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ORIGINAL ARTICLE

Autism Symptoms and Problem Behaviors in Children with and without Developmental Regression

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Abstract Although frequently observed in individuals with ASD, little is known about the nature of developmental regression amongst this population. Previous studies have found varying results in differences in ASD symptoms, cognitive functioning, and comorbid problems between individuals who have and have not regressed in skills. The purpose of the present study was to examine potential differences in ASD symptom domains (i.e., nonverbal communication/socialization, verbal communication, social relationships, insistence on sameness/restricted interests) and challenging behaviors across groups. Participants included 160 total children with ASD (M = 8.17, SD = 4.12), of which 70 reported a history of developmental regression. Results indicated significant differences between groups on ASD severity, nonverbal communication/socialization, verbal communication, social relationships, and internalizing problems. No difference was found on insistence on sameness/restricted interests and externalizing problems. Implications of these data are discussed.

Keywords Autism spectrum disorder · Symptomology · Challenging behaviors · Problem behaviors · Developmental regression

Autism spectrum disorder (ASD) is a neurodevelopmental disorder defined by marked impairments in social communication and social interaction and the presence of restricted and repetitive behaviors and interests (American Psychiatric Association (APA) 2013). Although often diagnosed at 3 years of age or older (Daniels and Mandell 2013), caregiver concerns regarding development usually begin between the first and second year of life for a child later diagnosed with autism (De Giacomo and

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Fombonne 1998). Typical onset of ASD symptomology is gradual throughout the first several years of life, where lack of developmental progression and autism symptoms become more prominent with age (Werner et al. 2005). However, there appears to be a subset of individuals with ASD who experience a late onset pattern (Ozonoff et al. 2008).

Developmental regression is among these late ASD onset patterns. Regression involves a loss of previously acquired skills and typically occurs prior to the age of 3 years old (Gadow et al. 2017; Kalb et al. 2010; Ozonoff et al. 2008). The rate of regression in the ASD population varies depending on sample characteristics, definition of regression, and assessment methods used within studies; though, it is estimated that regression occurs in approximately one-third of individuals with ASD (Gadow et al. 2017; Jones and Campbell 2010; Ozonoff et al. 2008). Regression can occur within multiple developmental domains (e.g., social, communication, motor). Most children who experience regression demonstrate both language and social skill losses (Kalb et al. 2010; Ozonoff et al. 2008). Developmental regression was traditionally conceptualized as a loss of skills (particularly language) following otherwise typical development, and evidence suggests that this form of regression does exist (Ozonoff et al. 2008; Saint-Georges et al. 2010). However, regression of skills following early developmental abnormalities (e.g., social and/or communication delays) has been found to be significantly more prevalent within the ASD population (Kalb et al. 2010; Ozonoff et al. 2008; Werner et al. 2005). Though distinct, researchers have found that individuals who experience regression after demonstrating no early symptoms do not differ in functional outcomes from individuals who regress after exhibiting delays (Shumway et al. 2011).

Presentation differences between individuals with ASD who experienced regression and individuals who did not regress remains less clear. Some researchers suggest that individuals with ASD who have a history of regression present as a distinct phenotype that should be studied separately from those without regression (Matson et al. 2010), while other researchers argue that no differences exist between those who have and have not regressed (Jones and Campbell 2010; Kalb et al. 2010). Within the studies that demonstrate significant differences between these groups, poorer prognoses across domains (e.g., verbal, social, behavior) are reported for individuals with ASD who had regressed (Kalb et al. 2010). Gadow et al. (2017) found that children with a history of regression were more likely to meet criteria for intellectual disability, have a diagnosis of epilepsy, have more severe ASD symptoms particularly within the communication domain, and have higher endorsements of schizophrenia spectrum symptoms (e.g. disorganized behavior and avolition). Hansen et al. (2008) also reported that children who have experienced regression demonstrated lower scores across measures of communication skills; however, these differences were small. Further, Bernabei et al. (2007) found that the poorer performance in communication and social functioning of individuals who regressed compared to those who did not regress may become more significant over time.

