Title page

Manuscript Title: Relationship between Repetitive Behaviour and Fear across Normative Development, Autism Spectrum Disorder, and Down Syndrome

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Running Title: Relationship between Insistence on Sameness and Fear

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Lay Abstract

The present study had two aims: First, we compare levels of repetitive behaviours (RRB) across two groups of typically-developing children, children with Autism Spectrum Disorder (ASD), and children with Down syndrome (DS). Second, we explore the relationship between fear and repetitive behaviours in these four groups of children. Parents of 41 children with ASD, 38 children with DS, 45 typically developing (TD) children matched on the mental age (MA) to the DS group, and 42 chronological age (CA) matched TD children completed measures of RRB and fear. Results show that children with ASD had the highest RRB levels followed by DS, TD MA, and TD CA children. In the ASD group, higher levels of fear were related to higher RRB levels. Similar associations between RRB and fear were found among DS and TD MA children, but not in older TD CA children. This study provided evidence of the fear-RRB association in children with ASD, DS, and two groups of TD children.

Scientific Abstract

The present study had two aims: firstly to compare levels of repetitive behaviours (RRB) across two groups of typically-developing children, and two disorders: Autism Spectrum Disorder (ASD) and Down syndrome (DS), and secondly to explore the relationship between fear and repetitive behaviours in these four groups. Parents of 41 offspring with ASD (M_{age}= 123.39 months, SD_{age}= 27.67), 38 offspring with DS (M_{age}= 125.37 months, SD_{age}= 45.71), 45 typically developing children matched to the mental age (MA) of the DS group (TD; M_{age}= 51.13 months, SD_{age}= 22.1), and 42 chronological age (CA; M_{age}= 117.93 months, SD_{age}= 22.91) matched TD children, completed measures of RRB and fear. ANOVAs revealed differences across the four groups on the RRB subscale scores: "Just Right" $F(3,162)= 16.62$, $p< .001$; Rigid Routines $F(3,162)= 52.76$, $p< .001$; Sensory behaviours $F(3,162)= 23.26$, $p< .001$. Post-hoc comparisons revealed that children with ASD had the highest RRB levels followed by DS, TD MA and TD CA children. In children with ASD, higher levels of fear were related to higher RRB levels. Similar, albeit less strong, patterns of associations was found among DS and TD MA children but not in older TD CA children. This study provided evidence of a fear-RRB association in children with ASD, DS, and two groups of TD children.

Key Words: Repetitive Behaviour, Fear, Autism, Down Syndrome, Typical Development

Introduction

Restricted and repetitive behaviour (RRB) is a heterogeneous group of behaviours ranging from simple tics and sensory-motor stereotypies, (referred to as repetitive sensory motor behaviours; RSM), to more complex forms of rigid, routinized and ritualistic behaviours that may be described as rigidity/insistence on sameness (RIS) (Berkson & Tupa, 2000; Leekam, Prior, & Uljarević, 2011; Lewis & Kim, 2009). From the original description provided by Kanner (1943) up to the latest incarnation of international diagnostic criteria (DSM-5: APA, 2013), RRB have been considered a defining feature of Autism Spectrum Disorder (ASD).

RRB are not unique to ASD, however. They are seen throughout normative developmentand across a range of neuropsychiatric and neurodevelopmental disorders including Tourette's syndrome, obsessive-compulsive disorder (OCD) and intellectual disabilities (ID) (Langen, Durston, Kas, Van Engeland & Staal, 2011), where RRB constitute a major barrier to normal functioning. Indeed in ASD, RRB negatively impact learning and social adaptation, and present a significant source of stress and management challenge for parents (Leekam, Prior, & Uljarević, 2011). Unlike atypical RRB, typical RRB are transitory and play an adaptive role in development. The mechanisms underlying the transition from typical into atypical RRB are not well understood, however (Evans et al., 2015). Thus the development of targeted early interventions geared to RRB is limited (Grahame et al., 2015).

In normative development, some aspects of RRB, such as rigidity and insistence on sameness (RIS), serve to constrain the unpredictability of the environment and to ward off fear and anxiety, thus serving as an early form of emotion and arousal regulation (Evans et al., 1999; Gesell et al., 1974; Leekam et al., 2011; Zohar & Felz, 2001). In young children, normative RIS are likely to occur at times of transition (such as bedtime), and are accompanied by normative fear (Evans, Gray, & Leckman, 1999). Normative fear and

anxiety, such fear of the dark, and separation/stranger anxiety, occurs in the context of typical development. This aspect of typical development may be distinguished from phobia and clinical anxiety based on the fact that they are largely adaptive mechanism for coping with particular developmental challenged; occur at specific stages of development; and are transitory in nature (Brooker, Buss, Lemery-Chalfant, Aksan, Davidson, Goldsmith, 2014; Evans et al., 1999; Gullone, 2000). The link between normal childhood RIS and fear/anxiety parallels findings in ASD (Lidstone, Uljarević, et al., 2014; Rodgers, Glod, Connolly, & McConachie, 2012), and in obsessive compulsive disorder (OCD), where particular behaviours are performed for purposes of anxiety reduction (Mataix-Cols, Rosario-Campos, & Leckman, 2005). However, it is not clear why and how the RRB-Fear/anxiety link transitions from adaptive to maladaptive forms that reinforce anxiety in clinical conditions.

Cross-sectional questionnaire studies suggest that RIS begin around the age of 24 months, peak around 72 months, and steadily wane afterwards (Arnott et al., 2010; Cevikaslan et al., 2013; Evans et al., 1997; Leekam et al., 2007). A similar timeline has been observed for normative fear (Evans et al., 1999), which are in turn, concomitant to developments in executive functioning (EF) skills, most importantly response inhibition and set-shifting abilities (Zelazo et al., 2003). Therefore, it seems that as cognitive and emotion-related strategies for managing and regulating arousal and fear develop, children become less reliant on RIS.

In ASD, where problems in EF (Hill, 2004; Robinson, Goddard, Dritschel, Wisley, & Howlin, 2009) and emotion regulation (Mazefsky et al., 2013) have been noted, RIS continue to serve to reduce fear/anxiety. Although RIS may be an efficient short-term strategy, given their developmentally inappropriate nature, they will likely reinforce and sustain fear and anxiety long term (Rodgers et al., 2012). This pattern closely mirrors EF-anxiety-RRB relationships noted in OCD (Evans, Iobst, & Lewis, 2004). Furthermore, in children with ID

such as Down syndrome (DS), where there is a dys-synchrony between chronological and mental age, RIS persist for a longer period relative to typically developing children (TD) of a similar chronological age, and RIS continues to be used as a means for managing fear and unpredictability (Evans et al., 2005).

