The effect of cognitive skills and autism spectrum disorder on stereotyped behaviors in infants and toddlers

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A B S T R A C T
Stereotyped behaviors are prominent in both the ASD and ID populations; stereotypies can impede social skill acquisition, interfere with learning, and adversely affect an individual’s quality of life. The current study explored the effect of cognitive skills and autism spectrum disorder (ASD) on the rate of stereotypies in 2019 children aged 17–39 months. Cognitive abilities were assessed using the cognitive developmental quotient (DQ) on the Battelle Developmental Inventory, Second Edition (BDI-2); two levels of cognitive skill were used: (1) low (cognitive DQ less than or equal to 70), and (2) typical (cognitive DQ greater than 70). Stereotypies were examined utilizing the Baby and Infant Screen for Children with autism Traits, Part 3 (BISCUIT-Part 3). Children with ASD were found to have greater rates of overall stereotyped behaviors compared to children with atypical development, regardless of cognitive level; however, children with ASD and typical cognitive ability evinced the highest rate of stereotypies. An examination of specific stereotyped behaviors (i.e., unusual play with objects, repeated and unusual vocalizations, repeated and unusual body movements) revealed disparate results. Research and clinical implications regarding these findings are discussed.

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1. Introduction
The Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5) identifies stereotypies as repetitive, apparently purposeless motor behavior that seems driven; when identified as a clinical concern, stereotypic behaviors interfere with daily functioning and may or may not cause self-injury (Matson & LoVullo, 2009; Reed, Hirst, & Hyman, 2012). Simple stereotypies, such as body rocking, are common in typically developing infants and toddlers and are considered a normal part of development. However, some stereotypies may persist into adulthood such as hair twirling or finger drumming. These behaviors are often exacerbated by stress or boredom (Goldman et al., 2009).

Stereotypies may become the focus of clinical attention when the behaviors cause injury or persist with great frequency into later developmental periods (Matson, Kiely, & Bamburg, 1997). Stereotypies are differentiated from motor abnormalities such as tics by virtue of the ability for stereotypies to be voluntarily suppressible, at least for a time (Goldman et al., 2009). Examples of common stereotypies in individuals with developmental disabilities include hand
flapping, body rocking, spinning objects, finger waving, repetition of words or non-functional sounds, and unprovoked giggling or yelling (Duncan, Matson, Bamberg, Cherry, & Buckley, 1999; MacDonald et al., 2007; Singer, 2009). Even when stereotypies consist of seemingly harmless behaviors, stereotypies may command a great deal of the individual’s attention and impede social inclusion, developmentally normative opportunities, and the learning process (MacDonald et al., 2007; Matson & Nebel-Schwalm, 2007). This condition may be particularly problematic for individuals with disorders that put them at increased risk of delays in intellectual, mental health, or adaptive functioning (Matson, Dempsey, & Fodstad, 2009a; Matson, Rivet, Fodstad, Dempsey, & Boisjoli, 2009; Smith & Matson, 2010a, 2010b, 2010c).

Restricted, repetitive patterns of behavior, interests, or activities are a core feature of autism spectrum disorder (ASD), and stereotyped motor movements or speech are commonly associated with the disorder (Matson & Wilkins, 2008b). However, stereotypies are frequently seen within the context of other disorders as well. Increased risk factors for stereotypic movement disorders include neurogenetic factors (e.g., Lesch–Nyhan syndrome) as well as environmental factors including social isolation or an environment with insufficient stimulation (Singer, 2010). Stereotypies are common in those with intellectual disabilities (ID) due to a variety of etiologies (Matson & Dempsey, 2008; Matson & Rivet, 2008). Carminati, Gerber, and Constantin (2005) found that stereotypic behaviors were among the most commonly reported problem behaviors in a day program setting for individuals with intellectual disabilities. Results of some studies have shown that children with an ASD exhibit even more motor stereotypies than atypically developing children without ASD or individuals with ID only (Carcani-Rathwell, Rab-Hasketh, & Santosh, 2006; Goldman et al., 2009; Matson, Cooper, Malone, & Moskow, 2008; Matson & Wilkins, 2008a).

