Diagnosis Advancements

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Relationship between Stereotyped Behaviors and Restricted Interests (SBRIs) measured on the Autism Diagnostic Observation Schedule (ADOS) and diagnostic results.

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Abstract

Introduction

This study investigated possible relationships between Stereotyped Behaviors and Restricted Interests (SBRIs) measured on the Autism Diagnostic Observation Schedule (ADOS) and diagnostic results in 119 children ages 16 months to 15 years of age. Sub-categories of SBRI symptoms, including sensory behaviors, complex motor mannerisms, and restricted interests and unusual behaviors on the ADOS were compared to diagnostic findings.

Results

Results indicated that mean SBRI scores varied across diagnostic categories with higher scores associated with more severe ADOS classifications and DSM-IV diagnoses. Furthermore, certain sub-categories of SBRI symptoms varied more significantly across diagnostic results.

Conclusion

These results support diagnostic validity of the SBRI domain for use with the ADOS and DSM-IV, with particular significance due to recent revisions of both (ADOS-2 and DSM-5).

Introduction

Stereotyped behaviors and restricted interests (SBRIs; also referred to as restricted and repetitive behaviors) are a core symptom area associated with Autism Spectrum Disorders (ASDs). The Diagnostic and Statistical Manual of Mental Disorders (DSM) guides the definition of this subset of symptoms and classifies SBRIs as restricted interests (e.g. preoccupation with trains), non-functional routines and rituals (e.g. always must drive a certain route), repetitive motor mannerisms (e.g. hand flapping), preoccupation with parts (e.g. fixating on the eyes of a baby doll), and sensory behaviors (e.g. seeking tactile input) [1,2,3].

Assessment of SBRI symptoms is necessary for diagnosis, treatment planning, and measuring response to intervention. Additionally, they are important to evaluate due to the impact they have on a child's functioning in areas such as learning, communication and social

interaction[4]. However, these symptoms have not received significant emphasis in many diagnostic tools until just recently. Part of this previous lack of emphasis may be due to perceived difficulties with diagnostic differentiation of these symptoms. For example, SBRI's have often been viewed as having been already considered within communication and social deficit symptoms [5]. Additionally, SBRI symptoms are also present in children with non-spectrum diagnoses, particularly with other developmental disorders and with intellectual disability [6,7], as well as in typically developing children [5,8,9]. Despite this, SBRIs have been proven to be an integral symptom domain of ASDs [1,7,10].

Recent revisions regarding inclusion of this symptom category have been made in diagnostic instruments including the Diagnostic and Statistical Manual of Mental Disorders-Fifth Edition (DSM-5)[2] and the Autism Diagnostic Observation Schedule-Second Edition (ADOS-2)[11]. The DSM is the primary diagnostic manual used by psychologists and clinicians to diagnose ASDs. Diagnoses considered ASDs in the DSM-IV include Autistic Disorder, Asperger's Disorder, and Pervasive Developmental Disorder Not Otherwise Specified (PDD-NOS). The DSM-IV requires that at least one SBRI symptom be present for a formal diagnosis of Autistic Disorder and Asperger's Disorder. PDD-NOS, often referred to as a milder form of ASD, require social deficits as well as deficits in communication, or SBRIs, or both. The fifth edition of the DSM was released in May, 2013. Clinicians and researchers are currently in a transitional period regarding adoption of the DSM-5 revised criteria. Significant changes to the diagnostic criteria of autism spectrum disorders have been made. No longer are there three separate diagnoses, but one "umbrella" diagnosis of Autism Spectrum Disorder which is further classified with severity level ratings. Included in revisions is the requirement of more SBRIs necessary for a diagnosis of ASD [2].

The ADOS is a standardized observational measure that assesses communication, social interaction, play, and stereotyped behaviors and restricted interests associated with ASDs [12]. It is considered a gold standard assessment tool for the diagnosis of ASDs, both for clinical and research purposes. The ADOS consists of four "modules" or forms, which involve various activities and/or questions based on the child's developmental and verbal abilities. Individual items are scored and transferred to an algorithm form where they are then compared to diagnostic cutoffs indicating a diagnostic classification of "Autism" or "Autism Spectrum." Diagnostic cutoffs for the

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previous edition of the ADOS were used for the communication and social interaction domains. Stereotyped behaviors and restricted interests scores were tallied on the algorithm; however, their scores did not contribute to the diagnostic cutoff. The decision to exclude SBRIs in diagnostic cutoffs was originally made by Lord, Rutter, DiLavore and Risi (2009) because it was believed that the brief administration time of about 45 minutes did not provide enough opportunity to observe these symptoms [12]. However, further research indicated that the inclusion of SBRIs on the algorithm supported stronger predictive validity of the ADOS[13,14,15,16]. Therefore, the decision to include SBRIs in the diagnostic algorithm was revised in the most recent edition of the ADOS, the ADOS-2 [11].

