Autism and Child Psychopathology Series
Series Editor: Johnny L. Matson

Eynat Gal
Nurit Yirmiya
Editors

Repetitive and Restricted Behaviors and Interests in Autism Spectrum Disorders

From Neurobiology to Behavior

Springer

Autism and Child Psychopathology Series

Series Editor

Johnny L. Matson, Department of Psychology,
Louisiana State University, Baton Rouge, LA, USA

More information about this series at http://www.springer.com/series/8665

Eynat Gal • Nurit Yirmiya
Editors

Repetitive and Restricted Behaviors and Interests in Autism Spectrum Disorders

From Neurobiology to Behavior

Springer

Editors
Eynat Gal
Department of Occupational Therapy
University of Haifa
Mount Carmel, Haifa, Israel

Nurit Yirmiya
Department of Psychology
Hebrew University of Jerusalem
Mount Scopus, Jerusalem, Israel

ISSN 2192-922X ISSN 2192-9238 (electronic)
Autism and Child Psychopathology Series
ISBN 978-3-030-66444-2 ISBN 978-3-030-66445-9 (eBook)
https://doi.org/10.1007/978-3-030-66445-9

This Springer imprint is published by the registered company Springer Nature Switzerland AG
The registered company address is: Gewerbestrasse 11, 6330 Cham, Switzerland

Contents

Contributors

Ayelet Ben-Sasson Department of Occupational Therapy, University of Haifa, Mount Carmel, Haifa, Israel

Omri Bing The Paul Baerwald School of Social Work and Social Welfare, The Hebrew University of Jerusalem, Jerusalem, Israel

Bob Shapell School of Social Work, Tel Aviv University, Ramat Aviv, Israel

Simon M. Bury Olga Tennison Autism Research Centre, School of Psychology and Public Health, La Trobe University, Melbourne, VIC, Australia

Cheryl Dissanayake Olga Tennison Autism Research Centre, School of Psychology and Public Health, La Trobe University, Melbourne, VIC, Australia

Esther Dromi Constantiner School of Education, Tel Aviv University, Ramat Aviv, Israel

Roni Enten-Vissoker Individualized Bio-Nutrition Clinic, Haifa, Israel

Eynat Gal Department of Occupational Therapy, University of Haifa, Mount Carmel, Haifa, Israel

Parisa Ghanouni Dalhousie University, Halifax, NS, Canada

Yael Goldfarb Department of Occupational Therapy, University of Haifa, Haifa, Israel

Gal College, Loewenstein Rehabilitation Hospital, Raanana, Israel

Darren Hedley Olga Tennison Autism Research Centre, School of Psychology and Public Health, La Trobe University, Melbourne, VIC, Australia

Tal Jarus University of British Columbia, Vancouver, BC, Canada

Alison E. Lane School of Health Sciences, The University of Newcastle, Callaghan, NSW, Australia

Susan R. Leekam Wales Autism Research Centre, School of Psychology, Cardiff University, Cardiff, Wales, UK

Omer Linkovski Department of Psychiatry and Behavioral Sciences, School of Medicine, Stanford University, Stanford, CA, USA

Department of Medical Neurobiology, The Hebrew University-Hadassah Medical School, Jerusalem, Israel

Aviva Mimouni-Bloch The Pediatric Neurology and Developmental Unit, Loewenstein Rehabilitation Medical Center, Raanana, Israel

Sackler Faculty of Medicine, Tel Aviv University, Tel-Aviv, Israel

Daniel Offen Sagol School of Neuroscience, Tel Aviv University, Tel Aviv, Israel

Sacklar School of Medicine, Department of Human Genetics and Biochemistry, Tel Aviv University, Tel Aviv, Israel

Alona Oren The Pediatric Neurology and Developmental Unit, Loewenstein Rehabilitation Medical Center, Raanana, Israel

Nisim Perets Sagol School of Neuroscience, Tel Aviv University, Tel Aviv, Israel

Shani Poleg Sackler Faculty of Medicine, Tel Aviv University, Tel Aviv, Israel

Alliyza Esther Qin Shi Lim College of Education, Psychology and Social Work, Flinders University, Adelaide, SA, Australia

Cory Shulman The Autism Center of Hebrew University, The Paul Baerwald School of Social Work and Social Welfare, The Hebrew University of Jerusalem, Jerusalem, Israel

Kevin Stephenson Department of Pediatrics and Psychology, Nationwide Children's Hospital, Columbus, OH, USA

Mirko Uljarević Melbourne School of Psychological Sciences, University of Melbourne, Melbourne, VIC, Australia

Maya Yaari Olga Tennison Autism Research Centre, School of Psychology and Public Health, La Trobe University, Melbourne, VIC, Australia

Nurit Yirmiya Department of Psychology, The Hebrew University of Jerusalem, Mount Scopus, Jerusalem, Israel

Robyn L. Young College of Education, Psychology and Social Work, Flinders University, Adelaide, SA, Australia

Ditza A. Zachor Sackler Faculty of Medicine, Tel Aviv University, Tel Aviv, Israel

The Autism Center/ALUT, Shamir (Assaf Harofeh) Medical Center, Zerifin, Israel

Osnat Zafrani Autism Treatment and Research Center – Association for Children at Risk, Givat Shmuel, Israel

Chapter 1
Introduction: Repetitive and Restricted Behaviors and Interests in Autism Spectrum Disorders

Eynat Gal and Nurit Yirmiya

Autism spectrum disorder (ASD) is a neurodevelopmental condition characterized by the DSM-5 definition by two major developmental deficits: (a) impairments of social-emotional reciprocity, deficits in non-verbal social communication and deficits in reciprocal relationship and (b) restricted and repetitive patterns of behavior, interests or activities (RRBI). The DSM-5 also defines a severity rating applied for both domains of impairment, ranging from "Level 1: requiring support" to "Level 3: requiring substantial support".

The onset of the above-mentioned symptoms occurs in the early developmental period, and these symptoms result in impairment in daily functioning. Research shows that the diagnosis of ASD remains stable across the life span of an individual (Esbensen, Seltzer, Lam, & Bodfish, 2009). Restricted and repetitive patterns of behavior and interests, one of the core criteria for ASD, is an umbrella term for the broad class of behaviors linked by repetition, rigidity, invariance, and inappropriateness to the place and context (Turner, 1997). These behaviors and interests were originally identified by Leo Kanner, who suggested that they are characterized by invariant nature, high frequency, repetition, and desire of sameness (Kanner, 1943).

The DSM-5 (APA, 2013) classifies this domain into four types of symptoms: (1) repetitive and stereotyped speech, movement or use of objects; (2) routines, rituals and resistance to change; (3) circumscribed and restricted interests, and (4) hypo- or hyper-reactivity to sensory input, including unusual sensory interests. Being the defining core features of ASD, repetitive behaviors and movements, rituals or special interests are prevalent across all individuals with ASD. However, the diagnostic

E. Gal (✉)
Department of Occupational Therapy, University of Haifa, Mount Carmel, Haifa, Israel

N. Yirmiya
Department of Psychology, The Hebrew University of Jerusalem, Mount Scopus, Jerusalem, Israel

E. Gal, N. Yirmiya (eds.), *Repetitive and Restricted Behaviors and Interests in Autism Spectrum Disorders*, Autism and Child Psychopathology Series,
https://doi.org/10.1007/978-3-030-66445-9_1

criteria for autism include the presence or history of any two of the four types of RRBI symptoms.

Repetitive and stereotyped speech, movement or use of objects. The first DSM-5 criterion of RRBI relates to repetitive and stereotyped speech, movement or use of objects, such as simple motor stereotypies, lining up toys or flipping objects, echolalia, and idiosyncratic phrases. Repetitive and stereotyped use of language, such as repeating favorite sounds, words, sentences or songs or asking the same question over and over again (APA, 2013), is the first common form of such ritualized behavior. Repetitive verbalization can be communicative in nature, representing the child's processing difficulties and/or their emotional state. Indeed, this criterion was previously included in the social communication part of the Autism definition (DSM-IV; APA, 1994). However, it may have a non-communicative purpose, where a response is not expected.

One of the most common RRBI in ASD is stereotyped movements (SM) or stereotypies, which have been described as patterned movements that are highly consistent, invariant and repetitious; excessive in rate, frequency and/or amplitude; and inappropriate and odd and lack an obvious goal (Turner, 1999). These motor behaviors are not specific to ASD. In fact, they characterize early typical development and usually diminish by 2 years of age (Fyfield, 2014). They, however, differ significantly from aimed and planned movements shown along the lifespan in typically developed individuals. Typical examples of such movements in ASD are rhythmic body rocking in a sitting or standing position, various arm and hand movements such as arm waving and hand flapping, and repetitive pacing and jumping (Schopler, 1995).

Stereotyped movements are often problematic for the observers as they may differ in their degree of "inappropriateness" or "oddness" in various contexts. For example, repetitive leg movement together with concentration may look more "appropriate" than hand flapping in front of the eyes or intensive rocking movements while standing and talking. However, this criterion is prone to subjective and cultural-oriented judgment (Gal, 2011).

The third characteristic of the first RRBI category is the repetitive manipulation of objects, which relates to the repetition of the same motor activity used to manipulate the physical environment. Typical repetitive manipulation of objects among individuals with ASD may include lining up objects, flicking light switches, or displaying repetitive manipulation of an object such as a string, rubber tubing or toy. A child may stack blocks over and over again without demonstrating pride in the accomplishment, dump toys or turn light switches on and off repeatedly. This kind of RRBI characterizes the play of children with ASD, who, for example, rather than playing imaginary play such as a racing or driving game may be preoccupied with spinning the wheels on a toy car (Gal, 2011).

Routines, Rituals and Resistance to Change

The second category of the RRBI criterion of the DSM-5 includes routines, rituals and resistance to change, such as extreme distress at small changes, difficulties with transitions, rigid thinking patterns, greeting rituals and a need to take the same route

or eat the same food every day (APA, 2013). Indeed, many individuals with ASD appear to adhere inflexibly to routines, do not tolerate changes in routine, and suffer from subsequent anxiety when novel events occur unexpectedly. First-hand accounts of individuals with ASD suggest an extreme need for structure and predictability. Changes in the routines may, therefore, cause extreme distress, resulting in insistence on inflexible routines, which, in turn, may serve as a strategy to overcome change-related anxiety. Along with the adherence to routines, many individuals with ASD perform ritualistic behaviors that are often compulsive in nature, such as a demand for consistency in their environment or an apparent compulsion to always act in exactly the same way at a specific time (Schopler, 1995) even if the behavior may be irrelevant and inappropriate to the environment's needs and demands. For example, a child with ASD may feel compelled to repetitively play with water whenever he sees a tap. Another may have a strong need to put things away even when they are still needed. Some insist on eating the same foods at the same time and use the same utensils or eat foods of a specific brand only. These behaviors may reflect the discomfort of the individual with ASD and simultaneously may create great difficulties for their social environment and may negatively affect their ability to participate in everyday life.

Circumscribed and Restricted Interests

The third category of the RRBI criterion is circumscribed and restricted interests such as strong attachment to or preoccupation with unusual objects and excessively circumscribed or perseverative interests (APA, 2013). Indeed, the development of unusual, narrow and circumscribed interests or an obsession or fascination with a particular topic is a hallmark symptom attributed to individuals on the autism spectrum, described as a specific field of knowledge that the individual is passionate about or restricted and circumscribed interests that are abnormal in intensity (Klin, Danovitch, Merz, & Volkmar, 2007). These behaviors may range from unusual activities such as memorizing serial numbers to more typical hobbies such as an intense interest in math. For children, one way through which these interests are shown is attachment to a specific object, often different in nature than the soft and cuddly transitional objects that children with typical development tend to be attached to. Some children with ASD carry these objects around and refuse to part with them. Others may show an extreme reaction—which may look bizarre—whenever they come into contact with a particular object (Volkmar, 2005). Later in life, children with ASD may develop specific interests or a peculiar fascination with subjects rather than objects. Typical examples are phone numbers, weather reports, and commercials. Such interests may be more complex, such as diagrams, maps, or stories by a specific writer (Schopler, 1995). The narrow interests, often pursued to the exclusion of other more appropriate behaviors, can substantially interfere with social functioning and academic success. Coupled with impairments in social development, which is a clear mark of the disorder, the narrow interests create further

"weirdness" and the preoccupations, rigidity, and invariant nature of activities in individuals with ASD prevent them from developing peer relations (Cohen & Volkmar, 1997).

However, these special interests also have the potential to induce motivation as they lead to a positive feeling by engaging in activities associated with the area of interest. For example, a child who obsesses over dates and other numbers may enjoy assisting in a library. In adults with ASD, the aspired goal of matching skills, special interests and jobs, possibly satisfies intrinsic motivation and promotes employees with ASD in various aspects of their jobs and career, however, barriers are high and compromise is often essential (Goldfarb, Gal, & Golan, 2019).

Hypo- or Hyper-Reactivity to Sensory Input/Unusual Sensory Interests

The fourth and final category of RRBI in the DSM-5 is hypo- or hyper-reactivity to sensory input/unusual sensory interests, for example, apparent indifference to pain/temperature, adverse response to specific sounds or textures, excessive smelling or touching of objects, and visual fascination with lights or movement (APA, 2013). While sensory differences have been documented in Kanner's early reports of ASD (Kanner, 1943), they were not included as diagnostic criteria in the following DSMs. However, Over the past two decades, professionals in numerous disciplines have increasingly recognized the sensory features of ASD (Ben-Sasson, Gal, Fluss, Katz-Zetler, & Cermak, 2019; DuBois, Lymer, Gibson, Desarkar, & Nalder, 2017), estimating their prevalence to be 60–95% (Lane, Molloy, & Bishop, 2014; Tomchek & Dunn, 2007), and associating them with repetitive behaviors (Gabriels et al., 2008; Gal, Dyck, & Passmore, 2010; Joyce, Honey, Leekam, Barrett, & Rodgers, 2017). In addition, almost all first-hand accounts of people with ASD include sensory issues (e.g., Chamak, Bonniau, Jaunay, & Cohen, 2008). In the current DSM-5 (APA, 2013) sensory characteristics were re-included as part of the second diagnostic criterion, namely RRBI.

Sensory characteristics also called sensory features, refer to patterns of behavior that are suggestive of differences in the manner in which daily sensory stimuli are processed. These features have often been referred to as impairments in sensory modulation, in which an individual has difficulty regulating and organizing the type and intensity of behavioral responses to sensory inputs to match environmental demands. For example, covering the ears in response to an unexpected sound or failure to respond to a painful stimulus (Schaaf & Lane, 2015). These features can manifest in response to touch, sight, sound, taste, smell, and movement, with many individuals presenting several types of symptoms. Sensory features can be classified into three patterns known as sensory over-responsivity (SOR), such as sensitivity to sounds, touch, and food taste or smells; sensory under-responsivity (SUR), such as failure to notice more salient stimuli and sensation seeking, such as fascination and intense interests in sensory stimuli (Miller, Anzalone, Lane, Cermak, & Osten,

2007). These impairments in sensory modulation are further known to present significant restrictions in participation in daily life activities of people with ASD (Dunn, Little, Dean, Robertson, & Evans, 2016; Schaaf, Toth-Cohen, Johnson, Outten, & Benevides, 2011). In fact, according to both research and first-hand accounts, all RRBI have numerous implications on the lives of people with ASD and their families. For example, Stereotyped movements (SM) may fulfill the inner needs of an individual with ASD, but they often appear bizarre and differ significantly from normal movements, and therefore may challenge children in social play, and academic learning, constituting a significant social barrier (Joosten & Bundy, 2010). "High-level" RRBI, such as insistence on sameness or restricted interests, may appear to be inappropriate and therefore can be socially stigmatizing (Cunningham & Schreibman, 2008). Moreover, they may pose difficulties in the process of finding and maintaining employment and may present challenges in interactions with employers and co-workers (Weissman-Nitsan, Schreuer, & Gal, 2019).

This book starts by describing developmental and neurobiological aspects of RRBI, moves on to relations with other ASD characteristics and participation in everyday life, all the way to assessment and intervention which addresses RRBI along the life span.

We set the tone for the book by including the opinions of individuals with ASD speaking about their RRBIs in the chapter titled "It's in my Nature – Subjective Meanings of RRBIs Voiced by Adults with ASD" by Goldfarb, Zafrani, and Gal. In the various diagnostic systems, RRBI are included as core symptoms of ASD. The conventional medical practice is to immediately consider symptoms as behaviors that indicate disease, illness, and psychopathology and therefore, should be treated and eliminated. However, the neurodiversity movement, with its emphasis on partnership with individuals with autism regarding their lives, has resulted in increased public awareness about ASD. First-hand accounts of individuals with ASD speaking about their reveal that RRBI actually help them cope in everyday life and regulate arousal, attention, sensations, and emotions; they also assist in feeling secure and coping with social interactions and unexpected and undesired changes. Thus, RRBIs that are helpful to individuals with ASD should not be treated by others merely as symptoms that need to be eliminated. It is in the eye of the beholder, and the unique experiences and needs of individuals with ASD need to be accepted. As a young woman with ASD commented about her repetitive behavior with her mobile phone: "*No one would tell a handicapped man sitting in a wheelchair to get up and start walking. It's the same thing; it doesn't distract me, just the opposite; to me, it's accessibility*." We hope that, while reading this book, one will continue to hold and honor these personal accounts.

In the next two chapters, Poleg and Zachor review potential neurological mechanisms underlying RRBIs in ASD, and Perets and Offen describe animal models for autism, with a focus on RRBIs. Regarding psychological theories, Poleg and Zachor suggest that the executive function deficit theory—which involves mechanisms of planning, controlling, and regulating higher-order mental processes—offers a good explanation for the observed RRBI. Structural brain abnormalities in the basal

ganglia and the striatum are suggested, with some data supporting the link between basal ganglia size, shape, or volume and the severity of RRBI among individuals with ASD; however, Poleg and Zachor conclude by stating that there are no conclusive and convincing data from clinical trials regarding the significance of structural changes in the brains of individuals with ASD. Next, regarding genetic abnormalities, ASD evidently has a robust genetic component, with strong familial inheritance patterns and the potential involvement of approximately 1000 genes. Research supports that RRBIs have a strong genetic component, and there is some evidence that an imbalance in the neurotransmission system of dopamine, GABA, and serotonin (5-HT) have a role in RRBI. Perets and Offen describe animal models for autism, with a focus on RRBI. They describe findings from three different mouse models: a mouse model based on mutations found in humans with ASD (the mouse genome is altered to include the same mutations as those found in humans with ASD), a mouse model of ASD involving neurodevelopmental processes during pregnancy, and a multifactorial mouse model that includes a combination of factors other than genetic mutations or biological markers that account for the observed ASD-related symptoms. The studies on these mouse models—similar to the research on neurological mechanisms among humans—emphasize our current understanding of ASD in humans, that is, some ASD symptoms may or may not be associated with various genetic and environmental factors, and the etiology of ASD and RRBI in most individuals with these diagnoses remains an enigma.

The next chapter describes RRBI. Uljarević, Hedley, Linkovski, and Leekam, addresses the underlying mechanisms and developmental trajectories of restricted and repetitive behaviors (RRB) in typical and atypical development. These authors mainly focus on RRB which typically precede IS, and offer a summary regarding the conceptualization and classification of RRB among children with typical and atypical development. RRB most often emerge earlier in development and fade throughout childhood whereas IS typically appear later and more gradually throughout development. They suggest that RRB are common among children with typical development and among children with diagnoses other than ASD. Therefore, a dimensional approach rather than a disorder-based approach may be more appropriate for conceptualization, research and intervention because RRB are prevalent in many neurodevelopmental, psychiatric, and genetic disorders.

In the following chapter, Lane presents an in-depth account of sensory subtypes associated with ASD. The focus of this chapter is on sensory features—which are patterns of behavior that are suggestive of differences in the manner in which daily sensory stimuli are processed—and on sensory modulation—which is defined as the ability of the central nervous system to regulate its responses to sensory input. The four sensory quadrants—poor registration, sensory sensitivity, sensory avoidance, and sensory-seeking behaviors—and the structures, mechanisms, and impairment associated with the various sensory subtypes are described, along with the seven proposed sensory subtype models in ASD, which identify distinct patterns of sensory features among toddlers, children, and adolescents but not adults. Lane concludes that more studies are needed to delineate the various sensory subtypes and their developmental trajectories to allow more specific assessments and

interventions. More data would also be helpful in distinguishing between sensory phenomena, which should be targeted for intervention versus this that actually assist individuals with autism in their coping with everyday activities and thus should be maintained and tailored for further development and enhanced well-being.

