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Brief Report

The relationship between subtypes of repetitive behaviors and anxiety in children with autism spectrum disorder

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ABSTRACT

Background: Many children with autism spectrum disorder (ASD) experience comorbid anxiety problems. Research suggests that anxiety symptoms are related to restricted and repetitive behaviors (RRBs) in general, and insistence on sameness behaviors, in particular. However, the extent to which other subtypes of RRBs are related to anxiety is less clear.

Method: This study evaluated the relationship between anxiety symptoms and subtypes of restricted and repetitive behaviors (RRBs) using the Repetitive Behavior Scale-Revised (RBS-R) in a large sample of children ages 68 months to 18 years-old with autism spectrum disorder (ASD) from the Simons Simplex Collection (N = 2093).

Results: An exploratory factor analysis suggested a four-factor solution to the RBS-R. All four factors were significantly correlated with each other and with anxiety symptoms ($ps \leq 0.001$). A hierarchical regression controlling for age and IQ indicated that the Sameness and Self-Injury factors accounted for unique variance in anxiety. There were no interactions between IQ and any of the RRB factors in predicting anxiety.

Conclusions: Results suggest that researchers and clinicians should consider the role of anxiety when understanding and treating RRBs in children with ASD. High endorsement of items that load on the Sameness and Self-Injury subscales of the RBS-R might be used to identify children who are at risk for anxiety but who may not show overt symptoms.

1. Introduction

Recent research suggests at least 40% of children with autism spectrum disorder (ASD) meet diagnostic criteria for an anxiety disorder (van Steensel, Bögels, & Perrin, 2011), and many more experience anxiety symptoms at a subclinical level (Rodgers, Glod, Connolly, & Mcconachie, 2012). Although anxiety is positively related to age and intellectual ability in individuals with ASD (Gotham, Brunwasser, & Lord, 2015), its relationship with core symptoms of ASD is not well understood. Several studies suggest anxiety symptoms are related to restricted and repetitive behaviors (RRBs; Factor, Condy, Farley, & Scarpa, 2016; Gotham et al., 2013; Lidstone et al., 2014; Rodgers, Riby, Janes, Connolly, & Mcconachie, 2012). This finding is consistent with the presence of repetitive behaviors in individuals with Obsessive-Compulsive and Related Disorders (OCRD; American Psychiatric Association, 2013), which are thought to be functionally related to anxiety as a way to alleviate distress caused by obsessions (Jiujias, Kelley, & Hall, 2017; Starcevic et al., 2011). However, the functional relationship between anxiety and RRBs in ASD is less clear (Joosten, Bundy, & Einfeld, 2008).

Studies that have examined subtypes of RRBs have typically found that insistence on sameness (IS) behaviors are associated with

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anxiety, while repetitive sensory motor (RSM) behaviors are not (Factor et al., 2016; Gotham et al., 2013; Lidstone et al., 2014; Rodgers, Glod et al., 2012). However, at least two studies have found a relationship between RSM behaviors and anxiety (Joyce, Honey, Leekam, Barrett, & Rodgers, 2017; Wigham, Rodgers, South, McConachie, & Freeston, 2015), suggesting that the unique relationship between IS and anxiety is not entirely clear-cut.

The differential association between these two RRB subtypes and anxiety symptoms suggests RRB subtypes may serve different functions in individuals with ASD compared to other populations. However, there are some limitations in this work. First, there is disagreement as to the structure of RRBs in individuals with ASD (Joseph, Thurm, Farmer, & Shumway, 2013). The two subtypes of RRBs that have been examined in relation to anxiety have been identified primarily by factor analyses of the Autism Diagnostic Inventory-Revised (ADI-R; Rutter, Le Couteur, & Lord, 2003) and the Repetitive Behaviour Questionnaire-2 (RBQ-2; Joyce et al., 2017; Lidstone et al., 2014; Rodgers, Riby et al., 2012). The ADI-R has only a few items that measure RRBs and may not capture the full range of RRBs. Therefore, it may fail to distinguish between meaningful subtypes of higher-order RRBs in individuals with ASD: IS behaviors, compulsive/ritualized behaviors, and circumscribed/restricted interests (e.g., Leekam, Prior, & Uljarevic, 2011). It is quite possible that these behaviors have differential relationships with anxiety.