Previous studies have shown diagnostic differences in SBRIs in children with ASD, developmental delays, and typical development [17, 18, 10, 19]. However, limited research has examined diagnostic differences across autism spectrum diagnostic categories on the ADOS and within the DSM-IV (including Autistic Disorder, Asperger's Disorder, and PDD-NOS). Kim and Lord (2010) investigated the prevalence and severity of SBRIs measured on the ADOS across 665 children diagnosed with autism, PDD-NOS, non-spectrum delays, and typically developing [14]. They found that the prevalence of SBRI symptoms was the same for autism and PDD-NOS groups. However, the severity of SBRI symptoms measured by the ADOS was higher for children with autism versus PDD-NOS diagnoses. Findings also indicated that SBRI scores on the ADOS did not change over time for children with autism spectrum and non-spectrum delays. They also assessed what factors were related to having SBRIs, such as non-verbal IQ and age. Non-verbal IQ was more highly related to SBRIs in older children in all groups than younger children with autism. Additionally, certain subtypes of SBRIs including sensory interests, hand and finger mannerisms, and complex motor mannerisms were more related to non-verbal IQ, age, and diagnosis [14].

The purpose of this study is to investigate whether SBRI symptoms are related to diagnostic results specifically when utilizing the ADOS and the DSM-IV. Additionally, it will examine whether specific sub-types of SBRIs are more closely related with certain diagnostic findings. This paper will add to existing literature on the importance of SBRIs as a symptoms category of autism spectrum disorders and their role in diagnosis, as well as factors that may or may not be related to SBRI symptoms, including age and gender.

Materials and Methods

Participants

Participants were 103 males (87%) and 16 females (13%) ages 16 months to 15 years with a mean age of 7 years. Participants had the following DSM-IV diagnoses: 94 (79%) with Autistic Disorder, 9 (7%) with Asperger's Disorder, 14 (12%) with PDD-NOS, and 2 (2%) with non-

Table 1: Age cohorts and gender distribution across 119 children ages 16 months to 15 years.

Age		1	2-3	4-6	7-9	10+	
Total Children in age		3	12	43	38	23	
Gender	Male	2	10	37	36	18	
	Female	1	2	6	2	5	

spectrum developmental delays. Additionally, 91 (76%) participants had an ADOS classification of "Autism", 26 (22%) with "Autism Spectrum", and 2 (2%) with no classification. Regarding module, 67 (57%) participants completed Module 1, 31 (26%) completed Module 2, 17 (14%) completed Module 3, and 4 (3%) completed Module 4.

Procedures

Data was obtained from children participating in a number of on-going research studies conducted at the Johnson Center for Child Health and Development. These studies included administration of two diagnostic assessments, the ADOS and Autism Diagnostic Interview-Revised (ADI-R) for diagnostic purposes only. A DSM-IV diagnosis was determined based on results of these assessments. Language and intellectual assessments were not included as part of these studies, therefore, this information was not available. The ADOSs and ADI-Rs were administered by a single clinical psychologist with research training on both measures. Information collected for each participant included the 1) ADOS SBRI score, 2) ADOS SBRI sub-category scores for sensory behaviors (item D-1), complex motor (item D-2), and restricted interests and unusual behaviors (item D-4), 3) ADOS diagnostic classification, 4) DSM-IV diagnosis, 5) ADOS module, 6) age and 7) gender. In order to adequately examine the relationship between age and SBRI score, the samples of participants were divided into age cohorts comprising: ages 2 and under (3, 3%), 2-3 years (12, 10%), 4-6 years (43, 36%), 7-9 years (38, 32%), and 10 years and above (23, 19%). The Austin Multi-Institutional Review Board approved this study (Table 1).