Despite these findings, several researchers have demonstrated no such difference within these groups. In a range of studies comparing ASD groups who have and have not experienced regression, children did not differ on autism symptoms (Gadow et al. 2017; Jones and Campbell 2010; Shumway et al. 2011), intellectual ability (Hansen et al. 2008; Shumway et al. 2011; Werner et al. 2005), adaptive behavior (Hansen et al.

2008; Shumway et al. 2011; Werner et al. 2005), seizure risk (Hansen et al. 2008), sleep problems (Hansen et al. 2008), or behavioral concerns (Hansen et al. 2008; Jones and Campbell 2010; Werner et al. 2005).

Although developmental regression in ASD has been addressed in the research for some time, much remains unclear regarding this late onset pattern and its implications for outcomes (Matson and Kozlowski 2010). Additional research is necessary to provide more accurate prognostic information regarding regression in autism. The present study extends findings from a previous research study (Matson et al. 2010) by further examining differences in overall ASD severity, ASD symptom domains, and internalizing and externalizing behaviors in a larger sample of children with and without a history of regression. Because participants of the current study were of a wider age range than in previous studies, examination of outcome differences several years after the reported regression occurred was possible. Further adding strength to the study, regression was loosely defined to include both children with regression from typical development and regression following delays. Developing a greater understanding of symptom differences at a domain level may lead to a better understanding of developmental trajectories and thus improve treatment services.

Methods

Participants

This study was approved by the Louisiana State University Institutional Review Board. All procedures in the study were performed in accordance with the ethical standards of the Louisiana State University Institutional Review Board, as well as the 1964 Helsinki declaration and its later amendments. Informed consent was obtained from all participants included in the study. The participants were children who received an evaluation from the Louisiana State University psychological services clinic and were referred for concerns regarding developmental delays. Evaluations were conducted by graduate students in a clinical psychology doctoral program supervised by a licensed clinical psychologist who specializes in the assessment of developmental disorders. Diagnoses were made based upon developmental history, in-clinic observation, various ratings scales, and clinical judgment. Parents/caregivers and children (when appropriate) were consented for participation in research, and data obtained from assessments were entered into an archival database that continues to expand with ongoing assessments. The version of the database used in this study included participants recruited between September 2006 to December 2016, resulting in 201 total children aged 2 to 16 years (M = 8.26, SD = 3.97).

For the purposes of the current study, only children with a diagnosis of ASD were included, which resulted in 160 children (age range 2–16 years, M = 8.17, SD = 4.12) (Table 1). The sample was composed of 83.1% males and 16.3% females. Race/ethnicity was reported as 7.5% African American, 58.1% Caucasian, 1.9% Hispanic, and 32.5% other or unspecified ethnicity. Of the 160 children with ASD, 43.75% (N = 70) were reported to have a history of developmental regression. Regression was indicted by parents/caregivers on the clinic's family information form by answering the question: "Was there a period of time during development that your child lost skills?"

Table 1 Participant demographics separated by group

	Total *N* = 160	ASD only *N* = 90	ASD+R *N* = 70
Age M (SD)	8.17 (4.12)	8.73 (4.25)	7.47 (3.86)
Gender			
Male	83.1%	84.3%	82.9%
Female	16.3%	15.6%	17.1%
Race/Ethnicity			
AA	7.5%	10.0%	4.3%
White	58.1%	55.6%	89.6%
Hispanic	1.9%	2.2%	2.1%
Other	32.5%	32.2%	32.8%

AA African American

Children whose parents/caregivers responded "yes" were coded as the "ASD + R" group (*M* = 7.47 years old, *SD* = 3.86 years) and those who responded "no" were coded as the "ASD only" group (*M* = 8.73 years old, *SD* = 4.25 years). Parents/caregivers were then asked to describe the nature of skills lost. Due to the open-ended nature of the question, it was possible for parents to report regression in multiple domains. Skills that were indicated as lost included speech (e.g., "was babbling then stopped," "lost words"), motor skills (e.g., rolling over, crawling), social skills (e.g., "engagement with people he knew," eye contact), and adaptive skills (e.g., toilet training).

