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BEHAVIORAL INTERVENTIONS FOR HIGHER-ORDER RESTRICTED AND REPETITIVE BEHAVIORS IN AUTISM: A SYSTEMATIC REVIEW AND META-ANALYSIS

A dissertation submitted in partial fulfillment

of the requirements for the degree of

DOCTOR OF PSYCHOLOGY

to the faculty of the

DEPARTMENT OF PSYCHOLOGY

of

ST. JOHN'S COLLEGE OF LIBERAL ARTS AND SCIENCES

at

ST. JOHN'S UNIVERSITY

New York

by

Emily Nichols

Date Submitted: _____ Date Approved: _____

_____ _____

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ABSTRACT

BEHAVIORAL INTERVENTIONS FOR HIGHER-ORDER RESTRICTED AND REPETITIVE BEHAVIORS IN AUTISM: A SYSTEMATIC REVIEW AND META-ANALYSIS

Emily Nichols

Although higher-order restricted and repetitive behaviors (H-RRBs) are associated with social, emotional, and behavioral difficulties in individuals with Autism Spectrum Disorder (ASD), there is a paucity of literature on behavioral interventions to treat this core symptom. Through a systematic search, 29 intervention studies that target H-RRBs were identified and analyzed in terms of (a) participant characteristics, (b) setting, (c) targeted behaviors, (d) intervention procedures, (e) experimental design, (f) outcomes and certainty of evidence, and (g) generalization and maintenance of outcomes. Overall, 41% of studies (*n*=12) yielded large effect sizes, 34% yielded medium effects (*n*=10), and 17% yielded small effects (*n*=5). Results suggest that studies yielding large effect sizes were more likely to be rooted in applied behavior analysis (ABA) than cognitive behavioral therapy (CBT), more likely to use function-based than non-function-based interventions, more likely to use both antecedent- and consequence-based interventions versus either one alone, more likely to use single-subject designs than group designs, and were more likely to be classified as focused intervention practices than comprehensive treatment models or manualized

treatment programs. Overall, results suggest that ABA and CBT approaches to treating H-RRBs show promise. Directions for future research are discussed.

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INTRODUCTION

Restricted and repetitive behaviors (RRBs), along with social-communication impairments, are a core feature of Autism Spectrum Disorder (ASD; American Psychiatric Association [APA], 2013). Although RRBs are observed in both typically developing children and those with developmental delays (Harrop, McConachie, Emsley, Leadbitter, & Green, 2014), these behaviors are more intense and occur more frequently in children with ASD (Bodfish, Symons, Parker, & Lewis, 2000; Matson, Dempsey, & Fodstad, 2009). RRBs encompass four different sub-symptom areas, according to the newly revised *Diagnostic and Statistical Manual of Mental Disorders* (DSM-5; APA, 2013) diagnostic criteria for ASD. These include: (a) stereotyped motor movements, use of objects, or speech; (b) insistence on sameness, inflexible adherence to routines or ritualized patterns; (c) circumscribed interests, and (d) hyper- or hyporeactivity to sensory input or unusual interest in sensory aspects of the environment (APA, 2013). Although the majority of the literature on ASD has historically focused on the social and communication deficits that are characteristic of the disorder (Lewis & Bodfish, 1998; Turner, 1999), researchers have increasingly started to investigate the phenomenology and mechanisms of RRBs (Boyd, McDonough, & Bodfish, 2012). However, research on interventions to treat RRBs is still lacking (Boyd et al., 2012; Harrop, 2015).

Classifying Restricted and Repetitive Behaviors

Since the term "restricted and repetitive" encompasses a broad range of heterogeneous behaviors, the various RRBs are often separated into two classes (Turner, 1999). *Lower-order* restricted and repetitive behaviors (L-RRBs), such as stereotypic (seemingly purposeless, repeated) motor movements and self-injurious behaviors, are

characterized by repetitive movements (Turner, 1999). *Higher-order*, more cognitively complex restricted and repetitive behaviors (H-RRBs), such as compulsions, are characterized by an inflexible adherence to some rule or mental framework (e.g., needing things to be "just so," Boyd et. al., 2012). H-RRBs also include ritualistic/sameness behaviors (e.g., a need for consistency in both activities and in the environment) and circumscribed interests, which are abnormally intense or focused interests that are highly specific (Lam & Aman, 2007). This systematic review examines behavioral interventions for H-RRBs (i.e., compulsions, ritualistic/sameness behavior, circumscribed interests) as well as the overall behavioral inflexibility related to these symptoms.

The Importance of Studying Higher-Order Restricted and Repetitive Behaviors

RRBs are associated with a host of challenging problems for individuals with ASD and their caregivers (Lovass, Koegel, & Schreibman 1979). Parents report that RRBs are the most stressful symptom of ASD to manage (Bishop, Richler, Cain, & Lord, 2006) and studies have found that these behaviors take up "a substantial amount of the child's daily time and energy in classroom, community, and home settings" (Patterson, Smith, & Jelen, 2010, p. 323). RRBs are not only associated with interferences in learning and increased caregiver stress, they are also related to sleep disturbances (Abel, Schwichtenberg, Brodhead, & Christ, 2018; Hundley, Shui, & Malow, 2016) and—if routines are disrupted— physical aggression toward others (Rispoli, Camargo, Machalicek, & Lang, 2014).

Although both lower- and higher-order RRBs have a substantial negative impact on quality of life, H-RRBs may be particularly important to study. Historically, interventions for RRBs have focused on reducing L-RRBs (Bodfish, 2004; Leekam,

Prior, & Uljaveric, 2012), while comparatively few intervention studies target H-RRBs (Boyd & Wakeford, 2013; Harrop, 2015). This is especially concerning, given that some researchers have suggested that H-RRBs can be even more impairing than L-RRBs to both individuals with ASD (Bodfish, 2004) and their caregivers (South, Ozonoff, & McMahon, 2005). For example, Bodfish (2004) argued that L- RRBs "do not seem to produce the kind of all-encompassing problems that the more general pattern of behavioral rigidity (e.g., inflexibility, resistance to change, need for sameness, restricted interests) seems to produce for persons with autism" (p. 323). Indeed, reports from parents of individuals with ASD revealed that their child's inflexibility in the face of change and the continual talking about restricted interests are the most difficult aspects of the disorder to regularly manage (South et al., 2005). For children with ASD, challenges with inflexibility negatively affect several domains of their lives including their play, conversations, eating habits (Koegel et al., 2012), and potentially even learning acquisition among school-aged children with ASD (Troyb et al., 2016).

Further, while L-RRBs have been found to remain stable or decrease in severity throughout childhood (Kim & Lord, 2010), H-RRBs, such as insistence on sameness, have been found to persist and even worsen over time in ASD (Lam & Aman, 2007; Richler, Huerta, Bishop, & Lord, 2010). Additionally, circumscribed interests, rituals, and compulsions appear to be more prevalent than stereotyped movements and self-injurious behaviors in adults with ASD (Esbensen, Seltzer, Lam, & Bodfish, 2009). These data suggest that H-RRBs do not simply abate with time in those with ASD, warranting the study of interventions specifically targeting this category of behaviors. Overall, the fact that H-RRBs make up such ubiquitous and enduring symptoms which interfere with the

daily functioning of individuals with ASD as well as their families and caretakers, yet are the least studied in the intervention literature, motivated this author to conduct this systematic review of interventions to address H-RRBs.

Behavioral Interventions for Restricted and Repetitive Behaviors

Antecedent- and consequence-based interventions. Behavioral interventions for ASD have traditionally been rooted in applied behavior analysis (ABA, Bearss et. al., 2015; Boyd & Wakeford, 2013), which seeks to modify target behaviors by applying the principles of operant conditioning and its related procedures. The efficacy of behavioral interventions based on the principles of ABA for individuals with ASD has been well documented in the literature (e.g., National Autism Center, 2015; Wong et al., 2015). These behavioral interventions are typically described as being consequence- or antecedent-based. In *consequence-based* interventions, RRBs are targeted by eliminating or decreasing the reinforcement an individual receives for engaging in such behaviors and increasing the reinforcement they receive for engaging in alternative/incompatible behaviors or displaying the absence of a target behavior (National Autism Center, 2015). *Antecedent-based* interventions typically involve (1) environmental modifications or making changes to the individual's routine, and/or (2) environmental or skill enrichment to reduce the rates of RRBs or to prevent their occurrence (Boyd, et al., 2012; Rapp & Vollmer, 2005). Although one systematic review examined the efficacy of antecedent-based, reinforcement or skills-based, consequence-based, or mixed interventions in reducing RRBs (Mulligan, Healy, Lydon, Moran, & Foody, 2014), only two out of the 59 studies they identified targeted H-RRBs, which is the purpose of the present review. Additionally, beyond the broad categories of antecedent- and

consequence-based, understanding which specific intervention strategies (e.g., reinforcement, functional communication training, video modeling) are effective in reducing H-RRBs will enable parents and providers to make informed decisions when deciding on appropriate treatment.

