

## **Executive function skills are linked to restricted and repetitive behaviours: Three correlational meta analyses**

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**Abstract:**

There is a consensus on the centrality of restricted and repetitive behaviours (RRBs) in the diagnosis of Autism Spectrum Disorder (ASD), yet the origins of these behaviours are still debated. We reconsider whether executive function (EF) accounts of RRBs should be revisited. EF deficits and high levels of RRBs are often pronounced in individuals with ASD and are also prevalent in young typically developing children. Despite this, the evidence is mixed, and there has been no systematic attempt to evaluate the relationship across studies and between task batteries. We examine recent evidence, and in three highly powered random-effects analyses (N= 2964), examine the strength of the association between RRB levels and performance on set shifting, inhibitory control, and parental-report based EF batteries. The analyses confirm significant associations between high levels of the behaviours and poor EF skills. Moreover, the associations remained stable across typical development and in individuals with ASD and across different types of EF measures. These meta-analyses consolidate recent evidence identifying that cognitive mechanisms correlate with high RRBs that are seen in individuals with ASD, as well as in typical development. We propose that the EF account may be critical for guiding future interventions in ASD research.

*Keywords: systematic review, meta-analysis, executive function, restricted and repetitive behaviours, autism*

**Lay summary:**

Restricted and repetitive behaviours (RRBs) are diagnostic criteria for Autism yet also common in typical development, and if they persist over time some can have a negative impact on learning and social acceptance. The present meta-analyses found that high levels of RRBs related to poor performance on set-shifting and inhibitory control tasks, as well as high ratings on parental report scales. Future studies should create interventions that aim to improve these skills as they may help manage challenging RRBs.

Many major puzzles in our understanding of Autism Spectrum Disorder (ASD) surround the nature of one of the two central diagnostic features, restricted and repetitive behaviours (RRBs) (American Psychiatric Association, 2013). In particular, the literature is unclear about how these behaviours relate to other aspects of the diagnosis, including comorbid social-communicational problems and underlying cognitive skills (e.g. Lopez, et al., 2007; Van Eylen, et al., 2015). In the 1990s great expectations were placed on cognitive models to explain the joint components of the diagnosis, notably theory of mind (Baron-Cohen, 1995), central coherence (Frith, 1989; 2008) and executive function (EF: Russell, 1997). The first two of these cognitive accounts are either agnostic about the link between cognitive factors and RRBs (e.g. Frith, 2008) or simply suggest that the explanation of the social/communication problem, like undeveloped ‘theory of mind’, is ‘related’ to these RRBs (Baron-Cohen, 2000, pp. 78-79) without explanation. The third set of theories has been more explicit about hypothesised links between EF skills and these behaviours (Russell, 1997). However, more recent analyses raise doubts about whether this third candidate theory can explain RRBs, or even the social and communicative difficulties in ASD. We re-open this debate, first by outlining the centrality of RRBs to the current diagnosis of ASD, then by summarising a shift in the theoretical focus of accounts of the nature of these behaviours, playing down these cognitive accounts. We re-evaluate the possible role of EF and present three meta-analyses to re-examine the evidence for a possible role of EF skills in this neglected diagnostic feature of ASD.

## **[1] Restrictive and Repetitive Behaviour in Autism Spectrum Disorder**

RRBs have been a part of the definition of ASD since Kanner's (1943) and Asperger's (1944) original descriptions of this neurodevelopmental disorder. Kanner (1943, p 245, his italics), for example, writes of an '*anxiously obsessive desire for the maintenance of sameness*', and it is not surprising that contemporary analyses of ASD show a high comorbidity between ASD and anxiety problems (e.g., Rodgers et al., 2012; Lidstone et al., 2014). Since the 1990s (DSM-IV: APA, 1994, ICD-10: WHO, 1992), RRBs have been divided into four sub-groups: stereotypies, preoccupation with objects, restricted interests and non-functional routines. They are manifest in actions that range from rocking and hand flapping to very specific food and routine preferences, such as eating only pizza. Parents often identify these behaviours as the most challenging ASD characteristics to manage (e.g. South, Ozonoff & McMahon, 2005), as they can create barriers to learning opportunities and social interactions (Harrop, McBee & Boyd, 2016). Analysis of the origins and nature of RRBs can guide research on the outcomes of the disorder, and help design interventions to remediate these behaviours

We conducted a search of RRBs in the ISI (Clarivate Analytics) Search Engine. Following the definitional changes in the DSM-5 (2013), output more than doubled: 767 articles in the 5-7 years since the change between 2014 and 2020 (to April) compared to 258 articles in the six years between 2008 and 2013. The rapid increase in research has led to more thorough analyses, often concerning how the broad range of RRBs cluster together. This has highlighted a lack of a universal definition

of the term. Factor analytic studies, for example, increasingly divide RRBs into dichotomous groups: “low-” and “high-level” behaviours (Cuccaro et al., 2003; Honey, Rodgers & McConachie, 2012). The “low-level” RRBs consist of motor actions like rocking or hand flapping and a preoccupation with objects (including collecting unusual items, like fluff from carpets), whereas the “high-level” behaviours consist of restricted interests and non-functional routines, like obsessively repeating facts about a special interest, such as Star Wars or Harry Potter (Turner, 1999)

### **Measurement Issues**

There is no ‘gold standard’ RRB measure. Widely used questionnaires implement a variety of response methods ranging from calculating frequency or intensity to identifying which behaviours are repeated (Honey, et al., 2012). Some also include several of these metrics, making it difficult to compare the same behaviours, yet alone different ones, across different measures (South, Ozonoff & McMahon, 2007). In addition to the various metrics, types of assessments also differ. Although some measures comprise observations and interviews used to diagnose ASD (e.g. the Autism Diagnostic Observational Scale, ADOS: Kim, & Lord, 2010, and the Autism Diagnostic Interview-Revised, ADI-R: Rutter, LeCouteur & Lord, 2003), others involve parental questionnaires that were created for the sole purpose of assessing RRBs (e.g. the Repetitive Behavior Questionnaire, RBQ: Turner, 1995). To complicate matters further, some scales such as the ADI-R consist of 12 items, while others, like the Repetitive Behaviour Scale-Revised (RBS-R: Lam & Aman, 2007), include up to 44 items divided into as many as six sub-scales. This diversity between measures has made it difficult to draw any conclusions concerning which of the

existing tools are sensitive enough to do more than capture the wide variety of RRBs. This is especially the case as most measures have not been used frequently enough to test and analyse their concurrent and construct validity (Honey et al. 2012). This meta-analysis explores the diversity between measures in more depth to examine whether such differences should be considered further.

Evidence for measurement difficulties can be seen in South, Ozonoff and McMahon's (2007) study, assessing one group of individuals with ASD on three measures (ADOS, ADI-R and the Repetitive Behaviour Interview, RBI: Turner, 1997). They found concurrent validity in terms of associations between their cognitive flexibility measures and RRBs using the ADOS and ADI-R, but not with the more specific RBI. These differences could be caused by the different levels of detail in each measure. Specifically, the ADI and ADOS are commonly used to diagnose ASD and hence rely on fewer questions, whereas the RBI is more comprehensive and created for the sole purpose of assessing the nature and extent of RRBs. Not only do findings like these imply that measurement issues may have negative implications for our understanding of the construct itself, they also stress the need for a systematic review of the RRB tools. Given the complexity of RRBs, it is perhaps naive to assume that we can develop one 'gold standard' measure. Nonetheless, researchers should consider the different features of the available measures against a range of criteria, and in the light of the specific question they are asking (Honey et al., 2012). This would then also make it easier to evaluate the various theoretical accounts in the field.

## **[2] The move towards a more complete account of Restrictive and Repetitive Behaviours.**

The past two decades have witnessed a shift of emphasis within theoretical analyses of the nature and origins of RRBs. For the decades before and after Russell's (1997) influential analysis, many researchers focused on the link between a delay in the control of action and the persistence of these behaviours. The typical increase in both lower- and higher-level behaviours towards the end of the preschool period and decline thereafter coincides with the manifestation of EF skills. This close correlation suggested a causal relationship (Turner, 1997). Over the past decade this view has received much critical scrutiny:

“Taking a developmental perspective, it seems unlikely that EF could have a direct causal role since RRBs emerge so early in typical development, hence it may be more appropriate to consider the *effect* of repetitive behaviors on neurocognitive functioning, than any causal role” (Leekam, Prior & Uljarevic, 2011, p 578).

In this section, we review the an alternative to the EF theory which is hinted at in this quotation – the developmental trajectory account. We will argue that neither is incompatible with an EF approach: indeed this review was motivated by the possibility that EF account could benefit from a more holistic combination with this area of theorization.