Assessments

Autism Diagnostic Observation Schedule (ADOS)
The ADOS [12] is a standardized observational assessment for symptoms related to autism in the areas of communication, social interaction, and interests and behaviors. Specific activities are initiated based on the individual's age and verbal abilities. Particular items are transferred to a diagnostic algorithm form in which scores are totaled in the areas of Communication, Social Interaction, and Communication and Social Interaction Total. Derived scores are compared to pre-determined cutoffs that indicate whether a classification of "Autism", "Autism Spectrum", or none, is met.
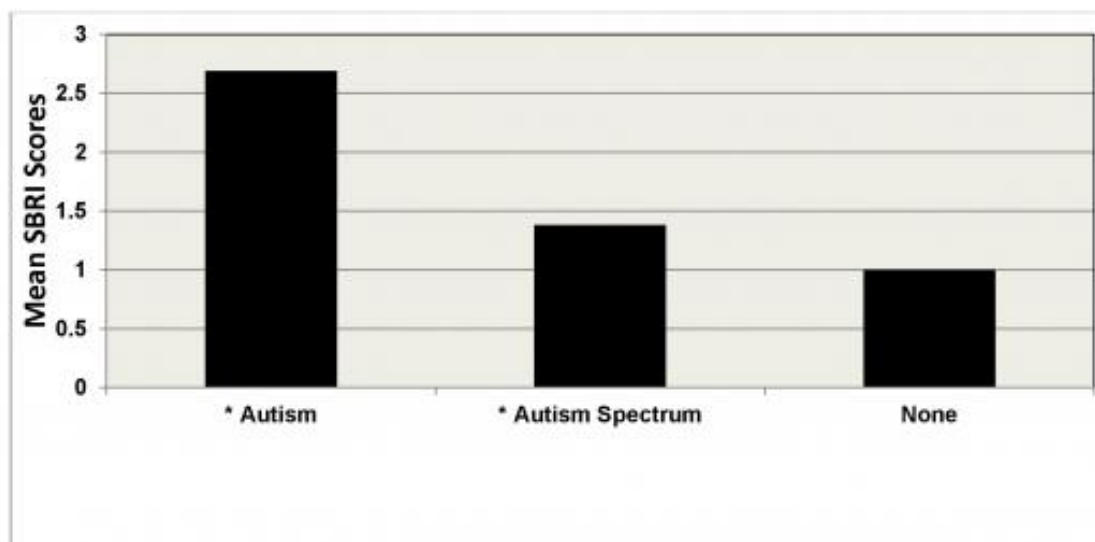
Figure 1: Differences in mean Stereotyped Behaviors and Restricted Interests (SBRI) scores across ADOS classifications in 119 children ages 16 months to fifteen years. Significant difference (*p < 0.001; Autism SEM= 0.15, Autism Spectrum SEM= 0.18).
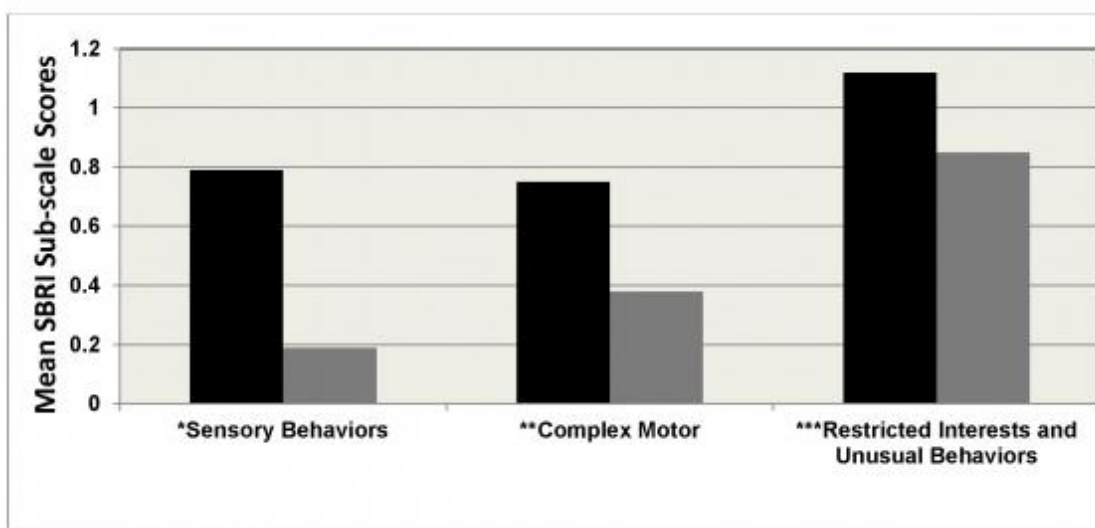

Figure 2: Differences in mean sensory behaviors, complex motor, and restricted interests and unusual behaviors scores across ADOS classifications in 119 children ages 16 months to fifteen years. Significant difference (*p < 0.001; Autism SEM= 0.08, Autism Spectrum SEM= 0.08, **p < 0.05; Autism SEM= 0.09, Autism Spectrum SEM= 0.12, ***p < 0.1; Autism SEM= 0.07, Autism Spectrum SEM= 0.12) Key: Black= Autism; Grey= Autism Spectrum.