Cognitive Behavioral Therapy. While many interventions for ASD have been informed by ABA, cognitive behavioral therapy (CBT) — more specifically, exposure and response prevention (ERP)—has recently begun to inform the intervention research on H-RRBs in ASD (Boyd et al., 2012). CBT emphasizes the relationship between an individual's thoughts, emotions, and behaviors (Luxford, Hadwin, & Kovshoff, 2017), incorporating both classical and operant conditioning with cognitive interventions. For example, acceptance and commitment therapy training (ACTraining), a CBT-based intervention that includes cognitive defusion exercises, acceptance strategies, present moment awareness, and flexible self-perspective taking, has been used to promote behavioral flexibility during play in children with ASD (Szabo, 2019).

CBT can focus on behavioral interventions (such as exposure) or cognitive interventions (such as cognitive restructuring or cognitive defusion), or both, with ERP-based CBT demonstrating the strongest empirical support for the treatment of OCD (Simpson, Neria, Lewis-Fernandez, & Schneier, 2010). ERP is a specific form of CBT which involves gradually exposing an individual to a feared stimulus that elicits anxiety/obsessions and subsequent compulsions, while the response prevention component involves extinguishing the compulsive behaviors (i.e.,, refraining from the rituals that typically follow the obsessive thought/urge/impulse) (Boyd et al., 2012). Given that compulsions and ritualistic/sameness behavior seen in ASD appear to share

some similarities with the obsessions and compulsions seen in OCD (if they both serve the function of escape or avoidance), researchers have recently started to examine the utility of using ERP to treat H-RRBs in individuals with ASD (Boyd, Woodward, & Bodfish, 2011; Boyd et al., 2012). Appendix A provides a brief description of some common interventions used in managing RRBs.

Function-based versus non-function-based interventions. Central to ABA is conducting a functional behavior assessment (FBA), the goal of which is to understand the function or purpose of the target behavior – that is, *why* an individual engages in the target behavior (Boyd & Wakeford, 2013). Although traditionally RRBs were thought to be maintained primarily through automatic/sensory reinforcement, recent studies suggest that they appear to serve a variety of functions (Cunningham & Schreibman 2008; Kennedy, Myer, Knowles, & Shulka, 2000). For example, two studies using functional analysis determined that H-RRBs were maintained by social attention (Fisher, Rodriguez, & Owen, 2013; Rehfeldt & Chambers 2003) in some individuals with ASD, while another suggested that these inflexible behaviors serve to escape or avoid social interactions and aversive task demands in other individuals with ASD (Szabo, 2019). These differences illustrate the importance of identifying the function of H-RRBs in order to select the most appropriate intervention (Boyd et al., 2012; Factor et al., 2016).

Function-based behavioral interventions are those that are derived from the results of a functional behavior assessment (FBA) of the target behavior before starting treatment, while non-function-based behavioral interventions use the principles of ABA, but are not based on a prior FBA (Mulligan et al., 2014). Although Patterson, Smith, and Jelen (2010) systematically reviewed function-based behavioral interventions for RRBs,

only one out of their ten studies targeted a higher-order RRB. Similarly, Mulligan and colleagues (2014) compared function-based to non-function-based interventions for the treatment of RRBs in ASD; however, just one of the 37 function-based interventions targeted H-RRBs (Wolff, Hupp, & Symons, 2013), and only one of the 22 non-function-based interventions targeted H-RRBs (Boyd, McDonough, Rupp, Khan, & Bodfish, 2011). Thus, although previous research has underscored the benefits of functional assessment in the treatment of RRBs (Boyd et al., 2012; Leekam et al., 2012), to date no systematic review has directly compared function-based and non-function-based behavioral interventions specifically targeting H-RRBs, as in the present review.

Focused versus comprehensive interventions. Interventions for ASD can also be categorized as either focused intervention practices (FIPs), such as reinforcement and prompting to teach specific skills (Boyd et al., 2012; Odom, Sally, Hatton, & Rogers, 2010), or comprehensive treatment models (CTMs) designed to improve a broader array of symptoms (National Research Council, 2001). CTMs, such as Lovaas' UCLA Model or Early Intensive Behavioral Intervention (EIBI), Pivotal Response Treatment (PRT), Early Start Denver Model (ESDM), and TEACCH, typically address a variety of developmental and/or behavioral domains that include the core features of autism (e.g., social-communicative skills, communication, play, RRBs), among other skills such as pre-academic skills (Odom et al., 2010; National Research Council, 2001). CTMs must also have a manual or curriculum that clearly describes the procedures as well as a clear conceptual framework (Odom et al., 2010). CTMs are also intensive, typically occurring

between 25-40 hours per week, and occur over an extended period of time (e.g., 1-3 years) (Odom et al., 2010).

Typically FIPs and CTMs are contrasted when evaluating the efficacy of symptom reduction for those with ASD; however, recent studies suggest that the use of a third method, manualized treatment packages (MTPs), may be a promising intervention for reducing H-RRBs (Lin & Koegel, 2018; Vause et al., 2017). MTPs are a category created by this author to classify those intervention programs that are similar to CTMs in that they are manualized and "branded" interventions (e.g., "Denver Model") comprising multiple components, but they are unlike CTMs in that they focus on just one symptom or a narrower range of symptoms (e.g., just H-RRBs versus multiple developmental domains) and, consequently, are less intensive in hours per week (e.g., 1-2 hours per week for MTPs). However, MTPs also differ from FIPs due to their structure and manualized format as well as their incorporation of multiple FIPs, rather than a single FIP.

The National Standards Project (NSP; 2015) identified several FIPs (e.g., reinforcement, response interruption and redirection) that are effective in reducing RRBs (National Autism Center, 2015). However, the NSP did not differentiate between lower- and higher-order behaviors in its definition of RRBs, obscuring whether or not these interventions are effective for the latter. Previous systematic reviews have examined FIPs for both lower- and H-RRBs; however, when examining interventions for H-RRBs, these reviews only included single-subject designs and not group designs, did not compare function- and non-function-based interventions, and were selective, rather than comprehensive systematic reviews (Boyd et al., 2012; Raulston & Machalicek, 2018).

Additionally, Raulston and Machalicek's (2018) review was restricted to interventions for children under the age of six. Given that some H-RRBs, such as circumscribed interests, appear to worsen in severity as children age (South et al., 2005) and may be targets for intervention in individuals older than six-years-old, the present review expands upon Raulston and Machalicek's (2018) findings by including interventions for H-RRBs in individuals of all ages with ASD. Further, although previous reviews have examined the use of FIPs, CTMs and what this author refers to as MTPs in the treatment of RRBs, to date no systematic review has compared all three types of intervention in the treatment of H-RRBs.

Present Study and Hypotheses

Despite the publication of several review articles on behavioral interventions for RRBs, no prior review has systematically examined the efficacy of both function-based and non-function based behavioral interventions to address H-RRBs. This is an important omission, given that parents of children with ASD often report that their children's perseverative interests and rigid adherence to routines are two of the most challenging features of the disorder that they encounter every day (South, et al., 2005).

Thus, the current systematic review updates and expands upon these previous reviews by (a) directly comparing the efficacy of function-based and non-function-based behavioral interventions as well as examining (b) both antecedent- and consequence-based interventions, (c) both focused practices and comprehensive treatment models and manualized treatment programs, and (d) both single-subject designs and group designs in the treatment of H-RRBs for individuals with ASD of all ages. Often, reviews of interventions for RRBs focus on lower-order behaviors or report on

both higher- and lower-order RRBs in aggregate, making it difficult to determine whether these interventions are effective at reducing H-RRBs. Further, Boyd and colleagues (2012) suggested that future research on interventions for RRBs should focus more on the underlying behavioral inflexibility to foster positive outcomes for individuals with ASD and their families. Therefore, in addition to including studies that target a specific higher-order RRB, this review includes interventions that seek to promote overall behavioral flexibility (i.e., the ability of an individual to adapt his behavior in response to changes in the environment; Brown & Tait, 2015). This review differs from previous reviews of RRBs by focusing solely on interventions targeting H-RRBs and the behavioral inflexibility that underlies these symptoms. By creating a separate systematic review of interventions for this understudied feature, researchers and practitioners will have a clearer idea of which treatments are promising for ameliorating this core symptom of ASD. Additionally, unlike selective reviews (e.g., Boyd et al., 2012; Raulston & Machalicek, 2018), the current review employed a systematic search that attempted to identify all relevant studies using an explicit and reproducible methodology.

In sum, the main objective of the current study is to systematically review single-subject design and group design studies that have used behavioral interventions to reduce H-RRBs and identify: (a) whether function-based or non function-based interventions are more effective in reducing H-RRBs; (b) whether antecedent-based, consequence-based, or mixed interventions are more effective in reducing H-RRBs; (c) whether FIPs, MTPs, or CTMs are more commonly used and effective in reducing H-RRBs, and (d) which specific intervention strategies (e.g., reinforcement, response interruption and redirection) demonstrate the most success in reducing H-RRBs.

Method

Identification of Studies

Search procedures. The following electronic databases were searched for references from 1994 up to March 1st 2020: ERIC (Education Resources Information Center), PsycINFO, Web of Science and PubMed. The keywords fields in all four databases were searched using the Boolean terms (Autis*) or (Asperger*) or (ASD) or (PDD*), and the following terms related to RRBs: stereotyp*; perseverative interest*; ritual* rigid*; flexib*; compuls* compulsive behavior; sameness; circumscribed interests; obsessive interests; preferred interests; special interests; restricted interest; preferred topic; perseverative topic; repetitive behavior; obsessive behavior; arranging and ordering; behavior modification; applied behavior analysis; behavioral intervention; behavioral strategies; cognitive behavioral treatment; cognitive behavioral intervention. The search was limited to peer-reviewed articles published in English. Appendix B displays the study selection procedure based on the Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA, Moher, Liberati, Tetzlaff, & Altman, 2009) process.