The section regarding the descriptive nature of RRBI is concluded with a chapter by Shulman and Bing, who address differences between females and males with ASD with regards to RRBI. Different methodologies including quantitative and qualitative paradigms, clinical and epidemiological studies, and methodologies including different informants using different instruments have repetitively demonstrated that more males than females are affected with ASD, a finding that remains unexplained. In this chapter, Shulman and Bing discuss these sex/gender differences in relation to RRBI. They offer convincing data that girls are diagnosed at an older age than boys and present a different clinical picture of behavioural symptoms, including RRBI from males. The sex/gender differences change over the course of life, with relatively few differences in toddlerhood; more differences emerging in early childhood, school-age, and early adolescence; and changes occurring again in later adolescence and adulthood, after which there seems to be a reduction in the sex/gender differences in RRBI. Overall, females present fewer of the diagnostic RRBI criteria than males but more self-injurious behaviors, compulsive behaviors, and insistence on sameness than males They argue that the fact that most diagnostic instruments were standardized with males hinders our ability to diagnose girls and to capture their unique RRBI profiles. The chapter includes a review of the developmental trajectory of RRBIs in males and females as well as the strengths and weaknesses of the most common diagnostic systems in identifying the sex/gender differences in RRBI. It is clear that more research comparing males and females with ASD is required, and not only in relation to RRBI.

The measurement of RRBI in ASD is presented in the subsequent chapter by Young and Lim. They outline the challenges involved in operationalizing these behaviors and discuss the general strengths and weaknesses of the various available types of assessment tools: questionnaires, interviews, and direct observation scales. The available specific instruments for assessment during infancy, childhood, adolescence, and adulthood, as well as those suitable across the life span are each reviewed, including the most common assessment measures utilized to assess RRBI: the repetitive behavior scale-revised (RBS-R), the autism diagnostic interview-revised (ADI-R), and the repetitive behavior questionnaire (RBQ). Many assessment tools exist; however, considering the heterogeneity of RRBI, the authors suggest that no single assessment tool is appropriate. They further discuss the fact that no assessment tool was designed and validated to specifically assess RRBI as defined in DSM-5 and state that future research should address this need.

The next four chapters focus on the relations between RRBI and language, social interactions, anxiety, and eating difficulties and disorders, respectively. Dromi, Oren, and Mimouni-Bloch review language development among children with ASD. They discuss speech production, comprehension, and development and review research regarding echolalia, verbal rituals, stereotyped language, and memorized speech, which are part of the symptoms listed in DSM-5 under RRBIs. Dromi

and colleagues argue that the distinction between communication, language, and speech is vital for describing the linguistic profile of children with ASD because young children with ASD are more likely to have weaker language comprehension skills than speech production skills, whereas the opposite is true for typically developing children. Important linguistic milestones, skills, and abilities such as babbling, age of first words, pitch, voice, intonation, pronoun reversals, and difficulties in linguistic inferences of metaphors and idioms are also reviewed. Owing to the paucity of research, the relations between severity of RRBI and various language skills is not well known; thus, future research should address this issue considering severity of symptoms, level of functioning, and subtypes of RRBI across the lifespan. The next chapter, by Ghanouni and Jarus, discusses challenges in social interactions and communication skills in relation to RRBI. After reviewing the challenges that individuals with ASD experience in social interactions, such as difficulties in perspective-taking, cognitive empathy, and affective empathy, and in processing social stimuli and drawing attention to them, Ghanouni and Jarus describe common underlying mechanisms for RRBI and impairments in social interactions, such as sensory processing and executive functioning deficits and the possibilities that impairments in social interactions result from RRBI and/or vice versa. One example which can fit all three possibilities is the transaction among anxiety, RRBI and social impairments in that any one or two of the three—RRBI, social difficulties or/and anxiety can initiate and then result in heightened levels of one or more of these, as well as other components. The role of anxiety in ASD and RRBI is presented in the next chapter by Ben-Sasson and Stephenson, who in their in-depth discussion, describe RRBI in anxiety-related disorders (ARDs) and in ASD and examine the important question similar to the egg-and-chicken problem—Does anxiety precede and lead to RRBI? Does RRBI cause anxiety? Is there a third mechanism associated with both? They offer a detailed description regarding the developmental trajectory of anxiety among individuals with ASD and on how to make a differential diagnosis between ASD and ARDs including how to distinguish between RRBIs that are most likely associated with ASD versus OCD. For some individuals with ASD, anxiety may lead to RRBI while for others or at other times, the opposite may be true. It is important to remember that RRBIs actually help some people with ASD to better cope with their anxiety and stress, yet negative reactions from the environment may hinder this adaptive function. Therefore, it is necessary to carefully evaluate RRBI in contexts, to assure that they are not disapproved of, and looked at negatively, just because they seem odd to the lay person.

Eating and feeding problems are prevalent among individuals with autism across the life span. These difficulties are discussed in the next chapter by Enten-Vissoker. Selective eating or food selectivity is the most prevalent eating problem among children with ASD, as displayed by insistence on specific foods, specific methods of food preparation, and mealtime routines. The estimate of young children with ASD who show eating problems is as high 90% compared to about 25–30% among typically developing children. Food selectivity and restriction is a risk factor for undernutrition, suboptimal growth, social deficits, poor academic progress, deficiencies of vitamins, minerals and amino acids as well as overweight for those with binge

eating. Turning to eating disorders, ASD symptoms are prevalent among people with eating disorders, with some evidence that adolescents who develop eating disorders have a childhood history of more repetitive, self-injurious, and compulsive behaviors and insistence on sameness, compared to adolescents who do not develop eating disorders. Although it is difficult to separate physiological aspects of feeding difficulties from behavioral aspects, it is safe to conclude that RRBI and a range of eating problems are interrelated in ASD, with sensory impairment as a possible underlying mechanism. More research is needed to clarify which RRBI are associated with which eating and feeding difficulties for people with ASD across the life span and range of levels of functioning.

The following two chapters focus on intervention. Yaari and Dissanayake offer a thorough evaluation of early intervention regarding RRBI. They highlight that most early intervention programs target symptoms associated with social communication challenges and outcomes rather than RRBI. Although RRBI clearly interfere with learning and social interactions, most families and professionals concentrate more on the social communication difficulties. Both comprehensive treatment models (CTMs) and focused intervention practices (FIPs) involving RRBIs as an outcome and as a predictor of early intervention are described. Family members, professionals, and others have an important role in ensuring that RRBI are carefully evaluated to assure that RRBI that are or may be helpful to individual with ASD are distinguished from those that are not. The take-home message is that the existing data regarding the efficacy of early intervention in the context of RRBI is inconsistent, and therefore more early intervention programs and studies are necessary to achieve a more in-depth understanding of factors that help in reducing RRBI that hinder adaptive daily life skills and well-being. Gal and Ben-Sasson present the "RepMod", a whole person–environment-focused intervention model that integrates sensory-based and behavior-based intervention techniques, addressing RRBI in individuals with ASD. The strengths of the model lie in its functional analysis regarding the differential advantage or disadvantage of RRBI in the life each individual. Thus, RRBI considered to hinder adaptation and well-being are targeted as behaviors that should be reduced or eliminated, whereas RRBI that are judged as potential assets are integrated into daily life activities to increase adaptation and well-being. The model is dynamic such that each cycle of evaluation, intervention, and post-intervention re-evaluation forms the next entry level for further intervention.

We conclude with a chapter by Bury, Hedley, and Uljarević, on RRBI in the workplace, which brings us back to the personal accounts presented in the first chapter in that RRBI may also serve assets and not necessarily only as undesired symptoms. Individuals with ASD often experience significant difficulties obtaining and maintaining employment. This is reflected in high rates of unemployment and underemployment worldwide. Social communication challenges can lead to difficulties through recruitment and employment processes. In addition, RRBI can present significant barriers to the employment. However, with sufficient support, individuals with autism can not only succeed in the workplace but may even outperform their colleagues in certain domains. This may be due to skills, or "talents", related to RRBI; for example, attention to detail and tolerance for repetitive tasks. A

critical appraisal of the evidence regarding the special talents of individuals with ASD as it pertains to RRBI, is offered, followed by evidence supporting the "autism advantage" in employment specifically. Finally, the authors discuss the benefits of adopting an individualized approach to research and practice, to best identify and support individual strengths and challenges within the workplace.

We end this chapter by referring to precision medicine which advocates considering individual variability in genes, environment, and lifestyle for each person in treatment and intervention. We hope this book contributes to thinking about RRBI in individuals with ASD using this model. It is interesting to note that a common goal in ASD intervention models is to promote and enhance social communication and interaction skills and to reduce or eliminate RRBI. This practice is most likely based on typical development and the desire to assist individuals with ASD achieve skills that are considered as reflecting typical development. Yet, this approach may miss out on the strengths of some RRBI as described by individuals with ASD. Therefore, we emphasize the need to evaluate RRBI in context and to distinguish among RRBI that are helpful and may promote development and well-being from those that are not. Accordingly, this book includes interdisciplinary, international, evidence-based knowledge addressing RRBI in people with ASD. It is primarily suitable for professionals and students who are working with people with ASD across the life-span. We hope that family members, as well as some individuals with ASD will also find it helpful and interesting.

References

American Psychiatric Association. (1994). *Diagnostic and statistical manual of mental disorders* (4th ed.). Washington, DC: Author.

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders (DSM-5)*. Washington, DC: American Psychiatric Publishing.

Ben-Sasson, A., Gal, E., Fluss, R., Katz-Zetler, N., & Cermak, S. (2019). Update of a meta-analysis of sensory symptoms in ASD: A new decade of research. *Journal of Autism and Developmental Disorders, 49*(12), 4974–4996.

Chamak, B., Bonniau, B., Jaunay, E., & Cohen, D. (2008). What can we learn about autism from autistic persons? *Psychotherapy and Psychosomatics, 77*(5), 271–279.

Cohen, D. J., & Volkmar, F. R. (1997). *Handbook of autism and pervasive developmental disorders* (2nd ed.). New York: Wiley.

Cunningham, A. B., & Schreibman, L. (2008). Stereotypy in autism: The importance of function. *Research in Autism Spectrum Disorders, 2*, 469–479. https://doi.org/10.1016/j.rasd.2007.09.006.

DuBois, D., Lymer, E., Gibson, B. E., Desarkar, P., & Nalder, E. (2017). Assessing sensory processing dysfunction in adults and adolescents with autism spectrum disorder: A scoping review. *Brain Sciences, 7*, 108. https://doi.org/10.3390/brainsci7080108.

Dunn, W., Little, L., Dean, E., Robertson, S., & Evans, B. (2016). The state of the science on sensory factors and their impact on daily life for children: A scoping review. *OTJR: Occupation, Participation and Health, 36*(2_suppl), 3S–26S. https://doi.org/10.1177/1539449215617923.

Esbensen, A. J., Seltzer, M. M., Lam, K. S., & Bodfish, J. W. (2009). Age-related differences in restricted repetitive behaviors in autism spectrum disorders. *Journal of Autism and Developmental Disorders, 39*, 57–66. https://doi.org/10.1007/s10803-008-0599-x.

Fyfield, R. (2014). *The rise and fall of repetitive behaviors in a community sample of infants and toddlers* (PhD thesis). Cardiff University.

Gabriels, R. L., Agnew, J. A., Miller, L. J., Gralla, J., Pan, Z., Goldson, E., et al. (2008). Is there a relationship between restricted, repetitive, stereotyped behaviors and interests and abnormal sensory response in children with autism spectrum disorders? *Research in Autism Spectrum Disorders, 2*(4), 660–670.

Gal, E. (2011). Nosology and theories of rituals and stereotypies. In J. Matson & P. Sturmey (Eds.), *International handbook of autism and pervasive developmental disorders*. New York: Springer-Verlag.

Gal, E., Dyck, M. J., & Passmore, A. (2010). Relationships between stereotyped movements and sensory processing disorders in children with and without developmental or sensory disorders. *American Journal of Occupational Therapy, 64*, 453–461. https://doi.org/10.5014/ajot.2010.09075.

Goldfarb, Y., Gal, E., & Golan, O. (2019). A conflict of interests: The complex role of special interests in employment success of adults with ASD. *Journal of Autism and Developmental Disorders, 49*(9), 3915–3923.

Joosten, A. V., & Bundy, A. C. (2010). Sensory processing and stereotypical and repetitive behaviour in children with autism and intellectual disability. *Australian Occupational Therapy Journal, 57*, 366–372. https://doi.org/10.1111/j.1440-1630.2009.00835.x.

Joyce, C., Honey, E., Leekam, S. R., Barrett, S. L., & Rodgers, J. (2017). Anxiety, intolerance of uncertainty and restricted and repetitive behaviour: Insights directly from young people with ASD. *Journal of Autism and Developmental Disorders, 47*, 3789–3802. https://doi.org/10.1007/s10803-017-3027-2.

Kanner, L. J. (1943). Autistic disturbances of affective contact. *The Nervous Child, 2*(3), 217–250.

Klin, A., Danovitch, J. H., Merz, A. B., & Volkmar, F. R. (2007). Circumscribed interests in higher functioning individuals with autism spectrum disorders: An exploratory study. *Research and Practice for Persons with Severe Disabilities, 32*, 89–100. https://doi.org/10.2511/rpsd.32.2.89.

Lane, A. E., Molloy, C. A., & Bishop, S. L. (2014). Classification of children with autism spectrum disorder by sensory subtype: A case for sensory-based phenotypes. *Autism Research, 7*(3), 322–333. https://doi.org/10.1002/aur.1368.

Miller, L. J., Anzalone, M. E., Lane, S. J., Cermak, S. A., & Osten, E. T. (2007). Concept evolution in sensory integration: A proposed nosology for diagnosis. *The American Journal of Occupational Therapy, 61*(2), 135–140. https://doi.org/10.5014/ajot.61.2.135.

Schaaf, R. C., & Lane, A. E. (2015). Toward a best-practice protocol for assessment of sensory features in ASD. *Journal of Autism and Developmental Disorders, 45*(5), 1380–1395. https://doi.org/10.1007/s10803-014-2299-z.

Schaaf, R. C., Toth-Cohen, S., Johnson, S. L., Outten, G., & Benevides, T. W. (2011). The everyday routines of families of children with autism: Examining the impact of sensory processing difficulties on the family. *Autism, 15*(3), 373–389. https://doi.org/10.1177/1362361310386505.

Schopler, E. (1995). *Parent survival manual*. New York: Plenum Press.

Tomchek, S. D., & Dunn, W. (2007). Sensory processing in children with and without autism: A comparative study using the short sensory profile. *The American Journal of Occupational Therapy, 61*(2), 190–200. https://doi.org/10.5014/ajot.61.2.190.

Turner, M. (1997). Towards an executive dysfunction account of repetitive behaviour in autism. In J. Russell (Ed.), *Autism as an executive disorder* (pp. 57–100). Oxford, UK: Oxford University Press.

Turner, M. (1999). Annotation: Repetitive behaviour in autism: A review of psychological research. *The Journal of Child Psychology and Psychiatry, 40*(6), 839–849.

Volkmar, F. R. (2005). *Handbook of autism and pervasive developmental disorders: Assessment, interventions, and policy*. Hoboken, NJ: John Wiley & Sons, Inc.

Weissman-Nitsan, M., Schreuer, N., & Gal, E. (2019). Employers' perspectives regarding reasonable accommodations for employees with autism spectrum disorder. *Journal of Management & Organization, 25*, 481–498. https://doi.org/10.1017/jmo.2018.59.

Chapter 2
"It's in my Nature" – Subjective Meanings of Repetitive and Restricted Behaviors and Interests Voiced by Adults with Autism Spectrum Disorders

Yael Goldfarb, Osnat Zafrani, and Eynat Gal

Introduction

Restricted and Repetitive patterns of Behaviors and Interests (RRBIs) are a core feature of ASD, encompassing behaviors such as: stereotyped or repetitive motor movements, use of objects, or speech; insistence on sameness; restricted interests; and hyper- or hypo-reactivity to sensory input (American Psychiatric Association, 2013). While a growing body of research explores the complexity of RRBIs and their integral role in symptoms of ASD, the different features of RRBIs and their causes remain only partially studied (Berry, Russell, & Frost, 2018). An important step towards understanding the full scope of RRBIs is listening to firsthand accounts of individuals with ASD, who, through their lived-experiences can be considered experts (Gillespie-Lynch, Kapp, Brooks, Pickens, & Schwartzman, 2017). This approach stresses the importance of listening to the subjective experiences that individuals with ASD encounter, with the personal meanings they hold.

Y. Goldfarb (✉)
Department of Occupational Therapy, University of Haifa, Haifa, Israel

Gal College, Loewenstein Rehabilitation Hospital, Raanana, Israel

O. Zafrani
Autism Treatment and Research Center – Association for Children at Risk, Givat Shmuel, Israel

E. Gal
Department of Occupational Therapy, University of Haifa, Mount Carmel, Haifa, Israel

E. Gal, N. Yirmiya (eds.), *Repetitive and Restricted Behaviors and Interests in Autism Spectrum Disorders*, Autism and Child Psychopathology Series,
https://doi.org/10.1007/978-3-030-66445-9_2

First-Hand Accounts of RRBI

Qualitative research addressing lived experiences of individuals with ASD covers several areas, at different stages of life (e.g. perception of self, interaction with others, experiences at school, and factors related to employment), with only a few relating to RRBIs (DePape & Lindsay, 2016). Joyce, Honey, Leekam, Barrett, and Rodgers (2017) investigated experiences of anxiety and their relation to RRBIs through self-reports. Other researchers addressed the unique sensory perceptual experiences of adults with ASD, employing different methodological approaches (Belek, 2019; Jones, Quigney, & Huws, 2003; Smith & Sharp, 2013). First-hand experiences of restrictive interests were also shared in studies addressing personal identity and employment (Krieger, Kinebanian, Prodinger, & Heigl, 2012; Marks, Scharader, Longaker, & Levine, 2000), but findings were not related to the wider framework of RRBI.

Another rich source for first-hand experiences can be found in autobiographies written by individuals with ASD (Grandin, 1995; Shore, 2001; Tammet, 2007; Williams, 2007), and qualitative research interpreting such publications (Ashby & Causton-Theoharis, 2009; Chamak, Bonniau, Jaunay, & Cohen, 2008; Davidson, 2010). Disparities between subjective experiences of individuals with ASD and widely held perceptions in scientific literature, suggest the existence of different views on the core difficulties that individuals with ASD encounter (Chamak et al., 2008). Whereas the scientific literature often regards social and cognitive difficulties as the core challenges of ASD, adults with ASD indicate the centrality of physical and sensorial manifestations in their subjective experiences (Belek, 2019; Davidson, 2010). Given these findings, first-hand accounts add an important and essential layer to understanding the full scope of RRBIs and their expressions in the lives of individuals with ASD.

First-Hand Accounts in the Current Chapter: Participants, Data Collection and Interpretation

The content of this chapter is based on 10 qualitative interviews held with individuals with ASD.[1] All participants (five females and five males) reported having an official ASD diagnosis, issued by a mental-health professional (psychiatrist/psychologist). They were recruited through relevant internet forums, direct contacts with individuals with ASD who maintain blogs on the internet, and organizations offering services to adults with ASD. Names and any identifying information were changed, in order to provide confidentiality.

[1] Interviews were conducted by the second author, as part of a thesis, submitted in partial fulfillment of the requirements for the MA degree, at the University of Haifa, Faculty of Social Welfare and Health Science, Department of Occupational Therapy (December, 2014).

Participants were quite heterogeneous in terms of age, education, employment, and living arrangements. The age range was between 18 and 54. Educational attainments included: high school education, vocational training, and academic studies (B.A. and M.A. degrees). Two of the respondents were employed, one was unemployed, one was amidst post-high-school national volunteer service, and five were studying at the time of the interview (one in high school, and four in academia). Living arrangements included: Living with parents, independently, in university dormitories and in supported housing.

More specifically: The youngest participant (Mila, age of 18) was completing her last year of high-school education. Eli (20) was in post-high-school national volunteer service. Both were living with their parents. Four participants, Naomi (24), Zoe (20), David (23), and Daniel (22) were studying for an academic degree. All four of them lived in university dormitories, and participated in a support program for students with ASD. Lia (28), who lived with her parents, did not acquire tertiary education, and was employed in the job market. Gabriella (42) completed vocational training, lived independently, and was not employed at time of the interview. Both Edi (42) and Neil (54), had an MA degree, but did not work in the field they studied. Edi was unemployed at the time of the interview, living with his wife and child. Neil lived in supported housing, working in customer service.

