 (9.706)	-	-
Clinical	-	6 (11.1)	≥ 40 (90th)
Subclinical	-	3(5.6)	38 – 39 (86th to 88th)
Non-Clinical	-	45 (83.3)	≤ 37 (84th and below)

Procedures

The present study's data was used from an existing de-identified dataset part of a larger ongoing study. The Louisiana State University institutional review board approved the study prior to data collection. Written informed consent was collected from the parent of each child. All participants received a comprehensive battery of assessments, which included the *BISCUIT-Part 3* and *PSI-4-SF*. These measures were administered by trained graduate students or completed independently by the parent informant with a trained administrator available to answer questions. Prior to performing analyses, personal identifiers were removed from the database.

Statistical Analyses

A standard multiple linear regression was conducted to determine the predictive influence of specific challenging behaviors (i.e., SIB, aggressive/disruptive behavior, and stereotypy) on parent stress of young children with ASD. All statistical analyses were conducted using SPSS 27.0 statistical software. As this study aimed to identify the predictive influence of challenging behaviors as a whole on parent stress and the unique contribution of each challenging behavior,

the standard multiple regression was determined to be the most appropriate analysis. A standard multiple regression is one of the three major strategies of multiple regression. The primary distinctions of a standard multiple regression include independent variables (IVs) that are simultaneously entered into the regression, with each IV assigned only the area of its unique contribution to the model (Tabachnick & Fidell, 2013).

The Parental Distress domain score obtained from the *PSI-4-SF* was selected as the continuous dependent variable. The total severity score of the following *BISCUIT-Part 3* factors including, 1) aggressive/disruptive behavior, 2) stereotypy, and 3) SIB, were selected as the predictor independent variables. It should be noted that the *BISCUIT-Part 3* factor scores are a summation of each item's Likert-type scale rating (i.e., "0 - not different; no problem," "1 - somewhat different; mild impairment," or "2 - very different; severe impairment"). Although this Likert-type scale rating is recognized as ordinal, the independent variables were treated as continuous for the standard multiple linear regression (Hair et al., 2014; Tabachnick & Fidell, 2013).

To determine a sample size with sufficient power, a priori power analysis was conducted in G*Power (Faul et al., 2009). A power of 0.80, a medium effect size of 0.25, and an alpha of 0.05 were selected to determine the necessary sample size to yield statistically sound results. According to Cohen (1988), a power of 0.80 was selected as an adequate means to detect an existing effect. The medium effect size of 0.25 was selected to be in the range and consistent with prior research examining parent stress and challenging behavior in children with ASD (Davis & Carter, 2008; Lecavalier, 2006; Tomanik et al., 2004). The a priori power analysis for a standard multiple regression of 3 predictor variables indicated an adequate total sample size to be at least 48 participants, confirming the final sample size of 54 primary caregivers of young children diagnosed with ASD to be satisfactory for further analysis.

Bivariate analyses revealed SIB factor severity to have a significant positive correlation (r = 0.309) with Parental Distress scores. Surprisingly, aggressive/disruptive behavior was not significantly correlated with Parental Distress, with an almost negligible correlation (r = 0.082, $p > 0.05$). However, the factor of stereotypy demonstrated a more nearly acceptable yet nonsignificant correlation with Parent Distress (r = .237, $p > 0.05$).

Regarding multiple regression model building, the independent variables of stereotypy and aggressive/disruptive behavior were included in the standard multiple regression model, regardless of their nonsignificant bivariate correlations with Parent Distress. This decision was made with consideration to both data-driven and theory-driven model building techniques (Beckstead, 2012; Friedman & Wall, 2005). It is important to note that challenging behaviors such as aggressive/destructive behaviors, SIB, and stereotypy often co-occur within the ASD population (Hong et al., 2018; Jang et al., 2011; Horner, 2002), a finding that has been replicated in the current study's sample (see Table 5 below).