Inclusion and exclusion criteria. To be included in this review, the study must have met the following criteria. First, the study must have included at least one participant with a *DSM-IV* diagnosis of Autistic disorder, Asperger syndrome, or Pervasive Developmental Disorder – Not Otherwise Specified (PDD-NOS) or a *DSM-5* diagnosis of Autism Spectrum Disorder.

Second, the study had to implement a behavioral intervention with the goal of reducing the frequency or severity of one or more H-RRBs or increasing overall

behavioral flexibility. *Behavioral intervention* was defined as a procedure involving manipulation of environmental antecedents and/or consequences in order to increase or decrease a targeted behavior (National Autism Center, 2015). Studies in which the intervention was cognitive-behavioral in nature were also included. *Cognitive-behavioral interventions* were defined as procedures which seek to modify behavior by teaching individuals to understand and change their thoughts, emotions, and behaviors (Luxford, Hadwin, & Kovshoff, 2017).

Initially, titles and abstracts were screened for interventions targeting RRBs in general. If it could not be determined from the title and abstract whether the intervention targeted a lower versus higher-order RRB, the full text of the article was referred to. Studies that did not include a behavioral intervention targeting the reduction of at least one H-RRB were excluded from this review. Studies were also excluded for the following reasons: (a) studies used medical or pharmacological interventions, sensory integration treatment, dietary modifications, or other non-behavioral treatments (e.g., music therapy, massage therapy, auditory integration training, equine-assisted therapy, exercise); (b) studies described clinicians' impressions of interventions or focused solely on the description of H-RRBs or their assessment; (c) studies were not empirical or data-based (i.e., descriptive case studies without quantitative data) (d) studies did not include at least one individual with ASD, and (e) studies were written in a language other than English. Review articles, meta-analyses, non-peer reviewed articles, conference abstracts, books, and chapters were also excluded. Studies focusing on the treatment of OCD were also excluded, as the extant literature suggests that symptoms of this disorder may serve different functions compared to H-RRBs (Kerns et al., 2014); however, studies that aimed

to treat "OCD-like behaviors" (referred to as OCBs, Vause et al., 2017) in individuals with ASD who did not have a diagnosis of OCD were included. These inclusion/exclusion criteria are similar to those used in recent systematic reviews of interventions for individuals with ASD, other developmental disabilities, and/or specific interventions (e.g., Harrop, Amsbary, Towner-Wright, Reichow, & Boyd, 2019; Palmen, Didden, & Lang, 2012; Patterson et al., 2009). Finally, although some clinicians and parents consider circumscribed interests (CIs) to be "islands of ability" (Mercier et al., 2000) which can be used to promote social behaviors or decrease inappropriate behaviors (Boyd et al., 2012), this review focuses on interventions seeking to *reduce* H-RRBs. Therefore, studies that incorporate CIs into interventions to address other behaviors, rather than target the reduction of CIs or other H-RRBs as an outcome variable, were excluded.

Inter-rater reliability for inclusion criteria. To ensure reliable application of inclusion/exclusion criteria and to determine initial eligibility of studies in the review, two research assistants (R.A.s) from St. John's University — a graduate student R.A. (AV) and an undergraduate R.A. (HZ) — were trained in coding specific categories using 22 practice articles. In order for R.A.s to move on from practice articles to the real articles, they needed to achieve 80% inter-rater reliability (IRR) on the practice articles. Two rounds of double coding were conducted. In the first round, each R.A. independently coded the same two practice articles as the principal investigator and initial IRR was obtained by calculating percent agreement (e.g., dividing the number of agreements regarding inclusion of an article by the sum of the agreements and disagreements between raters and multiplying by 100). The initial IRR for the two practice articles was

100%. The next round of coding followed the same format with 20 practice articles being double-coded. Mean IRR was 90% (AV = 100%; HZ = 80%).

Study Selection. The initial search yielded 6,533 studies. Retrieved studies were initially searched for duplicates, which left a remaining 2,862 papers to be screened. Titles and abstracts of 50% of the studies were randomly assigned to the R.A.s to determine if they met the inclusion criteria. The principal investigator coded the other 50% of studies. Additionally, 50% of articles assigned to the R.A.s were *double coded* by the primary investigator. Agreement as to whether or not an article should be included was obtained by entering the agreement data into Excel. On this initial screening of titles and abstracts for inclusion, mean IRR = 96% (AV = 95%; HZ = 96%). In instances where the R.A. and principal investigator disagreed on whether an article should be included, they reviewed the article and discussed the study until the disagreements were reconciled.

After articles were excluded based on title and abstract, 403 articles were identified for full-text screening. At this stage, all eligible papers were screened in full by the principal investigator and 50% of these articles were double-coded by the graduate student R.A (AV). Disagreements between reviewers were resolved by consensus and the agreement rate was 86%. In total, 374 papers were removed due to not meeting the eligibility criteria, leaving 29 studies to be included in the review (see Appendix B).

Data Extraction and Coding

Studies that met the inclusion criteria were summarized in terms of the following nine variables: (a) participant characteristics (e.g., age, number, comorbid diagnoses, medications, and cognitive functioning), (b) the setting in which the intervention was conducted (e.g., home, school, clinic), (c) targeted behaviors (i.e., compulsions,,

ritualistic/sameness behaviors, circumscribed interests, or behavioral inflexibility), (d) intervention procedures, including whether a study used antecedent-and/or consequence-based strategies, and whether an intervention was classified as an FIP, CTM, or MTP, (e) theoretical framework (e.g., ABA, CBT), (f) experimental design (e.g., single-case designs, group-based designs, randomized controlled trials), (g) outcomes of the intervention, (h) certainty of evidence, and (i) generalization and maintenance of outcomes.

Inter-rater reliability for data extraction and coding. To establish IRR on the coding of the aforementioned nine variables, the author coded each article and an R.A. double coded 100% of the articles. IRR was calculated in Excel using percent agreement and IRR = 88% (84-92%). Disagreements were resolved by discussion between the two reviewers and, when necessary, an expert on interventions for individuals with ASD was consulted to resolve any outstanding disagreements.

Quality of Research Evaluation

Evaluation of intervention outcomes. Intervention outcomes were evaluated based on the experimental design of the study. For studies employing group designs or whose data were analyzed at the group level, standardized mean difference was calculated to estimate the difference between the treatment and control conditions. Specifically, Hedges' *g* was used to calculate effect sizes. Scores were described as having a small (0.2), medium (0.5), or large (0.8) effect size (Cohen, 1988). For studies employing single-case design, Parker and Vannest's (2009) Nonoverlap of All Pairs (NAP) was calculated. NAP is an index of data overlap between single-subject design phases and is comparable to other single-subject outcome measures (e.g., Percent of

Nonoverlapping Data, Percent of All Nonoverlapping Data). However, NAP is less sensitive to the influence of outliers compared to these measures (Ramdoss et al., 2011). NAP is also less subject to human error and offers greater score precision, demonstrated by narrower confidence intervals (Parker & Vannest, 2009) . NAP summarizes the overlap between all pairs of baseline (phase A) and intervention (phase B) data points. NAP was calculated by adding the number of comparison pairs not showing overlap and the number of tied comparison pairs, then dividing by the total number of comparisons (Parker & Vannest, 2009). Based on the recommendations outlined by Parker and Vannest (2009), NAP scores between 0 and .65 were classified as "weak effects," .66–.92 as "medium effects," and .92–1.0 as ''strong effects." These methods for evaluating outcome have been used in recent systematic reviews of interventions for individuals with ASD (e.g., Lang et al., 2011; Ramdoss et al., 2011).

Certainty of evidence. Certainty of evidence was determined by evaluating the results in terms of study design and additional methodological factors (Schlosser & Sigafoos, 2007). The certainty of evidence for each study was rated as "suggestive," "preponderant," or "conclusive" (Simeonsson & Bailey, 1991; Smith, 1981). Studies were rated as having *suggestive* evidence—the lowest level of certainty— if they (1) lacked a true experimental design (e.g., group design with random assignment and a control group, multiple-baseline or an ABAB design), (2) did not report adequate inter-observer agreement (e.g., coefficients of 80% or higher collected for at least 20% of sessions); or (3) did not operationally define the intervention procedures or outcome variables. In order to be rated as having a *preponderance* of evidence, studies must have met the following criteria: (1) experimental design must have been used; (2) sufficient

interobserver agreement data (e.g., 20% of sessions and 80% or better agreement) must have been reported; (3) intervention and outcome variables must have been operationally defined; and (4) the study must have included enough details about the intervention to allow for replication. Finally, studies that met the four criteria of preponderance in addition to (5) controlling for alternative explanations of intervention effects were rated as *conclusive*. This method of evaluating the certainty of evidence has been used in several recent systematic reviews of interventions for individuals with ASD (Chan et al., 2009; Lang, Regester, Lauderdale, Ashbaugh, & Haring, 2010; Neely, Gerow, Rispoli, Lang & Pullen, 2016; Ramdoss et al., 2012; Verschurr, Didden, Lang, Sigafoos, & Huskens, 2014).