In addition, neither the ADI-R nor the RBQ-2 measure self-injurious behavior (SIB) as a distinct subtype of RRB, despite evidence that SIB and stereotyped behaviors are topographically similar and related over time in children with developmental disabilities (Rojahn, Barnard-Brak, Medeiros, & Schroeder, 2016). SIB may be an important subtype of RRB to consider in relation to anxiety, given the presence of repetitive SIBs such as hair-pulling and skin picking in OCD spectrum disorders, and the recent finding of an association between anxiety disorders and SIBs in youth with ASD (Kerns et al., 2015).

The Repetitive Behavior Scale-Revised (RBS-R; Bodfish, Symons, Parker, & Lewis, 2000) is a caregiver questionnaire that measures the breadth of RRBs that occur in individuals with ASD, including SIB. Recent factor analytic research suggests that it is comprised of two to five factors (Bishop et al., 2013; Georgiades, Papageorgiou, & Anagnostou, 2010; Lam & Aman, 2006; Mirenda et al., 2010). One recent study examined the relationship between subtypes of RRBs and anxiety symptoms in 72, 5- to 17-year-old children with ASD using the RBS-R (Stratis & Lecavalier, 2013) and the factor structure identified by Lam and Aman (2006). There was a bivariate association between all five subtypes of RRBs (Rituals/Sameness Behavior, Compulsive Behavior, Stereotypic Behavior, SIB, and Restricted Interests) and anxiety symptoms. However, only Rituals/Sameness Behavior was uniquely associated with anxiety. SIB was not an independent predictor of anxiety; however, the relationship was moderated by level of adaptive functioning. Specifically, SIB was a negative predictor of anxiety in children whose adaptive skills were one standard deviation below the sample mean, a positive predictor in children whose skills were one standard deviation above the mean, and not a predictor for children at the mean. This finding suggests that adaptive and/or intellectual functioning may interact with specific RRB subtypes, particularly SIB, in predicting anxiety in youth with ASD. This possibility is further supported by research suggesting that patterns of age-related reductions in specific subtypes of RRBs are dependent on intellectual functioning, with less reduction in stereotyped behavior compared to other types of RRBs among individuals with ASD and co-morbid intellectual disability (Esbensen, Seltzer, Lam, & Bodfish, 2009).

In this study, we used the RBS-R to further examine the relationship between specific subtypes of RRBs and anxiety symptoms in a large, well-characterized sample of youth with ASD. Given Stratis and Lecavalier's (2013) finding of an interaction between SIB and adaptive functioning in predicting anxiety, we also examined whether any relationships between RRB subtypes and anxiety were moderated by IQ. Given some disagreement regarding the preferred factor structure of the RBS-R (Joseph et al., 2013), we ran an exploratory factor analysis (EFA) of the RBS-R to identify meaningful subtypes of RRBs prior to examining their relationship to anxiety. Based on previous research, we hypothesized that we would observe a positive relationship between anxiety problems and ritualistic/sameness behaviors, but not stereotypic/RSM behavior. A clear hypothesis regarding the relationship between anxiety and compulsive/ritualized behaviors, circumscribed/restricted interests, and self-injury was not identified.

2. Method

2.1. Participants

The data in this study were collected through the Simons Simplex Collection (SSC). The SSC is a permanent repository of genetic and behavioral samples from 2600 simplex families. Each family included a child with ASD between the ages of 4 and 18 years, their parents, and typically developing siblings (Fischbach & Lord, 2010). Data collection for the SSC occurred at 12 university-affiliated research clinics in North America; recruitment was conducted through and partnerships with local service providers, parent and advocacy groups, web postings, and radio and television ads (Fischbach & Lord, 2010). A complete description of the SSC can be accessed from https://sfari.org/resources/autism-cohorts/simons-simplex-collection.

We analyzed data from all children age 68 months to 18 years-old with ASD in the SSC (N = 2093) who had data for the RBS-R and Child Behavior Checklist, School-Age version (CBCL/6-18; see Table 1 for participant demographics). All participants were diagnosed using DSM-IV-TR criteria, and met criteria for ASD on the Autism Diagnostic Observation Schedule (Lord, Rutter, DiLavore, & Risi, 1999) and Autism Diagnostic Interview-Revised.