The interview protocol included questions formulated to encourage respondents to pay attention to their everyday lives' RRBI (Repetitive and Restricted Behaviors and Interests) experiences, including their subjective motivations and possible implications. For example: "Could you share such experiences (of RRBIs), and the thoughts and emotions that accompany them?"; "Why do you think you engage in (specifying a behavior that was mentioned)?"; "How would you complete the sentence 'I (type of behavior) because…'", "When I (type of behavior) it makes me feel..."?.

Data were analyzed using a thematic approach, retrieving main themes and categories that arise from the data. In the interviews, participants described their perspectives about RRBIs that accompany their daily lives, characteristics of RRBIs and patterns of change across the life-span. Subjective meanings and reasons for RRBIs were also described at length, giving an important insight to this significant aspect.

In this chapter we will focus on the subjective meanings of RRBIs as described in first-hand accounts of adults with ASD. The content of the interviews will give voice to the experiences of the participants, in their own words (though, translated from Hebrew to English).

Subjective Meaning of RRBIs

Throughout the interviews a wide variety of subjective reasons and meanings for the existence of RRBIs were described. Most of them were shared by a number of participants, while a few were described by a single person, but left a strong impression as they gave clear illustrations to situations and feelings that might be shared by

others. Mostly, a single behavior was given a number of reasons. The following sections will list five central categories of meanings given to RRBIs by the participants: (a) arousal and attention regulation; (b) sensory regulation; (c) emotional regulation; (d) providing a sense of security and coping with unexpected changes in routine; and (e) managing social communication and social interaction. The implications of RRBIs regarding participation in daily activities will be considered. At the concluding segment of this chapter, we will review the described experiences in light of current literature and suggest implications for further research and practice.

Arousal and Attention Regulation

Repetitive behaviors (e.g. pacing, body rocking, rubbing surfaces repetitively, spinning or twirling objects repetitively, picking the nose and making repetitive sounds) were described as measures that help those engaging in them to carry out different activities, stay attentive and maintain adaptive levels of arousal and concentration. Repetitive behaviors such as rubbing surfaces or manipulating objects were mentioned as actions supporting activities requiring attention, such as learning, reading, working on the computer or dealing with social situations. Eli described the subjective meaning of repetitively rubbing surfaces:

> say, it sometimes happens that I feel… that things take too long so somehow there is a way that I… avoid losing attention. it helps me.

In another interview held with Naomi, the interviewer was aware that she was constantly touching objects during the conversation. In reply to an inquiry about the behavior, Naomi mentioned that constantly rubbing or manipulating objects helps her stay attentive and alert during an interview or a lecture. She described the tactile hand stimuli as a natural state, while stopping the behavior requires thought and attention. She mentioned an event that happened during a lecture in which she participated as part of her academic requirements:

> My professor hates it when I play with my cell-phone during class. At some point I had a fight about it and I was already prepared that when my professor will comment about it again I was going to answer him t…(slightly stuttering) tell me, would you also tell a handicapped man sitting in a wheelchair to get up and start walking? It's the same thing, it doesn't distract me, just the opposite, to me it's accessibility… it makes me concentrate… not doing anything with my hands requires thinking. Doing something with my hands is in my nature!

While self-injurious behaviors are not included in the DSM-5 definition of ASD, in the interviews they were often described. For example, non- regulated arousal level was described by Naomi as a reason for self-injurious behaviors. Specifically, she described deeply scratching her back in the morning, resulting in a sensation which assists her with the transition from a state of sleep to being awake:

> It's like, arousing. In winter I can't wake up without it… wake up in the morning… I simply scratch totally hard. It… warms me up and brings the body to a kind of sensation, a sort of movement, because otherwise I don't really have a sensation. Especially when I wake up unnaturally… then I need to force myself to wake up and then like… the pain is the thing that… actually tells me to wake up.

Sensory Regulation

RRBIs were often described as sensory regulators, needed on occasions of environmental disruptions of the sensory balance. In cases of sensory over-responsivity, the purpose of repetitive behavior was to block stimuli, diminish its intensity or avoid it. Avoidance was sometimes mentioned as a way of minimizing sensory over-responsivity. Examples given were avoiding the use of public restrooms due to smell, wearing specific fabrics or tight clothes only, and avoiding certain foods (due to certain textures, temperatures, and tastes).

The need to relieve sensory stimulation was described throughout different sensory systems: The tactile system – over-sensitivity to types of tactile stimuli that are experienced as painful, and tend to be avoided (e.g. sensitivity to soap and soap foam as it results in a burning sensation); The taste system – food selectivity due to over-sensitivity to certain tastes; The olfactory system – hyper-sensitivity to smells that was described to result in nausea in case of unpleasant smells, or alternatively unique enjoyment in case of pleasant smells (such as the blossom of a certain tree); The auditory system – loud noises led to the behavior of ear poking, in attempt to block the volume of the stimulus or diminish it; The visual system – bright or flashing lights were experienced as overwhelming, causing a feeling of overload.

Naomi shared her overload experience in which she used different behavioral measures in attempt to block stimuli through various sensory systems simultaneously. In her description, she addressed the relationship between sensory overload and emotional overload:

> I was barely functioning. I sat like that on a chair with my hands stuck in my ears for an hour… and the fact that I didn't have the ability to speak with my mom made it difficult for me in a level that I couldn't hear her voice, I couldn't do anything, I just sat like that… It seals, it closes, it's totally no communication, like I can't deal with any communication, I didn't have any desire, I really preferred that nothing. I wanted to shut myself down.

Sensory overload was also described as resulting in the experience of physical pain:

> I feel attacked, I feel that my senses are being flooded, I feel that my ears are going to explode and everything… it's like I feel like… my personal space is actually being intruded by noise. (David)

Additional descriptions were given of over-stimulation, resulting from a number of simultaneous stimuli in various sensory modalities, leading to a feeling of sensory overload. Neil shared an example of sensory over-responsivity to simultaneous intense auditory and visual stimuli while participating in a wedding:

> Sensory explosion… mostly noise, ah… at weddings all those flickering lights around, and the, ah… what else? Ah… a few months ago on one occasion there was so much noise and flashing lights, I started to cry from that combination… someone turns you around and your head is ah… ah… all the time, ah… in sort of a dizziness, something like that… I started to scream out there.

Neil also described using various RRBIs as a strategy to relieve the severe physical sensations he experiences. These include movements that allow proprioceptive stimulus or reducing stimuli to a minimum by being alone for several hours to several days, meanwhile engaging in circumscribed interests.

Interesting examples that demonstrate sensory under-responsivity were also given by the participants. In these cases, sensory under-responsivity was described as resulting in seeking of strong stimuli. Stimulations described are stronger in intensity then the acceptable norm, or even considered painful. Examples shared were hugging intensely, taking a very hot shower or closing a drawer on the fingers in order to squeeze them. Cravings for stimulation were commonly fulfilled by repetitive movements. Keeping in motion was described as a natural state, while trying to stop the movement was described as an action requiring personal resources. Constant movement of body-parts, jumping, repetitive manipulations of an object, or rubbing various surfaces or textures, were all described as actions that fulfill the intense need to move. David shared the many efforts needed to control this urge:

> Many times, I need to… make many efforts, say in job interviews, to avoid, as much as I can, moving my fingers. For this reason, I have to admit that I must be at my best on job interviews. I have to sleep well, I have to eat right and be as calm as possible. That's the only way I can control these urges…

Another unconventional pattern of behavior related to somatic experience is actively seeking multiple channels of stimuli at the same time. David described the daily activity of reading a book. While most people attend mostly to visual input while reading, he reads while activating multiple sensory channels concurrently:

> Lately I am convinced that I think through my hands and legs, or sometimes through what I see with my eyes, or through all senses, what I 'see' with my senses… it's hard to think in a conventional way. Through unconventional actions I sometimes sharpen my brain… it would be hard for me to read a book and derive information from it if it doesn't have a physical presence that I can feel at the palm of my hand. I can smell it, I can hear the pages turning and I can see the text… the physical presence is very important to me. I feel that… without the physical sensations I'll sometimes ask myself if I even really exist.

Repetitive movements can also function as a form of pain relief. Naomi described how she waves her hands during pain:

> During pain my mother always says that I go like this (demonstrates waving her hands on both sides of her head), but it's only during physical pain. I don't know how to scream when I feel pain and I wouldn't want to, but I don't know, it's like I'm releasing pain. It moves the sensation from the painful area to a different place almost immediately.

Emotional Regulation

Repetitive movements were often described as a form of self-help, relieving tension in situations that cause stress or over-excitement. Repetitive movements mentioned in this content were legs shaking, body rocking, pacing from one side of the room to the other, manipulating objects and more. Stressful times are accompanied by repetitive behaviors that are more frequent, and have higher intensity. Naomi explained how actions that she refers to as "autistic behaviors" help her manage high levels of stress or excitement, even high levels of happiness:

> It's related to pressure, stress, anxiety, depression. When everything is okay, and as usual and everything's great you will hardly find autistic behaviors, only when there's great joy, you know, there is joy that you can hardly bear to contain. And then yes, but you know in most… in normal situations, in so called standard and calm situations, you won't find autistic behaviors… they truly help. There is no other way.

Striking examples describing repetitive action in relation to emotional stress addressed self-injurious behaviors. On some occasions, extreme emotional experiences might lead to self-injury such as self-beating, banging the head on the wall, or self-induced vomiting, in order to substitute mental pain with physical pain. David described incidents of self-disappointment, that triggered self-injurious behavior:

> It happens when I feel frustrated or when I feel atomic pressure inside my head, when things get out of control or I feel like I did something inappropriate, or when I fail, I hate failing, it makes me take a hostile action towards myself… cause I can't stand myself at that moment… it's a maladaptive attempt to find some kind of a way to communicate with myself.

Naomi also described self-injurious behavior as a result of severe mental distress:

> In difficult times I had a thing for turning mental pain into physical pain, really scratching with metal wires until… I drew blood… it hurt… it was really serious violence towards myself for the sake of turning mental pain into physical pain… It indicates emotional distress that is very very very very strong, so that alone is the reason that it is a problem.

Providing a Sense of Security and Coping with Unexpected Changes in Routine

Familiar activities were mentioned as providing a sense of security and enjoyment. Examples shared were repeatedly reading the same segment in a book, re-watching television episodes, or listening to a song on repeat. Going back to a familiar experience enables to re-experience the positive feelings that accompany it again. Repetitive movements, or alternatively, avoidance of movement, also offer a sense of security as they are familiar activities. Zoe described the calming effect of engaging in a repetitive action:

> Moving it again and again… also gives me a sense of certainty, like… that I have something to do, it's good for me. I need to hold my cell phone and then I… I prefer to hold something

> in my hand… It's repeating something again and again, you see? You'll see later on that… I see the same TV show again and again and again it's, like not trying anything new, like a cycle, coming back to the same place every time.

The role of enabling a sense of security was also given to inanimate objects. Emotional connections to objects were often described in the interviews, and emotional interpretations were often given in relation to the inanimate surrounding. Zoe elaborated about her relationship to objects:

> I feel more attached to objects then to people, it… it can be, even though I can be a very friendly person so… yeh, when I come to think of it, objects make me feel more secure then people… objects they have a… ah, predictable in some kind of a way. Even though a cellphone is also not always predictable but ah… but all in all they are more predictable then people… you know how they will react, and they are always there. They are available, you don't need to chase after them and… they'll go anywhere with me.

Daniel described the meaning he finds in objects, along with hoarding behaviors:

> Hoarding often gives a sense of security, it makes me feel secure that I'm… that I'm surrounded with areas I have control over, and I'm surrounded with things I feel very strongly connected to… It's kind of, that you have your social safety net for feeling secure, like you have your academics… these are the nets that I have… so I also have this safety net, security… a network of energy, a network of memories, an emotional network, and it can be expressed by objects.

Indeed, being in a familiar environment was associated with a feeling of security and certainty. For example, familiar places are often preferred, while new places are actively avoided. Eli described how a familiar place offers solid ground for dealing with problems that might occur:

> I notice that when the bus arrives… as it arrives to a familiar area I feel more calm, because then I feel that if something happens I won't have a problem, I know, I know the area, I know what to do in the area, it is, it's mo(re)… (stutters), it's a safe area.

In contrast to the comforting feeling of familiarity, changes in routine were described as leading to feelings of anxiety and anxiousness. Zoe shared the negative feelings that accompany changes in routine:

> I need to know where I am, who I am… it's like I think that… I talked about it many times with my psychologist, that… I am always afraid that… if change will come it won't be me anymore… how will I think of myself? it's like the environment is what defines me.

David shared his feelings regarding unexpected changes and not knowing how to act in these circumstances:

> I am somewhat a control freak… I need things to occur exactly like I want them, as fast as I want them… I find it very difficult to compromise. I know how to compromise but I need to make myself sometimes…I don't know how to react when something unexpected happens, unless I am prepared for the unexpected.

An attempt to try and gain control over the environment was sometimes reflected in behavioral patterns such as walking in repetitive patterns. Zoe shared the habit of stepping on a single color when walking on checkered floors:

> I'll tell you the truth, many times when I'll walk on something checkered… I do it very precisely and I don't even notice… I walk only on the… there is ah… I step only on the black ones and vice versa. It's like, the black seems stronger, and my foot seems more, psychologically like… again, I don't even notice, it's like I do it casually.

Further along in the interview, Zoe also shared the need to be prepared in advance for what may come, and described the unpleasant physical sensations that accompany uncertainty:

> I need some kind of a basic feeling that I know what is happening… I know where I'm going, I know that I'm going home and I know the way. If I don't know the way then I… then I already go crazy… well, maybe not crazy, but I… it's an unpleasant feeling even in my throat also like… some kind of nausea maybe.

Managing Social Communication and Social Interaction

Restrictive interests and repetitive use of language were described as behaviors that are often aimed at dealing with difficulties in communication. Participants described being more confident when they talked about their special interest, subjects on which they consider themselves experts. Lia stated that her special interest area at childhood was biology. She described how her special interest helped her start a conversation:

> I would make up all kinds of questions, biology was my area, and I would come and ask. Felt awkward to come and start a conversation for no reason.

Another use of speech and language was described as an attempt to make sense of social situations and sometimes even personal emotions. Daniel described how internal idiosyncratic speech helps him make sense of the world:

> I have a kind of language. I have personal concepts that… the way I make meaning of things that I don't really understand, when I don't understand things I need to color them in strong colors in order to see them… and I need to give them very specific definitions, very certain characteristics, it isn't always in touch with reality.

David describes how reading a book repeatedly allows him to reflect on the 'real world'. He found engaging in special interests useful for trying to understand and make sense of social situations and life events:

> There are times when I feel it is parallel to our world so I say why not use it… to try and create an allegory or a comparison.

Subjective Implications of RRBIs in Daily Life

Another important aspect of RRBIs addressed in the interviews, is the way they interact with daily living, and their influence on a variety of daily activities such as: self-care, post-secondary studies and work, leisure and social activities. Interrelations

between RRBIs and participation can be divided to RRBIs that promote participation and RRBIs that inhibit participation. Examples of each kind will allow us to understand their function in daily activities, through the eyes of individuals with ASD.

RRBIs that Promote Participation

Strict organization of a daily routine was described as a way that helps individuals with ASD prevent the occurrence of unexpected events, and thus promotes daily functioning. Daily routine enables to follow a clear set of rules that reflect personal preferences, and a clear knowledge of what's ahead. Knowing what is about to happen helps reduce stress that accompanies the fear of the unknown. Eli described how his daily routine helps him:

> It's organization. I want to organize, to know that it's permanent, that there are rules… a clear set of rules… it gives a good feeling, that there is a certain shape… I think I need order, so I can make sure I don't neglect anything, and that I can make sure that each thing, like, receives the time of the day that I think it should receive… I think it's only because I'm organized, it's like, not scary, it doesn't… nothing unexpected will happen.

Engaging in special interests was described as the behavior that holds the most positive implications for individuals with ASD. Special interests were mentioned as contributing to participation in normative settings such as military or national service (considered a normative phase for young adults in Israel), college studies and employment. Interviewees were enrolled in academic programs in their areas of interest such as history, religion, art, science and communication. A preference for having jobs related to the special interests was clearly expressed. For example, Gabriella mentioned an interest in private investigation that was expressed through a job in court, typing statements of claim and defense and investigation documents. Another example was found in David's choice to work in a museum. He described this positive experience:

> I am just fascinated by it… how I love the world of art and creativity, I was never happy at school because I didn't have extracurricular activities in these areas… that's why I loved working in the art museum… I felt I am making the most of my abilities in an area that interests me.

A match between special interests and work/academics was perceived to contribute to success. Participants characterized themselves as having high curiosity for their areas of interest, which also led them to meet the academic requirements in those fields. Opinions about increased engagement in special interests were mostly positive and overall, they were experienced as an advantage, rather than a disadvantage as the term "excessive engagement" may hint. Daniel, for example, opposes this term and prefers to use the term "extended engagement", that reflects a more positive view of the matter:

> But I think more about Albert Einstein, who wrote about the rays of light and invented the formula $E=MC^2$. Meanwhile… while other guys in the same field were stuck in a totally different phase, because he dedicated more time… on account of his personal life… and yes I think we should change the term 'excessive engagement' cause… excessive sounds like ah… it sounds negative… say intensified engagement not excessive or let's say… extended engagement.

Another way in which special interests promote participation is by using the acquired knowledge in daily functioning. Extensive knowledge in fields such as photography, art, cooking, baking, history, and the bible was mentioned. This knowledge was manifested in daily activities such as taking pictures and posting them on 'facebook', watching sports, taking photos on family occasions, cooking, reading, watching movies and more. Interest areas were a source for leisure activities and hobbies.

The repetitiveness embedded in the special interests was also mentioned as helpful in promoting learning (e.g. reading the same text repeatedly promotes understanding), understanding emotions (of self and others), understanding values, and possible behaviors.

Special interests were also regarded as useful for dealing with the challenges of social communication. Interviewees found it easier to participate in conversations about their areas of interest. Extensive knowledge in a field of interest enhanced self-confidence and self-efficacy with regard to the social surroundings. Eli described the positive attitudes he received in regard to his knowledge in his special area of interest:

> When I was a student it was a lot of fun because I felt everybody knew I am smart and counted on me… I had self-confidence… people listened to me, I felt understood

RRBI that Inhibit Participation

Repetitive behaviors that help regulate sensory stimuli were mentioned with ambivalence. The positive effects on sensory regulation was contrasted by negative reactions from the social environment. Whereas the repetitive activity may help maintain attention and concentration, the negative reactions inhibit their positive effects. Edi shared his thoughts about the negative reactions to his repetitive behavior of picking at his nose while at work:

> The main problem, I think, is when I am at a job that I have to make an effort not to do it, because it just isn't appropriate… it's out of line, for me to sit at the cashier and pick at my nose… it didn't happen every day, just sometimes. The other cashiers… I would hear a little whispering behind my back or something like that, but even if they didn't talk about it it's obvious it is disturbing… the problem was evident… it was very awkward.

Naomi elaborated about her need to keep occupied by repetitive movement of objects, an activity that helped her concentrate, but was frowned upon by her college professors. She described the efforts required to avoid the repetitive motor activity and their toll on her:

> So I'll stop, and I'll endure the need to avoid it, because… it's so called 'not appropriate'…people don't like it… all those hand movements and stuff, so I really try to be considerate… it's a big problem for other people… it's their problem…but my problem is that my professors have a problem with it… it's a part of me, again, I can't… I am aware of it being a part of me and I am not always fully aware of every little presentation of it.

Due to negative reactions to repetitive behaviors, interviewees mentioned they prefer to engage in these behaviors in private.

> My family is a problem, my mom used to think we need to look into it, what's the deal, why do I do these things, that I have to play with my hands… now she calmed down. The truth is that now I do it in my room so she doesn't see. (Lia)

Negative reactions of other people lead to a preference of engaging in repetitive activities in privacy and can further lead to avoidance of social gatherings that demand the inhibition of such behaviors. The fear of not being able to control the behavior and being found odd can thus further inhibit social participation.

The overall feelings of sensory or emotional overload described in the interviews, can lead to a general impact on daily functioning. Sensory overload can last changing periods of time (hours to days), and cause confusion and distraction. These were described as leading to avoidance of daily activities such as grocery shopping or driving and can negatively affect financial conduct and the feeling of personal safety. Neil described his feelings while experiencing an overload:

> I start to feel confused, distracted and ah… I don't notice what's happening, it can happen while I drive, even though I didn't get into severe accidents…we're working on it…

Naomi described the complexity of every day self-care activities as a result of tactile over-reactivity, and the strategies she uses to manage it:

> It's so annoying, it irritates me. Yeh, I avoid combing my hair, it's like I'll use conditioner three times just to avoid combing my hair which gives me the nerves… yes, it's just unpleasant… the feeling of tangled hair in my hands is very unpleasant… and facial cleanser… ah, and I don't use a towel… I don't know, I guess I just can't… so I don't touch my face with a towel at all, I wash my face end then I just leave it like that.