Results

The systematic search procedures and the application of the predetermined inclusion and exclusion criteria resulted in the inclusion of 29 studies in this review. Table 1 summarizes the participant characteristics and setting. Table 2 summarizes the (a) dependent variables (target behaviors; i.e., the specific type of H-RRB), (b) intervention procedures (c) study design (d) intervention outcomes and certainty of evidence and (e) generalization and maintenance of the 29 included studies.

Participants

Collectively, the 29 studies provided direct intervention to a total of 284 participants with ASD. The majority of studies (59%; $n = 17$) included participants between the ages of four and 12 years, while six studies (21%) included participants between the ages of 13 and 18 years [1, 9, 10, 12, 22, 24]. Five studies (17%) included children younger than four years [2, 7, 15, 20, 23]. Finally, one study (3%; Wolff, Hupp, & Symons, 2013) included adult participants with ASD ranging from 42-54 years of age. Severity of cognitive functioning or adaptive behavior was reported for the individual participant(s) with ASD in 21 studies (72%), 15 of which reported that participants had a comorbid diagnosis of ID [71%; 3, 7, 8, 9, 10, 11, 13 15, 17, 18, 20, 21, 26, 27, 28]. Five of the studies (17%) reported comorbid diagnoses other than ID across participants and these most commonly included internalizing and externalizing disorders[1, 3, 6, 9, 27].

Settings and Intervention Agents

Descriptions of the setting and deliverer of the intervention were provided for all studies. Interventions were implemented in outpatient clinics or University clinics (37%; $n = 11$), schools (28%; $n = 7$), homes (17%; $n = 5$), residential schools (7%; $n = 2$),

preschools (3%; *n* = 1), group homes (3%; *n* = 1), community settings (3%; *n* = 1) and an inpatient unit (3%; *n* = 1). For some participants, interventions occurred in more than one setting. The majority of the studies (66%, *n* = 19) used a trained therapist/researcher to implement the intervention. Six of the interventions (20%) were implemented by teachers and four (14%) were implemented by parents.

Target Behaviors

The majority of studies (34%; *n* = 10) targeted multiple domains of H-RRBs [2, 3, 7, 8, 14, 15, 23, 26, 27, 28]. Six studies (21%) exclusively targeted ritualistic/sameness behaviors, including resistance to changing routines, insistence on wearing certain clothing, and rigid adherence to rules and routines [1, 4, 5, 16, 17, 25]. Six studies (20%) exclusively targeted compulsive behaviors, including arranging and ordering objects, labeling objects, counting, and checking [9, 11, 18, 22, 24, 29]. Five studies (17%) exclusively targeted perseverative speech about restricted interests [6, 10, 19, 21] or repetitive communication during social interactions [12]. Two studies (*n* = 7%) exclusively targeted overall behavioral flexibility [13, 20]..

Outcome Measures

Sixteen studies (59%) used direct observation to collect data on the target behaviors [2, 5, 6, 9, 10, 11, 12, 15, 16, 17, 18, 21, 22, 23, 25, 29]. Direct observation typically involves continuous recording or periodic sampling of a behavior of interest, whereas indirect observation relies on the use of rating scales, questionnaires, or interviews to make estimations about the frequency or severity of a behavior (Fisher, Piazza, & Roane 2013). Eight studies (28%) relied on indirect outcome measures [1, 4, 7, 8, 20, 26, 27, 28], the majority of which were standardized, validated interviews or

questionnaires about RRBs that can utilize different respondents (clinician, parent, teacher, self). These questionnaires and interviews included the Repetitive Behavior Scale- Revised (RBS-R; Bodfish et al. 1999); the Repetitive Behavior Questionnaire (RBQ; Turner 1999); the Dutch version of the revised Behavior Flexibility Rating Scale (BFRS-R; Green et al., 2007); the Children's Yale-Brown Obsessive–Compulsive Scale (CY-BOCS; Goodman et al. 1992); and the Child Obsessive–Compulsive Impact Scale-Revised Parent (COIS-RP; Piacentini et al. 2007); the Rigidity Rating Scale- Child and Parent Report (Boon, 2017); the Parent OCB Rating Scale (Vause et al., 2017); the MGH YouthCare Social Competency/Social Skill Development Scale (SCDS; Cotugno, 2009); and project-developed teacher and therapist rating scales (Boyd, Woodward, & Bodfish, 2011). Five studies utilized both direct and indirect measures of the target behavior [3, 13, 14, 19, 24].

Functional Assessment

Approximately half of the studies (n = 14) assessed for the function of the target behavior prior to implementing the intervention. Ten of these 14 studies used direct measures such as a functional analyses [6, 9, 10, 14, 19, 21, 22, 23, 25, 29], three studies [18, 26, 27] used a combination of direct and indirect measures, such as the Questions about Behavioral Function (QABF; Matson & Vollmer, 1995) rating scale, and one study (Mansdorf, 2013) only used indirect measures to determine the function of the H-RRB.

The reported functions of the H-RRBs varied across studies depending on the specific H-RRB, with attention seeming to be the most common function across all H-RRBs, followed by automatic reinforcement (nonsocial), and lastly escape/avoidance. Specifically, four studies examining the function of verbal perseverations about restricted

interests concluded that perseverative speech was maintained by attention [6, 10, 19, 21]. Two studies found that compulsive behaviors were maintained by automatic reinforcement for each of the participants [22, 29]. Neil and colleagues (2017) found that, for one participant, compulsive behaviors appeared to be maintained by both automatic reinforcement and access to attention. Other studies found that behaviors spanning the three categories of H-RRBs were largely maintained by automatic reinforcement [15, 26] and attention [26]. Szabo (2019) found that ritualistic and sameness behaviors were maintained by avoidance. Following a functional analysis, Kuhn and colleagues (2009) observed that undifferentiated rates of the ritualistic behavior (excessive straightening) occurred across all conditions; consequently, the authors were unable to identify a clear behavioral function of the target behavior. Two studies trained parents to conduct an FBA; however, neither study reported the specific function of the individuals' target behavior [14, 23]. Finally, Vause et al., (2020) used both direct and indirect measures to assess the function of participants' target behaviors, but did not report the specific functions of these behaviors. Of note, in their Discussion section, the authors anecdotally mentioned that their Fb-CBT intervention "treated compulsions that were maintained by escape from anxiety and other functions such as sensory stimulation and parental attention," [Vause et al., 2020, p.2385], though they did not identify the specific function for each participant in the Results section.

Intervention Procedures

Antecedent- and consequence-based interventions. The majority of studies (62%; *n* = 18) used a combination of antecedent- and consequence-based strategies in their interventions to reduce H-RRBs or promote behavioral flexibility. These strategies

included differential reinforcement [5, 10, 13, 14, 17, 18, 20, 23, 24, 25, 26, 27, 28], extinction [5, 9, 10, 18, 22, 25, 26, 27], teaching skills [14, 18, 23, 25, 26, 27], prompting [5, 8, 10, 20, 22, 28], providing choices [13, 14, 28], self-management [13, 17], environmental modifications [14, 24], functional communication training (FCT) [9, 23], redirection [8, 11], blocking [9, 22], abolishing operations components [11], and teaching trials with lag reinforcement schedules [16]. Twenty-four percent of studies (n = 7) only used consequence-based strategies in their interventions [2, 3, 6, 12, 15, 21, 29]. These consequence-based strategies included differential reinforcement of alternative behaviors (DRA) [6, 15, 21], extinction [3, 21, 29], differential reinforcement of variability (DRV) with response interruption and redirection (RIRD) [2], response blocking [15], and lag schedules of reinforcement [12]. Only a few studies (14%, n = 4) relied solely on antecedent-based strategies to reduce H-RRBs. These strategies included mindfulness training [1], social skills training [4], and noncontingent reinforcement [NCR; 19], as well as environmental modification, creating social routines, and building emotional reciprocity [20] .

Theoretical Framework. Seventy-two percent of studies (n = 21) were rooted in the principles of ABA and sought to promote behavioral change by applying the principles of operant conditioning and its related procedures. Twenty-one percent of studies (n = 6) were classified as CBT-based interventions [1, 3, 18, 25, 26, 27], some of which incorporated the use of ERP [3, 18, 26, 27] and CBT skills training (e.g., decisional balance, positive self-talk, and cognitive restructuring) into their interventions to reduce ritualistic and compulsive behaviors [18, 26, 27]. One study (Boon, 2017) used mindfulness training to reduce rigidity involving routines, need for sameness, and

inflexibility during transitions among adolescents with ASD. Finally, after noting that DRA and extinction were unsuccessful in decreasing inflexible behaviors in children with ASD when playing games, Szabo (2019) used "acceptance and commitment therapy" training (ACTraining) including defusion exercises, acceptance strategies, present moment awareness, and flexible self-perspective taking to promote behavioral flexibility during play.

Two studies were based on frameworks that fell outside the scope of traditional ABA or CBT [4, 7] interventions. Cotugno (2009) explored the effects of a social competence and social skills program based on a cognitive-developmental framework to address difficulties with transitions and flexibility in children with ASD. Gengoux and colleagues (2019) piloted a Developmental Reciprocity Treatment (DRT) parent-training program based on developmental treatment approaches, which rely on the use of stable social relationships and play, to treat compulsive and restricted behaviors in children with ASD.