2.2. Measures

2.2.1. Repetitive behavior scales – revised (RBS-R; Bodfish et al., 2000)

The RBS-R is a caregiver-report questionnaire that measures a wide range of RRBs in individuals with ASD. It includes 43 items

Table 1
Demographic Information for Study Participants.

	Percent / Mean (SD)	Range
Sex		
Female	13.4	
Male	86.6	
Race		
African American	3.8	
Asian	3.9	
More than one race	7.5	
Native American	0.2	
Native Hawaiian	0.1	
Not specified	0.9	
Other	4.4	
White	79.1	
Age (months)	123.50 (37.52)	68–216
Full scale IQ	81.55 (28.48)	7–167
CBCL Anxiety T-Score	61.69 (8.71)	50–80
RBS-R Factors		
Rituals/Sameness Factor	13.17 (8.42)	0–40
Compulsive Behavior Factor	2.93 (3.08)	0–18
Stereotypic Behavior Factor	5.40 (4.23)	0–24
SIB Factor	2.01 (2.70)	0–21

rated on a four-point scale from “behavior does not occur” to “behavior occurs and is a severe problem.” The published measure includes six conceptually grouped subscales of RRBs: Stereotyped behavior, Self-Injurious behavior, Compulsive Behavior, Ritualistic behavior, Sameness behavior, and Restricted behavior.

2.2.2. Child behavior checklist (CBCL; Achenbach & Rescorla, 2001)

The CBCL is a widely used caregiver-report measure that identifies common behavior problems in children and adolescents aged 6–18. The CBCL is comprised of 118 items that are rated on a 3-point scale from “Not true” to “Very true or often true.” The Anxiety Problems T-score, which is normed to a mean of 50 and standard deviation of 10, was used to measure anxiety symptoms.

2.2.3. Intellectual quotient (IQ)

Full scale IQ scores were derived from the Differential Abilities Scales, 2nd Edition (DAS-II; Elliott, 2007), the Mullen Scales of Early Learning (MSEL; Mullen, 1995), the Wechsler Intelligence Scale for Children, 4th Edition (Wechsler, 2004), or the Wechsler Abbreviated Scale of Intelligence (WASI: Wechsler, 1999), based on an established test hierarchy. When available, standard scores were used. In cases in which a norm-referenced standard score could not be calculated due to the youth’s age or developmental level, a ratio IQ was derived by dividing the youth’s mental age equivalent by chronological age and multiplying by 100. For this sample, 87.00% of youth were administered the DAS-II, 7.22% the MSEL, and 5.69% the WISC-4 or WASI.

2.3. Data analysis

We conducted an EFA (with principal axis factoring and promax rotation) of the RBS-R to determine which subscales to include in the regression model. We used eigenvalues above 1.0, an examination of the scree plot, and interpretability to guide the selection of a factor solution. We used Lam and Aman’s (2006) decision rule for item loadings: items comprised a factor if they loaded 0.35 or higher and if this loading was at least 0.10 higher than the loading on any other factor.

We used Pearson correlations to examine the relationship between the RRB subtypes, CBCL Anxiety Problem T-score, full scale IQ, and age. Significance level was set at $p \leq 0.001$ due to the large sample size. We conducted a hierarchical regression analysis to examine whether any of the RRB subtypes were unique predictors of anxiety after controlling for age and IQ, and whether any of the RRB subtypes interacted with IQ in predicting anxiety. All predictors were mean-centered. Chronological age and IQ were entered in the first step, the four RRB subtypes in the second step, and the interaction terms for IQ and each RRB subtype in the third step.

3. Results

3.1. Exploratory factor analysis of the RBS-R

Examination of the factor solutions for the RBS-R suggested a two-, three-, four-, or five-factor solution. These criteria suggested two, three, four, and five factor solutions, consistent with Lam and Aman (2006) and Bishop et al. (2013). Both Lam and Aman (using an independent sample) and Bishop et al. (using a subset of the current sample) adopted a five-factor solution. Like Bishop et al., we found that only 2 items loaded on the fifth factor (Restricted Interests). Given the recommendation of a minimum of 3 items to comprise a factor (e.g., Costello & Osborne, 2005), we selected the four-factor solution. Eight items from the RBS-R were omitted

Table 2
Factor Loadings for EFA of Repetitive Behavior Scale - Revised.