Leekam, et al.'s (2011) developmental account suggests that RRBs are immature responses that are maintained more strongly within the behavioural repertoire of individuals with ASD. To explain this process, they suggest that neurobiological changes must be traced alongside behavioural ones. They highlight the importance of the development of the corticostriatal circuits in early childhood. This account is largely based upon Thelen's (1981) perspective on skilled motor action in which a high prevalence of stereotypies in the first year of life is caused by slow cortical maturation, as motor actions are not yet under

voluntary control (Tinbergen, 1951). At the end of the first year, motor behaviours become more goal directed, and RRBs more varied. This suggests that RRBs are more likely to be released within specific events which provoke extreme arousal states (high or low), and that triggers for RRBs need to be understood within a context that balances developmental and environmental factors. Leekam et al. propose that Thelen's account can be applied to the broader category of RRBs in ASD. Accordingly, these behaviours are immature responses that are a normal part of early development, which come increasingly under control as infants begin to develop goal-directed actions. The developmental approach would benefit from being linked to a cognitive model that explains how repetitive behaviour changes with age. Leekam et al's (2011) observation that stereotypies reduce over time in typical development is widely supported in the literature (e.g. Cevikaslan et al., 2014; Uljarevic et al., 2017)

It is plausible that typically developing infants acquisition of goal-directed actions explains the reduction in lower level behaviours but this theory is less likely to account for the higher-level RRBs, as these follow a different trajectory, in which they first increase, then decline around the age of 5-6 (e.g. Cevikaslan et al., 2014; Evans et al, 1997; Uljarevic et al., 2017). Without an additional dimension, this account would struggle to explain what purpose the higher level RRBs have, and what it is that drives such changes.

In addition to focusing on their developmental trajectory, Leekam et al. (2011) suggest that RRBs become more likely to be triggered by specific events, particularly extreme physiological arousal states (high or low). This echoes an early RRB

account that they are caused by hyper- or hypo-arousal (Hutt & Hutt, 1965). The hyper-arousal prediction suggests that these behaviours are coping mechanisms that develop to reduce high-arousal or anxiety. Goodall and Corbett (1982) expanded this theory by proposing that RRBs may develop to regulate under-arousal caused by a lack of stimulation from the environment. The suggestion that anxiety plays a central role in RRBs, is perhaps not surprising, as it was highlighted in Kanner's (1943) original paper. A recent meta-analysis by Steensel, Bogels and Perrin (2011), however, refocused interest in this account by identifying that as many as 40% of individuals with ASD also met the criteria for an anxiety disorder. Indeed higher-level RRBs commonly associate with anxiety levels in children who are typically developing (Evans, Gray, & Leckman, 1999; Laing et al., 2009; Zohar & Felz, 2011) and with ASD (Rodgers, et al., 2012; Uljarevic & Evans, 2017). Findings like these have suggested that higher-level RRBs serve the purpose of controlling the environment and thus reducing anxiety. Some support for this can be seen in a study by Lydon, Healy and Dwyer (2013) that investigated heart rate before, during, and after challenging behaviours in three children diagnosed with ASD. Their findings suggest that, for some individuals with ASD, the behaviours serve to increase arousal and to sustain a preferred state of heightened arousal. Despite the interesting focus, these results do not reveal a causal pathway through which arousal and anxiety lead to the manifestation of these behaviours. This highlights the need to re-open the EF account as it is possible that anxiety and RRBs are part of a causal chain between poor cognitive control leading to hyper-attentiveness to negative information, creating anxiety and then RRBs (e.g., Spiker et al., 2012).

The emphasis on goal directed actions in the developmental account leads easily to the proposal that the different RRB trajectories are driven by an individual's EF skills. A widely cited definition of EF skills is "the ability to maintain an appropriate problem-solving set for attainment of a future goal" (Ozonoff et al., 1991, p. 1083). Examples of these skills are planning, inhibitory control and the flexibility of thought and action. Although Leekam et al. suggest that EF deficits are not vital for the development of RRBs, they stress the importance of corticostriatal circuits. As Langen et al. (2011, p2) state "cognitive models have provided valuable hypotheses for how neurobiological circuitry might be disturbed in repetitive behaviour". Given that the main function of the corticostriatal circuit is to control goal-directed behaviour, this statement points to EF processes, or even bidirectional influences between behaviour and neurophysiology. Evans, Lewis and Iobst (2004) suggest that variable EF skills and RRB trajectories across disorders may be caused by how different cognitive processes are governed by different regions of the orbitofrontal cortex. There is ample support for variable RRB levels across disorders. For example, individuals with Williams Syndrome have been found to engage in more stereotypies than those with Prader-Willi syndrome (Royston et al., 2018). Individuals with ASD and OCD are thought to engage in significantly more RRBs than typically developing children (Zandt, Prior and Kyrios (2007). Similar variability has been found in the EF literature as the same study by Zandt, Prior and Kyrios found that individuals with ASD performed better on inhibitory control tasks than individuals with OCD. The EF hypothesis could then possibly account for the frequency of RRBs in ASD, as individuals with ASD may be more impaired on EF skills. At the same time, it can help explain the wide range of these behaviours, as particular

skills may be responsible for individual RRBs. Moreover, it can account for the heterogeneity within disorders, as well as the change from RRBs across typical and atypical development.

### **Do individuals with ASD show EF impairments?**

One reason why the link between EF and RRBs has been contested is that extensive research, numerous reviews, and meta-analyses on the definition of EF impairments in ASD over the past fifteen years, is that the role that these skills play in the disorder remains unclear. For example, Geurts, Corbett and Solomon's (2009), discerned no firm evidence in 29 studies for a cognitive flexibility deficit in adults with ASD. They focused largely on tasks that they considered to have high ecological validity, using mechanistic approaches (e.g. task switching paradigms that warned participants about a rule change, and presented switch trials throughout the task). However, they found clear impairments on tasks that did not meet this criterion, such as the Wisconsin Card Sorting Task (WCST: Berg, 1948). Despite these positive results, Geurts, et al's (2009) paper has been widely cited as evidence against the EF hypothesis (Web of Science= 162) and it steered some researchers away from the EF explanation.

Perhaps paradoxically, Geurts and colleagues' subsequent research has identified the EF profile in ASD. In several meta-analyses, they have found strong prepotent response inhibition and interference control inhibition impairments ( $n=41$ ,  $g=.55$  and  $.31$ , respectively) (Geurts, van den Bergh & Ruzzano, 2014), as well as planning impairments ( $n=50$ ,  $g= 0.52$ ) (Olde Dubbelink & Geurts, 2017) in individuals with ASD. These positive links with inhibition and planning cast doubt on the

suggestion that individuals with ASD are unimpaired on EF skills. In addition, recent meta-analyses by Lai et al. (2017) and Demetriou et al. (2018) find even stronger evidence for the view that overall EF performance, as well as performance on separate EF skills are essential to control thoughts and behaviours in individuals with ASD. Demetriou et al.'s analysis consisted of 235 studies (n= ASD= 6816, Control= 7265). They found a moderate effect size ( $g= 0.49$ ) for overall measures, suggesting that individuals with ASD are significantly more impaired on EF than control groups. This effect also applied evenly across the 6 individual EF domains (concept formation, mental flexibility, fluency, planning, inhibition and working memory) ( $g=0.46-0.55$ ). Lai et al.'s analysis, on the other hand, of 98 studies (n=5991, ASD= 2985, Control= 3005), concentrated on a narrower age range of younger children and adolescents. Another difference was that Lai et al's. analysis only examined individual EF domains (verbal and spatial working memory, flexibility, inhibition, generativity and planning). Like Demetriou, they found moderate to strong effect sizes for all individual skills ( $g=.57-.67$ ), although a lower inhibition effect ( $g=.41$ ). These recent and more thorough analyses suggest both that EF impairments play a crucial role in ASD and that separate skills are important, despite controversy of what overarching EF is. Thus it is now more relevant to examine the theoretical and clinical implications of the EF account. Initially we wanted to examine the associations between RRB levels and Miyake et al.'s (2000) three "foundational" EF skills; set shifting, inhibitory control and working memory. Unfortunately, not enough studies (<10) have examined the relationship between RRBs and working memory, so our analyses only focus on set shifting and inhibitory control.