Focused, comprehensive, and manualized interventions. The studies in this review were split almost evenly between two categories: FIPs and MTPs. Fifty-two percent of studies (n = 15) used one or more FIPs, and were classified as such. Forty-eight percent of studies (n = 14) were classified as MTPs, as they each used a manualized protocol to guide their intervention. Of note, four of the reviewed intervention studies targeted a broader array of ASD core features [4, 7, 20, 28] and/or their intervention sessions lasted longer than the typical 1-2 hours per week of most manualized treatment sessions [4-10 hours/week for study #20, one 4-hour plus five 30-minute sessions for study #25, 7 hours/week for 16 weeks for study #28]; however,

given that none of those four studies met Odom et. al's (2010) intensity/duration criteria to be classified as CTMs (i.e., 25-40 hours per week over 1-3 years), we categorized these studies as MTPs.

Twenty-one percent of the MTP studies (n = 3) utilized function-based CBT (Fb-CBT), a manualized intervention that combines adapted CBT (e.g., ERP) with ABA interventions (e.g., extinction, FCT, differential reinforcement) (Vause et al., 2018). Fb-CBT involved nine weekly, two-hour sessions designed to target compulsive and ritualistic behaviors in youth with ASD, and included the following components: 1) Psychoeducation and Mapping, 2) Individual treatment for OCBs using functional assessment and CBT, 3) Cognitive-behavioral skills training, ERP, and positive reinforcement, 4) Parent Training, and 5) Parent- and child-led intervention [18, 26, 27]. Boon's (2017) CBT-based mindfulness curriculum, which provided participants with psychoeducation about mindfulness, experiential practice of mindfulness, and opportunities to engage in journaling and group discussions about their practice, was also delivered over nine weeks, with sessions each lasting 2.5 hours. Szabo's (2019) ACTraining intervention was the shortest in duration compared to the other CBT-based MTP studies, as participants received four hours of ACTraining during one session, followed by five subsequent 30 minute training sessions.

Half of the MTP studies (n = 7) heavily emphasized parent training [2, 7, 8, 13, 14, 23, 28]. Two studies used PRT to address H-RRBs in children with ASD [13, 28]. Specifically, Lin and Koegel (2018) conducted a parent-implemented, manualized self-management intervention that incorporated elements of PRT (e.g., reinforcing attempts, providing child choice, task variation), which was delivered to parents during

60-minute sessions twice a week for 10-12 weeks. Ventola and colleagues (2016) used a more intensive approach, where parents of children with ASD received two hours of coaching per week and the child received five hours of direct intervention per week for 16 weeks to promote the child's social skills and reduce their H-RRBs. Boyd and colleagues (2010) developed Family-Implemented Treatment for Behavioral Inflexibility (FITBI), an intervention comprised of response interruption and redirection (RIRD) and DRV, which was delivered over the course of 12 weekly 60-120 minute sessions. Gengoux et al.'s (2019) DRT program, which taught parents to follow their child's lead during play, respond to their interaction attempts, and create predictable routines, was implemented during 12 weekly 90-minute sessions. Grahame and colleagues (2015) piloted a 2-hours-per-week Managing Repetitive Behaviours Programme (MRB) for eight weeks, in which the researchers delivered psychoeducation about RRBs to caregivers of children with ASD, helped them understand the function of their child's behavior, and taught caregivers specific techniques to reduce their child's rigidity and preoccupations [8]. In another study [23], parents received ten weekly, 90-minute sessions of the Family-based Management of Behavioral Excesses of Autism Program (FMBEAP), during which they were taught to identify the antecedents and consequences of their child's H-RRB and use structured play activities to reduce these symptoms. One study used a collaborative parent education program [14], where parents worked with the experimenter twice a week to identify the function of their child's H-RRBs and select intervention strategies to address the target behavior; additionally parents received feedback on their implementation of intervention strategies.

While the majority of MTP studies lasted between 9 and 16 weeks, two studies spanned out across a longer duration [4, 20]. Cotugno's (2009) social competence and social skills intervention, which targeted a variety of ASD symptoms, including flexibility, was delivered over 1-hour weekly sessions for 30 weeks. The longest, most intensive intervention was a low-intensity behavioral treatment (LIBT) program that also addressed a broad array of ASD symptoms, including flexibility, and was delivered over 4-10 hours per week for two years.

Study Design

Of the 29 studies, 20 studies used single-subject design (SSD) and 9 used a group design. Of the 20 studies using SSD, 95% (n = 19) employed an experimental design; one study was pre-experimental because the experimenter used an AB design [3]. Seven of the 19 studies employing an experimental design used a multiple baseline or multiple probe design [2, 13, 14, 15, 17, 18, 25], seven used a reversal or withdrawal design [5, 6, 9, 16, 19, 24, 29], and five used a multielement or alternating treatments design [10, 11, 12, 21, 22]. Of the nine studies using a group design, 33% (n = 3) were randomized controlled trials [8, 26, 27]. The remaining five group studies were quasi-experimental because they used pre-post measures without a control group [1, 4, 7, 20, 23, 28].

Outcome for H-RRBs

Single-subject designs. Table 2 reports the specific outcomes for each study. Results for the effect sizes of studies utilizing SSD are categorized as having a small effect if the NAP score ranges from 0 to .66, a medium effect if NAP ranges from .66 to .92, and a large effect if NAP ranges from .93 to 1.00 (per criteria from Parker & Vannest, 2009). Half of the 20 studies that utilized SSD (50%; n = 10) demonstrated

strong effects. Most of the SSD studies yielding strong effects used FIPs comprised of differential reinforcement with extinction [5, 10, 21], extinction [29], NCR [19], , provision of structured leisure activities with differential reinforcement of incompatible behaviors [DRI; 24], and self-monitoring with DRV [17]. The remaining three SSD studies yielding strong effects used MTPs including parent-implemented PRT with self-monitoring [13], functional behavior-based CBT (Fb-CBT) [18], and ACTtraining following a functional analysis [25].

Forty-five percent of SSD studies demonstrated medium effect sizes (n = 9). Each of these interventions, with the exception of one parent education program [14], were classified as FIPs which included: lag reinforcement schedules [12, 16], DRI [6], DRV with RIRD [2], DRA with response blocking [15], matched items with prompts and response blocking [22], FCT, extinction, and response blocking [9], and incorporating an abolishing operations component into a play intervention [11]. None of the SSD studies demonstrated small effects. For one study [3], NAP could not be calculated due to missing data; following an intervention that used ERP, the authors reported that the percentage of time the participants engaged in an academic task increased, as did the average latency time before the participants began engaging in H-RRBs.

Group Designs. For the nine studies that analyzed data at the group level, Hedges' g corrected for small sample bias was used to describe small (0.2), medium (0.5), and large (0.8) effects. Both of the MTP group studies that used Fb-CBT yielded large effects [26, 27]. An MTP that used PRT resulted in reductions in H-RRBs for all participants and produced medium effects [28]. Following an 8-week parent-group intervention, children with ASD showed significant improvements on parent ratings of

preoccupations with restricted interests as well as some improvements on their ratings of rigidity and insistence on sameness; however, improvements in rigidity and insistence on sameness were not statistically significant [8]. The remaining four group studies yielded small effects. These interventions were classified as MTPs and included: a school-based mindfulness intervention [1], a DRT parent-training program [7], LIBT for H-RRBs [20], and FMBEAP [23]. For one group MTP study [4], effect sizes could not be calculated due to missing data; however, parent ratings indicated significant improvement on flexibility with transitions following a 30-week social competence and social skills training intervention.

Function- vs. non-function based interventions. With respect to outcomes, effect sizes were larger overall for function-based SSD studies (mean NAP = 89.77) compared to SSD studies whose interventions were not based on function (mean NAP = 78.72). However, for Single Subject Design studies, the differences between Fx-based and non-fx-based interventions were not statistically significant (p = 0.7640). A similar pattern emerged for group design studies; on average, function-based group interventions yielded larger effect sizes (mean Hedges' g = 1.09) than group interventions that were not function-based (mean Hedges' g = 0.39). For Group Design studies, the differences between Fx-based and non-Fx-based interventions were statistically significant (p = 0.0146).

Intervention Procedures. Of the 12 studies that yielded strong effects, 75% (n = 9) used both antecedent- and consequence-based strategies in their interventions [5, 10, 13, 17, 18, 24, 25, 26, 27], the majority of which (67%; n = 6) were classified as MTPs [13, 18, 24, 25, 26, 27]. Seventeen percent of studies that yielded strong effects (n = 2)

used consequence-based intervention strategies only, both of which were FIPs [21, 29], and 8% (n = 1) used an antecedent FIP (NCR) [19]. One MTP study which demonstrated moderate to strong effects [23] incorporated both antecedent- and consequence-based strategies into its intervention.

Of the 11 studies that yielded moderate effects on the target behavior, 64% (n = 7) incorporated both antecedent- and consequence-based strategies into their interventions [8, 9, 11, 14, 16, 22, 28], the majority of which (57%; n = 4) were classified as MTPs [8, 9, 14, 28]. Thirty-six percent of studies that yielded moderate effects (n = 4) used consequence-based intervention strategies only [3, 6, 12, 15], the majority of which (75%; n = 3) were classified as FIPs [6, 12, 15].