Implications for Research and Practice

First-hand accounts of individuals with ASD offer vivid examples of RRBIs, their underlying meaning, and possible explanations for engaging in them. In the following section we review the data in light of recent research and offer suggestions for further research and practice.

RRBIs in Adulthood

While most research on RRBI's is focused at childhood, the limited studies examining the prevalence of RRBIs across the life span do not reach clear conclusions. Some researchers suggest a general decrease in frequency and severity of RRBI over time, while others show that behaviors such as routines and resistance to change remain evident in adulthood (Leekam, Prior, & Uljarevic, 2011). In our sample of adults with ASD who are cognitively able, RRBIs were not only very prevalent, but were also found to have clear implications and impact on daily life. Our interviewees shared a wide range of RRBIs, not only those that are considered "high-level" behaviors (e.g. compulsive and ritualistic behavior, insistence on sameness) expected from people with ASD level 1, but also reflecting the category of "low-level" behaviors (e.g. stereotyped movement, repetitive manipulation of objects) and self-injurious behaviors. While some researchers presume low-level behaviors to be more prevalent in younger ages (Richler, Huerta, Bishop, & Lord, 2010), our sample supports studies indicating behaviors matching both levels among individuals with ASD, regardless of age or cognitive ability (Gal, 2006; Leekam et al., 2011). Negative responses to repetitive or stereotypic behaviors by the social environment, may cause these behaviors to be hidden, therefore less prevalent in studies based on observations or parent's reports.

The Centrality of Sensory Sensitivity

Unique modalities of sensory processing were described by the participants, supporting the central role of sensory sensitivities in the diagnosis of ASD (Gal, Dyck, & Passmore, 2009; Hazen, Stornelli, O'Rourke, Koesterer, & McDougle, 2014; Uljarević, Richdale, Evans, Cai, & Leekam, 2017). Sensory abnormalities are mentioned as one of four sub-categories of RRBIs composing the criteria for ASD in DSM-5. Subjective experiences of adults with ASD described in this chapter, point out the centrality of sensory processing irregularities, not only as behaviors meant to avoid or seek stimuli, but as a reason for engaging in various other RRBIs. Sensory sensitivities were linked to behaviors appearing in different categories in the DSM-5. For example, the need to inhibit sensory stimuli was answered by repetitive behaviors on some occasions, while at others, engagement in special interests was used as a calming strategy after experiencing sensory overload.

Emotional and Psychological Aspects

Regulation difficulties shared by our interviewees were not exclusively related to sensory experiences. Difficulties in emotional regulation were widely mentioned as regulated by RRBIs. The association between RRBIs and anxiety deriving from our data is supported by research, indicating associations between the two variables. Research findings support the role of RRBIs as a coping response that individuals with ASD use to reduce anxiety (Joosten, Bundy, & Einfeld, 2009; Joyce et al., 2017; Spiker, Lin, Van Dyke, & Wood, 2012).

Intolerance to uncertainty was also mentioned in the current interviews and manifested through actions taken to avoid uncertainty and enhance certainty. Experiences demonstrated a need for fixated routines, a preference for familiar activities and places and even familiar objects. Our findings, illustrating the role of intolerance to uncertainty as part of the regulatory purposes if RRBIs are complemented with quantitative data giving evidence to this relation (Uljarević et al., 2017; Wigham, Rodgers, South, McConachie, & Freeston, 2015).

Another finding related to emotional distress that requires further attention is self-injurious behaviors. These behaviors are not currently part of the ASD diagnosis criteria in DSM-5, yet about half of the participants in our sample mentioned self-injurious behaviors as part of their RRBIs. While self-injurious behaviors were sometimes related to a need for sensory stimulation, at other times the source of the behavior was described as severe emotional overload. The actions were described as attempts to transform unbearable emotional pain to physical pain that might be easier to bear. These findings suggest that these behaviors occur in adulthood and can attest to severe mental distress. More research of the phenomena and a wider set of quantitative data may shed more light on this issue and offer appropriate clinical interventions.

Implications for Daily Functioning

It is evident from our data that adults with ASD perceive some RRBIs to be adaptive, promoting participation in daily activities. These findings support the hypothesis that RRBIs are used as regulatory mechanisms of sensory abnormalities (Gal et al., 2009) and thus improve daily functioning. For instance, insistence on sameness allows to keep a regular routine and repetitive behaviors can help manage sensory stimuli and emotional distress. Generally speaking, it seems that RRBIs helps adults with ASD make sense of an otherwise chaotic reality.

Among the different categories of RRBIs, special interests stand out as having positive effects in relation to various goals: anxiety regulation, social communication, self-esteem, learning, vocational and leisure activities. While traditional approaches highlighted the deficits of engaging in special interests, different views (Bross & Travers, 2017; Grove, Hoekstra, Wierda, & Begeer, 2018; Koenig &

Williams, 2017; Winter-Messiers et al., 2007), which our findings support, suggest their positive effects on different aspects of daily functioning. Specifically, subjective experiences described in this chapter are complemented by a recent study associating special interests with higher subjective well-being, and satisfaction across life domains such as social contact and leisure (Grove et al., 2018).

The possible positive effects of RRBIs should be taken into account by practitioners supporting individuals with ASD and their families. That is, while aiming to address RRBIs in interventions, it is important to differentiate between RRBIs that interfere with participation and should therefore be decreased, from those that may assist functioning, and should thus be accepted as they promote participation in everyday life. Such an approach may be more adaptive then an attempt to extinct repetitive and restrictive behaviors while disregarding their subjective significance for some individuals with ASD.

And last, special focus should be given to the way repetitive behaviors appear to the social environment of individuals with ASD. We identified a behavioral cycle of the need to perform repetitive and restrictive behaviors in private, due to social judgement, leading to avoidance from daily activities and further inhibiting social participation. Without recognition, adaptive RRBIs might be avoided due to negative social responses. Therefore, acknowledging the function of RRBIs for individuals with ASD is an important step in raising awareness and offering appropriate accommodations for those behaviors which assist individuals with ASD in their daily functioning and promote subjective well-being.

Indeed, the importance of environmental awareness to the needs of adults with ASD was evident in the study of MacLeod, Allan, Lewis, and Robertson (2018), who interviewed students with ASD about their experiences of success. Assumptions of the social environment about ASD and barriers imposed by others were mentioned as obstacles to success, forcing the students to actively resist deficit-based interpretations and demand proper accommodations. Concurrently, experiences of support and adjustment had a central role in promoting success. The need to negotiate adjustments can cause stress for those who are able to self-advocate and pose a barrier to those who find it difficult to actively express their needs (MacLeod et al., 2018). To demonstrate from our sample, Naomi eloquently described the way using her cell-phone during a lecture improves her ability to concentrate: "*to me it's accessibility*". If her college professor was aware that manipulating an object can help her to attend the lecture, the behavior might not be frowned upon. An employer might offer an employee diagnosed with ASD, a private working space in which s/he can engage in a repetitive behavior without receiving negative responses from colleagues or disturbing the work of others. Awareness of the social surrounding could lead to higher consideration that in turn may help reduce the inhibiting effects of RRBIs and enhance their adaptive effects.

To conclude, findings described in this chapter join and complement the growing body of literature considering autistic behaviors not only from a deficit point of view, but also as having an inner system of meaning, aimed at helping individuals with ASD function better in everyday life. Understanding the possible explanatory models that identify individuals with ASD as experts on their own lives, can

promote and help develop relevant interventions to support and improve their quality of life. Expanding knowledge about RRBIs, their subjective meanings and their positive characteristics can help destigmatize attitudes towards individuals with ASD and facilitate integration in social, academic and vocational environments. It is our hope that the information in this chapter is a step forward towards achieving this goal.

References

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders (DSM-5)*. Washington, DC: American Psychiatric Pub.

Ashby, C. E., & Causton-Theoharis, J. N. (2009). Disqualified in the human race: A close reading of the autobiographies of individuals identified as autistic. *International Journal of Inclusive Education, 13*(5), 501–516.

Belek, B. (2019). Articulating sensory sensitivity: From bodies with autism to autistic bodies. *Medical Anthropology, 38*(1), 30–43.

Berry, K., Russell, K., & Frost, K. (2018). Restricted and repetitive behaviors in autism spectrum disorder: A review of associated features and presentation across clinical populations. *Current Developmental Disorders Reports, 5*(2), 108–115.

Bross, L. A., & Travers, J. C. (2017). Special interest areas and employment skills programming for secondary students with autism. *Teaching Exceptional Children, 50*(2), 74–83.

Chamak, B., Bonniau, B., Jaunay, E., & Cohen, D. (2008). What can we learn about autism from autistic persons? *Psychotherapy and Psychosomatics, 77*(5), 271–279.

Davidson, J. (2010). 'It cuts both ways': A relational approach to access and accommodation for autism. *Social Science and Medicine, 70*(2), 305–312.

DePape, A. M., & Lindsay, S. (2016). Lived experiences from the perspective of individuals with autism spectrum disorder: A qualitative meta-synthesis. *Focus on Autism and Other Developmental Disabilities, 31*(1), 60–71.

Gal, E. (2006). *An investigation of the relationship of sensory differences and intellectual level to stereotyped and self- injurious movement in children*. Australia: Curtin University: Unpublished dissertation.

Gal, E., Dyck, M. J., & Passmore, A. (2009). The relationship between stereotyped movements and self-injurious behavior in children with developmental or sensory disabilities. *Research in Developmental Disabilities, 30*(2), 342–352.

Gillespie-Lynch, K., Kapp, S. K., Brooks, P. J., Pickens, J., & Schwartzman, B. (2017). Whose expertise is it? Evidence for autistic adults as critical autism experts. *Frontiers in Psychology, 8*, 438.

Grandin, T. (1995). *Thinking in pictures: And other reports from my life with autism*. New York: Doubleday.

Grove, R., Hoekstra, R. A., Wierda, M., & Begeer, S. (2018). Special interests and subjective well-being in autistic adults. *Autism Research, 11*(5), 766–775.

Hazen, E. P., Stornelli, J. L., O'Rourke, J. A., Koesterer, K., & McDougle, C. J. (2014). Sensory symptoms in autism spectrum disorders. *Harvard Review of Psychiatry, 22*(2), 112–124.

Jones, R. S., Quigney, C., & Huws, J. C. (2003). First-hand accounts of sensory perceptual experiences in autism: A qualitative analysis. *Journal of Intellectual and Developmental Disability, 28*(2), 112–121.

Joosten, A. V., Bundy, A. C., & Einfeld, S. L. (2009). Intrinsic and extrinsic motivation for stereotypic and repetitive behavior. *Journal of Autism and Developmental Disorders, 39*(3), 521–531.

Joyce, C., Honey, E., Leekam, S. R., Barrett, S. L., & Rodgers, J. (2017). Anxiety, intolerance of uncertainty and restricted and repetitive behavior: Insights directly from young people with ASD. *Journal of Autism and Developmental Disorders, 47*(12), 3789–3802.

Koenig, K. P., & Williams, L. H. (2017). Characterization and utilization of preferred interests: A survey of adults on the autism spectrum. *Occupational Therapy in Mental Health, 33*(2), 129–140.

Krieger, B., Kinebanian, A., Prodinger, B., & Heigl, F. (2012). Becoming a member of the work force: Perceptions of adults with Asperger syndrome. *Work, 43*, 141–157.

Leekam, S. R., Prior, M. R., & Uljarevic, M. (2011). Restricted and repetitive behaviors in autism spectrum disorders: A review of research in the last decade. *Psychological Bulletin, 137*(4), 562.

MacLeod, A., Allan, J., Lewis, A., & Robertson, C. (2018). 'Here I come again': The cost of success for higher education students diagnosed with autism. *International Journal of Inclusive Education, 22*(6), 683–697.

Marks, S. U., Scharader, C., Longaker, T., & Levine, M. (2000). Portraits of three adolescent students with Asperger's syndrome: Personal stories and how they can inform practice. *Research and Practice for Persons with Severe Disabilities, 25*, 3–17.

Richler, J., Huerta, M., Bishop, S., & Lord, C. (2010). Developmental trajectories of restricted and repetitive behaviors and interests in children with autism spectrum disorders. *Development and Psychopathology, 22*, 55–69.

Shore, S. M. (2001). *Beyond the wall: Personal experiences with autism and Asperger syndrome*. Shawnee Mission, KS: Autism Asperger Publishing Company.

Smith, T., & Sharp, J. (2013). Fascination and isolation: A grounded theory exploration of unusual sensory experiences in adults with Asperger syndrome. *Journal of Autism and Developmental Disorders, 43*, 891–910.

Spiker, M. A., Lin, C. E., Van Dyke, M., & Wood, J. J. (2012). Restricted interests and anxiety in children with autism. *Autism, 16*(3), 306–320.

Tammet, D. (2007). *Born on a blue day: Inside the extraordinary mind of an autistic savant*. Simon and Schuster.

Uljarević, M., Richdale, A. L., Evans, D. W., Cai, R. Y., & Leekam, S. R. (2017). Interrelationship between insistence on sameness, effortful control and anxiety in adolescents and young adults with autism spectrum disorder (ASD). *Molecular Autism, 8*(1), 36.

Wigham, S., Rodgers, J., South, M., McConachie, H., & Freeston, M. (2015). The interplay between sensory processing abnormalities, intolerance of uncertainty, anxiety and restricted and repetitive behaviors in autism spectrum disorder. *Journal of Autism and Developmental Disorders, 45*(4), 943–952.

Williams, D. (2007). *Nobody nowhere: The extraordinary autobiography of an autistic*. London: Doubleday.

Winter-Messiers, M. A., Herr, C. M., Wood, C. E., Brooks, A. P., Gates, M. A. M., Houston, T. L., & Tingstad, K. I. (2007). How far can Brian ride the daylight 4449 express? A strength-based model of Asperger Syndrome based on special interest areas. *Focus on Autism and Other Developmental Disabilities, 22*(2), 67–79.

Chapter 3
Neurological Mechanisms Underlying Repetitive and Restricted Behaviors in Autism Spectrum Disorders

Shani Poleg and Ditza A. Zachor

Repetitive and restricted behaviors and interests (RRBI's) described in individuals with Autism Spectrum Disorder (ASD) and defined in the DSM-5 criteria are diverse (American Psychiatric Association, 2013). They encompass repetitive behaviors, such as stereotyped motor movements (i.e. hand flapping), repetitive manipulation of objects (i.e. spinning objects), repetitive and stereotyped language (i.e. immediate and delayed echolalia), repetitive sensory behaviors (i.e. visual inspection of objects), and insistence on sameness (i.e. compulsions, rituals and routines, and narrow and circumscribed interests) (Jiujias, Kelley, Hall, & Kelley, 2017; Kim & Lord, 2010; M. Lewis & Kim, 2009b; Mark H. Lewis & Bodfish, 1998; Stratis & Lecavalier, 2013; Turner, 1999).

Research based on standardized diagnosing tools such as the Repetitive Behavior Scale-Revised (RBS-R) (Bodfish, Symons, Parker, & Lewis, 2000), the Autism Diagnostic Interview-Revised (ADI-R) (Lord, Rutter, & LeCouteur, 1994), and the Autism Diagnostic Observation Schedule (ADOS) (Lord et al., 2000) grouped these RRBI's into two distinct categories: lower-level behaviors that include the repetitive behaviors, and high-level ones that include ritualistic habits and a strict adherence to well-established routines (Bishop et al., 2013; Kim & Lord, 2010; Richler, Huerta, Bishop, & Lord, 2010; Stratis & Lecavalier, 2013). The trajectory of RRBI's presentation changes with age and level of cognition (Jiujias et al., 2017; Stratis & Lecavalier, 2013) and may increase in severity (Richler et al., 2010). In addition, restricted interests and rituals were shown to be more prevalent in children with higher scores of non-verbal IQ (NVIQ) than repetitive sensory motor behaviors (Bishop, Richler, & Lord, 2006).

S. Poleg (✉)
Sackler Faculty of Medicine, Tel Aviv University, Tel Aviv, Israel

D. A. Zachor
Sackler Faculty of Medicine, Tel Aviv University, Tel Aviv, Israel

The Autism Center/ALUT, Shamir (Assaf Harofeh) Medical Center, Zerifin, Israel

E. Gal, N. Yirmiya (eds.), *Repetitive and Restricted Behaviors and Interests in Autism Spectrum Disorders*, Autism and Child Psychopathology Series,
https://doi.org/10.1007/978-3-030-66445-9_3

Many theories regarding the neurological mechanisms underlying RRBI's in ASD have been suggested, ranging from psychological, structural and anatomical abnormalities to molecular and genetic impairments. In this chapter we elaborate on some of the leading theories concerning the neurological basis of RRBI's in ASD.

RRBI's and Psychological Theories

Researchers throughout the years have attempted to explain what drives individuals with ASD to execute RRBI's. Very early works hypothesized that individuals with ASD have a constant nonspecific activation of the ascending reticular activating system, and therefore wish to lower that arousal by engaging in RRBI's to block new sensory input (B. Y. C. Hutt & Hutt, 1965; C. Hutt, Hutt, Lee, & Ounsted, 1964; Turner, 1999). Others refer to RRBI's as a homeostatic mechanism, claiming that individuals with ASD tend to perform RRBI's in situations involving lack of stimulation, or over stimulation from their environment (Sloman, LaRue, Weiss, & Hansford, 2013; Turner, 1999). However, a later work suggested that stereotyped behaviors were more likely to occur under conditions of low stimulation, and were less likely during conditions involving social contact (Hall, Thorns, & Oliver, 2003). Lovaas, Newsome, and Hickman (1987), hypothesized that RRBI's stem from self-stimulatory behaviors, and therefore act as operant responses, meaning that the consequences of the behaviors act to reinforce the continuous use of RRBI's by the individual.

Some other theories concerning RRBI's' etiology in individuals with ASD are based on the leading psychological theories for explaining the core symptoms of ASD: "Theory of Mind" (ToM), "weak central coherence" and "executive function deficits". For example, it has been suggested that individuals with ASD have "weak central coherence": as a result of deficits in information processing, they tend to focus on trivial details and miss the greater social context, leading to restricted and repetitive interests (Frith & Happe, 1994).

The "executive functioning deficits" theory has traditionally been thought of as the primary explanation for RRBI's in ASD (Rosenthal et al., 2013). Executive functioning is an umbrella term, used to describe a wide range of behaviors involved in planning, controlling, and regulating higher-order mental processes (Rosenthal et al., 2013). Deficits in cognitive flexibility- the ability to shift thoughts and adapt behaviors to the changing environment- was consistently associated with the presence of RRBI's in ASD (Street, 1994; Turner, 1999). Other executive function impairments, such as inhibitory and attentional deficits, were also suggested as part of RRBI's etiology in individuals with ASD (Mostert-Kerckhoffs, Staal, & Houben, 2015; Schmitt, White, Cook, Sweeney, & Mosconi, 2017). In a study on 64 high-functioning individuals with ASD and 53 typically developing (TD) comparison participants, individuals with ASD showed significantly higher cognitive control dysfunctions. Moreover, the severity of inhibitory control and attentional flexibility deficits predicted the severity of RRBI's in everyday life. Specifically, response

inhibition in reaction to visual information, and task switching in reaction to auditory information, predicted motor and sensory stereotyped behavior (Mostert-Kerckhoffs et al., 2015).

Additionally, there are those who hypothesized that RRBI's in ASD are correlated with anxiety levels (Sullivan et al., 2014). For example, it has been suggested that RRBI's serve as a coping strategy for lowering high levels of anxiety in individuals with ASD. The anxiety may be caused by deficits in ToM, the understanding of other people's mental state, which is the leading theory for explaining social-communication impairments in ASD (Baron-Cohen, 1989; Turner, 1999). In a different study, the correlation between anxiety levels and RRBI's in individuals with ASD was compared with that of individuals with Down Syndrome and typically developing (TD) individuals. The researchers used questionnaires completed by the parents of the participants concerning their RRBI's and anxiety levels. The results yielded a correlation between higher levels of anxiety and higher levels of RRBI's only among individuals with ASD (D. W. Evans, 2017). This issue was addressed in an additional study, in which self-report questionnaires were used rather than parental reports. The results also indicated a significant relation between RRBI's and anxiety, as reported by the individuals with ASD themselves (Joyce et al., 2017).