### **Are EF skills related to the high levels of RRB in ASD?**

A spurt of new research offers renewed support linking elevated RRB levels to EF impairments such as set shifting (e.g. Jones et al, 2017; Miller, Ragozzino, Cook, Sweeney & Mosconi, 2015), inhibitory control (e.g. Jones et al, 2017; Thakkar et al., 2008; Mosconi et al., 2010) and planning (e.g. Van Eylen et al., 2015). Jones et al. (2017), for example, investigated the relationship between RRBs and multiple EF skills in 100 adolescents with ASD and found significant associations with set shifting and inhibitory control, but not planning. Similarly, Miller et al. (2015) found that in individuals with ASD their overall set shifting errors predicted RRB levels. Studies like these have led to the call for set shifting interventions to remediate RRBs in ASD (e.g. Mostert-Kerckhoffs et al., 2015), and highlight the need to re-open the EF account. Some studies suggest that we need to consider EF skills in combination with genetic components, specifically, the need for gene-brain-behaviour models of ASD using set shifting (Yerys et al., 2009) or inhibitory control (Thakkar et al., 2008) as possible links between the components. Thakkar et al., for example, found that elevated RRB levels in ASD participants related to hyperactive response monitoring in the rostral anterior cingulate cortex (rACC) during an antisaccade task. These findings complement a genetic account (see Lewis & Kim, 2009) but also highlight the importance of cognitive factors, strengthening the view that the EF account must be re-examined. Such links, however, are not pervasive, as some recent investigations have also failed to find relationships between RRBs and set shifting (Ozonoff et al., 2004), inhibitory control (Joseph & Tager-Flusberg, 2004) and planning (e.g. Jones et al., 2017). The inconsistent literature makes it timely to examine the relationships further through a

meta-analytic framework to assess the strengths of the proposed relationships, and evaluate if EF interventions may have the potential to help manage challenging RRBs.

### **Task impurity**

Several explanations have been given for the inconsistent findings in the literature. First, like RRBs, EF measures have consistently been scrutinised in terms of their ecological validity (e.g. Kenworthy et al., 2008; Rabbitt, 1997). Given that the executive system incorporates a variety of skills (Miyake et al., 2000) it is not surprising that psychometric measures need to accommodate such diversity. Geurts, Van Den Bergh and Ruzzano's (2014) meta-analysis, for example, confirmed that WCST impairments that relate to RRBs may identify cognitive inflexibility, but might also identify difficulties with staying on task, learning from feedback and/or inhibiting irrelevant information. EF tasks have commonly been criticised for their complex structures (Burgess et al., 1998), and the impure nature of the WCST task has been highlighted as a clear example. It may also tap into cognitive flexibility (Everett et al., 2000), working memory (Medalia, Revheim & Casey, 2001) and inhibitory control (Geurts, Corbett & Solomon, 2009) skills. Nevertheless, Miyake et al.'s (2000) confirmatory factor analysis identified that the WCST task loaded onto the factor 'shifting' and not the two other factors. Thus, the overall conclusion that there are no clear shifting impairments in individuals with ASD may be mistaken.

Findings like these led to the development of EF rating scales completed by parents or teachers, such as the Behaviour Rating Inventory of Executive Function (BRIEF: Gioia et al., 2000). Whereas psychometric tasks require a response to a single

event and are conducted in carefully controlled environments, EF performance in the real world involves a stream of tasks (Dawson & Marcotte, 2017). The BRIEF consist of two smaller scales, the The Behavioral Regulation Index (BRI) and the Metacognition Index (MI). The BRI consists of four skills: Shift, Inhibit, Self-Monitoring and Emotional Control. The MI comprises five skills: Plan/Organize, Initiate, Task Monitoring, Working Memory, and Organization of Materials. The outcomes on both scales of the BRIEF have been found to be consistent with clinical expectations; they correlate with biological markers, and even show predictive relationships with academic skills (Isquith, Roth, & Gioia, 2013). This leads nicely into a second possible reason why the inconsistent findings as EF rating measures may have moderated the results. It has been suggested that rating scales have a higher ecological validity (Rabbitt, 1997), and consequently may be the only measures that can reliably predict EF impairments. To date, many studies find that parent-based EF ratings are not correlated with scores on performance based EF tasks in children with ASD (Teunisse et al., 2012) or other neurodevelopmental disorders such as ADHD (Mahone & Hoffman, 2007; Toplak et al., 2008). Mahone and Hoffman, for example found that 2-5 year old children with ADHD improved significantly with age on performance-based EF measures. In contrast, parent ratings remained relatively stable during this age range. These patterns again suggest that parent ratings are measuring a different aspect of the EF construct than most performance-based measures. Despite widely reported concerns like these, researchers often interpret the findings in rating scales and performance-based tasks in the same way. This may be problematic, as Toplak, West and Stanovich (2013) found not only low reliability between scales and psychometric measures ( $r = .19$ ), they also found that each

assessed different levels of cognition, namely cognitive abilities and goal pursuit achievement. As well as highlighting the need to examine potential moderating factors further, these findings emphasise the rationale for the third meta-analysis, reported below. Parent-based ratings may be useful when behavioural problems interfere with performance-based testing, and they can provide supplemental data that may relate more to “real-life” situations. We examine if measuring EF skills by behaviour vs. parental report makes a difference in regards to their relationship with RRBs.

### **Predictions**

Numerous possible explanations for RRBs have been proposed but their cause is unknown, since no hypothesis has yet stood up to rigorous evaluation. The nature of the database has shifted slightly since Leekam et al’s (2011) review, making it appropriate to re-examine the link between EF skills and RRBs. Not only have recent studies found strong links with EF skills, there is also not enough evidence to support an alternative framework to explain the developmental trajectory of these behaviours. Nonetheless, there is still ample evidence to suggest that some task or sample characteristics may play a key role in the relationship, albeit if the EF impairment may not be able to explain the full picture.

In order to assess the relationships between RRBs and set shifting, inhibition and parental control scores, a correlational meta-analytic approach was applied. This type of methodology is useful as it assesses the overall strength of relationships by combining data from all the available findings in the literature. One criticism of the approach is that analyses may combine results that are not comparable, since they have implemented different measures or statistical methods (Rosenthal & DiMatteo,

2001). Other authors however, argue that a certain degree of dissimilarity needs to be accepted in order to allow for generalisation (Smith, Glass & Miller, 1980). Meta-analysis can give us an indication of the strength of a relationship, but it has been further suggested that it does not necessarily give us a clearer understanding of its nature, particularly any directions of causality. Nonetheless, correlational relationships specify important research needs and identify children who may benefit from specific interventions.

The inconsistent literature reported above makes it difficult to make strong predictions. Previous meta-analyses indicate strong general EF impairments in ASD, apart from inhibition, in which the role is less clear (Lai et al., 2017). It is therefore possible that we find stronger effects in the first meta-analysis to be conducted, on the relationship between set shifting and RRBS, than in our second on the links between repetitive behaviour and inhibitory control. For the parental report analysis, we may find a strong overall association of EF with RRBS, as not only are we looking at the Meta-cognition Index (MI; Gioia et al., 2000) scale in which shifting and inhibition are combined, but parental report measures have also been argued to be more ecologically valid than psychometric measures (Kenworthy et al., 2008). If we take the inconsistent evidence into account, it is also possible that we will not find significant relationships between any of the EF skills and repetitive behaviours, calling the EF hypothesis into question. If we do find an overall relationship we need to highlight moderators that should be explored further.

## **Method**

### **Systematic literature Search and inclusion criteria:**

This systematic review and meta-analysis was performed in accordance with the PRISMA guidelines (Moher et al., 2009) and those specifically for correlational meta-analysis (Quintana, 2015). To collect the relevant data on the relationship between EF abilities and levels of RRBs, we searched Scopus and the ISI Search Engines [initially on 10.10.2017]. The following combinations of keywords were used: restricted, repetitive behaviours OR stereotypes OR insistence of sameness OR circumscribed interests AND executive function OR set shifting OR planning OR working memory OR inhibition OR inhibitory control OR BRIEF). These produced 177 results in Scopus and 138 in ISI. We closely examined previous reviews and asked leading researchers in the field (n=10) to provide unpublished data on the topic to avoid the risk of possible publication bias, or inaccessible data that we needed to calculate an effect size. Two provided additional data on set-shifting. The results made it possible to run set shifting, inhibitory control and parental-report based questionnaire analyses, but not planning and working memory as too few studies (<10) measured the relationships between these skills and RRBs. See Figure 1 for PRISMA flow diagram of the studies that were included in the meta-analysis, and Tables 4, 5 and 6 for summary data of the studies included in the set shifting, inhibitory control and parental report analyses respectively.

### **Statistical dependence of the samples**

If a paper reported multiple effect sizes, these were included and treated as separate studies if they fulfilled one of three criteria:

1. The effect sizes were independent and representative of different diagnostic groups (Borenstein et al., 2009).
2. Individual differences were examined within a specific participant group (e.g. ASD individuals divided into two groups, low- and high- functioning individuals, based on their IQ scores).
3. A study assessed participants on multiple tasks that measured different EF skills (e.g. one set shifting and one inhibition task).