While inclusion of nonsignificant bivariate correlations potentially complicates the interpretation of the results and increases statistical noise in the model, exclusion of the challenging behaviors factors could have led to the loss of valuable real-life variance due to the cooccurring nature of challenging behaviors in ASD (Beckstead, 2012; Friedman & Wall, 2005). Therefore, the decision to include all three factors was made to avoid the loss of real-life variance. Furthermore, existing studies on the influence of challenging behaviors on parent stress in ASD have measured and analyzed challenging behaviors in their totality or as a sum total of externalizing behavior, rather than examining the differential effects of each behavior (Davis & Carter, 2008; Hastings et al., 2005; Lecavalier, 2006). For this reason, the inclusion of all three factors was the most appropriate response to replicate existing research while also examining the deferential influence each behavior contributes to the relationship.

Table 5. Summary of Pearson Correlations between *BISCUIT-Part 3* Challenging Behavior factors and *PSI-4-SF* Parent Distress Raw Scores.

	1	2	3	4
1. PSI-PD	—			
2. AGG	0.082	—		
3. STR	0.237	0.501**	—	
4. SIB	0.309*	0.670**	0.393**	—

**. Correlation is significant at the 0.01 level (2-tailed).
*. Correlation is significant at the 0.05 level (2-tailed).

Preliminary analyses were run to ensure all assumptions of the data's characteristics were met before proceeding with the parametric test of standard multiple regression and subsequently interpreting statistical results. First, the assumption of linearity between the dependent and independent variables was met collectively and individually by assessing partial regression plots and a plot of studentized residuals against the predicted values; the use of total fitted lines aided in the interpretations. Independence of errors was also present with evidence of a Durbin-Watson statistic of 2.047, indicating no correlation between residuals (Field, 2009). The assumption of homoscedasticity was also met and determined by visual inspection of a plot of studentized residuals versus unstandardized predicted values; a fitted line was also used to aid in the interpretation. No evidence of multicollinearity was present, as all independent variable correlations were below 0.70 (see Table 1), and collaterality statistical tolerance values were greater than 0.1 (Hair et al., 2014). Furthermore, no studentized deleted residuals were greater than ±3 standard deviations, indicating no existing outliers were present. Additionally, leverage values were no greater than 0.2, indicating all values as safe to interpret (Huber, 1981). Evidence of influential points was not present, indicated by the measure of Cook's Distance influences values all above 1.0 (Cook and Weisberg, 1982). Lastly, the assumption of normality was met by examining a Q-Q Plot of the studentized residuals, which resulted in the residuals appearing close to normal for the analysis to proceed and indicated no data transformations were necessary.

Results

A standard multiple regression was run to predict PSI-4-SF Parental Distress domain scores from the BISCUIT-Part 3 severity factors of aggressive/disruptive behavior, self-injurious behavior, and stereotypy in young children with ASD. The overall multiple regression model significantly predicted PSI-4-SF Parental Distress domain scores, R2 = .112, F (3, 50) = 3.232, *p* = .030. Interestingly, SIB added statistical significance to the prediction, p = .016. Surprisingly, the other independent variables of aggressive/disruptive behavior, *p* = .087, and stereotypy, *p* = .135, did not add statistical significance to the standard multiple regression model. Regression coefficients and standard errors for these analyses can be found in Table 7 (below), listed as Model 1.

Considering SIB was the only significant predictive factor of Parental Distress, a follow-up simple linear regression was run to understand the effect of SIB on PSI-4-SF Parental Distress scores. The goals of these secondary analyses were to reveal the level of predictive validity, explain variance potentially lost from Model 1, and demonstrate the deferential predictive influence of SIB on Parental Distress when statistically isolated from other challenging behaviors. All assumptions of the data's characteristics were met before proceeding with the parametric linear regression and statistical interpretations. For instance, linearity was met by visual inspection of a scatterplot with a regression line, showing Parental Distress scores against SIB severity. Independence of errors was also present with evidence of a Durbin-Watson statistic of 2.218, indicating no correlation between residuals (Field, 2009). Visual inspection of these two plots indicated a linear relationship between the variables. Lastly, homoscedasticity and normality of the residuals were also present, and no outliers were identified. Following the additional preliminary and linear regression analyses, the results again established the statistically predictive nature of SIB severity on PSI-4-SF Parental Distress scores of young children with ASD, F (1, 52) = 5.492, p = .023. However, the