ASD and ID commonly co-occur, with estimates of ID in individuals with ASD ranging from about 50–70% (Artigas-Pallarés, Rigau-Ratera, & García-Nonell, 2007; LaMalfa, Lassi, Bertelli, Salvini, & Placidi, 2004). Among individuals with ID, estimates of ASD vary widely from 4%–40% (Matson & Shoemaker, 2009). Researchers have found that stereotypies are correlated with severity of autism symptoms (Bodfish, Symons, Parker, & Lewis, 2000; Campbell et al., 1990), and with greater intellectual disability (Bishop, Richler, & Lord, 2006; Militerni, Bravaccio, Falco, Fico, & Palermo, 2002). In comparing restricted, repetitive behavior (RRB) differences between individuals with ASD and varying levels of cognitive abilities, some researchers suggest that intelligence may be significantly related to the type of RRBs present (Gabriels, Cuccaro, Hill, Ivers, & Goldson, 2005; Goldman et al., 2009; Rapin & Katzman, 1998; Turner, 1999). ‘High level’ RRBs are defined as those behaviors that are more complex and thus, demand greater ability; ‘high level’ RRBs are found more often in children with ASD and typical cognitive skills and include unusual attachment to objects, repetitive speech, and circumscribed interests (Gabriels et al., 2005). ‘Low level’ RRBs are less sophisticated and require less skill. Researchers have indicated this level of RRBs is observed more often in children with ASD and low intellectual functioning and include sensory and motor stereotypies and self-injurious behaviors (Gabriels et al., 2005; Goldman et al., 2009).

Of note, the stereotyped behaviors and RRBs characteristic of ASD can be observed and identified as early as infancyhood (Fodstad, Rojahn, & Matson, 2012; Matson, Dempsey, & Fodstad, 2009b). Researchers have found that, as early as 10 months of age, parents of children with ASD report significantly higher rates of RRBs compared to parents of typically developing peers (Werner, Dawson, Munson, & Osterling, 2005). RRBs in infants and toddlers with ASD have also been found to worsen in frequency and presentation into childhood (Guthrie, Swineford, Nottke, & Wetherby, 2013). Though some people experience a decrease in RRB symptoms over the lifespan, a majority of individuals with ASD demonstrate long term impairment (Seltzer, Shattuck, Abbeduto, & Greenberg, 2004).

With regard to stereotypies, little research has been conducted on differences between individuals with ASD and varying levels of cognitive abilities, and how these groups may compare to atypically developing individuals without ASD. The present study sought to explore rates of stereotyped behaviors in infants and toddlers with ASD and with atypical development demonstrating different levels of cognitive skills. Comparisons of overall rate of stereotypies as well as rates of specific stereotyped behaviors such as unusual play with objects, unusual and repetitive vocalizations, and unusual and repetitive body movements were examined.

2. Method

2.1. Participants

Two thousand, nineteen infants and toddlers between the ages of 17 and 39 months (M = 25.65, SD = 4.65) were included in the study. The total sample consisted of 71.20% males and 28.80% females. 49.62% of the total participants were Caucasian (n = 1101), 39.30% were African American (n = 872), 2.25% were Hispanic (n = 50), and 8.83% were of other or unspecified ethnicity (n = 196). This sample was obtained via EarlySteps, Louisiana’s early intervention program under the Individuals with Disabilities Education Act, Part C, which provides services to infants and toddlers from birth to 3 years. A licensed psychologist with over 30 years of experience assigned ASD diagnoses using a variety of measures including the Modified Checklist for Autism in Toddlers (M-CHAT; Robins, Fein, & Barton, 1999), the Battelle Developmental Inventory, Second Edition (BDI-2; Newborg, 2005), the DSM-V criteria, and clinical judgment.