It is well agreed that ASD is a highly heterogeneous neurodevelopmental disorder with complex genetic origin and epigenetic impact (a probable contributing environmental component). Yet, the exact etiology remains unknown. Recent advances in the field of neuroscience have pointed out that specific behaviors are mediated not by impairments in one brain region but rather by subtle alterations across multiple brain regions, neurotransmitter systems, and synaptic processes that converge as neural circuits. This would also be true for ASD, as researchers currently consider it a brain network connectivity disorder (Just, Cherkassky, Keller, & Minshew, 2004; Monk et al., 2009; Nomi & Uddin, 2015; Shih et al., 2010), in which multiple genes coding for synaptic functioning are involved, leading to impairments in large-scale neuronal networks (Muhle, Reed, Stratigos, & Veenstra-vanderweele, 2018). Hence, it is possible that ASD behaviors that are explained by different psychological theories, such as RRBI's, originate from specific genetic and molecular etiologies and are related to specific brain structure and/or functional abnormalities.

Structural Abnormalities

Another hypothesis regarding the etiology of RRBI's in individuals with ASD suggests that some anatomical changes in certain brain structures are correlated with the severity of the RRBI's among individuals with ASD.

Most studies have focused on the basal ganglia and the striatum. The basal ganglia are a group of interconnected subcortical nuclei, the primary afferent structure being the striatum. In most mammals, including humans, the striatum contains the caudate and putamen, which are divided by the fibers of the internal capsule (Albin,

Young, Penney, Roger, & Young, 1989). The basal ganglia and the striatum are associated with a number of roles, ranging from cognition to motor movements (Stocco, Lebiere, & Anderson, 2011). In addition, the classic inhibitory neurotransmitter of the basal ganglia is the neurotransmitter GABA (Albin et al., 1989), whereas dopamine and glutamate are the dominant excitatory neurotransmitters (Caravaggio et al., 2016). The basal ganglia activity is traditionally divided to two pathways. The "direct pathway" describes the inhibitory effect of the striatum on the substantia nigra reticulata (SNr) and the globus pallidus interna (GPi). This inhibition of SNr/GPi causes a dis-inhibitory effect on the thalamus. The "indirect pathway" has the opposite effect on the thalamus. This pathway proceeds through the globus pallidus externa (GPe) and the sub thalamic nucleus (STN), leading to an inhibitory effect on the thalamus (Stocco et al., 2011). These pathways are involved in the control of motor movements in a subtle and complex pattern, since the basal ganglia have a role in the generation and execution of context-dependent behaviors (Y. Smith, Bevan, Shink, & Bolam, 1998).

In a study that compared high functioning individuals with ASD aged 6–17 to a TD comparison group, it was shown that the individuals with ASD had significant impairments in several measures of motor control (Newschaffer, Denckla, Landa, & Mostofsky, 2006). In addition, there is a resemblance between the ritualistic, repetitive, stereotyped and compulsive behavior in ASD and behaviors observed in other psychiatric disorders, for example obsessive-compulsive disorder (OCD) Boyer & Lie'nard, 2006; D. W. Evans et al., 1997; Langen, Durston, Kas, Van Engeland, & Staal, 2011). It was also reported that there may be a connection between basal ganglia lesions and obsessive-compulsive behavior (Graybiel & Rauch, 2000; Langen et al., 2011). Hence, it is reasonable to investigate brain structure abnormalities, especially basal ganglia changes, in the context of RRBI's in ASD.

In a study of 35 high functioning individuals with ASD and 36 TD individuals matched for age, gender and IQ (Ranson et al., 1999), the participants went through volumetric measurements of basal ganglia components (e.g. the bilateral caudate, putamen and globus pallidus) using Magnetic Resonance Imaging (MRI). The research revealed changes in the volume of the caudate nucleus, which was larger in participants diagnosed with ASD. Also, the increased caudate nucleus volume in ASD participants was correlative to an increase in their total brain volume, a phenomena that has been reported in several other studies (Anthony et al., 1989; Lainhart et al., 1997). The researchers claim that caudate volume in individuals with autism was associated with severity of compulsive behavior, difficulties with minor changes in routine, and motor stereotypies (Ranson et al., 1999).

The correlation between basal ganglia size, shape or volume and the degree of RRBI's severity in ASD was examined in additional structural studies. Hollander et al. (2005) revealed a larger right caudate volume in ASD compared with TD individuals. They also pointed out a correlation between right caudate and total putamen volumes and repetitive behavior scores on the ADI repetitive behavior scores (ADI-C), especially in the higher order OCD-like repetitive behaviors.

In addition, the differences in basal ganglia size in participants with ASD who were not on medication compared to TD individuals was investigated in another study which indicated a significant enlargement of the total caudate nucleus and bilateral putamen in the group with ASD. However, no significant correlations between ADI-R scores for RRBI's and basal ganglia structural changes were found (Langen, Durston, Staal, Palmen, & Van Engeland, 2007). Moreover, other studies indicate a positive correlation between basal ganglia volumes and metabolic activity and ASD (Buchsbaum et al., 2006; Voelbel, Bates, Buckman, Pandina, & Hendren, 2006). However, it is worth noting that some studies found no such correlations (Gaffney, Kuperman, Tsai, & Minchin, 1989; Goldman, O'Briend, Filipekf, Rapina, & Herbertg, 2013) or an opposite correlation (Estes et al., 2012).

A slightly different study using MRI focused on the cortico-striatal connectivity and not on the structural changes in volume. In this study, differences were found among the 50 participants with ASD, compared with 52 TD individuals (Abbott et al., 2018). The study's results suggested a possible association between RRBI's, as measured by the Repetitive Behavior Scale-Revised (RBS-R) scores, and imbalanced cortico-striatal intrinsic functional connectivity (iFC) in ASD, being increased for limbic circuits, but reduced for frontoparietal and motor circuits (Abbott et al., 2018).

Besides the basal ganglia, other brain structures were investigated in the context of RRBI's in ASD. One such study used MRI to test the gray matter volume of 24 participants with ASD compared with 23 TD individuals. The results suggested that the volumes of the medial frontal gyri, left pre-central gyrus, right post-central gyrus, right fusiform gyrus, caudate nuclei, and left hippocampus were larger in the ASD group relative to the TD individuals, while regions exhibiting smaller volumes in the ASD group were observed exclusively in the cerebellum (Rojas et al., 2006). Additionally, significant partial correlations were found between a measure of RRBI's in the ADI-R repetitive and stereotyped behavior domain and the volumes of the caudate nuclei, multiple frontal and temporal regions, and the cerebellum (Rojas et al., 2006). In a meta-analysis that investigated the differences between gray matter morphometric changes in boys diagnosed with ASD compared with girls, the girls showed less severe RRBI's as measured by the ADI-R. In addition, gray matter patterns in the motor cortex, the supplementary motor area (SMA), the cerebellum, the fusiform gyrus and the amygdala accurately differentiated between girls and boys with ASD, but not TD boys and girls. Moreover, gray matter pattern differences in the motor cortex, the SMA and in parts of the cerebellum were correlated with RRBI's severity in girls, whereas gray matter patterns in the right putamen were correlated with RRBI's in boys (Supekar & Menon, 2015).

Currently, it seems that there are no conclusive and convincing data from clinical trials regarding the significance of structural changes in the brains of individuals with ASD in general, and in the basal ganglia in particular, in relation to the prevalence and severity of RRBI's.

Genetic Abnormalities

Thanks to great advancements in technology over the past decade, it is now clear that ASD has a robust genetic component, since it has strong familial inheritance patterns and approximately 1000 genes are potentially implicated (Ramaswami & Geschwind, 2018). The new discoveries in this field have led to the ability to determine the etiology of 10–20% of ASD cases (Geschwind, 2011). There are no common mutations that are responsible for most cases of ASD (Abrahams & Geschwind, 2009; Geschwind, 2011), and the roles of the genes implicated in ASD vary, from synaptic proteins, RNA processing, and many others (Geschwind, 2011). Gene-gene interactions and epigenetic mechanisms are also considered part of the genetic causes of ASD (Ma et al., 2005).

Focusing on the genetic basis of RRBI's as one of the core features of ASD, some researchers have tried to find correlations between genetic variations and RRBI's in ASD. One such study was based on major Genome-Wide Association Study (GWAS(data from large ASD family cohorts. They used the ADI-R to assess RRBI's. Their results revealed that seven out of the 12 RRBI's that were measured were significantly familial and variable, and hence were subjected to further investigations using the GWAS. These RRBI's included circumscribed interests (68), repetitive use of objects (69), compulsions/rituals (70), unusual sensory interests (71), general sensitivity to noise (72), unusual attachments to objects (76), and stereotyped body movements (78). Eventually, after applying diverse methods of research and analysis, the researchers suggested two novel risk genes, SLC35B1 and PHB, for RRBI's in ASD (Cantor et al., 2017).

A second group of researchers tried to identify common variants that are associated with RRBI's. They performed the GWAS using the Autism Genetic Resource Exchange (AGRE) dataset and pointed out three Single Nucleotide Polymorphisms (SNPs) that might be correlated to RRBI's, one of which was also linked to schizophrenia. Additionally, all of the most significantly associated SNPs were found to be in close proximity to genes that are involved in neuron development (Tao et al., 2016). A third study approached the heritability issue of RRBI's in ASD from a different angle. The researchers examined the connection between severity of RRBI's among parents of children diagnosed with ASD as measured by the Interest in Patterns and Resistance to Changes subscales of the Autism Quotient, and the severity of RRBI's in the ASD diagnosed children themselves, measured by the Autism Diagnostic Observation Schedule RRB (ADOS RRB) standardized domain score. The results suggested that such a connection exists, as having both parents within the top 20% of RRBI's scores was associated with increased RRBI's scores for their children (Uljarevi, Evans, Alvares, & Whitehouse, 2016).

In a different genetic study, the researchers chose to focus on oxytocin (Francis, Kim, et al., 2016). Oxytocin is a neuropeptide that may have a role in the pathophysiology of ASD, and has even been suggested as a potential treatment for ASD (Anagnostou et al., 2012). This study tested whether there is a

correlation between SNPs in genes related to oxytocin or arginine-vasopressin systems, and ASD and its core symptoms, social communication deficits and RRBI's. The study results indicate that there is a correlation, with an especially significant association between SNP in the gene for oxytocin and having an ASD diagnosis (Francis, Kistner-griffin, et al., 2016). Another study by the same group also found a correlation between variants and SNPs in the receptors for oxytocin and vasopressin and being diagnosed with ASD. Specifically, variants correlated to RRBI's severity as measured by the ADOS RRB scores (Francis, Kim, et al., 2016).

Others have tried to clarify the previously assumed connection between RRBI's and several genetic disorders. In one study, the RRBI's were characterized using a questionnaire (Repetitive Behavior Questionnaire, "RBQ") with 797 participants who were diagnosed with specific genetic disorders such as Angelman, Cornelia de Lange, Cri du Chat, Fragile X, Lowe, Prader-Willi and Smith Magenis syndromes and a group of individuals with intellectual disability of heterogeneous etiologies. Comparing these genetic disorders, revealed they differ in their profile of RRBI. In Fragile X syndrome, hand stereotypies, lining up objects, restricted conversation, preference for routine and echolalia were the most prevalent forms of repetitive behavior. In Prader-Willi syndrome (PWS), hoarding and a preference for routine were more prevalent than other stereotyped behaviors. Attachment to objects was highly prevalent within the Cri du Chat syndrome and attachment to people was highly prevalent within the Smith-Magenis syndrome. Individuals with Angelman syndrome showed more heterogenicity in their RRBI's profile (Moss, Oliver, Arron, Burbidge, & Berg, 2009).These findings may suggest that in these known genetic disorders, there is a connection between the specific genetic abnormality and the clinical manifestations of RRBI's. The results support the claim that there is extreme heterogeneity of repetitive behavior across genetic syndromes, highlighting syndrome-specific profiles. A special focus in research was on PWS, a genetic disorder caused by the absence of paternally inherited genes in the 15q11-q13 region, as many individuals with PWS are diagnosed with ASD and high levels of RRBI's were also described (Bittel & Butler, 2005; E. M. Dykens, Cassidy, & King, 1999; E. Dykens & Shah, 2003; M. Lewis & Kim, 2009b; State & Dykens, 2000; Veltman, Craig, & Bolton, 2005).

Furthermore, in a study of more than 3000 twin pairs, it was found that RRBI's was both highly heritable and showed no shared environmental influence in univariate models (Ronald et al., 2006; Ronald, Happé, & Plomin, 2005), thereby adding reliability to the hypothesis that RRBI's etiology has a strong genetic basis.

As of today, the efforts to discover the genetic basis of ASD and its core symptoms, including RRBI's, are ongoing. Studies are being conducted worldwide in the hope of discovering new risk genes for ASD, in order to understand the impaired genetic mechanisms underlying this disorder and to find tailored treatments for ASD core symptoms.

Molecular (Neurotransmitters) Abnormalities

The last theory that will be presented in this chapter regarding the neurobiological basis of RRBI's in ASD implies that these repetitive behaviors are a result of an imbalance in neurotransmission systems.

There are data showing that dopamine has a role in RRBI's. First of all, dopamine is a principal player in the nigrostriatal dopamine pathway. As was mentioned earlier in this chapter, anatomical and physiological changes in the basal ganglia structures are considered by many to be a plausible cause for RRBI's in ASD (Mark H. Lewis & Bodfish, 1998; Scheel-Krüger et al., 1978; Scheel-Kruger et al., 1980). Additionally, stereotypic behaviors can be induced in many mammals, humans among them, by direct and indirect dopamine agonists (Mark H. Lewis & Baumeister, 1982; Mark H. Lewis & Bodfish, 1998). Early works in animal models also exhibit the importance of dopaminergic activity in the basal ganglia for RRBI's, as dopamine or dopamine agonists can cause RRBI's (Ernst & Smelik, 1966; M. Lewis & Kim, 2009a). Furthermore, it was demonstrated that inhibiting dopamine synthesis resulted in a reduction in RRBI's, while treating animals with a dopamine precursor caused increased RRBI's patterns (Kennes, Odberg, & de Rycke, 1988; Mark H. Lewis & Bodfish, 1998). However, a clinical study on individuals with intellectual disability who also had RRBI's revealed they had low plasma levels of homovanillic acid (HVA), a dopamine metabolite (M. H. Lewis et al., 1996; Mark H. Lewis & Bodfish, 1998). Later works indicated that having mutations in the dopamine signaling pathway was correlated with having an ASD diagnosis (Hamilton et al., 2014; Nguyen et al., 2014), and also with a specific type of RRBI's ("insistence on sameness") in ASD as measured by the ADI-R (Staal, De Krom, & De Jonge, 2012).

Gamma-aminobutyric acid (GABA) signaling pathways may also play a major role in the mechanism causing RRBI's in ASD. Many researchers report a correlation between changes in components of the GABA signaling pathways and having an ASD diagnosis (Barnby et al., 2005; Coghlan, Horder, Inkster, Mendez, & Declan, 2012; Collins et al., 2006; Fatemi, Reutiman, Folsom, & Thoras, 2009; M. Lewis & Kim, 2009a; Ma et al., 2005). In addition, like dopamine, GABA has a role in the nigrostriatal dopamine system, which was mentioned in this chapter several times as a suggested mechanism that causes RRBI's in ASD (Mark H. Lewis & Baumeister, 1982). Moreover, an animal study demonstrated that dysfunction of the MECP2 gene (a mutation in this gene may cause Rett syndrome) in GABAergic neurons can harm its function, and contribute to numerous neuropsychiatric phenotypes and symptoms, RRBI's among them (Chao et al., 2011). In an MRS study on 18 children with primary complex motor stereotypies (CMS) and 24 TD individuals, it was demonstrated that children with CMS had lower levels of GABA in the anterior cingulate cortex (ACC) and in the striatum. Furthermore, within the CMS group, a reduced GABA/creatinine ratio in the ACC was significantly associated with greater severity of motor stereotypies (Harris et al., 2016). Moreover, in two mouse models for ASD, improvement in RRBI's was demonstrated after treatment with $GABA_B$ receptor agonists (Silverman et al., 2015).

In addition to dopamine and GABA, there is a claim that serotonin (5-HT) is also related to RRBI's in ASD, since 5-HT axons project from the raphe nuclei to basal ganglia structures, thereby creating dopamine-5-HT interactions (Mark H. Lewis & Bodfish, 1998). Like other neurotransmitters, genetic changes in genes involved in the 5-HT signaling were implicated in ASD (Chakrabotri et al., 2016; R. M. Smith, Banks, Hansen, Sadee, & Herman, 2014; Yonan et al., 2003). In an early work in this field, it was found that 6 out of 23 participants with ASD had higher blood 5-HT levels, compared with individuals with typical development and those with intellectual disability without autism (Schain & Freedman, 1961). Furthermore, some consider serotonin reuptake inhibitors (SSRIs) to be an effective treatment for RRBI's (Sutcliffe et al., 2005). Additionally, reduction of central nervous system serotonin, induced by acute tryptophan depletion, caused a worsening of stereotyped behavior (Cook & Leventhal, 1996). However, results of a systematic review conclude that although SSRIs are sometimes given to reduce obsessive-compulsive behaviors, there is no evidence to support the use of SSRIs to treat autism in children, and there is limited evidence to suggest the effectiveness of SSRIs in adults with autism (Williams, Brignell, Randall, Silove, & Hazell, 2013).

Alongside the other aforementioned neurotransmitters, the glutamatergic system, which is a primary excitatory system involved in cognitive functions such as learning and memory (Jamain et al., 2002), also plays a key role in ASD pathogenesis (Yang & Chang, 2014). A correlation was found between genetic abnormalities in the glutamatergic system's components and an ASD diagnosis (Jamain et al., 2002; Nguyen et al., 2014). Moreover, similarly to GABA, the dopaminergic system can modulate glutamatergic signaling (Li, Wang, & Gao, 2012; Nguyen et al., 2014), therefore it is probable that disturbances in glutamate signaling are also a part of the etiology of RRBI's.

In conclusion, many neurotransmission systems are implicated in ASD, and may be a part of the etiology and pathophysiology of RRBI's in ASD. However, the data from clinical studies on individuals with ASD in this field are rather sparse, and there is a need for more clinical evidence in order to support these theories.

Summary

Currently, the etiology of ASD in general, and of its specific core domains, the social-communication impairments and RRBI's in particular, is not fully understood. Hence, suggested mechanisms regarding the pathophysiology of RRBI's in ASD are copious, and range from psychological theories to genetic, anatomical, functional, and metabolic abnormalities. Considering the growing impact of the field of genetics on diagnosis of complex disorders in general, and of ASD in particular, it is likely that further genetic advancement will aid in uncovering the neurobiological basis of RRBI's in ASD. The understanding of the underlying mechanisms of RRBI's may lead to the development of personalized biological treatments to improve these restricted and repetitive behaviors that negatively impact the quality of life in ASD.

References

Abbott, A. E., Linke, A. C., Nair, A., Jahedi, A., Alba, L. A., Keown, C. L., et al. (2018). Repetitive behaviors in autism are linked to imbalance of corticostriatal connectivity: A functional connectivity MRI study. *Social Cognitive and Affective Neuroscience, 13*(1), 32–42. https://doi.org/10.1093/scan/nsx129.

Abrahams, B. S., & Geschwind, D. H. (2009). Advances in autism genetics: On the threshold of a new neurobiology. *Nature Reviews Genettics, 9*(5), 341–355. https://doi.org/10.1038/nrg2346.Advances.

Albin, R. L., Young, A. B., Penney, J. B., Roger, L. A., & Young, B. B. (1989). The functional anatomy of basal ganglia disorders. *Trends in Neurosciences, 12*(10), 366–375.

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). Retrieved from https://www.autismspeaks.org/what-autism/diagnosis/dsm-5-diagnostic-criteria.

Anagnostou, E., Soorya, L., Chaplin, W., Bartz, J. A., Halpern, D., Wasserman, S., et al. (2012). Intranasal oxytocin versus placebo in the treatment of adults with autism spectrum disorders: A randomized controlled trial. *Molecular Autism, 3*(1), 16. https://doi.org/10.1186/2040-2392-3-16.

Anthony, B., Luthert, P., Bolton, P., Le Couteur, A., Rutter, M., & Harding, B. (1989). Autism and Megalencephaly. *The Lancet, 341*, 1225–1226.

Barnby, G., Abbott, A., Sykes, N., Morris, A., Weeks, D. E., Mott, R., et al. (2005). Candidate-gene screening and association analysis at the autism-susceptibility locus on chromosome 16p: Evidence of association at GRIN2A and ABAT. *American Journal of Human Genetics, 76*(6), 950–966.

