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Restricted and Repetitive Behaviors in Autism Spectrum Disorders: A Review of Research in the Last Decade

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Restricted and repetitive behaviors (RRBs) are a core feature of autism spectrum disorders. They constitute a major barrier to learning and social adaptation, but research on their definition, cause, and capacity for change has been relatively neglected. The last decade of research has brought new measurement techniques that have improved the description of RRBs. Research has also identified distinctive subtypes of RRBs in autism spectrum disorders. Research on potential causal origins and immediate triggers for RRBs is still at an early stage. However, promising new ideas and evidence are emerging from neurobiology and developmental psychology that identify neural adaptation, lack of environmental stimulation, arousal, and adaptive functions as key factors for the onset and maintenance of RRBs. Further research is needed to understand how these factors interact with each other to create and sustain atypical levels of RRB. The literature indicates that RRBs have the potential to spontaneously reduce across time, and this is enhanced for those with increased age and cognitive and language ability. Research on interventions is sparse. Pharmacological treatments can be helpful in some children but have adverse side effects. Behavioral intervention methods provide the better intervention option with positive effects, but a more systematic and targeted approach is urgently needed. Evidence suggests that we will learn best from the last decade of research by taking a developmental perspective, by directing future research toward subtypes of RRBs, and by implementing early intervention targeted to improve RRBs before these behaviors become entrenched.

Keywords: restricted and repetitive behaviors (RRBs), autism spectrum disorders (ASDs)

The autistic child desires to live in a static world, a world in which no change is tolerated. The status quo must be maintained at all cost. Only the child himself may sometimes take it upon himself to modify existing combinations. But no one else may do so without arousing unhappiness and anger. It is remarkable the extent to which children will go to assure the perseveration of sameness. (Kanner, 1973, p. 63)

Most conspicuous in this respect were his stereotypic movements. He would suddenly start to beat rhythmically on his thighs, bang loudly on the table, hit the wall, hit another person or jump around the room.(Asperger, 1952/1991, p. 43)

From the first conceptualization of autism as a disorder, restricted, repetitive, and stereotyped behaviors (RRBs) have been

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included as a core feature of autism, along with the hallmark symptoms of social and communication problems (Asperger, 1944; Kanner, 1943). After more than 60 years, this view has not changed in essence. However the central role of RRBs in the diagnostic description of autism has begun to be challenged by some, and this raises implications at a practical and policy level for diagnosis, prevalence estimates, and intervention (Bishop & Lord, 2010). In this article we review the literature from the last decade related to restricted and repetitive behaviors in autism, including arguments for and against the centrality of its role, and we identify signposts for new directions for research and practice for the next 10 years.

Restricted and repetitive behaviors form a class of behaviors characterized by high frequency, repetition in an invariant manner, and desire for sameness in the environment (Kanner, 1943). Restrictedness is apparent in the narrowness of focus, inflexibility and perseveration in interests and activities, and insistence that aspects of the environment stay the same. Repetition is manifested in rhythmic motor stereotypies, repetitive speech, routines, and rituals. In the last decade, there has been a growth of research into both restrictedness and repetitive aspects of RRBs in autism and several salient points emerge. The first is that research developments have taken place across quite different fields of developmental psychology, cognitive psychology, neurobiology, and psychiatry, often in isolation from the work in other areas. The second is that expansion in empirical work, particularly in very specific

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methodological and descriptive areas has not been matched by advancement in theoretical explanation. In sum there is a lack of integration in research on RRBs in autism and a need for a broader focus that pulls together disparate research areas. This review article aims to bring together different approaches in order to move the field forward. Given the relatively early stage of research at the present time, the goal is to draw out the evidence from a range of research areas and to structure this around particular themes in a first step toward the development of a conceptual framework that will allow a set of predictions to be tested.

In this article we review literature across the last decade (from 1999 to the present). Computerized searches of MEDLINE, PsychINFO, PubMed and Web of Science databases were performed for the years 1999 to 2009 in order to find eligible studies. During the final stages of writing, a second search was carried out to trawl for all studies, including those published in 2010. Our search included a combination of the following terms: autism, Asperger's syndrome, pervasive developmental disorders, repetitive, stereotyped, ritualistic, restricted, self-stimulatory, behaviors. We did additional manual searching of relevant journals (Journal of Autism and Developmental Disorders, Autism, The Journal of Child Psychology and Psychiatry, Journal of Child Psychology and Psychiatry, Development and Psychopathology). We also searched reference lists of review articles and lists of publications of researchers working in this field. The search was limited to articles written in English.

As a result of this search and appraisal of the literature, three main themes regarding RRBs emerged: (a) definition, (b) cause, and (c) change. We therefore structured our review around these themes. Each theme encompasses a number of questions. With respect to definition, we ask questions that relate to classification and measurement and to the distinguishing characteristics of RRB in ASD. For example, "Are there subtypes of RRB? How should they be measured? Are RRBs different in children with autism from those found in children who do not have autism? What is their status as core or peripheral features in the definition of autism?" Our analysis of this literature revealed that in the last 10 years there have been considerable advances in the identification and categorization of RRBs and increased understanding of the relation between repetitive behaviors and other factors.

In comparison with the advances in knowledge about the definition of RRBs, we know very little about why RRBs happen and about their potential for change. So, with respect to the second major issue of cause, we asked the following questions: "What are the underlying neurobiological, developmental and cognitive influences which evoke RRBs? How do the environment and particular contexts trigger and maintain RRBs?" We were able to identify several theoretical frameworks that appear to offer promising analyses about both the distal origins of RRBs and the proximal causal factors that might trigger them. However, research that might test predictions within these frameworks is still in its infancy. As a result, knowledge about how and why RRBs might develop, change, and improve is very limited and little is known about effective interventions for RRB.

Therefore, with respect to the third issue of change, we asked a number of key questions relating to both the natural history of RRB and to intervention. For example, "What potential is there for RRBs to spontaneously change across time? How do pharmaco-

logical and behavioral interventions affect repetitive behavior outcomes?"

Throughout the review, we refer to RRBs in the light of developmental considerations. Since repetitive behaviors have long been considered to be important for enhancing muscular, neural, and cognitive development in typical infants (Gesell, Ames, & Ilg, 1974; Piaget, 1950/1952), and yet dysfunctional when seen in ASD and other disorders, we include evidence on their frequency and function in typically developing children and their changing patterns across age. We focus our review on child populations, although acknowledging studies with adult samples where these studies help to confirm or supplement evidence about RRBs in children.

Definition

In the last decade, most of the research effort has concentrated on the description of RRBs, helping to map out the conceptualization and measurement of RRBs in children with ASD. This is a worthwhile enterprise, as careful description should facilitate more effective research in areas of cause and treatment. Clear definitions can provide conceptual distinctions that differentiate subtypes of RRB and guide research on origins and outcomes. Clarity of definition and rigorous measurement will also assist the design of interventions that are targeted specifically toward improving RRBs. In this section, we review evidence on classification and measurement. We also discuss evidence regarding the distinguishing characteristics of RRBs in ASD compared with other disorders and the question of whether RRBs have a central and defining role or a peripheral role in the characterization of ASD.

Classification

What are restricted and repetitive behaviors (RRBs)? Research descriptions have been broadly guided by the classifications outlined by the Diagnostic and Statistical Manual of Mental Disorders (4th ed.; DSM-IV; American Psychiatric Association, 1994) and the International Classification of Diseases (10th rev.; (ICD-10; World Health Organization, 1993). These classification systems provide the diagnostic criteria for Pervasive Developmental Disorders, including the following conditions: autistic disorder, Asperger syndrome, atypical autism, and pervasive developmental disorder not otherwise specified (PDD-NOS). All of these conditions are commonly described under the heading of autism spectrum disorder (ASD). Restricted and repetitive behaviors as described by DSM-IV and ICD-10 subdivide into four subtypes: (a) preoccupation with restricted interests; (b) nonfunctional routines or rituals; (c) repetitive motor mannerisms (stereotypies); and (d) persistent preoccupation with parts of objects. In the autism literature, the two latter subtypes are often described together as lower level repetitive behaviors (Prior & Macmillan, 1973; Turner, 1999). These are considered to be more characteristic of younger and lower functioning children and are also found in children with intellectual disability and postencephalitic or other brain-based impairments. Most children with ASD show these kinds of repetitive and stereotyped motor behaviors at some point in their development, either with their own body parts, such as trunk and limb movements, hand and finger flicking, rocking, or tapping or with involvement of objects, such as spinning wheels, repetitive pouring of sand or water, and so forth. This raises another potentially important further subclassification, repetitive motor behaviors with and without objects.

Repetitive motor behaviors often have a strong sensory component, such as spinning objects or the self. This class of behaviors is therefore also categorized by some researchers as repetitive sensory and motor behaviors.1 This classification takes account of the sensory feedback that the repetitive actions are assumed to give the child, and it also implies a physiological basis that is driving the behavior (Lovaas, Newsom, & Hickman, 1987). However, it is important to note that repetitive motor behaviors do not by necessity include sensory feedback, as they can involve repetitive actions such as lining up objects or using objects in nonfunctional and invariant ways (stacking chairs for example). Likewise, atypical sensory behaviors are not universally repetitive (e.g., preference for bright, shiny objects or dislike of touch). However, sensory experiences such as overload may be a trigger for RRBs related to increase in arousal level. In order to understand the nature of the relation between sensory and repetitive behaviors, it is important to make a clear conceptual distinction. For example, while there is evidence that sensory features and repetitive behaviors cooccur (Boyd, McBee, Holtzclaw, Baranek, & Bodfish, 2009; Chen, Rodgers, & McConachie, 2009; Gabriels et al., 2008), it is possible that different types of sensory features are differentially associated with different varieties of repetitive behaviors. This is a question for future research.

The two other RRB subtypes described by DSM-IV and ICD-10, (a) preoccupation with restricted interests and (b) nonfunctional routines or rituals, are also often described together as "higher level behaviors" (Turner, 1999). Within this higher order category, routines and rituals represent insistence on sameness (IS; e.g., Szatmari et al., 2006). This is an original feature of autism documented by Kanner (1943) and also described as "the desire for sameness" (Prior & Macmillan, 1973). It includes inflexible adherence to specific routines or rituals, insistence on particular foods, wearing only certain items of clothing, and resistance to change in the environment. Like the repetitive sensory and motor behaviors, this factor emerges reliably from factor analytic studies (see Table 1). A second subtype within the higher order category involves intense interests or preoccupation with particular objects, activities, or information or with selected topics (e.g., the planets, football, collecting objects). These behaviors are also often described as "circumscribed interests" that are pursued obsessively and sometimes to the exclusion of all other activities (see Attwood,

While the subtype boundaries above are useful, it is easy to see that, except perhaps for the first category of stereotyped motor behaviors, there are permeable boundaries between RRB subtypes. For example, it is difficult to specifically categorize repetitive play actions with cars to the exclusion of all other interests, since this seems to be relevant to all subcategories. Phenotypic complexity forms a dimension running across all RRBs, from the most primitive repetitive body movements to highly sophisticated obsessive interests, such as discussing the detailed history of particular wars or encyclopedic knowledge of the planets. While not unequivocal, the literature indicates that lower level RRBs are more apparent in younger and more developmentally delayed cases and preoccupations, special interests, and obsessions more often found in older and more able cases with higher language and cognitive capacities

(Barrett, Prior, & Manjiviona, 2004; Bishop, Richler, & Lord, 2006; Esbensen, Seltzer, Lam, & Bodfish, 2009; Richler, Huerta, Bishop, & Lord, 2010). Nevertheless low-level RRBs continue to be seen in high functioning groups (South, Ozonoff, & Mahon, 2005). At the more elaborated level, some behaviors begin to shade over into similar features seen in Obsessive Compulsive Disorder (OCD), and this can sometimes lead to an OCD diagnosis in a child as an addition or an alternative to ASD (Zandt, Prior, & Kyrios, 2007).

Another crosscutting theme concerns the influence of communication competence in children in relation to type and severity of RRBs. For example, Barrett et al. (2004) found that the lower functioning children in their sample showed both the lowest level of pragmatic language skills and the most severe and frequent RRBs. This was particularly the case for data coming from teacher ratings of these children. Hus, Pickles, Cook, Risi, and Lord, (2007) described a complex set of relationships between repetitive sensory and motor actions, verbal IQ, and verbal and nonverbal communication, with the lower functioning group showing greater frequency of these low-level repetitive behaviors. However the IS factor was not related to these variables.

Of course, it is children with good language skills who are able to develop more sophisticated routines and interests about which they are likely to talk incessantly. Indeed, circumscribed interests involving particular expertise can sometimes be an asset, as older more able individuals find a niche in employment that makes good use of their special interests and expertise, for example high-level computer skills (see Attwood, 2003; Howlin, 2003). Related to this point is the symptom of repetitive vocalization and language, in particular repetitive questioning, that often does not feature in classifications and measurement of RRBs but is a hallmark feature of communication impairment in ASD. Repetitive questioning can serve a variety of purposes for the child, including a form of echolalia, an obsessive interest with a particular topic or event, an effort to keep a conversation going, or an expression of anxiety or lack of understanding and may differ in function from other classes of RRBs described above. The lack of clear understanding of triggers and functions of RRBs makes it hard to decide where repetitive questioning fits in to the overall picture.

To summarize, there are different types of RRBs that vary in complexity and sophistication. These are often described as high-and low-level repetitive behaviors. Although these behavioral classes differ in their form, there are overlaps between them and, importantly, developmental factors of cognitive and communicative level appear to be strong influences on these different forms.

Measurement

The conceptualization of RRBs has been highly influenced by the types of measurement tools used, as these set the boundaries for the phenomena under focus. A large range of methods has been developed, including questionnaires completed by parents and/or teachers, either concurrently or retrospectively; interviews with

¹ We use the term "repetitive sensory and motor behaviors" rather than the term repetitive "sensorimotor behaviors," which is often used in the literature on repetitive behaviors, in order to distinguish sensory and motor behavior types from each other and from the more specific reference to sensorimotor behaviors originally defined by Piaget (1950/1952).

parents/caretakers; structured and unstructured observations of children; the use of videotaped material from children's early life; and reported case studies. Different forms of method highlight different types and qualities of repetitive behavior. For example, laboratory-based observation studies focus on stereotyped behaviors and tend to exclude obsessive compulsive-like behaviors that are more accurately reported by parents and teachers (Barrett, Prior, & Manjiviona, 2004; Zandt, Prior, & Kyrios, 2007). On the other hand, observation studies use detailed measures of duration and frequency that are not available in informant reports of severity. Even within particular forms of methodology, there are remarkable differences in the behaviors measured, such as in the relative focus on compulsive-like versus motor behaviors or inclusion of self-injurious behaviors or in the form of scaling used (e.g., frequency versus duration).

Currently there has been no systematic attempt to examine consistency of results across different forms of measurement (observation, interview, questionnaire) or within methods, such as different versions of interview or from different sources, including child, parent, or teacher reports. This is important to do as some research using standardized interview methods, such as the Autism Diagnostic Interview-Revised (ADI-R), or observation methods, such as the Autism Diagnostic Observation Schedule (ADOS), show inconsistent results for RRBs over time (Charman, Taylor, Drew, Cockerill, Brown, & Baird, 2005; Lord, Risi, DiLavore, Shulman, Thurm, & Pickles, 2006). In addition, research has not yet adequately examined the comparative validity and reliability of particular items measured or assessment of the overall psychometric integrity of measuring instruments. A detailed and systematic analysis of methodology is greatly needed. This work is the subject of another ongoing review article (Honey, Rodgers, & McConachie, in press) and therefore we simply summarize some of the more commonly used measures below and in Table 1. In Table 1, we have selected studies that include analyses of the relation between RRB and developmental variables such as chronological or mental age, given that in our scrutiny of the literature, developmental level emerged as an important factor across different studies. We also separately classified the main purpose of each study according to three different goals: classification (e.g., factor analysis grouping); distinguishing features, (identification of features that differentiate ASD), and developmental change, (e.g., change in RRBs specifically measured within a longitudinal or cross-sectional design).