Despite the recognition that RIS exists in both typical and atypical populations, previous research has focused almost exclusively on on ASD. Indeed, typical and atypical RRB may be subserved by common neural mechanisms (Evans et al., 2004; Leekam et al., 2011). In this study we focus on RIS behavior with the aim of exploring one of the elements (relationship with fear) of the theory of RIS described above, across two groups of typically-developing children and in two neurodevelopmental disorders-ASD and DS. Apart from providing group-based comparisons, we explore the fear-RIS link. We hypothesize that in ASD, higher levels of fear will be related to higher RIS levels, irrespective of age. We predict that a similar (albeit less pronounced) pattern of associations will be noted in children with DS, and in younger TD children (matched on mental age), but not for older TD children where one might expect that more mature forms of emotional regulation will be employed to manage potential fear and stress.

Methods

Sample

Participants were one-hundred-sixty-six parents and children. Forty one children received a clinical diagnosis of ASD, 38 children had Down Syndrome (DS), 45 were typically developing (TD) children matched to the mental age (MA) of the DS group, and 42 were chronological age (CA) matched TD children. Cognitive level was assessed with the Bayley Scales of Infant Development (Bayley, 1993) (for children < 30 months) or the Stanford-Binet Intelligence Scale (Thorndike, Hagen, & Sattler, 1986). MA was calculated

using the CA X IQ/100 equation. All children with ASD were within the normal range of cognitive functioning. TD CA matched children were not administered IQ assessments (see Table 1).

Table 1 Here

Procedures

Families were contacted through the day-care centres and school systems in the Greater New Orleans, Louisiana area, including those serving families with special needs. Parents provided informed consent. Identification of children with DS was based on physical phenotypic characteristics and parental confirmation of DS diagnoses. Diagnostic status of the children with ASD was reported by parents based on information from their family physician, as well as information provided by school psychologists, and/or speech and language therapists. None of the children with DS had co-morbid diagnosis of ASD.

Measures

The Childhood Routines Inventory (CRI; Evans et al., 1997) is a 19 item, parent-report questionnaire designed to measure rigid, routinized and ritualistic behaviours that may be described as rigidity/insistence on sameness (RIS). Each item is rated on the frequency/intensity. The CRI provides a total score and 'Just Right' (e.g., preference for particular ways of doing things), Rigid Routines (e.g., performing the same task repeatedly) and Sensory RIS (e. g. preference for certain articles of clothing due to being sensitive to how clothing feels) subscales. The CRI has excellent internal consistency and test-retest reliability (Evans et al., 1997; Cevikaslan et al., 2013).

Fear Inventory (Evans et al., 2005) is a 69-item parent-report inventory rating the intensity of each item on 5-point Likert scale, resulting in a total score and the following subscales: situations and places, harm, medical situations, animals, social situations, natural

environment, and strangers. The Fear Inventory has excellent internal consistency, face validity and assesses a range of experiences, both real and imaginary (Evans et al., 2005).

Results

CRI Factor Comparison

We first compared RIS scores across the four diagnostic groups (Table 2), ANOVAs were as follows: "Just Right" $F(3,162)= 16.62$, $p< .001$; Rigid Routines $F(3,162)= 52.76$, $p< .001$; Sensory behaviours $F(3,162)= 23.26$, $p< .001$. Post hoc tests are presented in Table 2. An identical pattern of results emerged when the effects of age and IQ were controlled for in Analysis of Co-variance (ANCOVAs) for all three CRI factors.

Table 2 here

RRB and Fear

Due to multiple associations, the significance level was set at .01. In the ASD group, "Just Right", Rigid Routines and Sensory behaviours were all significantly associated with total fear ($r= .48$, $r= .44$, $r= .44$, respectively, all $p \leq .009$). For fear subscales, the "Just Right" factor was associated with fear of situations and places ($r= .46$), and environmental fear ($r= .49$); Rigid Routines with fear of situations and places ($r= .42$), medical ($r= .48$) and environmental fear ($r= .43$); Sensory behaviours with medical ($r= .45$) and social fear ($r= .40$), all $p < .01$. In the DS group, both "Just Right" and Rigid Routines factors were associated with environmental fear ($r= .43$, and $r= .52$, respectively, both $p \leq .009$). In the TD MA group, Sensory behaviours were associated with total and environmental ($r= .41$ and .42 respectively, both $p < .01$). No significant associations were found in the TD CA group.

Discussion

This study compared ASD, DS and two groups of TD children on rigid, routinized patterns of behaviour, and the links between fear and RIS across these four groups.

ASD children had significantly higher Rigid Routines and Sensory behaviour scores than DS children, and although the "Just Right" scores were also higher, the difference was not statistically significant. The DS group had higher RIS scores than both TD (CA and MA) groups, which confirms findings reported previously (Evans & Grey, 2000; Evans, Kleinpeter, Slane and Boomer, 2014; Glenn & Cunningham, 2007). To our knowledge, although several studies have compared RIS and other types of RRB between ASD and groups with intellectual disabilities (see Leekam, Prior, & Uljarevic, 2011), this is the first study directly comparing ASD and DS groups. In general, previous research suggests that while other groups might display higher levels of specific behaviours (e.g., collecting/hoarding in Prader-Willi syndrome, see Greaves, Prince, Evans, & Charman, 2006), RRB in ASD appear to be distributed across a wide range of behaviours (Leekam et al., 2011) which supports findings from our study in terms of the ASD vs DS comparison.

We hypothesised that the strongest associations between fear and RRB would be for participants with ASD, followed by DS and younger TD children, and that this pattern would not be observed in older TD children. Our results support this hypothesis. These findings also support previous work on the RIS-anxiety association in ASD (Lidstone, Uljarevic, et al., 2014; Rodgers et al., 2012) and in DS (Evans et al., 2000; Evans et al., 2014). As noted above, RRB duringearly typical development provide a means of warding of normative fear, whose rise developmentally coincides with increasing RRB. However, as executive functions such as response inhibition and set shifting emerge and develop, RIS begin to wane, becoming less effective as a means of regulating fear and anxiety (Evans et al., 2004). Therefore, although RIS are adaptive in early development, their persistence can become maladaptive, serving to negatively reinforce fear and anxiety.

The emergence of response inhibition and set shifting abilities in early development is believed to be subserved by maturation of the orbitofrontal cortex (OFC) and the consolidation of pathways between the OFC, the anterior cingulate cortex (ACC), and striatal and limbic regions (in particular the caudate and amygdala, respectively) (see Judge, Evans, Schroepfer, & Gross, 2011 for an overview). These networks have also been implicated in the reappraisal of the emotional significance of various stimuli. For example OFC activation supresses the amygdala's response to negatively valanced stimuli (Silvers, Buhle, & Ochsner, 2014). Significant problems in EF (Hill, 2004), emotion regulation skills (Mazefsky et al., 2013), and structural and functional atypicalities of the OFC-ACC-limbic-striatal networks are well established in ASD as well as in OCD research (Schultz et al., 1999; Evans et al., 2004). Thus, we extrapolate that RRB will continue to serve their roles in regulating fear and anxiety. However being developmentally inappropriate, RIS will not serve their functions efficiently, rather RIS will reinforce fear and anxiety in clinical populations. This is in contrast to TD populations where more mature regulation strategies are in use. Findings from our study support this differential fear-RIS pattern in children with ASD and older TD children. Furthermore, our findings that reveal a positive fear-RIS association in DS individuals and young MA matched TD children are consistent with the already recognized role of RIS in managing fear during this particular developmental period. Our findings have potential clinical implications as they suggest that interventions such as Cognitive Behavior Therapy (Reaven, Blakeley-Smith, Culhane-Shelburne, & Hepburn, 2012; Sukhodolsky, Bloch, Panza, & Reichow, 2013) that emphasize emotion regulation and coping skills may have beneficial effects on reducing RIS in ASD.