Participants were divided into four groups: an ASD group with typical cognitive ability (n = 194), an ASD group with low cognitive ability (n = 131), an atypical group with typical cognitive ability (n = 1694), and an atypical group with low cognitive ability (n = 200). Atypical groups did not meet criteria for ASD but were atypically developing (e.g., global developmental delay, cerebral palsy, Down syndrome, premature birth, seizure disorder, hydrocephalus). Because measures
of intellectual functioning (i.e., IQ) are somewhat unstable in young children (Matson, Mahan, Hess, & Fodstad, 2010), cognitive developmental quotient (DQ) was used to measure cognitive ability. Identifying strengths and weaknesses in regards to developmental milestones is a more precise and applicable measure for intervention purposes in this young population (Matson et al., 2010). Participants were categorized as having low cognitive ability if their cognitive DQ was less than or equal to 70 on the BDI-2. A cognitive DQ score over 70 indicated typical cognitive ability. Complete demographic information can be found in Table 1.

### 2.2. Measures

#### 2.2.1. Baby and Infant Screen for Children with Autistic Traits – Part 3 (BISCUIT-Part 3)

The BISCUIT-Part 3 is comprised of 15 items that assess challenging behaviors that commonly occur with ASD. Challenging behaviors include the following areas: aggression, disruption, self-injury, and stereotypies. Items are rated as "0 = not a problem or impairment," "1 = mild problem or impairment," and "2 = severe problem or impairment." The internal consistency coefficient of the BISCUIT-Part 3 is .91 (Matson, Wilkins, et al., 2009). The Stereotypies subscale was used in this study and consists of three items: unusual play with objects, repeated and unusual vocalizations, and repeated and unusual body movements. The overall rate of stereotyped behavior was represented by total score on the Stereotypies subscale of the BISCUIT-Part 3.

#### 2.2.2. Battelle Developmental Inventory, Second Edition (BDI-2)

The BDI-2 is an infant and observation based assessment that measures developmental skills from birth to 7 years 11 months. This measure consists of 450 items and is comprised of the Adaptive, Personal-Social, Communication, Motor, and Cognitive domains (Newborg, 2005). Items are scored as "0 = no ability in the skill," "1 = emerging ability," and "2 = ability at the skill." The BDI-2 has been found to have acceptable test–retest reliability and excellent internal consistency (Alfonso, Rentz, & Chung, 2010; Newborg, 2005). The Cognitive domain was used in the current study. The Cognitive domain consists of 105 items measuring "mental" and "intellectual" skills and is divided into three subdomains: (1) Attention and Memory, (2) Reasoning and Academic Skills, and (3) Perception and Concepts. The Attention and Memory subdomain consists of 30 items measuring a child’s visual and auditory ability to attend to environmental stimuli. The 35 items in the Reasoning and Academic Skills subdomain assess critical thinking skills, and the 40 items in the Perception and Concepts subdomain measures the child’s active sensorimotor interactions with the environment.

### 2.3. Procedure

The current study was approved by the Louisiana State University Institutional Review Board. Informed consent was obtained from informants of all participants. Informants included parents, other caregivers, and legal guardians. Qualified and trained interviewers (e.g., social worker, physical therapist, speech-language pathologist, occupational therapist) administered the BISCUIT-Part 3 and the BDI-2. The measures were scored by the same interviewers.

### 2.4. Statistical analyses

A priori analyses were conducted to determine potential differences among groups in terms of age, race, or gender. Chi square tests indicated no significant differences for gender $\chi^2(3) = 6.61, p = .09$ or race $\chi^2(9) = 15.43, p = .08$. An analysis of variance (ANOVA) was conducted to investigate potential differences in age. Significant differences in age were observed between groups, $F(3, 2215) = 8.59, p < .001$. Scheffé post hoc comparisons indicated that the atypical group with typical cognitive DQ ($M = 25.38$ months, $SD = 4.61$) was significantly younger than the atypical group with low cognitive DQ ($M = 26.69$ months, $SD = 5.00$); no significant differences were observed in other groups. However, because this difference was limited only to the atypical groups, age was not added as a covariate in subsequent analyses. Rather, results of comparisons between the two atypical groups were interpreted with caution.

### Table 1

Demographic information.