Finally, of the three studies that yielded weak effects, 67% (n = 2) were classified as FIPs that relied solely on antecedent-based intervention strategies [1,7] and the remaining study was classified as an MTP that used both antecedent- and consequence-based strategies in its intervention [20] .

Theoretical Framework. Of the 12 intervention studies that yielded strong effects, 67% (n = 8) were rooted in the principles of ABA [5 10 13 17 19 21 24 29] and 33% (n = 4) were based on a CBT framework [18 25 26 27]. However, For both SSD and Group Design studies, the difference between ABA and CBT interventions were not statistically significant (p = 0.4789 and p = 0.2049, respectively). One study that yielded moderate to strong effects was rooted in ABA. All of the 11 studies that yielded moderate effects on the target behavior were rooted in ABA. Finally, of the three studies that yielded small effect sizes, one was based on a CBT framework [1], one was based on a

cognitive-developmental framework [4], and one was based on a developmental framework [7].

Certainty of Evidence

Twenty-eight percent of studies (*n* = 8) were classified as providing a *suggestive* level of certainty because they lacked sufficient baseline data [2], utilized an AB or pre-test/post-test design [3, 4, 7, 23, 28], failed to report inter-observer agreement data [17] or did not demonstrate treatment effects [20]. Thirty-one percent of studies (*n* = 9) were classified as providing a *preponderant* level of certainty [8, 9, 12 13, 14, 15, 16, 21, 24]. Within this category, four studies did not attempt to control for alternative explanations for reductions in the target behavior [13, 14, 15, 16]. The remaining studies in this category either demonstrated mixed effects for participants [12] or did not have a sufficient sample size [8, 9, 21, 24]. The majority of studies (41%; *n* = 12) were classified as providing a *conclusive* level of certainty [1, 5, 6, 10, 11, 18, 19, 22, 25**,** 26, 27, 29]. In addition to meeting the criteria of the suggestive and preponderant classifications, these studies controlled for alternative explanations of treatment effects.

Generalization and Maintenance

Of the 29 studies, 57% (*n* = 17) collected maintenance data. Maintenance was evaluated between a minimum of two weeks post-intervention [13, 29] and a maximum of 20 months post-intervention [18]. Overall, almost all of these 17 studies reported that improvements were maintained for all participants following treatment, with two exceptions [2, 24]. Boyd, Woodward, and Bodfish (2011) noted that maintenance effects were found for four out of five participants four weeks post-intervention [2]. Sigafoos

and colleagues (2009) found an increasing trend in object rearrangement at the 3-month follow-up, suggesting the possible need for booster sessions [24].

Thirty-one percent of the included studies (n = 9) collected generalization data. In all but one of these nine studies, intervention effects generalized across activities [13, 25], behaviors [14, 16], and people [3, 6, 22, 29]. One study found that the parent group intervention for managing RRBs did not generalize to the school setting and suggested that H-RRBs that occur in the home may differ from those that occur in school [8].

Discussion

This systematic review and meta-analysis summarizes 29 studies involving the use of behavioral and cognitive-behavioral interventions to reduce H-RRBs in individuals with ASD. Overall, the vast majority of studies were rooted in ABA (72%) with substantially fewer studies (21%) using CBT. In the majority of the studies, individuals with ASD demonstrated improvement in their target behavior, suggesting that behavior analytic and cognitive behavioral approaches to reducing H-RRBs are promising avenues to further investigate.

This review suggests that a functional assessment should be completed prior to implementing an intervention for H-RRBs. Almost two-thirds of the studies that used function-based interventions reported large effects, in contrast to approximately only a quarter of studies that used non-function-based interventions yielding large effects. Although the differences between mean effect sizes for function versus non-function-based interventions was not found to be statistically significant. Notably, qualitative analysis revealed that non-function based SSD studies were more likely to use consequence-based intervention procedures. This extends upon previous research that found consequence-based interventions to be effective for the treatment of lower-order RRBs irrespective of function. Nearly 88% of function-based interventions that produced large effects also provided a conclusive level of certainty of evidence, compared to just 25% of non-function-based interventions that reported large effects. This suggests that non-function-based interventions are also more likely to be lacking in methodological rigor and should be interpreted with caution. This review found eight studies that not only

produced large effect sizes on the target behavior but also provided a conclusive level of certainty of evidence.

The majority of studies within this category (and overall) incorporated a mix of antecedent- and consequence-based strategies into their interventions. The most effective mixed interventions, which also provided a conclusive level of certainty of evidence, included a function-based MTP that used CBT-skills training combined with DRA and extinction to treat compulsive and ritualistic/sameness behaviors [Fb-CBT; 18, 26, 27], a function-based Acceptance and Commitment Training (ACTraining) to decrease inflexible behaviors (25), and function-based FIPs rooted in ABA that used prompting combined with DRA and extinction to address verbal perseverations about restricted interests [5, 10]. Maintenance data suggests that these six studies produced lasting effects after the conclusion of the intervention.

Two additional FIPs rooted in ABA yielded large effect sizes and provided a conclusive level of certainty of evidence [19, 29]. In one study, a function-based antecedent intervention (NCR) was found to significantly reduce perseverative speech [19]. This finding is notable, as it contradicts the conclusion from Patterson et al.'s (2010) review that NCR alone was ineffective in reducing RRBs. However, it should be noted that in their review, NCR was used to reduce a lower-order RRB (i.e., face-rubbing) rather than a higher-order RRB, such as verbal perseverations. In another study, following a functional analysis, a consequence-based intervention comprised of response blocking and redirection, as well as extinction, significantly reduced participants' compulsive and ritualistic behaviors [29]. This finding supports those found in Mulligan et al. 's (2014)

review, which stated that consequence-based interventions that were function-based were effective.

Approximately half of the reviewed studies reported delivering interventions to individuals with ASD who had a comorbid diagnosis of ID. Of these 15 studies, only four yielded large effect sizes and provided a conclusive level of certainty. Notably, the majority of these four effective studies utilized Fb-CBT, a function-based MTP rooted in CBT that includes CBT skills training and ERP [18, 26, 27]. Moreover, intervention effects were maintained up to 20 month post-intervention, suggesting that Fb-CBT can produce positive and lasting effects on H-RRBs in individuals with ASD and comorbid ID. This is especially noteworthy, as Boyd et al., (2012) suggested that, given the emphasis in CBT on more cognitive elements, such as cognitive restructuring, CBT may be more appropriate for "individuals with intact cognitive abilities" (p.1242). However, it should be noted that the authors of the Fb-CBT studies targeting OCBs reported treatment modifications to traditional CBT for OCD (e.g., emphasis on visuals, choice of response modality, use of concrete/tangible exercises) and noted that the extent to which cognitive restructuring was used varied among participants, as it was dependent upon the identification of a distinct, interfering thought. Therefore, it is possible that the most effective type of CBT interventions for compulsive and ritualistic/sameness behaviors in individuals with ASD and comorbid ID is modified ERP, which would make sense, given that ERP is considered the first-line treatment for OCD symptoms (Rosa-Alcázar et al. 2015), which share many features with H-RRBs. Of note, an additional MTP intervention rooted in CBT demonstrated strong treatment effects that were sustained at nine weeks, generalized to novel people and activities, and provided a conclusive level of certainty

[19]; however, cognitive functioning was not reported. After function-based DRA plus extinction failed to produce marked progress in participants' behavioral flexibility, Szbao (2019) found that ACTraining, which included several cognitive components (e.g., defusion, present moment awareness, flexible perspective-taking), increased behavioral flexibility in children with ASD. However, given information regarding the participants' cognitive functioning was not provided, it is unclear if this treatment is effective for individuals with ASD who also have a diagnosis of ID.

With respect to target behaviors, the majority of the reviewed interventions targeted multiple subtypes of H-RRBs. Interventions that targeted at least one H-RRB in the ritualistic/sameness domain were the most effective, with 32% of these interventions yielding large effect sizes, compared to 27% of interventions targeting at least one H-RRB in the compulsive domain and 23% of interventions targeting at least one H-RRB in the restricted interests domain. Of note, interventions that were highly effective in treating ritualistic/sameness behaviors were less likely to be function-based (57%) compared to highly effective interventions for compulsive and restricted behaviors, all of which were function-based. The reason for this remains unclear, as there were no other notable differences between these studies, such as the setting or intervention provider, or treatment duration, except for the fact that interventions that were highly effective in treating compulsive and restricted behaviors were also more likely to treat individuals with comorbid ID; none of the highly effective interventions for treating ritualistic/sameness behaviors reported that their participants also had a comorbid diagnosis of ID.

Although parental reports in previous research indicate that their child's perseverative speech about restricted interests is one of the most difficult symptoms of ASD for them to manage (South et al., 2005), only a handful of the reviewed studies explicitly targeted this H-RRB. All interventions targeting verbal perseverations about restricted interests were function-based FIPs rooted in ABA, most of which used consequence-based strategies such as DRA/DRI and extinction [6, 12, 21] to treat the target behavior. Although each of these studies produced medium effect sizes, only one [6] provided a conclusive level of certainty. Two studies in which antecedent strategies were used in the intervention [10, 19] produced large effect sizes and provided a conclusive level of certainty, suggesting that interventions which incorporate antecedent strategies, such as prompting and NCR, may be even more effective than interventions which only use consequence-based strategies in treating verbal perseverations about restricted interests.