This rule did not apply, however, if the same participant group was tested on several set shifting or inhibitory control tasks, if a study included correlations for several task outcomes (e.g., perseverative errors and reaction time), or if participants were assessed on multiple RRB measures. To include the same comparison group in the same analysis several times would have violated the assumption of statistical independence, rendered the standard errors and thus made the confidence intervals inaccurate. We created further inclusion criteria for our analyses when this occurred:

1. If a study reported several outcome measures, we always chose the most widely reported outcomes for our analysis, as these were better comparisons. As a result, if a study: reported perseverative errors and reaction times (e.g. Dichter et al., 2010), we always chose perseverative errors; if frequency and duration were presented (e.g. LeMonda et. al, 2012), we included the effect size for frequency; if it reported commission (incorrect button press) and omission (no button press) rates

for set shifting scores (De Vries & Geurts, 2012), we reported the effect size for commission rates, as these are more comparable to perseverative errors and frequency scores.

2. If a study reported several correlations for different EF tasks with the same measure outcome, we included the correlation from the most widely used task. For example, in Van Eylen's (2015) study, the correlation for the WCST task was chosen over the Switch task (Rubia, Smith & Taylor, 2007), and the Go/No-Go task was chosen (e.g. Fillmore et al., 2006) over the Flanker task (Christ et al., 2011). In Mostert Kerckhoffs et al.'s (2015) study, effect sizes were listed for the auditory stimulus condition (SSA) and the visual stimulus condition (SSV) (tasks from the Amsterdam Neuropsychological Tasks, De Sonneville, 1999). We reported the correlation for the visual task, since other widely used shifting tasks (e.g. the WCST) rely heavily on visual skills. Finally, in Joseph and Tager-Flusberg's (2004) study, two types of inhibition tasks were reported, the Day and Night (Gerstadt, Hong & Diamond, 1994), and the Knock and Tap (Korkman, Kirk & Kemp, 1998) task. We decided to report the correlation for the Knock and Tap task, since it relies heavily on motor skills, making it similar to the frequently reported Walk/Don't Walk task, while the Day and Night task requires good verbal skills which are known to be compromised in ASD.

3. If participants in a study were assessed on multiple RRB measures we included the most widely used one. For instance, in Van Eylen et al.'s (2015) study, effect sizes for the social responsiveness scale (SRS, Roeyers et al., 2011) and the repetitive behaviour scale-revised (RBS-R, Bodfish et al., 1999) were reported. We used the RBS-R correlation, as this is more

widely used (Honey et al., 2012). In other studies, behaviours were measured through two widely used diagnostic measures, the Autism Diagnostic Interview (ADI, Le Couteur et al., 1989) and the Autism Diagnostic Observation Schedule (ADOS, Lord, et al., 1989). When a study provided correlations for both of these measures, we report the observational ADOS, as it includes a wide range of behaviours and is based on observation (following Turner, 1999). This exclusion criteria also meant that we included some non-traditional RRB measures such as the CY-BOCS (Zandt, 2009). We decided to include these measures as no traditional measures were used and we wanted to include as many studies as we possibly could.

### **Statistical analyses**

We ran random-effects models to estimate the overall means, to account for heterogeneity within studies, since a wide variety of tasks had been used to assess both RRBs and EF skills. Pearson *r*-values were converted to *z* scores to ensure that measures were normally distributed. For this analysis, the packages “metafor” (Viechtbauer, 2010) and “robumeta” (Fisher & Tipton, 2015) for *R* (R Development Core Team, 2015) were used. Following Cohen (1988), we interpreted a correlation coefficient of .10 as weak, of .30 as moderate and .50 or larger as strong. Between studies heterogeneity for each measure was assessed using the index of inconsistency (*I*<sup>2</sup>). This calculates a percentage of heterogeneity resulting from study differences that is not due to chance; therefore, larger values indicate greater heterogeneity. Forest plots were created for all analyses.

### **Measures of data quality**

We assessed whether non-significant results may have been suppressed from the literature. As the response rate to our e-mails asking for unpublished data was poor (2 out of 10 requests), this was particularly important. We examined publication bias through funnel plots, as studies with stronger effects may be more likely to get published and thereby be included in a meta-analysis. However, this type of analysis only offers a subjective measure of potential publication bias. Egger's regression test (Egger et al, 1997) was employed to offer an objective view. This is best suited to small meta-analyses (<25 studies) and evaluates if effect estimates and sampling variances for each study are related.

### **Moderator analyses**

In all comparisons, we ran meta-regression analyses to identify potential moderators for the relationships. These were: age, diagnosis (ASD versus TD), type of RRB scale (diagnostic versus RRB specific) and testing modality (experimenter-administered versus computer-administered). Age and diagnosis were examined further to explore the developmental trajectory for the relationship between EF skills and RRBs. If the continuous age effect was significant, we ran an additional analysis in which we split the factor into three age categories: child (0-11 years old), adolescent (12-18 years old) and adult (19 and above), following Van Eylen et al. (2015). This was to pinpoint whether the relationship is at its strongest during a particular stage of development. We explored the moderating effect of testing modality (computerised versus experimenter administered) because it has been suggested that the difficulties of individuals with ASD in experimenter-administered EF tasks may be due to their social interaction problems (e.g. Perner & Lang, 1999). For the RRB scale moderator analysis, we divided the scales into

two types of assessment: diagnostic and specific. The diagnostic measures comprised observations and interviews used to diagnose ASD (e.g. the ADOS and ADI-R), whereas the specific measures were created for the sole purpose of measuring RRBs (e.g. the RBQ). We explored the differences between these two types as, although they have a similar structure, large differences are found between them. This is likely to reflect the depth of analysis. Whereas the ADI-R uses 12 items to assess RRBs, the RBS-R includes 44-item questions divided into six sub-scales (Lam & Aman, 2007). These differences might produce variations in the results. For our set shifting analysis, we ran a moderator analysis that examined types of EF task (WCST versus others), since research has found particularly strong relationships between performance on the WCST task and RRB levels (e.g. South et al, 2007). We wanted, additionally, to examine the effect of IQ on the relationship between EF and RRB levels, but were unable to do so as insufficient information was available.

## **Results**

The first analysis tested the hypothesis that there is an association between high levels of RRBs and poor performance on set-shifting tasks. The second examined the strength of the relationship between RRBs and inhibitory control measures. The final analysis investigated if a similar relationship can be found between high RRB levels and performance on parental-rated EF measures. Note that in all analyses the EF measure is of errors, so both scores (the EF measure and RRBs) are scored in the same direction with higher values indicative of poor psychological functioning.

### **Meta analysis 1: RRB levels and performance and set shifting tasks**

The performance based set shifting analysis revealed a summary correlation and 95% CI indicative of a significant, but modest relationship with RRB levels [ $r = 0.31$ ; 95% CI (0.19, 0.41),  $p < 0.0001$ ]. Figure 2 presents a forest plot of effect sizes. The contour-enhanced funnel plot (Figure 3) indicates a low risk of publication bias, as it does not show an over-representation of effect sizes in the significance contour and points fell on both sides of the summary effect size. Egger's regression confirmed this by revealing no overall evidence of small study bias ( $p = .44$ ). Since there was no sign of publication bias we did not run a trim-and-fill analysis (Vevea & Wood, 2005). A set of influence diagnostics, derived from standard linear regression, identified none of the studies as potential outliers (Viechtbauer & Cheung 2010). The degree of heterogeneity between effect sizes,  $I^2 = 65.80\%$  (95% CI; 42.2, 83.2), represents moderate variance. Given that a heterogeneity score around 25.00% is considered low, 50.00% moderate, and 75.00% high (Higgins et al., 2003), we can infer that 65.80% of the proportion of observed variation can be attributed to the actual difference between the studies, suggesting that a few moderators may have had an influence on the results. Accordingly, moderator analyses were performed to identify sources of heterogeneity.

Moderator analyses: We found no moderating effects for age, diagnosis, type of RRB scale, testing mode or type of EF scale.

Table 1 summarizes the effects of each moderator.

### **Meta analysis 2: Inhibitory control scores and RRB levels**

A significant, weak to modest, relationship was found between the inhibitory control measures and repetitive behaviour levels [ $r = 0.21$ ; 95% CI (.04, 0.37),  $p = .02$ ]. See Figure 4 for forest plot. Egger's regression found no evidence for study bias

( $p = 0.38$ ). A contour-enhanced funnel plot showed a low risk of publication bias (see Figure 5). A set of diagnostics derived from standard linear regression identified none of the studies as potential outliers (Viechtbauer & Cheung 2010). The  $I^2$  for the inhibitory control analysis was 77.75% (95% CI; 59.00, 91.18), so moderator analyses were performed to identify sources of heterogeneity.