<table>
<thead>
<tr>
<th></th>
<th>ASD + typical cognitive DQ (n = 194)</th>
<th>ASD + low cognitive DQ (n = 131)</th>
<th>Atyp + typical cognitive DQ (n = 1694)</th>
<th>Atyp + low cognitive DQ (n = 200)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Mean age (in months)</td>
<td>26.41</td>
<td>26.52</td>
<td>25.38</td>
<td>26.69</td>
</tr>
<tr>
<td>Gender</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Male</td>
<td>78.35%</td>
<td>70.23%</td>
<td>70.13%</td>
<td>74.00%</td>
</tr>
<tr>
<td>Female</td>
<td>21.65%</td>
<td>29.77%</td>
<td>29.87%</td>
<td>26.00%</td>
</tr>
<tr>
<td>Race</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>African American</td>
<td>45.88%</td>
<td>45.04%</td>
<td>37.25%</td>
<td>46.50%</td>
</tr>
<tr>
<td>Caucasian</td>
<td>46.38%</td>
<td>41.98%</td>
<td>51.42%</td>
<td>42.50%</td>
</tr>
<tr>
<td>Hispanic</td>
<td>1.55%</td>
<td>2.29%</td>
<td>2.30%</td>
<td>2.50%</td>
</tr>
<tr>
<td>Other/unspecified</td>
<td>6.19%</td>
<td>10.69%</td>
<td>9.03%</td>
<td>8.50%</td>
</tr>
</tbody>
</table>
An ANOVA was conducted with group as the independent variable (IV) and the overall rate of stereotyped behavior as the dependent variable (DV). Follow up Scheffé post hoc comparisons were conducted. Further, in order to compare individual stereotyped behaviors among the four groups, a multivariate analysis of variance (MANOVA) was used. Group served as the IV; the DVs included unusual play with objects, repeated and unusual vocalizations, and repeated and unusual body movements. Three follow up ANOVAs were conducted to examine specific stereotyped behavior differences more comprehensively between groups. A Bonferroni-adjusted significance level of .017 was applied (i.e., p of .05 divided by a total of 3 items analyzed). Lastly, Scheffé post hoc tests were conducted to account for unequal sample sizes and to examine differences more extensively.

3. Results

The initial ANOVA indicated significant differences in overall rate of stereotyped behaviors between groups, $F(3, 2215) = 66.44, p < .001$, partial $\eta^2 = .08$; children with ASD and a typical cognitive DQ on BDI-2 ($M = 1.61, SD = 1.86$) displayed significantly more stereotyped behaviors than children with ASD with a low cognitive DQ ($M = 1.05, SD = 1.60$), children with atypical development with a typical cognitive DQ ($M = 0.43, SD = 1.01$), and children with atypical development with a low cognitive DQ score ($M = 0.65, SD = 1.28$). Children with ASD and low cognitive skills demonstrated significantly more stereotyped behaviors than children in both atypical groups. The atypical groups did not significantly differ on overall stereotypy. Results for the total stereotyped behavior comparison are presented in Table 2.

A MANOVA was then conducted to identify particular stereotypes related to cognitive skills in infants and toddlers with ASD and atypical development. The MANOVA yielded significant differences in challenging behaviors among groups, $F(9, 6645) = 24.66, p < .001$, partial $\eta^2 = .03$. Follow-up ANOVAs yielded significant differences among groups on all three behaviors: unusual play with objects, $F(3, 2215) = 53.49, p < .001$, partial $\eta^2 = .07$, repeated and unusual vocalizations, $F(3, 2215) = 33.67, p < .001$, partial $\eta^2 = .04$, and repeated and unusual body movements, $F(3, 2215) = 39.50, p < .001$, partial $\eta^2 = .05$.

Results from Scheffé post hoc tests indicate that the infants and toddlers with ASD with a typical cognitive DQ ($M = 0.42, SD = 0.71$) did not significantly differ on unusual play with objects from children with ASD with a low cognitive DQ ($M = 0.32, SD = 0.66$). Both ASD groups evinced significantly more unusual play with objects than atypically developing children with a typical cognitive DQ ($M = 0.08, SD = 0.32$) and atypically developing children with a low cognitive DQ ($M = 0.12, SD = 0.42$). The atypical groups did not significantly differ on unusual play with objects.

