Attentional processes involved in the development of set shifting and restricted and repetitive behaviours in young typically developing children and children with autism.

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List of abbreviations

TD - Typically developing

DD- Developmental delay

ASD - Autism Spectrum Disorder

OCD – Obsessive Compulsive Disorder

ADHD – Attention Deficit Hyperactivity Disorder

EF - Executive function

DCCS- Dimension Change Card Sort

WCST- Wisconsin Card Sort Task

LI- Learned Irrelevance

NP- Negative priming

RRB - Restricted and repetitive behaviour

CCC- Cognitive Complexity and Control theory

CCC-r – Cognitive Complexity and Control-revised theory

Acknowledgements

Although it is my name on the front page of this thesis, in reality a wide variety of people contributed to this work. Firstly, I would like to thank my two supervisors, Charlie Lewis and Calum Hartley for your insightful comments and encouragement over the years. A special thank you goes out to Charlie. Without your guidance, encouragement and feedback this PhD would not have been achievable. The joy and enthusiasm you have for your research has been motivational for me. I could not have asked for a better supervisor.

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Thesis abstract

It is widely known that young typically developing (TD) children and many individuals with autism (ASD) perform poorly on executive function (EF) tasks. In pre-schoolers, these skills develop rapidly between the ages of 3 and 4 and are often measured through the Dimensional Change Card Sort (DCCS) task. This is also around the same time that restricted and repetitive behaviours (RRBs), a diagnostic characteristic for ASD, peak in typical development. These findings have led to an increasing interest in the relationship between EF skills and RRBs, but the studies have produced mixed findings. To our knowledge no meta-analyses have been carried out to examine the relationship between RRB scores and performance on EF measures. Moreover, no studies have yet pinpointed what it is about these skills or behaviours that make them associate so highly. This thesis therefore presents a series of experiments that firstly aim to examine the strength of the relationship between the behaviours and performance on EF tasks. Secondly, examine the relationship between different sub-groups of RRBs and various set shifting processes, such as the ability to shift away from dominant stimuli, and the ability to activate previously ignored stimuli. Finally, examine training implications for the skills by assessing if a short-term training program can improve the scores and possibly have an impact on the behaviours.

In chapter 1, we conduct three meta-analyses to examine the relationship between RRB scores and performance on set shifting and inhibitory control tasks, as well as scores on EF parental report measures. We found significant correlations of medium strength in all three analyses. Moreover, whereas age and the type of RRB scale moderated the inhibitory control and parental report results; diagnosis, testing modality, and type of EF measure did not have an overall impact on the results. These findings suggest that the EF hypothesis may play a crucial role in the development of RRBs, or vice versa. Future research should focus

on disentangling different EF measures to pinpoint what it is about the tasks that make them associate with the behaviours.

In chapter 2, the focus is on set shifting, the individual EF skill that showed the strongest association with RRBs. Our aim in this chapter is to uncover what causes the correlations between the behaviours, and performance on the Wisconsin Card Sort Task, (WCST) but not the much simpler DCCS. We review the main theoretical frameworks that have attempted to explain two types of errors; the ability to shift away from dominant stimuli and the ability to activate previously irrelevant stimuli. Whereas research on the DCCS suggests that children find both errors difficult, research on the WCST suggests that adults find it more difficult to activate previously irrelevant responses. We argue that the different findings are not evidence for different developmental trajectories in children and adults. Instead, the tasks differ crucially in a way that only the design in the adult task isolates the errors properly and is consequently a pure measure of the two shifting processes. Our review concludes that both the ability to shift away from dominant stimuli and activate previously irrelevant stimuli play key roles in set shifting development, yet only the ability to activate previously irrelevant stimuli may be able to explain the high levels of RRBs in young TD children and individuals with ASD.

In chapter 3 we assessed the two predictions in chapter 2 in more depth, through two experiments that compared different variations on the standard DCCS with a new method in which the relevant response is no longer available. We found an age-related shift in which pre-schoolers learned to pass all task versions around the age of four, offering support for the proposition that the ability to attend to previously irrelevant aspects of the environment play a key role in set shifting development. We also found support for the prediction that a child's problems with activating a previously irrelevant cue (rule activation) may reveal biases of attention that explain the persistence of RRBs in typical and atypical development. We explain these through an attentional framework that suggests that the behaviours, and poor task performance is caused by difficulties with overriding automatic avoidance responses. These are responses that have been created over time as a person continuously ignores a response or an activity.

In chapter 4, we evaluated the training literature to address why there are a lack of training studies on the topic. We also made suggestions for future training interventions. More specifically, we stress that EF interventions can be challenging and expensive, as they often require a high level of resources, such as parent training, or supervision of adults or teachers. Moreover, it has been questioned if such interventions can offer long-term training effectiveness, and generalise to situations outside of the lab. Future research should therefore develop a brief and cost-effective EF training program that requires low resources, and can be easily implemented in schools to examine the long-term effectiveness of this type of intervention, as well as if training can have an overall impact on RRB scores.

In chapter 5, we examined the effectiveness of a brief training program to assess if pre-schoolers and children with ASD can be trained on tasks that measure their ability to activate previously irrelevant rules, and if training has the potential to influence the frequency and nature of their reported RRBs. We found highly significant training effects, and no change in set shifting performance in the control condition. We also found a small, yet not significant, decline in the RRB scores for the TD children after training. These findings propose that a brief rule activation training program may aid set shifting development and thereby be useful in a school setting. The RRB findings are less positive however, perhaps suggesting that to see an effect on the RRBs a training program may need to involve more sessions and run over a longer period of time. Overall, the results in this thesis provide evidence for the view that rule activation errors play a key role in the development of set shifting skills in pre-schoolers and individuals with ASD. Moreover, these errors may play a crucial role in the development of RRBs, or vice versa.

Statement of Author Contribution

In the Chapter entitled, "Executive function skills are linked to restricted and repetitive behaviours in Autism Spectrum Disorder: Three correlational meta-analyses", the authors agree to the following contributions:

Rebecca K. Iversen — 75 % (Data collection, Experimental design, analysis and writing)

Signed: Date: 30/09/2019

Professor Charlie Lewis — 25 % (writing, and review)

Signed: __________________________ Date: 30/09/2019

Chapter 1:

Executive function skills are linked to restricted and repetitive behaviours in Autism Spectrum Disorder: Three correlational meta-analyses

Text as it appears in Iversen, R. & Lewis, C. (2019). Executive function skills are linked to restricted and repetitive behaviours in Autism Spectrum Disorder: Three correlational meta-analyses. Manuscript in preparation.

Abstract

Despite the increasing centrality of restricted and repetitive behaviours (RRBs) in the diagnosis of Autism Spectrum Disorder, 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 autism (e.g. South et al., 2007) and are also prevalent in young typically developing children (e.g. Evans et al, 1997; Tregay, 2009). 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 present three random-effects analyses (N= 2895) to examine the strength of the association between RRB levels and performance on set shifting, inhibitory control, and parental-report based EF batteries. The results showed moderate but significant associations between high levels of the behaviours and poor EF skills. Moreover, the associations remained stable across typical development and in children with autism spectrum disorder and across different types of EF measures. In keeping with the recent evidence that we discuss these meta-analyses suggest that cognitive mechanisms may underpin the high RRBs that are seen in individuals with autism, as well as in typical development. We propose that the EF account may be critical for guiding diagnosis and future interventions in autism research.

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

1. Introduction

Many major puzzles in our understanding of Autism Spectrum Disorder 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, i.e. how it is associated with differences in social communication difficulties and underlying cognitive skills (e.g. Van Eylen, Boets, Steyaert, Wagemans & Noens, 2015; Lopez, Lincoln, Ozonoff & Lai, 2007). In the 1990s great expectations were placed on cognitive models, which might explain the joint problems that make up 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 repetitive behaviour (e.g. Frith, 2008) or simply suggest that the explanation of the social/communication problem, like undeveloped 'theory of mind', is 'related' to these repetitive behaviours (Baron-Cohen, 2000, pp. 78-79) without offering an 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, let alone their links with the second core diagnostic feature of ASD, social and communicative difficulties. We re-open the debate on this literature, 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 origins and nature of these behaviours. These play down the 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: Theoretical panacea or methodological quagmire?

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, Glod, Connolly & McConachie, 2012; Lidstone et al., 2014). Since the 1990s (DSM-IV: APA, 1994, ICD-10: WHO, 1992), the behaviours have been divided into four sub-groups: stereotypies, preoccupation with objects, restricted interests and non-functional routines. They are highly frequent and their repetition occurs in an invariant manner. They are manifest in actions that range from rocking and hand flapping to very specific food and routine preferences, such as eating only pizza. It is often reported (e.g., South, Ozonoff & McMahon, 2005) that parents identify these behaviours as the most challenging ASD characteristics to manage and beyond the preschool years they often create barriers to learning opportunities and social interactions (Harrop, McBee & Boyd, 2016). Analysis of the origins and nature of RRBs can not only guide research on the outcomes of the disorder, but also help design interventions that target these behaviours. Nevertheless, the behaviours have long been the neglected characteristic of ASD (Kasaris & Lawton, 2010), and research and diagnostic criteria have only recently suggested that they may have a more central and defining role.

An emerging body of research has highlighted the early appearance and the continuing importance of the RRBs in ASD. Kim and Lord (2010), for example, suggest that they may be the earliest emerging sign of the disorder. Indeed, Lord, Risi, DiLavore, Shulman, Thurm and Pickles (2006) found that repetitive behaviours at the age of two were a better predictor, than the social communication and interaction impairments of an ASD diagnosis at the age of nine. Findings like these have led to two major changes in how the disorder is defined and diagnosed in the fifth edition of The Diagnostic and Statistical Manual of Mental Disorders (DSM-5, APA, 2013). First, the DSM-5 collapsed the diagnostic triad of impairments (social communication, interaction and RRBs) into a dyad (social communication/interaction and RRBs). Secondly, it changed the diagnostic criteria so that two out of four types of RRBs (stereotyped or repetitive speech or motor movements, excessive adherence to routine, highly restricted interests, and hypo- or hyper-reactivity to sensory input or unusual interests in sensory aspects of the environment) have to be met, in contrast to one out of four in the DSM-IV (APA, 2000). The effect of these changes in classification mean that the behaviours may have a more central and defining role in the characterisation of ASD rather than being thought of as coping mechanisms for social interaction impairments (Baron-Cohen, 1989). This change in the DSM criteria emphasizes a need to re-examine the major hypotheses concerning the nature and origins of RRBs, particularly in terms of their variety and prevalence over development. Yet factors, such as the lack of a universal definition and measurement issues, make it hard to evaluate the strengths of the main theoretical contenders for explaining the behaviours.

Definitional issues concerning restrictive and repetitive behaviours

We conducted a search of RRBs in the ISI (Clarivate Analytics) Search Engine. Since the definitional changes in the DSM-5 in 2013, output more than doubled: 718 articles in the 5;7 years since the change between 2014 and 2019 (to July) compared to 317 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 suggest that the RRBs can be divided into dichotomous groups, "low-level" 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). Splitting RRBs into these dichotomous groups may have beneficial theoretical implications, as it has been argued that there are different causes for different behaviours (Constantino, 2011). It has been suggested that sensory and motor behaviours beyond infancy only persist in developmentally younger individuals (Szatmarti et al., 2006; Kim & Lord, 2010), or those who have experienced severe neglect (e.g. Bishop, Richler & Lord, 2006; Rutter et al., 2007). "Higher-level" RRBs, on the other hand, are argued to be more adaptive in the preschool years, because of an increasing need for individuals to regulate their own behaviours (e.g. Evans, Lewis & Iobst, 2004). As a result, they are thought to be more prevalent in more developmentally able individuals (Bishop, Richler & Lord, 2006).

The assumption that the type of an RRB reflects an individual's levels of functioning has, however, been challenged. Some researchers have found that sensory and motor behaviour is not only present in individuals with low IQ, but also higher functioning individuals (e.g. Szatmari et al., 1989; South, Ozonoff, & Mahon, 2005). Although higher functioning individuals with ASD appear to engage in less "low-level" behaviour, it has even been suggested that this may simply be a result of how these individuals have learned to camouflage their difficulties or behaviours, for example during social interaction (e.g. Hull et al., 2017). For higher-level RRBs, on the other hand, it is only children with high intellectual functioning who may be more able to develop more sophisticated routines and interests that fit this classification. Hus, Pickles, Cook, Risi, and Lord, (2007), for example, found relationships between verbal and non-verbal communication and lower-level RRBs, but no such relationships with higher-levels RRBs. Sub-type findings like these are of interest, as they propose that the RRB aetiology may be much more complex than was first suggested.

The idea that different behaviours may have different causes has then consequently resulted in the development of a wide variety of measurement tools that all measure the behaviours differently.

Measurement Issues

There is no 'gold standard' RRB measure. Widely used questionnaires implement a variety of response methods ranging from, for example, calculating frequency or intensity, to identifying whether a behaviour is present or absent (Honey, et al., 2012). Some measures also include several of these metrics, making it difficult to compare the same behaviours, let alone different behaviours, across different measures (South, Ozonoff & McMahon, 2007). In addition to the various metrics, types of assessments also differ. Whereas some of the measures comprise observations and interviews used to diagnose ASD (e.g. the Autism Diagnostic Observational Scale, ADOS: Kim, & Lord, 2010; the Autism Diagnostic Interview-Revised, ADI-R: Rutter, LeCouteur & Lord, 2003), others are 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 rely on 12 items, while others, such as 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 a majority of the measures have not been used frequently enough to test and analyse their concurrent and construct validity (Honey, McConachie, Turner, & Rodgers, 2012). The current meta-analysis explores the diversity between measures in more depth to examine whether such differences should be considered further.

Evidence for the measurement difficulties can be seen in South, Ozonoff and McMahon's (2007) study, which assessed 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 the same associations, using the more specific RBI. These differences could be caused by the different levels of details that each measure involves. More 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 to move forward. Given the complexity of RRBs, it is perhaps naive to assume that we can develop one 'gold standard' measure. Nonetheless, criteria should be developed to help researchers 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.

Over the past two decades there has been a shift that emphasizes the 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 pattern of an increase in both lower and higher order behaviours towards the end of the preschool period and a decline thereafter coinciding with manifestation of EF (executive function) skills were taken to indicate a close correlation if not a causal relationship (Turner, 1997). Over the past decade this view has received much critical scrutiny:

> "There is little evidence for robust associations between repetitive behaviour and specific cognitive, sensory or motor impairments. Thus, abnormalities in these domains identified in individuals with autism would not appear to provide much useful information relevant to the pathophysiology of restricted repetitive behavior" (Lewis & Kim, 2009, p. 117).
>
> "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 two alternatives to the EF account which are hinted at in these quotations – the neurobiological and developmental trajectory accounts. We will argue that neither is incompatible with an EF approach: indeed this review was motivated by the possibility that both alternatives could benefit from a more holistic combination with this area of theorization.

The first area, neurobiological accounts, encompasses a wide variety of possible mechanisms concerning genetics and neurological links. These have each provided valuable, but incomplete, information about ASD. They start with the genetic association between 36% of monozygous compared with 0% of dizygous twins, where one has received a diagnosis of ASD (Folstein & Rutter, 1977) A recent meta-analysis by Tick, Bolton, Happé, Rutter and Rjisdijk's (2016) offers stronger support for a hereditary component. This group of seven studies and sample of 6413 twin pairs found almost perfect correlations for monozygotic twins (MZ) (r= .98), whereas the dizygotic (DZ) correlation was .53. These findings offer support for the view that ASD has a strong genetic aetiology, but the genetic basis of RRBs has been harder to pinpoint. Whitehouse and Lewis (2015) suggest that there is limited evidence for specific genes or loci that may control RRBs. They stress that even in Prader Willi Syndrome, a disorder in which genetic loci are known (e.g. 15q11-13), there has not been clear progress in the attempt to detect alterations in the specific genes that have been associated with RRB levels. There is also not much progress on how this can relate to RRBs in other disorders, such as Obsessive Compulsive Disorder, Tourette Syndrome, ASD and Fragile X Syndrome (e.g. Lewis & Bodfish 1998; Moss et al. 2009). Whitehouse and Lewis conclude that genes may provide a necessary, but not sufficient, cause of RRBs.

A second strand of biological research examines the connections between RRBs in ASD and other disorders in terms of neuropathological changes in the cortical-basal ganglia pathways (Langen, Kas, Staal, van Engeland, & Durston, 2011). This account suggests that lower- and higher-level RRBs may be linked to separate regions in the corticostriatal circuitry, where the main function is to control goal-directed behaviour. A disruption within the basal ganglia or between striatal and forebrain structures is hypothesised to lead to dysfunctional feedback to frontocortical areas, which may then lead to RRBs. More specifically, Langen et al. contend that RRBs occur if one of three corticostriatial circuits (sensorimotor, associative and limbic loop) is damaged, and that the location of the damage determines the type of RRB. The sensorimotor loop (the motor and pre-motor cortex) is thought to be responsible for lower-level RRBs, the associative loop for rigidity or the inappropriate repetition of a goal (the dorsolateral prefrontal cortex), and the limbic loop (the lateral orbitofrontal and anterior cingulate cortex) is thought to mediate some higher-level RRBs, such as obsessions.

This account might explain the connections between RRBs and various disorders. First, a review by Morand-Beaulieu et al (2017) concludes that the thinning of sensorimotor loop impairments in Tourette Syndrome are thought to be involved in the development of tics (Sowell et al., 2008). Secondly, impairments in the limbic loop have been associated with obsessions and compulsions (Menzies, Chamberlain, Laird, Thelen, Sahakian & Bullmore, 2008). Thirdly, the same framework can also account for overall RRB levels in Obsessive Compulsive Disorder, as positron-emission tomography studies have shown that the limbic circuit, or more specifically an overactivity of the striatal-orbitofrontal circuitry, is involved in the development of the compulsions and obsessions (e.g. Remijnse et al., 2006). Finally, impairments in the limbic loop, or more specifically the anterior cingulate cortex, have been found to be associated with overall RRB levels in ASD (Zhou, Shi, Cui, Wang & Luo, 2016; Shafritz, Dichter, Baranek & Belger, 2008; Thakkar, Polli, Manoach, Joseph, Tuch, Hadjikhani, & Barton 2008).

Despite these interesting patterns, links between cortical structures and the functions of RRBs are hard to draw. For a start, post mortem research show differences in neural development (Avino et al., 2018; Zikopoulos et al., 2018) but do not identify links with RRBs. A review by Amaral, Schumann and Nordahl (2008), for example, concluded that the few studies that have examined post-mortem tissue findings in ASD have been inconsistent, perhaps because some have included individuals with seizures in their clinical histories (e.g. Kemper & Bauman, 1993; Bailey et al., 1999). Secondly, the corticostriatal circuit account cannot explain which neurobiological mechanisms mediate the reduction in RRBs that we commonly see in typically developing children and their persistence in developmental disorders.

Lewis and Kim (2009) acknowledge these limitations and suggest that in order to explain the wide variety of RRBs it is not enough to consider genetic factors and neuroadaptations in cortical-basal ganglia pathways. They suggest that interactions between these and early experience-dependent factors (e.g. restricted environments) must also be considered and, more specifically, that RRBs may be mediated by a circuitry that involves a large number of genes, given the complexity and heterogeneity of the behaviours. RRBs may then arise if one or a few of these genes mutate and interact with experiential factors, as it will cause disruption to the circuitry.

The environmental restriction account that Lewis and Kim (2009) highlight is based on findings in humans and animal models, and suggests that restricted environments can lead to higher RRB levels. For example, a longitudinal study of Romanian adoptees showed that environmental restriction induced RRBs in children (Rutter et al., 1999). Moreover, stereotypies in rats decrease after they are introduced to an enriched environment (Hornig, Weissenbock, Horscroft & Lipkin, 1999). Although animal models may not appear relevant to ASD at first glance, Lewis and Kim argue that deficits in early social and communicative behaviour are likely to impair experience-dependent brain development, which may then exacerbate RRBs. Two issues arise from these environmental enrichment studies. First, they have only focussed on stereotypies and thereby only provide information about specific RRBs. Secondly, they do not account for how genes and environment interact. As a result, they do not provide us with a psychological model of how they operate.

An account that considers the same factors as Lewis and Kim's review, but also focuses on the RRB trajectory itself, is Leekam, et al.'s (2011) developmental account. This suggests that RRBs are immature responses that are maintained more strongly within the behavioural repertoire of individuals with ASD. In order to explain this process, it is suggested that neurobiological changes must be traced alongside behavioural ones. They also highlight the importance of the development of the corticostriatal circuits in early childhood. The developmental account is largely based upon Thelen's (1981) view that stereotypies play a role in the development of skilled motor action. More specifically, the 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, suggesting that RRBs are more likely to be triggered by specific events, since more extreme arousal states (high or low) are needed to release the behaviours. 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 that we can see 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. Just as with the neurobiological account, it appears that 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) proposition that stereotypies reduce over time in typical development is widely supported in the literature (e.g., Mirkovic et al., 2017; Cevikaslan, Evans, Dedeoglu, Kalaca & Yazgan, 2014). It is plausible that lower level behaviours may reduce as infants develop goal-directed actions. It is more tricky to use this theory to account for the higher-level RRBs, however, as they have been found to follow a different trajectory, in which they first increase, then decline around the age of 5-6 (e.g. Evans et al, 1997; Mirkovic et al., 2017; Cenikaslan et al., 2014). Without an additional dimension, this account would struggle to explain what purpose the higher-level RRBs behaviours have, and what it is that drives their trajectory.

In addition to focusing on their developmental trajectory, Leekam et al. suggest that RRBs become more likely to be triggered by specific events, since extreme arousal states (high or low) are needed to release these behaviours. This echoes an early RRB account that the behaviours are caused by hyper- or hypo-arousal (Hutt & Hutt, 1965). The hyper-arousal prediction suggests that the behaviours are coping mechanisms that develop to reduce high-arousal or anxiety. A later account by Goodall and Corbett (1982) expanded this theory by proposing that RRBs may develop to regulate under-arousal that occurs due to a lack of stimulation from the environment. The suggestion that anxiety plays a central role in RRBs, is perhaps not surprising, as anxiety was highlighted in Kanner's (1943) original description of the behaviours. A recent meta-analysis by Steensel and Bogels (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. Recent sub-group analyses have expanded on previous research and found that higher-level RRBs only associate with anxiety levels in samples of typically developing children (Evans, Gray, & Leckman, 1999; Laing, Fernyhough, Turner & Freeston, 2009; Zohar & Felz, 2011) and children with ASD (Rodgers, et al., 2012; Uljarevic & Evans, 2016). Findings like these have suggested that higher-level RRBs serve the purpose of controlling the environment and thus reduce anxiety. Despite their 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 associated because poor cognitive control may lead to hyper-attentiveness to negative information that creates anxiety which then leads to RRBs (e.g., Spiker et al., 2012).

The emphasis on goal directed actions in the developmental account leads easily into the proposal that the different RRB trajectories are driven by an individual's executive function (EF) skills.

What are 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. Given the broad nature of the EF concept, it has been widely researched in the hope of understanding the neurodevelopmental progression of EF (e.g. Bardikoff & Sabbagh, 2017). Miyake, Friedman, Witzki, Howerter and Wager's (2000) seminal paper, assessed if the EF concept could be defined as a single self-regulatory ability (unity), or as a range of skills that are all essential to control thoughts and behaviours (diversity). To do this, they examined the relationship between three commonly studied EF skills: inhibition of prepotent responses, shifting of mental sets, and information updating and monitoring (also referred to as working memory). Inhibition of prepotent responses is defined as the ability to suppress a dominant or automatic response (Friedman, 2016), shifting, as the capacity to switch between mental sets or tasks (Monsell, 2003), and updating and monitoring, as the ability to pick out relevant information from the environment, and replace it with irrelevant information (Friedman, 2016). They concluded that the three skills share some common variance, but they can be divided into three separate factors. They demonstrated this statistically using structural equation modelling within what they termed a 'unity with diversity model' which shows links between the three components but clear differences between the latent variables identifying each construct (Miyake et al., 2000)

Recent analyses on preschoolers and school-aged children have questioned these results. Some, for example, find that whereas a unitary factor structure is the best fit for preschoolers (e.g. Willoughby, Wirth & Blair, 2012), a more diverse factor structure is the best fit for school-aged children (e.g. Rose, Feldman & Jankowski, 2011). The two-factor structures either support a working memory and shifting model (e.g. van der Sluis, de jong & Van der Leij, 2007), or a model consisting of working memory and a combined inhibition and shifting factor (e.g Lee, Bull & Ho, 2013; Brydges, Fox, Reid & Anderson, 2014). To try to get to the bottom of the inconsistent findings that confirmatory factor analyses (CFA) has produced across the life span, Karr, Areshenkoff, Rast, Hifer, Iverson and Garcia-Barrera's (2018) re-analysed 46 CFAs (N= 9756). They found support for a greater unity of the EF skills in children (one to two-model factor), and a more diverse framework in adolescents and adults (two to three-model factor). These findings highlight the need to measure EF skills separately and consider age when examining their development and their relationship with RRBs over time.

Although both Leekam et al.'s, and Lewis and Kim's reviews suggest that EF deficits are not vital for the development of RRBs, they both stress the importance of corticostriatal circuits. Moreover, Langen et al. (2011, p2) state that "cognitive models have provided valuable hypotheses for how neurobiological circuitry might be disturbed in repetitive behaviour". Considering the fact that the main function of the corticostriatal circuit is to control goal-directed behaviour, this statement points to EF processes. These skills may then play a crucial role in the relationship between RRB levels and impairments in the corticostriatal circuit. 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). Moreover, individuals with ASD and OCD are thought to engage in significantly more RRBs than typically developing children. Similar variability has been found in the EF literature as the same study by Zandt, Prior and Kyrios (2007) found that individuals with ASD and OCD performed worse on inhibitory control tasks than typically developing individuals. The EF hypothesis could then possibly account for the frequency of the behaviours in individuals with ASD and

OCD. At the same time, it can help explain the wide range of RRBs, as different skills may be responsible for different behaviours. Moreover, it can account for the heterogeneity within disorders, as well as the change from RRBs in typically developing children to those in developmental disorders.

Brief section on how EF skills develop

A wide range of theories have been developed in an attempt to explain the development of EF skills. Three influential EF theories will now be briefly presented to demonstrate that there are gaps in our EF knowledge that needs to be addressed before a comprehensive EF theory can be developed. The first of the influential theories is the neural network model, developed by Morton and Munakata (2002). This theory suggests that working memory improvements drive EF development. According to this theory, the neural model consists of an input layer, an output layer and a layer of hidden units that interact with each other. The interactions between these layers strengthen active connections over time and consequently help override latent responses in EF tasks. The connections also get strengthened over time as individuals are more capable of sustaining the active representation of the task instructions with age. Although this theory has many strengths, it is unclear how the concept of different dimensions has come about as a developmental process. According to the theory, the neural network codes for common features (e.g it only learns that something is blue because something else is blue), highlighting the need to explain the development of abstract representations. In addition to this, the framework also struggles to explain poor performance on the Dimension Change Card Sort (DCCS, Zelazo et al., 2003) task, as young children are often unable to switch from, for example, colour to shape on this task despite being provided with instructions prior to each trial.

The next two influential EF theories that will be presented combine a structural-hierarchical and functional approach. The first is Fuster's theory of PFC functions, and the second is Zelazo's cognitive control and complexity theory (CCC) theory. According to Fuster's (2008) theory, the main role of the pre-frontal cortex is to produce and integrate novel and complex behavioural structures. More specifically, goal-directed sequential actions have a particular temporal gestalt that is defined by the goal of an action and the interactions among its components. Consequently, goal-directed actions arise due to an interplay between the environment and the organism. This interplay is driven by the PFC as it selects and orders individual actions towards a goal and adjusts them if necessary. Despite having interesting implications for conceptualizing EF, this theory does not explain how the hierarchies develop. In other words, this is a factor that needs to be tackled in order to provide a comprehensive theory of the development of EF skills.

Finally, the CCC account by Zelazo and colleagues (2003) suggests that children formulate plans in terms of rules, and that the complexity of the rule systems change with age. As with the other two accounts, some issues arise with this theory. Several studies for example find that 3-year-olds can, under certain circumstances, shift between different sorting dimensions in the DCCS (e.g., Fisher, 2011; Jordan & Morton, 2008). Findings like these cannot be explained through the CCC framework, and the theory is thereby unable to account for the full development of EF skills. In conclusion, this brief presentation of three influential EF theories suggests that they have conceptual problems, and that there are currently no developmental theories of EF that are comprehensive enough to explain the full development of EF skills. It is therefore currently unclear whether EF undergoes quantitative change (e.g., increase in inhibitory control or stronger working memory activation), or if it is defined by qualitative changes and develops in terms of a sequence of hierarchical levels. In addition to the issues identified above, none of the theories also consider the relationship between the development of EF skills and RRBs. Future research should therefore systematically address the limitations raised by the EF account by teasing apart different components in EF tasks and tracing their development over time. Research should also examine their developmental trajectory alongside the development of RRB scores to pinpoint what it is that is contributing to the relationship between EF skills and the behaviours.