Moderator analyses: These revealed that part of the heterogeneity in the model between inhibitory control performance and RRB levels was caused by an age effect [ $Q(1) = 4.32$ ,  $p = 0.04$ ]. We examined this effect further and found a positive relationship between inhibitory control and RRBs in adolescents ( $r = 0.29$ ,  $p < .0001$ , CI (0.16-0.42),  $k = 5$ ) and adults ( $r = 0.50$ ,  $p = .0002$ , CI (0.24-0.78),  $k = 4$ ), but not in children ( $r = 0.02$ ,  $p = .88$ , CI (-0.25-0.29),  $k = 9$ ). The strength of the relationship between RRB levels and inhibitory control becomes stronger with age.

We found no effects for diagnosis, testing mode or type of RRB scale. Table 2 summarises of the effects of each moderator.

### **Meta analysis 3: Parent-rated EF scores and RRB levels**

The parent-rated EF analysis showed a summary correlation and 95% CI indicative of a significant, modest, relationship with repetitive behaviour levels [ $r = 0.33$ ; 95% CI (0.04, 0.62),  $p < 0.03$ ]. See Figure 6 for a forest plot. Egger's regression test showed no evidence for small study bias ( $p = 0.44$ ). Our contour-enhanced funnel plot presented in Figure 7 indicated a low risk of publication bias. A set of diagnostics derived from standard linear regression identified none of the

studies as potential outliers (Viechtbauer & Cheung 2010). The degree of heterogeneity between effect sizes was 90.98% (95% CI; 78.3, 97.7). This suggests that a high proportion of observed variation can be attributed to the actual difference between the studies. We carried out moderator analyses to identify the sources of this heterogeneity.

Moderator analyses: These revealed that part of the heterogeneity in the model correlating parent-rated EF measures and RRB levels was caused by the type of RRB measure used, [ $Q(1) = 8.50, p = 0.004$ ]. We split the measures into two factors: diagnostic and RRB specific. We found a positive relationship between parent rated measure and RRBs when assessed through a RRB specific measure ( $r = 0.50, p < .0001, CI(0.28-0.73), k = 7$ ), but not when conducted using a diagnostic measure ( $r = -0.23, CI(-0.86-0.39), p = 0.46, k = 3$ ). This analysis thereby suggests that the relationship between parent ratings and RRBs are stronger in studies that examine RRBs through measures that were created for the sole purpose of measuring RRBs. Finally, we found no evidence to suggest that the relationship between parent rated EF measures and RRB levels was caused by an age effect or diagnosis. Table 3 summarises the effects for all of the moderator analyses.

## **Discussion**

These meta-analyses are the first of their kind to gather all of the available evidence concerning the relationship between RRB levels and performance on set-shifting, inhibitory control and EF parental-report ratings. They revealed moderate but significant associations between high levels of RRBs and errors in two behavioural measures, set shifting and inhibitory control, as well as parental-report. Age and the type of RRB scale moderated the inhibitory control and parental

report results, respectively. Yet diagnosis, testing modality, and type of EF measure did not have an impact on the results. We discuss three implications of these findings, which we examine in relation to each other. First, the significant relationships in each meta-analysis suggest that recent analyses of RRBs have been hasty to reject the EF hypothesis. These skills may play a role in the development of the behaviours or vice versa. Secondly, the extent to which age moderates inhibition should be researched further, as this finding may offer support to a framework in which EF skills must be considered in combination with developmental factors. Finally, future research should examine whether individual factors involved in each type of EF measure may pinpoint what relates them to repetitive behaviours.

The significant associations between RRB levels and poor EF skills suggest that attention needs to be re-focused on the EF account, as the relationship between EF impairments and RRB may be more central in ASD than what has been suggested in key analyses of the origins of repetitive behaviour (Leekam, Prior & Uljarevic, 2011; Lewis & Kim, 2009). Two findings are of particular interest. First set shifting effects were stronger than those for inhibitory control. For ASD individuals, this finding is perhaps not surprising, considering that impairments have often been identified for set shifting skills, but the role of inhibition has been less consistent (Lai et al., 2017). Secondly, the strongest effects were uncovered in the parental report measures. These need to be considered further as they may support the view that these measures are more ecologically valid than psychometric assessments (Rabbitt, 1997). We return to these two effects in more detail. Our findings nevertheless offer support to the recent meta-analyses that show strong evidence for the view that overall EF performance, as well as performance

on separate EF skills are identified in individuals with ASD and appear to be essential to their control of thoughts and behaviours (Demetriou et al., 2018; Lai et al., 2017). They are also consistent with a spurt of recent research linking elevated RRB levels to set shifting (e.g. Miller, et al., 2015) and inhibitory control impairments (e.g. Mosconi et al., 2010).

Despite the significant correlations in the meta-analyses, it is unlikely that the EF account explains the full range and intensity of behaviours which are so prevalent in typical preschoolers and persist in ASD. Autism has a strong genetic component (Tick et al., 2016), but this needs to be partly channelled through other non-shared environmental factors identified by Sandin et al. (2017). The associations we identified highlight that issues to do with self-control are involved in the manifestation of repetitive behaviours, and the need not only to re-open the EF account, but also to explore the relationship within longitudinal research designs, training studies and the possible mutual influences of genetic factors and nonshared environmental influences on the development of EF.

In addition to the research that has found strong evidence for a hereditary component in ASD, studies on the topic (e.g. Van Eylen et al., 2015) and indeed our analyses, have suggested that sample characteristics may moderate the relationships between EF skills and RRBs. Age moderated the relationship with inhibitory control in adolescents and adults, but not children. As previous research has shown that young children with or without ASD engage in high levels of RRBs, and that children with ASD show strong evidence for set shifting but not inhibitory control impairments (Lai et al., 2017), these findings suggest that inhibitory control skills may not initially play a role in the development of RRBs. Indeed, it is possible

that the inhibitory control skills only play a role in the development of higher-level RRBs which necessarily develop later, consistent with Mosconi et al's (2009) finding of relationships between these skills and higher-level RRBs in adolescents, but not children.

Alternatively, it is possible that such age-related findings are caused by measurement issues. The inhibitory control tasks for adolescents and adults used in the current meta-analysis tend to include a wider range of skills than those for children. In the widely used Stroop task, participants identify a colour name printed in different coloured inks, which then interferes with naming the colour of the ink. This involves inhibition of an overlearned response, but it also requires set shifting skills in order to successfully switch between a wide variety of stimuli. This differs in complexity from the child-friendly "knock-don't-knock" task, where participants first match the actions of the examiner (knocking the table top with their knuckles or flat of their palm) and then have to respond with the opposite action to the action of the examiner. Although this task is difficult for children, set shifting demands are not high. The same measurement issues are not present in all the studies included in the set shifting analysis, as two of the widely-used tasks in this meta-analysis were the ID/ED and the WCST. These are very similar as both require the ability to identify a relevant rule, maintain it and shift between different rules, making it possible that set shifting or working memory skills moderate the analysis. There is evidence to suggest that simpler forms of set shifting in children do not relate as closely to RRBs (Dichter et al., 2010). Thus, task demands in tests for adults and children might explain variations between studies between these groups.

In addition to the age-related findings in our inhibitory control analysis, we also identified stronger correlations between RRBs and parental report measures when the skills were measured through RRB specific assessments, not clinical measures. Parental questionnaires are rated by the individuals who know the children best, allowing them to consider behaviours across a wider range of situations and settings, and potentially providing a better perspective on a child's behaviours than a brief test in a clinical setting. The RRB specific measures also cover a wider range of behaviours in a single scale so they consequently identify more, making it possible that the questionnaires tap onto some RRBs that the clinical tools do not. Nevertheless, this association may be explained through the fact that both EF parental reports and RRB specific measures are scored by parents. These factors highlight another potential measurement issue in the literature, and emphasises the need to create more robust and convergent measures to tackle this inconsistency.

Instead of moving away from the EF account, recent research and the results of these meta-analyses lead to a need to consider issues to do with control in explaining RRBs. We are not arguing that these higher functions can explain why such behaviours continue, as the amount of variance still to be accounted for in each analysis was large. We suggest that EFs should be explored in combination with other models. For example, a cognitive framework would offer support for Leekam et al.'s developmental account to explain why stereotypies reduce over time in typical development. Specifically, we suggest that these so-called "immature responses" must be driven by cognitive processes, which take into account that the behaviours reduce as infants develop goal-directed actions. Moreover, a cognitive framework is not incompatible with claims that genetic factors

(Mohrle et al., 2020; Lewis & Kim, 2009) and an imbalance of corticostriatal connectivity play important roles in the development of RRBs (Abbott et al., 2018). Abbot et al. found decreasing connectivity in limbic corticostriatal circuits with age and an underconnectivity in frontoparietal and motor circuits that did not increase with age. A joint model could account for why these behaviours change with age in typical development, and why they persist in individuals with ASD.