It is important to note that the support provided here for the mechanisms underlying progression from adaptive to maladaptive behaviours is limited by several factors. The cross-sectional nature of the study and the fact that EF skills were not directly assessed in this

study, serve as limitations to be addressed in future research. Future studies should also explore the interaction between EF development and developmental changes in the presentation of RRB, and their links to emotion regulation, to determine how these changes map onto the developing neural substrates that the comprise the OFC-striato-limbic network. Other limitations include the relative imbalance in the male-female ratio across diagnostic groups – only two female participants were in the ASD cohort as well as the fact that some of the children scored at or near the floor levels on certain Fear Inventory subscales. Also, ASD is estimated to occur in 5-10% of probands with DS (Kent, Evans, Paul & Sharp, 1999; Moss, Richards, Nelson, Oliver, 2013). Here, we relied on parental reports of child and family psychiatric, developmental, and medical history, that indicated that none of the participants with DS had been diagnosed with ASD.

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In normative development, some aspects of RRB, such as rigidity and insistence on sameness (RIS), serve to constrain the unpredictability of the environment and to ward off ~~fears~~fear and anxiety, thus serving as an early form of emotion and arousal regulation (Evans et al., 1999; Gesell et al., 1974; Leekam et al., 2011; Zohar & Felz, 2001). In young children, normative RIS are likely to occur at times of transition (such as bedtime), and are accompanied by normative ~~fears~~fear (Evans, Gray, & Leckman, 1999). Normative fear and

anxiety, such fear of the dark, and separation/stranger anxiety, occurs in the context of typical development. This aspect of typical development may be distinguished from phobia and clinical anxiety based on the fact that they are largely adaptive mechanism for coping with particular developmental challenged; occur at specific stages of development; and are transitory in nature (Brooker, Buss, Lemery-Chalfant, Aksan, Davidson, Goldsmith, 2014; Evans et al., 1999; Gullone, 2000). The link between normal childhood RIS and fear/anxiety ~~reflects~~parallels findings in ASD (Lidstone, Uljarević, et al., 2014; Rodgers, Glod, Connolly, & McConachie, 2012), and in obsessive compulsive disorder (OCD), where particular behaviours are performed for purposes of anxiety reduction (Mataix-Cols, Rosario-Campos, & Leckman, 2005). However, it is not clear why and how the RRB-Fear/anxiety link transitions from adaptive to maladaptive forms that reinforce anxiety in clinical conditions.

Cross-sectional questionnaire studies suggest that RIS begin around the age of 24 months, peak around 72 months, and steadily wane afterwards (Arnott et al., 2010; Cevikaslan et al., 2013; Evans et al., 1997; Leekam et al., 2007). A similar timeline has been observed for normative fear~~s~~ (Evans et al., 1999), which are in turn, concomitant to developments in executive functioning (EF) skills, most importantly response inhibition and set-shifting abilities (Zelazo et al., 2003). Therefore, it seems that as cognitive and emotion-related strategies for managing and regulating arousal and fear develop, children become less reliant on RIS.

In ASD, where problems in EF (Hill, 2004; Robinson, Goddard, Dritschel, Wisley, & Howlin, 2009) and emotion regulation (Mazefsky et al., 2013) have been noted, RIS continue to serve to reduce fear/anxiety. Although RIS may be an efficient short-term strategy, given their developmentally inappropriate nature, they will likely reinforce and sustain fear~~s~~ and anxiety long term (Rodgers et al., 2012). This pattern closely mirrors EF-anxiety-RRB relationships noted in OCD (Evans, Iobst, & Lewis, 2004). Furthermore, in children with ID

such as Down syndrome (DS), where there is a dys-synchrony between chronological and mental age, RIS persist for a longer period relative to typically developing children (TD) of a similar chronological age, and RIS continues to be used as a means for managing fears and unpredictability (Evans et al., 2005).

Despite the recognition that RIS exists in both typical and atypical populations, pPrevious research on RIS has focused almost exclusively on mainly on ASD. despite the fact that RIS intersect a wide range of disorders and extend well into the general population; Indeed, tTypical and atypical RRB may also be subserved by common neural similar mechanisms (Evans et al., 2004; Leekam et al., 2011). In this study we focus on RIS behaviors with the aimThe aim of this study was of to exploringe one of the elements (relationship with fear) of the theory of RRB RIS described above, across two groups of typically-developing children normative development and in two neurodevelopmental disorders-ASD and DS. Apart from providing group-based comparisons, we explore the fear-RRB RIS link. We hypothesize that in ASD, higher levels of fear will be related to higher RRB RIS levels, irrespective of their age. We predict that a similar (albeit less pronounced) pattern of associations will be noted in children with DS, and in younger TD children (matched on mental age), but not for older TD children where one might expect that more mature forms of emotional regulation will be employed to manage potential fear and stress.

Methods

Sample

Participants were one-hundred-sixty-six parents and children. Forty one children received a clinical diagnosis of ASD, 38 children had Down Syndrome (DS), 45 were typically developing (TD) children matched to the mental age (MA) of the DS group, and 42 were chronological age (CA) matched TD children. Cognitive level was assessed with the

Bayley Scales of Infant Development (Bayley, 1993) (for children < 30 months) or the Stanford-Binet Intelligence Scale (Thorndike, Hagen, & Sattler, 1986). MA was calculated using the CA X IQ/100 equation. All children with ~~children with~~ ASD~~children~~ were within the normal range of cognitive functioning. TD CA matched children were not administered IQ assessments (see Table 1).

Table 1 Here

Procedures

Families were contacted through the day-care centr~~e~~s and school systems in the Greater New Orleans, Louisiana ~~LA~~ area, including those serving families with special needs. Parents provided informed consent. Identification of children with DS was based on physical phenotypic characteristics and parental confirmation of DS diagnoses. Diagnostic status of the children with ASD was reported by parents based on information from their family physician, as well as information provided by school psychologists, and/or speech and language therapists. None of the children with DS had co-morbid diagnosis of ASD.