secondary analyses resulted in a lower adjusted R2 = 0.78, as SIB accounted for 7.8% of the variation in Parental Distress scores, which is also considered a small effect size according to Cohen (1988). Regression coefficients and standard errors for these analytical findings can be found in Table 6 (below), listed as Model 2.

Table 6. Regressions of parent-reported severity of challenging behaviors from *BISCUIT-Part 3* factors and Parental Distress domain of *PSI-4-SF*.

PSI-PD	*B*	95% CI for *B*		*SE B*	β	α	R^2	ΔR^2	f^2
		LL	*UL*						
Model 1							.162	.112**	.193
Constant	23.554**	18.961	28.147	2.287		<.001**			
AGG	-.611	-1.314	.092	.350	-.324	.087			
SIB	4.167*	.813	7.522	1.670	.437*	.016*			
STR	1.198	-.386	2.783	.789	.228	.135			
Model 2							.096	.078**	.106
Constant	23.851**	20.721	26.982			<.001**			
SIB	2.949*	.424	5.475	1.259	.309*	.023*			

Note. Model = "Enter" method is SPSS Statistics; *PSI-PD* = Parental Distress domain score of *PSI-4-SF*; AGG = aggressive/destructive behavior factor severity score of the *BISCUIT-Part 3*; SIB = self-injurious behavior factor severity score of the BISCUIT-Part 3; STR = stereotypy factor severity score of the *BISCUIT-Part 3*; *B* = unstandardized regression coefficient; CI = confidence interval; *LL* = lower limit; *UL* = upper limit; *SE B* = standard error of the coefficient; β =standardized coefficient; R^2 = coefficient of determination; ΔR^2 = adjusted R^2.
*$p < .05$. ** $p < .01$.

Discussion

To gain a better understanding of the predictive relationship that specific challenging behaviors have on parent stress of young children with ASD, this study examined the predictive influence of common challenging behaviors (i.e., aggressive/disruptive behavior, stereotypy, and SIB) on parenting stress of young children (i.e., infants and toddlers) with ASD. A standard multiple linear regression was conducted to examine this relationship and answer the following questions: 1) Do parent-reported severity of challenging behaviors on the *BISCUIT-Part 3* (i.e., aggression/disruption, stereotypy, SIBs) collectively predict parent stress measured by the Parental Distress domain score on the *PSI-4-SF*? 2) Do any of these challenging behaviors factor severity scores from the *BISCUIT-Part 3* (i.e., aggression/disruption, stereotypy, or SIB), uniquely predict the scores of Parental Distress from the *PSI-4-SF*? Statistical results yielded both expected and surprising findings.

While Model 1 significantly predicted total *PSI-4-SF* Parental Distress domain scores ($R2 = .112$, $p = .030$), SIB severity was the only challenging behavior to uniquely predicted Parental Distress scores ($p = .016$). This relationship held up when a follow-up linear regression was conducted to exclude the factor severity scores of stereotypy and aggressive/disruptive behavior and to identify the amount of real-life variance that was added when challenging behaviors were analyzed as a whole. While the second regression still yielded significant small effects, the adjusted R2 changed from .112 in Model 1 to .078 in Model 2, which demonstrated that all challenging behaviors were the best fit when predicting parent stress of young children with ASD.