A correlational meta-analysis has high informative value in the current context, as it identifies shared variance in large chunks of data between factors notwithstanding this analytic strength, it is important to emphasize also that it does not determine cause or effect. The literature is not yet positioned to conclude on the direction of causality and this remains an important objective for future research. In addition, the current state of the literature has also identified some limitations, such as the difficulty with determining if cognitive abilities confound the picture. Some studies controlled for Full-Scale IQ (FSIQ) and found that the correlation remained (e.g. Jones et al., 2017; Van Eylen et al., 2015), others controlled for VIQ and found that the relationship disappeared (Yerys et al., 2009). Moreover, whereas some studies in our analyses included representative samples of individuals below the average IQ range (Reed et al., 2011; Jones et al., 2018), most of them were confined to individuals in the average IQ range, making it more difficult to explore the role of IQ in the relationship. Due to the limited amount of data on the topic, we were also unable to assess the relationship between RRBs and EF skills in other developmental disorders in which the behaviours and the EF difficulties are prevalent, such as OCD and Williams Syndrome. Surprisingly, we

found a small amount of variance between the EF skills and RRBs. This can suggest that moderators influence the results. For example, a specific RRB sub-type, such as higher-level behaviour may have had an impact on the correlation. For example, it is possible that insistence of sameness and intense interests rely on set shifting and inhibitory control skills as the behaviours require the ability to modify a cognitive rule (Turner, 1997) and inhibit incorrect responses (Diamond, 2013).

Alternatively, gender could have played a role. Recent research by Antezana et al., (2019) found that boys with ASD engaged in high levels of stereotyped behaviours and restricted interests items, and girls in high levels of compulsive, insistence of sameness and self-injurious behaviours. Despite findings like these, it was not possible to include sub-types or gender differences as moderators in our analyses as only one of the studies provided gender data (Bölte et al., 2011) and only a few studies examined behaviours in sub-groups (Boyd et al., 2009; D’Cruz et al., 2013). Future studies should examine this further as Bölte et al. found stronger correlations in males and Boyd et al. found that shifting and inhibition predicted rituals and sameness, but not restricted interests.

The task impurity issues that were highlighted in this analysis highlight that we should assess if similar associations can be identified between RRB levels and other EF skills, such as planning and working memory. We could not consider these in our analyses due to the lack of studies on the topic. Future studies should examine the relationship between the skills and behaviour as the limited research in the area on planning (e.g. Jones et al., 2018; Van Eylen et al., 2014; Sachse et al., 2013) and working memory (Joseph & Tager-Flusberg, 2004; Van Eylen et al., 2014) skills have not predicted RRB scores. Future

research should also focus on disentangling different EF measures to pinpoint what it is about the tasks that make them correlate with these behaviours. We suggest that set shifting measures are of particular interest. Not only did they produce stronger associations than the inhibitory control measures, they were also the only skills that predicted RRBs in children. This is in line with a previous review by Geurts et al. (2009) which concluded that isolating crucial cognitive processes will aid in ultimately resolving the gap between inflexibility in daily life, and the ability measured in the set shifting tasks. Recent developments highlight that set shifting processes such as the ability to activate previously irrelevant stimuli may be of further interest, as these errors have been found to play an important role in set shifting development in both children and adults (e.g. Müller et al., 2006; Maes et al., 2004; 2006). Moreover, a wide variety of RRB measures are used, where the response options range from identifying whether a behaviour is present/not present, its frequency, intensity or its impact upon others, making it hard to compare the responses. Some studies also implement non-traditional measures that are not RRB specific (e.g. CY-BOCS) and some state that they measure RRBs with scales that do not focus on the four types of RRBs identified in the ICD-10 (e.g. the SRS mannerism scale), making it difficult to assess about the full picture. Future research should separate subgroups of lower-level and higher-level behaviours. These two subtypes appear may have different causes and could therefore help to explain the inconsistent results, as well as why we found no associations between high RRBs and poor inhibitory control skills in children. To examine these factors in more depth may help identify more causal links in the relationship

between specific set shifting errors and RRBs. It may also result in clinical or educational interventions that have the potential to help manage difficult and persistent RRBs.

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## Figures

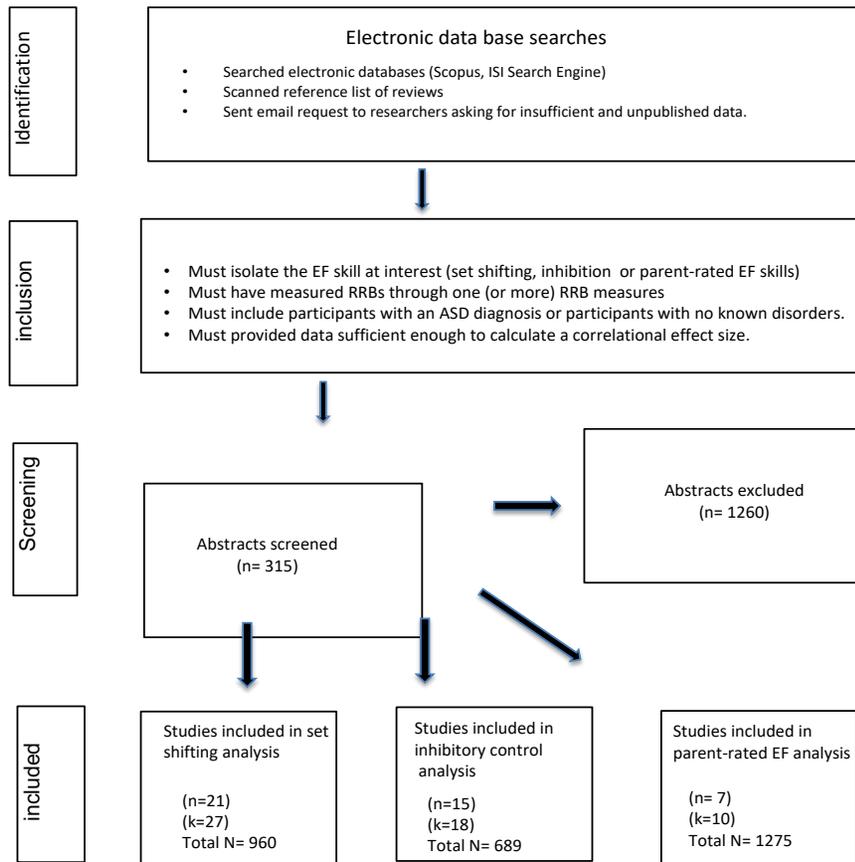


Figure 1: PRISMA flow diagram of the studies that were included in the meta-analysis