Measure

The Autism Spectrum Disorders Assessment Battery for Children (ASD-Child) are three separate scales developed to assess ASD symptoms, comorbid problems, and problem behaviors in children age 2 to 16 years old. The Autism Spectrum Disorders-Diagnostic Child Version (ASD-DC; Matson and Gonzalez 2007a) is an informant-based rating scale designed to assess for ASD. It consists of 40 items, and parents/caregivers rate the child's behaviors to the "extent that it is/was ever a problem." By comparing the child to same aged peers, parents/caregivers rate the child's behavior as "0" = not different/no impairment, "1" = somewhat different/mild impairment, or "2" = very different/severe impairment. All items are summed together to produce a total score. Factor analysis of the measure's items yielded the following factors: nonverbal communication/socialization, verbal communication, social relationships, and insistence on sameness/restricted interests (RRB; Matson et al. 2009). The ASD-DC has good psychometrics, with internal consistency at .99, test-retest reliability κω=. 77, and inter-rater reliability κω=. 67 (Matson et al. 2008b). The measure also has good sensitivity (84.3%), specificity (98.2%), and overall correct classification rate (91.3%; Matson et al. 2008b).

The Autism Spectrum Disorders-Problem Behavior-Child Version (ASD-PBC; Matson and Gonzalez 2007b) is an informant-based rating scale that assesses for common challenging behaviors in children with ASD, including aggressive, self-injurious, and stereotypic behaviors. It was designed to be an initial screen for further assessment of problem behaviors. The ASD-PBC consists of 18 items, and, just as in the ASD-DC, parents/caregivers rate the child's behavior as "0" = not different/no impairment, "1" = somewhat different/mild impairment, or "2" = very different/severe impairment. The ASD-PBC has two factors: externalizing (e.g., kicking objects, aggression towards others, property destruction) and internalizing problems (e.g., unusual play with objects, playing with own saliva, mouthing or swallowing objects). The ASD-PBC has high internal consistency (α = .90), fair test-retest reliability (κ = .64) and fair inter-rater reliability (κ = .49; Matson et al. 2008a).

Statistical Analysis

All statistical analyses were conducted with SPSS 22.0. A priori analyses were conducted to explore potential group differences in gender, race/ethnicity, and age. Chi-square analyses revealed no significant difference between groups on gender, $\chi^2(1) = .06$, $p > .05$, or race/ethnicity, $\chi^2(3) = 3.18$, $p > .05$. An independent samples *t*-test indicated no significant difference between groups on age, $t(150) = 1.88$, $p > .05$.

To examine differences between the ASD only and ASD + R groups, several independent samples *t*-tests were conducted with various factors as the dependent variable (i.e., ASD-DC total score, ASD-DC Nonverbal communication/socialization score, ASD-DC Verbal communication score, ASD-DC Social relationships, ASD-DC RRB, ASD-PB Externalizing, ASD-PB Internalizing).

Results

The age at regression for this sample was found to be 20.96 months (*SD* = 8.65), with the range of reported age at regression between 6 and 48 months. Regarding skills lost, 94% were reported to lose speech (e.g., stopped saying babbling or saying words), 28% lost social skills (e.g., eye contact, interest in interacting with others), 8% lost nonverbal language skills (e.g., stopped pointing), and 6% lost motor skills (e.g., stopped crawling). Regression of skills in more than one domain was reported in 27% of participants.

Results are shown in Table 2. Independent samples *t*-tests were conducted to examine differences between groups. Significant differences were found between the ASD only and ASD + R groups on total ASD severity (i.e., ASD-DC total score), $t(146) = -2.42$, $p < .05$; the ASD-DC Nonverbal communication/socialization score, $t(149) = -2.23$, $p < .05$; ASD-DC Verbal communication score, $t(148) = -3.16$, $p < .01$; and ASD-DC Social relationships score, $t(151) = -2.69$, $p < .01$. No significant difference was found between groups on the ASD-DC RRB score, $t(152) = -1.48$, $p > .05$. Regarding problem behaviors, a significant difference was found between groups on the Internalizing problems scale, $t(150) = -2.56$, $p < .05$, but not on the Externalizing problems scale, $t(151) = -0.72$, $p > .05$.