Baron-Cohen, S. (1989). Do autistic children have obsessions and compulsions? *British Journal of Clinical Psycology, 28*(3), 193–200.

Bishop, S. L., Hus, V., Duncan, A., Huerta, M., Gotham, K., Pickles, A., et al. (2013). Subcategories of restricted and repetitive behaviors in children with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 43*(6), 1287–1297. https://doi.org/10.1007/s10803-012-1671-0.

Bishop, S. L., Richler, J., & Lord, C. (2006). Association between restricted and repetitive behaviors and nonverbal IQ in children with Autism Spectrum Disorders. *Child Neuropsychology, 12*(4–5), 247–267. https://doi.org/10.1080/09297040600630288.

Bittel, D. C., & Butler, M. G. (2005). Prader – Willi syndrome: Clinical genetics, cytogenetics and molecular biology. *Expert Reviews in Molecular Medicine, 7*(14), 1–20. https://doi.org/10.1017/S1462399405009531.

Bodfish, J. W., Symons, F. J., Parker, D. E., & Lewis, M. H. (2000). Varieties of repetitive behaviour in autism: Comparison to mental retardation. *Journal of Autism and Developmental Disorders, 30*(3), 237–243. https://doi.org/10.1023/A:1005596502855.

Boyer, P., & Lie'nard, P. (2006). Why ritualized behavior? Precaution Systems and action parsing in developmental, pathological and cultural rituals. *Behavioral and Brain Science, 29*, 595–650. https://doi.org/10.1017/S0140525X06009332.

Buchsbaum, M. S., Hazlett, E. A., Ph, D., Licalzi, E. M., Cartwright, C., & Hollander, E. (2006). Volumetric analysis and three-dimensional glucose metabolic mapping of the striatum and thalamus in patients with autism spectrum disorders. *American Journal of Psychiatry, 163*(7), 1252–1263.

Cantor, R. M., Navarro, L., Won, H., Walker, R. L., Lowe, J. K., & Geschwind, D. H. (2017). ASD restricted and repetitive behaviors associated at 17q21.33: Genes prioritized by expression in fetal brains. *Molecular Psychiatry, 00*(September 2016), 1–8. https://doi.org/10.1038/mp.2017.114.

Caravaggio, F., Nakajima, S., Plitman, E., Gerretsen, P., Chung, J. K., Iwata, Y., & Graff-Guerrero, A. (2016). The effect of striatal dopamine depletion on striatal and cortical glutamate: A mini-

review. *Progress in Neuro-Psychopharmacology and Biological Psychiatry, 65*, 49–53. https://doi.org/10.1016/j.pnpbp.2015.08.013.

Chakrabotri, B., Verma, D., Karmakar, A., Jaiswal, P., Sanyal, A., Paul, D., et al. (2016). Genetic variants of MAOB affect serotonin level and specific behavioral attributes to increase autism spectrum disorder (ASD) susceptibility in males. *Progress in Neuropsychopharmacology and Biological Psychiatry, 71*, 123–136. https://doi.org/10.1016/j.pnpbp.2016.07.001.

Chao, H., Chen, H., Samaco, R. C., Xue, M., Yoo, J., Neul, J. L., et al. (2011). GABAergic dysfunction mediates autism-like stereotypies and Rett syndrome phenotypes. *Nature, 468*(7321), 263–269. https://doi.org/10.1038/nature09582.GABAergic.

Coghlan, S., Horder, J., Inkster, B., Mendez, M. A., & Declan, G. (2012). GABA system dysfunction in autism and related disorders: From synapse to symptoms. *Neuroscience & Biobehavioral Reviews, 36*(9), 2044–2055. https://doi.org/10.1016/j.neubiorev.2012.07.005.GABA.

Collins, A. L., Ma, D., Whitehead, P. L., Martin, E. R., Wright, H. H., Abramson, R. K., et al. (2006). Investigation of autism and GABA receptor subunit genes in multiple ethnic groups. *Neurogenetics, 7*(3), 167–174. https://doi.org/10.1007/s10048-006-0045-1.

Cook, E. H., & Leventhal, B. L. (1996). The serotonin system in autism. *Current Opinion in Pediatrics, 8*(4), 348–354. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/9053096.

Dykens, E. M., Cassidy, S. B., & King, B. H. (1999). Maladaptive behavior differences in Prader-Willi syndrome due to parental deletion versus maternal uniparental disomy. *American Journal on Mental Retardation, 104*(1), 67–77.

Dykens, E., & Shah, B. (2003). Psychiatric disorders in Prader-Willi syndrome epidemiology and management. *CNS Drugs, 17*(3), 167–178.

Ernst, A. M., & Smelik, P. G. (1966). Site of action of dopamine and apomorphine on compulsive gnawing behaviour in rats. *Experientia, 22*(12), 837–838. https://doi.org/10.1007/BF01897450.

Estes, A., Shaw, D. W. W., Sparks, B. F., Friedman, S., Giedd, J. N., Dawson, G., et al. (2012). Basal ganglia morphometry and repetitive behavior in young children with autism spectrum disorder. *Autism Research, 4*(3), 212–220. https://doi.org/10.1002/aur.193.Basal.

Evans, D. W. (2017). Relationship between repetitive behaviour and fear across normative development, autism spectrum disorder, and down syndrome. *Autism Research, 10*(3), 502–507. https://doi.org/10.1002/aur.1674.

Evans, D. W., Leckmn, J. F., Carter, A., Raznick, J. S., Henshaw, D., King, R. A., & Pauls, D. (1997). Rituals, habit and perfectionism: The prevalence and development of compulsive-like behavior in normal young children. *Child Development, 68*(1), 58–68.

Fatemi, S. H., Reutiman, T. T., Folsom, T. D., & Thoras, P. D. (2009). GABA A receptor downregulation in brains of subjects with autism. *Journal of Autism and Developmental Disorders, 39*, 223–230. https://doi.org/10.1007/s10803-008-0646-7.

Francis, S. M., Kim, S., Kistner-griffin, E., Guter, S., Cook, E. H., Jacob, S., & Ebstein, R. P. (2016). ASD and genetic associations with receptors for oxytocin and vasopressin—AVPR1A, AVPR1B, and OXTR. *Frontiers in Neuroscience, 10*(November), 1–10. https://doi.org/10.3389/fnins.2016.00516.

Francis, S. M., Kistner-griffin, E., Yan, Z., Guter, S., Cook, E. H., & Jacob, S. (2016). Variants in adjacent oxytocin/vasopressin gene region and associations with ASD diagnosis and other autism related endophenotypes. *Frontiers in Neuroscience, 10*(May), 195. https://doi.org/10.3389/fnins.2016.00195.

Frith, U., & Happe, F. (1994). Autism: beyond " theory of mind". *Cognition, 50*, 115–132.

Gaffney, G. R., Kuperman, S., Tsai, L. Y., & Minchin, S. (1989). Forebrain structure in infantile autism. *Journal of the American Academy of Child and Adolescent Psychiatry, 28*(4), 534–537. https://doi.org/10.1097/00004583-198907000-00011.

Geschwind, D. H. (2011). Genetics of autism spectrum disorders. *Trends in Cognitive Sciences, 15*(9), 409–416. https://doi.org/10.1016/j.tics.2011.07.003.

Goldman, S., O'Briend, L. M., Filipekf, P. A., Rapina, I., & Herbertg, M. R. (2013). Motor stereotypies and volumetric brain alterations in children with Autistic Disorder. *Research in Autism Spectrum Disorders, 7*(1), 82–92. https://doi.org/10.1016/j.rasd.2012.07.005.Motor.

Graybiel, A. M., & Rauch, S. L. (2000). Toward a neurobiology of obsessive-compulsive disorder. *Neuron, 28*(4), 343–347.

Hall, S., Thorns, T., & Oliver, C. (2003). Structural and environmental characteristics of stereotyped behaviors. *American Journal of Mental Retardation, 108*(6), 391–402.

Hamilton, P. J., Campbell, N. G., Sharma, S., Erreger, K., Herborg Hansen, F., Saunders, C., et al. (2014). De novo mutation in the dopamine transporter gene associates dopamine dysfunction with autism spectrum disorder Peter. *Molecular Psychiatry, 18*(12), 1315–1323. https://doi.org/10.1038/mp.2013.102.De.

Harris, A. D., Singer, H. S., Horska, A., Kline, T., Ryan, M., Edden, R. A., & Mahone, E. M. (2016). GABA and glutamate in children with primary complex motor stereotypies: A1H MRS study at 7T. *American Journal of Neuroradiology, 37*(3), 552–557. https://doi.org/10.3174/ajnr.A4547.GABA.

Hollander, E., Anagnostou, E., Chaplin, W., Esposito, K., Haznedar, M. M., Licalzi, E., … Buchsbaum, M. (2005). Striatal volume on magnetic resonance imaging and repetitive behaviors in autism. *Biological Psychiatry, 58*(3), 226–232. https://doi.org/10.1016/j.biopsych.2005.03.040

Hutt, B. Y. C., & Hutt, S. J. (1965). Effects of environmental complexity on stereoptyped behaviours of children. *Animal Behaviour, 13*(1), 1–4.

Hutt, C., Hutt, S. J., Lee, D., & Ounsted, C. (1964). Arousal and childhood autism. *Nature, 204*(4961), 908–909.

Jamain, S., Betancur, C., Quach, H., Philippe, A., Fellous, M., Giros, B., et al. (2002). Linkage and association of the glutamate receptor 6 gene with autism. *Molecular Psychiatry, 7*(3), 302–310. https://doi.org/10.1038/sj/mp/4000979.

Jiujias, M., Kelley, E., Hall, L., & Kelley, E. (2017). Restricted, repetitive behaviors in autism spectrum disorder and obsessive – compulsive disorder: a comparative review. *Child Psychiatry and Human Development, 48*(6), 944–959. https://doi.org/10.1007/s10578-017-0717-0.

Joyce, C., Honey, E., Leekam, S. R., Barrett, S. L., Rodgers, J., & Joyce, C. (2017). Anxiety, intolerance of uncertainty and restricted and repetitive behaviour: Insights directly from young people with ASD. *Journal of Autism and Developmental Disorders, 47*(12), 3789–3802. https://doi.org/10.1007/s10803-017-3027-2.

Just, M. A., Cherkassky, V. L., Keller, T. A., & Minshew, N. J. (2004). Cortical activation and synchronization during sentence comprehension in high-functioning autism: Evidence of underconnectivity. *Brain, 127*, 1811–1821. https://doi.org/10.1093/brain/awh199.

Kennes, F. O., Odberg, Y. B., & de Rycke, P. H. (1988). Changes in naxolone and heloperidol effects during the development of captivity-induced jumping stereotypy in bank voles. *European Journal of Pharmacology, 153*(1), 19–24.

Kim, S. H., & Lord, C. (2010). Restricted and repetitive behaviors in toddlers and preschoolers with autism spectrum disorders based on the Autism Diagnostic Observation Schedule (ADOS). *Autism Research, 3*(4), 162–173. https://doi.org/10.1002/aur.142.

Lainhart, J. E., Piven, J., Wzorek, M., Ph, D., Landa, R., Ph, D., et al. (1997). Macrocephaly in children and adults with autism. *Journal of the American Academy of Child and Adolescent Psychiatry, 36*(2), 282–290. https://doi.org/10.1097/00004583-199702000-00019.

Langen, M., Durston, S., Kas, M. J. H., Van Engeland, H., & Staal, W. G. (2011). The neurobiology of repetitive behavior … and men. *Neuroscience and Biobehavioral Reviews, 35*(1), 356–365. https://doi.org/10.1016/j.neubiorev.2010.02.005.

Langen, M., Durston, S., Staal, W. G., Palmen, S. J. M. C., & Van Engeland, H. (2007). Caudate nucleus is enlarged in high-functioning medication-naive subjects with autism. *Biological Psychiatry, 62*(3), 262–266. https://doi.org/10.1016/j.biopsych.2006.09.040.

Lewis, M., & Kim, S.-J. (2009a). The pathophysiology of restricted repetitive behavior. *Journal of Neurodevelopmental Disorders, 1*(2), 114–132. https://doi.org/10.1007/s11689-009-9019-6.

Lewis, M., & Kim, S. (2009b). The pathophysiology of restricted repetitive behavior. *Journal of Neurodevelopmental Disorders, 1*(June), 114–132. https://doi.org/10.1007/s11689-009-9019-6.

Lewis, M. H., Bodfish, J. W., Powell, S. B., Wiest, K., Darling, M., & Golden, R. N. (1996). Plasma HVA in adults with mental retardation and stereotyped behavior: biochemical evidence for a dopamine deficiency model. *American Journal of Mental Retardation : AJMR, 100*(4), 413–418. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/8718995.

Lewis, M. H., & Baumeister, A. A. (1982). Stereotyped mannerisms in mentally retarded persons: Animal models and theoretical analyses. *International Review of Research in Mental Retardation, 11*, 123–161.

Lewis, M. H., & Bodfish, J. W. (1998). Repetitive behavior disorders in autism. *Mental Retardation and Developmental Disabilities Research Reviews, 4*, 80–89.

Li, Y.-C., Wang, M.-J., & Gao, W.-J. (2012). Hyperdopaminergic modulation of inhibitory transmission is dependent on GSK-3β signaling-mediated trafficking of GABAA receptors. *Journal of Neurochemistry, 122*(2), 308–320. https://doi.org/10.1111/j.1471-4159.2012.07790.x.

Lord, C., Risi, S., Lambrecht, L., Cook, E. H. J., Leventhal, B. L., DiLavore, P. C., et al. (2000). The Autism Diagnostic Schedule – Generic: A standard measures of social and communication deficits associated with the spectrum of autism. *Journal of Autism and Developmental Disorders, 30*(3), 205–223. https://doi.org/10.1023/A:1005592401947.

Lord, C., Rutter, M., & LeCouteur, A. (1994). Autism diagnostic interview: A revised version of a diagnostic interview for caregivers of individual s with possible pervasive developmental disorders. *Journal of Autism and Developmental Disorders, 24*(5), 659–685. Retrieved from http://search.ebscohost.com/login.aspx?direct=true&db=epref&AN=JADD.BD.FEI.LORD.ADIRVD&site=ehost-live.

Lovaas, I., Newsome, C., & Hickman, C. (1987). Self-stimulatory behavior and perceptual reinforcement. *Journal of Applied Behavior Analysis, 20*(1), 45–68.

Ma, D. Q., Whitehead, P. L., Menold, M. M., Martin, E. R., Mei, H., Ritchie, M. D., et al. (2005). Identification of significant association and gene-gene interaction of GABA receptor subunit genes in autism. *The American Journal of Human Genetics, 77*(3), 377–388.

Monk, C. S., Peltier, S. J., Wiggins, J. L., Weng, S. J., Carrasco, M., Risi, S., & Lord, C. (2009). Abnormalities of intrinsic functional connectivity in autism spectrum disorders. *NeuroImage, 47*(2), 764–772. https://doi.org/10.1016/j.neuroimage.2009.04.069.

Moss, J., Oliver, C., Arron, K., Burbidge, C., & Berg, K. (2009). The prevalence and phenomenology of repetitive behavior in genetic syndromes. *Journal of Autism and Developmental Disorders, 39*(4), 572–588. https://doi.org/10.1007/s10803-008-0655-6.

Mostert-Kerckhoffs, M. A. L., Staal, W. G., & Houben, R. H. (2015). Stop and change : Inhibition and flexibility skills are related to repetitive behavior in children and young adults with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 45*(10), 3148–3158. https://doi.org/10.1007/s10803-015-2473-y.

Muhle, R. A., Reed, H. E., Stratigos, K. A., & Veenstra-vanderweele, J. (2018). The emerging clinical neuroscience of autism spectrum disorder A review. *JAMA Psychiatry, 75*(5), 514–523. https://doi.org/10.1001/jamapsychiatry.2017.4685.

Newschaffer, C. J., Denckla, Æ. M. B., Landa, R., & Mostofsky, Æ. S. H. (2006). Motor signs distinguish children with high functioning autism and Asperger's syndrome from controls. *Journal of Autism Developmentall Disorder, 36*(1), 613–621. https://doi.org/10.1007/s10803-006-0109-y.

Nguyen, M., Roth, A., Kyzar, E. J., Poudel, M. K., Wong, K., Stewart, A. M., & Kalueff, A. V. (2014). Decoding the contribution of dopaminergic genes and pathways to autism spectrum disorder (ASD). *Neurochemistry International, 66*(1), 15–26. https://doi.org/10.1016/j.neuint.2014.01.002.

Nomi, J. S., & Uddin, L. Q. (2015). Developmental changes in large-scale network connectivity in autism. *NeuroImage: Clinical, 7*, 732–741. https://doi.org/10.1016/j.nicl.2015.02.024.

Ramaswami, G., & Geschwind, D. H. (2018). Genetics of autism spectrum disorder. In *Handbook of clinical neurology* (Vol. 147, 1st ed.). https://doi.org/10.1016/B978-0-444-63233-3.00021-X.

Ranson, B. J., Piven, J., Lonnie, L., Vest, C., Mohamed, S., Bailey, J., et al. (1999). An MRI study of the basal ganglia in autism. *Progress in Neuro-Psychopharmacology and Biological Psychiatry, 23*(Apa 1995), 613–624.

Richler, J., Huerta, M., Bishop, S. L., & Lord, C. (2010). Developmental trajectories of restricted and repetitive behaviors and interests in children with autism spectrum disorders. *Development and Psychopathology, 22*(1), 55–69. https://doi.org/10.1017/S0954579409990265.

Rojas, D. C., Peterson, E., Winterrowd, E., Reite, M. L., Rogers, S. J., & Tregellas, J. R. (2006). Regional gray matter volumetric changes in autism associated with social and repetitive behavior symptoms. *BMC Psychiatry, 6*(1), 56. https://doi.org/10.1186/1471-244X-6-56.

Ronald, A., Happ, F., Bolton, P., Sych, F. R. C. P., Butcher, L. E. E. M., Price, T. S., & Wheelwright, S. (2006). Genetic heterogeneity between the three components of the autism spectrum: A twin study. *Journal of the American Academy of Child & Adolescent Psychiatry, 45*(6), 691–699. https://doi.org/10.1097/01.chi.0000215325.13058.9d.

Ronald, A., Happé, F., & Plomin, R. (2005). The genetic relationship between individual differences in social and nonsocial behaviours characteristic of autism. *Developmental Science, 8*(5), 444–458. https://doi.org/10.1111/j.1467-7687.2005.00433.x.

Rosenthal, M., Wallace, G. L., Lawson, R., Wills, M. C., Dixon, E., Yerys, B. E., & Kenworthy, L. (2013). Impairments in real-world executive function increase from childhood to adolescence in autism spectrum disorders. *Neuropsychology, 27*(1), 13–18. https://doi.org/10.1037/a0031299.

Schain, R. J., & Freedman, D. X. (1961). Studies on 5-bydroxyindole metabolism in autistic and other mentally retarded children. *The Journal of Pediatrics, 58*(3), 315–320.

Scheel-Krüger, J., Arnt, J., Braestrup, C., Christensen, A. V., Cools, A. R., & Magelund, G. (1978). GABA-dopamine interaction in substantia nigra and nucleus accumbens -relevance to behavioral stimulation and stereotyped behavior. *Advances in Biochemical Psychopharmacology, 19*, 343–346. Retrieved from http://www.ncbi.nlm.nih.gov/pubmed/567933.

Scheel-Kruger, J., Arnt, J., Magelund, G., Olianas, M., Przewlocka, B., & Christensen, A. V. (1980). Behavioural functions of GABA in basal ganglia and limbic system. *Brain Research Bulletin, 5*(2), 261–267.

Schmitt, L. M., White, S. P., Cook, E. H., Sweeney, J. A., & Mosconi, M. W. (2017). Cognitive mechanisms of inhibitory control deficits in autism spectrum disorder. *Journal of Child Psychology and Psychiatry, 59*(5), 586–595. https://doi.org/10.1111/jcpp.12837.

Shih, P., Shen, M., Öttl, B., Keehn, B., Gaffrey, M. S., & Müller, R. A. (2010). Atypical network connectivity for imitation in autism spectrum disorder. *Neuropsychologia, 48*(10), 2931–2939. https://doi.org/10.1016/j.neuropsychologia.2010.05.035.

Silverman, J. L., Pride, M. C., Hayes, J. E., Puhger, K. R., Butler-struben, H. M., Baker, S., & Crawley, J. N. (2015). GABA B receptor agonist R-baclofen reverses social deficits and reduces repetitive behavior in two mouse models of autism. *Neuropsychopharmacology, 40*(9), 2228–2239. https://doi.org/10.1038/npp.2015.66.