Measures

The Childhood Routines Inventory (CRI; Evans et al., 1997) is a 19 item, parent-report questionnaire designed to measure rigid, routinized and ritualistic behaviours that may be described as rigidity/insistence on sameness (RIS). Each item is rated on the frequency/intensity. The CRI ~~It~~ provides a total score and 'Just Right' (e.g., preference for particular ways of doing things), Rigid Routines (e.g., performing the same task repeatedly) and Sensory RIS (e. g. preference for certain articles of clothing due to being sensitive to how clothing feels) subscales. The CRI has excellent internal consistency and test-retest reliability (Evans et al., 1997; Cevikaslan et al., 2013).

Fear Inventory (Evans et al., 2005) is a 69-item parent-report inventory rating the intensity of each item on 5-point Likert scale, resulting in a total score and the following subscales: situations and places, harm, medical situations, animals, social situations, natural environment, and strangers. The Ffear Inventory has excellent internal consistency, face validity and assesses a range of experiences, both real and imaginary (Evans et al., 2005).

Results

CRI Factor Comparison

We first compared RIS scores across the four diagnostic groups (Table 2), ANOVAs were as follows: "Just Right" $F(3,162)= 16.62$, $p< .001$; Rigid Routines $F(3,162)= 52.76$, $p< .001$; Sensory behaviours $F(3,162)= 23.26$, $p< .001$. Post hoc tests are presented in Table 2. An identical pattern of results emerged when the effects of age and IQ were controlled for in Analysis of Co-variance (ANCOVAs) for all three CRI factors.

Table 2 here

RRB and Fear

Due to multiple associations, the significance level was set at .01. In the ASD group, "Just Right", Rigid Routines and Sensory behaviours were all significantly associated with total fear s ($r= .48$, $r= .44$, $r= .44$, respectively, all $p \leq .009$). For fear subscales, the "Just Right" factor was associated with fears of situations and places ($r= .46$), and environmental fears ($r= .49$); Rigid Routines with fears of situations and places ($r= .42$), medical ($r= .48$) and environmental fears ($r= .43$); Sensory behaviours with medical ($r= .45$) and social fears ($r= .40$), all $p < .01$. In the DS group, both "Just Right" and Rigid Routines factors were associated with environmental fearfears ($r= .43$, and $r= .52$, respectively, both $p \leq .009$). In the TD MA group, Sensory behaviours were associated with total and environmental ($r= .41$

and .42 respectively, both $p < .01$). No significant associations were found in the TD CA group.

Discussion

This study compared ASD, DS and two groups of TD children on rigid, routinized patterns of behaviour, and the links between ~~fears~~fear and ~~RRB~~ RIS across these four groups.

ASD children had significantly higher Rigid Routines and Sensory behaviour scores than DS children, and although the "Just Right" scores were also higher, the difference was not statistically significant. The DS group had higher RIS scores than both TD (CA and MA) groups, which confirms findings reported previously (Evans & Grey, 2000; Evans, Kleinpeter, Slane and Boomer, 2014; Glenn & Cunningham, 2007). To our knowledge, although several studies have compared RIS and other types of RRB between ASD and groups with intellectual disabilities (see Leekam, Prior, & Uljarevic, 2011), this is the first study directly comparing ASD and DS groups. In general, previous research suggests that while other groups might display higher levels of specific behaviours (e.g., collecting/hoarding in Prader-Willi syndrome, see Greaves, Prince, Evans, & Charman, 2006), RRB~~s~~ in ASD appear to be distributed across a wide range of behaviours (Leekam et al., 2011) which supports findings from our study in terms of the ASD vs DS comparison.

We hypothesised that the strongest associations between fear and RRB would be for participants with ASD, followed by DS and younger TD children, and that this pattern would not be observed in older TD children. Our results support this hypothesis. These findings also support previous work on the RIS-anxiety association in ASD (Lidstone, Uljarevic, et al., 2014; Rodgers et al., 2012) and in DS (Evans et al., 2000; Evans et al., 2014). As noted above, RRB during early typical~~years of normative~~ development provide a means of warding of normative ~~fears~~fear, whose rise developmentally coincides with increasing RRB.

However, as executive functions such as ~~, in particular~~ response inhibition and set shifting~~,~~ emerge and ~~become more~~ develop~~ed~~, ~~RRB~~ RIS begin to wane, becoming less effective as a means of regulating fear and anxiety (Evans et al., 2004). Therefore, although ~~RRB~~ RIS are adaptive in early development, their persistence can become maladaptive, serving to negatively reinforce fear and anxiety.

The emergence of response inhibition and set shifting abilities in early development is believed to be subserved by maturation of the orbitofrontal cortex (OFC) and the consolidation of pathways between the OFC, the anterior cingulate cortex (ACC), and striatal and limbic regions (in particular the caudate and amygdala, respectively) (see Judge, Evans, Schroepfer, & Gross, 2011 for an overview). These networks have also been implicated in the reappraisal of the emotional significance of various stimuli. For example OFC activation supresses the amygdala's response to negatively valanced stimuli (Silvers, Buhle, & Ochsner, 2014). Significant problems in EF (Hill, 2004), emotion regulation skills (Mazefsky et al., 2013), and structural and functional atypicalities of the OFC-ACC-limbic-striatal networks are well established in ASD as well as in OCD research (Schultz et al., 1999; Evans et al., 2004). Thus, we extrapolate that RRB will continue to serve their roles in regulating fear and anxiety. However being developmentally inappropriate, ~~RRB~~ RIS will not serve their functions efficiently, rather ~~RRB~~RIS will reinforce fear~~s~~ and anxiety in clinical populations. This is in contrast to TD populations where more mature regulation strategies are in use. Findings from our study support this differential fear-~~RRB~~ RIS pattern in children with ASD and older TD children. Furthermore, our findings that reveal a positive fear-~~RRB~~ RIS association in DS individuals and young MA matched TD children are consistent with the already recognized role of ~~RRB~~ RIS in managing fear during this particular developmental period. Our findings have potential clinical implications as they suggest that interventions such as Cognitive Behavior Therapy (Reaven, Blakeley-Smith, Culhane-Shelburne, &

Hepburn, 2012; Sukhodolsky, Bloch, Panza, & Reichow, 2013) that emphasize emotion regulation and coping skills may have beneficial effects on reducing RIS in ASD.

It is important to note that the support provided here for the mechanisms underlying progression from adaptive to maladaptive behaviours is limited by several factors. The cross-sectional nature of the study and the fact that EF skills were not directly assessed in this study, serve as limitations to be addressed in future research. Future studies should also explore the interaction between EF development and developmental changes in the presentation of RRB, and their links to emotion regulation, to determine how these changes map onto the developing neural substrates that the comprise the OFC-striato-limbic network. Other limitations include the relative imbalance in the male-female ratio across diagnostic groups – only two female participants were in the ASD cohort as well as the fact that some of the children scored at or near the floor levels on certain Fear Inventory subscales. Also, ASD is estimated to occur in 5-10% of probands with DS (Kent, Evans, Paul & Sharp, 1999; Moss, Richards, Nelson, Oliver, 2013). Here, we relied on parental reports of child and family psychiatric, developmental, and medical history, that indicated that none of the participants with DS had been diagnosed with ASD.