Original RBS-R Items:	Factor 1: Rituals/ Sameness	Factor 2: Stereotypic Behavior	Factor 3: Compulsive Behavior	Factor 4: SIB
Stereotyped Behavior Subscale				
1. Whole Body	0.01	**0.54**	−0.06	0.01
2. Head	−0.01	**0.40**	0.02	0.06
3. Hand/Finger	−0.05	**0.68**	−0.06	−0.08
4. Locomotion	−0.01	**0.61**	−0.07	0.06
5. Object Usage	−0.03	**0.55**	0.03	0.10
6. Sensory	0.07	**0.48**	0.06	0.09
Self-Injurious Behavior Subscale				
7. Hits self with Body Part	0.02	0.10	−0.08	**0.64**
8. Hits Self Against Surface or Object	0.04	0.03	−0.02	**0.64**
9. Hits Self with Object	0.06	−0.01	−0.07	**0.64**
10. Bites Self	−0.02	0.08	0.01	**0.38**
11. Pulls	−0.03	−0.05	0.09	**0.48**
12. Rubs or Scratches Self	−0.04	−0.01	0.13	**0.44**
14. Skin Picking	0.01	−0.12	0.16	**0.35**
Compulsive Behavior Subscale				
15. Arranging/Ordering	0.06	−0.05	**0.61**	0.04
16. Completeness	−0.03	0.01	**0.70**	0.01
17. Washing/Cleaning	−0.11	−0.01	**0.53**	0.10
18. Checking	−0.09	0.01	**0.58**	−0.01
19. Counting	0.06	0.05	**0.43**	−0.02
21. Repeating	0.17	0.13	**0.42**	−0.02
Ritualistic Behavior Subscale				
23. Eating/Mealtime	**0.35**	0.06	0.17	−0.03
26. Travel/Transportation	**0.48**	0.01	0.22	−0.02
27. Play/Leisure	**0.55**	−0.02	0.13	−0.04
28. Communication/Social Interactions	**0.5**	0.04	0.03	−0.08
Sameness Behavior Subscale				
30. Objects to Visiting New Places	**0.61**	0.00	−0.11	0.02
31. Becomes Upset if Interrupted	**0.77**	0.00	−0.19	0.06
33. Insists on Sitting at the Same Place	**0.6**	−0.13	0.10	0.04
34. Dislikes Changes in Appearance/Behavior of Others	**0.57**	−0.04	0.12	0.00
36. Likes Same Movie/Piece of Music Played Continually	**0.37**	0.22	0.07	−0.04
37. Resists Changing Activities; Difficulty with Transitions	**0.88**	−0.01	−0.30	0.07
38. Insist on Same Routine/Schedule Everyday	**0.84**	−0.01	−0.08	0.00
39. Insists that Specific Things Take Place at Specific Times	**0.76**	−0.06	0.01	0.03
Restricted Behavior Subscale				
40. Fascination, Preoccupation with One Subject/Activity	**0.49**	0.17	−0.07	−0.07
41. Strongly attached to One Specific Object	**0.41**	0.15	0.10	−0.02
42. Preoccupation with Part(s) of Object Rather than the Whole Object	0.16	**0.42**	0.16	−0.05
43. Fascination, Preoccupation with Movement/Things that Move	0.09	**0.48**	0.14	−0.10

Note: Bold indicates that item loaded most heavily and corresponds to the subscale scoring (.35) and items that failed to load with any single factor were omitted.

because they did not meet the decision rule for item loadings (see Table 2). We named the final four factors Rituals/Sameness (14 items; α = .88), Compulsive (6 items; α = .75), Stereotypic (8 items; α = .78), and SIB (7 items; α = .71). Cronbach's alphas for each factor were all ≥.70, suggesting adequate internal consistency. These factors explained 23.56%, 5.00%, 3.08%, and 2.19% of the total variance of the measure, respectively.

3.2. Relationship between RRB subtypes and anxiety problems

All four subtypes of RRBs were moderately to strongly positively correlated with each other (*r*s = .28–.58). Each RRB subtype was also positively correlated with anxiety problems, although effect sizes varied from weak to moderate (Table 3). Age and full-scale IQ were weakly positively correlated with anxiety problems, and negatively correlated with several of the RRB subtypes, although effect sizes were generally small.