Sloman, K. N., LaRue, R. H., Weiss, M. J., & Hansford, A. S. (2013). Best practices in the assessment and treatment of stereotypical behaviour in individuals with autism. In J. M. Zachor & A. Ditza (Eds.), *Understanding autism* (pp. 29–37). Nova Science Publishers.

Smith, R. M., Banks, W., Hansen, E., Sadee, W., & Herman, G. E. (2014). Family-based clinical associations and functional characterization of the serotonin 2A receptor gene (HTR2A) in autism spectrum disorder. *Autism Research, 7*(4), 459–467. https://doi.org/10.1002/aur.1383.

Smith, Y., Bevan, M. D., Shink, E., & Bolam, J. P. (1998). Microcircuitry of the direct and indirect pathways of the basal ganglia. *Neuroscience, 86*(2), 353–387.

Staal, W. G., De Krom, M., & De Jonge, M. V. (2012). Brief report: The dopamine-3-receptor gene (DRD3) is associated with specific repetitive behavior in autism spectrum disorder (ASD). *Journal of Autism and Developmental Disorders, 42*(5), 885–888. https://doi.org/10.1007/s10803-011-1312-z.

State, M. W., & Dykens, E. M. (2000). Genetics of childhood disorders: XV. Prader-Willi syndrome: Genes, brain, and behavior. *Developmental Neurobiology, 39*(6), 797–800. https://doi.org/10.1097/00004583-200006000-00021.

Stocco, A., Lebiere, C., & Anderson, J. R. (2011). Conditional routing of information to the cortex: A model of the basal Ganglia's role in cognitive coordination. *Psychologic Review, 117*(2), 541–574. https://doi.org/10.1037/a0019077.Conditional.

Stratis, E. A., & Lecavalier, L. (2013). Restricted and repetitive behaviors and psychiatric symptoms in youth with autism spectrum disorders. *Research in Autism Spectrum Disorders, 7*(6), 757–766. https://doi.org/10.1016/j.rasd.2013.02.017.

Street, D. (1994). The psychology of perservative and stereotyped behaviour. *Progress in Neurobiology, 44*(2), 221–231.

Sullivan, J., Rodgers, J., Lidstone, J., Uljarevic, M., Mcconachie, H., Freeston, M., et al. (2014). Research in Autism Spectrum Disorders Relations among restricted and repetitive behaviors, anxiety and sensory features in children with autism spectrum disorders. *Research in Autism Spectrum Disorders, 8*(2), 82–92. https://doi.org/10.1016/j.rasd.2013.10.001.

Supekar, K., & Menon, V. (2015). Sex differences in structural organization of motor systems and their dissociable links with repetitive/restricted behaviors in children with autism. *Molecular Autism, 6*(50). https://doi.org/10.1186/s13229-015-0042-z.

Sutcliffe, J. S., Delahanty, R. J., Prasad, H. C., Mccauley, J. L., Han, Q., Jiang, L., et al. (2005). Allelic heterogeneity at the serotonin transporter locus (SLC6A4) confers susceptibility to autism and rigid-compulsive behaviors. *American Journal of Human Genetics, 77*, 265–279.

Tao, Y., Gao, H., Ackerman, B., Guo, W., Saffen, D., & Shugart, Y. Y. (2016). Evidence for contribution of common genetic variants within chromosome 8p21.2-8p21.1 to restricted and repetitive behaviors in autism spectrum disorders. *BMC Genomics, 17*(163), 1–15. https://doi.org/10.1186/s12864-016-2475-y.

Turner, M. (1999). Repetitive behaviour in autism: A review of psychological research. *Journal of Child Psychology and Psychiatry, 40*(6), 839–849.

Uljarevi, M., Evans, D. W., Alvares, G. A., & Whitehouse, A. J. O. (2016). Short report: Relationship between restricted and repetitive behaviours in children with autism spectrum disorder and their parents. *Molecular Autism, 7*(29), 1–5. https://doi.org/10.1186/s13229-016-0091-y.

Veltman, M. W. M., Craig, E. E., & Bolton, P. F. (2005). Autism spectrum disorders in Prader – Willi and Angelman syndromes: A systematic review. *Psychiatric Genetics, 15*(4), 243–254.

Voelbel, G. T., Bates, M. E., Buckman, J. F., Pandina, G., & Hendren, R. L. (2006). Caudate nucleus volume and cognitive performance: Are they related in childhood psychopathology? *Biological Psychiatry, 60*(9), 942–950. https://doi.org/10.1016/j.biopsych.2006.03.071.

Williams, K., Brignell, A., Randall, M., Silove, N., & Hazell, P. (2013). Selective serotonin reuptake inhibitors (SSRIs) for autism spectrum disorders (ASD) (Review). *Cochrane Database of Systematic Reviews*, ((8), CD004677. https://doi.org/10.1002/14651858.CD004677.pub3. https://www.cochranelibrary.com/.

Yang, P., & Chang, C.-L. (2014). Glutamate-mediated signaling and autism spectrum disorders: Emerging treatment targets. *Current Pharmaceutical Design, 20*(32), 5186–5193. https://doi.org/10.2174/1381612819666140110120725.

Yonan, A. L., Alarco, M., Cheng, R., Magnusson, P. K. E., Spence, S. J., Palmer, A. A., et al. (2003). A genomewide screen of 345 families for autism-susceptibility loci. *American Journal of Human Genetics, 73*(4), 886–897.

Chapter 4
Repeated Behavioral Patterns in Animal Models of Autism

Nisim Perets and Daniel Offen

ASD is a neurodevelopmental disorder diagnosed mainly by behavioral symptoms, with the core deficits modeled in animals for bio-psychiatric research and drug development purposes. The vast majority of animal models of ASD are based on mice research, because (A) mice are relatively easy to genetically manipulate (much easier than rats for example, yet harder than flies) and (B) they are mammals which present relatively complex behaviors, and therefore have become the main focus for bio-psychiatric studies.

The variety of animal models of ASD can be divided into three groups according to the different approaches of medical and biological knowledge regarding ASD, collected from human research. The first cluster of animal models is the genetically modified animals whose genome was altered to include genetic mutations that were found in ASD patients (Bakker et al., 1994; Peça et al., 2011; Tabuchi et al., 2007). These mutations potentially lead to ASD symptoms. The second cluster of models include ASD symptoms in animals. These models aim to mimic the cases in which ASD symptoms may have developed as a result of viral/bacterial infection during different trimesters of the pregnancy that led to deficits in neurodevelopmental processes (Patterson, 2009; Zerbo et al., 2013). The third cluster is the multi-factorial models of ASD which are based on combinations of factors that lead to ASD symptoms (Scattoni, Shruti, Ricceri, & Crawley, 2008). This cluster of animal models mimics the clinical cases of patients whose behavioral and cognitive symptoms match the ASD diagnosis, yet neither specific biological markers nor genetic mutations can be traced and account for the deficits.

N. Perets (✉)
Sagol School of Neuroscience, Tel Aviv University, Tel Aviv, Israel

D. Offen
Sagol School of Neuroscience, Tel Aviv University, Tel Aviv, Israel

Sacklar School of Medicine, Department of Human Genetics and Biochemistry, Tel Aviv University, Tel Aviv, Israel

E. Gal, N. Yirmiya (eds.), *Repetitive and Restricted Behaviors and Interests in Autism Spectrum Disorders*, Autism and Child Psychopathology Series,
https://doi.org/10.1007/978-3-030-66445-9_4

This chapter will review the most common animal models of the three different clusters, including their advantages and drawbacks, while emphasizing the repetitive and obsessive disorders as expressed in the various animal models.

Before diving deeper into the different phenotypes of the different models of ASD, we discuss fundamental questions regarding animal behavior: How can we differentiate between normal behavior of animals and "ASD-like" symptoms? What are the behavioral tests that can reliably measure those symptoms? Animal behaviors (especially mice) have been extensively studied during the last few decades. Their reactions to different types of stimuli in natural and controlled environments are well described and highly predictable. Therefore, by using genetic or developmental manipulations, the differences in responses to known stimuli between "ASD mice" and healthy controls can be measured. Based on the mice behavioral repertoire, several behavioral examinations have been developed and validated. The same behavioral examinations are used across the different models, sometimes with slight adaptations.

ASD in humans is characterized by two core symptoms, which include (1) Persistent deficits in social communication and social interaction across multiple contexts, and (2) Restricted, repetitive patterns of behavior, interests, or activities (DSM5). Therefore, the behavioral tests to measure ASD-like behaviors among mice focus on those domains with the relevant adaptations to rodent's natural behaviors.

ASD-Like Mice Models Based on Genetic Manipulation

Genetic disruptions were found highly related to several types of ASD. Even though the causative genes and their downstream impacts are largely unknown, familial and twin studies indicate that ASD has an extremely high genetic background.

Shank3 Knockout/Mutation

One of the most common genetic causes for ASD is a mutation in the SHANK3 gene. Different malfunctions in SHANK3 are responsible for around 1–2% of ASD cases in humans (Harony-Nicolas et al., 2017). Symptomatically, mutation in SHANK3 (usually knockout of different areas of the gene) can be characterized in neonatal developmental delay, absent to severely delayed speech, autistic behavior and also minor dysmorphic features (Durand et al., 2007). SHANK3 is an excitatory synapses postsynaptic protein. SHANK proteins are used as anchors of the synapse and are used for their stabilization (Durand et al., 2007). Disruption at the genetic level is thought to be highly related to the development of 22q13 deletion syndrome (Phelan–McDermid syndrome) and other types of ASDs (Peça et al., 2011).

Mouse models were established based on Shank3 knockout or mutations. These mice present the core symptoms of ASD including increased repetitive behaviors such as digging and self-grooming, and deficits in social behaviors (Peça et al., 2011; Yoo et al., 2019). Their repetitive behavior patterns of self-grooming may be related to the level of self -injury. Correlations between neuroanatomical alterations and ASD symptoms, though not strong, have been found. For example, increased striatum size (caudate in particular) was suggested to be related to the repetitive behavior patterns in ASD patients (Marieke et al., 2009). The caudate striatum of the Shank3 mutant mice was also found to have anatomical abnormalities yet is was smaller compared to control group. Moreover, Shank3 mutant mice have morphological differences in their dendritic spines such as significant reduction in the mean thickness and length (Peça et al., 2011). Altogether, Shank3 mouse model for autism is considered valid and widely used to mimic ASD behaviors.

NLGN3/NLGN4 Mutation

Mice models expressing mutations/deletions in NLGN (mainly *NLGN4*) present behavioral changes that resemble the core symptoms of ASD, including impaired social interactions, deficient ultrasound vocalizations, and increased repetitive behaviors (El-Kordi et al., 2013; Stephane et al., 2008). Since the ratio of males to females in ASD is about 4:1 there is a clear sexual predisposition (Loomes, Hull, Polmear, & Mandy, 2017). Thus, not surprisingly, mutations on the X chromosome are highly linked to ASD. Several researchers reported that a mutation in two X-linked genes encoding neuroligins NLGN3 and NLGN4 is highly correlated to ASD (Stéphane et al., 2003). In the X chromosome, at least two loci have been found to be associated with the sexual predisposition to ASD. A locus at Xq13–21 (Shao et al., 2002) and Xp22.3 de novo chromosomal deletions (Thomas et al, 1999). Within the deleted interval on Xp22.3, researchers have identified KIAA1260, a transcript which corresponds to NLGN4, a member of the neuroligin family (Stéphane et al., 2003).

Neuroligin (NLGN), is a cell adhesion protein on the postsynaptic membrane of neurons. This protein plays an important role in mediating the formation and maintenance of the synapse (Fabrichny et al, 2007; Scheiffele et al, 2000). In humans, familial deletion within *NLGN4* was found to be associated with autism and Tourette syndrome (Lawson-Yuen, Juan-Sebastian, Steve, & Picker, 2008). Neurophysiologically, researchers have shown that loss of Neuroligin-4 causes a profound impairment of glycinergic synaptic transmission and a decrease in glycinergic synapse numbers (Zhang, Gokce, Hale, Brose, & Südhof, 2018). Other studies claimed that some of the behavioral alterations of this mice model can be explained by increased inhibitory synaptic transmission, yet with no noticeable effect on excitatory synapses (Tabuchi et al., 2007).

Altogether, mutations in neuroligin are highly associated with ASD in humans and autism-like behaviors in mice models. These models are based on the synaptic dysfunctions led by mutations in a protein which is responsible for the synaptic stability and plasticity.

Social Interaction Tests for Rodents

In order to perform valid pre-clinical trials in animal models, there is a need to use standard and well-established behavioral experiments that can differentiate between normal and autistic-like phenotypes. The different tests aim to exhibit differences in the behaviors of social interaction and restricted, repeated behaviors. Since mice have a high sociability baseline, several social interaction tests have been established to quantify the normal sociability score. This score can then be compared to an ASD mice model which is significantly lower.

Dyadic Reciprocal Social Interaction Test

In this test, two young-adult male mice are placed in a cage for 10–30 minutes and are filmed. One of them is an ASD-like mouse while the other is a healthy control mouse. They are matched for age (4–5 weeks old) before sexual adulthood. These mice have never met before and were in social deprivation (i.e. alone in a cage) for 2 hours prior to their meeting. During the test, the behavior of the ASD-like mouse is constantly measured and classified according to several categories. The first category is social interaction, and it contains (a) the time the ASD-like mouse spent in nose-to nose interaction with the healthy mouse, (b) the time it spent in nose-to genitals interaction with the healthy mouse and (c) the time it spent fighting with the healthy mouse (for mice, fighting is considered to be a normal strategy for establishing social ranking). The second category is the antisocial interaction and it contains the amount of time that the ASD-like mouse actively avoided interactions initiated by the healthy mouse. The third category refers to the time the ASD-like mouse spent in stereotypical movements. While humans can display a variety of stereotypical movements such as hand waves, head nodding, clapping etc., mice have limited repertoire of stereotypical movements. The operationalization of stereotypical movements of ASD-like mice is defined by the increased rate of self-grooming and digging compared to healthy mice (Perets, Hertz, London, & Offen, 2018; Perets, Oron, Eliot, & Offen, 2020; Segal-Gavish et al., 2016). Thus, this category contains the time mice spend in self-grooming and the time spend in digging. The advantage of this test is that it involves minimal interference to the mice's natural behavior, and it is easily performed and reliable. The critics of this test mainly focus on the interpretation of the results. Some suggest that the social behaviors measured in this test are influenced by "resident-intruder" phenomenon that is an inherent quality of

rodents. That is, a healthy mouse will be interested in a new mouse in its territory because it is an intruder and not because the resident mouse is sociable. Yet, since there is a significant delta between the scores of healthy mice and ASD-like mice in this test, it is considered relevant and captures real behavioral differences between the models.

Three Chambers Social Interaction Test

This test measures social interactions and novelty. The test is performed in two stages: The first stage examines whether the tested mouse would rather spend time in the presence of another mouse or randomly divide its time between another mouse and a novel object. For this test, one healthy mouse is placed inside a round chamber with thin bars so it can't get out but can be clearly seen, and in the other chamber there is a visible object. The tested mouse can wander freely between the chambers while filmed and the time it spends closely to each of the chambers is measured. Healthy mice will tend to spend significantly more time next to the chamber containing the mouse while ASD-like mice will tend to randomly wonder between the chambers, though in some ASD-like phenotype, the mice also spend more time in the presence of the other mouse. In the next stage, the object in the second chamber is replaced by a new mouse. A healthy mouse would rather spend time next to the chamber of the new mouse instead of the familiar one, while the ASD-like mouse would usually randomly wonder between the chambers. The ASD-like models that spend more time next to the chamber with the mouse in the first stage should now spend equal amount of time between the chambers. The advantage of this test is that it is highly controlled, and the social interaction can easily be measured. Stereotypical movements of self-grooming can also be measured during this test. There are no influences of resident/intruder phenomenon and the interaction is completely initiated by the tested mouse. The drawback of this test is that is barely mimics natural rodent behavior since it is highly controlled (Moy et al., 2007).

Male to Female Ultrasonic Communication

This test is based on the natural behavior of male to female courtship vocal communications. When an adult male mouse meets a sexually mature female, he emits ultrasonic vocalizations (30-100 kHz) (Holy & Guo, 2005). These vocalizations can be reordered via an ultrasonic microphone. The duration of this test can vary from 5 minutes to several days of interactions between the male and the female. Several features can be measured and analyzed, such as the number and duration of the syllables. Advanced algorithms of deep learning allow detailed classification of each syllable and language-like statistical analysis to examine context specific messages that may be conveyed during this interaction (Miller et al., 2008). Interestingly, it

has been reported that some ASD-like mice models present deficiencies in ultrasonic communication (Scattoni et al., 2008; Woehr, 2014). Therefore, this form of communication can be used as a behavioral test of ASD-like phenotype, based on the core symptoms that ASD patients suffer from such as deficits in verbal communication. The advantage of this test is that it is very easy to perform, and it examines natural behavior that is highly relevant to ASD. The analysis can reveal both low- and high-resolution differences between normal and ASD-like mice using automatic and unbiased technology. Since it has been reported that the females do not make the same ultrasonic vocalizations during courtship, the main disadvantage of this test is that it measures the male's response to females only, and cannot measure the vocal response of the female to the male. Several researchers have reported that some of the ultrasonic vocalizations may belong to the females rather to the males, and they therefore suggest performing this test with mute females. Yet, most of the literature agrees that the vast majority of syllables is recorded from the males only (D'Amato & Moles, 2001; Moles, Costantini, Garbugino, Zanettini, & D'Amato, 2007) This was firmly confirmed by using female's urine to evoke males' vocalization while males' urine did not seem to make females vocalize similarly (Whitney & Nyby, 1979).

Stereotypical Movements Tests for Rodents

Self-Grooming Independent of Social Context

Self-grooming is a typical behavior of mice. Yet, compared to normal mice, ASD-like mice model present a significantly higher amount of time invested in this behavior. In this test a single mouse is placed in a clean cage, wood chips-free, for 20 minutes and its behavior is recorded. The amount of time spent on self-grooming is measured and compared to healthy control mice.

Digging Independent of Social Context

Digging is also a typical behavior of mice, yet compared to normal mice, ASD-like mice model presents a significantly higher amount of time invested on this behavior. In this test, a single mouse is placed in a cage with wood chips for 20 minutes and its behavior is recorded. The amount of time spent digging is measured and compared to healthy control mice. Both of these tests are easily performed and analyzed, yet it is important to mention that isolated mice tend to be at high levels of stress, which naturally influences these behaviors.

Another aspect of the repetitive-behaviors phenotype in ASD-like mice is the obsessive interest in objects compared to healthy mice. This can be reflected in their behavior in the following tests:

Marble Burying Test

Mice tend to bury food and objects. Yet ASD-like mice models tend to do this significantly more. Thus, this behavior can be used to differentiate between ASD-like mice and healthy mice. In this test, several marbles are equally distributed in the mouse's home cage and several hours afterwards, the number of buried marbles is counted. This test can quantify the repetitive/obsessive behaviors of ASD-like mice compared to healthy mice (Malkova, Collin, Hsiao, Moore, & Patterson, 2012).

Wheel Jam Test

This test uses the natural attraction of mice to run on a wheel. Both healthy and ASD- like mice display a fondness to run on the wheel when given free access to it. After the mice have gotten accustomed to the wheel in their cage, the wheel is jammed and cannot spin. Healthy mice tend to try and move the wheel for a while and then they neglect it, while ASD mice will spend more time trying to move it. The drawback of this test is mainly the interpretation of its results and the analysis, since it is hard to accurately measure how much time each mouse really spent trying to move the wheel (Zilkha, Kuperman, & Kimchi, 2017).

Morris Water Maze/Water T-Maze

In both of these tests, each mouse learns to swim towards a platform. After the mouse learns where the location of the platform is and uses the shortest trajectory towards it, the location of the platform is changed and the time it takes the mouse to reach the new location is measured. After several trials healthy mice will present learning abilities and will again swim to the platform in the shortest trajectory, while ASD-like mice will take much longer to adapt. In the Morris water maze, the shape of the pool is round while in water T-maze the pool is in a T shape. Both tests are common for cognitive rigidity examination. A drawback of them is that they both involve swimming, thus may involve high levels of stress to the mice (Tsai et al., 2012).