~~Conclusion~~

~~This study provided the evidence of the fear-RRB association in children with ASD, DS, and two groups of TD children.~~

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Table 1. Demographic Information

	CA (months)		MA (months)		IQ		Gender (N)	
	Mean	**SD**	**Mean**	**SD**	**Mean**	**SD**	**Male**	**Female**
ASD	123.39	27.67	84.40	37.43	68.15	23.69	38	3
DS	125.37	45.71	52.11	13.31	42.35	7.56	17	21
TD CA	117.93	22.91	NA	NA	NA	NA	15	27
TD MA	51.13	22.1	56.69	24.51	111.91	9.51	24	21

Table 2. Comparison of CRI Scores across four groups

	ASD[a] (Mean, SD)	DS[b] (Mean, SD)	TD MA[c] (Mean, SD)	TD CA[d] (Mean, SD)	Post Hoc comparison (*p*, Cohen's d)
"Just Right"	24.27 (8.01)	20.53 (7.1)	19.11 (6.59)	14.09 (4.46)	a > d ($p < .001$, d= 1.57) a > c ($p = .003$, d= .70) a ≈ b ($p = .07$, d= .49) b > d ($p < .001$, d= 1.09) b ≈ c ($p = .78$, d= .20)
Rigid Routine	23.29 (4.86)	17.12 (5.36)	13.81 (5.49)	10.5 (3.5)	a > d ($p < .001$, d= 3.02) a > c ($p < .001$, d= 1.83) a > b ($p < .001$, d= 1.21) b > d ($p < .001$, d= 1.46) b > c ($p = .019$, d= .61)
Sensory Behaviours	15.76 (4.4)	11.39 (4.38)	10.57 (4.02)	8.64 (3.19)	a > d ($p < .001$, d= 1.86) a > c ($p < .001$, d= 1.23) a > b ($p < .001$, d= .99) b > d ($p = .016$, d= .71) b ≈ c ($p = .80$, d= .19)

Note: [a]=ASD; [b]=DS; [c]= TD MA; [d]=TD CA.

Dear Dr. Amaral,

Below, you will find your, and the reviewers' comments and suggestions, and a point-by-point response.

David W. Evans

EDITOR

Thank you for submitting the manuscript entitled, "Relationship between Repetitive Behaviours and Fears across Normative Development, Autism Spectrum Disorder and Down Syndrome" (Manuscript ID: AUR-16-0128) to Autism Research. The manuscript has now been reviewed by two experts in the field and although clearly of value, the manuscript does require revision before further consideration for publication in Autism Research.
The comments of the reviewers are enclosed below. Final acceptance of the paper is contingent upon the manuscript being revised acceptably, taking into consideration the reviewers' comments. At the time of resubmission please provide a detailed point-by-point response to all the reviewers' comments and concerns. I will determine whether the paper has been sufficiently revised to deal with the concerns of the reviewers.

Response: We are very pleased to have the opportunity to revise and resubmit out paper. We welcome these comments and suggestions from the reviewers which were very constructive and have significantly improved the quality of our original submission. We have attempted to address each of the reviewers' comments (please see point by point responses below) and we hope we have done so in a satisfactory manner.

Autism Research reviews: Point-by-point response

REVIEWER 1:

Comment 1: Need to define "normative fears" and possibly some examples, early in the introduction. How normative are they? Perhaps a couple of example questions from each of the questionnaires would be helpful when talking about the measures.

Response 1: We have now defined the term "normative fears" noting that they appear as a normal part of the development and serve adaptive functions and noting the criteria that distinguish them from the clinical fears, we have also added three examples of normative fears (stranger and separation anxiety, and fears of the dark) that are reflected in the Fear Survey. Please see track changes on Page 4, line 24 and Page 5, lines 1-6.

Comment 2: LA should be spelled out to Louisiana; non-US citizens may confuse it with Los Angeles

Response 2: We have spelled out Louisiana.

Comment 3: Surprising to find no co-morbid ASD in DS given previously reported high rates of ASD in DS. How was this ascertained or ruled out? Were any ASD-type measures given to DS folks? If not sufficiently rigorous distinction, this should be noted as a limitation.

Response 3: This is an excellent point. We asked parents to report on the child and family psychiatric/neurodevelopmental and other medical history and none of the parents have reported that their child with Down syndrome had co-morbid ASD diagnosis. However, we did not use any ASD specific interview nor questionnaire measure and it is possible that some of the children could have had ASD traits expressed at the subthreshold level or even at the diagnostic level but have not received clinical diagnosis. We agree with the reviewer this should be included as a limitation, we how now done this in the final paragraph of the manuscript. Please see track changes on Page 11, lines 11-16.

Comment 4: Why no significant associations between fear and anxiety in the TD group? Are there floor effects?

Response 4: we have found associations between total fear score and sensory subscale of CRI (r= .41, p < .01) as well as between environmental fear score and sensory CRI subscale (r= .42, p < .01) for the typically developing group matched on mental age with Down Syndrome group but no significant associations in TD group matched with Down Syndrome group on chronological age i. e. for older TD group. We have explored the potential presence of floor and ceiling effects for both total fear schedule score as well as for subscale scores (we have copied tables separately showing detailed descriptive statistics as well as % of sample scoring at floor level across four groups). Following standards for similar types of measures (Lim, Seubsman, & Sleigh, 2008 for example), floor effects should be less than 20% of sample in order for a scale to capture the full range of potential responses. As can be seen, ≥ 20% criteria was met for: Situation and Places subscale across all 4 groups, Harm subscale for ASD, Animals for ASD and both TD groups, Social Situations in DS and TD MA group, and Strangers in DS group. Percentage of individuals scoring at or near the floor level across the four samples was low. Pattern of scores is similar in TD CA and TD MA groups and reflects what we would expect from the developmental change in the content of typical fears and there are also parallels with DS sample group. The fact that relationship between total fear score and CRI was for example found in both DS and TD MA but not TD CA group even though none of the groups had high % of individuals scoring at the floor level and furthermore there were no significant differences between total fear scores between these groups suggests that floor effects are not likely to be the explanation of our pattern of findings. As we have suggested, we feel that the explanation that in older typically developing children more mature forms of regulation than repetitive behaviours will be employed to manage potential fear and stress fits well with the pattern of findings. However, we do agree with the reviewer that it is good to err on the side of caution and do not discard the influence of less spread of fear scores in TD CA group. Therefore we have added this as a limitation, please see track changes on Page 11, lines 11-12.