After controlling for age and IQ and other RRB factors, Rituals/Sameness and SIB factors were each independent predictors of anxiety problems. None of the interaction terms were significant (see Table 4).

Table 3
Bivariate Correlations Between Study Variables.

	Age	Full Scale IQ	CBCL Anxiety T-Score	Rituals/ Sameness Factor	Compulsive Factor	Stereotypic Factor
Age						
Full Scale IQ	−.02					
CBCL Anxiety T-Score	.09*	.11*				
Rituals/Sameness Factor	−.05	−.04	.38*			
Compulsive Factor	−.06	−.23*	.17*	.58*		
Stereotypic Factor	−.18*	−.31*	.16*	.48*	.47*	
SIB Factor	.02	−.16*	.22*	.33*	.28*	.39*

* $p < .001$.

Table 4
Summary of Hierarchical Regression Analysis for Variables Predicting CBCL Anxiety T-Scores.

N = 2088	Model 1		Model 2		Model 3	
Variable	***β***	***t***	***β***	***t***	***β***	***t***
Age	.09	4.15*	.11	5.40*	.11	5.49*
Full Scale IQ	.11	5.15*	.14	6.69*	.15	4.60*
Rituals/Sameness Factor			.36	13.60*	.35	13.34*
Compulsive Factor			−.04	−1.56	−.04	−1.42
Stereotypic Factor			.03	1.08	.03	.97
SIB Factor			.12	5.42*	.12	5.50*
Rituals/Sameness Factor X IQ					.04	1.59
Stereotypic Factor X IQ					−.01	−.34
Compulsive Factor X IQ					−.01	−.23
SIB Factor X IQ					.03	1.12
R^2	.02		.18		.18	
F Change	21.39*		103.04*		1.32	

* $p < .001$.

4. Discussion

The results of this study provide further evidence for a relationship between restricted and repetitive behaviors and anxiety in youth with ASD. Using the RBS-R and a large, well-characterized sample, we were able to examine the differential relationships between anxiety and a broader range of RRB subtypes than previous research (Factor et al., 2016; Gotham et al., 2013; Lidstone et al., 2014; Rodgers, Glod et al., 2012). Although RRBs, anxiety and IQ have already been analyzed with this sample through the ADI-R (Gotham et al., 2013), we have used the RBS-R because it is a more comprehensive measure of RRBs.

The EFA revealed a 4-factor solution for the RBS-R, comprised of Rituals/Sameness, Compulsive Behavior, Stereotypic Behavior, and SIB factors. Consistent with previous factor analyses, items from the original Ritualistic and Sameness subscales of the RBS-R loaded on a single factor (Bishop et al., 2013; Lam & Aman, 2006). It is unclear whether the lack of a robust Restricted/Circumscribed Interests factor is due to the underrepresentation of restricted and circumscribed interests on the RBS-R or whether they are not distinct from other IS behaviors in youth with ASD. We checked the current analysis with the factors from Bishop et al. (2013) and they were virtually identical, so we retained the current analysis due to the lack of consensus on RRB factors using the RBS-R. Studies using the RBQ-2 and ADI-R have also found that circumscribed interest items load onto a Sameness factor (Bishop et al., 2013; Gotham et al., 2013; Honey, McConachie, Turner, & Rodgers, 2012). It has been also suggested that both IS behaviors and restricted/circumscribed interests may be coping responses to anxiety in children with ASD (Spiker, Lin, Van Dyke, & Wood, 2012). It is important for future research to consider additional ways of measuring this subtype of RRB to determine whether it is distinct from IS behaviors, and if so, how it relates to anxiety in individuals with ASD.