Parent Report Methods

Parent interviews. Clinical interview methods collect information on repetitive behaviors, social interaction and communication. One commonly used clinical interview, the Diagnostic Interview for Social Communication Disorders (DISCO; Wing, Leekam, Libby, Gould, & Larcombe, 2002) includes a total of 47 items with separate sections on stereotypies, routines, circumscribed interests, repetitive speech, and sensory features. In contrast, the most popular parent interview method used for research on RRBs is The ADI-R (Lord, Rutter, & Le Couteur, 1994), which relies on 12 items only. The inclusion of different numbers, types, and weighting of items introduces further potential for variable results and, consequently, for differing interpretations of the significance of different behaviors. For example, factor analytic stud-

ies using parent interview data have been exclusively based on the ADI-R (see Table 1).

Most factor analytic studies report two factors that correspond with the higher level and lower level classes described earlier. These are (a) repetitive sensory motor behaviors and (b) insistence on sameness. These factor analytic studies are detailed in Table 1 and demonstrate that although the boundary between the groupings is not entirely distinct, it is broadly distinguished by the quality of rigidity and dislike of change in the insistence on sameness factor and by motor rhythmicity and atypical sensory responses in the repetitive motor and sensory factor. Repetitive sensory and motor behaviors are especially common and often severe in ASD as well as in developmentally delayed groups, even where the latter groups do not have the social impairments of autism. The two-factor structure has been replicated in at least one study reporting on a non-English speaking sample (Papageorgiou, Georgiades, & Mavreas, 2008). Not all research studies have found evidence for two factors using ADI-R data however, and there are several reports of three factors emerging from factor analysis (Honey, McConachie, Randle, Shearer, & Le Couteur, 2008; Lam, Bodfish, & Piven, 2008). The age of different samples may explain these variable findings. In addition, different studies also use different numbers of ADI-R items which may account for the varying results (see Table 1, subsection on Classification, for details of age and number of items).

Parent questionnaires. A number of specialized parent questionnaires have been designed to target solely repetitive behaviors. Unlike the measures above, these questionnaires are not usually part of a diagnostic assessment battery. These questionnaires include many of the same items as found in ADI-R but offer a larger and more differentiated set of items. Questionnaires also differ from each other in terms of the balance of items measuring motor, sensory, or compulsive-like behaviors. Of these, the most commonly used over the last 10 years are seen in Table 1. These are (a) the Repetitive Behaviors Scale (Bodfish, Symons, & Lewis, 1999) with five dimensions of stereotyped, self-injurious, and compulsive behaviors, plus ritualistic/sameness and restricted interests dimensions; (b) the Childhood Routines Inventory (CRI; Evans et al., 1997) with two factors, "just right" and "repetitive activities"; and (c) the 33-item Repetitive Behavior Questionnaire (RBQ; Turner, 1995), which also appears in a 55-item interview version (Turner, 1995). The RBQ was recently revised into the 20-item RBQ-2 (Leekam et al., 2007). This measure selects items from the Diagnostic Interview for Social and Communication Disorder (DISCO; Wing et al., 2002) that match most closely with corresponding items from the original RBQ. Although all items in the RBO-2 were designed for children with ASD and the items in the DISCO have been used extensively in this population, so far this questionnaire has been published only with a typical child population at 15 months of age and 24 months of age (Leekam et al., 2007). In children aged 24 months, four factors resembling the DSM-IV classification were found in factor analytic research (motor, sensory, rigidity/routines, restricted interests) and these combined into two clusters ("sensory and motor" and "insistence on sameness"). For children at 15 months, the data were best described in terms of the two groupings only (Arnott et al., 2010).

In the last 2 years, a number of other new measures have appeared in the literature. These include another questionnaire coincidentally also named the Repetitive Behavior Questionnaire

(RBQ; Moss & Oliver, 2008) but which is completely unconnected to Turner's (1995) RBQ and with the later RBQ-2 (Leekam et al., 2007). Moss and Oliver's separately named RBQ has been used to examine RRBs in a number of genetic syndromes. In addition, other questionnaire and interview methods have appeared that are adapted for very young children (Matson, Dempsey, & Fodstad, 2009). Results of studies using these questionnaires are described later in this section.

Observation methods. Observation methods come in both structured and unstructured forms. The ADOS-Generic (ADOS-G; Lord et al., 2000) is a semistructured observation tool used for the diagnosis of ASD. RRBs that are seen during the assessment are recorded by a trained observer and RRB items are recorded. Although early research studies did not show differences between autism and some neurodevelopmental comparison groups (Lord et al., 2006; Ozonoff, South, & Miller, 2000), in other studies differences have been found. For example, recent research using versions of the ADOS designed for prelinguistic children (PL-ADOS; DiLavore, Lord, & Rutter, 1995) and toddlers (ADOS-T; Lord, Luyster, Gotham, & Guthrie, 2010), has shown increased prevalence and severity of RRBs in children with ASD compared with those who had nonspectrum disorders (i.e., those with language impairment or mild intellectual disability) or typical development (Kim & Lord, 2010).

Other investigations (Ozonoff, Macari, Young, Goldring, Thompson, & Rogers, 2008) of RRBs in young children at risk for ASD have used newly developed structured observation instruments, such as the Autism Observation Scale for Infants (AOSI; Bryson, Zwaigenbaum, McDermott, Rombough, & Brian, 2008) and the Communication and Symbolic Behavior Scales Developmental Profile (CSBS-DP; Wetherby & Prizant, 2002; for review see Yirmiya & Charman, 2010). These observation methods have elicited some striking results, indicating that repetitive behaviors, particularly those involving motor stereotypies and sensory responses to objects may be one of the first places to look for early markers of ASD in the second year of life (Rogers, 2009).

Observation methods have advantages but they also have disadvantages. They carry challenges of diversity of observation contexts and of behavior sampling and scoring methods (direct, indirect, structured, semi structured, unstructured). The difficulties inherent in this methodology were illustrated in a study by Gardenier, MacDonald, and Green (2004), who observed proportions of observation periods containing stereotypy. They compared 10-s partial-interval recording (PIR) estimates and momentary time sampling (MTS) estimates (using 10-, 20-, and 30-s intervals) of the actual durations of stereotypic behavior in young children with ASD. The partial-interval recording method overestimated the relative duration of stereotypy, while momentary time sampling both over and underestimated the relative duration of stereotypy. This study highlights the method dependence of findings using these kinds of sampling and measuring techniques. Variability and reliability can be especially problematic when observations are not tightly structured and standardized. Recently developed systematic observational methods that screen for developmental abnormalities, such as the CSBS-DP and AOSI, mentioned above, hold promise for eliciting more valid data because these include agreed operational definitions of specific behaviors.

There are other methodological considerations to take into account when evaluating studies of RRB. One is the fact that a

number of studies rely on retrospective clinical records in deriving both diagnostic and behavioral data on children and their RRBs, especially when there are long time intervals between initial measurement and later data mining (e.g., Carcani-Rathwell, Rabe-Hasketh, & Santosh, 2006). Many aspects of the study of autism have changed over time, including diagnostic criteria; hence conclusions must be cautious with retrospective data. Age specificity for symptoms and behaviors is also an issue. For example, the current *DSM-IV* criterion relating to RRBs of inflexible adherence to routines is not emphasized in early identification of autism because it is difficult to measure in infants. Likewise a number of rituals and routines are difficult to measure by time-limited observation since they need longer time frames to qualify as rituals and routines.

While progress is evident in developing measurement tools, more replication is needed as well as comparison across methods (e.g., RBQ, AOSI, DISCO, ADI, etc.). Probably the most important issue for future research is to reach a consensus on the operationalization of interviewer, questionnaire, and observational measures. Currently the ADI-R has been the most widely used measure, but this measure is not comprehensive. Observation measures on the other hand may yield different results and focus on different behaviors. Such agreement on operational measurements could lead to greater accuracy in comparisons across studies. Meanwhile, as parent report and observations represent such different ways of recording repetitive behaviors, researchers should be wary of mixing together results from these different forms of method unless a concerted comparison is the aim of the study being reported.

Distinctiveness

Are there distinguishing characteristics of RRBs in ASD? One empirical issue concerns the distinctiveness in the topography of RRBs in children with ASD in comparison with children who do not have autism. A different nosological issue relates to the status of RRB symptoms as defining, core symptoms for ASD rather than as peripheral, noncore features. We discuss both issues below.

RRBs in ASD compared with other clinical groups. stricted and repetitive behaviors are not only found in ASD. They are also found in a range of other disorders such as Tourette Syndrome, Fragile X, Rett's Disorder, Parkinson's Disease, Obsessive Compulsive Disorder, Down's Syndrome, dementia, deafness, blindness, schizophrenia, and intellectual disabilities. It is evident from literature describing the topography of RRBs that these behaviors occur at higher frequencies in ASD than in other disorders (see Matson, Dempsey, & Fodstad, 2009, for a recent review). However, the question of the distinctiveness of RRBs in ASD compared with other clinical groups in terms of their form, pattern, frequency, or intensity requires further examination. Probably the clearest finding coming from comparative studies relates to stereotyped motor and sensory behaviors that are not distinctive to ASD but are part of the behavioral pattern seen in children with developmental delay or intellectual disability and have been reported in a number of childhood disorders. Using the RBQ (a new questionnaire not related to the original Turner, 1995, RBQ or the more recent RBQ-2, Leekam et al., 2007), Moss, Oliver, Arron, Burbridge, and Berg (2009) examined the prevalence and phenomenology of RRBs in the genetic syndromes of Angleman, Cornelia de Lange, Fragile-X, Cri du Chat, Lowe, Prader-Willi, and Smith-Magenis, all of which are associated with intellectual disability. Motor stereotypies were common across all syndromes. Within these disorders significant positive associations were found between ASD symptoms as assessed by the Autism Screening Questionnaire (ASQ; Moss, 2005) and RRBs in Fragile X and Cri du Chat syndromes.

When higher level insistence on sameness-related RRBs are examined, they are also commonly reported across other childhood disorders, including anxiety disorders. The common features of compulsive behavior, obsessions, sameness, and repetitive speech, show more variability across genetic syndromes (Moss et al., 2009), where IQs are often low. In a study using the CRI (Evans et al., 1997), high levels of RRBs were found in both Prader-Willi syndrome and ASD groups; and, when developmental level was controlled, the two groups did not differ in overall frequency. A few differences in frequency of type were found within insistence on sameness behaviors, as Prader-Willi children were more likely to collect or hoard objects while those with ASD showed more lining up of objects, specific food preferences, and attention to detail (Greaves, Prince, Evans, & Charman, 2006). One study comparing higher functioning groups of children and adolescents with OCD and with ASD (Zandt, Prior, & Kyrios, 2007) found that insistence on sameness behaviors and repetitive motor behaviors, as well as total RRBs were similar in these groups and both groups showed significantly more RRBs than control typical children. However the OCD group had higher frequencies of obsessions and compulsions than the ASD group. In cases where ASD children did exhibit compulsions, these were less sophisticated in nature than those in OCD cases. In a sample of low functioning children, Hus et al. (2007) found that while repetitive sensory motor actions were more common in low-functioning children, IS was relatively independent of gender, age, IQ, and symptom domains, as assessed via the ADI-R and the ADOS (see also Bishop, Richler, & Lord, 2006), perhaps questioning claims of lower level + higher level RRBs as a function of age and developmental level factors.

There has also been exploration of differences within the ASD category itself. South, Ozonoff, and McMahon (2005) compared RRBs in 8- to 20-year-olds with either high functioning autism (HFA) or Asperger's syndrome but found no significant age differences between these groups, possibly because of their higher functioning status. More recently, Matson and colleagues (2009), examined subtypes of RRBs in very young children aged 17-37 months by comparing children with autism, PDD-NOS, or a non-ASD medical condition likely to result in a developmental delay. The highest rate of RRBs occurred in the autism group, followed by PDD-NOS and atypical groups. The latter was the least likely to show RRBs (this is not surprising as the presence of RRBs forms part of differential diagnosis decisions for autism). Findings suggested that such behaviors can be identified very early in autism, especially limited interests, repetitive motor movements, and eye gaze abnormalities, reinforcing the desirability of early identification and intervention aimed at modifying these challenging behaviors. However, we know that cognitive capacities can exert an influence on symptomatology (e.g., Lam & Aman, 2007; Hus et al., 2007) and are likely to moderate age effects. Thus, older and higher functioning children show more circumscribed interests and preoccupations, and lower functioning

children show more repetitive sensory and motor behaviors (South, Ozonoff, & McMahon, 2005).

The picture emerging from most studies comparing RRBs in ASD and other clinical groups is that it is the frequency of RRBs rather than their systematic form or pattern that mark any distinction between ASD and other clinical groups (Bodfish, Symons, Parker, & Lewis, 2000). RRBs in ASD appear to be distributed across a wide range of behaviors, whereas more specific RRBs are seen in other groups, for example, obsessions and compulsions in OCD or hoarding in PW. The distinctive difference in RRB frequency in ASD however is strongly related to developmental level, with repetitive stereotyped and sensory behaviors being more frequent in younger children and those with intellectual disability (Richler, Bishop, Kleinke, & Lord, 2007; Lam & Aman, 2007; Militerni, Bravaccio, Falco, Fico, & Palermo, 2002). The results of factor analytic and other studies that use the ADI-R highlight this effect of developmental level and IQ on RRBs (see Table 1) and the point will be further emphasized when considering the potential for change in RRBs across time, discussed later in this article.

Distinctiveness of RRB as a core impairment. have considered distinguishing features of ASD in terms of form, frequency, and age and level of functioning differences. Another way to evaluate the distinctiveness of RRB in ASD is in terms of their significance as a core impairment required for a diagnosis of ASD (DSM IV, ICD-10). Traditionally, RRBs have been part of the constellation of impairments constituting ASD and are assumed to be a defined behavioral dimension that is reliably related to the social and communicative deficits. However, some researchers have questioned this assumption. For example, Constantino, Gruber, Davis, Hayes, Passanante, & Przybeck (2004) assessed the factor structures of both the ADI-R and their own Social Responsiveness Scale across a range of child psychiatric disorders and found, for both instruments, a single continuously distributed underlying (unitary) factor rather than the traditional three domains of social, communication, and repetitive behaviors, leading them to query the concept of separable domains.

In contrast to the view of a single dimension, Happé, Ronald, and Plomin (2006) and Happé and Ronald (2008) argue for independence or "fractionation" of social and nonsocial (RRB) symptoms of ASD, suggesting that while these different impairments may be coincidentally associated in some children, they have distinctive genetic etiologies. Support for this claim has been based in part on the results of a large twin study (Ronald, Happé, & Plomin, 2005; Ronald, Happé, Price, Baron-Cohen, & Plomin, 2006). The study was representative of the whole population with relatively few children with ASD included. In this study, 7- and 8-year-old children were rated by parents and teachers on their social, communication, and RRB traits. The results showed that these traits were only modestly correlated. Model fitting analyses that compared the traits across twins and also looked at extreme traits, found that the genetic influences on social traits were to some extent independent of the genetic influence on communication and/or RRBs. When assessed as traits in the normal population therefore, it appears that while there is some overlap between the symptoms phenotypically, RRBs can also dissociate from other social and communication symptoms.