Table 1. Fear Descriptives in Autism Spectrum Disorder

	Mean	SD	Range	Theoretical Range	% (N) Floor	% (N) Ceiling
Fear Total	135.5	55.74	72-276	69-345	0	0
Situation and Places	25.51	13.73	15-58	15-75	25.6	0

Harm	19.76	12.17	12-59	12-60	29.3	0
Medical Situations	31.65	10.22	11-45	9-45	8.1	7.3
Animals	13.07	7.82	7-35	7-35	27.5	5
Social Situations	16.80	8.98	8-37	8-40	19.5	0
Natural Environment	19.52	7.95	9-36	9-45	2.5	0
Strangers	8.23	3.83	5-21	5-25	17.9	0

Table 2. Fear Descriptives in Down Syndrome

	Mean	SD	Range	Theoretical Range	% (N) Floor	% (N) Ceiling
Fear Total	125.51	37.75	72-239	69-345	0	0
Situation and Places	22.66	10.61	15-75	15-75	26.3	2.6
Harm	21.73	10.14	12-51	12-60	18.9	0
Medical Situations	24.66	10.47	9-45	9-45	7.9	2.6
Animals	15.81	7.03	7-35	7-35	13.2	2.6
Social Situations	12.23	6.23	8-40	8-40	36.8	2.6
Natural Environment	20.89	7.43	9-34	9-45	5.3	0
Strangers	8.52	3.58	5-20	5-25	21.1	0

Table 3. Fear Descriptive in Mental Age matched Typically Developing Children

	Mean	SD	Range	Theoretical Range	% (N) Floor	% (N) Ceiling
Fear Total	118.89	33.90	71-203	69-345	2.2	0
Situation and Places	18.09	4.73	15-31	15-75	55.6	0
Harm	23.55	11.75	12-52	12-60	15.6	0
Medical Situations	21.62	9.55	9-42	9-45	11.1	0
Animals	12.69	5.75	7-34	7-35	20	0

Social Situations	13	4.83	8-32	8-40	22.2	0
Natural Environment	19.02	6.38	9-33	9-45	6.7	0
Strangers	10.91	3.92	5-19	5-25	6.7	0

Table 4. Fear Descriptives in Chronological Age matched Typically Developing Children

	Mean	SD	Range	Theoretical Range	% (N) Floor	% (N) Ceiling
Fear Total	110.1	29.9	69-176	69-345	5.3	0
Situation and Places	17.21	3.57	15-30	15-75	53.7	0
Harm	24.46	10.85	12-56	12-60	9.8	0
Medical Situations	20.50	8.64	9-40	9-45	10.5	0
Animals	10.27	3.69	7-18	7-35	41.5	0
Social Situations	13.05	4.28	8-27	8-40	17.1	0
Natural Environment	15.35	4.59	9-27	9-45	10	0
Strangers	8.22	2.58	5-15	5-25	19	0

Comment 5: I know this is a short report, but it seems that a scatter plot/s or box plots or other way of showing the data (means and variability across participants and groups) would be a helpful addition. Dealing with point 4 above, also degree of overlap (and individual differences) in DS vis-à-vis ASD.

Response 5: Please see our response above, we hope that detailed descriptive information provided across four tables above provides sufficient information. We have also explored the differences in terms of fear scores across the four groups, ANOVAs were as follows: Total Fear $F= 2.62$, $p= .053$; Situation and Places $F= 7.66$, $p< .001$; Harm $F= 1.33$, $p= .265$; Medical

Situations F= 10.09, p< .001; Animals F= 5.23, p= .002; Social Situations F= 3.97, p= .009; Natural Environment F= 4.94, p= .003; Strangers F= 5.88, p= .001. Post hoc tests are presented in Table 5 below.

Table 5. Comparison of Fear scores across four groups

	Post Hoc comparison (p)
Fear Total	a > d (p= .038); b ≈ c ≈ d.
Situation and Places	a > c (p= .001); a > d (p< .001), b > d (p= .039); b ≈ c.
Harm	a ≈ b ≈ c ≈ d.
Medical Situations	a > b (p= .012); a > c (p< .001), a > d (p< .001); b ≈ c ≈ d.
Animals	b > d (p= .001); a ≈ b; a ≈ c ≈ d.
Social Situations	a > b (p= .011); a > c (p= .036), a > d (p= .045); b ≈ c ≈ d.
Natural Environment	a > d (p= .03); b > d (p= .002); a ≈ b ≈ c.
Strangers	a < c (p= .004); b < c (p= .013); c > d (p= .003); a ≈ b ≈ d

Note: a=ASD; b=DS; c= TD MA; d=TD CA.

Comment 6: 2nd sentence of Discussion is incomplete. First sentence of third paragraph of Discussion has a tense mismatch "will" instead of "would".

Response 6: This has been changed.

Comment 7: Were there other measures of EF or anxiety administered? If so, I think a brief mention of the analyses would be useful even if—especially if—there were no significant results. We're trying as a field to develop both theory and measures and these can be hard to match up, so underreporting of data isn't useful for anyone. We need to think more carefully about the theory-measurement distance.

Response 7: Having EF measures, either in the form of parental report (such as BRIEFF or even Effortful Scale from Rothbart's Temperament Questionnaires) or experimental tasks to tease apart the influence of response inhibition and set shifting components of EF would indeed have

been beneficial and provide more detailed testing of our theory. For these reasons we have noted the lack of EF measure as a limitation in the discussion section:

"The cross-sectional nature of the study and the fact that EF skills were not directly assessed in this study, serve as limitations to be addressed in future research. Future studies should also explore the interaction between EF development and developmental changes in the presentation of RRB, and their links to emotion regulation, to determine how these changes map onto the developing neural substrates that the comprise the OFC-striato-limbic network."

Similarly, having dedicated measure of anxiety that has been shown to perform well psychometrically and also in terms of sensitivity and specificity in both general populations as well in ASD group such as for example the Spence Children's Anxiety Scale-Parent Version (Spence, 1999; Nauta et al., 2004), the Screen for Childhood Anxiety Related Emotional Disorders (Birmaher et al., 1995; 1999) or even ASD specific anxiety measure such as ASC-ASD (Rodgers, Wigham, McConachie, Freeston, Honey, & Parr, 2016) would have been beneficial. However, in relation to this, we would like to point out that emphasis of our paper to explore the relationship between repetitive behaviors and normative fears. We are hoping to build on results presented here in future studies that would, as we noted above, combine the use of EF tasks, measures of both normative fears and clinical fears and anxiety, ideally within longitudinal design.

Comment 8: The conclusion is incomplete. Flesh out, or eliminate.

Response 8: We deleted the conclusion. We agree that it did not add to the manuscript.

REVIEWER 2:

Comment 1: Review use of serial comma: http://blog.apastyle.org/apastyle/2011/04/usingserial-commas.html. This relates to using a comma to separate the last two items in a series to reduce possible confusion.

Response 1: We have added the comma in the title.