The ASD group with a typical cognitive DQ ($M = 0.60, SD = 0.83$) displayed significantly more repetitive and unusual vocalizations compared to the ASD group with a low cognitive DQ ($M = 0.31, SD = 0.67$), the atypical group with a typical cognitive DQ ($M = 0.18, SD = 0.50$), and the atypical group with a low cognitive DQ ($M = 0.25, SD = 0.58$). The children with ASD with a low cognitive DQ and the atypical groups did not significantly differ on vocalizations.

The ASD group with a typical cognitive DQ ($M = 0.58, SD = 0.81$) evinced significantly more repetitive and unusual body movements than the atypical group with a typical cognitive DQ ($M = 0.17, SD = 0.49$) and the atypical group with a low cognitive DQ ($M = 0.29, SD = 0.63$). Both the ASD group with a low cognitive DQ ($M = 0.43, SD = 0.76$) and the atypical group with a low cognitive DQ evinced significantly more repetitive and unusual body movements than the atypical group with a typical cognitive DQ. The ASD group with a low cognitive DQ did not significantly differ from the children with ASD with a typical cognitive DQ or the children with atypical development with a low Cognitive DQ. Item means for each group and comparison results are presented in Table 3.

### Table 2
Comparison of total stereotyped behavior between groups.

<table>
<thead>
<tr>
<th></th>
<th>ASD + typical cognitive DQ</th>
<th>ASD + low cognitive DQ</th>
<th>Atyp + typical cognitive DQ</th>
<th>Atyp + low cognitive DQ</th>
</tr>
</thead>
<tbody>
<tr>
<td>M (SD)</td>
<td>1.61 (1.86)</td>
<td>1.05 (1.60)</td>
<td>0.43 (1.01)</td>
<td>0.65 (1.28)</td>
</tr>
</tbody>
</table>

Note: *M* = mean; *SD* = standard deviation.

* Significantly different from ASD + typical cognitive DQ, $p < .001$.

### Table 3
Item comparison between groups.

<table>
<thead>
<tr>
<th>Stereotypies</th>
<th>ASD + typical cognitive DQ</th>
<th>ASD + low cognitive DQ</th>
<th>Atyp + typical cognitive DQ</th>
<th>Atyp + low cognitive DQ</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>M (SD)</td>
<td>M (SD)</td>
<td>M (SD)</td>
<td>M (SD)</td>
</tr>
<tr>
<td>Unusual play with objects</td>
<td>0.42 (0.21)</td>
<td>0.32 (0.66)</td>
<td>0.08 (0.32)</td>
<td>0.12 (0.42)</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Repeated and unusual vocalizations</td>
<td>0.60 (0.83)</td>
<td>0.31 (0.67)</td>
<td>0.18 (0.50)</td>
<td>0.24 (0.58)</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Repeated and unusual body movements</td>
<td>0.58 (0.81)</td>
<td>0.43 (0.76)</td>
<td>0.17 (0.49)</td>
<td>0.29 (0.63)</td>
</tr>
</tbody>
</table>

* Significantly different from ASD + typical cognitive DQ, $p < .005$.

* Significantly different from ASD + low cognitive DQ, $p < .05$.

* Significantly different from Atyp + low cognitive DQ, $p < .05$. 
4. Discussion

Consistent with previous research (Carcani-Rathwell et al., 2006; Fodstad et al., 2012; Goldman et al., 2009; Matson & Dempsey, 2008; Matson, Dempsey et al., 2009b), infants and toddlers with ASD, regardless of cognitive skill level, demonstrated significantly greater overall stereotypies compared to atypically developing peers of typical or low cognitive ability. Because restricted, repetitive, and stereotyped patterns of behavior serves as one of the diagnostic domains for ASD, this finding is not surprising. However, in contrast with previous research (Bishop et al., 2006; Militeri et al., 2002), results indicate that infants and toddlers with ASD and typical cognitive skills, displayed significantly greater overall stereotypy than children with ASD and low cognitive skills. Further, three distinct relationships were found when examining the specific stereotyped behaviors of unusual play with objects, repetitive and unusual vocalizations, and repetitive and unusual body movements.