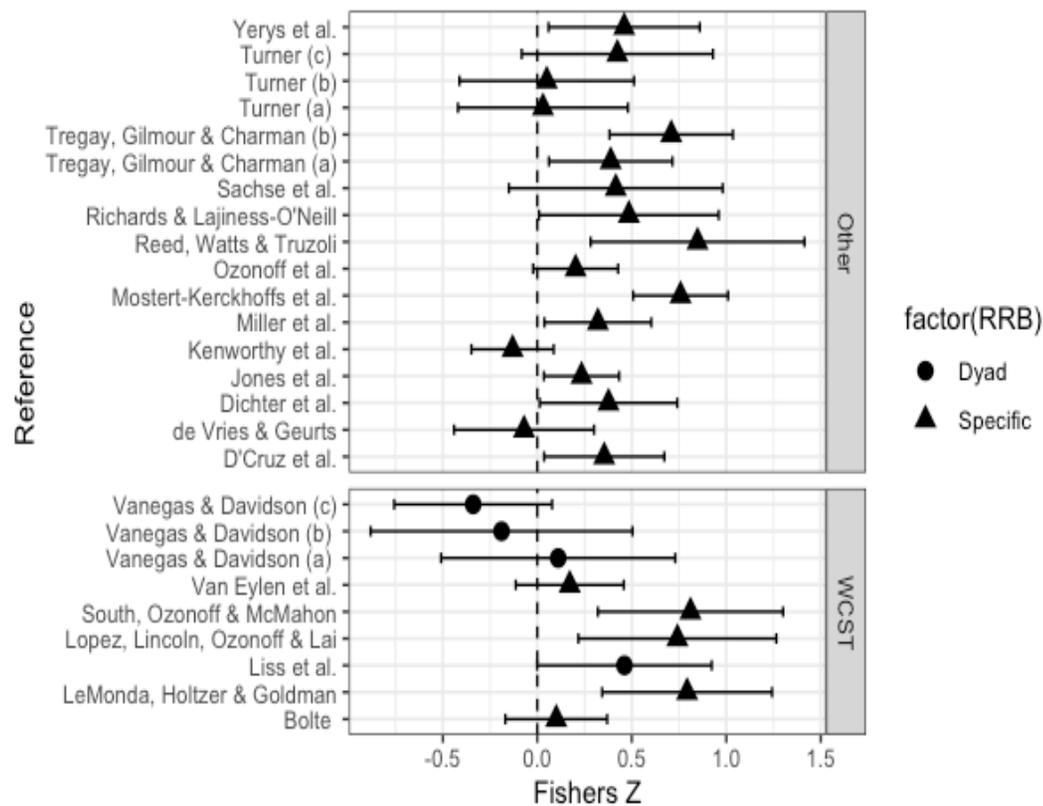
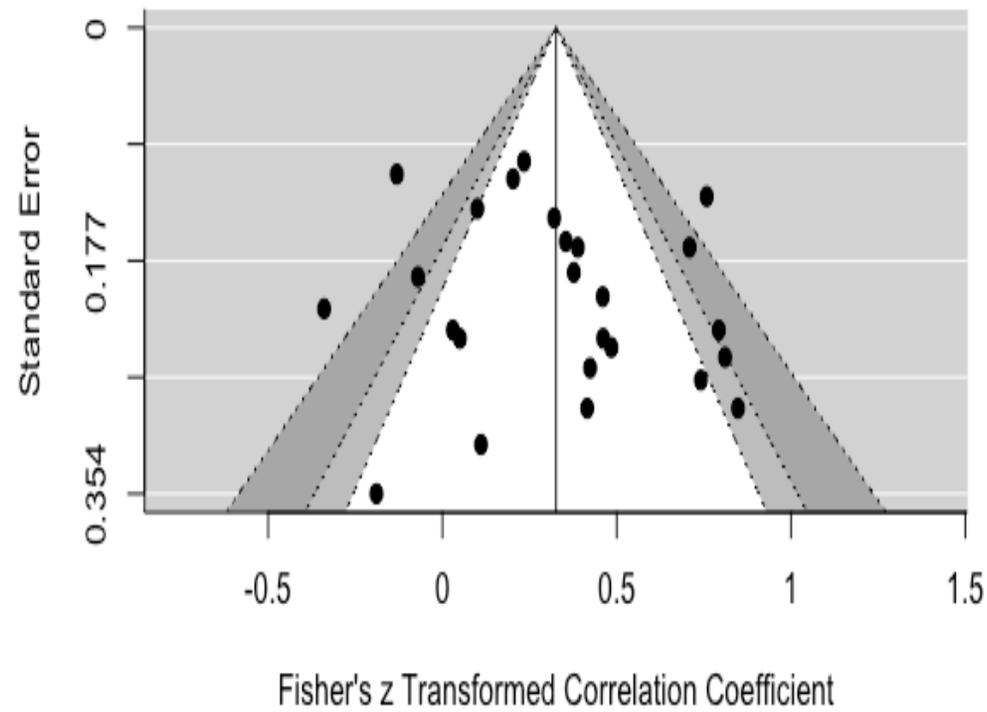


Figure 2. A forest plot containing effect sizes and 95% confidence intervals for the relationship between RRBs and set shifting performance with the impact of RRB scale and task (WCST versus other). Dyad refers to a measure that is used to diagnose autism (e.g. ADOS) whereas a specific measure is a questionnaire that only examines repetitive behaviour (e.g. RBQ).



*Figure 3.* Contour-enhanced funnel plot showing standard error against the effect sizes (Fisher z Transformed Correlation Coefficient) of the association between RRBs and set shifting performance. Contour lines are at 1%, 5%, and 10% levels of statistical significance.

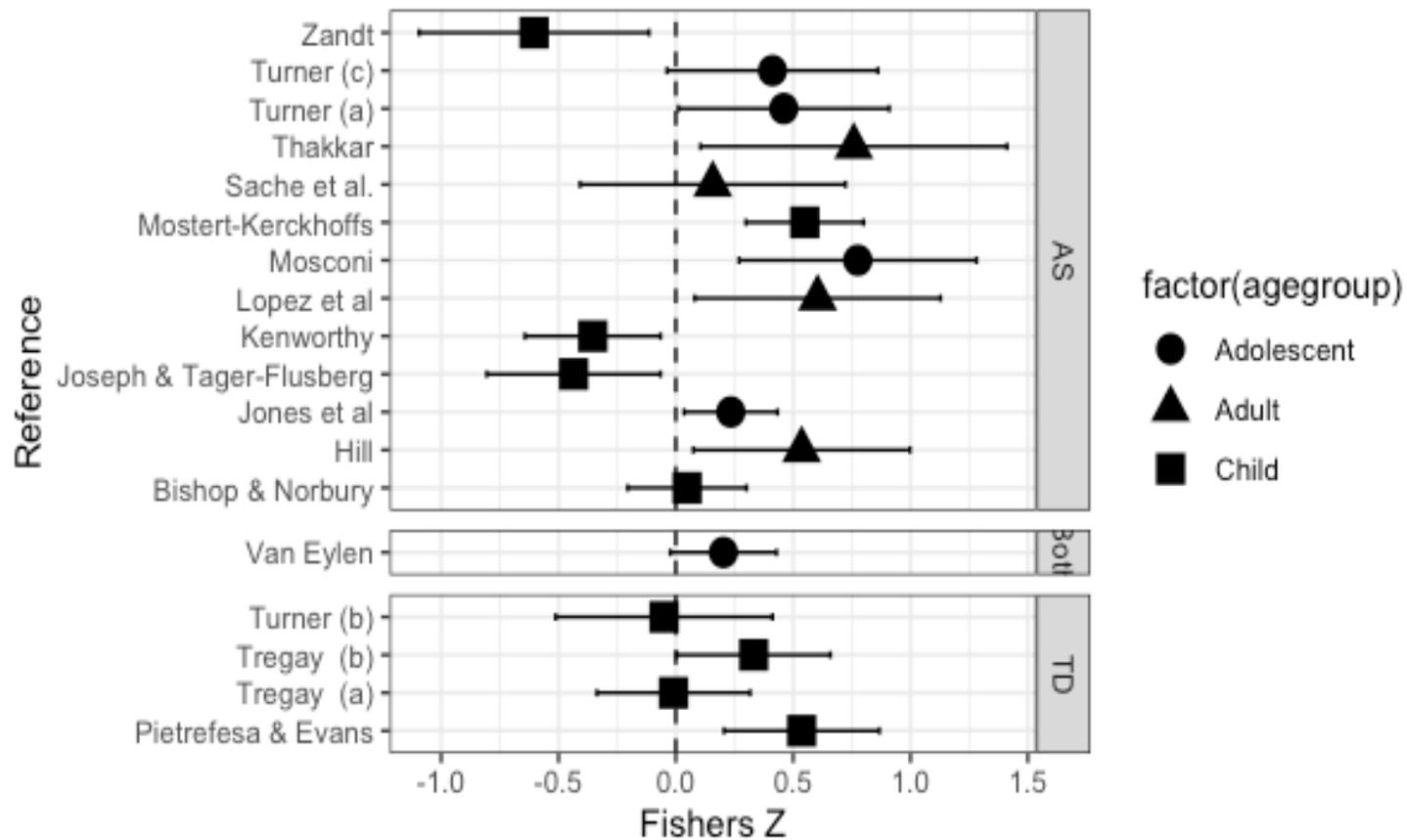
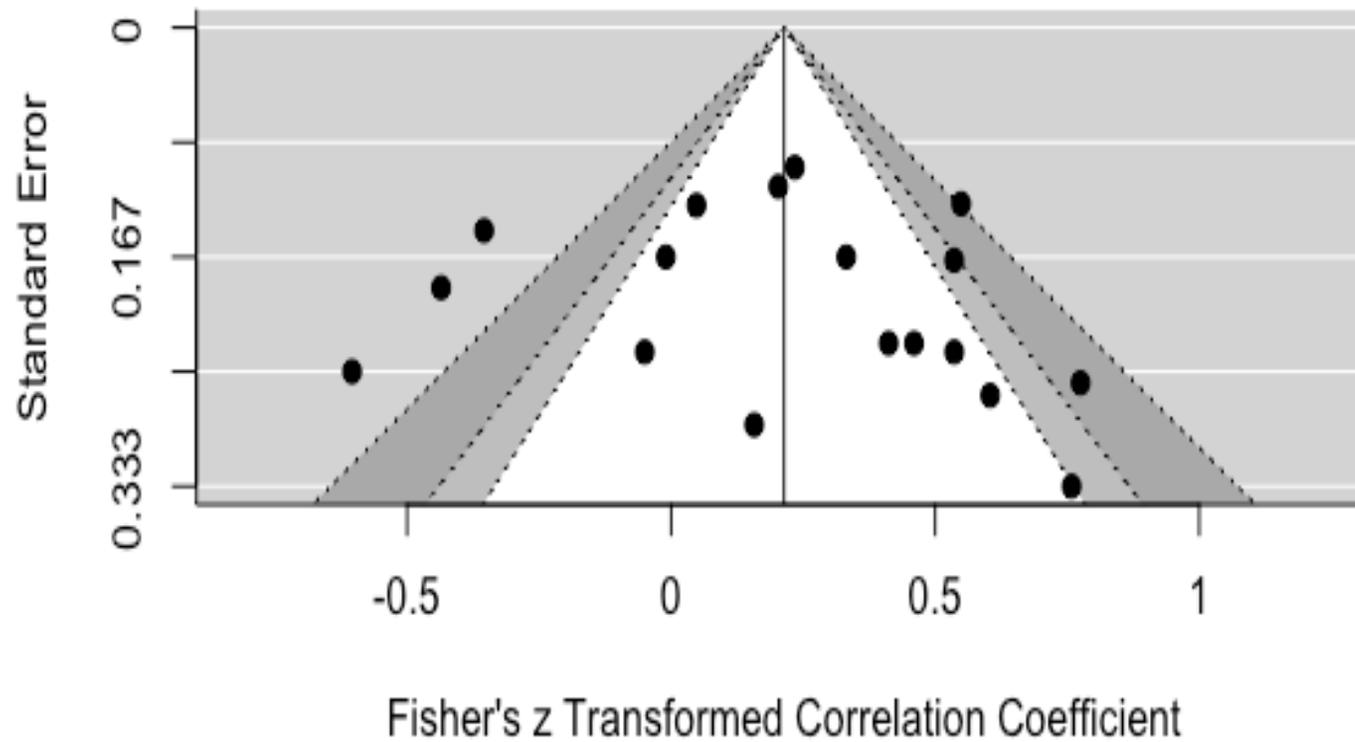