Lay Abstract

Comment 2: Lines 13-15. You mention 4 groups of children; however, you have not yet explained that there are two TD groups.

Response 2: We now mention in the first sentence of the Lay Abstract that there are two TD groups

Comment 3: Line 24. Comma in a series again and revisions will be needed for this throughout the paper.

Response 3: We have applied this rule throughout the manuscript.

Scientific Abstract

Comment 4: Lines 10-12. Check consistency in capitalization. In Lay Abstract Autism Spectrum Disorder is capitalized and not here.

Response 4: We believe we have now addressed this, and all instances are upper case.

Comment 5: Line 16. Same note as above in that you refer to 4 groups but have not explained that there are 2 TD groups, which makes it confusing.

Response 5: We have made the edit in the first sentence of the Scientific Abstract.

Comment 6: Introduction The introduction feels a little unfocused; it could benefit from a close edit and some reorganization.

Response 6: Thank you for this suggestion, we have made several additions and also reorganized the structure of the introduction, we hope that it now reads better and feels more focused.

Comment 7: Page 4, Line 19. No "s" needed for Autism Spectrum Disorder

Response 7: We have removed the "s" – except in the titles of published works in the References section.

Comment 8: Page 4, Line 19-22. Consistency is needed regarding whether you are using RRB or RRBs and clarify throughout.

Response 8: We now write it as RRB throughout the manuscript.

Comment 9: Page 4. Line 38. You mention how research on RRB has been neglected which has limited early interventions; however, you don't come back to this point in the Discussion. You might consider the benefit of remarking on how your findings could help with this or revising this point as it seems a bit out of place.

Response 9: This is an excellent point. We have now provided discussion on potential clinical/treatment implications of our findings, please see track changes on Page 10, lines 24-25 and Page 11, line 1-2.

Comment 10: Page 4, Lines 49-50. It might be helpful to provide an example of normative fears. It was very helpful to suggest when normative RIS are likely to occur with the example so this would be along those lines.

Response 10: We have now defined the term "normative fears" noting that they appear as a normal part of the development and serve adaptive functions and noting the criteria that distinguish them from the clinical fears, we have also added three examples of normative fears (stranger and separation anxiety, and fears of the dark) that are reflected in the Fear Survey. Please see track changes on Page 4, line 24 and Page 5, lines 1-6.

Comment 11: Page 4, Lines 52-54. It is unclear what you are suggesting by saying the link between normal childhood RIS and fear/anxiety reflect findings in ASD and then how that relates to OCD. I find that the normative RIS and fear/anxiety are quite different than what we see in children with ASD and in OCD where the children are under much distress to complete the compulsion to prevent a dreaded outcome.

Response 11: we have now change the word "reflects" to "parallels" as it reflects better what we wanted to express. Review makes a very interesting point. Our previous research (Evans et al., 1997; 1999) and findings from others (Zohar & Felz, 2001; Laing, Fernyhough, Turner, & Freeston, 2009) all suggest clear relationship between fears/anxiety and rigid routines and rituals during typical development. For example, Lang et al. (2009) administered semi-structured interviews to a group of 142 typically developing children in order to explore the content of their fears and their relations with ritualistic behaviors and identified that the regular performance of

rituals was indeed related to anxiety—children performed ritualistic actions and preferred routines and rigidity as ways of constraining the unpredictability of environment and warding off anxieties and fears. Interruption (or inability to perform) of rigid rituals was associated with significant discomfort and increase in anxiety mirroring therefore (although to a milder extent) ego-dystonic nature of these behaviors seen in OCD with an important note that, as we have discussed they are transitory in nature and abate over development. We do agree with the reviewer that it is important to precisely map out the role of particular behaviors with being particularly mindful of Tinbergen's (1951) distinction between evolutionary origin (distal cause) and the immediate antecedents (triggers or proximal causes) of behaviorsthat are distinct from the original cause of the behavior, and establish whether phenomenologically similar behaviors might play different roles in different groups.

Comment 12: Page 5, Lines 5-8. I am wondering if you mean to say RIS here rather than RRB or whether you are using them interchangeably. In addition, the switching of these terms is used throughout the intro with the primary focus on RIS. You note at the beginning that RRB encompasses RIS and RSM; however RSM is not mentioned throughout, although your measure (CRI) has a sensory component. As mentioned above, I am wondering whether a more thorough description of these components and which one (s) exactly you are addressing in this paper would be helpful.

Response 12: thank you for pointing this out, we have indeed meant to say RIS rather than to use more genera category of RRB. We have provided a definition of RIS in the introduction and indeed most of our theorizing relates to RIS. We do agree with the reviewer that focus might have been shifting at several points, particularly in the last paragraph of the introduction and we have now made it more explicit that we are focusing on RIS category. Also, we have made it explicit in the methods section (please see track changes on Page 7, lines 18-19), where we are describing CRI that this questionnaire was designed to measure rigid, routinized and ritualistic behaviours that may be described as RIS and that sensory subscale also covers this type of behaviors that has sensory component.

Comment 13: Page 5, Lines 10-12. Are these cross-sectional studies you are referencing in TD children?

Response 13: Indeed, we provide this statement to make this clear, "Cross-sectional questionnaire studies suggest that…"

Comment 14: 13. Page 5, Lines 52-56. It would be helpful to clarify the difference between typical and atypical RRB.

Response 14: We have re-written the second paragraph of the Introduction (Page 4) and note that the distinction between typical and atypical RRB is not clear, aside from the fact that the evidence suggests that RRB in ASD "negatively impact learning and social adaptation, and present a significant source of stress and management challenge for parents."

Method

Comment 15: Page 6, Lines 41-42. Since it is not typical for a representative group of children with ASD to all have cognitive functioning in the normal range, did you exclude children with lower cognitive functioning. If so, why?

Response 15: Thank you for this excellent question. We did this to try to differentiate ASD symptoms *per se*, as apart from intellectual disability. Many children with ID exhibit RRB, and so we are trying to differentiate (cognitive) developmental variables from neurodevelopmental status. We also wanted to represent a group with ASD but no ID, a well-defined group with ID but no ASD (DS), and two comparisons that match to the MA and the CA of the DS group.

Comment 16: Table 1 info. As mentioned above, there is a concern about so few girls in the ASD group and so many in the TD groups. I realize it is more difficult to find girls with ASD; however, there are findings in multiple studies of higher rates of RRBs in boys than girls with ASD. I am not aware of whether this follows with RIS in normative development. This could really affect your findings overall. Did you look at gender differences? Is it possible to gather data on more girls with ASD or more TD boys?