Do individuals with ASD show EF impairments?

One reason why the link between EF and RRBs has been played down is that extensive research, numerous reviews, and meta-analyses on the definition and EF impairments in ASD conducted up to a decade ago, suggested that the role that these skills play in the etiology of the disorder remains unclear. Geurts, Corbett and Solomon (2009), for example, evaluated 29 studies and concluded that there is no firm evidence for a cognitive flexibility deficit in adults with ASD. The authors 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, Science Direct= 172 and PubMed= 59) 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 (n= 41, g= .55 and .31, respectively) (Geurts, van den Bergh & Ruzzano, 2014), as well as planning difficulties (n=50, g= 0.52) (Dubbelink & Geurts, 2017) in individuals with ASD. The positive links with inhibition and planning cast doubt on the suggestion that many individuals with ASD do not find the tasks difficult. More evidence for this can be seen in a meta-analysis by Landry (2015) that combined 31 studies and showed impaired WCST performance in individuals with ASD. Moreover, recent meta-analyses by Lai et al. (2017) as well as Demetriou et al. (2018) find even stronger evidence for the view that overall EF performance, as well as performance on separate EF skills play a role in controlling 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 the overall EF relationship, implying that individuals with ASD performed worse on EF tasks than the 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, was smaller as it included 98 studies (n=5991, ASD= 2985, Control= 3005), concentrating on younger samples of children and adolescents, in contrast to Demetriou's analysis that included a wide age range. Another difference between the studies 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 that EF impairments are likely to play a crucial role in ASD, as well as supporting the idea that separate skills are important, despite the controversy of what overarching EF is. Recent evidence therefore emphasises that it is now more relevant than before to examine the clinical implications for 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 skills.

Are executive function skills related to the high levels of RRB in ASD?

A spurt of new research offers renewed support linking elevated RRB levels to EF difficulties such as set shifting (e.g. Miller, Ragozzino, Cook, Sweeney & Mosconi, 2015; Jones et al, 2017), inhibitory control (e.g. Thakkar et al., 2008; Mosconi et al., 2010; Jones et al, 2017) and planning (e.g. Van Eylen, Boets, Steyart, Wagemans & Noens, 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. Moreover, Miller et al. (2015) found that in a sample of 60 individuals with ASD the overall set shifting errors predicted RRB levels. Studies like these have led to the suggestion that there is a need for immediate 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. Despite the positive results, some of these studies suggest that we need to consider EF skills in combination with genetic components. More specifically, the need for gene-brain-behaviour models of ASD has been highlighted, either using set shifting as a link between the components (Yerys et al., 2009) or inhibitory control (Thakkar et al., 2008). Thakkar et al., for example, found that elevated RRB levels in their ASD sample related to hyperactive response monitoring in the rostral anterior cinglulate cortex (rACC) during an antisaccade task. These findings complement Lewis and Kim's genetic account 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, Yerys, Anthony & Wallace, 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, as they suggest 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 has been argued to tap into cognitive flexibility

(Everett, Lavoie, Gagnon & Gosselin, 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 other two skills. 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, Isquith, Guy, & Kenworthy, 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 consists of two smaller scales, 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 of 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; correlate with biological markers, and even show predictive relationships with academic skills (Isquith, Roth, & Gioia, 2013). This leads nicely into a second possible reason for the inconsistent findings as EF rating measures may have moderated the results. More specifically, it has been suggested that rating scales have a higher ecological validity, and consequently may be the only measures that can reliably predict EF impairments. 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) did not only find low reliability between scales and psychometric measures ($r = .19$), they also found that they 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 that is reported below to examine if measuring EF skills by behaviour vs. parental report makes a difference in regards to their relationship with the behaviours.

Predictions

Numerous explanations for RRBs have been proposed, but the cause of the RRBs is unknown, since no hypothesis has yet stood up to rigorous evaluation. The nature of the debate has shifted slightly since Lewis and Kim's (2009) and Leekam et al's (2011) reviews, 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 propose that another framework is capable of explaining the full development of these behaviours. Nonetheless, there is still ample evidence to perhaps 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 approach is useful as it assesses the overall strength of the relationships by combining data from all of 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 statistical methods (Rosenthal & DiMatteo, 2001). Other authors however, argue that a certain degree of dissimilarity needs to be accepted in order to allow for generalisations (Smith et al., 1980). It has been further suggested that, while a correlational meta-analysis can give us an indication of the strength of the relationship, it cannot help us to get a clearer understanding of its nature, particularly any directions of causality. Nonetheless, correlational relationships offer valuable clues that help establish a need for more focused research and identify children who may benefit from specific interventions.

The inconsistent literature on the topic makes it difficult to make strong predictions. Previous EF meta-analyses indicate strong general EF impairments, 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 with RRBs, as not only are we looking at The Behavioral Regulation Index (BRI; Gioia, Isquith, Guy, & Kenworthy, 2000) scale in which shifting and inhibition is combined, parental report measures have been argued to be more ecologically valid, than psychometric measures (Kenworthy, Yerys, Anothony & Wallace, 2008). If we take the inconsistent evidence into account, there is also the possibility that we will not find any significant relationships between any of the EF skills and repetitive behaviours. This will call the EF hypothesis into question. If we do find an overall relationship we need to highlight moderators that should to be explored further'

2. Methodology

2.1 Systematic literature Search and inclusion criteria:

This systematic review and meta-analysis was performed in accordance with the PRISMA guidelines (Moher, Liberati, Tetzlaff & Altman, 2009) and those specifically for correlational meta-analysis (Quintana, 2015). To collect the relevant data that had examined the relationship between EF abilities and levels of RRBs, we searched Scopus and the ISI Search Engines [10.10.2017]. The following combinations of keywords were used: restricted, repetitive behaviours OR stereotypies OR insistence on sameness OR circumscribed interests AND executive function OR set shifting OR planning OR working memory OR inhibition OR inhibitory control OR BRIEF). Scopus produced 177 results and the ISI Search Engine produced 138 results. We also 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 for the set-shifting analysis. 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.

Statistical dependence of the samples

If a paper reported multiple effect sizes, they 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, Hedges, Higgings & Rothstein, 2009).
2. Individual differences were examined within a specific participant group (e.g. if individuals with ASD were 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 used outcome 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. Moreover, if a study reported frequency and duration (e.g. LeMonda et. al, 2012), we included the effect size that included frequency. Finally, if a study 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 this is 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, Kester, Bodner & Miles, 2011). Moreover, 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 decided to report the correlation for the visual task, since other widely used shifting tasks (e.g. the WCST) rely heavily on visual skills, making this task a better comparison. Finally, in Joseph and Tager-Flugberg'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 again included the most widely used measure. 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 decided to report the observational ADOS, as it includes a wide range of behaviours and is based on observation (following Turner, 1999).

Statistical analyses

We ran random-effects models to estimate the overall means and 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 not 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^2). 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 sought to assess 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 assessed publication bias through funnel plots, as studies with stronger effects may be more likely to get published and thereby also 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 therefore employed to offer an objective view. This test 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 specific) and testing modality (experimenter-administered versus computer-administered). Age and diagnosis were examined further in our analyses 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. This type of analysis was of particular interest as whereas the meta-analysis by Landry (2015) found that individuals with ASD performed significantly worse than the control group on the WCST, age did not predict their perseverative errors. We explored the moderating effect of testing modality (computerised versus experimenter administered) further, since it has been suggested that individuals with ASD only find experiment-administered EF tasks difficult due to the social nature of this task (e.g., Perner & Lang, 2002). For the RRB scale moderator analysis, we decided to divide the scales into two types of assessment: diagnostic and specific. The diagnostic measures comprised of 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 the diagnostic and RRB specific measures have a similar structure, big 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 type 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.

3. Results

The first analysis examined whether there is reliable evidence for 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 performance-based 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.

3.1 Meta analysis 1: The association between RRB levels and performance on 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 = 0.29; 95% CI (0.16, 0.40), $p < 0.0001$]. 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= .87). 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 = 65.64% (95% CI; 42.9, 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 65.64% 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 and Table 4 includes the details of the studies that were involved in the analysis.

3.2 Meta analysis 2: The association between inhibitory control scores and RRB levels

A significant, weak to modest, relationship was found between the inhibitory control measures and repetitive behaviour levels [$r = 0.20$; 95% CI (0.03, 0.37), $p = .01$]. See Figure 4 for forest plot. Egger's regression found no evidence for study bias ($p = 0.27$). 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 75.69% (95% CI; 55.25, 90.92), so moderator analyses were performed to identify sources of heterogeneity.

Moderator analyses: These revealed that part of the heterogeneity on the model between inhibitory control performance and RRB levels was caused by an age effect [Q(1)= 4.53, p= 0.03]. We examined this effect further and found a positive relationship between inhibitory control and RRBs in adolescents (r=0.29, p < .001, CI (0.14-0.45), k= 5) and adults (r=0.52, p < .001, CI (0.25-0.79), k= 4), but not in children (r= 0.00, p= 0.95, CI (-0.22-0.23), k=9). The strength of the relationship between RRB levels and inhibitory control seems to get stronger with age. We found no effects for diagnosis, testing mode or type of RRB scale. See Table 2 for a summary of the effects of each moderator and Table 5 for the details of the studies that were involved in the analysis.

3.3 Meta analysis 3: The association between the 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 = 0.32; 95% CI (0.07, 0.53), $p < 0.001$]. See Figure 6 for forest plot. Egger's regression test showed no evidence for small study bias (p = 0.43). 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 90.19% (95% CI; 77.3, 97.4). 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.

<u>Moderator analyses:</u> These revealed that part of the heterogeneity on the model between parent-rated EF measures and RRB levels was caused by the type of RRB measure used, [Q(1)= 8.83, p= 0.003. 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=0.47, CI(0.27-0.67), $p < .001$, k= 3), but not when conducted using a diagnostic measure (r= -0.23, CI(-0.85-0.38), p= 0.46, k= 3). This moderator analysis thereby suggests that the relationship between parent-rated measures 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 and Table 6 shows the details of the studies that were involved in the analysis.

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. The analyses revealed moderate but significant associations between high levels of RRBs and errors in two EF skills, set shifting and inhibitory control, as well as EF parental-report measures. Whereas age and the type of RRB scale moderated the inhibitory control and parental report results respectively; 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 turn and in relation to each other. First, the significant relationships in each meta-analysis suggests 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. 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 the different EF measures 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 EF impairments may be more central in the development of the behaviours or vice versa than what has been suggested in key analyses of the origins of repetitive behaviour (Leekam, Prior & Uljarevic, 2011; Lewis & Kim, 2009). Of particular interest are set shifting effects as these were stronger than those of inhibitory control. For ASD individuals, this finding is perhaps not surprising, considering that overall EF impairments have been identified for all EF skills, and that the role of inhibition has been less consistent (Lai et al., 2017). That the strongest effects were uncovered in the parental report measures needs to be considered further. This may offer support for the view that these measures are more ecologically valid than psychometric measures (Toplak, West & Stanovich, 2013). We return to these two effects in more detail. Our findings nevertheless offer support to recent meta-analyses that found strong evidence for the view that many individuals with ASD find tasks that target EF skills difficult, and that these skills may be essential in their control of thoughts and behaviours (Demetriou et al., 2018; Lai et al., 2017). They also offer support to a spurt of recent research that has linked elevated RRB levels to set shifting (e.g. Miller, et al., 2015) and inhibitory control impairments. (e.g. Thakkar et al., 2008; Mosconi et al., 2010).

Despite uncovering significant relationships between the skills and the behaviours, it is unlikely that the EF account explains the full range and intensity of behaviours which are so prevalent both in typical preschoolers and which 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 (see Sandin, et al., 2017). The associations identified in the current paper suggest that self-control difficulties may be involved in the manifestation of repetitive behaviours. The associations identify the need to re-open the EF account, but also to explore the relationship in terms of 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 previous research that has found strong evidence for a hereditary component in ASD, previous research on the topic (e.g. Van Eylen et al, 2015) and indeed our analyses, have also suggested that sample characteristics may moderate the relationships between EF skills and RRBs. We found that 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 may suggest that inhibitory control skills do not play a role in the initial development of RRBs. Indeed, it is possible that the inhibitory control skills only play a role in the development of higher-level RRBs which may develop later, following research by Mosconi et al (2009) that only found relationships between the skills and higher-level RRBs in adolescents, and not children.

Alternatively, it is possible that such age-related findings are caused by measurement issues. The nature of the inhibitory control tasks for adolescents and adults that were used in the current meta-analysis seem to include a wider range of skills than those for children. In the widely used Stroop task, participants are exposed to colour names that are printed in ink of different colours, which then interferes with naming the colour of the ink. This involves inhibition of an overlearned response, but it also requires set shifting skills as adults must successfully switch between a wide variety of stimuli. This differs in complexity from the child-friendly "knock-don't-knock" task, where children 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, it does not involve high levels of set shifting skills. The same measurement issues are not present in the studies included in the set shifting analysis, as two of the widely used set shifting tasks in this meta-analysis were the ID/ED and the WCST tasks. 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 measures, not clinical measures. This association can perhaps be explained through parental questionnaires being 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 measure a wider variety of them, 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 the role of self-control in the development of 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 executive functions should be explored in combination with other models. For example, it is not incompatible with Lewis and Kim's claims that genetic factors and neuroadaptations in cortical-basal ganglia pathways play important roles in the development of RRBs. Moreover, a cognitive framework would also offer support for Leekam et al.'s developmental account to explain why stereotypies reduce over time in typical development. More specifically, we suggest that these so-called "immature responses" must be driven by a cognitive model that means that the behaviours reduce as infants develop goal-directed actions. This would be able to account for why these behaviours change with age in typical development, and why the behaviours tend to persist in individuals with ASD.

Despite the great benefits a correlational meta-analysis has as it combines big chunks of data on a specific topic and examines the overall effect, it is unable to determine cause or effect. This makes it impossible to draw conclusions as to whether the behaviours cause the EF difficulties, or vice versa. In addition to these difficulties, the current state of the literature has also led to some limitations, such as the difficulty with determining if cognitive abilities confound the picture. Although some of the 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 had not. Due to the limited amount of studies 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.

The task impurity issues that were highlighted in this analysis highlights that future research should assess if similar associations can be identified between RRB levels and other EF skills, such as planning and working memory skills. Future research should also focus on disentangling different EF measures to pinpoint what it is about the tasks that make them associate with the behaviours. We suggest that set shifting measures are of particular interest, as not only did they produce stronger associations than the inhibitory control measures, they were also the only skills that predicted the behaviours in children. This is in line with a previous review by Geurts, Corbett and Solomon (2009) that concluded that isolating crucial cognitive processes will aid in ultimately resolving the gap between inflexibility in daily life, and that 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, the RRB literature is increasingly finding evidence to suggest that the behaviours should be researched in sub-groups of lower-level and higher-level behaviours. This is of interest as the behaviours 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 meaningful relationship between specific set shifting errors and RRBs, which can have clinical implications as interventions that have the potential to help manage difficult RRBs can be developed.

5.References

References marked with an asterisk indicate studies included in the meta-analysis.

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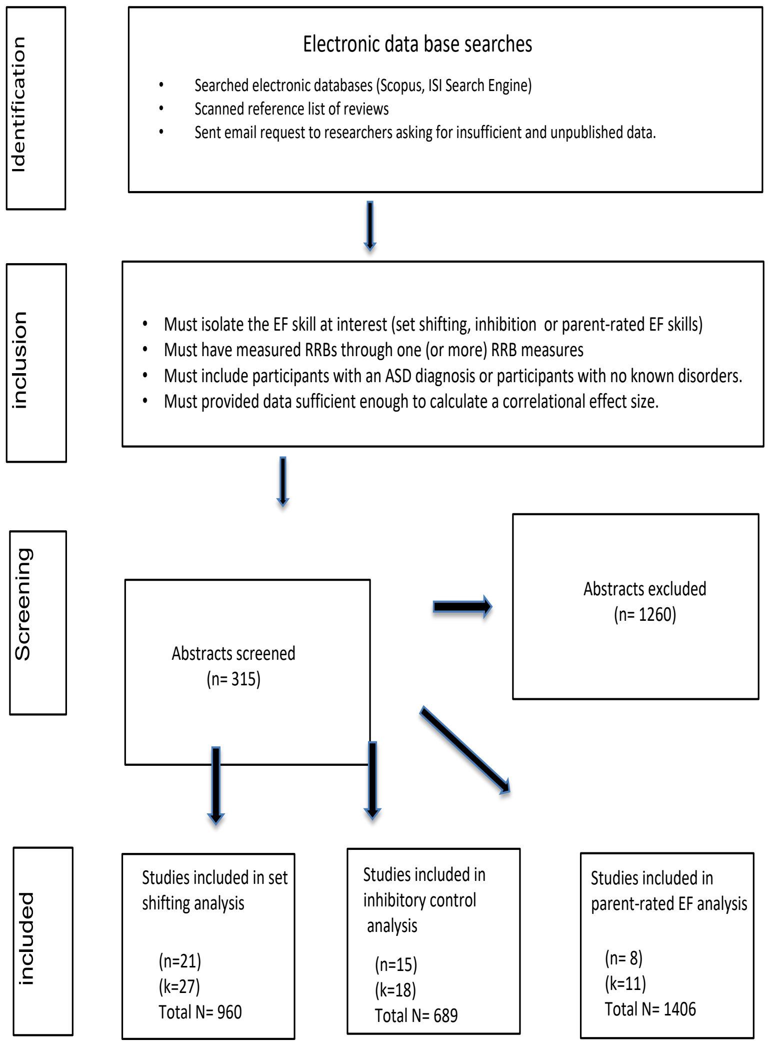

Figure 1: PRISMA flow diagram of the studies that were included in the meta-analysis

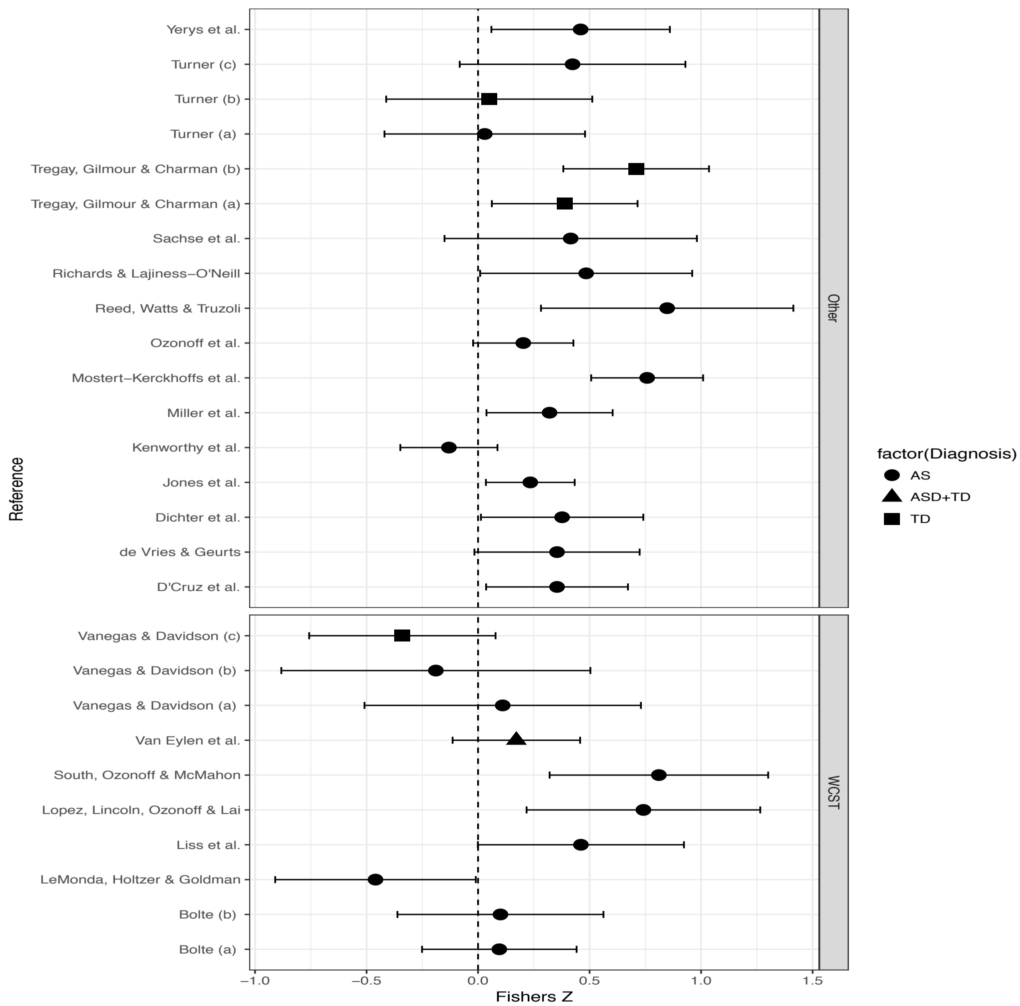

Figure 2. A forest plot containing effect sizes and 95% confidence intervals for the relationship between RRBs and set shifting performance with the impact of diagnosis and task (WCST versus other).

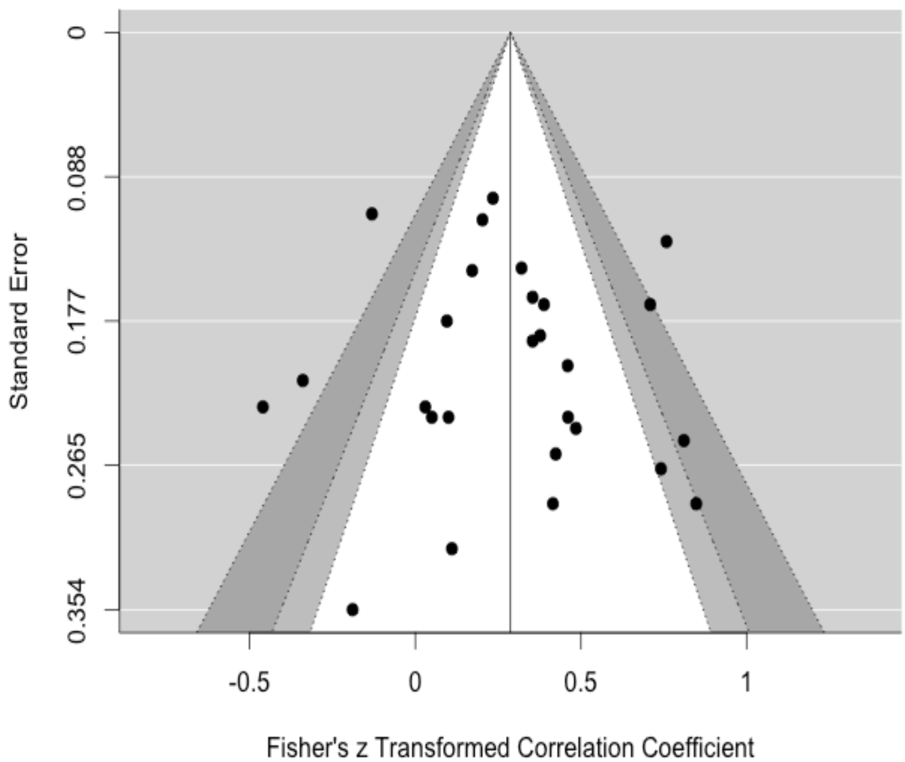

Figure 3. Contour-enhanced funnel plot showing standard error against the effect sizes (Fisher z Transformed Correlation Coefficient) of the association between RRBs and set shifting performance. Contour lines are at 1%, 5%, and 10% levels of statistical significance.

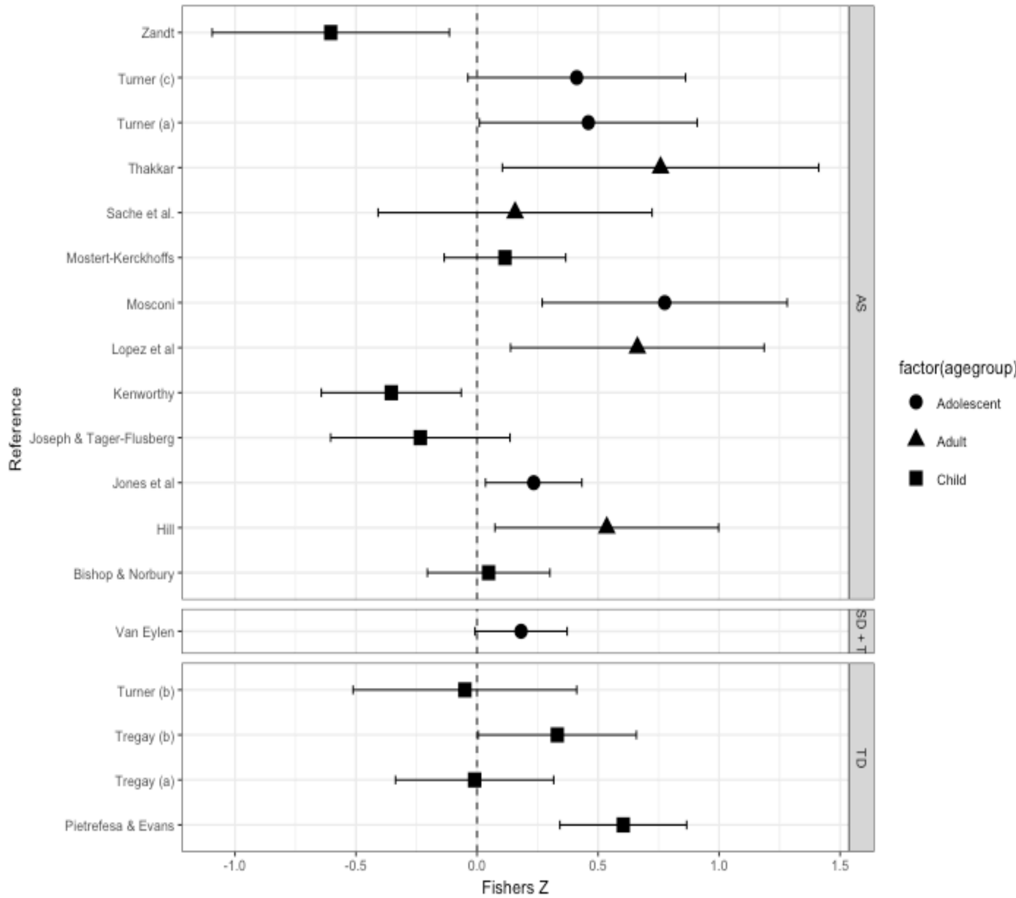

Figure 4. A forest plot containing effect sizes and 95% confidence intervals for the association between inhibitory control tasks and RRB levels and the impact on diagnosis and age groups.