While the results of this research suggest that social and nonsocial symptoms may be separate dimensions with different genetic underpinnings, Mandy and Skuse (2008) argue that existing research evidence does not yet directly test the claim for independence of three sets of autistic traits. Mandy and Skuse (2008) make a number of recommendations for further research, including the need to separate out measures of different types of high- and low-level RRBs to examine nonsocial traits alongside social traits and close examination of the social impairment traits in atypical autism and PDD-NOS, where RRBs are much less marked. On the other hand, evidence presented by Happé and Ronald (2008) makes a persuasive case that investigation of specific areas of the triad rather than its cooccurrence will offer a clearer way forward for diagnosis and research. For example, molecular genetic evidence points to symptom-specific genetic effects related to high levels of RRBs (Alarcón, Cantor, Lui, Gilliam, & Geschwind, 2002; Shao et al., 2003; Sutcliffe et al., 2005), while evidence from a new genome-wide association study has initially identified different single nucleotide polymorphisms (SNPs) that are separately associated with social and nonsocial traits in a general population sample (Ronald et al., 2010). Nevertheless, research that supports the case for a distinct fractionation of the autistic phenotype is still at an early stage. Much of the research on the behavioral phenotype relates to extreme traits in the typical general population, which includes samples at a single age of 8 years old, and uses specific measurement tools with limited psychometric properties. More research is needed to decide whether the fractionation view is the appropriate approach to take toward the triad of social, communication, and repetitive behavior impairments in autism.

The discussion raised by Mandy and Skuse (2008) and Happé and colleagues is relevant to issues that we raise later in our review regarding the causes of RRBs and their capacity for change. While autism features, including RRBs, are known to show continuity in the normal population, the developmental pattern of these behaviors may differ from that seen in children with ASD, who generally have significant levels of developmental delay.

Although RRBs in ASD have been compared with other samples, these tend to be syndromes where there is intellectual disability (e.g., Angelman syndrome, Fragile X), but there are no matched groups of typically developing children. With respect to RRBs in typical populations, these behaviors are known to reduce by 4 years of age (Evans et al., 1997) and to show much reduced frequency in comparison to high functioning children with ASD of the same age (South et al., 2005).

While further research evidence in this area is required, it is worth noting that restricted and repetitive behaviors (RRBs) are reliable predictors of a stable diagnosis between age 2 and 9 years (Lord & Luyster, 2006; Lord et al., 2006) and that all of the major instruments used in the diagnosis of ASD include RRB items. We believe that several questions need to be answered before the importance of the association between RRBs and socialcommunication symptoms is dismissed. The first relates to neurobiological evidence from animal models, discussed in a later section of this article, indicating that social and/or sensory isolation contributes to RRBs. The question of whether a similar association can be inferred beyond animal models to children with ASD remains to be established (but see Rutter et al., 1999, on findings of autism and autistic like traits in Romanian orphans). The second closely related question is whether experimental interventions that are designed specifically to improve social impairments will have an additional effect on improving RRBs. The evidence so far is suggestive (Loftin, Odom, & Lantz, 2008). Until

these questions are answered, we will continue to believe that RRBs play a central role to ASD with importance for development and with impact for other aspects of functioning.

Summary

Our review of the literature on the definition of RRBs aimed to answer questions about what RRBs look like, whether there are subtypes, how they can be measured, and how distinctive they are. Although we still do not have a comprehensive taxonomy of RRBs, the evidence suggests that RRBs form a structure that resembles *DSM–IV* subcategories and which further subdivides these into two classes of low-level and higher level RRBs. These classes of behaviors are also seen in other clinical groups and show continuity into nonclinical populations. In terms of distinctiveness of topography, the evidence shows that while RRBs in ASD may be more frequent, there is no distinctive RRB marker for ASD.

There is a wide range of different measurement tools for RRBs. The design and use of measurement tools has a circular effect on the way that RRBs have been conceptualized because the measurement instruments themselves (e.g., ADI-R) are clinical instruments and therefore clinical features in the diagnosis are being redescribed, restricting the range of possible phenomena under study. Therefore, to further advance our understanding of the definition of RRBs in ASD, careful consolidation of the diverse range of measurement instruments and their findings is greatly needed. Robust measurement for RRB needs to be applied across all types of research, including measures used for intervention, given the lack of attention to RRB-specific measures in intervention studies. A shared effort by researchers to compare across measurement tools will be an important step for the field.

The problem of circularity described above may be partially addressed by examining research that draws from wider populations, such as early screening studies that do not specifically focus on RRBs. Screening studies with large-scale normative populations have shown that social and communication characteristics, rather than RRBs, provide the first key indicators of ASD (Cox et al., 1999; Robins, Fein, Barton, & Green, 2001). A recent study that investigated agreement among four widely used diagnostic measures for toddlers found that many children in fact missed receiving a diagnosis on the ADI-R compared with other instruments because the ADI-R criteria require RRBs while some other instruments did not (Ventola et al., 2006). However, these findings should be reappraised given recent observational research evidence indicating that we need to look more closely at particular types of RRBs in infancy (Ozonoff et al., 2008). Whether or not RRBs are a key indicator of ASD early in life compared with social and communication features, there is evidence even from ADI-R studies that RRBs provide a stable predictor of ASD as children get older (Lord & Luyster, 2006; Lord et al., 2006). These findings speak to the debate about the distinctiveness of RRB as a core impairment and also the need to take account of developmental change in RRBs.

Several other important issues remain to be settled about the significance of associations between RRBs and other symptoms. One concerns the role of sensory symptoms that affect all sensory modalities, such as hyper- or hyposensitivity in vision, hearing, and touch. Only some of these sensory symptoms are described under the subtype of repetitive sensory and motor behaviors and

the majority are not included under the *DSM-IV* criteria. A key question is the role that sensory symptoms play in arousal, since as we discuss later, arousal may be a key feature in the manifestation of repetitive behaviors. There is little recent research on this, although progress on the link between arousal and sensory sensitivity is now being made. For example, Schoen, Miller, Brett-Green, & Nielsen, (2009) report atypical physiological arousal in children with ASD compared with children with Sensory Modulation Disorder (SMD), which is a subtype of Sensory Processing Disorder (SPD). SPD is diagnosed when sensory processing impairments are found in the absence of other disorders (Miller, Anzalone, Lane, Cermak, & Osten, 2007) and sensory modulation disorder is marked by difficulty in regulating sensory responding.

Another issue concerns the association between RRBs and social-communication symptoms referred to earlier. One proposal is that the genetic etiology of the social impairment in ASD itself may be distinctive from that for the genetic underpinning for RRBs. Currently we do not have sufficient evidence about the separateness of the genetic etiologies of these two symptom clusters, as they arise over time. Since both RRBs and social-communication impairment create particular types of physical, emotional, and social environments for the child however, the codependence of these symptoms may be important to study even if it turns out that there is independence in their etiology.

Finally, further work is needed on other factors that may influence the association between repetitive behaviors and social communication impairment. A prime candidate is imagination. Wing and Gould's (1979) original triad of features of autism, identifies two inversely related activities—impairments in imagination cooccurring with restricted, repetitive behaviors. A view of the triad of impairments, in which impaired imagination is linked with repetitive behavior has provided the descriptive basis for theoretical accounts of autism for many years (see Frith, 2003; Happé, 1994). Yet empirical research on this relation has scarcely been reported in the literature (Honey, Leekam, Turner, & McConachie, 2007). Further research is needed that encompasses samples of children with typical development and those with genetic and psychiatric disorders, as well as those with ASD, with special attention paid to developmental factors, such as developmental delays and rate of change.

Cause

Why do RRBs happen? This question refers both to distal origins and to the immediate proximal causes of RRBs, and it has received far less attention in the last 10 years compared with questions that relate to definition and description. The outcomes of developmental disorders like ASD emerge from complex interactions between behavioral, genetic, neurobiological, social, and cognitive factors across time, in which causal factors are probabilistic and are difficult to isolate singly (Gottlieb, 2007; Hay & Angold, 1993). While detailed theoretical work in this area is lacking, new work is emerging in areas of neurobiology, developmental psychology, and clinical work that has the potential to provide new insights on origins, triggers, and functions of RRB. Although this work is still at an early stage in explanatory terms, we can identify potential candidate factors that influence RRBs in order that theoretical predictions can guide future empirical work.

In the next part of this review, we look at neurobiological, neuropsychological and developmental psychological explanations.

Neurobiological Theories and Evidence

Neurobiological explanations of RRBs have been most extensively supported by research using animal models. The main claims in support of neurobiological theories have been recently outlined in comprehensive reviews by Lewis and Kim (2009), Langen, Durson, Kas, Engeland, and Staal (2011), and Langen, Kas, Staal, van Engeland, and Durston (2011), Lewis and Kim's account of gene-environment neuroadaptation starts with the evidence for genetic involvement in RRBs found in chromosomal mutations in a range of genetic disorders such as Tourette syndrome, Rett syndrome, Fragile X, Parkinson's disease, Prader Willi syndrome, and other conditions. Support for genetic involvement is also indicated through familial aggregation in factor analytic studies of RRBs (Szatmari et al., 2006). Families with high scores on the IS factor provided some linkage evidence in the 15q11-q13 region at the GABRB3 locus, an area which has been implicated in a number of genetic studies of autism (Shao et al., 2003). In addition to the genetic evidence, it is also well established that repetitive sensory and motor behaviors are an invariant outcome of experiential deprivation or restriction in all animal species tested. Furthermore, any insult to the CNS will produce some effects on human behaviors, and repetitive motor behaviors can also be induced by pharmacological agents. Given that stereotypies are common in many disorders and not discriminating of autism as distinct from other neurodevelopmental disorders, these phenotypically heterogeneous sets of behaviors can arise from many etiologies (Lewis & Kim, 2009).

It is proposed that gene expression is mediated by neural circuitry that makes up the basal ganglia. This circuitry involves a large number of genes, the mutations of any of which may result in disruption of the circuitry (Arnold, Sicard, Burroughs, Richter, & Kennedy, 2006; Di Giovanni, Di Matteo, Pierucci, Benigno, & Esposito, 2006). A similar explanation can be made with respect to perinatal, neonatal, and targeted CNS insults, which may also be implicated through the neurochemical pathways and transporter genes linked to this circuitry. The role of environment is a critical element in such a proposal as there is strong evidence that stereotyped motor behavior in mice (e.g., hind limb jumping) can be induced by restricting the environment early in life, and can be reversed by enriching the environment. In sum, it is claimed that changes take place to the cortical basal ganglia circuitry as an outcome of early experience-dependent development (Lewis & Kim, 2009). Evidence for an association between striatal volume and RRB in structural MRI studies provides the main evidence for this (Hollander et al., 2005; Langen et al., 2009; Rojas et al., 2006; Sears et al., 1999).

The use of mutant mouse models of various neurodevelopmental disorders (e.g., the GABRB3 homozygous knockout mouse, which shows stereotyped behavior) is yielding some interesting results pertinent to ASD and repetitive motor behaviors. Models encompass investigation of the effects of perinatal risk factors and teratogenic agents, along with the influence of enriched vs. deprived environments. In regard to the latter theory, that is, repetitive motor behaviors as a product of environmental restriction, these are the most common category of abnormal behavior in

Table 1
Restricted and Repetitive Behaviors (RRBs) and Their Relation With Developmental Characteristics in Children With Autism Spectrum Disorder (ASD)

Authors and purpose of the study	Subjects	RRBs measure	Developmental characteristics	Results
Cox et al., 1999 Developmental Change	N = 50 children prospectively identified with autism or PDDs at 1.7 and 3.5 yr.	ADI-R	CA only	At 20 months very few children with autism and PDDs showed definite abnormality on RRBs, although some children with autism (and fewer with PDDs) showed possible abnormality. At 42 months more children showed abnormality on hand and finger and complex mannerisms and repetitive use of objects.
Militerni et al., 2002 Developmental Change	 N = 121 ASD children divided into 2 age groups: toddlers (N = 75, mean CA = 3.4 yr, range: 2.4–4.1 yr) and children (N = 46, mean CA = 8.9 yr, range: 7.2–11.4 yr). 	Semi-structured questionnaire developed by authors	CA; IQ (GMDS; WISC-III)	Motor repetitive behaviors significantly more frequent in toddler than in the child group, and child group showed significantly more complex RRBs. Sensory repetitive behaviors more frequent in low IQ (<35) group, more complex motoric sequences were more frequent in the higher IQ (medium [36–70] and high [>70]) subgroups.
Fecteau et al., 2003 Developmental Change	N = 28 autistic individuals, mean CA = 13 yr (range: 7–20.4 yr), mean IQ = 83.79 (range: 40–108).	ADI-R	CA; IQ (WISC-III; WISC-R; WAIS-R)	Current ADI–R algorithm score improved in all 3 domains, least improvement was in RRBs, when compared with retrospective scores for the 4-to 5-year age children with improvements. Chronological age was associated with developmental changes for RRBs. No significant correlation was found between FSIQ and the level of change of RRBs.
Moore & Goodson, 2003 Developmental Change	N = 20 children with severe communication problems, assessed at 2.8 yr (ADI-R) and reassessed when aged between 4 and 5 years.	ADI-R	CA only	Number of repetitive behaviors between the ages of 2 and 4 years increased. At the age of 2, body mannerisms, repetitive use of objects, and unusual sensory interests were most frequently reported. Circumscribed interests, unusual preoccupations, compulsions and rituals, hand and finger mannerisms, and repetitive use of objects increased between 2 assessments (nonsignificant). Complex mannerisms decreased significantly. (table continues)

Table 1 (continued)

Authors and purpose of the study	Subjects	RRBs measure	Developmental characteristics	Results
Gabriels et al., 2005 Developmental Change	N=14 ASD individuals, divided in high NVIQ group (N=8, mean CA = 10.6 yr [SD=7] NVIQ > 97) and low NVIQ group $(N=6, \text{mean CA} = 10.8 \text{ yr } [SD=7] \text{ NVIQ} < 56).$	RBS-R (total scores)	IQ (Leiter-R); Adaptive functioning (VABS); Behaviors (ABC); Sleep problems (CSQ)	Significantly more RRBs in lower NVIQ group. When adjusted for multiple comparisons, the groups differed significantly on only one Sameness scale. Sameness higher in the Low NVIQ group. Total adaptive scores negatively correlated with total RBS-R scores. Total RBS-R scores more highly correlated with communication ability than with social ability (VABS). Parent ratings of stress levels and sleep problems in children also highly correlated with the presence of RRBs.
Werner & Dawson, 2005 Developmental Change Distinguishing Features	N = 72 ASD (mean CA = 3.6 yr, range: 2.8–4.3), $N = 34$ DD (mean CA = 3.7 yr, range: 2.7–4.7) and $N = 39$ TD children (mean CA = 27 months, range: 12–46). Groups matched on MA (MSEL).	EDI	CA only	Children with ASD had a highe level of RRB symptoms than typical children of 10–12-months, and higher than children with DD of 16–18 months.
Kim & Lord, 2010 Developmental Change	N = 121 children with autism, $N = 71$ children with PDD-NOS; $N = 90$ children with a nonspectrum disorder (NS), and $N = 173$ children with TD. Children were divided in 6 age cohorts for each diagnostic group (<18 months; 19–24; 25–30; 31–36; 37–42; and 43–56 months).	ADOS (PL-ADOS, ADOS-T)	CA NVIQ (MSEL, BSID)	RRBs in children with autism and PDD-NOS were significantly more prevalent and severe than in children with NS and TD at all ages. Prevalence (at least one RRB exceeded 90% for both autism and PDD-NOS groups. Prevalence of RRBs increased with age for children with autism, PDD-NOS, and NS but decreased with age for TD group. Severity of RRBs was independent of age for the autism and PDD-NOS groups. NVIQ was not a significant predictor of RRBs for children with autism at any age but was for children with PDD-NOS, NS, and TD olde than 25 months.
Cuccaro et al., 2003 Classification (Factor analysis using 12 ADI- R items for RRBs)	N = 292 autistic individuals, CA range: 3–21 years.	ADI-R Factor analysis yielded 2 factors. Factor 1 (repetitive sensory motor actions, RSMA): hand and finger mannerisms, unusual sensory interests, repetitive use of objects or parts of objects, other complex mannerisms or stereotyped body movements and rocking;	Level of functioning (VABS; ABC)	than 25 months. RSMA negatively correlated with the level of functioning (ABC). Resistance to change (RC) was not correlated with the level of functioning.