Response 16: Findings on the influence of gender on the expression of repetitive behaviors in both typical development and ASD have been mixed. Questionnaire studies of RRB in typical development reported no difference using the same measure that we have used for the current paper (Evans et al., 1997), while other studies reported that boys have higher total repetitive behavior scores (Leekam et al., 2007) and sensory interests (Arnott et al., 2010). Similarly, in an

ASD population, one study using both observational assessment (ADOS) and parent interview (Developmental, Dimensional, and Diagnostic Interview; Skuse et al., 2004), found that boys individuals (aged 3-18 years) had higher repetitive play with objects and circumscribed interests than girls (Mandy et al., 2012). However, Joseph, Thurm, Farmer, and Shumway (2013) found no influence of gender on any of the RBS-R parent questionnaire subscales.

We have indeed looked at the gender differences across three groups (since there were only 2 females in ASD group comparison was not performed), as can be seen from the Table 5, there were no significant differences within Down Syndrome, MA nor CA matched TD groups.

In addition to this, we've also explored gender differences across three groups in terms of total fear score as well as fear inventory subscale scores, similarly as with CRI scores, as can be seen from the Table 7 there were no significant differences within Down Syndrome, MA nor CA matched TD groups (with the exception of fear of animals for CA TD group [p= .033] but after correction for multiple comparisons that difference was no longer statistically significant).

However, we have now added relative imbalance in the male-female ratio across groups as a limitation, please see track changes on Page 11, lines 10-11.

Table 6. Gender Differences in CRI scores

	Down Syndrome			MA TD Group			CA TD Group		
	Male	Female	vs	Male	Female	vs	Male	Females	vs
"Just Right"	20.87 (7.54)	20.28 (6.95)	F= .06, p= .813	20.04 (5.43)	18.09 (7.69)	F= .96, p= .334	12.93 (3.59)	14.74 (4.82)	F= 1.60, p= .213
Repetitive Actions	16.78 (5.13)	17.36 (5.65)	F= .09, p= .763	14.14 (5.94)	13.48 (5.1)	F= .15, p= .699	9.13 (2.09)	11.29 (3.91)	F= 3.79, p= .058
Sensory	11.93 (4.63)	11 (4.25)	F= .39, p= .536	10.52 (4.19)	10.62 (3.94)	F= .01, p= .94	7.8 (3.78)	9.11 (2.78)	F= 1.65, p= .206

Table 7. Gender Differences in Fear Inventory scores

	Down Syndrome			MA TD Group			CA TD Group		
	Male	Female	vs	Male	Female	vs	Male	Females	vs
Fear Total	122.25 (43.37)	128 (33.76)	*F*=.206 *p*= .653	120.83 (37.82)	116.67 (29.57)	*F*=.16 *p*= .686	101.53 (30.34)	115.69 (28.89)	*F*= 2.09 *p*= .156
Situation and Places	24.82 (14.57)	20.9 (5.59)	*F*=1.29 *p*= .263	18.29 (4.84)	17.86 (4.7)	*F*=.09 *p*= .762	16.47 (2.39)	17.65 (4.08)	*F*= 1.05 *p*= .311
Harm	22.12 (9.71)	21.43 (10.68)	*F*=.04 *p*= .839	26.46 (12.6)	20.24 (9.97)	*F*=3.3 *p*= .076	22.53 (11.62)	25.58 (10.44)	*F*= .74 *p*= .394
Medical Situations	23.94 (11.33)	25.23 (9.96)	*F*=.14 *p*= .71	21.21 (9.35)	22.09 (9.99)	*F*=.09 *p*= .760	19.73 (8.31)	21 (9)	*F*= .19 *p*= .665
Animals	13.76 (4.94)	17.48 (8.08)	*F*=2.74 *p*= .106	11.46 (4.74)	14.09 (6.57)	*F*=2.43 *p*=.127	8.67 (2.91)	11.19 (3.83)	*F*=4.86 *p*= .033
Social Situations	12.65 (8.19)	11.9 (4.23)	*F*=.13 *p*= .72	13.25 (5.79)	12.71 (3.54)	*F*=.13 *p*=.715	11.53 (3.89)	13.92 (4.32)	*F*=3.13 *p*= .085
Natural Environment	20 (6.8)	21.62 (7.99)	*F*=.44 *p*= .512	19.46 (6.12)	18.52 (6.79)	*F*=.24 *p*=.63	14.8 (4.99)	15.68 (4.42)	*F*= .34 *p*= .565
Strangers	7.41 (2.03)	9.43 (4.29)	*F*=3.16 *p*= .084	10.71 (4.04)	11.14 (3.86)	*F*=.13 *p*= .715	7.8 (2.34)	8.48 (2.72)	*F*=.65 *p*= .426

Procedures

Comment 17: 16. Page 7, Lines 3-8. If children were recruited through day-care centers and school systems, how did they get a multidisciplinary diagnostic team eval? Was this done through the same study or prior to the study somehow? Were any standard measures used?

Response 17: That was based on the parental report. Parents indicated that ASD diagnoses that had been conferred by family primary care physician or pediatrician or speech and language pathologist. The daycare and school systems had clinical school psychologists on staff to help in the diagnoses. No standardized measures (aside from IQ) were used. We now clarify this information in the first paragraph on Page 7, lines 11 to 14.

Comment 18: Page 7, Line 34. Capitalize Fear.

Response 18: Edit made.

Results

Comment 19: Page 7, Line 45. RIS is the term used here again then RRB in the next paragraph. Speaks to the point above about cleaning this up some. It isn't clear to me whether RIS can include a Sensory behaviors scale as that seems more applicable to the way RSM is described above.

Response 19: we have now clarified this, please see our response to your comment 12 above.

Comment 20: Page 8, Lines 6-12 and throughout paper. Sometimes the term fear is used and sometimes fears is used in a way that is not consistent throughout.

Response 20: We now use "fear" throughout the manuscript.

Comment 21: Page 8, Line 19. Add " before Just Right"

Response 21: Edit made.

Discussion

Comment 22: Page 8, Line 42. Remove parentheses around 2000.

Response 22: Edit made.

Comment 23: Page 9, Line 58 and onto next page. This sentence is very long. Consider revising.

Response 23: Edit made.

Comment 24: You might consider bringing back in the idea of how this work could inform intervention as you mention in the introduction. There is evidence that behavioral intervention and cognitive behavioral therapy (see Judy Reaven's work) is effective in teaching children with ASD ways to self-regulate and/or face their fears. It is possible that, like many areas for children with ASD, they would benefit from explicit teaching of how to face and cope with fears so that

their reliance on RRBs could take the same form as their TD counterparts by using more cognitive and emotional forms of managing fears and anxieties when possible.

Response 24: We have now provided discussion on potential clinical/treatment implications of our findings, please see track changes on Page 10, lines 24-25 and Page 11, lines 1-2.

General Comment: In summary, I recommend that the authors address the substantive concerns detailed here, as well as the minor stylistic issues, and resubmit a revised version of the manuscript for final review.

Response: We thank the reviewer for very constructive feedback and useful suggestions, we hope that changes made address in a satisfactory manner all the points reviewer has raised.