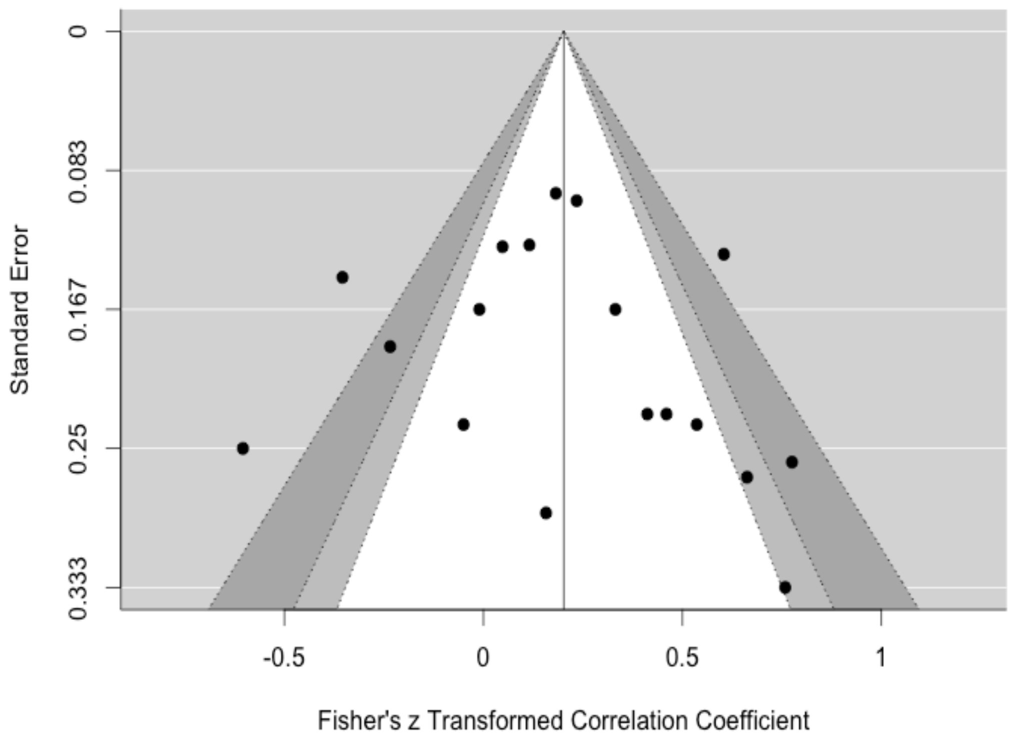

Figure 5. Funnel plot showing standard error of the effect size for the association between inhibitory control tasks and RRB levels. Contour lines are at 1%, 5%, and 10% levels of statistical significance

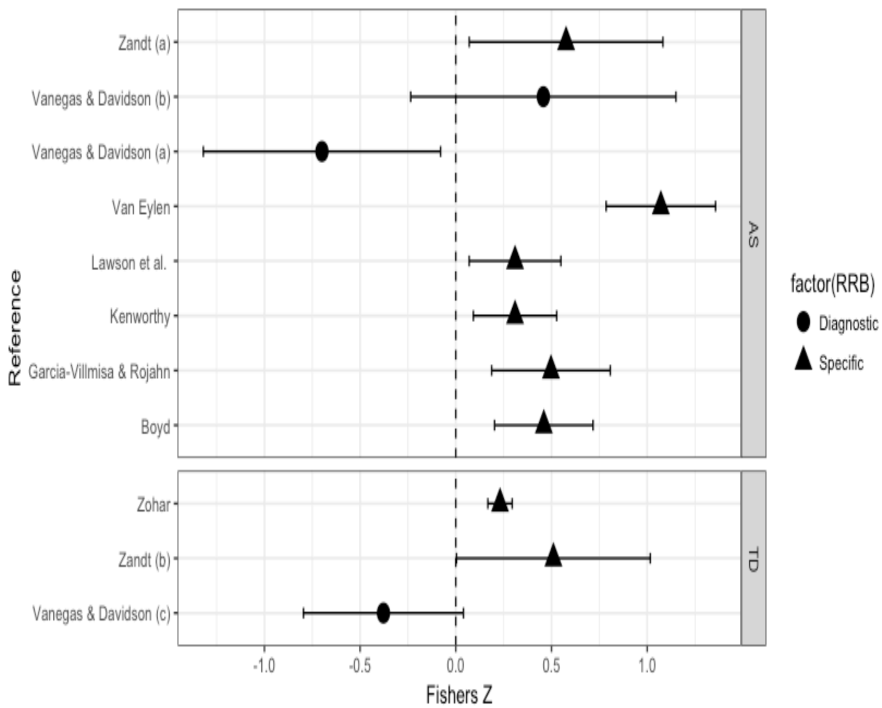

Figure 6. A forest plot containing effect sizes and 95% confidence intervals for the association between parent-rated EF tasks and RRB levels and the impact of diagnosis and diagnostic versus specific RRB measures.

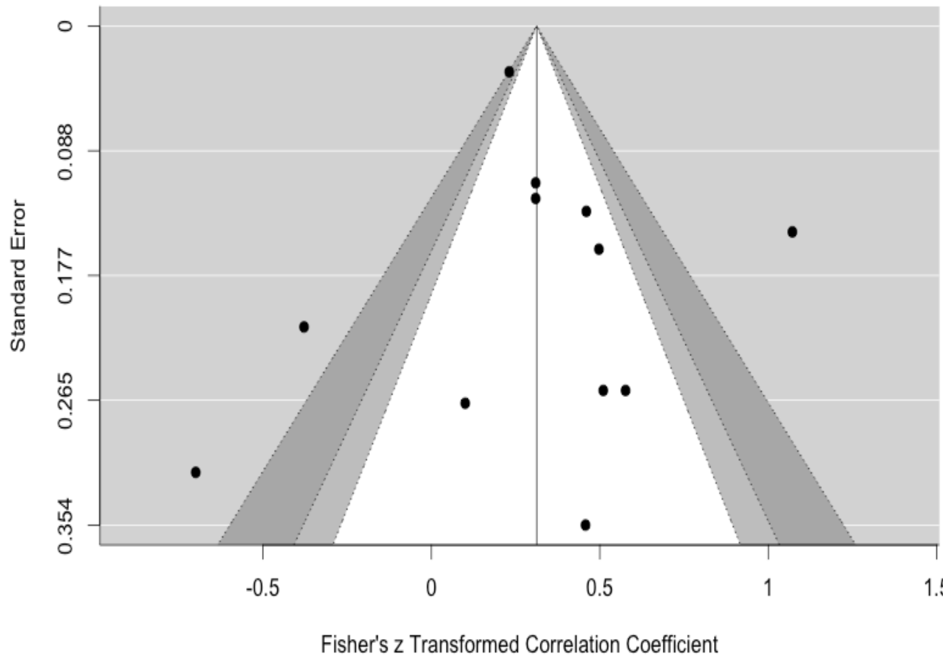

Figure 7. Funnel plot showing standard error of the effect size for the association between parent-rated EF task performance and RRBs levels. Contour lines are at 1%, 5%, and 10% levels of statistical significance.

Table 1:
The effect of each moderator on the overall effect size difference between RRB levels and set shifting performance

Moderator	*df*	Heterogeneity (Q)	*P*
Age (scale)	1	1.05	.30
Task modality	1	0.06	.79
Diagnosis	2	0.30	.85
RRB scale	1	2.53	.11
EF task (WCST versus other)	1	2.68	.10

Table 2:
The effect of each moderator on the overall effect size difference between RRB levels and inhibitory control measures

Moderator	*df*	Heterogeneity (Q)	*P*
Age Age groups:	2	7.92	0.01*
Child	*8*	*37.19*	*0.95*
Adolescent	*4*	*5.83*	*<. 001***
Adult	*3*	*2.37*	*<. 001***
Diagnosis	2	.15	.92
RRB scale	2	.21	.90
Task modality	1	0.65	.41

*p < .05. ** p < .01. *** p < .001

Table 3:

The effect of each moderator on the overall effect size difference between RRB levels and parent-rated EF measures

Moderator	*df*	Heterogeneity (Q)	*P*
Age	1	0.30	.58
Diagnosis	1	1.00	.31
RRB Measure	1	8.83	. 002*
Diagnostic	2	6.31	.46
Specific	7	37.33	<.001**

*p < .05. **p < .01. ***p < .001

Table 4:

Details of the studies that were included in the set shifting analysis

Study	Author(s)	Year	n	Age	Age group	Diagnosis	Diagnostic group	Sub-group	Shifting battery	Outcome measure	Experimenter/ computerised	RRB battery	RRB type	R
1	Liss et al.	2001	21	9.1	Child	ASD	AS	None	WCST	Perseverative responses	Experimenter	Wing behaviour questionnaire	Diagnostic	0.43
2	Lopez, Lincoln, Ozonoff & Lai	2005	17	29.1	Adult	ASD	AS	None	WCST + California Trials Test	Perseverative Responses + letter number task time	Experimenter	ADI-R, ADOS, GARS, ABC	Specific	0.6
3	Van Eylen et al.	2015	78	12.2	Child	ASD + TD	Both*	None	WCST	Perseverative responses	Computerised	RBS-R total	Specific	0.17
4	Vanegas & Davidson (a)	2015	13	9.9	Child	HFA	AS	Yes	WCST	Perseverative responses	Experimenter	SCQ	Diagnostic	0.11
5	Vanegas & Davidson (b)	2015	11	9.4	Child	ASD	AS	Yes	WCST	Perseverative responses	Experimenter	SCQ	Diagnostic	-0.18
6	Vanegas & Davidson (c)	2015	25	8.8	Child	TD	TD	Yes	WCST	Perseverative responses	Experimenter	SCQ	Diagnostic	-0.32
7	Dichter et al.	2010	32	9.9	Child	ASD	AS	None	DCCS	Accuracy-Single task Blocks	Computerised	RBS-R	Specific	0.36
8	D'Cruz et al.	2013	41	15.3	Adolescent	ASD	AS	None	Probabilistic reversal learning errors	Total regressive errors	Computerised	RBS-R, ADI	Specific	0.34
9	Reed, Watts & Truzoli	2013	15	8.3	Child	ASD	AS	None	Discrimination task	Perseverative responses	Experimenter	GARS	Specific	0.69
10	LeMonda, Holtzer & Goldman	2012	22	8.0	Child	ASD	AS	None	WCST	Categories completed	Experimenter	Play sessions	Specific	0.66
11	Yerys et al.	2009	27	10.2	Child	ASD	AS	None	ID/ED	Total errors	Computerised	ADI/ADI-R	Specific	0.43
12	Ozonoff et al.	2004	79	15.7	Adolescent	ASD	AS	None	ID/ED	Total errors	Computerised	ADOS-G stereotyped	Specific	0.2
13	South, Ozonoff & McMahon	2007	19	14.9	Adolescent	ASD	AS	None	WCST	Perseverative responses	Experimenter	ADOS	Specific	0.67
14	Mostert-Kerckhoffs et al.	2015	64	11.3	Child	ASD	AS	None	Set shifting SSV	SSV switch cost accuracy	Computerised	ADOS	Specific	0.64
15	Sachse et al.	2013	15	19.2	Adult	HFA	AS	None	ID/ED	Total errors	Computerised	ADOS	Specific	0.39
16	de Vries & Geurts	2012	31	10.5	Child	ASD	AS	None	Switch task	Comission	Computerised	ADI-R	Specific	-0.07
17	Miller et al.	2013	51	15.1	Adolescent	ASD	AS	None	PCET	Total errors	Computerised	ADI-R	Specific	0.31
18	Bolte (a)	2011	35	14.1	Adolescent	ASD	AS	None	WCST	Perseverative responses	Computerised	ADOS, ADI-R	Specific	0.09
19	Bolte (b)	2011	21	14.1	Adolescent	ASD	AS	None	WCST	Perseverative responses	Computerised	ADOS, ADI-R	Specific	0.1
20	Richards & Lajiness-O'Neill	2015	20	10.5	Child	ASD	AS	None	Navon set shift	Reaction Time	Computerised	ADOS	Specific	0.45
21	Turner (a)	1995	22	12	Child	HFA	AS	yes	ID/ED	Perseverative responses	Computerised	RBI	Specific	0.03
22	Turner (b)	1995	21	11.1	Child	TD	TD	Yes	ID/ED	Perseverative responses	Computerised	RBI	Specific	0.05
23	Turner (c)	1995	18	14	Adolescent	LDA	AS	Yes	ID/ED	Perseverative responses	Computerised	RBI	Specific	0.4
24	Tregay, Gilmour & Charman (a)	2009	39	4.0	Child	TD	TD	Yes	Card sorting test	Overall errors	Experimenter	CRI	Specific	0.19
25	Tregay, Gilmour & Charman (b)	2009	39	7.0	Child	TD	TD	Yes	Card sorting test	Overall errors	Experimenter	CRI	Specific	0.48
26	Jones et al.	2017	100	15.6	Adolescent	ASD	AS	None	Card sort	Number of errors	Experimenter	RBS-R restricted	Specific	0.23
27	Kenworthy et al.	2009	84	9.6	Child	ASD	AS	None	Creature counting	Number of errors	Experimenter	ADOS, ADI-R	Specific	-0.13

*Study was excluded from the diagnosis moderator analysis

Specific measures were created for the sole purpose of measuring RRBs, whereas the dyad measures also included social

Table 5:

Details of the studies that were included in the inhibitory control analysis

Study	Author(s)	Year	Sub group	n	Age	Age group	Diagnosis	Diagnostic group	Inhibition task	Outcome measure	Task modality	RRB measure	RRB type	R
1	Bishop & Norbury	2005	None	63	8.0	Child	AS	AS	Walk-don't walk	Number of trials correctly completed	Experimenter	SCQ & ADOS repetitive	Both	0.04
2	Hill et al.	2006	None	21	31.	Adult	AS	AS	Hayling overall	Overall score	Experimenter	AQ	Specific	0.49
3	Kenworthy et al.	2009	None	49	9.0	Child	AS	AS	Walk-don't walk	Overall errors	Experimenter	ADI & ADOS	Diagnostic	-0.34
4	Zandt	2009	None	19	11	Child	AS	AS	Walk-dont walk	Obsessions	Computerised	CY-BOCS	Specific	-0.54
5	Thakkar	2009	None	12	27	Adult	AS	AS	Antisaccade task	% of antisaccade errors	Computerised	ADI-R	Diagnostic	0.64
6	Mosconi	2009	None	18	17	Adolescent	AS	AS	Antisaccade task, prosaccade errors	% of antisaccade errors	Computerised	ADI-R	Diagnostic	0.65
7	Pietrefesa & Evans	2010	Yes	59	4.0	Child	TD	TD	Global-local Stroop Test	Reaction Time	Computerised	CRI	Specific	0.54
8	Van Eylen	2015	None	78	12.2	Adoles-cent	ASD and TD	Both*	Go-No go	% errors	Computerised	RBS-R total	Specific	0.20
9	Mostert-Kerckhoffs	2015	None	64	11.	Child	AS	AS	SSV inhibition	Total errors	Computerised	RSMB & SRS	Specific	0.12
10	Joseph & Tager-Flusberg	2004	None	31	8.1	Child	AS	AS	Knock and tap	Total errors	Experimenter	ADOS RRB	Diagnostic	-0.41
11	Jones et al	2017	None	100	15.	Adolescent	AS	AS	Opposite Worlds	Inhibition cost score	Experimenter	RBS-R restricted	Specific	0.23
12	Sache et al.	2013	None	15	19.	Adult	AS	AS	Stroop	Interference score	Computerised	ADOS	Diagnostic	0.15
13	Lopez et al	2005	None	17	29	Adult	AS	AS	Stroop	Switching time interference condition	Experimenter	ADOS-G, ADI-R, GARS, ABC-C	Both	0.58
14	Turner (a)	1995	Yes	22	12	Adolescent	AS	AS	Sequence Task	Immediate repetitions	Computerised	RBI overall	Specific	0.43
15	Turner (b)	1995	Yes	21	11.1	Child	TD	TD	Sequence Task	Immediate repetitions	Computerised	RBI overall	Specific	-0.05
16	Turner (c)	1995	Yes	22	14	Adolescent	AS	AS	Sequence Task	Immediate repetitions	Computerised	RBI overall	Specific	0.39
17	Tregay (a)	2009	Yes	39	4.0	Child	TD	TD	Luria's Hand Game	The total number of errors	Experimenter	CRI Overall	Specific	-0.01
18	Tregay (b)	2009	Yes	39	7.0	Child	TD	TD	Luria's Hand Game	The total number of errors	Experimenter	CRI Overall	Specific	0.32

*Study was excluded from the age diagnosis moderator analysis

Diagnostic measures comprised of observations and interviews used to diagnose ASD , specific measures were created for the sole purpose of measuring RRBs.

Table 6:
Details of the studies that were included in the parent-rated EF task analysis

Study number	Author(s)	Year	Sub-groups	n	Age	Age Group	Diagnosis	Diagnostic group	EF questionnaire	RRB Battery	RRB type	r
1	Van Eylen et al.	2015	None	111	12.2	Child	ASD	Both*	BRIEF Shift	RBS-R	Specific	0.79
2	Vanegas & Davidson (a)	2015	Yes	13	9.9	Child	HFA	AS	BRIEF	SQC	Dyad	-0.604
3	Vanegas & Davidson (b)	2015	Yes	11	9.4	Child	ASD	AS	BRIEF	SCQ	Dyad	0.428
4	Vanegas & Davidson (c)	2015	Yes	25	8.8	Child	TD	TD	BRIEF	SCQ	Dyad	-0.361
5	Kenworthy et al.	2009	None	84	9.6	Child	HFA	AS	BRIEF	ADOS and ADI	Specific	0.3
6	Lawson et al.	2015	None	70	10.0	Child	ASD	AS	BRIEF	CBCL	Specific	0.3
7	Boyd et al.	2009	None	61	10.2	Child	ASD	AS	BRIEF	RBS-R	Specific	0.43
8	Garcia-Villmisa & Rojahn	2009	None	43	2.79	Child	ASD	AS	DEX	RBS-R	Specific	0.46
9	Zandt (a)	2009	Yes	18	10.9	Child	ASD	AS	BRIEF	RBQ, CY-BOCS	Specific	0.52
10	Zandt (b)	2009	Yes	18	11.9	Child	TD	TD	BRIEF	RBQ, CY-BOCS	Specific	0.47
11	Zohar et al.	2016	None	952	3.4	Child	TD	TD	BRIEF	CRI	Specific	0.227

*Study was excluded from the age diagnosis moderator analysis

Specific measures were created for the sole purpose of measuring RRBs, whereas the dyad measures also included social communication.

Statement of Author Contribution

In the Chapter entitled, " What are the shared underlying mechanisms between high levels of RRBs and poor set shifting performance in preschoolers and children with ASD?" The authors agree to the following contributions:

Rebecca K. Iversen — 80 % (Data collection, Experimental design, analysis and writing)

Signed: Date: 30/09/2019

Professor Charlie Lewis — 20 % (Writing, Review)

Signed: ____________________________ Date: 30/09/2019

Chapter 2:

What are the shared underlying mechanisms between high levels of RRBs and poor set shifting performance in preschoolers and children with ASD?

Introduction

The meta-analysis in chapter 1 suggests that there are moderate but significant associations between high levels of restricted and repetitive behaviours (RRBs), and poor executive function (EF) skills measured through parental report measures, and set shifting and inhibitory control tasks. If poor EF skills lead to high levels of RRBs, these findings may have clinical implications, as training programs can be developed to help improve these skills, and consequently help manage repetitive behaviours that may be challenging. In order to devise successful interventions however, we need to pinpoint why EF difficulties associate with high levels of RRBs.

Out of the two individual EF skills that we included in our meta-analyses, the strongest relationship was identified with set shifting, suggesting that this may be an area of further interest. Set shifting skills are examined through a wide variety of tasks that all involve a shift to a new thought or action, according to changes in a specific situation (Diamond, 2013). A widely used task to measure the skills is the child-friendly Dimension Change Card Sort task (DCCS, Zelazo et al., 2003). This is a task in which children have to sort cards that vary on two sorting dimensions (e.g. red rabbits and blue boats) after one sorting rule (e.g. shape) then to sort the same cards by another, incompatible sorting rule (e.g. colour). It is a well replicated finding that performance on this task rapidly improves around the age of four (Muller et al., 2006; Zelazo et al., 2003), around the same time that RRBs peak in preschoolers (e.g. Leekam, 2007). Despite the coinciding age-related changes in DCCS performance and RRB scores, performance on the DCCS has not been found to predict RRBs (Dichter et al., 2013). This is surprising considering how performance of the Wisconsin Card Sort Task (WCST, Grant & Berg, 1948) consistently predicts RRBs in adults. In the WCST, individuals are presented with cards that vary along three dimensions (colour, shape and number), and individuals then have to determine the correct sorting rule (e.g. shape), and maintain it until a new sorting rule becomes relevant (e.g. colour). The different findings in the DCCS and the WCST make it possible that the tasks measure different set shifting errors, and that only WCST errors share an underlying process with the behaviours. This chapter will assess this further by systematically addressing different set shifting frameworks to examine if they can explain the behaviours, and thereby help guide the focus of future studies.

Most of the set shifting tasks that have found associations between the behaviours and skills explain the errors through perseverative errors, or the inability to shift away from a dominant response. In Kanner's (1943) original description of RRBs he makes the statement: "it is remarkable the extent to which children will go to assure the perseveration of sameness", p. 63. It is therefore unsurprising that RRBs are often interpreted to be a result of perseverative responding. The accounts that aim to explain perseverative responding are plentiful, proposing a wide variety of underlying mechanisms that may explain the sorting errors and the behaviours.

A well-researched theory that attributes a primary role for inhibitory control is Kirkham, Cruess and Diamond's (2003) attentional inertia hypothesis. This hypothesis suggests that individuals persevere on a rule due to difficulties with redirecting their attention once it is focussed on a particular response. It further suggests that the difficulties may diminish if children are encouraged to refocus their attention to a response by labelling the cards before sorting them, and that difficulties become worse when incorrect rules are made more salient (e.g.Kirkham et al., 2003). Whereas this explanation has face value, as prompts and visual reminders have been found to reduce ASD symptoms (Hodges et al., 2006), it struggles to explain why the WCST, but not the

DCCS task, predicts repetitive behaviours. It is possible that only the WCST predicts RRBs as it introduces more rule shifts and consequently requires higher levels of disengagement than the DCCS task. This is plausible as children must sort six cards before the rule change is introduced in the DCCS, whereas in the WCST adults must sort as many as ten cards before the first rule change is implemented. The additional sorts before each rule shift in the WCST may consequently make it difficult to disengage from a response, as an individual's attention may get "stickier" the more sorts s/he completes. This type of hypothesis would be consistent with findings in Doebel and Zelazo's (2015) meta-analysis that suggests that more pre-switch trials predict lower switching rates. Moreover, research has shown that verbal teacher prompts during peer interactions has decreased lower-level behaviours in children with ASD (Lee, Odum & Loftin, 2007).

Other accounts such as the active-latent account (Munakata, 1998) and the Cognitive Complexity and Control (CCC, Zelazo et al., 2003) theory attribute a secondary role for inhibition.

The CCC theory suggests that whereas young children are able to construct if-then rules that they can apply in card sorting tasks (e.g. if the card is blue it goes here and if the card is red it goes there), they are unable to construct an embedded if-if-then rule (e.g. if shape and rabbit then here, but if colour and red then there). Again, this theory has face value, as if children have not yet developed self-reflection skills to develop different rules for different situations, this may lead to high RRBs, such as circumscribed interests (Rajendran & Mitchell, 2007). It may, however, not account for the development of other behaviours such as stereotypies. This is not necessarily problematic as recent RRB theories suggest that the behaviours should be researched through sub-groups of lower and higher-level RRBs, as different behaviours may follow different trajectories (Turner, 1999). It also struggles to explain why only the WCST predicts RRBs, unless the complexity of the WCST plays a crucial role. This is possible as the WCST may require the development of more higher-order rules than the DCCS. Whereas only two higher-order rules needs to be created in the DCCS, the WCST requires three, one for each dimension.

The active-latent account takes on a different perspective by suggesting that strong memory representations make it difficult to override the initially relevant, but now irrelevant, stimuli (Morton & Munakata, 2002; Munakata, 1998). In the WCST, the rule change is sudden and adults are not reminded of the rules. Instead, individuals are given feedback ("correct" and "incorrect"). A memory confound may therefore be responsible for the association between high RRB levels and poor WCST performance. Morton and Munakata (2002) suggested that the most effective strategy to help children overcome perseveration is to scaffold the use of new rules, as this will lead to changes that favour the new rule. Moreover, research suggests that verbal prompts and visual reminders reduce ASD symptoms (Hodges et al., 2006). This account could consequently explain why no associations have been found with the DCCS as the rule change is emphasised in this task, and participants reminded of the rule prior to each trial, making it less likely that children will create strong memory representations.

So far we have reviewed accounts that suggest that perseverative responding develops as a result of an inability to redirect attention once focussed on a particular dimension, an inability to create higher-order rules, or a result of memory confounds. Whereas these accounts can account for poor performance on set shifting tasks, not enough evidence has yet been provided to account for why the WCST, but not the DCCS task, associates with high RRBs.

Another line of research that is receiving increasing attention is the suggestion that children and adults find it difficult to activate a previously ignored pre-switch dimension in the post-switch (Maes, Damen & Eling, 2004; Maes, Vich & Eling, 2006; Müller, Dick, Gela, Overton & Zelazo, 2006; Zelazo et al., 2003). This ability has been measured in tasks modelled on the DCCS and the WCST. Zelazo et al. measure the errors in preschoolers through a DCCS adaptation that they refer to as Negative Priming (NP). In this task, previously relevant sorting stimuli were replaced by new sorting exemplars from the same dimension (e.g. boat and house were replaced with rabbit and train), but previously irrelevant stimuli remained after the rule switch (e.g. sort by blue and red). It was argued that this task prevents perseverative responding, as children can no longer perseverate on previously relevant exemplars. Their results suggested that set shifting development consists in part of the ability to activate previously ignored stimuli. Doebel and Zelazo (2015) offer further support for this conclusion in a meta-analysis that combined the results in six studies that implemented similar NP tasks.

Similar set shifting adaptations have been created for the WCST, with one crucial difference, as the WCST adaptations replaced the previously relevant dimension (e.g. shape) with a novel dimension (e.g. size), instead of new task exemplars. More specifically, Maes, Damen and Eling (2004) created a learned irrelevance (LI) task, in which an individual had to first sort cards after shape (circles and squares), and ignore colour (blue and red), before they had to activate the previously irrelevant colour (blue and red) dimension, and ignore a novel size (small and big) dimension. In support of Zelazo et al's (2003) findings, Maes and colleagues found evidence to suggest that the ability to activate previously irrelevant responses plays an important role in the development of set shifting skills. In fact, they found that the ability to activate previously irrelevant stimuli was more difficult than the ability to shift away from dominant responses.

The different findings in the NP and LI tasks highlight that the ability to activate previously irrelevant stimuli may be more important in the development of set shifting skills than was first believed to be the case. It cannot, however, explain why the DCCS task has not been found to predict repetitive behaviours. The differences in the two tasks may mean that they measure different errors. More specifically, whereas the NP task replaces the relevant stimuli with new exemplars of the same dimension in the post-switch to prevent perseverative errors, this task may still be measuring perseveration. Research has shown that children persevered on dimensions in an adapted DCCS version (Hanania, 2010). In contrast, the LI task replaces the previously relevant dimension with a novel dimension making it impossible to perseverate on the rule. This may then make this task a purer measure of LI. No one has yet examined if performance on the NP and LI tasks predict RRBs.

Several frameworks have been developed to explain the difficulties with activating previously irrelevant stimuli. An account that aims to explain the findings on the DCCS-like adaptation is the Cognitive Complexity and Control-revised (CCC-r, Zelazo et al., 2003) theory. This account attributes a secondary role for inhibition by arguing that children fail the task because of the interfering effect that arises when a child needs to activate a previously irrelevant rule and suppress attention to previously relevant rules. In order to overcome the conflict that arises, a child needs to create higher-order rules to suppress the activation of the pre-switch rules, as well as to activate previously irrelevant rules. Like the CCC theory, this account struggles to explain why one task, but not the other, correlates with RRBs. Unless, the NP adaptation does not successfully measure the ability to activate previously irrelevant responses. If so, it may be that only the LI task successfully measures the errors, making it possible that the inability to activate previously irrelevant rules is the shared underlying mechanism for the behaviours and the errors, and that only the WCST adaptation successfully measures it.

An alternative explanation for the difficulties with activating previously ignored stimuli is offered through Neill's (1997) episodic retrieval account. This suggests that irrelevant stimuli are marked with a "do not respond" tag that conflicts with the situation when the stimuli turns relevant at a later stage, creating NP errors. This account is similar to the active-latent account (Morton & Munakata, 2002; Munakata, 1998) in many ways, as they both suggest that memory confounds make it difficult for a child to master set shifting tasks. This explanation is a contender, as individuals may mark stimuli with a "do not respond tag" in both tasks, which is then likely to make it difficult to overcome the memory tag when it later becomes relevant. With reference to the DCCS task however, it has been argued that children do not make memory tags, as they are reminded of the rules prior to each trial. If this is the case, memory may not be able to explain why children fail the NP version of the DCCS task, yet it can offer an explanation for, why one, but not the other predicts RRBs.

Finally, it is possible that an individual's difficulties with activating previously irrelevant responses can be explained through an automatic inhibition framework (Maes et al., 2004). This account suggests that the LI task involves automatic inhibition, as the purpose of this task is to pre-expose an individual to the target stimuli in pre-switch. This then suggests that the irrelevant part of a stimulus is continuously inhibited throughout the pre-test phase, since an individual's attention is directed to other relevant aspects. This then leads to automatic inhibition when a situation occurs in which an individual must activate previously ignored responses. This type of inhibition is arguably less effortful than the ability to shift away from previously dominant stimuli, as this is often said to require voluntary inhibition. More specifically, the aim of these tasks is for an individual's attention to be focused on the target stimulus in the pre-switch, for then to ascertain if they can successfully switch away from the stimuli when it later becomes irrelevant. If automatic inhibition is harder to overcome than controlled inhibition and only the LI task measures this ability, it may help explain why the WCST but not the DCCS task associates with RRBs. No one has yet examined this.