Table 1 (continued)

Authors and purpose of the study	Subjects	RRBs measure	Developmental characteristics	Results
Bishop, Richler, & Lord, 2006 Classification Developmental Change	 N = 830 children with ASD (N = 560 autism, N = 268 PDD-NOS, 2 AS). Mean CA = 4.8 yr (range: 1.2–11.9). 	Factor 2 (resistance to change, RC): difficulties with minor changes in routine or personal environment, resistance to trivial changes in the environment and compulsions/rituals. Unusual preoccupations, unusual attachment to objects, idiosyncratic negative responses, sensitivity to noise did not load on either of the factors. ADI-R Factor analysis yielded 2 factors. Factor 1 (sensory-motor behaviors, RSM): repetitive use of objects, unusual sensory interests, complex mannerisms, hand and finger mannerisms, unusual preoccupations; Factor 2 (insistence on sameness, IS): difficulties with minor changes in personal routine or environment, resistance to trivial changes in the environment, compulsions/ rituals, idiosyncratic negative responses, sensitivity to noise. Unusual attachment to objects did not load on either of the factors.	CA; IQ (MSEL; DAS)	CA positively associated with expression of self-injury, sensitivity to noise, circumscribed interests, difficulties with change in routine, resistance to trivial changes in the environment, and compulsions and rituals and negatively with repetitive use of objects and unusual sensory interests. NVIQ positively associated with circumscribed interests and negatively with self-injury, unusual preoccupations, repetitive use of objects, unusual sensory interests, hand/finger mannerisms, and complex mannerisms. Factor 1 had significant negative correlation with CA, factor 2 had positive correlation.
Szatmari et al., 2006 Classification (Factor analysis using 11 ADI-R items for RRBs)	N = 339 individuals with autism, mean CA = 8.4 yr ($SD = 5.5$). Mean IQ (Leiter) = 65.7 ($SD = 28.7$).	ADI-R Factor 1 (IS): difficulties with minor changes in personal routine or environment, resistance to trivial changes in the environment, compulsions/rituals; Factor 2 (RSM): hand and finger mannerisms, repetitive use of objects, unusual sensory interests, complex mannerisms, rocking. Circumscribed interests, unusual preoccupations, unusual attachment to objects did not	IQ (Leiter); VABS	RSM scores were negatively associated with adaptive skills. IS was positively correlated with communication and language symptoms.
Richler et al., 2007 Classification (Factor analysis using ADI-R items for RRBs)	N = 165 ASD (CA < 3 yr), N = 44 DD (1.1-2.9 yr) and $N = 65 \text{ TD } \text{children}$ (CA < 3 yr). Children were assessed when they were 2, 3, 5, and 9 yrs of age.	load on any of the factors. ADI-R Factor analysis yielded 2 factors. Factor 1 (RSM): repetitive use of objects, unusual sensory interests, hand/finger mannerisms, and other complex mannerisms; Factor 2 (IS): compulsions and rituals, difficulties with changes in routine, and resistance to trivial changes in the environment.	IQ as a matching variable (MSEL; BSID)	Higher prevalence of RSM behaviors in ASD than in DD and TD groups (unusual preoccupations, unusual sensory interests, repetitive use of objects, hand/finger mannerisms, complex mannerisms, abnormal/idiosyncratic response to sensory stimuli, difficulties with change, and unusual attachments).

Table 1 (continued)

Authors and purpose of the study	Subjects	RRBs measure	Developmental characteristics	Results
Honey et al., 2007 Classification (Factor analysis using 12 ADI- R items for RRBs) Developmental Change	N = 104 children with ASD or language disorders, CA range: 2–4 yr.	ADI-R Factor analysis yielded 3 factors. Factor 1 (RSM): hand and finger mannerisms, repetitive use of objects, unusual sensory interests, complex mannerisms, unusual fears, self-injury; Factor 2 (IS): difficulties with minor changes in personal routine or environment, resistance changes in environment, compulsions/ rituals, unusual fears, idiosyncratic negative responses; Factor 3 (circumscribed interests, CI): unusual preoccupations, unusual attachment to objects.	Ability (MSEL, VABS)	Ability was related to the degree of RRBs, i.e., children with better ability had fewer repetitive behaviors. The only exception was one cluster of relatively able children with greater than expected levels of RRBs. ADI-R repetitive behavior algorithm scores increased over time.
Lam, Bodfish, & Piven, 2008 Classification (Factor analysis using 10 ADI- R items for RRBs)	1.7-29). Mean IQ = 69.5	ADI-R Factor analysis yielded 3-factor solution. Factor 1 (RSM): repetitive use of objects, hand and finger mannerisms, and other complex mannerisms/stereotyped body movements items; Factor 2 (IS): difficulties with minor changes in personal routine and environment, resistance to trivial changes in the environment, and compulsions and rituals; Factor 3 (CI): circumscribed interests, unusual preoccupations, and unusual attachment to objects.	CA; IQ (MSEL, WAIS-R, WISC-III; Leiter-R); ADOS	Higher RMB scores associated with younger age, lower verbal IQ, greater social deficits and communication impairments, and loss of skills. Higher IS scores only associated with greater social deficits and communication impairments. CI factor showed no significant correlations with any of the variables.
Esbensen et al., 2009 Classification (Factor analysis using RBS-R) Developmental Change	N = 712 ASD individuals (62.2% with comorbid diagnosis of ID). Mean CA = 19.6 years (range: 2–62 yr).	RBS-R 5 subscales (Stereotyped Behavior, Self-injurious Behavior, Compulsive Behavior, Ritualistic/Sameness Behavior, and Restricted Interests).	CA; IQ (measure not reported)	Significant correlation between CA and all five subscales of RBS-R. Individuals with comorbid ID showed significantly more stereotyped movements and SIB than individuals with ASD alone.
Mooney et al., 2009 Classification (Factor analysis using 12 ADI- R items for RRBs)	N = 137 DD children with PDD and N = 61 DD children without PDD, all with CA 1.7–4.6 yr.	ADI-R Factor 1 (RSM): hand and finger mannerisms, repetitive use of objects, complex mannerisms; Factor 2 (IS): difficulties with minor changes in personal routine or environment, resistance to trivial changes in the environment, compulsions/rituals, unusual attachment to objects; Idiosyncratic negative responses, sensitivity to noise, unusual sensory interests, unusual preoccupations, self-injury did not load on any factors.	CA, developmental age (PEP-R), adaptive behavior (VABS)	Significant positive associations between IS and CA and developmental age in children with DD and PDD. RSM had a significant negative association with all developmental variables. Both IS and RSM had negative association with developmental variables in children with DD without PDD.

Table 1 (continued)

Authors and purpose of the study	Subjects	RRBs measure	Developmental characteristics	Results
Mirenda et al., 2010 Classification (Factor analysis using RBS-R)	N = 287 children with ASD, mean CA = 3.4 yr, range: 2–5.3.	Idiosyncratic negative responses, sensitivity to noise, unusual sensory interests, unusual preoccupations, self-injury did not load on any factors. RBS-R Factor analysis found that 3- and 5-factor RRBs models were the preferable models. Model III comprised of Factor I: Compulsive Ritualistic Sameness Behaviors (CRSB); Factor II: Self Injurious Behaviors (SIB); Factor III: Restricted Stereotyped	ADI-R; ADOS; VABS II; CBC; M-P-R	Factors in both 3- and 5-factor models were negatively correlated with the VABS II total score, most strongly for RSB (Model III) and Stereotypy (Model V). Only factors related to CRSB were strongly correlated with chronological age. Developmental index standary scores were not correlated.
Richler et al., 2010 Classification (Factor analysis using ADI-R items for RRBs) Developmental Change	 N = 192 children with ASD who were referred for a diagnosis when they were under the age of 3 and followed up at the age of 3, 5, and 9. 	Behaviors (RSB), Items 1–6 and 40–43. Model V comprised of Factor I: Stereotyped Behaviors; Factor II: Self-Injurious Behaviors (SIB); Factor III: Compulsive Behaviors (COMP); Factor IV: Ritualistic Sameness Behaviors (RITUAL/SAME); and Factor V: Restricted Behaviors (RESTR). Factor analysis yielded 2 factors. Factor 1 (RSM): repetitive use of objects, unusual sensory interests, hand/finger mannerisms, and other complex mannerisms; Factor 2 (IS): compulsions and rituals, difficulties with changes in routine, and	CA; IQ (MSEL)	Increasing CA was associated with decreasing RSM. At age 2, there was significant negative main effect for NVIQ, i.e., as NVIQ scores increased, RSMs decreased. Children with higher NVIQ scores at age 2 showed more pronounced decrease in RSM scores over time.
		resistance to trivial changes in the environment.		Higher CA associated with increasing IS; NVIQ at age 2 was not associated with change in IS; however, milder social/communicative impairments were.
Liss et al., 2001 Distinguishing Features	N = 35 HFA children (mean CA = 4.8 yr, SD = 1.38) and N = 31 age matched children with developmental language disorder. LFA children (Mean CA = 4.9 yr, SD = 1.38) with 17 age matched children with low IO	Wing Autism Diagnostic Interview Checklist	IQ (SB FE) Adaptive behavior (VABS)	Both autistic groups had significantly more RRBs than matched children. RRBs significantly correlated with adaptive behavior in the HFA group; for the LFA group there was no such correlation.
South, Ozonoff, & McMahon, 2005 Distinguishing Features	IQ. $N = 21$ HFA (mean CA = 14.1 yr, range: 8–20 yr), $N = 19$ AS (mean CA = 14.28 yr, range: 8–19 yr) and $N = 21$ TD individuals (mean CA = 13.34 yr, range: 7–19 yr) matched on VIQ, PIQ, and FSIQ.	RBI; YSII	CA only	The only significant between- group repetitive behavior differences were for lifetime severity of the Object Use and Rigid Routines categories. Severity of any repetitive behavior category not significantly correlated with age. Circumscribed Interests showed gradual increases in mean impairment over time. (table continues

Table 1 (continued)

Authors and purpose of the study	Subjects	RRBs measure	Developmental characteristics	Results
Cuccaro et al., 2007 Distinguishing Features	N = 33 pairs of AS and HFA individuals matched on age, sex, and IQ. Mean CA = 11.6 yr, range: 5.5–22.7 (AS), 10.7 yr, range: 4.8–21.9 (HFA), Mean IQ = 100, range: 75–138 (AS), 101, range: 75–146 (HFA).	ADI-R; RBS-R; ABC-C	Developmental level	No differences between groups in either intensity or the frequency score on RBS-R. No differences at the item level. No differences on ABC-C scale either.
Honey et al., 2007 Distinguishing Features	N = 79 ASD and $N = 117TD children. CA range: 2–4 and 6–8 yr.$	RBQ	Language level (items taken from DISCO); Play (APQ-R)	Significantly more RRBs in ASD than in TD children. RRBs were negatively associated with play in ASD but not TD children. RRBs in ASD were predicted by play and expressive and receptive language.
McDonald et al., 2007 Distinguishing Features	N = 30 ASD and $N = 30$ TD children. Within each group $N = 10$ 2-year-olds, 10 3-year-olds, and 10 4-year olds.	Direct observational protocol (NECC Early Core Skills Assessment Battery).	CA only	2-year-old ASD children had higher levels of motor, vocal, and total stereotypic behavior than 2-year-old TD children. Difference was more pronounced at 3 yr, and even more at the age of 4 yr.
Morgan, Wetherby, & Barber, 2008 Distinguishing Features	N = 50 ASD, $N = 25$ DD, and $N = 50$ TD children. Mean CA at ADOS assessment = 3.7 yr ($SD =$ 1.2) for ASD group and 3.9 yr ($SD = 1.2$) months for DD group.	RSMS	Developmental quotients (MSEL); Autism symptoms (ADOS)	Significantly more RSM with body and objects in ASD than the TD group and significantly more RSM with body and objects than the DD group. RSM with objects in 2nd year significantly predicted NVDQ and VDQ in the fourth year for the ASD group. Rate and restricted inventory of RSM with objects were negatively correlated with NVDQ and VDQ.
Ozonoff et al., 2008 Distinguishing Features	N = 66 1 yr infants. $N = 9$ children met ASD criteria by 3 yr, $N = 10$ met criteria for other DD, and $N = 47$ fell into no concerns group.	RRBs coded by blind raters from videos where 4 objects were presented to the infant, one at the time.	CA only	Significant group effect was observed for atypical uses of objects. Significantly more rotating, spinning, and unusual visual exploration of objects in ASD than in both other delays and the no concerns groups The most common atypical object use in the autism/ASD group was unusual visual exploration, shown by 7 of the 9 infants.
Watt et al., 2008 Distinguishing Features	N = 50 ASD, $N = 25$ DD, and $N = 50$ TD children, all with CA range: 1.5–2 yr.	RRBs were coded from videotaped Behavior Sample of the CSBS.	Developmental level (MSEL)	Significantly higher frequency and longer duration of RSB with objects, body, and sensory behaviors in ASD than in DD and TD groups. Significant correlations between RRBs with objects and developmental level on the symbolic composite in ASD group, and between RRBs with objects and the social composite. (table continues)

Table 1 (continued)

Authors and purpose of the study	Subjects	RRBs measure	Developmental characteristics	Results
Goldman et al., 2009 Distinguishing Features	N = 129 children with autism and N = 148 cognitively-matched non-autistic developmentally disordered (NADD) children divided into 2 subgroups: developmental language disorder and non-autism, low IQ group. Mean CA = 4.5 yr, range: 2.9–8.1.	Standardized play sessions coded for motor stereotypies.	IQ (Abstract/Visual Reasoning score of the SB); Diagnosis	Significant correlations between RRBs with objects in the 2nd year and verbal and nonverbal DQ on the MSEL at 3 years. Correlations no longer significant when controlling for developmental level on the symbolic composite in the 2nd year. More children with autism had stereotypies than NADD comparison children. Autism and NVIQ (<80) contributed independently to the occurrence, number, and variety of stereotypies. Autism contributed independently to gait and hand/finger stereotypies and NVIQ < 80 to head/trunk stereotypies.
Boyd et al., 2010 Distinguishing Features	N = 67 children with autism (mean CA = 4.3 yr, SD = 1.4), N = 42 with DD (mean CA = 4.1 yr, SD = 2). Children were matched on MA.	RBS-R (6 subscales)	Visual Reception (VR) scale of MSEL, 4 sensory measures (SEQ; SP; SPA; TDDT-R)	Children with autism had significantly higher scores than children with DD on all RBS-R subscales except for self-injury. There were significant correlations between hyperresponsiveness and the presence of stereotypies, compulsions, and rituals/sameness behaviors. For sensory seeking, a significant correlation was only found for ritualistic/sameness behaviors.