Autism Diagnostic Interview-Revised (ADI-R)

The ADI-R [20] is a standardized, comprehensive parent interview that collects information regarding the individual's developmental history as well as history of symptoms in areas associated with autism spectrum disorders. As with the ADOS, a scoring algorithm is used to determine whether or not a child meets particular cutoffs for a diagnostic classification of Autism.

Diagnostic and Statistical Manual of Mental Disorders-Fourth Edition-Text Revised (DSM-IV-TR)

The DSM-IV [1] provides standard criteria for the diagnosis of pervasive developmental disorders. Pervasive developmental disorders (commonly referred to as autism spectrum disorders) within the DSM-IV include Autistic Disorder, Asperger's Disorder, and Pervasive Developmental Disorder-Not Otherwise Specified. The DSM-IV was utilized in this study to provide a "best estimate" diagnosis from information obtained on the ADOS and ADI-R.

Statistical Analyses

An analysis of variance (ANOVA) was used to investigate the relationships between SBRI scores and ADOS classification, DSM-IV diagnosis, age and gender. An analysis of variance was also used to examine relationships between SBRI sub-category scores and ADOS classification, and DSM-IV diagnosis. To examine possible relationships
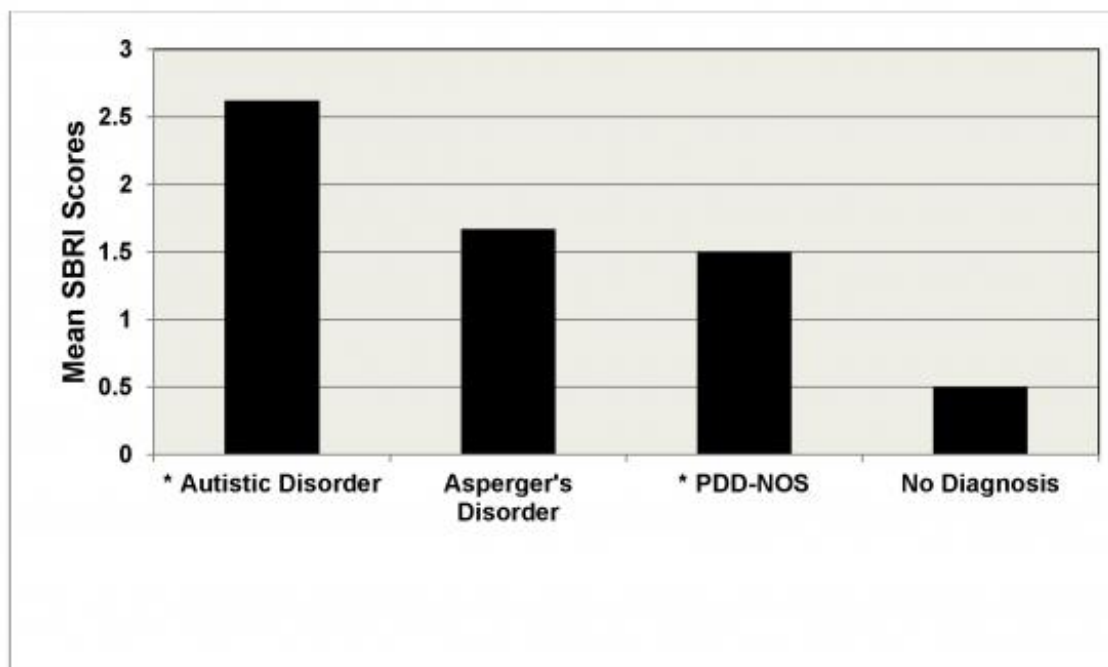
Figure 3: Differences in mean Stereotyped Behaviors and Restricted Interests (SBRI) scores across DSM-IV Diagnoses in 119 children ages 16 months to fifteen years. Significant difference (*$p < 0.01$; Autistic Disorder SEM= 0.15, PDD-NOS SEM= 0.25).

between ADOS Module and ADOS classification and DSM-IV diagnosis, a Fisher's Exact Test was used.