Despite the intriguing findings that the ability to activate previously ignored stimuli plays a crucial role in the development of set shifting skills in the child (Muller et al., 2006; Zelazo et al. 2003) and adult literature (e.g. Maes, Damen & Eling, 2005), no one has yet isolated this ability from the ability to switch away from relevant stimuli, and examined if this type of fine-grained analysis can help to explain the inconsistent results between the behaviours and performance on set shifting measures. Moreover, despite recent developments that suggest that RRBs should be examined through sub-groups, no one has yet compared performance on the different errors to scores on the two sub-groups of behaviours. In the next chapter in this thesis, we will therefore examine the relationships between sub-groups of RRBs and the two types of set shifting errors in more depth, as this may help explain the relationship between the two.

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Statement of Author Contribution

In the Chapter entitled,
"Do rule activation errors explain the persistence of repetitive behaviours in typical development and ASD?" the authors agree to the following contributions:

Rebecca K. Iversen — 80 % (data collection, analysis and writing)

Signed:  Date: 30/09/2019

Professor Charlie Lewis —20 % (writing, and review)

Signed:  Date: 30/09/2019

Chapter 3:

Do rule activation errors explain the persistence of repetitive behaviours in typical development and ASD?

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Abstract

In set shifting tasks, respondents need to suppress a relevant response and attend to previously irrelevant aspects of the environment. Preschoolers tested on the Dimensional Change Card Sort (DCCS) show dramatic change in this ability at age 4, but this does not relate to a similar decline in restricted and repetitive behaviours (RRBs), which peak in this age group and persist in Autism. We hypothesized that the difficulties with activating previously irrelevant aspects of the environment may play a role in linking set shifting to RRBs. Two studies compare variations on the standard DCCS with a new method in which the relevant response is no longer available. 177 typically developing (TD) children (m= 3.9 years: Experiment 1) and 90 children with Autism or developmental delay (DD) (m= 7.7 years: Experiment 2) were assessed on card-sorting tasks and RRBs, measured in a parental questionnaire. In both studies no differences were found between sorting performance on the tasks, but the TD children showed the expected 3-4 age shift. Moreover, the results showed that children who struggled to activate a previously irrelevant dimension engaged in more RRBs. This suggests that the child's problems with activating a previously irrelevant cue may reveal biases of attention that explain the persistence of RRBs in typical and atypical development.

Keywords: set shifting, restricted and repetitive behaviours, autism, pre-schoolers

1. Introduction

In the preschool years, children display Restricted and Repetitive Behaviours (RRBs), peaking at 62% of two year olds, but still evident in 58% of three and four year olds and 49% at age 5 (Evans, et al. 1997). These manifest themselves in terms of repetitive motor movements, a rigid adherence to routine, a preoccupation with restricted patterns of interest, or unusual sensory interests (Leekam, 2007). RRBs are important because they persist in some clinical populations, notably Autism Spectrum Disorder (ASD, Wolff et al., 2014), but also Williams syndrome (Royston et al., 2018), Obsessive Compulsive Disorder (OCD, Ruzzano, Borsboom & Geurts, 2015) and other syndromes (Greaves, Prince, Evans & Charman, 2006). Despite several attempts at identifying the processes involved in these behaviours, these are still unclear (e.g., Leekam, Prior, & Uljarevic, 2011; Lewis & Kim, 2009). Both these reviews downplay the role of executive function skills in the control of these behaviours, but three recent meta-analyses suggest that two of these skills (set shifting, inhibition) show consistent relationships with RRBs (**blinded*for*review***). In this paper, we explore recent developments in research on set shifting through a large normative study, and children with ASD, attempting to identify more precisely its relationship to RRBs.

Set shifting refers to the ability to switch flexibly between different mental sets (Monsell, 2003), as it helps a person to select and adapt different strategies depending on their immediate circumstances. For example, parents often tell their children that it is time to stop playing and get ready for school. A child with poor set shifting skills may have a tantrum as they find it difficult to shift from one activity to another, while her sibling with good set shifting skills would be more likely to adjust to such a change. Set shifting has long been argued to have clear parallels with RRBs (Turner, 1999; Boyd et al., 2011; Van

Eylen et al., 2015), as a child who struggles to shift between different response sets may also be likely to persevere on a particular activity at the exclusion of all others, and consequently engage in high levels of RRBs.

Despite the possible links between set shifting and RRBs, the evidence is contradictory and age-related. The Wisconsin Card Sort task (WCST, Grant & Berg, 1948) is the most widely used neuropsychological test to assess set shifting skills in adults. This task requires multiple shifts of attention, as the participant needs to sort cards from one dimension (e.g. colour) then suddenly change to another (e.g. shape or size) without warning. The WCST has long also been known to be a predictor of RRBs (e.g. Liss et al., 2001; Lopez, Lincoln, Ozonoff & Lai, 2005; South et al., 2007). Despite the elevated levels of RRBs in children, the same association has not been found with the much simpler Dimensional Change Card Sort (DCCS; Frye et al., 1995) (Dichter et al., 2010), a widely used task measuring set shifting skills in early childhood. This task involves only one shift in attention and the child is continually informed about the need to change their sorting strategy.

There are many variations of the DCCS (see meta-analysis by Doebel & Zelazo, 2015) and the WCST (e.g. Maes et al., 2004), in attempts to establish the cause of errors on both tasks. Traditionally, perseverative responding, or the inability to disengage attention from a previously relevant dimension, is regarded as the cause of poor performance. For example, in the standard DCCS the child may arrange cards into (e.g.) ‘dogs’ and ‘cars’, but then has to sort them into ‘blue’ and ‘yellow’ objects, disregarding the fact that each set consists of both dogs and cars. A perseveration with the first rule is found even when the properties of the second rule are changed. For example, in the

Partial Change condition, the first rule (e.g. the two shapes) remains the same while the exemplars of the second rule (e.g. colour) change (e.g. from white vs. green to yellow vs. black; see example in Figure 1).

Zelazo et al. (2003) carried out other DCCS tasks to test the hypothesis that failure in the task concerns the ability to hold in mind the two rules and to apply each in its appropriate setting (Cognitive Complexity and Control-revised theory-CCC-r, Zelazo et al., 2003). To test this theory, they conducted a Negative Priming (NP) manipulation in which the previously irrelevant dimension (e.g. colour in Figure 1) remains the same, while the previously relevant stimulus (e.g. shape) is changed (e.g. from dogs and cars to dinosaurs to birds). That three-year-old children fail to use the new rule in this condition was thought to provide strong evidence for the CCC-r theory in that it demonstrates a clear inability to reconcile the two relevant rules.

However, studies of adults (e.g. Maes, Damen & Eling, 2004; Owen et al., 1993) have increasingly found that the inability to attend to previously irrelevant information plays a key role in set shifting performance. This effect is often referred to as learned irrelevance (LI) in the adult literature (e.g. Owen et al., 1993), which is largely consistent with the term negative priming (NP, Zelazo et al., 2003) in the developmental literature. While LI and NP appear to be two different ways of describing an inability to activate previously ignored stimuli, the different task designs that they derive from make it possible to construct a greater diversity of card sort procedures, identifying individual skills which may explain the correlation with RRBs.

Maes et al. (2004) devised a variation of the WCST that resembles the DCCS. In their LI condition the previously irrelevant rule (e.g. colour) became relevant in the post-switch, but the previously relevant rule (e.g. shape) was replaced with a novel dimension (e.g. size). Thus, in this condition shape is no longer a factor as all of the stimuli contain the same information (e.g. in Figure 1 all of the shapes are birds). By removing the previously irrelevant stimulus, their LI condition may be a purer measure of the ability to activate previously irrelevant dimension than each of the other conditions illustrated in Figure 1. Previous studies suggest that children do not perseverate at the level of dimensions in the DCCS (Zelazo et al., 2003) Research by Hanania (2010) suggests otherwise as she found that children continued to persevere on a sorting dimension after she added a third phase to the standard DCCS task that introduced new sorting exemplars of the previously relevant dimension. Maes et al. (2004) also added a control task in which they measured whether errors could be caused by perseveration on the former relevant dimension. In their Perseveration task, the previously relevant dimension (e.g. shape in Figure 1) was retained but made irrelevant in the post-switch, whereas the previously irrelevant dimension (e.g. colour in Figure 1) was replaced with a novel dimension (e.g. number; see Figure 1). Adults made fewer errors in this condition than in the LI task, offering support for the view that the key to set shifting is the ability to apply a previously irrelevant rule.

To understand the critical differences between these tasks we must compare and contrast the different versions to see how they complement two key theoretical frameworks in the literature. First, Zelazo et al.'s (2003) CCC-r theory suggests that in order to pass the DCCS task children need create a rule structure that enables them simultaneously to reflect on the pre- and post-switch rules (e.g. "if shape game and if dog, then place here, but if colour game and white, then put there"). Three-year-olds can construct these types of rules during the pre-switch (e.g. "if white, then sort here, but if green, then sort there"), but cannot integrate the pre- and post-switch rules in the post-switch and default back to the dominant pre-switch rule. This theory can explain the results in the Partial Change and NP tasks. In the Partial Change version the pre-switch rule may maintain a high level of activation, which carries over into the post-switch stage. In the NP condition the rule that becomes relevant had been inhibited during the pre-switch phase. The CCC-r theory would then also make a similar prediction concerning a 3-4-year shift in performance on the perseveration task, since the previously relevant rule is still present in the post-switch in this version. However, this framework would predict that performance on the LI task should be significantly easier, as the previously relevant dimension is no longer involved in the post-switch, but instead replaced with a novel dimension. There is therefore no longer a conflict to reconcile between the rules in the pre- and post-switch in this version.

In contrast to the CCC-r theory, Maes et al. (2004) focused on adults' abilities to continue to attend to one dimension of a stimulus (perseveration) and then switch to a previously suppressed dimension (learned irrelevance). Their attentional theory concerns why adults continue to make errors, which they attribute to attentional biases, coupled with the weak associative strength that suppressed rules continue to have after a switch is made. We apply Maes et al.'s distinction to the 3-4-age shift in Zelazo's DCCS task. Consistent with the CCC-r theory, the attentional account would explain the standard DCCS performance and most of the variations in Figure 1 in terms of the associative strength of the pre-switch dimension and the suppression of other aspects of the stimulus. It would be hypothesized that 4-year-olds have developed two abilities. The first is to suppress a previously activated attentional bias, while the second is to activate a dimension of the stimulus that has been suppressed.

The two theories differ crucially in their explanation of performance in the LI condition. Following Maes et al., (2004) we predict that this version is equally difficult because the core skills within set shifting involve the re-activation of the previously suppressed dimension of the stimulus. The LI variation removes the perseverative component of the first dimension, so it becomes a pure test of the ability to activate a perceptual link, which has previously been suppressed. Indeed, Maes et al. would suggest that all the versions of the task presented in Figure 1 cause difficulties as they involve the automatic inhibition of the now relevant dimension, and they show that this presents problems even in adults performing tasks with as many trials and shifts as the WCST.

The second topic of this paper addresses how recent advances on set shifting development may relate to RRBs. According to the DSM-IV (APA, 1994), and ICD-10 (WHO, 1992) RRBs can be divided into four subgroups, these are: stereotypies, preoccupation with objects, restricted interests and non-functional routines. The first two of these subgroups are often combined and referred to as lower-level repetitive behaviours (Prior & Macmillan, 1973). Examples of these behaviours are hand flapping and repetitively ordering objects. The second sub-group is referred to as higher-level, and consist of circumscribed interests (CI), such as intense interests in selective topics (e.g. space, trains), and insistence of sameness (IS), such as excessive adherence to routines. Lower-level behaviours are pronounced in developmentally younger individuals (Hundley et al., 2016; Larkin, Meins, Centifanti, Fernyhough & Leekam, 2017; Szatmari et al., 2006) and in a wide variety of disorders (Berry, Russell & Frost, 2018), whereas higher-level behaviours are often referred to as ASD specific (Turner et al., 1999), but also prevalent in pre-schoolers (Evans et al., 1997). Research by Mosconi et al. (2009)

has also suggested that only higher-level RRBs can account for the behavioural flexibility problems that are widely documented in individuals with ASD.

The EF hypothesis conceptualises RRBs to reflect the inability to adapt flexibly to changing environmental cues (e.g. Russell, 1997). In particular, cognitive inflexibility is argued to be consistent with the repetitiveness and rigidity of the behaviours (Turner, 1999), and predominantly higher-level RRBs (Miller et al, 2015; Mosconi et al., 2009). However, key RRB reviews (e.g. Leekam, Prior & Uljarevic, 2011; Lewis & Kim, 2009) state that the EF hypothesis lacks specificity, and that two decades of research have not been able to validate the hypothesis. Instead researchers turn towards explanations of 'pathophysiology' (e.g. Lewis & Kim, 2009), developmental immaturity (e.g. Leekam et al., 2011), behavioural manifestation of anxiety (Wigham et al., 2015), or they focus on the impact that developmental and individual factors, such as IQ and age, have on the presentation of different RRB sub-groups (e.g. Berry et al., 2018). Over the past decade new evidence has emerged in favour of the EF hypothesis (e.g. D'Cruz et al., 2013; Miller et al., 2014; Mosconi et al. 2009; Yerys et al., 2009), often highlighting the link between RRBs and set shifting, mostly measured through the WCST (Lopez et al., 2005; South et al., 2007; Van Eylen et al., 2014). However, these studies have focused on adults and measured RRBs on a unitary scale. The lack of research in children is surprising, considering the prevalence of these behaviours, as is the lack of focus on sub-types of RRBs in correlational analyses, given the heterogeneity in the behaviours displayed in young children, and findings by Mosconi et al. (2009) that only higher-level RRBs can account for the behavioural flexibility problems that are widely documented in individuals with ASD. The studies presented here build on the new research finding a relationship between RRBs and set shifting in adults, and studies suggesting that the ability to activate previously irrelevant stimuli plays an important role in set shifting development in TD adults (e.g. Maes et al., 2004; Maes Vich & Eling, 2006), and cognitively able individuals with ASD (Turner et al., 1995). To our knowledge, no studies have yet examined these links in pre-schoolers, or in children with ASD.

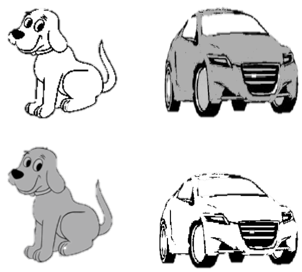
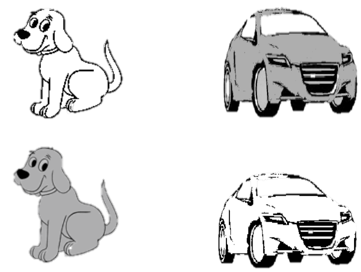
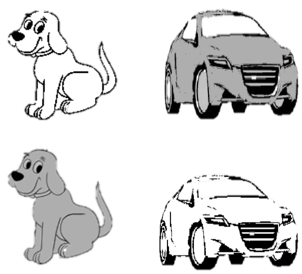
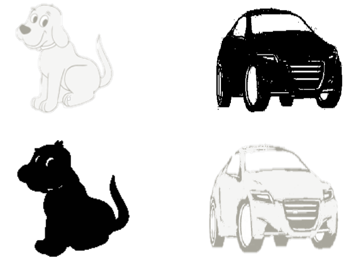
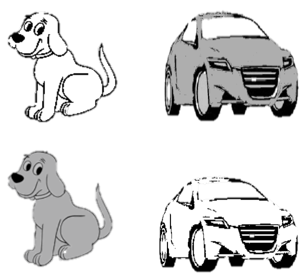
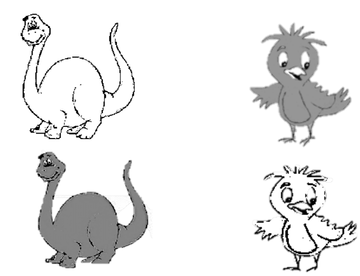
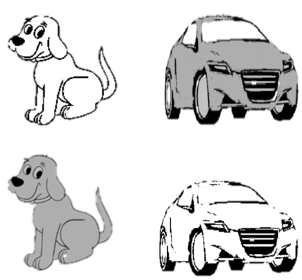
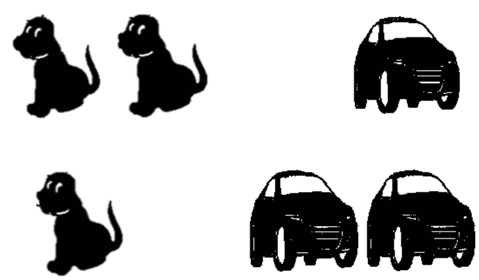
We will test two research questions, which compare the CCC-r explanation with an attentional explanation. The first research question concerns whether similar age-related patterns can be found between the ages of 3-4 in all five versions of the DCCS. Our second research question addresses the relationship between set shifting performance and RRBs. These factors were examined in two experiments, through a large normative study, and through samples of children with ASD and DD. We predict one out of three outcomes.

First, following research that has shown that RRBs start to decline around the age of four (Evans, et al., 1997; MacDonald et al., 2007; Uljarevic et al., 2017), and studies that have shown that a majority of children pass the DCCS around the same age (e.g. Doebel & Zelazo, 2015), performance on all of our task versions may predict a child's RRB levels. Confirmation of this hypothesis will, however, contradict the limited research on the topic, as Dichter et al. (2010) found that the better individuals performed on the DCCS tasks, the more RRBs they displayed.

Secondly, if we find support for the CCC-r framework, performance on all of our task versions apart from the LI task will associate with a child's reported RRBs. This type of result would offer support for the CCC since all four of these task versions children may persevere on the pre-switch dimension due to the inability to inhibit previously relevant rules, as well as redirect attention to previously ignored rules. Likewise, an individual may get stuck in a specific behaviour or thought due to difficulties with both inhibiting the dominant response, and activating a behaviour that was previously irrelevant to the situation. This hypothesis suggests that children at around the age of four develop a rule system that helps them process rules more flexibly so that they can override dominant rules, and activate previously irrelevant rules when appropriate. The same rule system may then also help children determine when it is appropriate to engage in different behaviours, and thus control their perseverative tendencies. However, this type of explanation may not hold up for two reasons. It would contradict previous research on the topic as it suggests that poor DCCS performance should predict less repetitive behaviour. In addition, studies by Evans et al. (1997) and Uljarevic et al. (2017) show that while children engage in fewer RRBs around the age of four, the behaviours were still prominent. As it has been repeatedly shown that children pass the DCCS by the age of five (Doebel & Zelazo, 2015), this implies that perseveration on a dominant rule is unlikely to account for the child gaining control over behaviours that are repeated.

Thirdly, given that previous research has established strong links between WCST performance and RRBs (e.g., Van Eylen et al., 2015), and that research suggests that errors on tasks like the WCST are mainly a result of the inability to activate previously ignored rules (Maes et al., 2004; Maes Vich & Eling, 2006), we may find that the LI condition is the only version of card sorting tasks that assesses biases of attention that resemble the RRBs. This prediction would support the attentional theory. More specifically, in a set shifting task, relevant and irrelevant stimuli are repeatedly presented. The attentional theory suggests that an initial analysis of the stimuli must take place in which it is decided if the stimuli must be attended to or ignored. This analysis consists of an excitatory process, where the selected object receives further analysis, and an inhibitory process, where ignored distracters are actively inhibited. In the LI task, the target dimension is repeatedly exposed as a distractor in the pre-test, meaning that it may be continuously inhibited throughout the pre-switch phase. Similarly, an individual who engages in high levels of RRBs may actively inhibit irrelevant activities, as well as attempts of redirection by other people. If an individual then inhibits certain activities or sorting stimuli over some time, they may end up creating an internal representation of the stimuli that eventually becomes automatic. An attentional orienting response is then only triggered when there is a mismatch between the current environment (e.g. when previously irrelevant behaviours/stimuli must be selected) and the internal representations of that environment. The difficulty with overriding this automatic response may then lead to set-shifting errors and repetitive cycles of well-learned behaviours.

Experiment 1 tests the three hypotheses listed above. Given the issues and contradictory evidence surrounding the first two we predicted that the evidence that we collected would support the attentional theory and that children's performance on the LI task would show clearer links with their parents' reports of their RRBs.

Standard DCCS	 Relevant= shape, Irrelevant= colour	 Relevant= colour, Irrelevant= shape
Partial Change	 Relevant= shape, Irrelevant= colour Relevant= shape, Irrelevant= colour	 Relevant= colour, Irrelevant= shape
Negative Priming	 Relevant= shape, Irrelevant= colour	 Relevant= colour, irrelevant= shape
Perseveration	 Relevant= shape, Irrelevant= colour	 Relevant= Number, Irrelevant= shape

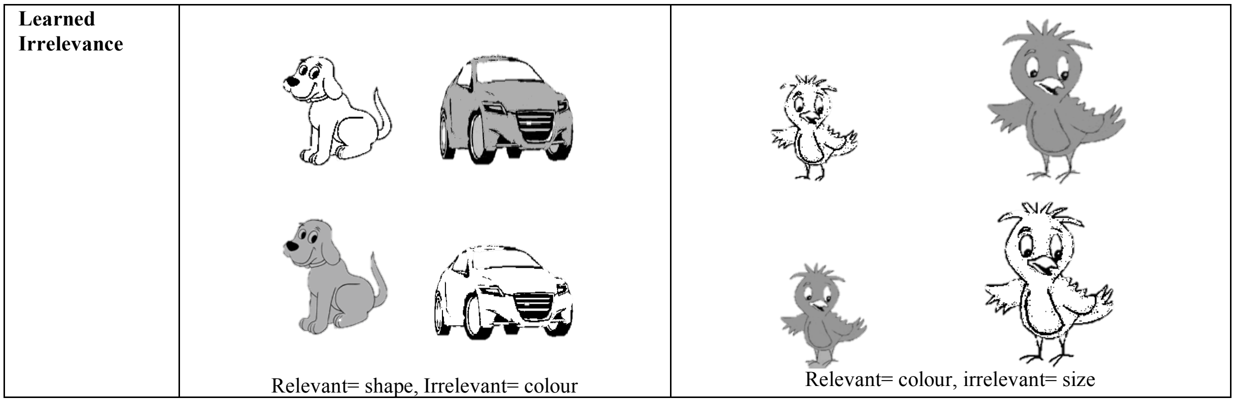


Figure 1. Card sorting conditions

2. Experiment 1

2.1 Participants:

Two hundred and two children aged between 33 to 64 months (mean= 46.6, SD= 7.0) were recruited from thirteen nurseries and primary schools. Eight of these children were omitted from the full analysis as they did not meet pre-switch criteria (sort 7 out of 8 sorts correctly), and eighteen were absent on the day of testing at that school. Parents of twenty-seven children declined to complete the questionnaires, and one nursery refused to distribute it (n=10).

The exclusion criteria meant that the card sorting analysis consisted of one-hundred and seventy-seven children, one-hundred and four were three years of age (mean=42.4, SD= 3.2), and seventy-three had turned four (mean=54.1, SD= 4.6). The full regression analysis consisted of 141 children, 89 were three years of age (mean= 42.3, SD= 3.3), and 52 had turned four (mean= 53.8 months, SD= 4.4). Checks using ANOVA revealed that there were no significant age differences between the 3- year-olds [$F(4, 84)=.94$, $p= .44$] *or* the 4-year olds [$F(4, 84)=.24$, $p= .92$] across the five sorting groups in the card sorting analysis. Moreover, there were no significant age [$F(1, 140)=.40$, $p=$

.81] or RRB score differences [$F(4, 140)=1.42$, $p= .23$] across the five sorting groups in the RRB analysis.

Repetitive Behaviours:

Restricted and repetitive behaviours were assessed through the 33-item parent-administered Repetitive Behaviour Questionnaire (RBQ, Turner, 1995). A total repetitive behaviour score was computed as well as a sensory/motor behaviour score and an insistence of sameness/circumscribed interest score, following guidelines by Zandt, Prior and Kyrios (2007). See Table 1 for RRB scores.

Table 1: *Participant scores on the RBQ. Mean (SD)*

Scores on RRB questionnaires	**3 year olds:**	**4 year olds:**
Overall:	9.93 (7.43)	8.21 (7.61)
Lower-level:	5.01 (4.37)	4.21 (4.48)
Higher-level:	3.43 (2.86)	2.88 (3.63)

2.2 Procedure:

** University ethics committee approved this study. Parents provided informed written consent, and children gave assent prior to participation. Each child was randomly allocated to one out of five experimenter administered card-sorting conditions, in which they had to switch to a shape, colour, size or number rule, depending on which task set they were presented with. All five card-sorting conditions followed the standard Dimensional Change Card Sort (DCCS; Frye, Zelazo & Palfai, 1995) procedure (See

Figure 1 for a pictorial representation of the conditions). The only differences were that a child was asked to sort eight instead of six test cards in each phase, and target cards were replaced after the rule switch in all tasks, apart from in the DCCS. Target cards were affixed to two sorting boxes (16cm x 13cm x 13cm). All test cards were 90mm x 90mm, and could display pictures of four different shapes (dogs, bikes, cars, apples), two sizes (small and big), two numbers (1 and 2) and eight colours (red, blue, purple, white, yellow, black, orange and grey). Children were given no feedback as to whether they sorted the cards correctly or not, but they were reminded of the rules prior to each trial. A child had to sort seven out of eight cards correctly in order to pass the pre- and the post-switch phases.

3. Results:

Card sorting comparisons

Table 2 presents the means and SDs for the different sorting tasks. We performed a series of Generalized Linear Mixed-Effects Models (Baayen, 2008; Jaeger, 2008), to examine whether the age-related changes that are widely seen in the DCCS were evident in the card sorting performance, and to compare the relative difficulty of the five sorting tasks. All models were conducted using the glmer function from the lme4 package in R (Bates, Maechler, Bolker, & Walker, 2015). We started with a null model containing random effects of participants and switch dimension on intercepts. We considered the effects of age group (3-and 4-year olds) and task type (DCCS, Partial Change, NP, Perseveration and LI), as main effects and interactions terms. Adding the fixed effect of age group to a model with only random effects significantly improved the model fit (χ

$^2(1) = 17.63$, p = <.0001). Closer inspection showed that 3-year olds performed significantly less accurately than the 4-year olds (estimate = 4.46, SE = .90, z = 4.93, p = <.0001). The same analysis also found that neither task type ($\chi^2(1) = 2.42$, p = .12), nor the interaction term type of task by age group improved the model fit ($\chi^2(2) =3.00$, p = .22), suggesting that whereas there was a significant age-related sorting effect for the overall sorting scores, the improvements were consistent across all tasks.

To check whether the inclusion of random effects was justified in the final model we compared models using likelihood ratio tests, using the REML = FALSE setting. Likelihood ratio tests with the same fixed effect but varying in random effects of participant, and dimension suggested that the inclusion of random effects of participant ($\chi^2(1) = 919.79$, p = <.0001) was justified. There was not, however, a significant difference between models including the full random effect structure and one containing only dimension ($\chi^2(1) = .09$, p = .77), suggesting that only the inclusion of the random effect of participant on intercepts was justified. See Table 3 for full model fit.

Table 2:
Sorting performance on the card-sorting tasks and t-test comparison by age

Sorting score	3 year olds: Mean(SD)	Pass(%)	Fail(%)	4 year olds: Mean(SD)	Pass(%)	Fail(%)	Age: t
Overall (n=177)	3.35 (3.3)	32 (29)	79 (71)	5.18 (4.5)	47 (64)	26 (36)	-3.5
DCCS (n=37)	2.36 (3.22)	4 (18)	18 (82)	4.73 (3.88)	9 (60)	6 (40)	-2.02
Partial Change (n=35)	3.86 (3.33)	8 (36)	14 (64)	5.00 (3.60)	9 (60)	6 (40)	-.98
NP (n=35)	3.45 (3.62)	7 (33)	14 (67)	4.60 (3.89)	9 (60)	6 (40)	-.90
Perseveration (n=35)	2.95 (3.40)	6 (25)	18 (75)	5.64 (3.71)	10 (71)	4 (29)	-2.21
LI (n=35)	4.19 (3.17)	7 (33)	14 (67)	6.00 (3.31)	10 (71)	4 (29)	-.16

Table 3:
Summary of general linear mixed effects model of overall card sorting performance, including fixed effects of age groups.