Note. ABC = Autism Behavior Checklist (Krug, Arick, & Almond, 1980); ABC-C = Aberrant Behavior Checklist-Community (Aman & Singh, 1994); ADI-R = Autism Diagnosis Interview-Revised (Lord, Rutter & Le Couteur, 1994); ADOS = Autism Diagnostic Observation Schedule (Lord et al. 2002); PL-ADOS = Pre-linguistic ADOS (DiLavore, Lord, & Rutter, 1995); ADOS-T = ADOS-Toddler Module (Lord, Luyster, Gotham, & Guthrie, 2010); APQ-R = Activities and Play Questionnaire-Revised (Honey et al., 2007); BSID = Bayley Scales of Infant Development (Bayley, 1993); CBC = Child Behavior Checklist (Achenbach & Rescorla 2000); CSQ = Child Sleep Questionnaire (Garcia & Wills, 2000); DAS = Differential Ability Scales (Elliott, 1990); DISCO = Diagnostic Interview for Social and Communication Disorders (Wing et al., 2002); EDI = Early Development Interview (Keller et al., 1987); GMDS = Griffiths Scale of Mental Development (Griffiths, 1992); Leiter = Leiter International Performance Sales (Levine, 1986); Leiter-R = Leiter International Performance Test-Revised (Roid & Miller, 1997); M-P-R = Merrill-Palmer-Revised Scales of Development (Roid and Sampers 2004); MSEL = The Mullen Scales of Early Learning (Mullen, 1995); PEP-R = Psycho Educational Profile-Revised (Schopler et al, 1990); RBI = Repetitive Behavior Interview (Turner, 1997); RBQ = Repetitive Behavior Questionnaire (Turner, 1996); RBS-R = Repetitive Behavior Scale-Revised (Bodfish et al., 2000); RSMS = Repetitive and Stereotyped Movement Scales: Companion to the CSBS (Wetherby & Morgan, 2007); SB FE = Stanford Binet, Fourth Edition (Thorndike, Hagen, & Sattler, 1986); SEQ = Sensory Experiences Questionnaire (Baranek et al., 2006); SP = Sensory Profile (Dunn, 1999); SPA = Sensory Processing Assessment for Young Children (Baranek, 1999); TDDT-R = Tactile Defensiveness and Discrimination Test Revised (Baranek, 1998); VABS = Vineland Adaptive Behavior Scales (Sparrow et al., 2005); WAIS-R = Wechsler Pre-School and Primary Scales of Children-Revised (Wechsler, 1991); WISC-III = Wechsler In

confined animals. Lewis, Tanimura, Lee, and Bodfish (2007) reported on three related studies looking at enrichment-related changes in neuronal structure and function relating to stereotypic behaviors in deer mice, including a focus on the role of cortical based ganglia circuitry. Low stereotypy plus enriched-environment deer mice showed higher neuronal functioning activity in motor cortex, striatus, nucleus accumbens, thalamus, and hippocampus, and also showed higher dendritic spine densities compared to nonenriched groups. They also showed better procedural learning

and reversal learning, although we note that the enriched *low-stereotypy* mice had the best chance to do well, given their less severe behavioral abnormalities. Enrichment effects were regionally selective for motor cortex and basal ganglia, and this is argued to be consistent with MRI studies of caudate volume associations with stereotypy in autism. The mouse studies summarized by Lewis et al. (2007) then, indicate selective effects on stereotypic behavior from enriched environmental experience, conditional on severity of initial levels of stereotyped behavior, as well as on

regionally selective brain differences in the motor cortex and basal ganglia. Moreover, the reported behavioral benefits were associated with better performance on reversal learning and procedural tasks. Attenuation of RRBs through exposure to more complex environments was also reported by Schneider, Turczak, and Przewlocki (2006). For example, exposing animals to more complex environments can attenuate or reverse some sequelae of CNS insults (infarcts, seizures, cortical lesion, brain injury etc.).

The effects of environmental enrichment on behavioral abnormalities in male rats exposed to valproic acid on day 12.5 of gestation (this elicits long term negative effects on post natal development) has also been examined (Schneider et al., 2006). This study compared Valproic acid (VPA) and non-VPA rats in enriched environments and also with a control group of nonenriched animals. Environmental enrichment reversed negative effects of VPA. Schneider et al.'s (2006) salient effects in rats exposed to VPA can be related to a model of RRBs in autism, that is, lower locomotor, repetitive/stereotypic activity, enhanced exploratory activity (in mazes), decreased anxiety, and increased social behaviors. The reduction in repetitive behaviors in this model points to the usefulness of enriched environment as treatment for autism as well as for other developmental disorders since "enforced interaction with the physical environment" (Schneider et al., 2006, p. 44) is important in stimulating brain changes in the rat. Schnieder et al. refer to Applied Behavior Analysis type treatments (Lovaas et al., 1987) as illustrating the validity of this theory of rehabilitation, and note the value of cognitive engagement and response contingencies along with physical exercise as parallels for intervention in autism (see also Dawson, 2008).

Drug induced RRBs have also been explored in animal models; and here, too, there is emphasis on the importance of the basal ganglia in the mediation of RRBs. While this is noted by many authors, the basal ganglia is a nondisorder-specific site, and basal ganglia influences on abnormal behavior have been applied to many different behavioral abnormalities and various disorders. Dopamine or dopamine agonists injected into the corpus striatum lead to increased stereotyped behavior in rats. Motor stimulatory effects of amphetamine, such as stereotypies, have been observed that can be reversed via intracortical infusion of DA or GABAergic agonists. Drug induced stereotypic behavior is sensitive to manipulations; for example, substantia nigra pars reticulate is implicated as a direct pathway and the subthalamic nucleus as an indirect pathway in rat studies. Lewis and Kim (2009) suggest that drug induced RRBs provide the strongest evidence related to pathophysiological mechanisms underlying these behaviors.

These recent findings in animal research raise important insights for understanding potential causal influences as well as for remediation of repetitive behaviors in ASD. It can be argued that the early onset of deficits in social, communicative, and adaptive behavior (arising from extreme social withdrawal) in infants and young children could interfere with experience dependent behavioral and brain development in early life, as children with ASD begin to create their own restricted environment. The message from these neurobiological findings supports the desirability of active and intensive intervention that acts upon that self-imposed constrained environment to enhance brain development and reduce stereotypies. Interventions that work by providing reward contingencies for alternative adaptive behaviors therefore may increase success in treating RRBs and associated anxiety.

While the potential implications from animal models might be encouraging, we should also be cautious about the evidence. First, as noted in reference to drug induced RRBs, explanations of basal ganglia influences on abnormal behavior have been broadly applied to many different behavioral abnormalities and various disorders. The claim that RRBs are the result of imbalanced activity along the direct and indirect pathways of the basal ganglia needs to be considered in the light of this fact (Schneider, Turczak, and Przewlocki, 2006). There is no evidence so far from postmortem investigations for abnormalities in basal ganglia or thalamus in autism; and the evidence in ASD rests mainly on the MRI studies of caudate volume associations with stereotypy, with some studies showing increased volume while others show no difference (see Langen, Durston, et al., 2011, for review). Second, there are limitations in the behavioral comparisons that can be made between animals and children. Apart from the fact that the evidence from animals is generally restricted to repetitive sensory and motor behaviors, these accounts do not seem to take into account those sensory and motor behaviors that occur in ASD individuals when stimulated by a deliberate sensory experience, for example repetitive light switching and self-spinning. Nor can they include the more sophisticated forms of RRBs-like elaborate forms of preoccupations found in very able children. Furthermore, they also do not take account of normal developmental processes that are likely to be seen in children, given that reduction of RRBs in normal infant development takes place over time without specific environmental intervention. Even when children have been subject to early and severe environmental deprivation, as in the case of children adopted from Romania, only a minority of severely deprived children were reported to show autistic or quasi-autistic features including repetitive behaviors, indiscriminate social approaches, and abnormal response to sensory stimulation (Rutter et al., 2007) and improvements in development were reported from 4 to 6 years of age (Rutter et al., 1999). For approximately half of these children, autistic symptoms disappeared quite soon after adoption and just a small percentage had more long-term symptoms. These studies showed that early and severe environmental privation can lead to autistic behaviors but do not inevitably do so. Finally, environment enrichment effects for caged animals in deprived environments and for children with ASD who create a self-imposed restricted environment may be quite different. We know that increasing intrusion and forced interaction for young autistic children (i.e., an enrichment context) can improve their learning, although this learning often does not generalize; and there seems to be a ceiling on how much progress is possible with severely intellectually disabled cases. Nevertheless, the proposals for gene-environment neuroadaptations and potential brain plasticity in response to environmental enrichment in animals do suggest potential avenues of intervention and change, and predictions can be tested to advance both theory and treatment.

The recent work of Langen and colleagues (Langen, Durston, et al., 2011; Langen, Kas, et al., 2011), also makes a new and valuable conceptual contribution to this area. These authors propose a classification of three parallel corticostriatal macro-circuits, each targeting primary motor and premotor and prefrontal cortical areas through feedback loops. These are the sensory motor loops, linked to motor and premotor cortex and related to simple motor stereotypies: the cognitive or associative loop connected to dorsolateral prefrontal cortex and implicating rigidity and obsessive

routine, and the limbic (motivation) circuit connecting to anterior cingulate and orbitalfrontal cortex and implicating compulsive and addictive behavior. Langen, Durston, et al. (2011), argue that the functional relations between these circuits may help account for distinctions between one type of RRB (motor stereotypies) compared to another (obsessions) but also their cooccurrence across the different clinical conditions of ASD, OCD, attention deficit hyperactivity disorder (ADHD), Parkinson's disease and Huntington's disease. While the structural and functional interactions between these brain areas may end up being more complex than their scheme suggests, their classification enables hypotheses to be tested about brain behavior links in RRBs and could help to clarify connections between ASD and other childhood disorders.

Neuropsychological Theories and Evidence

Theories relating frontal lobe or executive functioning (EF) capacities to ASD (Russell, 1997) and proposing a connection between EF and RRBs (Turner, 1997) highlight the EF impairments of poor regulation and control of behavior. Executive dysfunction encompasses problems with inhibition of inappropriate behavior, impaired generation of adaptive, organized, goaldirected behavior, failure to profit from feedback in the environment, lack of flexibility, and perseveration. Such impairments are characteristic of individuals who have sustained frontal lobe injury; but it is easy to see how they could be causally connected to the lack of goal-directed behavior, restricted interests, and stereotyped behaviors in ASD. For example, in a theoretical review of the neurocognitive literature related to Obsessive Compulsive Disorder (OCD), particularly EF task performance, Evans, Lewis, and Iobst (2004) proposed that repetitive rituals and compulsions may share a common neurobiology which is similar to that underlying the RRBs of typically developing children. Further, they argue that the processes inherent in cognitive EF tasks and central in emotional and behavioral self-regulation are governed by particular regions of the orbitofrontal cortices which have been implicated in OCD. The same explanation might apply to ASD. However it must be noted that there is a fundamental contrast between ASD and OCD in that the obsessive thoughts and behaviors in OCD are seen by the sufferers as unpleasant, distressing and contrary to their adjustment and well-being. We do not know if this is the case for ASD, and it can be argued that some RRBs are pleasurable and rewarding for them. Therefore it is questionable whether it is appropriate to apply similar developmental neurobiologically based localization theories to ASD.

Ten years ago, theoretical proposals related to EF impairments provided the major contender as an explanation for RRBs; with potential to account for a wide range of both low and higher level behaviors. Turner (1997, 1999), proposed two separate hypotheses, one relating to an inability to inhibit ongoing behavior and another related to an inability to generate novel behavior. A decade of research has not been able to fully substantiate either hypothesis. There have been mixed results concerning evidence for EF deficits, with a number of variables, including type of tests used, child age, overall cognitive ability, and language facility significantly modifying results in assessment tasks (Prior & Ozonoff, 2007).

Zandt, Prior, and Kyrios (2009) compared children and adolescents with OCD and those with high functioning ASD on a battery

of EF tests. They found that these two clinical groups did not differ from a typical group with equivalent measured intelligence on most tasks. There was a trend for children with ASD to show poorer generativity (on verbal fluency and concept generation tasks), while those with OCD tended to have impaired inhibition (e.g., the walk-don't walk task). Turner (1997) has also reported some evidence for generative impairments in ASD, but there is little support for deficits in inhibition capacities (e.g., Brian, Tipper, Weaver, & Bryson, 2003). Tregay, Gilmour, and Charman (2009) found no association between either inhibition or generativity measures. Likewise, research on set-shifting with children has revealed mixed results (Yerys et al., 2009). Repetitive behaviors, as measured by Zandt et al. (2009), did show some associations with EF, although these were small and variable across the clinical groups. It is important to note that, in this study, parents rated both OCD and ASD individuals to have significantly more difficulty with EF skills in their daily lives compared with typically developing children, as measured by the Behavior Rating Inventory for Executive Functioning questionnaire. Rated and psychometrically assessed measures of EF showed only weak correlations, again highlighting measurement challenges in EF research.

Overall, findings of executive dysfunction are very mixed and suggest a strong effect of type of assessment, especially when tests are highly rule bound; this is where children with ASD are likely to show deficits. There is a higher likelihood of finding EF-related impairments in children with lower general cognitive abilities (Prior & Ozonoff, 2007), hence, a significant influence of IQ and language ability is central to EF findings. Yerys et al. (2009) addressed the question of whether EF deficits might be causal in ASD or whether they are a secondary deficit. They compared 2- to 3-year-old children in four groups: ASD, developmentally delayed, typical development matched for chronological age (CA), and typical development matched for mental age (MA). Children were given a battery of visual-spatial EF tests. No specific deficits in the ASD group were found relative to controls. The authors suggest that EF deficits may emerge as a secondary deficit at least in higher functioning children. This conclusion would be helpful in rationalizing the very variable findings concerning EF capacities in very young children. 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. Again, lack of specificity is an issue since EF impairments are common across a range of childhood disorders, including OCD, ADHD, and conduct disorder (CD; Clark, Prior, & Kinsella, 2002). Neuropsychological profiles of EF deficits are not discriminating for ASD but appear in individuals with various behavioral and symptom profiles where behavioral dysregulation is part of the pathology.

Developmental Psychological Theories and Clinical Evidence

The developmental psychology approach to RRBs is best represented by the early work of Thelen (1981). Although this account predates the 10-year period of our review, we describe it here because the ideas within it offer a conceptual framework that can

be applied to clinical evidence and can raise predictions that are testable.

Thelen's theoretical account of the emergence of stereotypies in typical infant development begins with Tinbergen's (1951) distinction between evolutionary origin (distal cause) and the immediate antecedents (triggers or proximal causes) of these behaviors that are distinct from the original cause of the behavior. Evolutionary selection provides a slow period of cortical maturation, and this results in a prolonged developmental stage in which lower level rhythmical behaviors dominate. As these behaviors become drawn into the behavioral repertoire in the first months of life, adaptive functions emerge. Adaptive functions may include communication of affect to caregiver; the increase of vestibular, auditory, and visual neural stimulation; and cognitive development (e.g., means-end relations achieved by manipulating the interesting effects of actions on the external environment). While these adaptive functions are involuntary consequences of RRBs in the early months, they change with development to acquire a more voluntary, instrumental significance.