Results

SBRI scores across ADOS classifications
As predicted, mean SBRI scores varied significantly across ADOS diagnostic classifications. Scores were significantly higher with Autism compared to Autism Spectrum ADOS classifications ($p < 0.001$; Autism SEM= 0.15, Autism Spectrum SEM= 0.18) (Figure 1).

Analysis of SBRI subcategories across ADOS diagnostic classifications indicated that mean sensory behaviors and complex motor scores were significantly higher with Autism vs. Autism Spectrum ADOS classifications ($p < 0.001$; Autism SEM= 0.08, Autism Spectrum SEM= 0.08; $p < 0.05$; Autism SEM= 0.09, Autism Spectrum SEM= 0.12). There was a marginally significant difference in mean restricted interests and unusual behaviors scores across classifications ($p < 0.1$, Autism SEM= 0.07, Autism Spectrum SEM= 0.12) (Figure 2).

SBRI scores across DSM-IV diagnoses
Similar results were found when comparing mean SBRI scores across DSM-IV diagnoses, with higher scores associated with more "severe" diagnoses. Mean SBRI scores were significantly higher with DSM-IV diagnoses of Autistic Disorder compared to PDD-NOS ($p < 0.01$; Autistic Disorder SEM= 0.15, PDD-NOS SEM= 0.25) (Figure 3). Further analysis of SBRI sub-categories across diagnoses showed that mean sensory behaviors scores were

significantly higher for Autistic Disorder versus PDD-NOS ($p < 0.05$; Autistic Disorder SEM= 0.08, PDD-NOS SEM= 0.11). Mean complex motor scores were significantly higher for Autistic Disorder versus Asperger's Disorder ($p < 0.05$; Autistic Disorder SEM= 0.09, Asperger's Disorder SEM=0.0). Marginally significant differences in mean restricted interests and unusual behaviors scores were found between PDD-NOS and Asperger's Disorder, with scores slightly higher for Asperger's Disorder ($p < 0.1$; PDD-NOS SEM= 0.16, Asperger's Disorder SEM= 0.17) (Figure 4).

Additional Analyses
Mean SBRI scores did not vary significantly across age or gender. Additionally, the ADOS module administered was not significantly related to either the ADOS classification or DSM-IV diagnosis.

Discussion
The results of this study contribute to existing literature regarding the importance of the SBRI symptom category and its role in the diagnosis of ASDs [1, 7, 17, 18, 10, 19, 14]. We were able to identify diagnostic differences in severity of SBRIs measured on the ADOS across 119 children. These results extend the findings of previous studies demonstrating that SBRIs reliably occur in children diagnosed with ASDs [1, 7, 14]. Furthermore, results show that SBRI symptoms are related to diagnostic results using two primary diagnostic tools, the ADOS, and the DSM-IV, thus supporting diagnostic validity of this important subset of
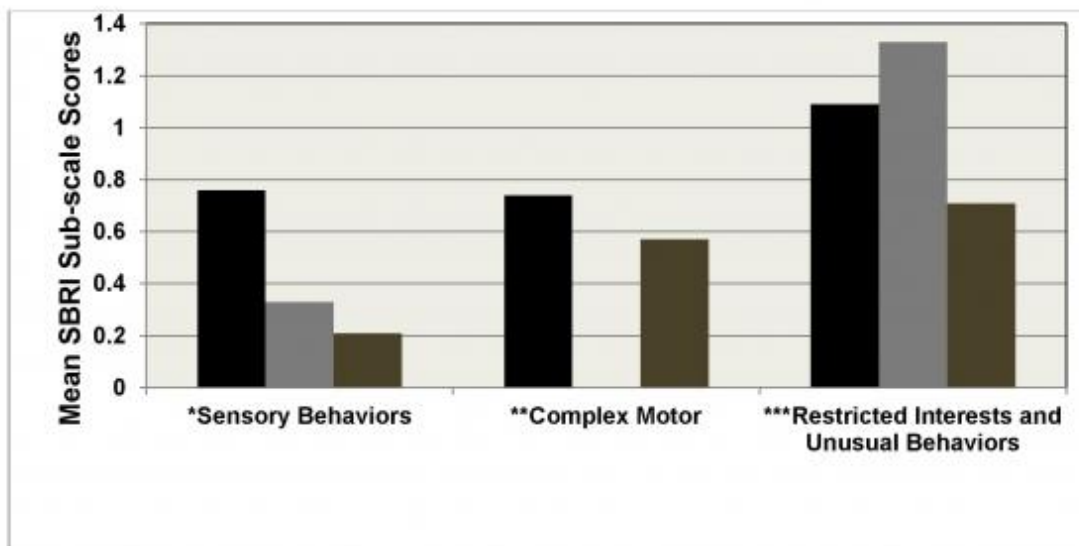
Figure 4: Differences in mean sensory behaviors, complex motor, and restricted interests and unusual behaviors scores across DSM-IV Diagnoses in 119 children ages 16 months to fifteen years. Significant difference (*p < 0.05; Autistic Disorder SEM= 0.08, PDD-NOS SEM= 0.11, **p < 0.05; Autistic Disorder SEM= 0.09, Asperger's Disorder SEM=0.0, *** p < 0.1; PDD-NOS SEM= 0.16, and Asperger's Disorder SEM= 0.17) Key: Black= Autistic Disorder; Light Grey= Asperger's Disorder; Dark Grey= PDD-NOS.