Table 2 Comparison of scores between ASD only and ASD+R groups

	ASD only *N* = 90	ASD + R *N* = 70
ASD-DC total	47.78 (17.36)	54.29 (14.73)*
Nonverbal communication	17.46 (7.00)	19.82 (5.83)*
Verbal communication	10.00 (4.47)	12.16 (3.76)*
Social relationships	9.36 (3.54)	10.84 (3.13)*
RRB	6.62 (2.92)	7.33 (2.99)
Externalizing	2.50 (2.47)	2.81 (2.91)
Internalizing	3.69 (2.99)	5.06 (3.63)*

* Indicates significant difference between ASD only and ASD + R groups, $p < .05$

Discussion

Interest in studying regression in individuals with ASD has grown because of the potential to study underlying causal mechanisms of autism, provide further means of categorization to this heterogeneous disorder, and assist with prognosis (Williams et al. 2015). The prevalence of regression is also higher in individuals with ASD than in other developmental conditions, which provides researchers with the opportunity to study this phenomenon (Wiggins et al. 2009; Williams et al. 2015). Consistent with previous researchers (Barger et al. 2013; Matson and Kozlowski 2010), the current study found that 43.75% of children with ASD experience regression of skills with the average age of onset at 20.96 months. Of those who regressed, the majority were reported to lose speech and/or social skills, with nearly 95% of caregivers of children who have lost skills reporting a regression in language. Though not diagnostic of ASD, it is important to note that many children with the disorder experience a loss of skills early in their development. Therefore, monitoring of skills in these domains at multiple time points in early development, particularly in children who may be at-risk for ASD, is important for early identification.

Results from current analyses comparing functional outcomes of children who have and have not regressed support previous research indicating that individuals with regression experience greater deficits in ASD-associated symptoms (i.e., ASD severity, nonverbal communication and socialization skills, verbal communication skills, social relationships), as well as in behavioral problems (Bernabei et al. 2007; Kalb et al. 2010; Luyster et al. 2005; Matson et al. 2010; Meilleur and Fombonne 2009). Therefore, those who lose skills early in development may exhibit a profile of more severe deficits both in ASD specific (i.e., verbal and nonverbal communication, socialization skills) and more global (i.e., behavioral problems) developmental areas. In regard to ASD symptom domain differences in the current study, results indicated that children who regressed continued to experience severe deficits that affect their communication skills and social relationships. Interestingly, no difference was found between groups on RRBs, suggesting that regression may not influence the presence or severity of these core ASD symptoms. This finding is in contrast to previous studies, which found that some of the greatest discriminators between children with ASD with and without regression were

RRBs (e.g., odd/restricted interests, engagement in behaviors that impair daily routines or activities, unusual play with objects; Matson et al. 2010; Meilleur and Fombonne 2009). As such, additional research is needed to confirm these results.

In relation to ASD-associated difficulties, the ASD + R group was found to exhibit greater ASD severity (particularly in the social communication and social interaction domain), and previous researchers have found that ASD severity is associated with higher rates of challenging behaviors (Jang et al. 2011). Therefore, it would be expected that the ASD + R group would evince higher rates of behavior problems across both internalizing and externalizing categories. However, results showed that the ASD + R group had higher rates of internalizing problem behaviors (e.g., unusual play with objects, playing with own saliva, mouthing/swallowing objects), but not externalizing problem behaviors (e.g., kicking objects, aggression towards others, property destruction). This indicates that regression may differentially influence specific presentations of comorbid symptomology. More research should be done examining the relationship between regression and problem behaviors; further subtyping presentations of challenging behavior, beyond internalizing and externalizing categories, would be beneficial in the examination of these group differences.