Fixed effects	Estimated Coefficient	SE	Wald confidence intervals 2.50 %	97.50 %	Z	Pr(>|z|
(Intercept)	-5.74	1.31	-8.50	-3.30	-4.40	1.1×10^{-5}
Age group	4.33	.89	2.69	2.23	4.86	1.2×10^{-6}
Random effects	Name	Variance	St. dev.			
Participant	(Intercept)	27.57	5.25			
	AIC	BIC	Loglik	Deviance		
	975.0	990.8	-484.5	969		

1433 observations, 179 participants

Repetitive behaviours

The second part of the analysis examined whether a child's RRB scores predicted their performance on the sorting tasks, especially the LI task. To examine this further we performed a series of Linear Mixed-Effects Models (Baayen, 2008; Jaeger, 2008), in which we modelled RRB scores as the dependent variable. In our first set of analyses we examined overall RRBs scores, whereas in the second and third set we modelled lower-level and higher-level RRB scores respectively. All models were conducted using the lmer function from the lme4 package in R (Bates, et al., 2015). We started with a null model containing random effects of participants and switch dimension on intercepts, and considered the main effects of overall sorting scores, sorting task, and age groups as main effects and interactions terms.

RRBs Overall

To add the fixed effect of sorting score to a null model that only included random effects significantly improved the model fit ($\chi^2(1) = 5.63$, $p = .02$). Adding type of task ($\chi^2(1) = 1.77$, $p = .18$) or age groups ($\chi^2(1) = 1.10$, $p = .29$) did not significantly improve the model fit. Finally, the interaction terms sorting score by task ($\chi^2(2) = 2.07$, $p = .35$), or sorting score by task by age group ($\chi^2(6) = 6.59$, $p = .36$) were not significant.

Despite the nonsignificant task effects, we wanted to further explore our three hypotheses to see if performance on any of the tasks played a crucial role in linking performance with the overall RRB scores. We found that adding the fixed effect of sorting score to a null model that only included random effects did not significantly improve the model fit for the DCCS ($\chi^2(1) = 2.00$, $p = .16$), partial-change ($\chi^2(1) = .02$, $p = .90$), NP ($\chi^2(1) = .28$, $p = .59$) and the perseveration task ($\chi^2(1) = 1.08$, $p = .30$). However, we did find that the model improved significantly for the LI scores ($\chi^2(1) = 5.41$, $p = .02$*, suggesting that this task is mostly responsible for the overall association with RRB levels.

Again, we ran likelihood ratio tests to check if the inclusion of random effects was justified in the final model. Whereas, the inclusion of random effects of participant ($\chi^2(1) = 58.42$, $p = 2.1 \times 10^{-14}$) was justified, there was not a significant difference between models including the full random effect structure and one containing only dimension ($\chi^2(1) = 0$, $p = 1$). This then suggests that only the inclusion of the random effect of participant on intercepts was justified. See Table 4 for final model fit for the LI task.

Table 4.

Summary of general linear mixed effects model of LI sorting scores on overall RRBs, including fixed effects of age groups.

Fixed effects	Estimated Coefficient	SE	Wald confidence intervals 2.50 %	97.50 %	t value	Pr(>|z|
Overall RRBs	.55	.08	.39	.71	6.66	$2.64 \times 10^{-7***}$
LI sorting score	-.03	.01	-.06	-.01	-2.4	.02*
Random effects	Name	Variance	St. dev.			
Participant	(Intercept)	.05	.22			
	AIC	BIC	Loglik	Deviance		
	1579.9	1599.0	1.571.9	866		

870 observations, 29 participants

Lower-level behaviours

Including the fixed effect of sorting score to a null model that only included random effects significantly improved the model fit ($\chi^2(1) = 5.98$, $p = .01$). Adding type of task ($\chi^2(1) = .51$, $p = .47$) or age groups ($\chi^2(1) = 1.03$, $p = .31$) did not significantly improve the model fit. Moreover, the interaction terms sorting score by task ($\chi^2(2)$ =. 52, $p = .77$), sorting score by age ($\chi^2(2) = 5.27$, $p = .07$) were not significant. Finally, we found no three-way interaction between sorting score, task and age group ($\chi^2(6) = 8.14$, $p = .22$).

Higher-level behaviours

We found that the addition of sorting score ($\chi^2(1) = 3.49$, $p = .06$), age groups ($\chi^2(1) = .93$, $p = .34$), type of task ($\chi^2(1) = 1.73$, $p = .19$), or the interaction term sorting score by age ($\chi^2(2) = 5.27$, $p = .07$) did not significantly improve the model fit when compared to a null model that only included random effects. The interaction terms sorting score by task ($\chi^2(3) = 7.86$, $p = .04$), however, significantly improved the model findings, suggesting that one of the tasks might play a crucial role in linking sorting performance with higher-level behaviours. Finally, the interaction term sorting score by task by age group ($\chi^2(6) = 8.14$, $p = .22$) was not significant.

We wanted to further explore the interaction between sorting and type of task to check whether a specific task played a crucial role in the link with higher-level behaviour scores. We found that adding the fixed effect of sorting score to a null model that only included random effects did not significantly improve the model fit for the DCCS ($\chi^2(1) = .76$, $p = .38$), partial-change ($\chi^2(1) = .47$, $p = .49$), perseveration ($\chi^2(1) = .14$, $p = .70$) or the negative priming task ($\chi^2(1) = .14$, $p = .70$). However, the model improved significantly for the LI scores ($\chi^2(1) = 7.90$, $p = .005$**), suggesting that this task is responsible for the overall association with higher-level RRBs.

Finally, likelihood ratio tests suggested that the inclusion of random effects of participant ($\chi^2(1) = 9.73$, $p = .002$) was justified. There was no significant difference between models including the full random effect structure and one containing only dimension ($\chi^2(1) = .11$, $p = .74$), suggesting that only the inclusion of the random effect of participant on intercepts was justified. See Table 5 for final model fit for the LI task.

Table 5.

Summary of general linear mixed effects model of LI sorting scores on higher-level RRBs, including fixed effects of age groups.

Fixed effects	**Estimated Coefficient**	**SE**	**Wald confidence intervals 2.50 %**	**97.50 %**	**t value**	**Pr(>|z|**
Higher-level RRBs	.51	.07	.37	.66	7.01	1.06×10^{-7}***
LI sorting score	-.04	.01	-.06	-.01	-3.09	.004**
Random effects	Name	Varianc e	St. dev.			
Participant	(Intercept)	.03	.16			
	AIC 661.7	BIC 677.6	Loglik -326.8	Devianc e 653.6		

406 observations, 29 participants

4. Discussion

We defer analysis of the findings in Experiment 1 until the General Discussion. The results from this typically developing sample appear to show that the LI task was no different from the version of the DCCS task devised by Zelazo et al., (2003), but it alone predicted the frequency of RRBs.

5. Experiment 2

Experiment 2 was designed to examine if we could identify the same relationship between LI scores and RRBs in children with ASD. To examine this relationship in children with ASD is of importance as RRBs decline in typically developing children around the same time that set shifting skills undergo a rapid development, but set shifting difficulties and behaviours may persist. In addition to examine the relationships in children with ASD, we also included a control group of children with DD. This was done to match children on receptive vocabulary and nonverbal intellectual abilities as well as to examine if our findings can be found in a group of older children whose diagnostic criteria does not include RRBs. Given the relative scarcity of these groups we employed a within-participants design. Finally, in addition to examining the relationship between RRBs and LI scores, we introduced the perseveration task as a control task. Perseverative errors were controlled for, as the idea that children perseverate on a dominant representation is so widespread that it has almost become commonplace in the set shifting literature. The P task was chosen as the control task as by replacing the previously irrelevant dimension with a novel dimension it prevents LI errors and may consequently be the only pure measure of P errors.

5.1 Participants

Sixty-six children with autism (ASD) aged between 52 and 141 months (mean=93.8, SD=22.8) and twenty-four children with developmental delays (DD) aged between 59 and 166 months (mean=101.0, SD=34.9) were recruited from seven special needs schools in Lancashire and Cheshire. Children with ASD had all received a diagnosis by a chartered educational or clinical psychologist, using standardised instruments (i.e. Autism Diagnostic Observation Scale and Autism Diagnostic Interview-Revised; Lord et al., 2002; Lord et al., 1994). The DD children had a diagnosis of Mild Learning Disorder (MLD), and no other known diagnoses. For seven children the parents did not return the questionnaires, and ten children were absent during one of the two testing sessions.

The within-participant analysis is based on the 79 children who completed both tasks (55 ASD and 24 DD), while the full regression analysis (children who had completed both sorting tasks and returned the questionnaire) consisted of 73 children (23 DD and 50 ASD). The two sample were matched on receptive vocabulary as measured by the British Picture Vocabulary Scale (BPVS; Dunn, Dunn, Whetton, & Pintilie 1982) and nonverbal intellectual abilities using the Leiter-R short form (Roid & Miller, 1997), which comprises four sub-tests of visualisation and reasoning. Eleven children did not complete the BPVS and the Leiter-r measures, as they were absent for one of the two testing sessions, or did not manage to sit through the tasks due to inattention. See Table 6 for a more detailed account of the two groups Leiter-r, BPVS and RRB scores.

Table 6.

Standardised test scores and RRB scores.

	ASD:	***DD:***
BPVS standardised score	*75.0 (8.4)*	*76.4 (8.2)*
Leiter-r standardised score	*32.5 (8.0)*	*31.1 (8.5)*
Overall RRBs	*28.7 (13.1)*	*27.3 (16.6)*
Lower-level RRBs	*12.9 (6.7)*	*11.4 (8.5)*
Higher-level RRBs	*14.3 (7.1)*	*14.1 (7.9)*

5.2 Procedure

Experiment 2 employed a within-participant design in which all of the children completed two card-sorting tasks across two sessions that were spaced one week apart. These tasks were the Perseveration and the LI task from experiment 1. Children were randomly allocated to a task set that contained two dimension switches: shape and colour, colour and size or shape and size. The order in which the two tasks and sorting dimensions were presented was counter-balanced. Each set included pictures of 6 different objects (house, train, horse, car, dog, bird), in 6 colours (yellow, black, white, purple, red and blue) and two sizes (small and big).

6. Results

To explore whether the same association between LI performance and RRB scores is found in children with DD and ASD, we performed a series of Linear Mixed-Effects Models where the dependent variable was the overall, lower, and higher-level RRB scores respectively. We started with a null model including participants, dimension and order as random effects. We then considered the effects of sorting score, diagnosis, task order, BPVS scores and Leiter-r scores as main effects and interactions terms.

Overall RRB scores

Adding the fixed effects of perseveration plus LI sorting score ($\chi^2(2) = 8.86$, p = .01*) and the interaction term perseveration by LI sorting score ($\chi^2(3) = 10.05$, p = .02*) improved model fit, suggesting that a child's overall sorting performance predicted their overall RRB scores. To examine this interaction further, we ran models that examined the sorting performance on the two tasks separately. Whereas adding the fixed effects of perseveration score ($\chi^2(1) = 2.14$, $p = .14$) did not improve the model, the addition of the LI score alone ($\chi^2(1) = 8.45$, p = .004**) did.

We then examined whether individual differences predicted additional variability in the LI score. Adding the effect of diagnosis ($\chi^2(1) = .05$, $p = .82$), task order ($\chi^2(1)$ = .1.42, $p = .23$), or the interaction term accuracy by diagnosis ($\chi^2(1) = .07$, $p = .97$) or accuracy by order ($\chi^2(1) = 1.75$, $p = .42$) did not improve the model fit. Furthermore, adding Leiter-r scores ($\chi^2(1)$ = .1.30, $p = .26$) or the BPVS scores ($\chi^2(1) = .73$, $p = .39$)

alone, or the interaction terms accuracy by Leiter-r ($\chi^2(2) = .3.50$, $p = .17$), or accuracy by BPVS ($\chi^2(2) = .13$, $p = .94$), did not improve model fit. Finally, combining accuracy, Leiter-r and the BPVS scores ($\chi^2(2) = 1.77$, $p = .41$), or the interaction term accuracy by BPVS by Leiter-r ($\chi^2(6) = 6.20$, $p = .40$) did not improve model fit.

Likelihood ratio tests with the same fixed effect but varying in random effects of participant, order and dimension suggested that the inclusion of random effects of participant ($\chi^2(1) = 391.59$, $p = <.001$***) was justified, whereas dimension ($\chi^2(1) = 0$, $p = 1$), or order ($\chi^2(1) = 0$, $p = 1$) on intercepts were not. See Table 7 for final model summary.

Table 7.

Summary of linear mixed effects model of the LI performance on the overall RRB score, including fixed effects of accuracy.

Fixed effects	**Estimated Coefficient**	**SE**	**Wald confidence intervals** 2.50 %	97.50 %	**t**	**Pr(>|z|**
(Intercept)	1.13	.07	.99	1.25	16.32	< 2 x 10^{16}***
LI score	-.04	.01	-.07	-.02	-2.97	.003**
Random effects	Name	Variance	St. dev.			
By-participant random intercepts	(Intercept)	.19	.44			
	AIC 5459.8	BIC 5482.6	Loglik -2725.9	Deviance 5451.8		

*2190 observations, 73 participants. ** Significant at 0.001 level, * Significant at 0.01 level*

Lower-level RRB scores

We modelled whether overall sorting performance predicted low-level RRB scores. Adding the fixed effects of perseveration plus LI sorting score ($\chi^2(2) = 8.07$, p = .02*) or the interaction term perseveration by LI sorting score ($\chi^2(3) = 10.46$, p = .02*) to a model with only random effects improved model fit, suggesting that a child's overall sorting performance predicted their lower-level RRB scores. Again, to examine this interaction further, we split the file and ran models that examined the sorting performance on the two tasks separately.

Adding the fixed effect of Perseveration sorting score ($\chi^2(1) = 2.57$, p = .10) did not improve the model score, whereas the LI score ($\chi^2(1) = 7.40$, p = .006**) did. Adding diagnosis ($\chi^2(1) = .15$, p = .70) or task order ($\chi^2(1) = 1.12$, p = .28), however, did not improve the model fit. Moreover, the interaction term LI score by diagnosis ($\chi^2(2) = .68$, p = .71) or LI score by order ($\chi^2(2) = 2.45$, p = .29) were non-significant. Finally, we found no interactions between LI score, diagnosis and order ($\chi^2(6) = 4.50$, p = .61). This suggests that whereas LI scores predicted low-level RRBs, their diagnostic group and the order in which they completed the two tasks did not.

Again, we examined whether individual differences predicted additional variability in the model. Adding the Leiter-r scores ($\chi^2(1) = 1.57$, p = .21) or the BPVS scores ($\chi^2(1) = 0$, p = 1) to a model with LI score did not improve model fit, nor did the interaction terms LI by Leiter-r ($\chi^2(2) = 2.24$, $p = .33$), LI by BPVS ($\chi^2(2) = .10$, $p = .95$), or LI by BPVS by Leiter ($\chi^2(6) = 6.69$, $p = .35$). Likelihood ratio tests suggested that the random effect of participant was justified ($\chi^2(1) = 136.98$, $p = < .0001$***). Given that there was no difference between the full random effects structure and the model with only order ($\chi^2(1) = 0$ $p = 1.$), or dimension ($\chi^2(1) = 0$, $p = 1$), this suggests that the inclusion of participant random effect, but not those of order and dimension, were justified. See Table 8 for final model summary.

Table 8.

Summary of linear mixed effects model of the lower-level RRB score including fixed effects of accuracy

Fixed effects	**Estimated Coefficient**	**SE**	**Wald confidence intervals 2.50 %**	**97.50 %**	**t**	**Pr(>|z|**
(Intercept)	1.15	.08	.99	1.32	14.02	$< 2 \times 10^{-16}$***
LI score	-.05	.02	-.09	-.02	--2.80	.006**
Random effects	Name	Variance	St. dev.			
By-participant random intercepts	(Intercept)	.25	.50			
	AIC	BIC	Loglik	Deviance		
	1073.5	1119.7	-527.7	1055.5		

876 observations, 73 participants. Significant codes : '' 0.01 '**' 0.001 '***' 0.0001*

Higher-level RRB scores

Our final set of analysis concerned whether overall sorting performance predicted higher-level RRB scores. Adding the fixed effects of perseveration plus LI sorting score ($\chi^2(2) = 4.75$, $p = .09$) or the interaction term perseveration by LI sorting score ($\chi^2(3) = 5.13$, $p = .16$) to a model with only random effects improved model fit, suggesting that a child's overall sorting performance did not predict their higher-level scores. As our prediction was that the LI score only would predict RRBs, we also examined sorting performance on the two tasks separately.

Again, adding the fixed effect of the perseveration score ($\chi^2(1) = .98$, $p = .32$), to a model with only random effects did not improve the model fit, whereas the addition of the LI score did ($\chi^2(1) = 4.63$, $p = .03$*). The addition of diagnosis ($\chi^2(1) = .02$, $p = .90$) or order ($\chi^2(1) = 1.70$, $p = .19$) to a model with only random effect did not improve the model. However, whereas the interaction term LI score by diagnosis ($\chi^2(2) = .33$, $p = .85$) was non-significant, accuracy by order $\chi^2(2) = 7.45$, $p = .02$*) improved the model fit. Closer inspection revealed that children performed worse on the LI task if they had completed the perseveration task first (mean= 2.3), than if they had completed the LI task first (mean= 3.2). Finally, we found no three-way interaction between LI score, order and diagnosis $\chi^2(4) = 1.83$, $p = .77$) These findings suggest that whereas accuracy on the LI task predicted higher-level RRB scores, performance was also moderated by the order in which children completed the tasks.

Again, we examined if individual differences predicted additional variability in the behaviour scores. Adding Leiter-r scores ($\chi^2(4) = .01$, $p = .93$) or BPVS scores ($\chi^2(1) = .45$, $p = .50$) to a model with accuracy did not improve model fit, nor did the interaction terms accuracy by order by Leiter-r ($\chi^2(4) = 6.37$, $p = .17$) or accuracy by order by BPVS ($\chi^2(4) = 1.37$, $p = .85$). Finally, the interaction terms accuracy by diagnosis plus BPVS and Leiter-r ($\chi^2(2) = .61$, $p =.74$) or accuracy by diagnosis by BPVS by Leiter-r ($\chi^2(12) = 12.40$, $p = .41$) did not improve the model fit.

Likelihood ratio tests suggested that whereas the inclusion of the random effects of participant was justified ($\chi^2(1) = 176.13$, $p = <.001$***), the random effects of dimension ($\chi^2(3) = 0$, $p = 1$), and order ($\chi^2(3) = 0$, $p = 1$), on intercepts were not. See Table 9 for final model summary.

Table 9.

Summary of linear mixed effects model of the higher-level RRB score, including fixed effects of accuracy, and the interaction term accuracy by order.

Fixed effects	**Estimated Coefficient**	**SE**	**Wald confidence intervals** 2.50 %	97.50 %	**t**	**Pr(>|z|**
(Intercept)	1.19	0.20	.79	1.60	5.82	1.4×10^{-7} ***
Accuracy	-.15	.05	-.24	-.06	-3.11	.003**
Order	-.08	.05	-0.35	-.20	-.54	.59
Accuracy: Order	.08	.03	.02	.14	2.45	.02*
Random effects	Name	Variance	St. dev.			
By-participant random intercepts	(Intercept)	.17	.41			
	AIC	BIC	Loglik	Deviance		
	2531.9	2561.9	-1260.0	2519.9		

1095 observations, 73 participants, 2 Orders. Significance codes: '' 0.01 '**' 0.001 '***' 0.0001*

7. Discussion

Experiment 2 replicated the findings in experiment 1 that only LI performance predicted high levels of RRBs in children with ASD and DD. These findings are of interest as whereas the behaviours are frequent in young typically developing, they are diagnostic criteria in ASD and may persist in other clinical groups.

8. General Discussion

This paper presents two experiments that examine the development of set shifting skills and RRBs in pre-schoolers, children with DD and children with ASD. Experiment 1 replicates the age-related shift between three and four in all of the DCCS task variations. It also offers support for previous studies that found that high RRB levels predict set-shifting performance in typical and atypical populations. However, these findings were only identified in the LI condition, a version that was modelled on Maes' LI task. In addition to the general relationships between RRB levels and LI performance we also found that LI performance predicted lower- and higher-level RRBs in ASD, yet only lower-level RRBs in preschoolers. We discuss the age-related findings first, then attempt to relate our correlational findings to our three study hypotheses.

The age-related changes shown in Experiment 1 offer support to data of Müller et al. (2006) that suggests that the ability to activate previously irrelevant responses plays an important part in the development of set shifting skills. Nonetheless, our findings may be novel as previous research has suggested that children persevere on dimensions in the DCCS (Hanania, 2010), meaning that Zelazo's (2003) NP task may not measure the ability to activate a previously irrelevant response, but instead the ability to switch away from a response. By separating P and LI errors through dimensions this paper provides strong evidence for the need to make a distinction between two abilities: to switch away from dominant responses, and to activate previously irrelevant responses. Our evidence suggests that both address common aspects of the development of set shifting skills. The similarity between the results on the LI task and the other DCCS variations in this paper may also suggest that children's main difficulty with the standard DCCS task is with reactivating previously suppressed responses, not to reconciliate the two rules and learn the appropriate settings to apply them. This possibility needs to be tested further through a within-participants design in typically developing children to control for between subject variance.

The second part of our study examined three hypotheses that concerned the relationship between set shifting performance and high levels of RRBs. The first hypothesis suggested that all of our sorting tasks would predict RRB levels. We based this on the findings that children pass the DCCS around the same age (e.g. Doebel & Zelazo, 2015) at which RRBs decline (e.g. Uljarevic et al., 2017). We did not find any evidence for this prediction, as only one of our conditions predicted the behaviours. The task that predicted the RRB levels was modelled on the WCST, a task that introduces more sorting exemplars than the Standard DCCS and may have consequently required higher levels of flexibility skills. Yet, the fact that we did not find the same associations with the Perseveration condition (the other version that was modelled on the WCST) makes it unlikely that the high flexibility levels alone can account for the relationship with RRB levels. In addition, by establishing a relationship with one of the tasks, we also did replicate Dichter et al's (2010) finding that better DCCS performance would predict higher RRB levels.

Our second prediction was that all of our task versions apart from the LI version would predict RRBs. This prediction was based on the CCC-r (Zelazo et al., 2003) framework that suggests that children may persevere on the pre-switch stimuli due to the inability to inhibit previously relevant rules, as well as redirect attention to previously ignored rules. To persevere on the pre-switch stimuli is possible in all of our tasks apart from the LI task, as this is the only condition in which the pre-switch stimuli is replaced with novel stimuli in the post-switch stage. Our results were quite the contrary however, as only the LI condition associated with RRB levels. The current study therefore does not offer support for the CCC-r framework. Instead, these findings offer partial support for research that has suggested that poor DCCS performance don't predict high RRB levels (Dichter et al., 2010), as we only found that a specific error predicted the behaviours. Moreover, our findings also offer support for the research suggesting that perseveration on a dominant rule is unlikely to account for the full development of the behaviours, or vice versa, as it may be that children learn to pass the DCCS task as they have learned to switch away from previously relevant responses (as measured in four of the sorting tasks), but not the ability to activate previously irrelevant responses. This would also help explain why studies by Evans et al. (1997) and Uljarevic et al. (2017) show that although children engage less in RRBs from around the age of four, the behaviours are still prominent.

This leaves us with the final hypothesis suggesting that only LI errors should predict RRBs. This prediction was based on a combination of previous research that has established strong links between WCST performance and RRBs (e.g. Van Eylen et al., 2015), as well as research that has suggested that WCST-like errors are mainly caused by the inability to activate previously ignored rules (Maes et al., 2004; 2006). The current study found support for this prediction. We did not however, find any support for previous findings that suggest that higher-level RRBs can account for the relationship with EF difficulties in individuals with ASD (Mosconi et al., 2009), as LI errors predicted both lower- and higher-level RRBs in ASD and, indeed, in children with developmental delay but not ASD. The relationship between LI errors and RRBs offers support for the attentional theory, as according to this framework the LI condition may be the only version that assesses biases of attention that resemble the RRBs. Both high levels of RRB and sorting errors may in part be caused by how the 'now-relevant' activity or response may have been suppressed in the pre-switch or in a previous situation. More specifically, in the LI task children are asked to activate a response that was previously exposed as a distractor, and it had been continuously inhibited throughout the pre-switch phase. It may be the case that if an individual inhibits an activity or disregards dimensions of a stimulus over a period of time, this process eventually becomes automatic. An attentional orienting response may only be triggered when there is a mismatch between the current environment and the internal representations of that environment. It may consequently be difficult to override this automatic response, leading to set-shifting errors and repetitive cycles of well-learned behaviours.

Somewhat surprisingly, we did not find any differences in RRB scores ASD and DD groups. We believe that several factors could have contributed to these results. First, it may be that some of the children in our DD group have ASD but have not received a diagnosis yet. This explanation would help explain the similar levels of RRBs in the two populations. Secondly, it may be that some of the DD children have an undetected developmental disorder in which RRBs are common. This is possible as although the behaviours are diagnostic criteria of ASD, they are also commonly found in other diagnoses. The results could therefore be caused by comorbidity. Thirdly, it is possible that our results are caused by the different sample sizes in the two groups as our study included fewer DD children than ASD. Consequently, the DD population is perhaps not representative. This explanation is however unlikely to hold up as the RRB differences between the two groups are highly non-significant so a bigger sample size is unlikely to alter the results. Finally, my results rely on parental report measures and it is possible that parents felt the need to 'please the experimenter' by providing higher scores on the questionnaires than they actually observed in real life. Future work should explore these possibilities further by including a second RRB measure scored by teachers or clinicians.

The paper had some limitations. In the second experiment, ASD and DD children were assessed on only two of the five card sorting tasks that the TD children completed in study one. This makes it difficult to conclude if the three remaining tasks also would have predicted the behaviours in children with ASD and DD. Moreover, the second experiment applied a within-subject design meaning that all of the ASD and DD children were assessed on both tasks. In order to avoid training effects, the sessions were spaced one week apart and the order in which children completed the two tasks was randomised. Although these manipulations are likely to have minimised training effects, we cannot be certain that the different designs did not have an impact on the results in the two experiments. Future studies should examine this further by applying a between- subject design in which ASD and DD children are assessed on one of the five tasks only. Despite these weaknesses, the studies also have numerous strengths. The big samples of TD and ASD children makes the study representative of their respective populations. The brief and engaging picture tasks also led to small drop-out rates, even with the inclusion of children with a wide range of intellectual abilities.

The findings in this paper may have clinical implications as never before has anyone disentangled set shifting processes in this way, and found that LI errors predict restricted and repetitive behaviours. Considering previous findings that WCST errors are mostly a result of LI errors (Maes et al, 2004), our findings may consequently explain the strong link between the WCST and the behaviours. Our findings may also have educational implications as they propose that future studies should train children on the ability to activate previously irrelevant stimuli.

In conclusion, the findings in this study highlight the importance of considering the ability to activate previously ignored responses in preschoolers, but also in children with diagnoses in which the RRBs are diagnostic criteria, such as ASD. Future studies should examine both the circumstances in which activation errors continue to cause problems in children, and even adults (following Maes et al., 2004) and the possibilities for training TD children and individuals with ASD to overcome these errors, as if they play an important role in development of RRBs and set shifting skills. Such training studies, if effective in improving these skills, would have clear clinical implications.

9. **References**

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Statement of Author Contribution

In the Chapter entitled, "Why are there so few set shifting interventions for individuals with autism?",
the authors agree to the following contributions:

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Chapter 4

Why are there so few set shifting interventions for individuals with autism?

The previous chapter replicated Zelazo et al's (2003) and Muller et al's (2006) findings that the abilities to inhibit previously relevant stimuli, as well as the ability to activate previously irrelevant stimuli, play important roles in the development of set shifting skills in young typically developing (TD) children. It also extended these findings to large samples of children with DD and ASD. Furthermore, it provided evidence to suggest that the inability to activate previously irrelevant stimuli predicted high levels of restricted and repetitive behaviours (RRBs) in all of the populations. These findings are novel and may have great implications for early intervention programs. Yet, very few set shifting interventions have been developed for children with ASD, and only one have measured if training has an impact on the RRBs (Varanda & Fernandes, 2012). It is currently unclear why this is. This chapter will outline the current state of set shifting interventions and highlight various factors that may explain why few interventions have been developed for individuals with ASD. This will help make future suggestions for training interventions.

To identify studies that had examined the effect of set shifting training, we searched Scopus and the ISI Search Engines [02.07.2019]. The following combinations of keywords were used: training OR randomised controlled trial AND executive function OR set shifting OR cognitive flexibility AND autism OR ASD OR pre-schoolers OR typically developing. Our initial search produced 140 results. Our inclusion criteria resulted in eight studies that will now be discussed further. Due to time constraints, we did limited time to screen the data, making it possible that some relevant training studies were missed out. See Table 1 for a description of the interventions discussed.