Proximal causes (immediate triggers) for repetitive behaviors differ from adaptive functions and also change with development. In the first year of life, when motor action is less under voluntary control, stereotypies are high in frequency and sensitive to being released by many triggers. At the end of the first year however, when RRBs are more varied and motor behavior more goal directed, more extreme arousal states (either high or low) are needed to release stereotypy, and they are more likely to be triggered by specific environmental stimuli, such as objects or events. Triggers for RRBs, then, need to be understood within a context that balances developmental and environmental factors.

Thelen's explanation is that stereotyped behaviors in the early months play a role in neuromuscular development and in the development of skilled motor action. Although initially driven by endogenous neural mechanisms, the repetitive behaviors themselves have an impact on the developmental system, creating a developmental transformation in the organization of behaviors. The implications of Thelen's account are that rhythmical behavioral patterns of repetition have a systemic effect on development that go beyond the behaviors themselves and may be related to other aspects of development, such as communication and language and social interaction (Iverson & Wozniak, 2007). We draw on these implications and apply Thelen's account of causal and functional aspects of infant stereotypies to the broader category of RRBs seen in ASD in order to clarify the relation between triggers and adaptive functions of RRBs in developmental terms.

Some interesting clinical research evidence has appeared in the last 10 years that, although not motivated from developmental theory, might be interpreted in the light of Thelen's concepts. While these studies do not focus on developmental change or on the coordination of motor and nonmotor systems in development, they draw on Thelen's concepts of adaptive functions and triggers. For example, in a recent Ph.D. study, Barber (2008) has investigated triggers and functions for repetitive and stereotyped behaviors using video-recorded behavior samples of 18–24-month-olds during the completion of the CSBS Behavior Sample (Wetherby & Prizant, 2002). Children with ASD showed lower proportions of well-regulated behavior during episodes of repetitive, stereotyped behavior compared to children with developmental delay and typical samples, matched for either chronological age or mental

age. Children with ASD also demonstrated more object-focused repetitive sensory and motor behaviors for the novel objects presented to them than the TD groups and fewer repetitive sensory motor behavior functions related to the meaningful use of objects than children in the comparison groups. The most common function for RRBs was the need to occupy self. This was defined as a repetitive stereotyped behavior that appeared to entertain the child when the child was disengaged or disinterested prior to its onset. There was also some soothing function. In addition, RRBs created a barrier function and interfered with social experience and new learning of all kinds. The functions underlying RRBs did not differ between the three groups. As Barber pointed out, no single theory of utility could be supported. Here we suggest that it might help to apply Thelen's account by considering how different adaptive functions (occupying self versus soothing) for different individuals might be related to arousal states and to particular triggers (e.g., manipulating objects, emotion). For example, high arousal might lead to a soothing function of RRBs while low arousal leads to manipulation of objects (occupying self).

Several studies have investigated emotional or motivational triggers for repetitive behaviors and their results are also consistent with Thelen's view that developmental changes need to be taken into account when considering events that trigger repetitive behaviors. Militerni et al. (2002), in a study with 2- to 4- and 7- to 11-year-old children with ASD found that almost three quarters of the RRBs they observed did not seem to be reactive to a particular emotional trigger, whilst the remainder were reactive. Results showed that simple motor behaviors and sensory stimulation were more common in the younger age group and that these were the kinds of behaviors (e.g., repetitive limb and trunk movements and high intensity sensory behaviors) that were reactive to emotional triggers, while repetitive complex sequences and repetitive language did not follow this path. These results are consistent with Thelen's analysis of developmental changes in the significance of triggering events.

Another recent study (Joosten, Bundy, & Einfeld, 2009) investigated motivational triggers for repetitive behaviors in 5- to 18year-old individuals with ASD plus intellectual disability (ID) and in those with ID alone. The Motivation Assessment Scale (MAS; Durand & Crimmins, 1988) was used and measures taken of intrinsic (sensory seeking and arousal factors) and extrinsic motivation (attention, gaining an object, or escape), to which were added items measuring anxiety as a further potential intrinsic motivator. The MAS questions were rated by teachers who knew the young people well and were familiar with their repetitive behaviors. Results showed anxiety to be a stronger motivator for RRBs than sensory seeking in the children with ASD plus ID, with the reverse relationship the case for children with ID only. For extrinsic motivation, there were again group trends, with escape and gaining an object most common for ASD plus ID and attention and escape most common for the ID group. Generally RRBs in ASD were driven by both intrinsic and extrinsic factors, but the particular importance of anxiety as assessed in this study is salient. The study presented some measurement challenges including the limits of validity in relying on teacher reports. It also raises interesting conceptual issues. Although anxiety is conceptualized as intrinsic, it can be driven by external situations that induce anxiety and that themselves require analysis as distal triggers.

The role of anxiety is a key factor that is increasingly being discussed in the literature on RRBs. Very early explanations of impaired reticular-activation-system functioning (Hutt & Hutt, 1965) proposed that repetitive behaviors might be caused by either hyper- or hypoarousal. That is, RRBs provide coping strategies for children with ASD to enable them to either regulate high levels of arousal or to reduce anxiety; or, in the case of hypoarousal, to increase sensory stimulation. Insufficient evidence was provided at the time to support either of these proposals. However, new evidence from animal models, especially work on restricted environments and experience, as well as new research on childhood anxiety, suggests that we need to revisit the issue of arousal and repetitive behaviors, and reconsider the results of this earlier work.

Anxieties over change related to the need for sameness are often intense, and increases in RRBs that occur when children are challenged, stressed, tense, or anxious may provide relief, as seen in the performance of compulsive behaviors in OCD and in outbursts of stereotyped behaviors in children with ASD that act as a barrier when children are faced with a challenging task. There is evidence in both the typical development and OCD literature for relationships between RRBs and anxiety and fears. With regard to typically developing children, Evans, Gray, and Leckman (1999) reported that repetitive and "just right" behaviors including bedtime rituals and hoarding objects were significantly related to overall fears and fear of strangers.

Although the case for anxiety as a proximal cause for RRBs seems compelling, the scientific evidence for the link between anxiety and RRB in ASD is still relatively sparse. Currently the limited evidence for a potential connection between RRBs and anxiety comes from a range of sources. For example, Kamp-Becker, Ghahreman, Smidt, and Remschmidt (2009) reporting on the dimensional structure of the autism phenotype using the ADI-R and ADOS found two factors in higher functioning ASD, social communication and anxious and compulsive behavior, which were themselves linked. A study by Tonge, Brereton, Gray, and Einfield (1999) reporting high anxiety in HFA and AS on the parent-rated Developmental and Behavior Checklist, also supported the significance of anxiety in behavioral aspects of autism. More comprehensive assessment was carried out by Gillot, Furniss, and Walter (2001), using the Spence Children's Anxiety Scale (Spence, 1998) and the Social Worries Questionnaire (Spence, 1995) with 10year-old children with ASD. This study showed higher mean levels of anxiety in ASD compared with levels in children with specific language impairment and in TD children, although most anxiety scores were not in the clinical range. The highest subscale scores were on Obsessive Compulsive type behaviors, and on separation anxiety, and children with ASD were also significantly higher on social anxiety. Children with ASD self-reported more social worries compared with the other groups. Parent-report measures too show high levels of social worries in children with ASD. However, it is worth noting that parent-reported and child self-reported symptoms of anxiety may not be concordant (White & Roberson-Nay, 2009), possibly due to problems with self-insight and with the unusual expression of symptoms of anxiety in ASD.

OCD is classified as a disorder with anxiety as its origins, thus this research may also speak to theories of the role of anxiety in ASD. Some of the neurobiological, genetic, and pharmacological findings associated with OCD have been extended to apply also to ASD (Evans et al., 2004) in terms of their etiological functions.

However, as mentioned above, it is not yet clear that anxiety linked to obsessions and compulsions seen in OCD is exactly the same as experienced in the sameness behaviors of ASD. In sum, research evidence supporting the increasingly popular proposal that anxiety and arousal are key causal factors in RRBs is still in its infancy and more research is needed to achieve a clear picture of the similarities and differences between RRBs and anxiety disorders, as well as any putative neurobiological associations.

Summary

In this section we focused on the question of why RRBs happen in children with ASD, taking theoretical perspectives from neurobiology, neuropsychology, and developmental psychology. The neurobiological account proposes that RRBs are the outcome of gene-environment neuroadaptations that arise from effects of genetic vulnerability and social isolation, with evidence from mouse models pointing to the likely importance of environmental restriction, whether real or self-imposed, as a candidate for increased RRBs. Most of the evidence highlights the presumed role of basal ganglia pathways. This neurobiological account allows predictions to be made about environmental effects on early brain development, and there are important implications here for intensive interventions that move children away from restricted patterns of self-chosen behavior toward alternative adaptive behaviors. On the other hand, the evidence is limited to animal models and, apart from selected evidence on the effects of institutionalization and privation in Romanian orphans whose preinstitutionalized behavior is largely unknown, this account has not been tested on human populations. Furthermore, apart from a very few studies that test maze reversal behavior in rats, the evidence and predictions are heavily restricted to repetitive sensory and motor behaviors rather than insistence on sameness behaviors.

While the neurocognitive account attempts to address both lower level repetitive sensory and motor behaviors and higher level insistence on sameness behaviors, the claims it makes for dysfunction in executive processes have not been sustained by converging evidence. Descriptively, children with ASD clearly appear to have poor regulation and control, but the proposed cognitive impairments of response inhibition, set shifting, generativity, planning, and their associations with repetitive behavior frequency have not been established. It is just as likely that neurocognitive functioning is a consequence rather than a cause of RRB, since all behavioral aspects of autism are both affected by, and also affect, the level of neurocognitive development. Given the reciprocal effects of EF and RRB associations across time, it might be more amenable to testing in older children and adults. A major challenge to this field is the traditional conceptual approach taken to viewing differing components of executive functions as a single construct related to frontal lobe functioning (Prior & Ozonoff, 2007). In sum, the evidence for the executive dysfunction account is very mixed, and conceptual clarity and specificity of theoretical prediction is lacking. Furthermore, developmental age and level of intelligence are strong influences in extant findings in this domain. Until these complexities are resolved, we recommend that researchers consider carefully before selecting components and measures of executive function and aim to move the field forward conceptually before retesting the same component constructs.

The developmental psychology account proposes that RRBs seen in children with ASD are immature behavioral responses that are a normal part of early development but have been maintained beyond the typical period of development. In typical development, these involuntary behaviors are triggered by arousal but come increasingly under voluntary control as infants begin to develop goal-directed actions. The balance between arousal and action in typical development is affected by systemic influences that connect motor behaviors to other domains of cognitive and emotional development. This account allows predictions to be made about developmental effects, triggers for RRBs, and functions of RRB. Like the neurobiological account, the developmental psychology account applies predominantly to motor stereotypies. Its generalizability to higher level insistence on sameness behaviors and obsessive interests is unknown. Some clinical findings related to triggers and adaptive functions, however, might fit into the conceptual framework outlined by Thelen and could also add conceptually to existing approaches to functional analysis (see Vollmer & Smith, 1996, for review). This approach also predicts that interventions that generate an optimal state of arousal in addition to encouraging alternative adaptive behaviors will be more effective than interventions that focus on only one of these aspects.

While the neurobiological, cognitive, and developmental approaches to the origins of RRB noted above need further explanation, evidence emerging from clinical studies indicates that a key trigger for repetitive behaviors is arousal. Unstructured environments may also act as a trigger. RRBs could function in the same way as in typical development, as self-regulating coping strategies that help to regulate hyperarousal or to increase sensory stimulation in hypo-arousal. In future work, it is likely that research will be more fruitful if we distinguish between lower level repetitive sensory and motor behaviors and higher level insistence on sameness behaviors in investigating the origins of RRBs.

To summarize, there may be a mixture of proximal causes or triggers for RRBs in ASD, including anxiety and communication difficulties that lead to frustration and maladaptive behavior; lack of stimulation in situations of self-induced withdrawal (since providing structure, stimulation, and purposeful activity can be shown to lead to reduction in RRBs); learned stereotypies (as seen in caged animals), and internal biological triggers operating independently of the environment. Currently, we do not understand how these factors relate to each other. But further research on this is very important in order for clinical interventions based on functional analysis of behavior to proceed. Future empirical work should target how proximal causal factors relate to each other systematically and take account of individual variation seen across children.

Change

In the third part of this review we examine the potential for RRBs to change across time. This section is divided into two. First we consider the evidence on changes in RRBs in terms of the natural history of development. Then we evaluate evidence of change in terms of response to specific interventions.

Developmental Trajectory of RRBs

What is the potential for change in RRBs in children with ASD? Do RRBs reduce with age and developmental level? Gaining a

clear understanding of the natural history of RRBs in children is important in order to understand what degree of change is possible and therefore to plan effective interventions. To date, evidence from short-term longitudinal follow-up studies and from cross-sectional studies has made it difficult to discern distinct, reliable patterns of increases or decreases in RRBs across time. However, it is possible to review the effect of developmental factors across time by examining research carried out within the larger context of ASD symptoms together with research that specifically examines developmental effects (see Table 1).

Change in early childhood. At the beginning of this decade, there were several follow-up studies in the literature that reported on change in core social, communication and RRBs between the 2nd year and the age of 4-5 years. These studies, using data collected from the ADI-R, indicated that there was little sign of atypicality in RRBs in children with ASD compared with those seen in typically developing children until the end of the 3rd or beginning of the 4th year, when more evidence of atypicality in RRBs appeared (Cox et al., 1999; Moore & Goodson, 2003; Stone et al., 1999). Characteristics of RRB in these studies included persistence of earlier repetitive sensory and motor behaviors that would be expected to have disappeared in typically developing children, in particular hand and finger mannerisms and repetitive use of objects. In addition, increase in preoccupations, circumscribed interests, and attachment to objects featured in those children with higher cognitive ability. The view that atypical RRB is uncommon in toddlerhood has continued to be supported in research using the ADI-R (Ventola et al., 2006), although one study using the ADI-R (Richler et al., 2007) showed a different result, reporting elevated frequencies of sensory and motor behaviors in children with ASD as young as 2 years compared with children with developmental disorders without ASD. Measurement issues might underlie these differences. As RRBs are common in typical development, parents may be unaware when they are interviewed using the ADI-R of atypicality in their child's behavior. Richler et al. (2010) included both parent report and also observations in their measurement of RRB.

As mentioned earlier, it is important to consider research evidence that is based on observational methods compared with parent report. In contrast with the evidence from the ADI-R parent interview, research using standardized observation methods suggests that the first signs of atypicality may appear earlier than age 3-4 years and may be seen by 18-24 months or before (Ozonoff et al., 2008; Rogers, 2009; Yirmiya & Charman, 2010). These signs of repetitive behavior include repetitive actions with objects (e.g., spinning, tapping, banging, rolling objects) and body (e.g., rubbing body, hand and finger mannerisms (Watt, Wetherby, Barber, & Morgan, 2008), unusual nonfunctional exploration of objects, differences in sensory reactions (unusual visual fixations on objects), object manipulation (spinning) (Ozonoff et al., 2008), and motor stereotypies, particular in arm and finger movements (Loh et al., 2007; Ozonoff et al., 2008). Some of these studies (Ozonoff et al., 2008) found higher frequencies of repetitive behaviors in high-risk autism sibling samples. Future research should establish systematic comparisons between observation and questionnaire methods to account for this discrepancy, especially if certain RRBs are emerging as potential candidates for early markers for autism in infancy.