Our study provides additional support for a significant relationship between anxiety and IS in ASD (Factor et al., 2016; Gotham et al., 2013; Lidstone et al., 2014; Rodgers, Glod et al., 2012). This relationship may indicate that ritualistic and IS behaviors serve as a way of coping with anxiety by attempting to control one's environment (Rodgers, Glod et al., 2012). Indeed, studies have found the association between anxiety and IS behaviors in children with ASD are mediated by sensory sensitivity (Black, Stevenson, & Segers, 2017) and sensory avoidance (Lidstone et al., 2014), suggesting IS behaviors reduce anxiety through avoidance of sensory stimulation. Alternatively, it has also been proposed that intolerance of uncertainty may drive the association between anxiety and IS in children with ASD, such that children with a low tolerance for uncertainty adhere to set routines to avoid anxiety (Boulter, Freeston, South, & Rodgers, 2014; Wigham et al., 2015). This latter possibility is consistent with recent qualitative findings from young people with ASD, who described engaging in routines as a means of stopping or avoiding uncertainty (Joyce et al., 2017). Both propositions suggest important clinical targets for reducing anxiety and IS behaviors in individuals with ASD (Rodgers et al., 2017)

Like other studies (Factor et al., 2016; Lidstone et al., 2014; Rodgers, Glod et al., 2012), we did not find a significant relationship between the Stereotypic factor and anxiety, over and above the relationship with other RRBs. This finding suggests that RSM

behaviors do not have the same relationship with anxiety as IS behaviors. It has been long been suggested that RSM behaviors serve to regulate arousal by providing stimulation through sensation seeking during underarousal (Lovaas, Newsom, & Hickman, 1987; Rincover, Newsom, & Carr, 1979; Wieseler, Hanson, Chamberlain, & Thompson, 1985); although more recent research suggests that they may also serve to avoid sensory input and reduce anxiety, as well as to gain attention, tangibles, and escape demands (Joosten & Bundy, 2010; Joosten et al., 2008; Joyce et al., 2017). Thus, a failure to find an association between RSM behaviors and anxiety in this and other studies may be due the many functions that RSM serve.

We did not find an independent relationship between the Compulsive factor and anxiety after controlling for other RRB factors, consistent with some prior research in ASD (Stratis & Lecavalier, 2013). This stands in contrast with what is known about OCD, for which a key function of compulsions is to alleviate anxiety caused by obsessions (Starcevic et al., 2011). These findings suggest that compulsive behaviors may not serve the same function in individuals with ASD as individuals without ASD (Jiujias et al., 2017). Further research is needed to explore the potential functions of compulsive RRBs in ASD.

Although limited work has examined the relationship between SIB and anxiety in ASD, our results extend research finding a significant relationship between these constructs in ASD (Kerns et al., 2015), and other disorders such as trichotillomania and skin picking (Grant, Redden, Leppink, & Chamberlain, 2017; Hayes, Storch, & Berlanga, 2009). Unlike Stratis and Lecavalier (2013), we observed a direct relationship between the SIB factor and anxiety, rather than a moderated effect. Our ability to find a significant interaction between SIB and IQ may have been limited by the limited number of participants with an intellectual disability in our sample. Thus, it is possible that the relationship between SIB and anxiety is higher among youth with lower IQ. However, our results suggest that SIB may be related to anxiety across the adaptive spectrum in individuals with ASD. It has been proposed that SIB may serve to alleviate anxiety associated with IS in individuals with ASD (Duerden et al., 2012). However, our finding of a significant association between Self-Injury and anxiety after controlling for Sameness suggests that IS behaviors cannot fully account for the relationship. Future research should further explore anxiety in relation to self-injurious RRBs in ASD specifically.

The current study has several limitations. First, although rituals/sameness and SIB were significantly related to anxiety in this sample, bivariate correlations were small to moderate, and the RRB factors explained a limited amount of variance in anxiety in the regression model (18%). This suggests that a number of additional things contribute to anxiety in youth with ASD. In addition, this sample was limited to school aged children from families without complicated medical histories, and many participants had average to above-average cognitive abilities. This information should be considered when comparing findings of this study with other samples of individuals with ASD. Further, both the CBCL and the RBS-R are parent-report measures, which can lead to shared method variance. Additional research using multiple informants, including teacher- and child-reported anxiety symptoms, and behavioral observations of RRBs would increase confidence in these findings. Finally, all measures were collected concurrently. Longitudinal data are needed to better understand the functional relationship between different RRB subtypes and anxiety and how they may influence each other over time.

5. Implications

In summary, our data suggest researchers and clinicians should consider the role of anxiety when understanding and treating RRBs in children with ASD. Specifically, high endorsement of items that load on the Rituals/Sameness and SIB subscales of the RBS-R might be used to identify children who are at risk for anxiety.

Conflict of interest

None of the authors have a conflict of interest.

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