The set shifting studies we identified in this review show that TD and ASD children can be trained on set shifting tasks. See Table 1 for more details. In fact, only two studies in our review did to not find that set shifting performance improved over time. Despite the positive training effects, several studies included insufficient control groups that were either trained on ToM tasks (Kloo & Perner, 2003; Fisher & Happè, 2005; Iversen, 2013) or on tasks that required low EF skills (Karbach & Kray, 2009). In fact, one study did not include a control group at all (Varanda & Fernandes, 2017). The different types of control groups are not ideal as they should be implemented to minimise the changes in all other variables except the one being tested. In addition to this, the study design in ASD and TD training research tends to differ. Whereas some of the TD training studies are brief, most of the ASD designs are intense and consist of as many as 28 sessions (e.g. Kenworthy et al., 2014). The ASD studies are also often intense as a session can last for, for example, 20-25 minutes (Traverso, Viterbori, & Usai, 2015) or as long as 30-40 minutes (e.g. Kenworthy et al., 2014). This can then lead to attrition rates as high as 26% (de Vries, Prins, Schmand & Geurts, 2015) and difficulties with implementing the interventions in everyday life, as the designs are both time and resource consuming. This review will consider these factors in more details to examine if ASD children can benefit from shorter interventions that require less resources.

A recent EF meta-analysis offer support for the set shifting findings in the paragraph above as it shows that EF interventions are effective in TD children (Kassai, Futo, Demetrovics & Takacs, 2019). These results are encouraging as EF skills have been found to predict school success (O'Shaughnessy et al., 2003), and mental and physical health (Moffitt et al., 2011). Unsurprisingly, this has led to the development of a wide variety of set shifting interventions for TD children. Surprisingly, there are considerably less set shifting interventions for children with ASD (Fisher & Happe, 2003). On the one hand, this is unexpected since interventions may have bigger preventive effects for this population, especially considering that a recent EF meta-analysis found a broad executive dysfunction in ASD that was relatively stable across development (Demetriou et al., 2017). On the other hand, researchers may be more apprehensive to invest time and resources in this area considering how previous EF training programs for children with ASD often produce less encouraging findings (e.g. Fisher & Happe, 2003).

There may be several reasons as to why training studies in this population have produced inconsistent findings. Previous TD and ASD interventions are for example, often time and resource consuming, as they require intense teacher or parent training and good student-teacher ratios (Traverso, Viterbori & Usai, 2015). Moreover, they have not included a comparable control group. Whereas one study allocated children to a control group that included low levels of EF training (de Vries, Prins, Schmand & Geurts. 2014), another study did not include a control group at all (Varanda & Fernandes, 2017). This makes it hard to assess the effectiveness of the training. Finally, some ASD interventions report big attrition rates (de Vries, Prins, Schmand & Geurts. 2014), and they do not provide much evidence in terms of long-term training and generalisation effects (Kenworthy et al., 2014). Whereas these factors can have a negative impact on interventions in TD and ASD populations, it is generally easier for TD children to take part in interventions as TD children do not have the same sensory difficulties and social engagement difficulties as their autistic peers, and these factors often lead to high dropout rates during intense interventions (Koenig, Feldman, Siegel, Cohen & Bleiweiss, 2014). The impact that these factors may have on the literature will now be discussed to determine if the inconsistent EF training literature is affected by these factors and if so, can less intense EF interventions that require fewer resources benefit children with ASD.

Many of the effective EF interventions for TD children have an intense nature and may therefore not be appropriate to replicate in samples of individuals with ASD. They can often be intense and involve many lengthy training sessions. Traverso, Viterbori and Usai (2015) for example trained seventy-five pre-schoolers on an EF training program that focused on working memory, inhibitory control and cognitive flexibility skills. They found strong training effects after twelve training sessions that each lasted for approximately 30 minutes. To replicate this design for children with ASD can be problematic, as this type of intervention would require a good attention span. One such intervention was the Unstuck and On Target (UOT) intervention carried out by Kenworthy et al (2014). This program was delivered through 28 sessions that all lasted between 30-40 minutes. Whereas, classroom behaviours, flexibility skills and problem solving skills improved, the training design meant that children had to have a mental age of more than 8 years old to take part in the intervention. This is not ideal for TD children or children with ASD, as this type of intervention would not be able to benefit them during the critical pre-school years when EF skills develop rapidly in TD children (Zelazo et al., 2003).

In addition to time commitment, the development of few EF interventions for children with ASD may also be influenced by how interventions often require both training and "homework" and consequently ask a lot from parents and/or teachers. A good example of this can be seen in a randomised controlled trial of the Tools of the Mind program (Bodrova & Leong, 1996), by Diamond et al. (2007). This is a school-based intervention that integrates opportunities for cognitive and socio-emotional abilities into different classroom activities throughout the day, as well as focuses on a child's ability to pay attention. This study found that pre-schoolers who attended the intervention benefitted significantly more than a control group. This is perhaps not surprising considering how dedicated teachers were to the study. They spent roughly 80% of their day promoting EF skills throughout one or two academic years. This type of commitment may lead to difficulties with recruitment as most mainstream and special schools do not have a good student–teacher ratio, especially considering the support many children in special schools may require to attend these sessions. Previous interventions also often required high levels of engagement from parents. Kenworthy et al's (2014) UoT intervention for example, trained parents and teachers to deliver the intervention. This type of training comes with some benefits, as teachers and parents can keep using the skills after the intervention has finished. Nonetheless, it can also lead to difficulties with recruitment, as the burden of the study may be too high for parents or teachers, resulting in high dropout rates.

There is also little evidence to suggest that interventions benefit children in the long-term. More specifically, some research on older adults suggest that the benefits diminish as soon as the training ends (Willis et al., 2006), less is known about the long-term effect in children. There is also little evidence to suggest that training generalises to everyday life skills, as many interventions take place in labs. Instead, future interventions should take place in classrooms but also in home settings to maximize the potential for generalisation of skills (Dingfelder & Mandell, 2011). If parents or teachers struggle to see the benefits of the intervention, they may consequently be reluctant to implement them, as uncertain situations can often be anxiety provoking for individuals with ASD (Rodgers & Ofield, 2018). The scarce evidence for the long-term benefits can then reduce the feasibility of an intervention, as it may be harder to get funding for the studies, especially if they require high levels of resources. In order to change this, more interventions need to measure the long-term effects of training, or indeed if training can generalise to related skills. These factors are better researched in TD children, as a recent meta-analysis found that training generalised to related skills (e.g. other tasks that measure the same EF skill), yet there was no convincing evidence for far-transfer (e.g. effects on untrained EF skills). Future research should address these issues by measuring if the trained skills generalize to similar skills, as well as unrelated skills.

Finally, interventions can be expensive to carry out. Big projects require big funds as they often require both research assistants and therapists. In addition to this, smaller scale training programs may be expensive for schools or parents as they may require expensive equipment such as, for example, a tablet. These costs may then make some interventions too expensive and thereby inaccessible for some parents. This may have a knock-on effect on the recruitment process.

In order to take these factors into account, shorter interventions that require fewer resources should be developed, so that more schools and/or families can take part. This may be effective as shorter training interventions that have included fewer resources have been found to benefit TD children. Kloo and Perner (2003) for example, carried out an intervention in which an experimenter trained children on set shifting tasks in a school-setting over four brief sessions that each lasted 15 minutes. They found training improvements on the set shifting tasks, and that training improved performance on a related and an unrelated task. There is also evidence to suggest that less comprehensive training studies are effective in children with ASD. Karbach and Kray (2009) for example, carried out a study that found that children with ASD improved on EF skills after an intervention that only involved four training sessions. Despite the positive results, they however, also found that the children who were allocated to the control control improved on the tasks too. Another similar study was carried out by Iversen (2013) and consisted of four brief set shifting training sessions and found that children with ASD improved on the tasks, and that training generalised to a structurally similar task. These two studies highlight that comprehensive intervention studies may not be necessary in this population.

Despite these positive training results, no brief studies have yet examined the long-term effects on this type of training design, or indeed, if training can have an impact on high levels of RRB scores. This is plausible, considering the highly significant relationships we found between set shifting and RRBs in our meta-analysis. To replicate this sort of methodology in children with ASD may help to make interventions more accessible.

In conclusion, we argue that there may be a lack of EF interventions in ASD for several reasons. Although previous interventions have been found to be effective, they are also often time consuming for researchers, teachers and parents, and they may require high levels of resources, as they often need to be conducted under the supervision of adults. They can also be expensive to carry out, and difficult to recruit for, as there is little evidence to suggest that they will be effective for families in the long run. Instead, previous research suggests that an intervention may not need all of these factors, as shorter and less intense interventions have been found to be effective. Future research should develop a training study that is cost and time efficient and requires little resources. This is essential as it may help make EF interventions more accessible to children with ASD, and consequently help reduce the gap between the amount of studies that have been carried out on TD children, and children with ASD. In the next chapter, we will therefore examine the efficacy of an EF intervention for young TD children and children with ASD that fits these criteria and can be easily implemented in schools.

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Table 1: *Details of the studies that were included in the training study review*

Study	Author(s)	Year	N (Training; control)	Age (Training; control)	Control group?	Diagnosis	EF task	Number of sessions	Training effective?	Near/ Far transfer	Attrition rates
1	de Vries, M., Prins, J. M. P., Schmand, B. A., & Geurts, H. M..	2015	37;38	8-12 year olds	Yes	ASD	Gender emotion task	25	< .001	Far and near: <.001	26%
2	Iversen, R.	2013	32	7.0	No	ASD	DCCS	4	Yes < .001	Far transfer	
3	Karbach, J., & Kray, J.	2009	56	9.2	Half of the children allocated to single-task training	TD	Task switching paradigm	4	Yes < .001	Near and far <.001	
4	Kenworthy, L., Anthony, L. G., Naiman, D. Q., Cannon, L., Wills, M. C., Caroline, L.-T., . . . Wallace, G. L.	2014	47;20	9.49	Social skills	ASD	UOT	28 each 30-40 min	Yes < .001 Both groups		
5	Kloo, D., & Perner, J.	2003	47	10.68	ToM	TD	DCCS	4 each 15min	>.20	Far <.001	3 children
6	Fisher, N., & Happé, F.	2005	10;7	10.68; 9.67	ToM	ASD	WCST	5-10 each 25 min	>.001	Far p=.01	
7	Traverso, L., Viterbori, P., & Usai, M.	2015	35;55	5.6;5.6	Yes	TD	Dots task	12 each 20-25 min	Yes < 0.05	NA	15 children
8	Varanda, C., & Fernandes, M.,	2017	10	5.5-13.5	No	ASD	WCST	14-21	Yes <0.05	NA	NA

Statement of Author Contribution

In the Chapter entitled, "Training preschoolers and children with ASD to grasp rule activation errors: does this influence their repetitive behaviours?"

the authors agree to the following contributions:

Rebecca K. Iversen — 80% (Experimental design, data collection, analysis and writing)

Signed: Date: 30/09/2019

Professor Charlie Lewis — 20% (Experimental design, writing, and review)

Signed: Date: 30/09/2019

Chapter 5

Training preschoolers and children with ASD to grasp rule activation errors: does this reduce their repetitive behaviours?

Text as it appears in Iversen, R. & Lewis, C. (2019). "Training preschoolers and children with ASD to grasp rule activation errors: does this reduce their repetitive behaviours?" Manuscript in preparation.

Abstract:

Typically developing (TD) pre-schoolers and children with autism (ASD) fail set shifting tasks because they cannot activate previously irrelevant aspects of the environment. This ability has been linked to the control of repetitive behaviours (RRBs). Employing a card sort procedure assessing learned irrelevance, this study tested whether 10 typically developing (TD) children (m= 3.7 years) and 10 children with Autism (ASD) (m= 6.3 years) could be trained in this ability across four weekly sessions and reduced RRB frequency, assessed at the start and the end of the study and one month later. Training produced clear improvements in understanding learned irrelevance, generalizing to another set shifting skill, with some gains in RRBs. These findings have clear implications for how we understand set shifting in preschoolers and individuals with ASD, and may help remediate RRBs.

Keywords: Set shifting, Restricted and Repetitive Behaviours, Training, Autism, Pre-schoolers

1. Introduction:

Research suggests that executive function (EF) skills are important in many aspects of life. They have, for example, been linked with a better quality of life (Davis et al., 2010), school readiness (Morrison et al., 2010) and success throughout the school years (Gathercole et al., 2004). A recent paper (**blinded*for*review***) proposes that the ability to activate previously irrelevant aspects of the environment (learned irrelevance; LI, Maes, 2004) plays a crucial role in the development of set shifting skills, and the control of restricted and repetitive behaviours (RRBs) in typically developing (TD) children and children with Autism Spectrum Disorder. (ASD). RRBs are diagnostic criteria for ASD together with communication and social interaction (DSM 5, APA, 2013), but also common in TD children where RRBs peak around the age of four (e.g. Tregay, 2009) and decline at about the same time that children learn to pass tasks that measure the ability to activate previously irrelevant aspects of the environment. In individuals with ASD the behaviours persist (e.g. Baron-Cohen, 2000) and set shifting task performance often remains poor (Demetriou, 2017). Despite the developmental overlap in TD children, no studies have yet examined training implications for rule activation in these populations. The aim of the current paper is therefore to examine if TD children and children with ASD can be trained on rule activation tasks, and to examine if training can influence the frequency and nature of a child's reported RRBs.

Thirty years ago it was proposed that executive function (EF) deficits may be the underlying cause of ASD (Russell, 1991). EF skills help maintain appropriate problem-solving strategies in order to reach a future goal (Ozonoff et al., 1991). The face validity of the EF account is strong, as inflexible behaviour is evident in the daily life of individuals with ASD (Geurts, Corbett & Solomin, 2009) and in TD pre-schoolers (e.g. Leekam et al., 2007). Nonetheless, the overall literature on the topic is mixed. Whereas a growing number of cross-sectional studies suggest that the cognitive inflexibility shown in set shifting tasks manifest as RRBs (e.g. Lopez et al, 2005; South et al., 2007), other studies have found no evidence for this (e.g. Ozonoff et al., 2004). Some research has also highlighted the importance of investigating the behaviours through sub-groups of lower- and higher-level RRBs, as they found that set-shifting and RRBs correlated with higher-level behaviours only (Faja & Darling, 2019; Mosconi et al., 2009). Examples of lower-level behaviours are hand flapping and repetitively ordering objects, whereas higher-level RRBs consist of circumscribed interests (CI), such as intense interests in selective topics, and insistence of sameness (IS), such as excessive adherence to routines (Prior & Macmillan, 1973).

Poor set shifting skills and high levels of RRBs have been thought to associate as they both reflect uncontrolled continuation of a response, also commonly referred to as perseverative responding. A majority of these studies have measured these executive skills through the adult-friendly Wisconsin Card Sort task (WCST). This requires multiple shifts of attention, as the participant needs to sort cards from one dimension (e.g. colour) then suddenly change to another (e.g. shape or size) without warning. In children, set shifting skills are commonly measured through the Dimensional Change Card Sort (DCCS), a task in which children have to sort cards after one dimension (e.g. shape) before a switch occurs and he or she must sort after a new dimension (e.g. colour).

Preschoolers tested on this task show dramatic improvement around the age of 4 (e.g. Zelazo et al., 2003), yet, the only study that has examined the relationship between the DCCS and RRBs has produced null results (Dichter et al., 2010). It is not yet known why poor performance on the WCST, but not the DCCS, predicts repetitive behaviours.

The WCST task may predict RRBs due to an inability to activate previously ignored responses, an ability to ignore dominant responses, or both simultaneously. A recent study modifying the DCCS (**blinded*for*review***) suggests that one skill, the ability to activate a previously ignored response, provides a key link with repetitive behaviour. It examined a child's reported RRB scores in terms of their performance on the standard DCCS and four DCCS adaptations that aimed to isolate the ability to shift away from relevant responses from the ability to activate previously ignored responses. In two of the task versions, children had to shift away from previously relevant stimuli by either selecting new exemplars of a previously irrelevant dimension (partial change), or stimuli from a novel dimension (perseveration). In the other two versions, children had to activate previously ignored stimuli by either suppressing exemplars of a previously relevant dimension (negative priming), or stimuli from a novel dimension (learned irrelevance). Preschoolers showed similar improvement in all of the tasks around the age of 4, whereas performance remained poor for children with ASD and developmental delays (DD). However, poor performance on only one task predicted RRB levels and this provided the purest measure of the ability to attend to a previously irrelevant dimension of the array – what are termed 'learned irrelevance' trials (Maes, 2004). These results suggest that children may struggle with selecting previously ignored stimuli that may lead to disrupted control of attention. To date the training implications for this ability have not been examined.

EF interventions may have clinical implications, but as yet these have not been fully specified. A recent meta-analysis by Kassai, Futo, Demetrovics and Takacs (2019) suggests that TD children can be trained on inhibitory control, cognitive flexibility, and working memory skills. The findings in ASD, however, are not as straightforward. A study by Varanda and Fernandes (2017), for example, produced positive findings. They trained 10 individuals with ASD on the WCST across 14-21 training sessions and found that cognitive flexibility improved with time. Yet, their study lacked a control group. Kenworthy et al. (2014) trained children with ASD on 28 problem solving, flexibility and planning skills, and found that they improved significantly compared to the 20 children they allocated to a social skills control group. Other studies have produced null findings. de Vries, Prins, Schmand and Geurts (2014) trained children with ASD on 21 sessions that focused on working memory (WM) or cognitive flexibility and compared to a control group given "mock-training". Trained children improved on WM, cognitive flexibility, attention and parent-rated EFs, but the improvement was not larger than that in the mock-training controls. Noteworthy, however, the tasks completed by the control group did require some WM skills and this could have consequently caused their improvement.

Despite some encouraging findings in TD and ASD children, previous training studies have often been intensive and time consuming - thereby not appropriate for many children with ASD. The same study by de Vries et al., (2014), for example, resulted in a 26 % drop-out, perhaps due to its rigorous nature. Less intensive interventions have been found to be successful in TD children, but they are under-researched in ASD. A brief intervention study on preschoolers that produced positive findings was carried out by Kloo and Perner (2003). They found that children improved on the DCCS after administering only two training sessions. The TD literature thereby suggests that intensive interventions may not be necessary. There is no doubt that short-term training may be beneficial to individuals with ASD, given their particular attentional difficulties (Noterdaeme, Amorosa, Mildenberger, Sitter, & Minow, 2001), yet there is little evidence for these types of interventions, perhaps as a result of the inconsistent findings for the more exhaustive programs.

Given these findings, the current study will implement a brief EF intervention that addresses the effect that training on attending to previously ignored rules within set shifting tasks, and the control of RRBs. This will be examined in preschoolers and individuals with ASD. In addition to measuring a particular EF skill (LI) that has not yet been trained, the current study will address other current issues in the literature. Given that previous training studies are often time consuming and consequently confounded with high attrition rates, the current investigation will measure the effects of 4 training sessions that last around 10 minutes. Some studies lack a control group (e.g. Varanda & Fernandes, 2017), and others include a control group that completes activities that require basic EF skills (de Vries, Prins, Schmand and Geurts, 2014). The current study will therefore include a control group that spends the same amount of time with the experimenter, but receives training with no obvious EF content.

Additionally, little is known about whether training on EF skills generalises to other similar competences. de Vries et al. (2014) found no convincing evidence of transfer to other EF tasks, but Kloo and Perner's (2003) brief intervention found that training on the DCCS or the false belief (FB) task improved performance on the other skill. Moreover, Karbach and Kray (2009) found transfer effects to a structurally similar, but new, switching task, as well as other EF tasks (e.g. working memory and inhibitory control) and fluid intelligence. These effects are not widely examined in ASD, but again there is some evidence for far-transfer effects as Fisher and Happé's study (2005) found that training children with ASD on EF tasks improved FB performance.

Finally, little is known about the long-term benefits of the training in the TD and ASD children. The previous ASD interventions that found evidence for significant training effects did not implement a follow-up session to assess the maintenance of the learned skill (Kenworthy et al., 2014; Karbach & Kray, 2009). The current study will therefore implement a two-month follow-up session in which training performance, and performance on a structurally similar perseveration task, and a more advanced Three-Dimension-Change-Card-Sorting (3DCCS, Deak & Wiseheart, 2015) task will be measured. The current study has three predictions:

1. Based on previous training findings, we predict that both of the training groups (TD and ASD) will improve on the tasks yet, given their more rapid development, the TD group will improve more.

2. Following the findings in a recent meta-analysis (***Blinded**), and the findings discussed above that children learn to master both perseveration and LI tasks around the age of four, we predict that training will generalise to the closely related perseveration task, but not the more advanced 3DCCS task.

3. Finally, given the evidence that activating previously irrelevant aspects of the environment contributes to RRBs, we predict that there will be moderate changes in RRB levels in the EF training group even after only four training sessions. Following recent cross-sectional ASD findings, the changes will only be seen in higher-level RRBs (Mosconi et al., 2009).

2. Method

2.1 Participants

Twenty children with autism (ASD) and twenty typically developing children were recruited from two nurseries and one specialist school in Lancashire. Children with ASD had all received a diagnosis by a qualified educational or clinical psychologist, using standardised instruments (i.e. Autism Diagnostic Observation Scale and Autism Diagnostic Interview-Revised; Lord et al., 1994; Lord et al., 2002). Children with ASD were closely matched on receptive vocabulary as measured by the British Picture Vocabulary Scale (BPVS; Dunn, Dunn, Whetton, & Pintilie 1982). Their non-verbal intellectual abilities were also measured using the Leiter-R short form (Roid & Miller, 1997). See Table 1 for participant characteristics. The Brief Assessment comprises four sub-tests of visualisation and reasoning that, together, provide a reliable measure of the child's IQ. *** University ethics committee approved this study. Parents provided informed consent, and children provided assent prior to participation.

Table 1. *Participant characteristics on age, receptive vocabulary and nonverbal intelligence Mean(SD)*

Participant group:	**ASD:**		**TD:**	
Measure	**Training (n=10)**	**Control (n=10)**	**Training (n=10)**	**Control (n=10)**
Chronological age (mean months)	79.6(17.2)	70.8(12.9)	43.8(3.9)	41.8(2.6)
BPVS: (mean standardised score)	53.9(8.6)	57.1(9.9)		
Leiter-r: (mean standardised score)	76.5(20.5)	82.4(10.8)		

2.2 Procedure

Repetitive behaviours

A 33-item parent-administered Repetitive Behaviour Questionnaire (RBQ; Turner, 1995) was administered to examine the level of restricted and repetitive behaviour displayed in the children. The RBQ consists of two sub-scales that consider routine behaviour, stereotypies, need for sameness, restricted behaviour, self-injurious behaviour and compulsive behaviour. The scoring of this questionnaire followed that of Zandt, Prior and Kyrios (2007) meaning that a child could obtain a score from 0-2 (or 3) on each question, and a total repetitive behaviour score could range from 0-54. Following a factor analysis by Honey et al (2012) an alternative method of scoring was also employed that divided the overall items into two sub-groups, sensory/motor behaviours and insistence of sameness/circumscribed interests. The sensory/motor behaviour sub-group contained 12 items and could obtain a score range of 0-24, whereas the insistence of sameness/circumscribed interest group contained 15 items and it was possible to attain a score range of 0-30.

Training design

This study used a within-participant design in which a child was randomly allocated to a training or control group. All children completed four training or control sessions that were spaced one week apart. Training occurred one week after the pre-test, followed by four weekly training sessions, a post-test phase and finally a two-month follow- up session. Tasks were administered in a counterbalanced order, and each of the training sessions lasted for no more than fifteen minutes. All target and test cards for the sorting tasks were 90mm x 90mm, and the target cards were affixed to two sorting boxes and remained visible during task administration.

Screening sessions

In the pre- and post- testing sessions, a child was tested on one of two versions of a Learned Irrelevance (LI), perseveration, and a Three Dimension Changes Card Sorting task (3DCCS; Narasimham & Deak, 2001). The order of the tasks was counterbalanced.

LI task

The LI pre-, post- and follow-up tasks were modelled on Maes et al's (2011) LI design. In the pre-switch stage, a child was asked to sort cards after one sorting dimension (e.g. colour) and ignoring a distractor (e.g. shape). After eight sorts a crucial post-switch stage was introduced in which the previously irrelevant sorting dimension turned relevant (e.g. shape), and a novel dimension was introduced as the irrelevant dimension (e.g. size). The sorting switches in these tasks measured whether a child's sorting errors were caused by a difficulty with suppressing previous distractors. See Figure 1 for a pictorial representation of one of the LI tasks. The full instructions for both tasks are in the appendix.

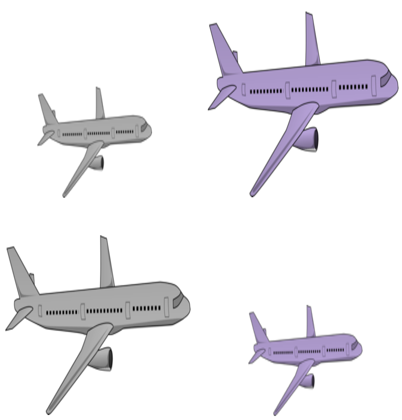
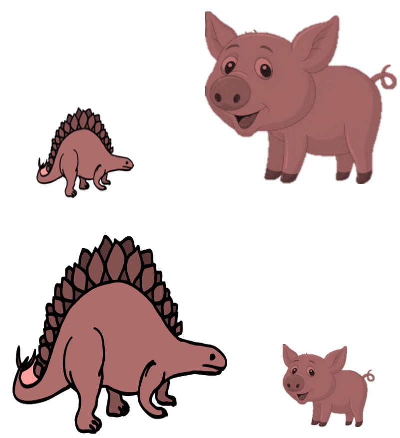
Pre-switch:	Post-switch:
 Relevant=Colour, Irrelevant= Size	 Relevant= Size, Irrelevant= Shape

Figure 1. *LI task example: In this example, the child must sort the big grey planes and the little purple planes by colour then sort the big dinosaurs and the little pigs by Size*

Perseveration task

The Perseveration pre-, post- and follow-up tasks were modelled on Maes et al's (2011) perseveration design. In the pre-switch stage, a child was asked to sort cards after a sorting dimension (e.g. shape). After eight sorts a crucial post-switch stage was introduced in which the previously relevant sorting dimension turned irrelevant (e.g. shape), and a novel dimension was introduced as the relevant dimension (e.g. size). This task measured whether a child’s sorting errors were caused by perseverative responding on a previously relevant dimension (i.e. shape). See Figure 2 for a pictorial representation of the stages of the Perseveration task

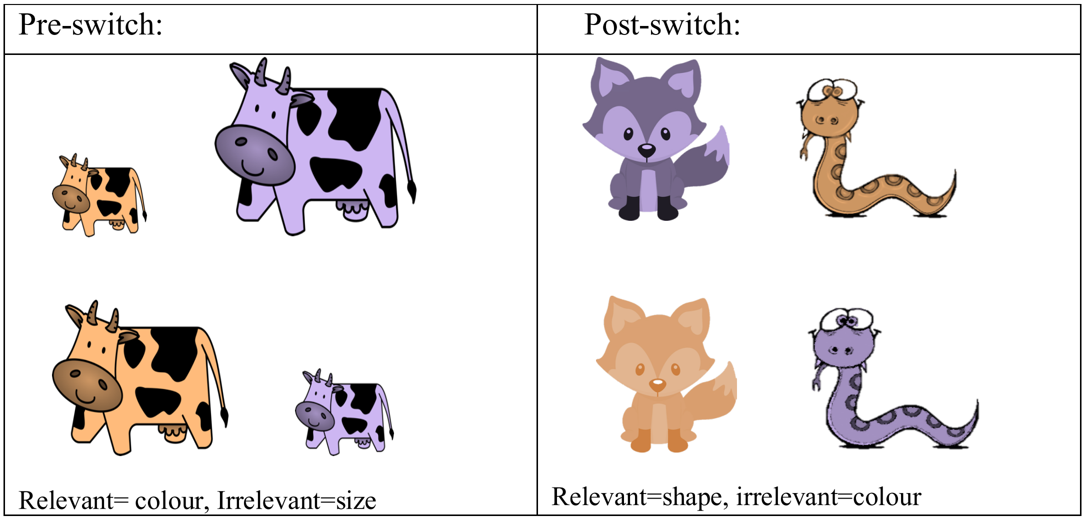


Figure 2. *Perseveration task example: In this example, the child must sort the big orange cow and the little purple cow by colour, then sort the orange cat and the purple snake by shape*

3DCCS

In the pre-test we also introduced the 3DCCS task, a three boxes card-sorting task. This is a modified version of The Dimensional Change Card Sort (DCCS; Frye, Zelazo & Palfai, 1995), and it consists of four sorting boxes, three sorting rules (size, colour and shape) and two rule switches. The sorting stimuli contained four target cards (in which a green frog of intermediate size is a distractor card), and eighteen tests cards that matched the target cards on one dimension each (e.g. a medium red dog or a large yellow fish). The order of the test cards was randomized, but no card occurred more than two times per rule switch, and no two combinations (e.g., medium and yellow) were presented more than once. See Figure 3 for a pictorial representation of the task.