It is important to understand how RRBs in typical development change across time in order to compare atypical trajectories in children with autism. There is very little longitudinal evidence on the developmental change of RRBs in typical infancy through to middle childhood. With respect to observational data, Thelen's (1979, 1981) work showed that repetitive stereotyped movements are extremely common in typical development in the first year of life; and, although their overall frequency reduces toward the end of the first year, they still remain relatively high and some stereotypies, particularly repetitive arm movements, begin to increase at around 9 months. McDonald et al. (2007), in an observational study, found that older children with ASD (aged 4 years) had higher frequencies of stereotyped behaviors than younger children with ASD (aged 2 and 3 years). This is a reversal of the age trend found in typically developing children, and also supports the idea that some children with ASD are delayed in their development of RRBs. A study by Iverson and Wozniak (2007), examined Thelen's claim that the developmental changes in RRBs are coordinated with the development of vocal and motor systems in the first 18 months of life. They found that in typically developing infants, repetitive arm and finger movements increased sharply during the month of babbling onset compared with either the month before or after this onset. For infant siblings of children with ASD who are at risk for ASD themselves, this developmental effect of repetitive arm movements linked to babbling was significantly less marked. Furthermore, these siblings were delayed in both developmental milestones and postural stability.

More evidence is available on the developmental trajectories of both typical and atypical development when parent report methods are used. For example, a questionnaire study with parents of typically developing children by Leekam et al. (2007) found that RRBs were common across a range of RRB types (motor, sensory, routines, interests) in 2-year-olds, with every item endorsed for 18%–30% of the sample. In the same sample, at 15-months, the frequency of motor behaviors, particularly hand movements, such as repetitively fiddling with toys, was even higher, with up to 60% endorsement (Arnott et al., 2010). Future research on typical development will provide important benchmarking of standardized typical RRB milestones that will help researchers and clinicians to identify which RRBs are appropriate or problematic at particular ages or cognitive development levels and what would be expected for a child's mental age according to typical pathways.

With respect to developmental change and mental age, research using parent report, particularly the ADI-R, shows that low levels of intellectual ability, language, and adaptive behavior are highly associated with the quantity of RRBs. Developmental and intellectual skills also help to mediate improvements in RRBs and influence a child's potential future RRB pathways (Berkson & Tupa, 2000; Hus et al., 2007; Szatmari et al., 2000). However, it is important to be aware that the relation between developmental skills and RRBs changes with age. In a cross-sectional study to examine interactions between IQ and age, Bishop et al. (2006) examined RRBs from the ADI-R in a sample of children with ASD (15 months to 12 years). Higher nonverbal IQ was positively associated with circumscribed interests but negatively associated with other RRBs, such as use of objects, resistance to change, rituals, and attachments to objects. In the case of these latter RRBs, their negative association with IQ increased with age so that by age 7 the association between RRB and IQ was at its strongest.

However there were some exceptions to this. Some low-frequency behaviors, such as sensitivity to noise, abnormal idiosyncratic responses, difficulties with change in routine, and resistance to change were not associated with IQ at any age. In addition, by 7 years, compulsions and rituals were found in both higher and lower ability groups.

In the only longitudinal study to date to examine repetitive behaviors in ASD across childhood, Richler et al. (2010) followed children's RRBs across four age periods, age 2, 3, 5, and 9. They also included separate analysis of repetitive sensory and motor behaviors and insistence on sameness behaviors. Results showed that repetitive sensory and motor behaviors remained high across these age points though they decreased in children with higher nonverbal IQs by age 9. In contrast, IS behaviors (e.g., routines and rituals), started at a low level at age 2 years and moderately increased in severity. IS behaviors were not associated with IQ scores at 2 years, but higher IS scores were associated with older ages and with milder social and communication impairments. This pattern is consistent with what would be expected in typical development but at an extremely delayed level. Children with or without ASD who have mental ages below age 2 years will show few IS behaviors, as these behaviors involve greater sophistication and awareness than simple motor behaviors. An interesting issue for future research is the extent to which this delay varies from the typical trajectory, especially in high functioning children with ASD, and in comparison with developmentally delayed children without ASD.

Change between childhood and adulthood. traces changes in RRB across childhood to adulthood gives some insights into lifetime change, but these studies are often limited by their design and methodology, with no known longitudinal studies sampling behaviors at multiple time points and existing studies tending to sample across very wide age ranges. However, Esbensen et al. (2009), using the Repetitive Behaviors Scale-R, found evidence of lower levels of RRBs in adults compared with children. Individuals with ASD who also had intellectual disability showed the highest levels of stereotyped and self-injurious behavior but did not differ on the other dimensions. A study by Murphy et al. (2005) examined frequency of different types of RRB in a follow-up interview study after 12 years and found that abnormal motor behaviors and responses to sensory stimulation reduced with age while routines and resistance to change did not. This is consistent with the notion of low and high level of RRBs being associated with age and level of functioning. Other research has examined change in RRBs in relation to other core social and communication symptoms. Fecteau, Mottron, Berthiaume, and Burack (2003), in a retrospective ADI-R follow-up of 4- to 5-yearolds when they reached young adulthood, found that improvement for RRB was not as great as for social and communication impairments. However, RRBs did improve for some items, such as repetitive use of objects. It would be interesting to know how the environmental experiences of these varying samples may have influenced patterns of symptom reduction.

To summarize, there is potential for improvement in RRBs across time, but this potential will depend on nonverbal IQ, language competence, and adaptive functioning. Evidence for developmental change is sparse, and further observational research in combination with parent report measures will help to identify if there are reliable differences in topography and/or frequency.

Meanwhile, it is important to take account of the fact that in typical development, different types of RRB, that is, low-level repetitive sensory and motor behaviors and high-level insistence on sameness behaviors, show different trajectories (Evans et al., 1997; Leekam et al., 2009). In children with ASD, both onset of, and reductions in, RRB may appear at a much later age than expected in typical development but may be consistent with other aspects of the child's developmental level. Planning of intervention and management of RRBs therefore needs to consider these behaviors against the profile of the child's cognitive and behavioral development in other domains of functioning.

Intervention

Intervention studies provide the opportunity to study potential for change in repetitive behavior as a result of a particular manipulation in the child's physiology or environment. Surprisingly, RRBs are less likely to be targeted in interventions compared with social and communication domains despite their prominence as management challenges and barriers to adaptive learning and as sources of burden for families. In this section we review recent research interventions that have relevance for RRBs in ASD: pharmacological treatments and behavioral intervention.

Pharmacological treatment. The groups of medications that have been most widely used in the pharmacologic treatment of ASD are atypical antipsychotics, serotonin reuptake inhibitors, and opioid antagonists. Research evidence on the effectiveness of each of these three classes of treatments indicates a limited degree of improvement for symptoms that are related to repetitive behaviors and, importantly, in some cases, adverse side effects.

Atypical antipsychotics (clozapine, risperidone, olanzapine, quetiapine, ziprasidone, aripiprazole) are pharmacologically serotonin and dopamine antagonists. They have largely replaced typical antipsychotics (such as haloperidol) in the treatment of various symptoms in ASD because of the significantly lower risk of extrapyramidal syndrome and tardive dyskinesia. Several studies in the last 10 years have examined the effects of resperidone in children with ASD. In one study (McCracken et al., 2002), 101 children (mean age, 8.8 years; range, 5-17 years) were randomly assigned to a risperidone or placebo group. After 8 weeks, risperidone was effective in reducing self-injurious behaviors, tantrums, and aggression as measured by the Aberrant Behavior Checklist, which measures irritability, social withdrawal, stereotypy, hyperactivity, and inappropriate speech. Children in the McCracken et al. (2002) study who showed positive response to risperidone were entered into a further two-part study by the Research Units on Pediatric Psychopharmacology (RUPP) Autism Network (2010). Part 1 was open-label risperidone treatment that lasted 16 weeks, and Part 2 was a randomized, double-blind, placebo substitution study of risperidone withdrawal that lasted 8 weeks. Risperidone had persistent efficacy for the above mentioned symptoms. However, after discontinuation, there was a rapid relapse in symptoms (RUPP, 2005). McDougle et al. (2005) used a database from these two studies (McCracken et al., 2002; RUPP, 2010) to test whether risperidone was superior to placebo for RRBs and whether it improved social relatedness and communication. Results showed that treatment with risperidone led to significant reduction in RRBs. There was a significant reduction in sensory and motor behaviors measured with the Ritvo-Freeman Real Life Rating

Scale and significant decrease in Children's Yale-Brown Obsessive Compulsive Scale (CY-BOCS; Goodman, Price, Rasmussen, & Mazure, 1989) and the maladaptive behavioral domain of the Vineland Adaptive Behavior Scales (Sparrow, Balla, & Cicchetti, 1984), but no significant effects were found on the other two core impairments (social interaction and communication). This treatment effect was maintained for 6 months. Another randomized, double-blind study (Shea et al., 2004) of 79 children aged 5-12 years with PDD, reported that risperidone was efficient in reducing symptoms in all subscales of the Aberrant Behavior Checklist, including irritability, lethargy and social withdrawal, stereotypic behavior, hyperactivity/noncompliance, and inappropriate speech, with the greatest improvement for irritability and hyperactivity. However, other types of RRBs (e.g., routines, compulsions) were not measured. Adverse effects of risperidone treatment in all these studies were relatively mild. The most common and significant adverse effect was weight gain in children, although a range of other side effects have also been reported, including increased somnolence (Weeden, Ehrhart, & Poling, 2009). Other atypical antipsychotics (Quetiapine, Ziprasidone) have been shown to be much less effective and with serious side effects (Cohen, Fitzgerald, Khan, & Khan, 2004; Findling, 2002; Martin, Koenig, Scahill, & Bregman, 1999).

Serotonin reuptake inhibitors (SRIs) are quite widely prescribed medications in individuals with ASD (Aman, Lam, & Van Bourgondien, 2005). The rationale for use of this group of medications lies in the hypothesized serotonin dysregulation in ASD and their effectiveness in reducing repetitive and obsessive behaviors in OCD (Soorya, Kiarashi, & Hollander, 2008). This group of SRIs includes agents such as clomipramine that inhibit reuptake of both norepinephrine and serotonin at the level of presynaptic membrane and agents that only inhibit reuptake of serotonin (selective serotonin reuptake inhibitors; SSRIs: fluoxetine, fluvoxamine, sertraline, paroxetine, citalopram).

Clomipramine was the subject of a number of early research studies that reported adverse reactions in both adults and children (Brodkin, McDougle, Naylor, Cohen, & Price, 1997; Sanchez et al., 1996). Since 1999, research has been focused mainly on SSRIs, particularly fluoxetine and citalopram. Hollander et al. (2005) evaluated fluoxetine in 45 children and adolescents aged 5-17 years for reduction of RRBs as measured by the CY-BOCS (Goodman et al., 1989). They found low-dose liquid fluoxetine superior to placebo with moderate to large effect sizes on the compulsion scale but not on the Clinical Global Impressions autism score. In a very recent, large-sample study report, King et al. (2009), using multiple sites and 5- to 17-year-old cases, found that another SSRI, citalopram, did not have any positive effects on behavior and was significantly associated with adverse side effects. An important additional finding was that around one third of placebo treated cases showed some behavioral improvement. Soorya et al., (2008) reviewed the literature on use of SSRIs, as well as risperidone and oxytocin, for RRBs in ASD and concluded that findings were mixed for commonly prescribed medications (both SRIs and SSRIs), with more research needed on pharmacological management of RRBs and special emphasis on individual variability in treatment response and on defining and evaluating hyperactivity, which is a common adverse effect of treatment with SRIs. A more recent review by Wink, Erickson, and McDougle (2010) concluded that efficacy of serotonin reuptake inhibitors is limited. In addition, Williams, Wheeler, Silove, and Hazell (2010) reviewed seven randomized, controlled studies of selective serotonin reuptake inhibitors in ASD and concluded that the evidence showed that SSRIs are not effective in children with ASD and that they can cause serious adverse effects.

Two reviews have reported on the effectiveness of opioid antagonists (Symons, Thompson, & Rodriguez, 2004; ElChaar, Maisch, Gianni, & Wehring, 2006), both of which concluded that naltrexone is effective for self-injurious behavior. However, one case study has reported increase in self injurious behavior after naltrexone treatment (Benjamin, Seek, Tresise, Price, & Gagnon, 1995).

Although research on the pharmacological treatment in ASD has shown that medications can be effective in reducing aggression toward self and others, inattention, motor hyperactivity, and other behavioral problems, achievements in improving core symptoms have not been encouraging. While there is evidence that some drugs, such as risperidone, might be effective in reducing sensory and motor behaviors and OCD-type behaviors as measured by the CY-BOCS, the direct or indirect mechanism for improvement is not clear since the main effects of risperidone relate to irritability and aggression rather than RRBs in particular. There is a surprising disconnection between the pre- and postmeasures of RRB taken in pharmacological intervention studies compared with standard measures of RRB used in most other research studies reported elsewhere in this article (see Table 1), and more robust and careful measurement of different types of RRB using standard measures would increase the usefulness of future research.

Behavioral intervention. Skill-based behavioral strategies that specifically target RRBs have been summarized in a number of review articles. A meta-analytic review of behavioral interventions by Horner, Carr, Strain, Todd, and Reed, (2002) covered research published since 1988 with special attention to behavioral interventions between 1996 to 2000, in children up to 8 years old with developmental disabilities including autism. Most frequently, these studies were of stereotypy, self-injury, and aggression. Horner et al.'s (2002) review consisted of only nine studies, most of which were single case or small-sample studies, with a consequent low overall sample size in the review. The authors provided a set of recommendations in which they noted the higher rates of RRBs in unstructured situations by comparison with those in highly structured situations.

Three other subsequent reviews, mostly of single case studies, have concluded that skill-based behavioral interventions (interventions aiming to increase particular behaviors), while not universally effective, can be successful in reducing maladaptive behaviors, including RRBs. A review by Rapp and Vollmer (2005) concluded that the use of antecedent behavioral interventions, such as general and specific environmental enrichment as well as consequent intervention (displacement of reinforcement), can lead to the reduction in stereotypy. Patterson, Smith, and Jelen (2010) included 10 single-case intervention studies (17 individuals with ASD in total), all of which were behaviorally based, in their review. This analysis showed that noncontingent reinforcement (the most frequently used among the studies included in analysis) was ineffective, while differential reinforcement of alternative behavior combined with extinction procedures as well as response interruption were effective. In addition, a recent, comprehensive research review of treatments for ASD aimed at distinguishing

established, emerging, unestablished, and harmful treatments the National Autism Center's (2009) National Standards Project concluded that RRBs could be decreased with the implementation of several of the established behavioral packages. In all of the above mentioned reviews, the critical importance of functional analyses in identifying and analyzing RRBs and their contextual influences (for example, whether they occur when the child is unoccupied and self-stimulating or being challenged in some way) for successful intervention has been stressed.

While RRBs are frequently the target of specific skill-based behavioral interventions, research on the effectiveness of comprehensive interventions (interventions that address numerous aspects simultaneously) have rarely included RRBs as an outcome measure (Howlin, Magiati, & Charman, 2009). Because of this, we carried out a separate computerized literature search (MEDLINE, PsychINFO, PubMed, and Web of Science) in order to specifically identify group-based studies on comprehensive interventions that reported changes in repetitive behaviors in ASD samples. We included behavioral, developmental, and educational interventions only. Unestablished behavioral treatments, as defined by the National Autism Center's (2009) National Standards Project, were excluded. The seven group-based behavioral intervention studies in the last 10 years that have specifically targeted RRBs as an outcome measure are reviewed here.