Unusual play with objects appeared to be endorsed at low rates by parents of infants and toddlers with and without ASD. However, the behavior seems to be autism-specific; infants and toddlers with ASD of low and typical cognitive levels did not differ from each other but evinced significantly more unusual play with objects than their atypically developing peers. These results suggest that unusual play with objects (e.g., twirling string, staring at a toy) may serve as a good indicator for possible ASD in the diagnosis of very young children.

Infants and toddlers with ASD and typical cognitive skills displayed the highest rate of repeated and unusual vocalizations. There were no significant differences in vocalizations between the ASD and low cognitive ability group and the two atypical groups. Perhaps within the infant and toddler population, repeated and unusual vocalizations can be categorized as a 'high level' restricted and repetitive behavior as repetitive language, rigidity, and circumscribed interests are in older populations (Gabriels et al., 2005; Rapin & Katzman, 1998; Turner, 1999). Thus, results of the current study indicate that the RRB dichotomy ('high level' versus 'low level' RRBs) should be reevaluated based on age. It is also possible the difference may be in part due to acquisition of verbal skills. The participants with ASD and low cognitive abilities may be more impaired verbally and perhaps make less vocalizations of any type.

Rates of repeated and unusual body movements (e.g., hand flapping, body rocking) did not differ between ASD groups. Infants and toddlers with ASD and typical cognitive abilities evinced significantly more repetitive body movements than both atypical groups. The ASD group with low cognitive skills did not differ from the atypical group with low cognitive skills; however, both groups showed more repeated and unusual body movements than the atypical group with typical cognitive abilities. Though the difference between the atypical groups may be due to the difference in age, the results obtained are consistent with previous research; children with low cognitive abilities engage in behaviors such as body rocking at a higher rate than peers with average cognitive skills. The lack of difference between the ASD group with low cognitive ability and the atypical group with low cognitive ability on repeated and unusual body movements is surprising and requires further research. This finding may be due to the heterogeneity of the atypical groups; individuals labeled with atypical development have a variety of impairments. Among these individuals, several disorders such as cerebral palsy could account for the higher rates of body movements.

Because stereotypies are socially stigmatizing, interfere with normal development, and impede on skill acquisition during intervention, it is important to target these behaviors in the assessment and treatment of individuals with ASD and/or ID (Gardenier, MacDonald, & Green, 2004; Klintwall & Eikeseth, 2012). The informed treatment of stereotypies should begin with the identification of the behavioral functions where possible (Cunningham & Schreibman, 2008; Matson & Minshawi, 2007). The function indicates the factors that maintain a behavior. Attention, escape, access to a preferred item, and automatic reinforcement are the most common functions (Tarbox et al., 2009). These problems tend to be compounded by problems in communication (Fodstad, Matson, Hess, & Neal, 2009). There is a trend in the research literature to classify stereotypy as self-stimulatory and thus, assume all stereotypies serve an automatic function. However, stereotyped behaviors can also be socially mediated or serve as an escape from task demands (Matson & Boisjoli, 2007).

Behavioral function can be assessed in a number of ways: indirectly through client or caregiver interview, descriptively through direct behavioral observation, or experimentally through functional analysis (Tarbox et al., 2009). Treatment of stereotypy should focus on the determined function. Common behavioral intervention procedures for decreasing stereotyped behaviors involve non-contingent attention, non-contingent access to matched stimulation, response cost, differential reinforcement of alternative behavior (DRA), differential reinforcement of other behavior (DRO) and response interruption and redirection (Ahearn, Clark, MacDonald, & Chung, 2007; Athens, Vollmer, Sloman, & Pipkin, 2008; Fritz, Iwata, Rolider, Camp, & Neidert, 2012; Love, Miguel, Fernand, & LaBrie, 2012; Matson & LoVullo, 2008).

Based on the results of the current study, the presence of ASD and level of cognitive ability seem to be related to the stereotypies exhibited by infants and toddlers. Because diagnoses are being made earlier in childhood, further research examining a greater variety of RRBs in young children with ASD and varying cognitive abilities would be beneficial. In examining the effect of cognitive abilities, stereotypies of individuals with ASD should also be compared to a typically developing group. Future research focusing on subtypes of RRBs and their correlates in infants and toddlers with ASD is imperative and may improve the assessment of and intervention for young children.

References