symptoms. Results also support that the gold standard diagnostic tool, the ADOS, provides a reliable opportunity to observe these symptoms [13]. This study also indicates the consistency in presence of SBRI symptoms across age and gender. Certain subtypes of SBRI symptoms were shown to be more closely related to diagnostic results. Higher scores in the areas of sensory behaviors and complex motor mannerisms were associated with more severe diagnostic categories. This is consistent with findings of Kim and Lord, 2010 [14]. Restricted interests and repetitive behaviors appeared slightly more consistent across diagnostic categories suggesting that both sensory behaviors and complex motor mannerisms might help support diagnostic differentiation.

Results of this study are of particular significance due to the evolving diagnostic frameworks used to diagnose ASDs. These include the recent revisions of the ADOS and DSM, both highly utilized for diagnosis. The increased emphasis on the SBRI symptom area in both of these tools is supported by our overall results indicating these symptoms are highly related to diagnostic conclusions. Symptoms in this category were present for all ASD diagnostic categories, including Autism Spectrum on the ADOS, and PDD-NOS diagnoses in the DSM-IV. This suggests that symptoms in this domain should be a required criteria for all ASDs, and therefore the newly, more broadly defined "Autism Spectrum Disorder" diagnosis in the DSM-5 [2].

Limitations and Future Directions
We compared SBRI symptoms across diagnostic results for children who already had, or were suspected of having, an ASD diagnosis. However, we did not investigate SBRI symptoms across groups of children who were typically developing or had other developmental disorder diagnoses. It would be useful for future studies to utilize a control group of typically developing children, as well as samples of children with other developmental disorders, to further support the validity of these results. An additional area of limitation is the Module analysis. The majority of participants completed Module 1; however, a number of subjects completed Modules 2-4. A weakness of this study is that the Module and number of items per Module was not controlled for. Certain sample sizes, including particular diagnostic groups (i.e. Asperger's Disorder) that were used for comparisons, had a rather small number of subjects. This made some analyses less reliable. Another limitation of the present study is that we did not examine subsets of SBRI symptoms across age or gender. It is recommended that this be examined to ascertain if certain subtypes of SBRI's, such as sensory behaviors or repetitive motor mannerisms, vary according to those variables.

Although this study extended previous studies that support that SBRI symptoms can be observed during the short administration time of the ADOS, it is unlikely that all SBRI symptoms can be reliably observed during this brief period. For example, some symptoms such as compulsive or ritualized behaviors may occur exclusively in certain contexts outside of the testing environment. Therefore, it is important that supplemental information is collected from caregivers, such as through the ADI-R, to fully assess these symptoms in regards to frequency, duration, and severity.
It would be interesting to expand upon results of this study using the more recently released ADOS-2, particularly because of the further emphasis placed on SBRI symptoms. The ADOS and ADI-R are often viewed as companion assessments and are both considered current gold standard diagnostic measures. Future studies should

investigate SBRI symptoms on both the ADOS-2 and ADI-R and how they relate to diagnostic findings. Furthermore, relationships between SBRI symptoms and diagnostic findings utilizing the DSM-5 should be explored.

Conclusion

The importance of "best practice" in the assessment of autism spectrum disorders is critical during this time of diagnostic transition. Best practice includes the use of reliable and valid tools such as the ADOS-2 and ADI-R in order to thoroughly assess symptom domains, support diagnostic conclusions, and aid in treatment planning. This study supports best practice by adding to the literature supporting the significance of SBRI symptoms as they relate to diagnosis.

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