Five of the seven studies were based on the principles of Applied Behavioral Analysis (ABA) and four of these reported improvements in RRB. The first of these, Sallows and Graupner (2005), compared an ABA program based on Lovaas et al. (1987) with parent-directed ABA therapy program. Children averaging 33-34 months, were matched on pretreatment IQ and randomly assigned to a clinic-directed group (N = 13), or parent directed group (N = 10) and received over 30–39 hrs per week treatment across 4 years with additional supervision. The parent-directed ABA group performed as well as the clinic-directed group, with significant improvements on RRBs (ADI-R) in both groups for those children described as "rapid learners," with increases in language and adaptive functioning and 48% achieving full-scale IQ in average range. Three studies by Ben Itzchak and colleagues also reported improvements in RRBs. Ben Itzchak and Zachor (2007) did not include a control group, so this intervention is difficult to evaluate. However, a more recent study (Ben Itzchak, Lahat, Burgh, & Zachor, 2008) followed Howlin et al.'s recommendations by including posttest repetitive behaviors as well as IQ measures. In this study, ABA was compared with other developmental treatments (speech, occupational, physical therapy) in 44 children diagnosed with autism; mean age, 27 months. Treatment (45hrs/week) was delivered in center based settings for 12 months. There were significant improvements in RRBs (measured via ADOS) and IQ improvements correlated significantly with the reduction in RRBs. There were significant improvements in social, communication, and play behaviors (measured via ADOS), which were again related to higher pretreatment cognitive levels. In another study, Ben Itzchak and Zachor (2009; sample overlap not stated) assigned 40 children diagnosed with autism to an ABA program and compared them with 28 assigned to eclectic treatment. Treatment was delivered for 45 hrs per week in center-based settings for 12 months for both groups. Significantly more children in the ABA group improved their diagnostic classification (i.e., moving to a less severe category) and had significant improvement

in ADOS measured RRBs. The improved group also had higher pretreatment verbal scores and better outcome scores on cognitive and adaptive skills. Finally, one study by Eldevik, Eikeseth, Jahr, and Smith (2006) that used ABA (N=15) compared with eclectic treatment (N=15) in children below the age of 6 years old, for 2 years (12 hrs per week), did not find significant improvements in RRBs in either group, despite some improvements in ABA relative to eclectic treatment for IQ, language, and communication. The poorer outcomes of the lower dose ABA studies reviewed here deserve notice.

It should be noted that while early intervention programs based on ABA focus on positive reinforcement and learning of alternative adaptive behaviors, other intervention programs take a different approach by focusing on enhancing the affective experience for the child with the goal of reducing over arousal and anxiety. One intervention that combines ABA techniques with a developmental affective relationship-based approach and is delivered by trained parents is the Early Start Denver Model (Rogers & Dawson, 2009). This is also an intensive method of intervention, delivered for 25-40 hrs per week over 2 years and based on developmentally sensitive assessment. A randomized control trial study of 48 infants who started this intervention at 18–30 months (Dawson et al., 2009) found significant improvements in adaptive behaviors and IQ and a change toward a milder PDD diagnosis over 2 years. The Repetitive Behavior Scale (Bodfish et al., 1999) used to measure RRBs during this intervention did not show any specific change across time. However, RRBs were not specifically targeted for intervention in this program.

Another randomized control study that also used an affective relationship-based approach and involved training of parents is the Parent Mediated Communication-focused treatment (PACT) trial (Green et al., 2010). This intervention works with parents to increase their responsiveness to acts of sharing attention and eye-gaze and helps to adapt communication to the child. After an initial orientation meeting, PACT group-families attended 2-hr biweekly clinic sessions and received a maximum of 18 sessions in 12 months. The PACT group also received treatment as usual (TAU). 152 children between 2 and 4 years and 11 months, were randomly assigned to PACT (N = 77) or TAU (N = 75). RRBs unexpectedly improved in both groups, a result that will guide future analysis of differential treatment response. Positive treatment effects for parental synchronous response to child, child initiations with parent and for parentchild shared attention were found. However, language and adaptive functioning treatment effects were small.

Beyond the studies discussed above, only a relatively small number of evaluation studies are sufficiently rigorous to underpin confident conclusions about treatment effects (see National Autism Center, 2009). The literature consists mainly of small-group or case studies of behavioral treatment using functional assessment and often predominantly targeting social skills. Strategic small *N* studies can however be informative. For example, Loftin et al. (2008) took the approach of assessing whether reduction in repetitive motor behaviors could be achieved in a training program that aimed to increase peer social interaction in three students with autism in a regular elementary grade classroom. Instruction in specific social skills produced increases in social initiations with collateral reductions in repetitive motor behaviors (see also Lee, Odom, &

Loftin, 2007, for additional demonstrations of this effect). The implication is that the competing stimulation of peer interaction, which was reinforcing for these individuals, reduced the need for repetitive motor behaviors. In general it seems that as individuals with ASD acquire new skills and competencies, RRBs appear to reduce in frequency. Such findings may be linked to arousal theories of RRB (Hutt & Hutt, 1965; Ornitz & Ritvo, 1968) with positive effects on modulation of arousal emerging in more competent individuals, as well as to the influence of more stimulating environments in eliciting more adaptive behavior. Such findings also address the debate raised earlier about the nature of the relation between RRBs and social-communication impairment, supporting the view that nonsocial and social impairments may be intrinsically linked for children with ASD, even if their distal etiologies are independent.

Summary

In this section we asked whether RRBs have the potential to spontaneously change across time and the extent to which specific pharmacological and behavioral interventions lead to improvement in RRBs. Whether referring either to spontaneous change or to change due to intervention, the answer is that changes can be seen in restricted repetitive behaviors. However, the potential for change in RRBs is affected by age and cognitive and language ability (see Table 1). There is also evidence that delays in both adaptive functioning and in other domains (motor skills, symbolic ability, social interaction) are associated with RRB (Cuccaro et al., 2003).

There is encouraging support for more targeted behavioral approaches to intervention involving careful identification of triggers and functions. At present, it seems that comprehensive interventions hold the most promise for successful treatment of ASD especially if targeted in the early years of development. Delivering early interventions during sensitive periods in brain development could have positive impacts on the developmental trajectory of neural systems in ASD. Development of the full clinical presentation of ASD together with associated problematic behaviors could be prevented if more appropriate behavioral patterns are established early (Dawson, 2008). However, there is still a considerable way to go before reaching this goal and improving the methodological quality of studies is challenging but essential. For example, in a systematic review, Patterson et al. (2010) evaluated the methodological quality of 10 studies of behavioral intervention for stereotypic and repetitive behaviors in ASD, using the American Academy for Cerebral Palsy and Developmental Medicine's scale. This scale, developed by Romeiser Logan, Hickman, Haris, & Heriza (2008), evaluates the methodological quality of singlesubject studies as either strong, moderate, or weak, using criteria based on (a) description of participants and settings, (b) independent and dependent variables, and (c) design and analysis. Patterson et al. (2010) found that out of 10 case studies that they included in their analysis, 9 studies had a rank of moderate quality and one was rated as weak, illustrating the methodological shortfalls in this literature. Greater focus is needed on the oft-noted individual variability in children in order to identify which children benefit and which do not. Inclusion of within-subject child, family, and environmental variables will be needed.

Pharmacological treatment has proven to be effective for treating irritability, inattention, and aggression in some cases; and, since these behaviors can present serious obstacles for the delivery of behavioral interventions, medications can be used to complement behavioral treatment in these cases. However, research examining the combined effects of pharmacological and behavioral interventions is lacking and is greatly needed (Weeden et al., 2009). Although effects of pharmacologic treatments on core symptoms of ASD have been disappointing thus far, this may be related to the limited understanding of the neurochemical basis of autism in the field and lack of integration across different disciplines. Improvements in our understanding of this area may in time result in the development of wider range of intervention options during early development.

One goal in behavioral intervention is to increase the repertoire of social and behavioral skills beyond restricted and repetitive behaviors, thereby loosening rigidity, facilitating more flexibility, and reducing repetitive behavior patterns. Another goal is to achieve more optimal regulation of arousal and anxiety states through interventions that target these factors. The most effective method for treating anxiety in children is Cognitive Behavioral Therapy (CBT). This has also been shown to be effective in reducing anxiety in children with ASD (e.g., Sofronoff, Attwood, & Hinton, 2007; Wood et al., 2009), but currently we do not know the extent to which it also serves to reduce RRBs. We await research findings demonstrating the association between reduced anxiety and reduced RRBs in ASD.

To fully understand the extent to which RRBs can change, we need to be careful to compare RRBs in ASD with those seen in the general population, and hence we recommend systematic assessment of developmental level. Since similarities and overlaps between ASD and other clinical or handicapped groups lie in the RRB domain rather more than in the social and communication domain, we also need to understand what contribution different early abilities make to different types of trajectories characteristic of children with ASD, OCD, ADHD, language impairment etc. Another way to approach change is to study very systematically what works and what does not work in particular interventions and which children are affected. With this in mind, a systematic, experimental approach is suggested that specifically targets RRBs within intervention programs and subsequently measures RRB outcomes along with outcomes for other behaviors, including social interaction, communication, imaginative activities, and sensory sensitivities.

Conclusions

The last decade of research has advanced our knowledge about restricted and repetitive behaviors in ASD at both theoretical and empirical levels. As Table 1 shows, there has been a shift across the last 10 years toward greater consistency of measurement and increasing use of subscales and factor analysis to interpret findings. There has also been growing awareness of how individual and developmental factors impact on the severity of RRBs and on their potential for change. While knowledge about RRBs in ASD still remains seriously limited and the future decade is likely to produce more substantial understanding, some important points have consistently emerged from the review that move our understanding forward and provide pointers to future research.

First, we know from factor analytic studies that there are several dimensions or subgroupings of RRBs that emerge reliably in children with ASD, in particular repetitive sensory and motor behaviors (RSM) and IS behaviors. Second, it is clear that the forms of RRBs seen in ASD are also found in many other neurodevelopmental and genetic conditions, although particular types of RRB may vary in frequency from those in ASD. Third, RRBs are part of normal development especially in infancy, and we know that RRBs are affected by age and developmental level in ASD. Fourth, although we do not know exactly why RRBs happen, explanations are advancing in the study of neurobiology, neurocognition, and developmental psychology. Currently, the importance of corticostriatal circuits, genetic vulnerability, environmental restriction, arousal, and development of goal-directed action may help us understand the distal origins of RRBs, while stress, anxiety, and arousal, as well as environmental deprivation, form important immediate triggers for RRBs deserving of more research. In this regard, recent mouse models may provide important clues to the role of environmental stimulation, or the lack of it, in relation to the development of RRBs.

These points raise potential for new research directions. One clear direction for future research is to clarify the relation between RRBs and other core social and communication impairments. This is important because the co-occurrence of social-communication impairments and RRBs is what distinguishes ASD from other clinical groups. Although contested by some, we believe that RRBs are central to ASD. We argue this case on the grounds that RRBs are important to development, with impact and implications for other aspects of functioning, that they are a stand-out feature of presentation in the vast majority of children with ASD, and that they are related to differing levels of severity of handicaps. Furthermore, evidence shows that RRBs are extremely basic behaviors found across the animal species and that they may arise in the presence of social isolation and sensory deprivation. It is likely then that higher level social, communicative and symbolic difficulties may be additional to RRBs in developmental terms.

This leads us to our next point, that the focus needs to turn toward a developmental approach to the study of restricted and repetitive behaviors. Following Thelen's developmental account, we believe that it may be helpful to think of RRBs as developmentally immature responses that have been maintained more strongly within the behavioral repertoire of individuals with ASD. These RRBs may serve various forms of adaptive function; however, they are not serving an appropriate developmental function for neural development and voluntary motor control. If we think about RRBs as immature or inappropriate responses, this helps us to redirect the search for specific causes at a neurobiological or neuropsychological level toward an approach that first takes account of developmental explanations. However, we still need to know much more about the link between these early RRBs and the class of higher level RRBs, such as routines, rituals, and special interests, which are classed as IS. Future longitudinal research will help to inform us about the dependence and independence of these two groups of behavior in relation to each other.

New research is emerging that will help to direct the path into the next decade of research on RRBs. This work will help clarify the contribution of different brain areas to the development of different types of RRBs (Langen, Durston, et al., 2011; Langen, Kas, et al., 2011), enable different brain imaging techniques to be combined to examine the neurophysiology of RRBs at different levels (Thakkar et al., 2008), and test developmental trajectories in brain and RRB relations (Langen et al., 2009). Currently, however, we still know relatively little about the developmental trajectories of RRB in either typical development or in ASD, although the picture suggests that RRBs do change and reduce across time even if there is no direct intervention. This offers potential promise for Lewis and Kim's (2009) ideas of plasticity and neuroadaptation. Nevertheless, RRBs can be pervasive and persistent and are hard to treat by means of interventions currently available. To date, findings indicate that pharmacological interventions provide only limited benefits; and, while behavioral interventions are more promising, both types of intervention need more development and evaluation with larger numbers of children. We strongly recommend a focus on early identification and intensive early intervention for infants and young children with autism before RRBs become entrenched and hard to alter. We regard this recommendation for early intervention as essential to more effectively tackle all varieties of repetitive behaviors, in the same way as it is for amelioration of the core social and communication deficits.

We identified three themes or issues for our review: definition, cause, and change. Research addressing each of these three themes has so far not been well connected. Yet effective intervention requires attention to all three of these issues. Intervention design should be guided by theoretical predictions about cause and consequence of RRBs, be able to identify and intervene on the proximal and distal factors that influence RRBs, be informed about what degree of change or improvement is possible and be able to measure this change accurately. In setting signposts for future research, we recommend further consolidation of different research areas.

First with respect to definitions, we recommend that there be greater consolidation in research on methodology. A consolidated focus should include rigorous evaluation of different techniques, examination of RRBs beyond the items that are recorded for a clinical diagnosis (e.g., ADI-R) and also careful comparative work of parent report measures and observation techniques. Further investigation of the distinction between, and measurement of, groupings of RRB (RSM and IS), obsessions and preoccupations, and special interests also need more attention.

With respect to cause and change, we recommend further conceptual work to build on existing explanatory accounts. This will require researchers to work across different disciplines to establish how neurobiology can account for developmental change. To achieve this, as Langen, Durston, et al. (2011) and Langen, Kas, et al. (2011) propose, studies are needed that can trace neurobiological changes alongside behavioral changes. We believe that this work needs to begin with early development in infancy, using typical development as a guide. Detailed psychological research on the origins of goal-directed motor behavior, cognition, and motivation-arousal should inform knowledge of the origins and development of basal ganglia, striatal and forebrain structures. Psychological models of dynamic, transactional and cumulative change will also help to improve understanding of the dysfunction in feedback that may be occurring within corticostriatal circuits (Langen, Durson, et al., 2011).

The need for well formulated intervention demands further research that connects hypotheses about distal causes to predictions about proximal causes. Here clinical researchers can work together with researchers trained in developmental psychology and neurobiology researchers to identify profiles of triggers and functions (e.g., the effect of being unoccupied) and types of RRB (e.g., the selection of particular special interests) that may be distinctive to particular individuals. This work will help to build understanding of how RRBs emerge across time. A prime candidate for explaining the ongoing shape and form of particular RRBs is level of ability; this will impact upon the relative balance of sensory and motor behaviors and the type of special interests selected. But other candidates are also becoming evident from research, though evidence is less established. These include arousal, anxiety, and motivation. We recommend that researchers working in clinical fields of anxiety beyond ASD continue to join forces with researchers working in autism to help explain the role of anxiety in the neurobiology and the development of RRBs and in the focus of treatment approaches.

While the major work ahead for the next decade might seem daunting, taking a broad, multidisciplinary approach will enable a stronger conceptual framework to be developed. We look forward to an exciting future decade of research on restricted and repetitive behaviors in autism.

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