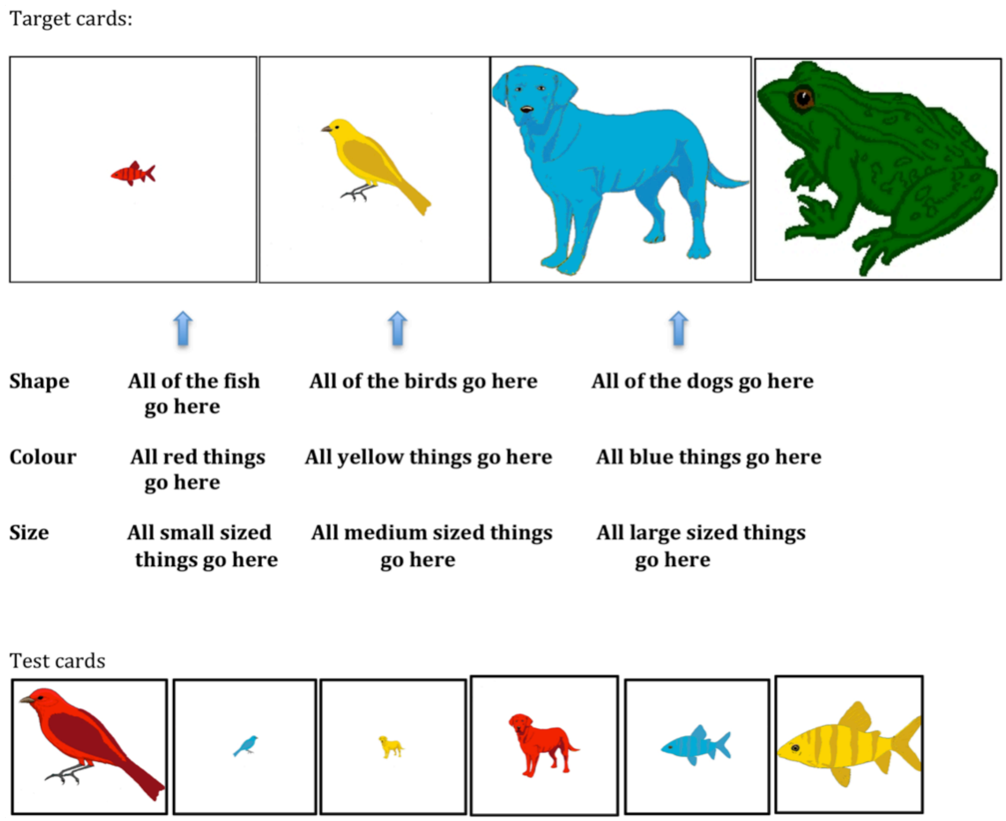


Figure 3. *Exemplars of the 3DCCS stimuli.*

Each test card could be sorted in a different box with a distinct target card, depending on the current rule (e.g. shape, colour or size).

Training group

The training group completed four sets of card sorting tasks that followed the standard Dimensional Change Card Sort procedure (DCCS; see Frye, Zelazo & Palfai, 1995). The only difference was that a child sorted eight instead of six test cards in each phase, and the target cards were replaced between the switching phases in all tasks. The order of the presentation of the four training sessions was counter-balanced, and the four sorting tasks included four switches in which two involved shape, one colour and one size. The four training sessions included cards that displayed pictures of fourteen different shapes (bird, house, train, horse, fish, boat, dinosaur, car, dog, bike, apple, bear, cake, shoe), two sizes (small and big) and eight colours (red, blue, purple, white, yellow, black, orange, grey).

Control group

The control group spent the same amount of contact time with the experimenter as the children in the training group (approximately 10 minutes per session). Instead of being trained on card sorting tasks, a child completed a 30-piece jigsaw, coloured in a picture, read or looked at pictures in a short book, and named dinosaurs or farm animals in a domino game. The order in which children did these tasks was counterbalanced.

Follow up sessions

Two follow-up sessions took place, one a week after the fourth training session and the second approximately two-months after that post-test session had taken place. Each session took between fifteen to twenty minutes and it consisted of a LI task, a perseveration task and a 3DCCS task. Again, the order of the tasks was counterbalanced and all of the tasks had novel testing exemplars.

3. Results

Scoring

In the perseveration and LI tasks, children received an overall score between 0-8, whereas in the 3DCCS task children could score between 0-12; depending on how many cards they sorted correctly after the two dimension switches. In order to be included in the study children had to sort 7 out of 8 sorts correctly in the pre-switch on the perseveration, LI and 3DCCS tasks. The inclusion criteria meant that three ASD children and two TD children were excluded from the study. Normality was assessed through the explore function, where the data distribution appeared normal.

Pre-test analysis

Normality was assessed through the explore function, where the data distribution appeared normal. Preliminary checks using ANOVAs explored if the initial performance of the groups were comparable. The results showed no chronological age differences between the two TD (*F(2,20)=1.74, p= .20),* or ASD groups *(F(1,19)=.1.74, p= .13)* and no differences between the ASD groups on the BPVS (*F(1, 18)= .35, p= .56),* or the Leiter-r (*F (1, 15)= .48, p= .48).* Furthermore, no baseline differences were found between the ASD groups on the LI (*F (1,19)=. 04, p= .85)*, Perseveration (*F (1, 19)= .01, p=. 95),* or the 3DCCS task *(F (1,19)=. 30, p= .59)*. Finally, no differences were found between the TD groups on the LI, (*F (1,19)=. 08, p= .78)*, Perseveration (*F (1,19)=. 09, p= .77),* or the 3DCCS task *(F (1, 19)=. 04, p= .84).* See Table 2 for the pre-test scores.

Table 2. *Pre-test scores Mean (S.D)*

Participant group:	ASD:		TD:	
	Training (n=10)	Control (n=10)	Training (n=10)	Control (n=10)
Perseveration pre-score	2.9(3.4)	2.8(3.1)	1.8 (2.3)	2.2(3.5)
LI pre-score	3.0(3.2)	3.3(3.6)	4.1(2.9)	4.5(3.4)
3DCCS pre-score	4.5(.99)	3.8(.87)	3.5 (1.05)	3.2(3.3)
Overall RRB score	32.3 (15.1)	26.2(15.5)	13.6(14.8)	8.8(6.3)
Lower-level	12.7(7.7)	11.4(7.2)	5.8(7.1)	2.6(2.4)
Higher-level	15.8(6.7)	11.9(7.5)	6.3(6.5)	5.0(3.6)

Training effects

We performed a series of Generalized Linear Mixed-Effects Models (Baayen, 2008; Jaeger, 2008). Each was conducted using the glmer function from the lme4 package in R (Bates, Maechler, Bolker, & Walker, 2015). We started with a null model containing random effects of participants on intercepts. For our first analysis, we considered if training on the LI task improved performance on the task. We modelled the pre-test scores as the dependent variable and considered the effects of the four training sessions, post-test and two-month follow-up scores, as well as diagnosis (TD and ASD) as main effects and interaction terms. We were also interested in whether individual differences predict additional training variability in the ASD group, so we considered the effects of the BPVS and Leiter-r scores on the performance. For our second analysis we modelled the pre-test scores as our dependent variable and the post-test and two-month follow-up scores as main effects and interaction terms. Here, we first modeled the overall sorting performance on the LI task, followed by the Perseveration and the 3DCCS performance. We considered the effects of group (training and control) and diagnosis (TD and ASD) on the sorting scores. For our final analysis we modelled the RRB pre-test scores as the dependent variable and the RRB post-test scores, group and diagnosis as main effects and interaction terms. See Table 9 for the mean scores on the training tasks and RRB questionnaire over time.

In order to address our hypothesis that children can be trained on the LI tasks, we added the fixed effects of post-test plus follow-up scores to a model with only random effects and found that it significantly improved the model fit ($\chi^2(2) = 30.5$, $p = 2.4 \times 10^{-7}$***). We inspected the model in more depth and found that the LI follow-up scores made the biggest difference (estimate= 3.49, SE= .72, 4.84, p= 1.27×10^{-6}*). Whereas adding diagnosis as a fixed effect ($\chi^2(1) = .54$, $p = .46$) or an interaction term ($\chi^2(2) = 1.82$, $p = .40$) did not improve model fit, adding testing group as a fixed effect ($\chi^2(1) = 7.17$, $p = .007$**) or an interaction term ($\chi^2(2) = 27.57$, $p = 1.03 \times 10^{-6}$***) did. More specifically, the training groups improved more than the control groups (estimate = 6.90, SE = 1.60, z = 4.29, $p = 1.77 \times 10^{-5}$ ***). Finally, we found no improvements when we added diagnosis to the interaction term LI scores by group ($\chi^2(1) = .27$, $p = .60$), and we found no three-way interaction between scores, diagnosis and group ($\chi^2(4) = 5.99$, $p = .20$). See Table 3 for full model fit.

Since we found strong differences in results between the trained and untrained groups we wanted to explore the changes in the trained group in more detail. We therefore examined the effect of each of the training sessions on the post and follow-up performance in this group**.** We found that adding the fixed effects of the training sessions, post-test and follow-up scores to a model with only random effects of participant significantly improved the model fit ($\chi^2(6) = 41.78$, $p = 2.03 \times 10^{-7***}$). The training groups improved the most at training one (estimate = 4.01, SE = .65, z = 5.00, $p = 9.8 \times 10^{-8}$***) and training three (estimate = .57, SE = .74, z = 3.16, $p = .03$*). Adding diagnosis as a fixed effect ($\chi^2(1) = 0$, $p = 1$), or an interaction term on the other hand ($\chi^2(2) = 4.15$, $p = .11$) did not improve the model fit. See Figure 4 for a representation of the LI performance for all of the groups, and Table 4 for full model fit.

Table 3. *The effect of training on LI post and follow-up performance*

Fixed effects	**Estimated Coefficient**	**SE**	**Wald confidence intervals** 2.50 %	**intervals** 97.50 %	**z**	**Pr(>|z|**
(Intercept)	1.51	2.34	- 3.07	6.09	.65	.51
LI post-score	.88	.81	-.71	2.47	1.08	.28
LI follow-up score	-7.11	2.44	-11.89	-2.32	-2.91	.003**
Group	-2 .49	1.34	.5.11	0.13	-1.86	.06
LI follow-up score x Group	6.85	1.60	-3.70	9.99	4.27	1.97×10^{-5} ***
Random effects	Name	Variance	St. dev.			
By-participant random intercepts	(Intercept)	3.63	1.91			
	AIC	BIC	Loglik	Deviance		
	241.4	264.0	-114.7	229.4		

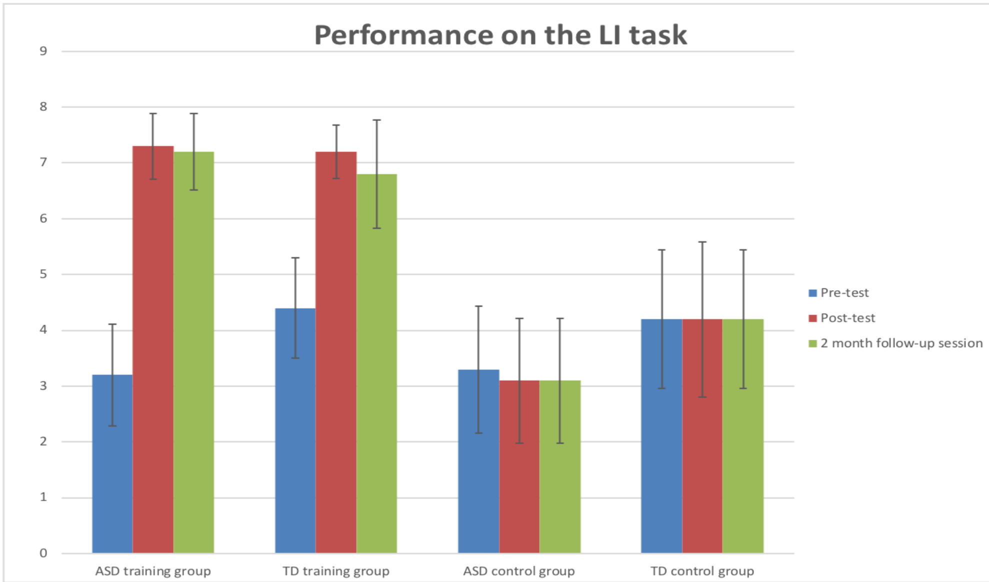


Figure 4. *Learned irrelevance performance in the post switch trials for all groups across time (max score = 8)*

Table 4. *The effect of each training session on the LI post and follow-up performance*

Fixed effects	**Estimated Coefficient**	**SE**	**Wald confidence intervals**		**z**	**Pr(>|z|**
			2.50 %	**97.50 %**		
(Intercept)	**-2.95**	**1.58**	**- 6.04**	**-7.31**	**-.27**	**.03***
Training 1	4.01	.65	2.73	2.31	5.00	9.8×10^{-8}***
Training 2	.55	.64	-.71	-1.62	.88	.57
Training 3	.57	.74	-.88	.18	3.16	.03*
Training 4	-.02	.97	-1.91	-1.21	3.45	.35
Post-test	-.81	1.49	-3.73	-3.73	2.37	.66
Two month follow-up	.69	1.32	-1.94	-2.39	3.19	.78
Random effects	Name	Variance	St. dev.			
By-participant random intercepts	(Intercept)	2.99	1.73			
	AIC	BIC	Loglik	Deviance		
	137.5	162.1	-60.8	121.5		

Number of observations: 320, Participants: 40

The effect of training on Perseveration performance

Following Fisher and Happé's (2005) findings that DCCS training generalized to non-trained tasks, we also wanted to examine if the training effects improved performance on tasks of similar nature. We found that adding the fixed effects of post- plus follow-up perseveration scores (χ 2(2) = .26, p = .61) to a model with only random effects did not improve the model. Moreover, adding diagnosis (χ 2(1) = 1.97, p = .16) or testing group alone ($\chi^2(1) = 0$, $p = 1$), or the interaction term scores by diagnosis ($\chi^2(2) = 2.43$, p = .30) did not improve model fit. When we added group as an interaction term however, the model fit improved ($\chi^2(1) = 27.54$, $p = 1.54 \times 10^{-7***}$), and after inspecting the model more closely it became clear that the trained group improved the most from pre-test to the two-month follow-up (estimate =-4.4, SE = 3.4, z = -1.3, p = 0.19) (see Figure 5). Finally, there was no three-way interaction between post score, diagnosis and group ($\chi^2(4)$ = 5.55, p = .23). See Figure 5 for the perseveration task performance across time.

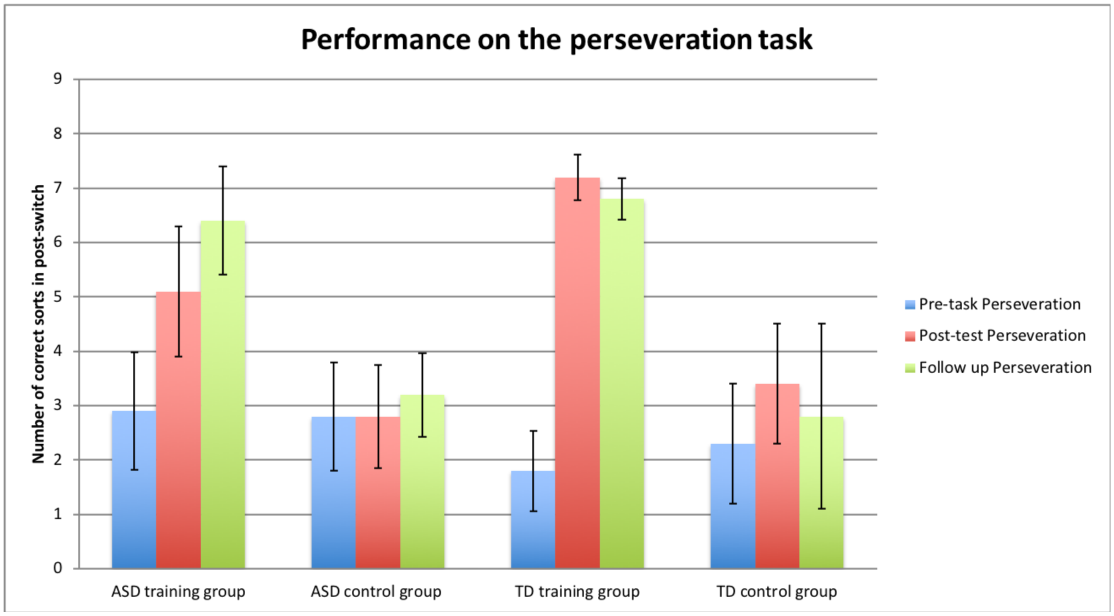


Figure 5. *Perseveration task performance for the training and control groups across time (max score = 8)*

The effect of training on performance on the 3DCCS task

In line with the previous analysis, we also wanted to examine if the training effects generalised to a task in which a reversal shift strategy could not take place. We found no improvements when we added the fixed effects of 3DCCS post-test plus follow-up score ($\chi^2(2) = 2.24$, $p = .33$) to a model with only random effects. We also found no improvement when adding group as a main effect ($\chi^2(3) = 2.31$, $p = .51$) or an interaction term ($\chi^2(4) = 3.57$, $p = .47$). Moreover, whereas adding diagnosis as a main effect did not improve model fit ($\chi^2(3) = 4.21$, $p = .24$), entering diagnosis as an interaction term did ($\chi^2(4) = 12.54$, $p = .01$*). Closer inspection revealed that the TD training group improved more on the 3DCCS than the ASD children (estimate =-1.48, SE = 0.53, z = -2.78, p = 0.006), suggesting that training generalized in the TD training sample. Finally, we found no three-way interaction between post score, diagnosis and group ($\chi^2(4) = 1.01$, $p = .90$). See Figure 6 for a pictorial representation of the change, and Table 5 for final model fit.

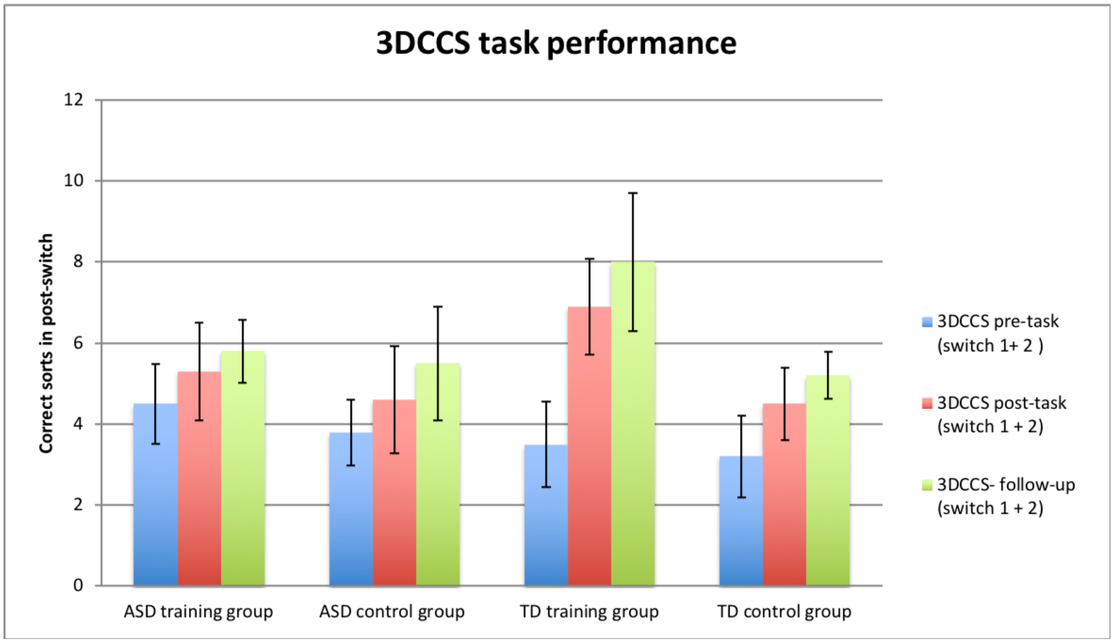


Figure 6. *3DCCS task performance (max score = 12)*

Table 5: *The effect of training on the 3DCCS post and follow-up performance*

Fixed effects	Estimated Coefficient	SE	Wald confidence intervals 2.50 %	97.50 %	z	Pr(>|z|
(Intercept)	.82	.99	-1.14	2.78	.82	.41
3DCCS post-score	.99	2.37	-3.66	5.64	.42	.68
3DCCS follow-up score	-2.81	2.53	-7.79	2.16	-1.11	.27
Diagnosis	-1 .47	.43	-2.52	-.43	-1.11	.006
Group	-.01	.43	-.85	.83	-.03	.98
3DCCS follow-up score x Diagnosis	-1.48	.53	.50	2.50	-2.77	.004**
Random effects	Name	Variance	St. dev.			
By-participant random intercepts	(Intercept)	1.27	1.13			
	AIC	BIC	Loglik	Deviance		
	546.1	575.3	-266.0	532.1		

Number of observations: 480, Participants: 40

Repetitive behaviour analyses

RRBs overall

In order to address our third hypothesis that training on the LI tasks may have an impact on a child's RRB scores, we examined the effect on the overall score first, followed by lower-level and higher level RRB scores. Adding the fixed effects of the overall RRB post scores to a model with only random effects significantly improved the model fit ($\chi^2(1) = 99.84$, $p = 2.2 \times 10^{-16***}$), where the post-scores were higher than the pre-scores (estimate= 3×10^{-1}, SE= 2.8×10^{-2}, t= 10.6). This may be expected as this analysis included training and control groups. Nonetheless we also found that adding group as a main effect ($\chi^2(1) = 1.98$, $p = .16$) or an interaction term ($\chi^2(2) = 3.50$, $p = .17$) did not improve the model fit. Interestingly, adding the main effect of diagnosis ($\chi^2(1) = 13.7$, $p = .0002$***) and the interaction term did ($\chi^2(2) = 14.39$, $p = .0007$***). We examined the model in more depth and found that the scores decreased more with time in the TD group (estimate= -.42, SE= .11, t= -3.88), something which could be a result of how the TD group showed stronger training effects. Finally, adding overall RRB scores, diagnosis and group as main effects ($\chi^2(2) = 2.79$, $p = .09$) or as interaction terms ($\chi^2(5) = 7.71$, $p = .17$) did not improve the model fit. See Table 6 for final model fit and Figure 7 for a pictorial representation of the change across time.

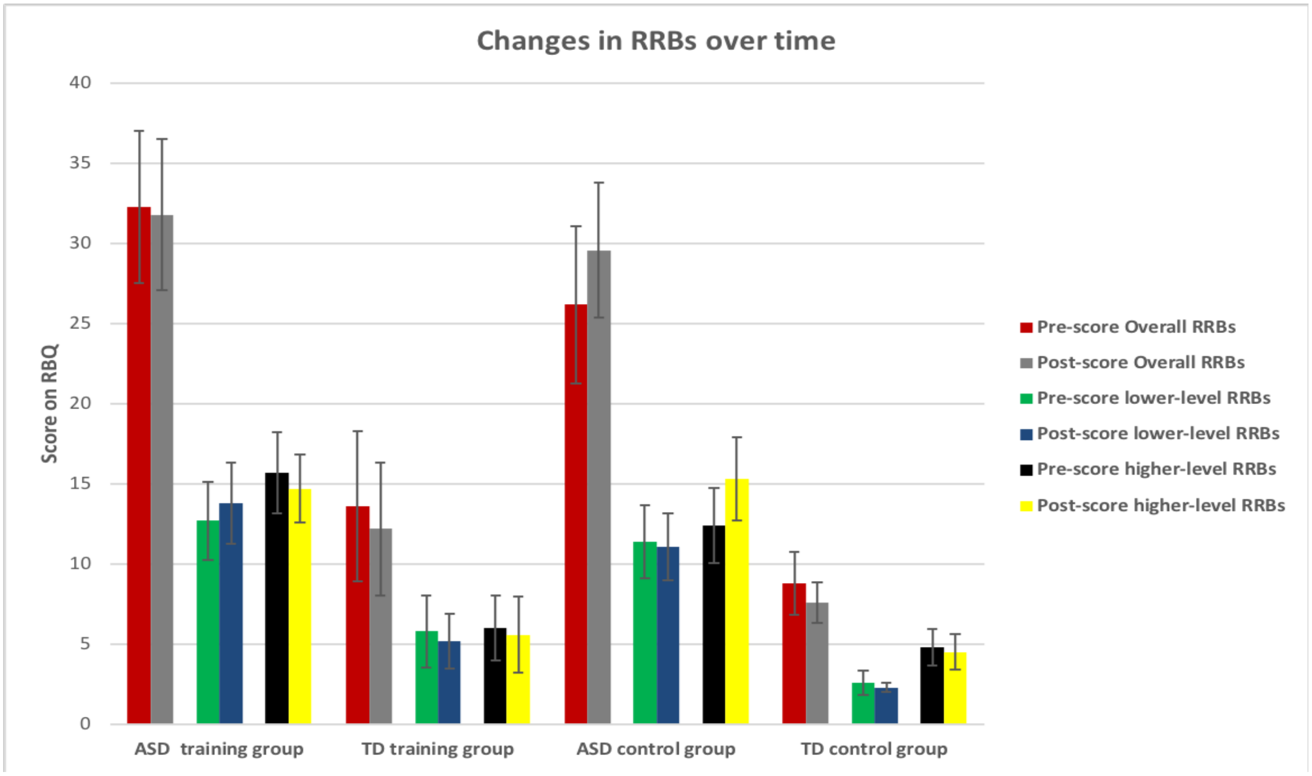


Figure 7. *Changes in RRB scores at pre-test, post-test and two-month follow-up*

Table 6. *The effect of training on the post-test overall RRB scores*

Fixed effects	Estimated Coefficient	SE	Wald confidence intervals 2.50 %	97.50 %	z	Pr(>|z|
(Intercept)	1.10	.18	.75	1.44	6.25	1.8×10^{-7}***
RRB post-score	.29	.03	.24	.35	10.24	$< 2 \times 10^{-16}$ ***
Diagnosis	-.42	.11	-.63	-.21	-3.88	4×10^{-4} ***
Random effects	Name	Variance	St. dev.			
By-participant random intercepts	(Intercept)	.09	.31			

Number of observations: 480, Participants: 40

Lower-level RRBs

Next, we examined whether training had an impact on the lower-level RRBs and found that adding the fixed effects of the sub-type of RRBs to a model with only random effects significantly improved the model fit ($\chi^2(1) = 6.46$, $p = .011$*). In this analysis the RRB scores were slightly lower at the post-test than the pre-test (estimate = .14, SE = .05, $t = 2.75$). Adding group as a main effect ($\chi^2(1) = .83$, $p = .36$) or an interaction term ($\chi^2(1) = 2.32$, $p = .31$) did not improve the model fit ($\chi^2(2) = 1.04$, $p = .59$). Yet, adding diagnosis as a main effect ($\chi^2(2) = 12.49$, $p = .0004$**) or an interaction term did ($\chi^2(2) = 12.39$, $p = .002$**). To examine this interaction further we investigated the lower-level RRBs scores for the ASD and TD groups separately. For the ASD group, the post-scores were higher than the pre-scores (estimate = .15, SE = .07, $t = 2.09$), whereas for the TD group the post-score were lower than the pre-scores (estimate = .07, SE = .07, $t = .93$). Finally, adding lower-level score, diagnosis and group as main effects ($\chi^2(1) = 1.14$, $p = .28$) or interaction terms ($\chi^2(5) = 7.36$, $p = .08$) did not improve the model fit. See Table 7 for final model fit, and Figure 7 for the changes in lower-level RRBs across time.