Figure 4. A forest plot containing effect sizes and 95% confidence intervals for the association between inhibitory control tasks and RRB levels and the impact on diagnosis and age groups



*Figure 5.* Funnel plot showing standard error of the effect size for the association between inhibitory control tasks and RRB levels. Contour lines are at 1%, 5%, and 10% levels of statistical significance

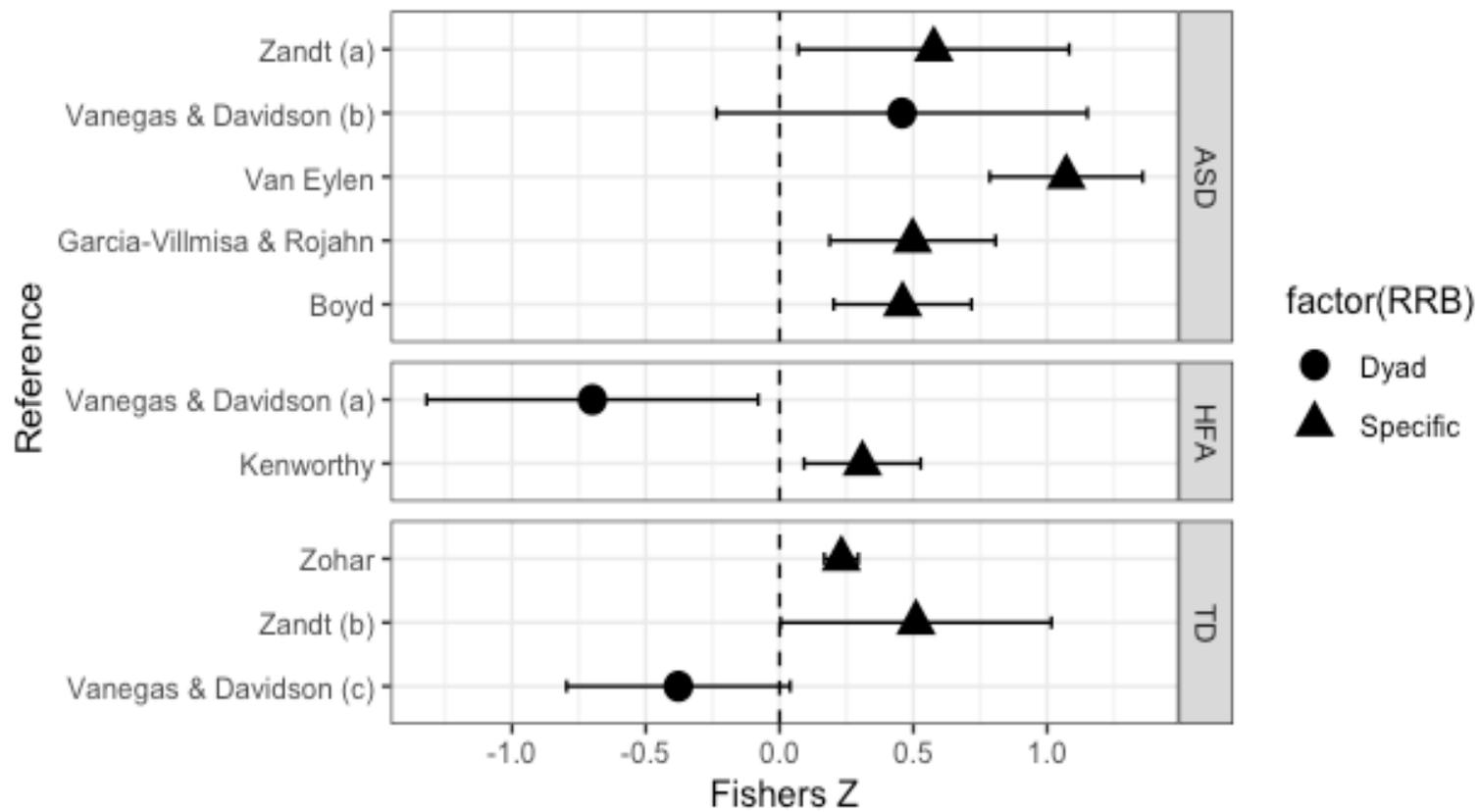
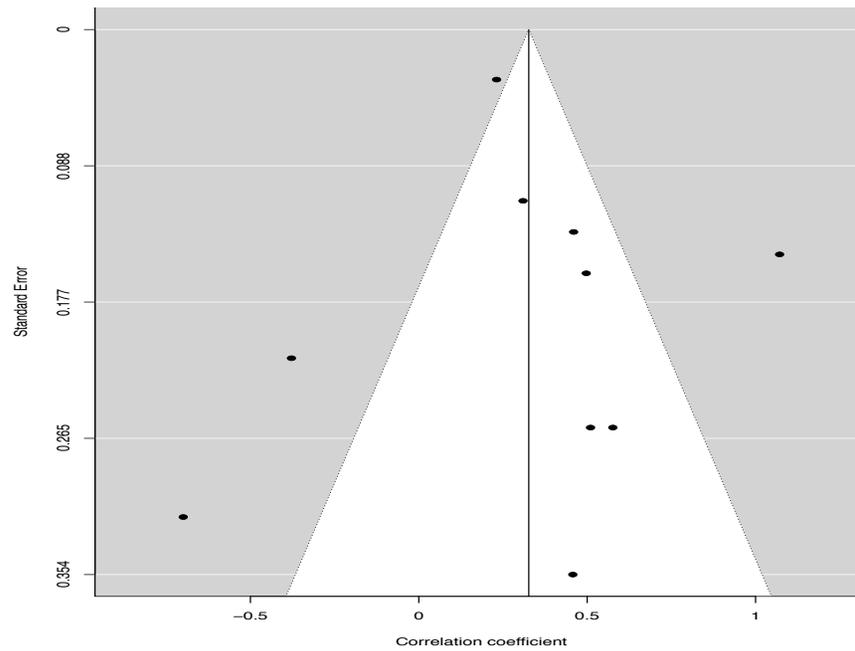


Figure 6. A forest plot containing effect sizes and 95% confidence intervals for the association between parent-rated EF tasks and RRB levels and the impact of diagnosis and diagnostic versus specific RRB measures.



*Figure 7.* Funnel plot showing standard error of the effect size for the association between parent-rated EF task performance and RRBs levels. Contour lines are at 1%, 5%, and 10% levels of statistical significance