Though this study provides support for the notion that regression negatively impacts prognoses in children with ASD, a number of researchers have found no significant difference in various outcomes for individuals with ASD who regressed from individuals with ASD who have not (Gadow et al. 2017; Hansen et al. 2008; Jones and Campbell 2010; Shumway et al. 2011; Werner et al. 2005). These differences may be due to how regression was defined in each study, as well as methodology, sampling method, and age of the individual at assessment. To allow for more precise conclusions on prognostic information, definitional issues in regression research must be addressed. As stated by other researchers, the definition of regression likely affects study results (The International Molecular Genetic Study of Autism Consortium (IMGSAC) et al. 2011; Wiggins et al. 2009), and various definitions and categorizations of regression have been used across the literature (e.g., language regression, social regression, language and social regression, mixed regression; Barger et al. 2013). Furthering our understanding of regression to determine a more precise definition would lead to a bettered ability to compare results across studies and thus more accurate estimations of developmental trajectories. Relatedly, although used widely in regression literature, conceptualization of regression as a dichotomous variable (i.e., ASD with or without history of regression) may limit research findings because the phenomenon may instead occur on a spectrum and result in various developmental and behavioral profiles (Ozonoff et al. 2008).

Methodological challenges should also be addressed. Parent-report is typically used to establish history of regression; however, other studies have utilized more formal criteria (e.g., response to questions on the Autism Diagnostic Interview-Revised; Lord et al. 1994). Parent-report is subject to bias. Further, parents typically do not have extensive training in child development, which may limit their ability to detect subtle skill losses and/or early symptoms consistent with ASD or developmental delay (Zwaigenbaum et al. 2007). In the present study, the open-ended question of the ASD-DC was used to elicit a broad range of responses regarding loss of developmental skills; however, without specific prompting for additional information, parents/caregivers may not have reported or been aware of all possible domains for potential skill loss. Prospective studies may be better able

to inform the nature and onset of regression in these children and thus better operationalize the phenomenon for future research. Further prospective research should be also conducted longitudinally to assess differential prognoses related to regression across development.

The present study also included children as young as 2 years old. Given that the age range for regression typically occurs between 18 to 36 months (Barger et al. 2013), it is possible that some children may not have experienced regression in skills yet at the time of assessment. Younger children were included in this study to provide information on individuals who may have regressed earlier; children in the ASD + R group were as young as 2 years and experienced regression as early as 6 months old. Thus, inclusion of younger children provides information on regression at very young ages; however, it is also possible that young participants in the ASD only group may not have experienced regression and thus may have been inaccurately classified due to their presentation at the time of assessment. Therefore, results should be interpreted with some caution.

Although a number of children with ASD regress, there is still limited knowledge of the nature of regression and how it may affect prognosis. Studies on regression may further elucidate the underlying mechanisms of autism and their associated behavioral deficits. Additional research is needed to study not only what neural mechanisms may be involved, but also how they influence the development of ASD and severity of impairments. Identifying domains that may distinguish between individuals with ASD only and individuals with ASD + R may help inform treatment planning and understanding the developmental trajectories of affected individuals. At present, it is unclear whether children with ASD who experience developmental regression have different outcomes than those who do not experience regression. Regardless, it is important to individualize treatment for individuals with ASD to serve their unique needs. Due to the limited understanding of regression, it is important for future research to continue to examine and define this phenomenon and identify what factors may be predictive of regression and how it occurs.

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Compliance with Ethical Standards

Ethical Approval All procedures performed in this study were in accordance with the ethical standards of the institution, and with the 1964 Helsinki declaration and its later amendments.

Informed Consent Informed consent was obtained from all parents/legal guardians of the participants in the study.

Conflict of Interest Jasper A. Estabillo and Paige E. Cervantes declare that they have no conflicts of interest. Johnny L. Matson's wife is the sole owner of the Autism Spectrum Disorders Assessment Battery for Children, which is sold by her company.

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