Strengths and Limitations of the Current Study

Overall, the findings of this study add to the limited body of research on behavioral interventions for H-RRBs. The principal strengths of this review include the use of a systematic search strategy employed across multiple databases, the use of three independent coders who not only determined which studies should be reviewed, but two of which also extracted the data, and the evaluation of certainty of evidence across studies. Additionally, review includes two unpublished dissertations whose findings would not have been known if the search had not included grey literature. Another noteworthy strength of this review is its focus on higher-order RRBs and underlying behavioral inflexibility; an area of research that has largely been understudied in the

Autism literature. By systematically reviewing and evaluating the quality of interventions for H-RRBs, this review provides clinicians with guidance on how to treat a core symptom of ASD.

In spite of the present review's strengths, it also has some limitations. First, in our search, we neglected to specify the terms "exposure and response prevention" and "exposure and ritual prevention." Although it is possible that articles containing these interventions would have been subsumed under the broader CBT category, it is possible that this oversight led to missed studies. Similarly, because our search was limited to studies written in English, other relevant contributions may have been omitted from this review. Another limitation of this review was the dichotomous coding of ABA and CBT studies. Several interventions that were categorized as CBT-based also included elements of ABA treatment, such as the use of functional analysis; therefore coding these studies as CBT may not have been an accurate reflection of the intervention procedures. Finally, treatment integrity was not coded in this review. Given that treatment integrity data provides useful information about the degree to which a treatment was implemented with accuracy and consistency and has been linked to treatment outcome, such information would have been helpful in comparing the efficacy of the reviewed interventions.

Future Directions

A secondary objective of this review was to encourage future research to further investigate the utility of behavioral interventions, including interventions beyond those offered by traditional ABA approaches, to treat H-RRBs. Several important implications for future directions emerged. Mulligan et al. (2014) previously noted that many presume repetitive behavior to be maintained by non-social reinforcement. However, this review

revealed that, while that is sometimes the case, H-RRBs can also serve the socially mediated functions of attention or escape. This finding suggests that it is particularly important to conduct an FBA before developing interventions to treat H-RRBs rather than just assuming the function is sensory/automatic.

Additionally, this review confirms previous research highlighting the paucity of studies on early childhood interventions for RRBs (Raulston & Machalicek, 2018). Given that ASD can be reliably diagnosed as early as age two and that H-RRBs have been found to persist and even worsen over time (Lam & Aman, 2007; Richler, Huerta, Bishop, & Lord, 2010), future research should examine the utility of these interventions with younger children. It is also worth noting that, although H-RRBs can significantly impact family functioning (South et al., 2005), only one quarter of studies in this review included parent training interventions to address H-RRBs. Although most of the studies that included a parent training component demonstrated moderate to strong effects that were maintained post-intervention, these studies were only able to provide a suggestive or preponderant level of certainty of evidence. As such, future research should continue to examine the role of caregivers in interventions for H-RRBs, as the incorporation of caregivers into treatment may be an important intervention component.

Despite being one of the most difficult symptoms of ASD for parents to cope with, interventions targeting verbal perseverations about restricted interest were the least studied in the intervention literature. Therefore, there is a need for future research to focus future intervention efforts on addressing this impairing behavior. Given that two studies in this review underscored the benefits of NCR and prompting on the reduction of perseverative speech [10, 19], these findings suggest that future studies targeting verbal

perseverations may benefit from incorporating antecedent strategies, such as NCR and prompting, into their interventions.

Although an ABA approach has historically been favored in the treatment of ASD core symptoms, half of the reviewed studies that both demonstrated strong effects on H-RRBs and provided a conclusive level of certainty were rooted in CBT, which has historically been understudied when exploring treatment options for individuals with ASD (Boyd et al., 2012). Therefore, future research should continue to explore the clinical utility of both individual and group CBT in treating H-RRBs.

As noted in other reviews of treatment for ASD core symptoms (Patterson et al., 2010), information on maintenance and generalizability of behavioral interventions to treat H-RRBs is lacking. Just over half of the reviewed studies collected information on the maintenance of intervention gains, and only a third of studies collected data on generalization. Given that H-RRBs can interfere with social, academic, and family functioning, future research should evaluate the conditions in which interventions address behavioral flexibility and their long-term effectiveness. Finally treatment integrity should be reported in future studies of H-RRBs to ensure that the obtained results are related to the actual implemented intervention, rather than the intervention as designed.

Implications for the Practice of School Psychology

Approximately one-third of the reviewed interventions (n = 10) took place in a school setting. The majority of these were FIPs rooted in ABA. Most of the school-based interventions yielded medium to large effect sizes and used a combination of antecedent- and consequence-based strategies including self-management, provision of structured leisure activities, differential reinforcement, and response blocking. However, only a third

of the school-based interventions provided a conclusive level of certainty [10, 11, 22]; therefore, the effectiveness of these interventions should be interpreted with caution. One function-based FIP that demonstrated large effect sizes and provided a conclusive level of certainty found that treatment effects on compulsive and ritualistic behaviors in students with ASD generalized to the classroom teacher. This finding suggests that school-based behavioral interventions can be successfully implemented by classroom teachers, although future research is needed to support this.

Of note, the majority of these school-based interventions were conducted at specialized or residential schools for students with developmental disabilities and/or disruptive behavior disorders. Given that, according to national data, the majority of students with ASD spend over 80% of their time in general education settings (US DOE, 2017), further research is needed to examine the utility of school-based behavioral interventions on the reduction of H-RRBs in mainstream classrooms.

Additionally, only three of the school-based interventions were based on a prior FBA [10, 15, 22]. It is possible that function-based treatments may be viewed as intensive and not feasible in school settings (Stormont et al., 2005). Given that this review found function-based interventions addressing H-RRBs to be more effective than non-function-based intervention, future research should examine the effectiveness of (and the possible barriers to) implementing function-based interventions to treat H-RRBs in the classroom.

Conclusion

In sum, the reviewed studies suggest some promising evidence for the treatment of H-RRBs in individuals with ASD using behavioral and cognitive-behavioral

interventions, particularly when both antecedent and consequence-based strategies are incorporated into the intervention. Moreover, interventions that were rooted in ABA and based on the hypothesized function of the behavior tended to yield large effect sizes compared to those that were not. Future research should investigate the efficacy of these interventions for children under the age of four. Additionally, more research on the effects of individual and group CBT in the reduction of H-RRBs is needed.

Appendix A

Description of Intervention Strategies to Manage RRBs

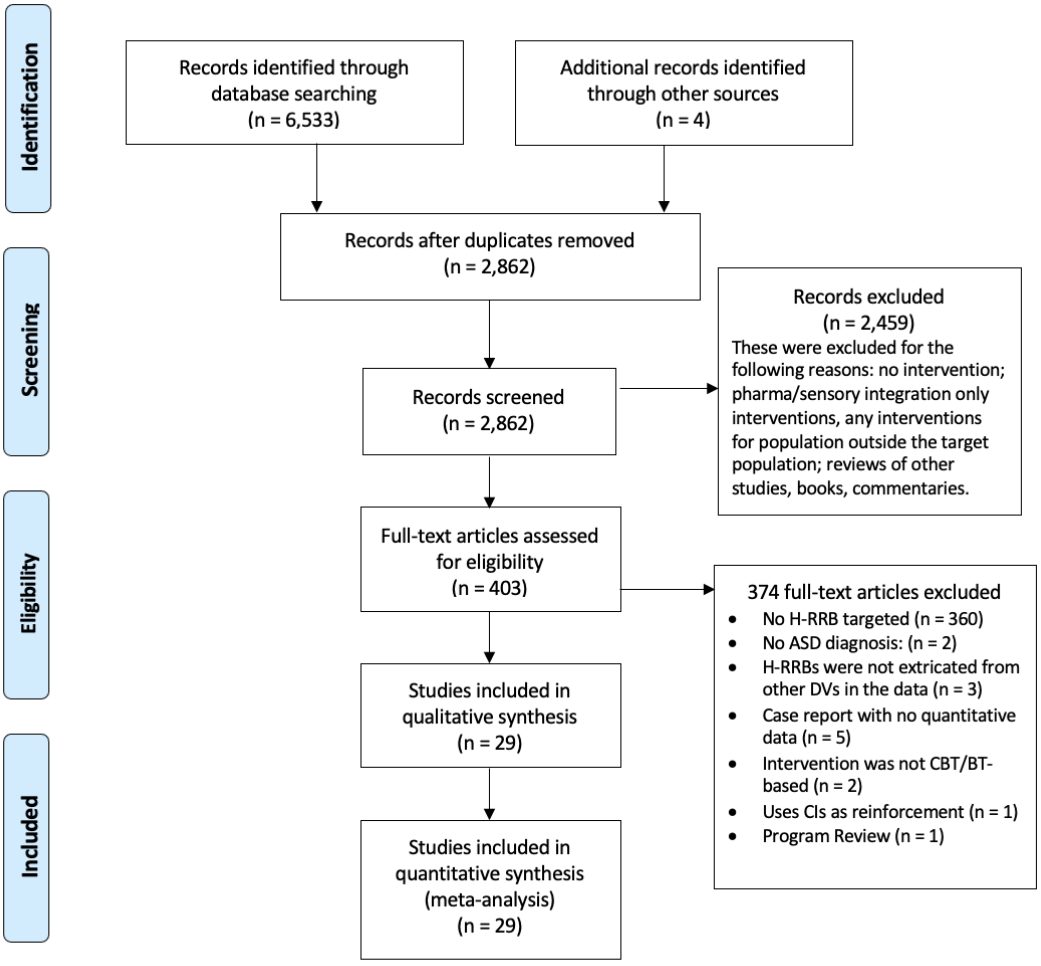
Strategy	Description
Behavioral interventions *Antecedent strategies*	
Video modeling	Individual watches a video of someone performing a desired behavior to eventually model the behavior
Visual schedules	Individual views a series of pictures/words that depict a sequence of events or activities
Noncontingent reinforcement (NCR)	Individual is given a reinforcer (independent of the occurrence of the target behavior) on a fixed-time schedule
Environmental enrichment	Individual is given non-contingent access to high-preference reinforcers, which compete with the hypothesized stimulation of the target behavior
Self-management	Individual is taught to monitor their behavior by recording the occurrence and absence of the target behavior
Discrimination training/stimulus control	Individual is taught to engage in a target behavior only in the presence of a specific stimulus
Skills teaching	Individual is taught adaptive play, leisure, or social interaction skills
Functional communication training (FCT)	Individual is taught appropriate communicative responses that can be used to obtain a desired reinforcer, rather than engaging in problem behavior