Table 7. *The effect of training on the post-test lower-level RRB scores*

Fixed effects	Estimated Coefficient	SE	Wald confidence intervals 2.50 %	97.50 %	z	Pr(>|z|
(Intercept)	1.46	.26	.95	1.96	5.66	1.3 x 10^{-6}***
Lower-RRB post-score	.12	.05	.02	.23	2.38	.02*
Diagnosis	-.58	.16	-.88	-.27	-3.66	.0008 ***
Random effects	Name	Variance	St. dev.			
By-participant random intercepts	(Intercept)	.20	.44			

Number of observations: 480, Participants: 4

Higher-level RRBs

Finally, we wanted to examine if training had an impact on the higher-level RRB scores. Adding the fixed effects of higher-level post scores to a model with only random effects significantly improved the model fit ($\chi^2(1) = 13.89$, $p = .0002$***), with post-scores being higher than the pre-scores (estimate = .15, SE = .04, $t = 3.94$). Moreover, adding group as a main effect ($\chi^2(2) = 3.44$, $p = .18$), or interaction term ($\chi^2(1) = 1.49$, $p = .22$) did not improve model fit. Yet, adding diagnosis as a main effect ($\chi^2(1) = 12.61$, $p = .0004$***) or as an interaction term did ($\chi^2(2) = 14.95$, $p = .0006$***). To examine this interaction further we investigated the higher-level RRBs scores for the ASD and TD groups separately. For the ASD group, the post-scores were higher than the pre-scores (estimate = .09, SE = .05, $t = 1.77$), whereas for the TD group the post-score were slightly lower than the pre-scores (estimate = .22, SE = .06, $t = 4.00$). Finally, adding higher-level RRB score, diagnosis and group as main effects ($\chi^2(1) = 2.03$, $p = .15$) or interaction terms ($\chi^2(2) = 2.61$, $p = .27$) did not improve the model fit. See Table 8 for final model fit, and Figure 7 for changes in the higher-level scores across time.

Table 8. *The effect of training on the post-score higher-level RRB scores*

Fixed effects	**Estimated Coefficient**	**SE**	**Wald confidence intervals** 2.50 %	97.5 %	**z**	**Pr(>|z|**
(Intercept)	**1.34**	**.21**	**.92**	**1.**	**6.3**	**5.1 x 10^{-8}*****
Higher-RRB post-score	-.03	.12	-.26	.19	-.30	.77
Diagnosis	-.52	.13	-.77	-.26	-3.97	.0002 ***
Post higher x Diagnosis	.13	.08	-.04	.29	1.53	.13
Random effects	Name	Variance	St. dev.			
By-participant random intercepts	(Intercept)	.11	.34			

Number of observations: 480, Participants: 40

4. **Discussion**

In line with the findings of a recent meta-analysis on EF intervention in TD children (Kassai, Futo, Demetrovics & Takacs, 2019), and in support of our first hypothesis, we found that TD children and children with ASD could be trained on the LI tasks after only four weekly training sessions each lasting around 10 minutes. Moreover, we found that the children in the control group did not improve at all on the tasks, and that the training effects were still present at the follow-up session two months after the training had commenced. In support of our second hypothesis we found that training on the LI task generalized to performance on the P task. Moreover, training also improved performance on the more complex 3DCCS task, but only significantly in the TD group. Finally, we found no evidence to suggest that our short-term training program had an impact on a child's RRB levels.

That children could be trained on the sorting task is noteworthy for three reasons. First it offers support for the findings in previous training studies on TD children by Kloo and Perner (2003) and Röthlisberger Neuenschwander, Cimeli, Michel and Roebers (2012). Although more studies show positive training effects (e.g. de Vries et al., 2014; Varanda & Fernandes, 2015) than not (Fisher and Happé, 2005), our findings provide evidence for the effectiveness of a brief training programme. One interpretation of previous findings is that Fisher and Happé's (2005) intervention was shorter (5-10 training sessions) than that of de Vries et al (21 sessions) but our study was much briefer and showed longer term effects.

Secondly, this study involved a control group who did not improve on the tasks. This control group received equal amounts of attention but no EF training. This is a strength as a previous study by de Vries et al. (2014) included control tasks that required a moderate amount of EF skills. This may have obscured the training effects as the design led to improvements in the control group. Our findings offer further support for this as we saw no improvements in the children who were simply spending time with a researcher (see Figure 4).

Thirdly, we also found that the training effects were present one month after the training had ceased. As identified in the introduction, insufficient attention has been paid to the need to investigate training effects beyond the immediate testing period. These findings may have clinical implications, as not only do our results suggest that children can be trained on the EF task, they also suggest that training on LI tasks may be beneficial in the long-term as the training effects were present after a month. Future research should however, examine longer-term effects.

In addition to the positive training effects, we also found that training significantly improved performance on the closely related perseveration task. Moreover, training on the 3DCCS only generalized in the TD training sample. These findings offer support to previous research that has shown that training on a switch task improved performance on a structurally similar task (e.g. Kloo & Perner; Karbach & Kray, 2009).These findings are of interest as they offer support to a recent meta-analysis by Kassai, Futo, Demetrovics and Takacs (2019) that found that pre-schoolers could be trained on structurally similar task, yet they found no convincing evidence of transfer to other EF tasks. These findings should be examined further in future studies as if they generalise in studies with bigger samples, it will demonstrate clear training benefit with educational and possible clinical implications.

Despite the fact that the LI task and the 3DCCS task are structurally similar, the finding that training did not generalize to the more complex task may be because the

3DCCS perhaps needs, for example, higher flexibility skills. Alternatively, it was a brief study so it is possible that training influences need more time for accommodation /incubation.

Finally, we did not find evidence for our hypothesis suggesting that activating previously irrelevant aspects of the environment contributes to RRBs. There may be several reasons for this. First of all, the intervention may not have made an impact on RRB levels due to the fact that it took place in a lab setting, instead of being implemented into activities at home or in the classroom. This is possible as Dingfelder and Mandell (2011) suggest that implementing interventions into a child's everyday activities maximizes the potential for generalization of skills. Alternatively, the intervention was brief, so although children improved on the LI task, four training sessions is perhaps not enough for training to have an impact on the persistent RRBs.

This study has some limitations, including relatively small sample sizes, which prevents meaningful investigation of moderator variables such as age. Moreover, the experimenter completed the pre- and post-test assessments as well as the training and was consequently not blind to group allocation. Future investigations should include larger samples and train teachers to complete the training as it would allow the researcher to be blind to group allocation at the pre- and post-testing sessions.

In conclusion, this study suggests that TD preschoolers as well as children with ASD can be trained on LI tasks, and that the training effects persist over time. We also found evidence for generalization to a structurally similar P task, and the more complex 3DCCS task in the TD group. Future research should establish interventions in which training on activating previously irrelevant responses should be incorporated into activities in the classroom.

5. References

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Table 9: Training tasks performance and RRB scores over time

	Training:		**Control:**	
	ASD:	**TD:**	**ASD:**	**TD:**
Pre: Perseveration:	2.9(3.4)	1.8 (2.3)	2.8(3.1)	2.3(3.5)
Pre: 3DCCS (switch 1+2)	4.5(.99)	3.5 (1.05)	4.1(.87)	3.2(1.0)
Pre: RRBs overall	32.3(15.0)	23.6(15.4)	26.2(15.5)	6.7(4.5)
Pre: Sensory/motor	14.3(7.6)	12.2(9.3)	10.8(7.0)	1.7(1.1)
Pre: IS/CI	15.7(7.2)	9.8(8.0)	12.4(7.8)	3.7(3.3)
Tr 1:	3.2(0.9)	4.4(0.9.)		
Tr. 2:	4.5(0.9)	4.3(1.0)		
Tr. 3:	3.6(1.2)	5.5(1.09)		
Tr. 4:	6.4(0.8)	7.6(0.2)		
Post: Perseveration:	5.1(1.2)	7.2(1.3)	2.8(3.0)	3.4(3.5)
Post: 3DCCS (switch 1+2):	5.3(1.2)	6.9(1.19)	4.6(1.3)	4.5.(0.8)
Follow-up: Perseveration:	6.3(3.1)	6.8(0.8)	3.9(2.4)	2.8(3.8)
Follow-up: NP:	7.2(0.7)	6.8(0.97)	4.5(3.4)	5.6(1.67)
Follow-up: 3DCCS (switch 1+2):	5.8(.77)	7.4(1.63)	5.5(1.4)	5.2(.58)
Follow-up: RRBs overall:	31.0(15.2)	13.2(15.1)	28.4(15.5)	7.6(6.1)
Follow-up: Sensory/motor:	13.4(9.2)	4.2(3.5)	11.0(6.04)	1.3(0.5)
Follow-up: IS/CI:	15.6(7.4)	7.7(11.02)	14.2(10.8)	5.6(6.0)

Instructions for the 3DCCS task

‘We are going to play the animal game. Let me tell you how to play the animal game. In the animal game, all dogs go in here, all fish go in here, and all birds go in here [pointing]. So, do you see this picture of a dog here? That’s to remind you that all dogs go in here. And do you see this picture of a fish . . . [etc.]? So, all dogs go in here, all fish go in here, and all birds go in here. Are you ready to play the animal game?’

Switch instruction:

Are you ready to play a new game? We’re going to play the colour game. Let me tell you how to play the colour game. In the colour game, all blue things go in here, all red things go in here, and . . . [pointing]. So, do you see this blue thing here? That’s to remind you that all blue things go in here. Do you see this red thing . . . [etc.]? So, all blue things go in here, all red things go in here and all yellow things go in here. Are you ready to play the colour game?

Chapter 6

General discussion

In this thesis, I have explored the relationships between EF skills and high levels of RRBs, through three meta-analyses, two cross-sectional studies, and a training study. Previous literature that has examined the association between RRB levels and poor EF skills have produced inconsistent results, leading to the suggestion that we must move away from the EF theory as a contender to explain the behaviours (e.g. Leekam et al., 2011). Instead, much of the focus has now shifted to the neurobiological (Lewis & Kim, 2009) and the developmental (Leekam, 2011) accounts. Yet in recent years, a new spurt of research on set shifting studies has yielded positive results, suggesting that we might need to re-open the EF account (e.g. South et al, 2007). Despite the new collection of positive studies, none of these have yet identified what it is with the set shifting tasks that predicts the behaviours, or indeed why RRBs consistently predicts poor performance on the complex WCST, but not the child-friendly DCCS task. Traditionally, perseverative responding is considered to be the cause of set shifting errors, and it is thought to have parallels with RRBs, as a child who struggles with switching away from a dominant response may also be more likely to select a particular activity at the exclusion of all others. Recent research however, has found evidence to suggest that children and adults fail set shifting adaptations due to an inability to activate previously ignored responses. Yet, no one has isolated different set shifting responses and examined their relationships with RRBs. Moreover, few interventions have examined if EF training can have an impact on RRBs, and the few who have did not include a control group or long-term measures (e.g. Varanda & Fernandes, 2017).

The aims of this thesis were thus, to first of all to examine the strength of the relationship between EF skills and RRBs through a meta-analysis. Secondly, to explore the relationships between RRBs and set shifting errors in more depth through various set shifting adaptations. Finally, to examine if a brief training intervention could improve shifting performance in a normative population as well as in samples of children with ASD and DD, and if this type of training could have the potential to reduce problematic RRBs.

The first part of this chapter will provide a summary of the findings in this thesis. It will then address what those findings tell us about the current state of the literature, considering how the systematic review in the first chapter highlighted that RRBs are likely to be caused by a mixture of neurobiological factors and developmental factors, some that are perhaps driven by cognitive factors. The second part of the thesis aims to pinpoint the underlying mechanisms that are responsible for the relationship between high levels of RRBs and poor set shifting skills in young typically developing children and children with ASD. More specifically, set shifting errors and poor EF skills are traditionally interpreted as the inability to inhibit dominant stimuli, yet recent research proposes that the ability to activate previously ignored stimuli may play a key role. The second part of this chapter will therefore review how my findings fit in with different theoretical accounts that aim to address these errors. Finally, the current chapter will conclude with the strengths and limitations of the thesis, and suggestions for future research

1. Summary of results

In chapter 1, three meta-analyses were carried out to examine the strength of the relationships between EF skills and RRBs. Searches in Scopus and the ISI Search Engine made it possible to run set shifting, inhibitory control and parental-report analyses. All three analyses produced moderate but significant relationships, yet the set shifting effects were stronger than the inhibitory control ones. Whereas age and type of RRB measure moderated the inhibition and parental-rating effects respectively, no other factors moderated the results. The results in chapter 1 highlighted the need to re-open the EF account. Although this study offers support for the view that the tasks associate with the behaviours, they cannot pinpoint what causes the relationships. It was therefore concluded that future research should focus on disentangling different EF measures, particularly set shifting, to pinpoint what it is about the tasks that make them associate with the behaviours.

Chapter 2 reviewed evidence and theoretical frameworks that attempt to explain why children find it difficult to inhibit dominant stimuli and activate previously irrelevant stimuli in DCCS-like tasks, whereas adults tested on WCST adaptations struggle more with the latter. This chapter also examined why strong relationships have been identified between RRBs and WCST performance in adults, yet not with the simpler DCCS task in children. This was done by systematically reviewing various set shifting accounts and assessing if they are capable of accounting for the prominent RRBs. Most of the accounts were not capable of explaining the different trajectories for the errors in children and adults, or why the DCCS has not predicted RRB scores. This chapter does however, raise the possibility that the different results in the two populations may be caused by how the WCST measure introduces a new dimension after the rule switch, and may therefore be the only pure measure of the errors. This posed a question of whether the ability to activate previously irrelevant stimuli can explain the relationship between high levels of set shifting errors and RRBs.

In chapter 3, we further explored the prediction that to pass set shifting tasks children must be able to suppress a relevant response, and learn to attend to previously irrelevant aspects of the environment. Previous research on the DCCS showed dramatic improvement in both abilities around the age of four, but that set shifting improvement does not relate to a similar decline in RRBs, which peak in this age group and persist in ASD. Chapter 2 suggested that a previous DCCS adaptation that aimed to measure this ability was not a pure measure of the ability to activate previously ignored stimuli, and hypothesised that the difficulties with activating previously irrelevant aspects of the environment play a role in linking set shifting to RRBs. In chapter two, two studies compared variations on the standard DCCS with a new method in which the relevant response was no longer available. As predicted, only the task in which the previously ignored stimuli had to be activated by ignoring a novel dimension predicted RRB levels. These findings were explained through an automatic inhibition account. Finally, this chapter offered support to previous research that suggested that only higher-level RRBs predict EF skills. Yet, these findings were only present in the TD children population as both sub-types of behaviours predicted the relationships in the ASD and DD.

The previous chapter suggested that the abilities to inhibit previously relevant stimuli, as well as to activate previously irrelevant stimuli, play important roles in the development of set shifting skills in TD children and children with ASD. It also found that the inability to activate previously irrelevant stimuli predicted high levels of restricted and repetitive behaviours (RRBs) in all populations. These findings are novel and may have great training implications. Yet, very few interventions have trained children on EF skills and measured if training has an impact on the RRBs (e.g.Varanda &

Fernandes, 2017). In chapter 4 we highlighted different factors that may explain why so few EF interventions have been developed for individuals with ASD. We argued that previous interventions are often time consuming for researchers, teachers and parents. They also often require high levels of resources as they need to be conducted under the supervision of adults. Moreover, these interventions can be expensive to carry out, and difficult to recruit for, as there is little evidence to suggest that they will be effective for families in the long run. Future research should develop shorter and less intense interventions, as they should make EF interventions more accessible to children with ASD. This should consequently help reduce the gap between the studies that have assessed training in TD and ASD children.

Chapter 5 extended the findings in chapter 3 and 4 to investigate training implications on the LI task. This was done to examine if children could be trained on the set shifting task, and, if training on these skills generalized to a structurally related task, as well as a more complex sorting task in which a reversal shift strategy could not take place. Finally, this chapter explored if a short training program could reduce a child's RRB score. In line with our prediction, we found that pre-schoolers and children with ASD could be trained on sorting tasks. Moreover, training generalized to the related task. There was also an improvement on the more complex task for the TD training group, although this improvement not significantly different than the improvement in the control groups. During a two-month follow-up session, we found that the training and crossover effects remained with time. Finally, this training program had no significant effects on RRB levels.

2 Integration of Results and Implications for the Literature.

2.1 Neurobiological account (Lewis & Kim, 2009)

This theory proposed that genetic factors and neuroadaptations in cortical-basal ganglia pathways play important roles in the development of RRBs, and that RRBs develop if one or a few of these genes mutate and interact with experiential factors, as it will cause disruption to the circuitry. Whereas this account was outside the scope of this thesis, our findings are not incompatible with this account, as we suggest that EF skills should be explored in combination with other models.

2.2 The developmental account (Leekam, 2011)

Leekam's (2011) developmental account suggests that RRBs are immature responses that persist in children with ASD. This account is based on research that has shown that RRBs are present in TD children until around the age of four (Evans et al., 2004), and that they move from lower-level behaviours in early infancy to higher-level behaviours in later infancy (e.g. Arnot et al., 2010). This thesis did not examine this hypothesis specifically; however, some of our data can be presented to help us conclude if chronological age had an impact on a child's RRB scores. If our findings offer support for the developmental account we should find age related changes in the TD, but not ASD population.

To explore if chronological age predicted RRB levels in the TD children, we ran Linear Mixed-Effects Models (Baayen, 2008; Jaeger, 2008) that showed that age did not predict overall RRB scores (χ 2(2) = 2.00, p = .37), lower-level scores (χ 2(1) = 1.56, p = .21) or higher-level scores (χ 2(1) = 1.49, p = .22). The findings in TD children are somewhat unexpected, but may simply be a result of how the RRB scores were not significantly different in three and four year old children. This is possibly due how our sample of pre-schoolers included more three-year-olds (n= 104) than four-year-olds (n=73), making the mean age high (46 months old).

The developmental trajectory for individuals with ASD is less clear-cut. On the one hand, Militerni, Bravaccio, Falco, Fico and Palermo (2002) found a similar developmental trajectory for ASD, as although lower-level RRBs were more common in young ASD children, higher-level behaviours were more common in older children. On the other hand, Berkson and Tupa (2000) found that the behaviours were both persistent and stable over time in individuals with ASD. To address these inconsistent findings, we examined if chronological age had an effect on the ASD children's RRB levels. The analysis found no evidence to suggest that age-related changes can explain overall ($\chi^2(2) =.06$, $p = .97$), lower-level ($\chi^2(1) =.17$, $p = .68$), or higher-level RRB scores ($\chi^2(1) = .67$, $p = .41$).

In line with the research above, studies have also suggested that children with ASD with higher intellectual abilities display fewer overall RRBs (e.g. Burton et al., 2008) and particularly fewer lower-level behaviours (e.g. Szatmari et al., 2006). Again, this thesis did not examine this hypothesis specifically, however, some of our data can be presented to explore if verbal (British Picture Vocabulary Scale, BPVS; Dunn, Dunn, Whetton, & Pintilie 1982), and receptive (Leiter-r, Roid & Miller, 1997) intelligence scores affected a child's RRB scores. We did not find any evidence to suggest that verbal intelligence played a role in the development of overall RRB ($\chi^2(1) =.88$, $p = .35$), lower-level ($\chi^2(1) =.002$, $p = .97$), or higher-level RRB scores ($\chi^2(1) = .008$, $p = .93$). Moreover, we found no evidence to suggest that receptive intelligence scores had an impact on the development of overall ($\chi^2(1)$ =.03, p = .86), lower-level ($\chi^2(1)$ =.17, p = .68), or higher-level RRBs scores ($\chi^2(1)$ = .01, p = .92).

In conclusion, the findings in this thesis do not offer support for the developmental account, as there is not much evidence to suggest that chronological age or developmental level can explain a child's RRB score on the whole, or through sub-groups of lower-level and higher-level behaviours.

2.3 The EF account (Russell, 1991)

This thesis offers support for the EF account. First, through the meta-analysis in chapter 1 that found significant relationships between RRB scores and performance on set shifting and inhibitory control measures, as well as scores on parental-report rating scales. The meta-analyses therefore suggests that recent analyses of RRBs have been hasty to reject the EF hypothesis. Further support for the EF account can also be seen in the cross-sectional studies in chapter 3, as they show that TD children and children with ASD find set shifting tasks difficult, and that the ability to activate irrelevant stimuli may help to explain the significant associations with high RRBs.

3 Implications for the ability to shift away from previously dominant stimuli.

3.1 Attentional inertia Account (Kirkham, Cruess & Diamond, 2003)

The results in this thesis do not offer support for the attentional inertia account, as Kirkham, Cruess and Diamond (2003) state that "children should be able to succeed if the previously relevant values on the now irrelevant dimension are no longer present in the stimuli (and they do)", p. 451. Instead, our results suggest that TD children, children with

ASD and DD failed set-shifting tasks due to an inability to switch away from dominant rules, but also to activate previously irrelevant stimuli. Additionally, we found no support to suggest that attentional inertia can account for the strong association between RRBs and set-shifting performance, as only the inability to activate previously irrelevant stimuli predicted RRBs.

3.2 CCC-theory (Zelazo et al., 2003)

No support was found for the CCC theory, an account that suggests that children perseverate on the DCCS task, as they are not able to create and apply higher order rules for the pre- and post-switch rules accordingly (e.g. "if colour game and red then here, if shape game and rabbit then there"). Instead, they perseverate on the pre-switch rules. The results in this thesis however, suggested that children failed set-shifting due to an inability to shift away from aspects of a situation that was previously irrelevant, as well as because of the inability to activate previously irrelevant stimuli. We also found no support to suggest that higher order rules can explain the strong association between RRBs and set-shifting performance, as only the inability to activate previously irrelevant stimuli predicted RRBs. In chapter 2, we addressed the possibility that the WCST may predict RRBs due to how it introduces three dimension and four exemplars of each dimension, so it requires more higher-order rules than the DCCS. Our design allowed us to examine this in more depth as all of our task versions (apart from the DCCS) introduced more higher-order rules than the standard version. Yet, only one task version (LI) predicted RRBs. It must be acknowledged that none of the task versions in this thesis required as many higher-order rules as the standard WCST.

3.3 Active-latent account (Munakata, 1998)

This thesis does not offer support for the active-latent memory account, an account that suggests that increasingly strong memory representations make it difficult to override the initially relevant, but now irrelevant, stimuli. In the WCST the rule change is sudden and adults are not reminded of the rules. Instead, individuals are given feedback ("correct" and "incorrect"). This differs from the DCCS design as children are reminded of the rules prior to every sort. It is therefore possible that a memory confound is responsible for the association between high RRB levels and poor WCST performance. However, we did not find any support for this framework, as all of our task versions followed the DCCS design in the way that children were reminded of the rules prior to every trial. If set-shifting difficulties and RRBs were a result of memory confound, none of the tasks should have predicted RRBs, yet the LI task did.

4 Implications for the ability to activate previously ignored stimuli.

4.1 CCC-r theory (Zelazo et al., 2003)

On the one hand, the findings in this thesis offer support for Zelazo et al.'s (2003) Cognitive Complexity and Control theory-revised (CCC-r). The CCC-r theory hypothesises that individuals perseverate on set shifting tasks as an individual struggle with activating rules that were relevant during the pre-switch, as well as suppressing attention to previously irrelevant rules. Our results suggest that TD children, as well as children with DD and ASD perform poorly on all of the tasks that require the ability to inhibit previously dominant rules, as well as activate previously irrelevant rules. This account is however, not able to account for why children performed poorly on the LI task as this is the only task in which it was not possible for children to perseverate on the pre-switch dimension. Moreover, it cannot explain why the LI condition is the only task that predicted RRBs. If higher-order rules played a part, all of our tasks apart from the LI manipulation should have predicted RRBs.

4.2 Episodic retrieval account (Neill, 1997)

The episodic retrieval framework suggests that an episodic memory trace is formed when a stimulus is first encountered in an irrelevant situation, marking the stimulus with a ("do not respond" tag). When the same stimulus then becomes relevant at a later time, the episodic "do not respond" memory is automatically retrieved. If this memory conflicts with current situational demands it produces NP errors. The results in this thesis do not offer support for this account for several reasons. Firstly, if this explanation would hold up the LI and NP task should have predicted RRBs. Secondly, all of our tasks are modelled on the DCCS task, meaning that the tasks should not rely on memory skills as children are always reminded of the rules prior to each trial. This then makes it unlikely that memory tags explain the association between RRBs and task performance on the LI task.

4.3 Attentional theory (Maes, Damen & Eling, 2004)

The findings in this thesis offered support for the attentional theory. This hypothesis was based on the automatic inhibition framework (see Maes et al, 2004). This framework suggests that individuals may struggle with set shifting tasks due to difficulties with overcoming both controlled and automatic inhibition. More specifically, the aim of the P tasks is to focus an individual's attention on the distractor in the pre-switch for then to measure if they are able to switch away from dominant stimuli when it later becomes irrelevant. This process is argued to require controlled inhibition. In contrast, the aim of the LI task is to pre-expose individuals to the target stimuli in pre-switch, meaning that the target stimulus is continuously inhibited throughout the pre-test phase since an individual's attention is directed to other relevant stimuli. This then leads to automatic inhibition when a child must activate previously ignored responses at a later stage. Maes et al. argue that this type of inhibition is less effortful than the ability to shift away from previously dominant inhibition, and that this can help explain why adults find the task that requires automatic inhibition harder. The fact that we found that children struggled with both of the tasks may simply be a result of immature inhibitory control, as this could lead to difficulties in both of the tasks. In terms of the relationship with RRBs, our findings only found evidence to suggest that difficulties with overriding automatic responses lead to high levels of shifting errors and repetition of well-learned behaviours. This finding is novel and if the WCST only is a pure measure of this type of errors it can help us explain why the WCST but not the DCCS task associates with RRBs.

The findings in this thesis propose that learned irrelevance is the critical factor in set shifting tasks that is responsible for the strong relationship with RRBs. We propose that this component is particularly relevant in the WCST, and that LI errors consequently explain the strong relationship between performance on this task and RRB scores. We argue that neither the traditional DCCS or the partial change task successfully measure this ability. Although the partial change condition replaces the previously relevant stimuli with new exemplars of the same dimension, we believe that children may still be perseverating on the previously relevant dimension. This account would explain why there is a stronger association between with WCST than the DCCS. It can also explain why we found an association between the frequency of RRBs and performance on the LI task but not the DCCS or partial change task. There are however, other differences between the DCCS and WCST that must be considered before we can be certain. The WCST requires higher levels of flexibility than the DCCS, as it introduces four dimensions and more overall switches. The nature of the task is also more complex as participants are not provided with rules, only feedback. The differences in complexity may therefore consequently help explain why the association has only been found in adults. In order to examine this further, future studies should assess children on similar task adaptations modelled on tasks that are more comparable to the traditional WCST, such as the Modified Wisconsin Card Sort Task (M-WCST, Schretlen, 2010).

5 Limitations

In addition to measuring RRBs through parent ratings, it would have strengthened our implications if we had included another type of measure, such as the RBQ-2 teacher ratings version or a clinical measure, such as the ADOS. It would have also been of interest to measure if LI scores predicted communication difficulties, the other diagnostic criteria for ASD, as well as to examine if training has an impact on this skill. In line with recent developments in RRB research, we could have also examined if anxiety played a role in the relationship.

As well as the general limitations above, there are also a few limitations that only apply to a few of our experiments. First of all, we did not measure intellectual abilities for our TD sample in the correlational study or in our training study. To examine this factor in the TD children would have helped us conclude if developmental level played a role on TD children's performance on the sorting adaptations or the RRB scores.

Furthermore, we did not introduce a control task in which both of the dimensions were changed in the post-switch. This would have been a useful control measure, as if set shifting consists of the ability to overcome dominant responses and the inability to activate previously irrelevant stimuli, then children should not find this task difficult. Moreover, performance on this task should not predict RRBs as our results suggest that the ability to activate previously irrelevant stimuli only is responsible for these errors. Finally, a limitation of our training study in chapter five was that we had a small sample size.

6 Future directions

Future research should explore the relationship between RRB scores and LI errors through longitudinal research designs that also examine the possible mutual influences of genetic factors and non-shared environmental influences on the development of EF. Moreover, future research should also develop training programs with larger sample sizes to increase power. Since the DCCS task is only appropriate for young TD children, future studies should also extend and create similar adaptations in other flexibility tasks that can be used in a wider age-range, such as the Switching Inhibition and Flexibility Task (SwiFt task; Carroll & Cragg, 2012). This should help to understand the association between LI errors and RRBs from childhood to adulthood in individuals in which the behaviours persist. Furthermore, future studies should further explore why the association with parental-report scales were only significant when measured through RRB specific measures. This can be examined by exploring if the same results appear through clinical rating measures, such as the ADOS. This type of analysis would help conclude if the results appear because the extensive nature of the measure examines a wider range of behaviours, or if the association is caused by how parents rated both scales. In addition to this, future studies should address the role of anxiety in the relationships between LI and RRBs, as well as the role that communication skills play in the relationship. Finally, future research should extend the research to other populations in which the behaviours are present, such as OCD and Attention Deficit Hyperactivity Disorder (ADHD).

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