Regarding the nonsignificant challenging behavior factor of aggressive/disruptive behavior in Model 1, the correlation coefficient measuring the relationship between the severity of aggressive/disruptive behavior and Parental Distress scores was nearly negligible. However, after the standard multiple regression was run, the beta weight of SIB aggression/disruption

demonstrated a negative relationship (b = -.324). Statistically, this change in the relationship can be explained by the aggressive/disruptive behavior factor acting as a suppressor variable. A suppressor variable is defined more traditionally as an independent variable that is not correlated with the dependent variable but is correlated with another independent variable and increases the variance explained (Cohen, 1988; Friedman & Wall, 2005; Horst, 1941). Additionally, it has been described as an independent variable that is added to the model and produces a negative beta weight, even though the variable demonstrates a non-negative correlation with the outcome variable (Darlington, 1968; Friedman & Wall, 2005). Various statistical theorists have explained suppression, more recently referred to as model enhancement, as a specific model-building technique that increases predictive validity and variance explained by the regression model (Friedman & Wall, 2005). While the benefits of a suppressor variable can be revealing, understanding its influence on a model can be challenging. Therefore, it is imperative that the statistical impact of the suppressor variable's improvement of the model be noted, and predictive interpretations of aggressive/disruptive behavior adding unique variance should not be made considering the lack of a significantly correlated or predictive relationship with Parent Distress. Relatedly, the correlation coefficient measuring the relationship between the severity of stereotypy and Parental Distress scores in Model 1 was also nonsignificant. The severity scores of stereotypy resulted in a nonsignificant correlation ($r = 0.239$, $p>0.05$) and nonpredictive relationship ($p = .135$) with Parental Distress. This nonsignificant correlation with Parental Distress may be explained due to the underpowered nature of this sample; therefore, interpretation of the amount of variance stereotypy severity adds to the prediction of Parental Distress is limited.

As hypothesized, the overall impact of the study's results corroborated previous research, indicating the overall severity of challenging behaviors to significantly predict parent stress (Davis & Carter, 2008; Hastings et al., 2005; Lecavalier, 2006). Surprisingly, the results of the differential

impact each challenging behavior had on parent stress only partially supported the original hypotheses, as the small but notable effects highlighted SIB as the only challenging behavior to predict Parental Distress uniquely. Consideration should be given to the novelty of these findings and the minimal effect demonstrated in small sample size, and results should be interpreted with caution.

The limitations of this study and future directions should also be discussed. One limitation of this study is the small effect sizes of Model 1 and Model 2, which measure the strength of the predictive relationship of Parent Distress from the severity of challenging behaviors, particularly SIB. The targeted sample size was initially determined a priori and based on a range of existing research to yield a medium f^2 effect size of 0.25. However, the regression outcomes revealed significant predictive relationships that consistently demonstrated small effects (Model 1 f^2=.193 and Model 2 f^2 = .106). According to Cohen (1988), the presence of a small effect in a sample size determined based on an expected medium effect reared the sample as underpowered; this increases the risk of making a Type I error and decreases its applicability to treatment settings. Therefore, future researchers should replicate these results in a larger sample size to increase their reliability and potential to identify other small effects. Furthermore, future research should aim to identify additional independent factors that might have a predictive influence on parent stress and account for more variance to create a stronger predictive model for young children with ASD. Some factors to consider are ASD symptom severity (Davis & Carter, 2008; Gabriels et al., 2008; Konstantareas & Papageorgiou, 2006), age of the parent or child, and gender differences (Duarte et al., 2005; Lau et al., 2014).

In addition, the current sample yielded generalizable rates comparable to prior studies on challenging behavior (Rojahn et al., 2009) and parent stress levels in young children with ASD (Davis & Carter, 2008; Lecavalier, 2006). Interestingly, the majority of Parental Distress scores

were below clinical or subclinical levels. This means that most parents were experiencing a typical or expected degree of stress related to their role as a parent, which was not likely impairing the parent-child relationship. Additionally, the majority of the young children's challenging behaviors fell in the minimal to no impairment range. This indicates that most parents perceived their child's current challenging behaviors as having little to no impact on their functioning when compared to their same-age peers. These descriptive frequencies limit the generalizability of these findings to parents experiencing normative levels of parent distress who have a child with ASD that is engaging in minimally severe challenging behavior(s). Future researchers should apply the current study's aim and methodology to a sample consisting of primarily young children experiencing moderate to severe impairment of challenging behaviors and parents experiencing higher levels of parent stress. Researchers should also explore how factors influencing parent stress may differ between challenging behavior severity groups and if these factors change as parents and children with ASD age. Targeting these populations could also help identify implications that may improve treatment plans addressing various levels of functional impairment while also adjusting for changes over time.