Consequence strategies	
Differential reinforcement	Individual is given reinforcement contingent upon: the absence of a target behavior (DRO); the presence of an alternative, appropriate behavior (DRA); occurrence of a behavior which in incompatible with the target behavior (DRI); engagement in varied/novel behavior (DRV)
Response cost	A reinforcer is removed when the target behavior occurs
Response interruption and redirection (RIRD)	When the target behavior occurs, the individual is interrupted and redirected to an alternative behavior
Extinction	Individual no longer receives reinforcement for a previously reinforced behavior
Cognitive-behavioral interventions	
Exposure and response prevention (ERP)	Individual is systematically introduced to a feared stimuli without engaging in anxiety-relieving behaviors

Adapted from Bodfish et al., 2012 and Mulligan et al., 2014

Appendix B

PRISMA Flowchart of Study Selection



Appendix C

Inclusion Criteria Reliability Form

Study ID	1. English publication	2. Peer-reviewed, data-based journal	3. Published between 1994-2019	4. Dx of Autistic Disorder, Asperger, PDD-NOS, or ASD?	5. Is it a behavioral or cognitive behavioral intervention?	6. Does it attempt to reduce an H-RRB or promote behavioral flexibility?

Table 1: Participant Characteristics and Setting of Selected Studies

Study	n	Age span (Mean)	Cognitive functioning
1. Boon, 2017	10	11-16 (14.875)	ID (none)
2. Boyd et al., 2011	5	3-5 (4)	VABS SS: 62-84 Chronological Age: 39-65 months
3. Boyd, Woodward, & Bodfish, 2011	5	5-11 (8.6)	ID IQ (severe)
4. Cotugno, 2009	18	7-11	ID (none)
5. Fisher et al., 2019	Experiment 1: 2 Experiment 2: 2	Experiment 1: 5, 7 Experiment 2: 4,5	NR
6. Fisher, Rodriguez, & Owen, 2013	1	14	NR
7. Gengoux et al., 2019	22	2-6 (3.7)	ID (severe)
8. Grahame et al., 2015	45 parents	3-7 (5.44)	ID (severe)
9. Kuhn et al., 2009	1	16	ID (moderate)
10. Kuntz et al., 2019	1	13	ID (mild)
11. Lang et al., 2010	2	5, 7	ID (severe)
12. Lee & Sturmey 2006	3	17-18	NR
13. Lin & Koegel, 2018	3	4-6	ID (unspecified)
14. Malmberg, 2007	6	4-10	NR
15. Mansdorf 2013	6	3-5	ID (mild)

16. Napolitano et al., 2010	6	6-10	NR
17. Newman, Reinecke, & Meinberg 2000	2	<5, 6	ID (unspecified)
18. Neil et al., 2017	1	11	ID (unspecified)
19. Noel & Rubow 2018	1	7	NR
20. Peters-Scheffer et al., 2013	40	3-7 (5.52)	ID (severe to none)
21. Rehfeldt & Chambers, 2003	1	23	ID (severe)
22. Rodriguez & Thompson, 2012	3	13-15	NR
23. Shiria et al., 2020	17 parents	2-4 (2.94)	ID (none)
24. Sigafoos et al., 2009	1	15	ID (none)
25. Szabo, 2019	3	8-10	NR
27. Vause et al., 2020	37	7-13 (9.92)	ID (unspecified)
28. Ventola et al., 2016	15	4-7 (6.11)	ID (moderate to severe)
29. Wolff, Hupp, & Symons, 2013	3	42-54	NR

Table 2: Descriptive Synthesis of Selected Studies

Study	Target behaviors	Intervention	Design
1. Boon, 2017	Rigidity involving routines, need for sameness, and inflexibility during transitions	*MTP*: Mindful moments for Teens	Multiple baseline across groups
2. Boyd et al., 2011	Perseverative interests, arranging and ordering, repetitive touching, hoarding object attachment	*MTP*: Family-Implemented Treatment for Behavioral Inflexibility (FITBI)	Multiple baseline across responses/settings
3. Boyd, Woodward, & Bodfish, 2011	Preoccupation with clocks, Nintendo game and Disney; completeness	*FIP*: ERP	Quasi-experimental pre-test post-test single subject design
4. Cotugno, 2009	Flexibility/transitions	*CTM*: Social Competency and Social Skills Training and Intervention Program	Quasi-experimental pretest post-test group design
5. Fisher et al., 2019	Tolerance of change (e.g., wearing a hat/ new bib using a new tray)	*FIP*: Prompting + DRA + extinction	Reversal
6. Fisher, Rodriguez, & Owen, 2013	Perseverative speech about comic book characters and violent topics	*FIP*: FA, DRI + extinction	Reversal
7. Gengoux et al., 2019	Unspecified compulsive and restrictive behavior	*CTM*: Developmental Reciprocity Treatment parent-training program	Quasi-experimental pre-test post-test
8. Grahame et al., 2015	rigidity, adherence to routine and insistence	*MTP*: Managing Repetitive Behaviors	Randomized controlled trial

	on sameness and preoccupation with restricted pattern of interest, limited play;	Parent Group Intervention	
9. Kuhn et al., 2009	straightening of trash and non-trash items	*FIP*: FA, FCT, extinction, and blocking	Reversal
10. Kuntz et al., 2019	Perseverative speech about historical and literary figures	*FIP*: FA; DRA + extinction + prompting	Multielement
11. Lang et al., 2010	Ordering/arranging, repetitive counting, repeatedly labeling toys	*FIP*: incorporated an AOC into a play intervention	Alternating treatments design with a baseline
12. Lee & Sturmey 2006	Response variability during social interactions	*FIP*: Implemented a lag 1 reinforcement schedule on varied social responding	Reversal and multielement
13. Lin & Koegel, 2018	Behavioral flexibility during play	*MTP*: Parent implemented PRT + SM	Non-concurrent multiple baseline across participants
14. Malmberg, 2007	Rigid & ritualistic behaviors and routines	*FIP*: FA; Parent Education Program	Multiple baseline across participants
15. Mansdorf, 2013	Ritualized play and eating; ordering and arranging	*FIP*: DRA + response blocking	Multiple baseline across behaviors
16. Napolitano et al., 2010	Response variability when creating a block structure	*FIP*: Teaching trials + lag reinforcement	Withdrawal
17. Newman, Reinecke, & Meinberg 2000	Ritualistic play with toy robot and ritualistic drawing	*FIP*: SM + DRV	Multiple baseline across participants
21. Rehfeldt & Chambers, 2003	Perseverative speech about a restricted interest (a) sirens or	*FIP*: FA; DRI + extinction	Multielement; reversal

	alarms, (b) dentist or doctor appointments, or (c) coughing		
22. Rodriguez & Thompson, 2012	Arranging and ordering, completeness, washing	*FIP*: FA; Matched item + prompts + response blocking	Multielement
23. Shiria et al., 2020	Unspecified behavioral excesses	*MTP*: FA; Family-based Management of Behavioral Excesses of Autism Program (FMBEAP)	Quasi-experimental pre-test post-test
24. Sigafoos et al., 2009	Object rearrangement	*FIP*: Provision of structured leisure activities + DRI	Withdrawal
25. Szabo, 2019	Inflexible behaviors related to changes in rules when playing games.	*MTP*: FA; ACTtraining	Non-concurrent multiple probe design across participants
26. Vause et al., 2017	Obsessive-compulsive behaviors	*MTP*: Function-Based CBT for OCBs in ASD	Randomized controlled trial
27. Vause et al., 2020	Obsessive-compulsive behaviors	*MTP*: Function-Based CBT for OCBs in ASD	Randomized controlled trial
28. Ventola et al., 2016	Unspecified H-RRBs	*CTM*: Pivotal Response Treatment	Quasi-experimental pre-test post-test
29. Wolff, Hupp, & Symons, 2013	Compulsions related to checking and touching; **r**itual involving fire alarm	*FIP:* FA; Response blocking and redirection, extinction	Withdrawal

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