It is also important to consider the appropriateness of the *PSI-4-SF* and the *BISCUIT-Part 3* when measuring stresses and challenging behavior based on parent reports. While the *PSI-4-SF* is one of the most commonly used measures of parent stress, it was not developed to measure the impact of challenging behavior on parent stress in populations with ASD. Relatedly, the *BISCUIT-Part 3* was normed on the studied sample. However, some items included aggregate behavioral descriptions, and limited the reportability of specific challenging behaviors. Therefore, future researchers are encouraged to use or create more discriminant measurement techniques for the population sample. These mediations may increase the validity of the predictive relationship between parent stress and challenging behavior in young children with ASD. Also, while this study

used the Parental Distress domain of the *PSI-4-SF* to measure parent stress, future researchers might consider including the Parent-Child Interaction domain to measure the extent to which parents feel satisfied with their child and their interactions with them (Abidin, 2012). Including this variable may account for additional parent stress factors influencing parenting behavior and child outcomes.

Lastly, many researchers have analyzed the influence of accompanying intellectual impairment has on challenging behaviors, as a comorbid diagnosis of ID has been found to increase the risk of a child engaging in these behaviors (Holden & Gitlesen, 2006; Jang et al., 2011; McClintock et al., 2003; Myrbakk & von Tetzchner, 2008). However, to receive a diagnosis of ID, an individual must have deficits in intellectual functioning, measured by clinical assessment and standardized IQ testing, with markedly impairing deficits in adaptive functioning and an onset of these symptoms in childhood (American Psychiatric Association, 2013; Bal et al., 2015; Papazoglou, 2014). Due to the participants' young age range, intellectual impairment was not accounted for in this study for the following reasons: standardized IQ testing is not valid until at least 5 to 6 years of age (Eysenck, 2012), and the robust evidence of young children with ASD experiencing adaptive deficits (Feige et al., 2021; Juergensen et al., 2018).

When considering the clinical implications of this study's findings, it is first recommended that these results be replicated. If shown to be reliable, it may be advantageous for practitioners to consider prioritizing the treatment of SIB. For instance, it is a common clinical practice to prioritize decreasing challenging behaviors in an applied early intervention treatment setting. This is due to their negative impact on learning, and a higher priority is given to more severe behaviors (Horner et al., 2002). Therefore, these findings possibly highlight the importance of targeting SIB in early intervention treatment to not only prevent self-injury or harm but decrease its negative impact on intervention and parent stress, in turn, potentially improving skill acquisition and the parent-child

relationship.

Overall, the results of this study revealed that challenging behaviors (i.e., aggression/disruption, stereotypy, and SIB) significantly predicted parent distress in a sample of young children with ASD demonstrating mild challenging behaviors. However, when differential effects were examined, SIB was found to be the single significant predictive factor of Parental Distress. Challenging behaviors are the most consistent child-related source of parent stress (Baker et al., 2002; Beck et al., 2004; Estes et al., 2009). Additionally, their presence in young children can interfere with skill acquisition during critical developmental periods (Fodstad et al., 2012; Itzchak et al., 2008; Itzchak & Zachor, 2011; Matson, 2007). Therefore, these results help fill a gap in understanding the understudied impact of challenging behaviors in young children with ASD on parent stress, both individually and collectively. Most young children with ASD may be too young to exhibit more severe challenging behaviors than older individuals (Fodstad et al., 2012). However, identifying the impact of small predictive effects is paramount to developing a more accurate understanding of how challenging behaviors potentially influence overall parent stress, parenting behavior, skill acquisition, and early intervention outcomes.

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Vita

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