Anxiety Depression and ADHD Tests for Rodents

Beside social interactions, stereotypical movements and repetitive behaviors, there are several other ASD related phenotypes that can be measured in mice. These include, for example, levels of anxiety, depression and hyperactivity. Here are common examples for such tests:

Open Field Test

Being one of the most common tests for anxiety and hyperactivity, this exam relies on the natural tendency of mice to explore new territories. In this test, a single mouse is placed in an empty box (~80x80cm) and the time it spends close to the edges of the box is compared to the time it spends in the center. Since spending time in the center of the box may expose the mouse to dangers, high anxiety levels will be characterized in avoiding it and these mice will spend significantly longer time along the edges of the box (Moy et al., 2007). ASD patients tend to have high levels of anxiety, a phenotype which also characterize ASD-like mice. They, therefore, tend to spend most of their time at the edges of the box compared to healthy mice that tend to be more in the center. A drawback of this test is that there is no full consensus regarding its duration which can vary from 20 minutes to a few hours. This variability is mainly because all mice, healthy and ASD-like, require an acclimation time to the box, therefore they sometimes show high levels of anxiety at the beginning of the test.

Elevated plus Maze Test

This test measures the anxiety levels of mice in a more controlled environment. In this test the mouse is placed in an elevated "plus" maze, the outer sides of its arms are covered and therefore it provides a feeling of safety, while the middle of the "plus" is exposed. The amount of time that the mouse moves in the exposed area is measured and indicates low anxiety levels. In this test, ASD-like mice are expected to spend more time in the covered places rather than the exposed areas in comparison to healthy mice (Han, Tai, Jones, Scheuer, & Catterall, 2014). This test is highly controlled and easier to interpret compared to the open field text.

These are some examples of behavioral tests that can differentiate between healthy and ASD-like behaviors. The literature is abundant with more behavioral examinations for these symptoms. Each has its advantages and drawbacks therefore it is highly recommended to perform several tests for the same phenotype.

Since ASD is a complicated disorder with a variety of causes that may lead to behavioral and cognitive alterations, it is not possible nor effective to establish only

one animal model that will aim to mimic all the causes for ASD. Rather, several different models have been established and aimed to imitate the main causes that may lead to ASD in humans.

ASD-Like Mice Model Based on Induced Inflammation during Pregnancy

Inducing by Poly (I:C)

Recent studies have spotlighted the often used to simulate viral infections (Fortier et al., 2004). This substance is often used to evoke the immune system and to increase its activity. Using the recent insights regarding the involvement of maternal over-activation of the immune system during pregnancy and increasing risks of ASD, mouse and rats models were established (Malkova et al., 2012; Patterson, 2011). These models aim to mimic the ASD cases caused by viral infection during pregnancy.

It has been reported that mice offspring of viral-induced pregnancy, mainly during the first trimester, had all core symptoms of autism-like phenotypes including repetitive behaviors and stereotypical movements, decreased social interaction and pup ultrasonic vocalizations (Malkova et al., 2012). Adult offspring of Poly I:C treated mothers usually display increased levels of GABAA receptor 2 immunoreactivity (Nyffeler, Meyer, Yee, Feldon, & Knuesel, 2006) as well as increased dopamine levels (Ozawa et al., 2006). Taken together, immune activation during pregnancy may lead to alterations and dysregulations in the inhibitory-excitatory (IE) balance in the brains of the offsprings (Meyer, Yee, & Feldon, 2007). Since IE imbalance in the brain was found highly relevant to ASD in humans (Fatemi et al., 2012), this model of immune activation during pregnancy is considered to be valid in mimicking both the core symptoms of ASD in humans as well as the neuropathologies.

ASD-Like Mice Model Based on Multifactorial Factors

Despite the accumulating knowledge regarding the genetic and environmental causes that may lead to ASD in humans, and the numerous mice and rats models that have developed to mimic those causes, most of ASD causes in humans remain unknown and are characterized as multifactorial causes for ASD.

To investigate the cases of ASD phenotypes that may be caused by multifactorial causes, a different approach has been taken. By comparing the baseline behaviors of several mice models, researchers have found one strain of mice with natural autistic-like behaviors (Moy et al., 2007). BTBR T + tf/J (BTBR) are inbred mice that were

found to have all the core symptoms of ASD, including repetitive behaviors of increased self-grooming and digging, reduction in ultrasonic communication and social interaction and also cognitive rigidity and learning disorders (McFarlane et al., 2008).

Interestingly, when trying to investigate the causes for the autistic-like phenotypes of the BTBR mice, researchers found correlations to post-mortem ASD human brains, thus this model is based on an "reverse engineering" approach. For example, BTBR mice present innate IE imbalance in the brain, a finding that is correlated with findings from post mortem ASD patients (Han et al., 2014). A genetic basis was also found to be involved in the autistic-like phenotype of BTBR mice. For example, it was found that BTBR mice present altered DNA methylation and cerebellar oxidative DNA damage which was found also in post mortem ASD human brains (Shpyleva et al., 2014). BTBR mice can be used for mimicking the ASD cases that are based on multifactorial reasons rather than a known point mutation or viral infection. Therefore, they can be used to test innovative treatments such as stem cells therapies, that are not aimed to target specific pathway but rather to ameliorate the core symptoms of ASD using regeneration and neurogenesis in the brain (Perets et al., 2017; Perets et al., 2018; Segal-Gavish et al., 2016). Altogether, BTBR mice are helpful in uncovering some of the mechanism of ASD in humans that cannot be attributed to genetic mutations or a viral infection during pregnancy. This mouse model can be coupled to other genetically modified mice to make findings regarding ASD in humans more general to the ASD spectrum.

Summary

ASD is a complex disorder that cannot be presented by one mice or rat model. Therefore, in order to cover most of the pathological causes leading to ASD there is a need to use several animal models. Interestingly, although the models are based on different biological mechanisms, all of them present the core symptoms of ASD including the increased repetitive behaviors, reduction in social interaction and ultrasonic communication.

References

Bakker, C. E., Verheij, C., Willemsen, R., van der Helm, R., Oerlemans, F., Vermey, M., et al. (1994). Fmr1 knockout mice: A model to study fragile X mental retardation. *Cell, 78*(1), 23–33.

D'Amato, F. R., & Moles, A. (2001). Ultrasonic vocalizations as an index of social memory in female mice. *Behavioral Neuroscience, 115*(4), 834.

Durand, C. M., Betancur, C., Boeckers, T. M., Bockmann, J., Chaste, P., Fauchereau, F., Nygren, G., Rastam, M., Gillberg, I. C., Anckarsäter, H., Sponheim, E., Goubran-Botros, H., Delorme,

R., Chabane, N., Mouren-Simeoni, M. C., de Mas, P., Bieth, E., Rogé, B., Héron, D., Burglen, L., et al. (2007). Mutations in the gene encoding the synaptic scaffolding protein SHANK3 are associated with autism spectrum disorders. *Nature Genetics, 39*(1), 25–27.

El-Kordi, A., Winkler, D., Hammerschmidt, K., Kästner, A., Krueger, D., Ronnenberg, A., Ritter, C., Jatho, J., Radyushkin, K., Bourgeron, T., Fischer, J., Brose, N., & Ehrenreich, H. (2013). Development of an autism severity score for mice using Nlgn4 null mutants as a construct-valid model of heritable monogenic autism. *Behavioural Brain Research, 251*, 41–49.

Fabrichny, I. P., Leone, P., Sulzenbacher, G., Comoletti, D., Miller, M. T., Taylor, P., Bourne, Y., & Marchot, P. (2007). Structural analysis of the synaptic protein neuroligin and its β-neurexin complex: Determinants for folding and cell adhesion. *Neuron, 56*(6), 979–991. https://doi.org/10.1016/j.neuron.2007.11.013.

Fatemi, S. H., Aldinger, K. A., Ashwood, P., Bauman, M. L., Blaha, C. D., Blatt, G. J., Chauhan, A., Chauhan, V., Dager, S. R., Dickson, P. E., Estes, A. M., Goldowitz, D., Heck, D. H., Kemper, T. L., King, B. H., Martin, L. A., Millen, K. J., Mittleman, G., Mosconi, M. W., Persico, A. M., et al. (2012). Consensus paper: Pathological role of the cerebellum in autism. *Cerebellum (London, England), 11*(3), 777–807.

Han, S., Tai, C., Jones, C. J., Scheuer, T., & Catterall, W. A. (2014). Enhancement of inhibitory neurotransmission by GABAA receptors having α2, 3-subunits ameliorates behavioral deficits in a mouse model of autism. *Neuron, 81*(6), 1282–1289.

Harony-Nicolas, H., Kay, M., Hoffmann, K., Klein, M., Bozdagi-Gunal, K., et al. (2017). Oxytocin improves behavioral and electrophysiological deficits in a novel Shank3-deficient rat. *eLife, 6*(1–23).

Holy, T. E., & Guo, Z. (2005). Ultrasonic songs of male mice. *PLoS Biology, 3*(12), e386.

Lawson-Yuen, A., Juan-Sebastian, S., Steve, S., & Picker, J. (2008). Familial deletion within NLGN4 associated with Autism and Tourette syndrome. *European Journal of Human Genetics: EJHG, 16*(5), 614–618.

Loomes, R., Hull, L., Polmear, W., & Mandy, L. (2017). What is the male-to-female ratio in Autism Spectrum disorder? A systematic review and meta-analysis. *Journal of the American Academy of Child and Adolescent Psychiatry, 56*(6), 466–474.

Malkova, N. V., Collin, Z. Y., Hsiao, E. Y., Moore, M. J., & Patterson, P. H. (2012). Maternal immune activation yields offspring displaying mouse versions of the three core symptoms of autism. *Brain, Behavior, and Immunity, 26*(4), 607–616.

Marieke, L., Schnack, H., Nederveen, H., Bos, D., Lahuis, B., et al. (2009). Changes in the developmental trajectories of striatum in autism. *Biological Psychiatry, 66*(4), 327–333.

McFarlane, H. G., Kusek, G. K., Yang, M., Phoenix, J. L., Bolivar, V. J., & Crawley, J. N. (2008). Autism-like behavioral phenotypes in BTBR T+tf/J mice. *Genes, Brain, and Behavior, 7*(2), 152–163.

Meyer, U., Yee, B. K., & Feldon, J. (2007). The neurodevelopmental impact of prenatal infections at different times of pregnancy: The earlier the worse? *The Neuroscientist, 13*(3), 241–256. https://doi.org/10.1177/1073858406296401.

Miller, J. E., Spiteri, E., Condro, M. C., Dosumu-Johnson, R. T., Geschwind, D. H., & White, S. A. (2008). Birdsong decreases protein levels of FoxP2, a molecule required for human speech. *Journal of Neurophysiology, 100*(4), 2015–2025.

Moles, A., Costantini, F., Garbugino, L., Zanettini, C., & D'Amato, F. R. (2007). Ultrasonic vocalizations emitted during dyadic interactions in female mice: A possible index of sociability? *Behavioural Brain Research, 182*(2), 223–230.

Moy, S. S., Nadler, J. J., Young, N. B., Perez, A., Holloway, L. P., et al. (2007). Mouse behavioral tasks relevant to autism: Phenotypes of 10 inbred strains. *Behavioural Brain Research, 176*(1), 4–20.

Nyffeler, M., Meyer, U., Yee, B. K., Feldon, J., & Knuesel, I. (2006). Maternal immune activation during pregnancy increases limbic GABAA receptor immunoreactivity in the adult offspring: Implications for schizophrenia. *Neuroscience, 143*(1), 51–62.

Ozawa, K., Hashimoto, K., Kishimoto, T., Shimizu, E., Ishikura, H., & Iyo, M. (2006). Immune activation during pregnancy in mice leads to dopaminergic hyperfunction and cognitive impairment in the offspring: A neurodevelopmental animal model of schizophrenia. *Biological Psychiatry, 59*(6), 546–554. https://doi-org.rproxy.tau.ac.il/10.1016/j.biopsych.2005.07.031.

Patterson, P. H. (2009). Immune involvement in schizophrenia and autism: Etiology, pathology and animal models. *Behavioural Brain Research, 204*(2), 313–321.

Patterson, P. H. (2011). Maternal infection and immune involvement in autism. *Trends in Molecular Medicine, 17*(7), 389–394.

Peça, J., Feliciano, C., Ting, J. T., Wang, W., Wells, M. F., & Feng, G. (2011). Shank3 mutant mice display autistic-like behaviours and striatal dysfunction. *Nature, 472*(7344), 437–442.

Perets, N., Hertz, S., London, M., & Offen, D. (2018). Intranasal administration of exosomes derived from mesenchymal stem cells ameliorates autistic-like behaviors of BTBR mice. *Molecular Autism, 9*(1), 57.

Perets, N., Oron, O., Eliot, E., & Offen, D. (2020). Exosomes derived from mesenchymal stem cells improved core symptoms of genetically modified mice model of autism Shank3B. *Molecular Autism*. https://doi.org/10.21203/rs.3.rs-17171/v2.

Perets, N., Segal-Gavish, H., Gothelf, Y., Barzilay, R., Barhum, Y., et al. (2017). Long term beneficial effect of neurotrophic factors-secreting mesenchymal stem cells transplantation in the BTBR mouse model of autism. *Behavioural Brain Research, 331*, 254–260.

Scattoni, M. L., Shruti, U., Ricceri, L., & Crawley, J. N. (2008). Unusual repertoire of vocalizations in the BTBR T+tf/J mouse model of autism. *PLoS One, 3*(8), e3067.

Segal-Gavish, H., Golan, K., Barak, N., Barzilay, R., Ganz, J., et al. (2016). Mesenchymal stem cell transplantation promotes neurogenesis and ameliorates autism related behaviors in BTBR mice. *Autism Research: Official Journal of the International Society for Autism Research, 9*(1), 17–32.

Shao, Y. M., Wolpert, C. M., Raiford, K. L., Menold, M. M., Donnelly, S. L., et al. (2002). Genomic screen and follow-up analysis for autistic disorder. *American Journal of Medical Genetics, 114*(1), 99–105.

Shpyleva, S., Ivanovsky, S., de Conti, A., Melnyk, S., Tryndyak, V., et al. (2014). Cerebellar oxidative DNA damage and altered DNA methylation in the BTBR T+ tf/J mouse model of autism and similarities with human post mortem cerebellum. *PLoS One, 9*(11), e113712.

Stéphane, J., Quach, H., Betancur, C., Råstam, M., Colineaux, C., et al. (2003). Mutations of the X-linked genes encoding Neuroligins NLGN3 and NLGN4 are associated with autism. *Nature Genetics, 34*(1), 27–29.

Stephane, J., Radyushkin, K., Hammerschmidt, K., Granon, S., Boretius, S., et al. (2008). Reduced social interaction and ultrasonic communication in a mouse model of monogenic heritable autism. *Proceedings of the National Academy of Sciences of the United States of America, 105*(5), 1710–1715.

Tabuchi, K., Blundell, J., Etherton, M. R., Hammer, R. E., Liu, X., et al. (2007). A neuroligin-3 mutation implicated in autism increases inhibitory synaptic transmission in mice. *Science, 318*(5847), 71–76.

Tsai, P. T., Hull, C., Chu, Y., Greene-Colozzi, E., Sadowski, A. R., et al. (2012). Autistic-like behaviour and cerebellar dysfunction in Purkinje cell Tsc1 mutant mice. *Nature, 488*(7413), 647–651.

Whitney, G., & Nyby, J. (1979). Cues that elicit ultrasounds from adult male mice. *American Zoologist, 19*(2), 457–463.

Woehr, M. (2014). Ultrasonic vocalizations in Shank mouse models for autism spectrum disorders: Detailed spectrographic analyses and developmental profiles. *Neuroscience and Biobehavioral Reviews, 43*, 199–212.

Yoo, Y., Yoo, T., Lee, S., Lee, J., Kim, D., et al. (2019). Shank3 mice carrying the human q321r mutation display enhanced self-grooming, abnormal electroencephalogram patterns, and suppressed neuronal excitability and seizure susceptibility. *Frontiers in Molecular Neuroscience, 12*, 1–23.

Zerbo, O., Iosif, A., Walker, C., Ozonoff, S., Hansen, R. L., & Hertz-Picciotto, I. (2013). Is maternal influenza or fever during pregnancy associated with autism or developmental delays? Results from the CHARGE (childhood autism risks from genetics and environment) study. *Journal of Autism and Developmental Disorders, 43*(1), 25–33.

Zhang, B., Gokce, O. W., Hale, D., Brose, N., & Südhof, T. C. (2018). Autism-associated Neuroligin-4 mutation selectively impairs Glycinergic synaptic transmission in mouse brainstem synapses. *The Journal of Experimental Medicine, 215*(6), 1543–1553.

Zilkha, N., Kuperman, Y., & Kimchi, T. (2017). High-fat diet exacerbates cognitive rigidity and social deficiency in the BTBR mouse model of autism. *Neuroscience, 345*, 142–154.

Chapter 5
Underlying Mechanisms of Restricted and Repetitive Behaviors Across Typical and Atypical Development

Mirko Uljarević, Darren Hedley, Omer Linkovski, and Susan R. Leekam

Introduction

Restricted and repetitive behaviors and interests (RRBI) have been considered as a core symptom of autism spectrum disorder (ASD) since the first clinical descriptions provided by Leo Kanner (1943). In the original sample of children with ASD, Kanner observed a range of RRBI including various motor mannerisms such as shaking of the head from side to side or jumping up and down repeatedly, use of objects in inflexible and highly repetitive manner (e.g., spinning round objects), unusual preoccupation and fascination with ordinary objects such as cardboard boxes and pencils, highly intense interests and very strong insistence that things need to be 'just so' (e.g. insistence that parts of the furniture and other objects need to be arranged in a certain way and becomming distressed if any change was made). Kanner considered RRBI to be an essential feature of ASD and this view has been supported through all the incarnations of international diagnostic systems. More

M. Uljarević (✉)
Melbourne School of Psychological Sciences, University of Melbourne, Melbourne, VIC, Australia
e-mail: mirko.uljarevic@unimelb.edu.au

D. Hedley
Olga Tennison Autism Research Centre, School of Psychology and Public Health, La Trobe University, Melbourne, VIC, Australia

O. Linkovski
Department of Psychiatry and Behavioral Sciences, School of Medicine, Stanford University, Stanford, CA, USA

Department of Medical Neurobiology, The Hebrew University-Hadassah Medical School, Jerusalem, Israel

S. R. Leekam
Wales Autism Research Centre, School of Psychology, Cardiff University, Wales, Cardiff, UK

E. Gal, N. Yirmiya (eds.), *Repetitive and Restricted Behaviors and Interests in Autism Spectrum Disorders*, Autism and Child Psychopathology Series,
https://doi.org/10.1007/978-3-030-66445-9_5

than 70 years since this pioneering work, it is now well established that RRBI are one of the earliest predictors of subsequent ASD diagnosis (Ozonoff et al., 2008; Wolff et al., 2014). RRBI, and specifically repetitive and restricted behaviors (RRB) have a significant impact on all aspects of functioning and long-term outcomes for affected individuals and can be a major source of stress and management challenge for caregivers and family members (Grahame et al., 2015; Harrop, McBee, & Boyd, 2016; Leekam, Prior, & Uljarević, 2011; South, Ozonoff, & McMahon, 2005). However, despite their prominence in terms of diagnosis and impact on the individuals and their families, we still lack a comprehensive understanding of the mechanisms underpinning the development and maintenance of RRB which precludes the development of individually tailored treatment options.

In this chapter we will argue, in line with the current dimensional models of psychopathology such as, for example, the Research Domain Criteria put forward by the National Institute of Mental Health (Insel, 2010), that one of the major obstacles to a more in-depth understanding of RRB in ASD is the fact that research thus far has been largely disorder-centric, both in terms of explanatory frameworks and measurement. Given that RRB are not specific to ASD, these behaviors offer a particularly good candidate for the dimensional approach. Indeed, a wide range of RRB are seen throughout normative development (Arnott et al., 2010; Evans et al., 1997; Leekam et al., 2007; Thelen, 1979, 1981; Uljarević et al., 2017) and are common in individuals with a range of neurodevelopmental and genetic disorders, individuals diagnosed with schizophrenia, Tourette's syndrome, Obsessive Compulsive disorder, Alzheimer's and other forms of dementias, as well as in individuals with sensory impairments such as loss of hearing and vision (Bodfish, Symons, Parker, & Lewis, 2000; Evans, Uljarević, Lusk, Loth, & Frazier, 2017). Importantly, while it has been suggested that during typical development different RRB domains serve a range of adaptive functions such as neuromuscular and general central nervous system development (Sprague & Newell, 1996; Thelen, 1979; Wolff, 1968) and self-regulation/fear reduction (Evans, Gray, & Leckman, 1999; Leekam et al., 2011; Uljarevic & Evans, 2017; Uljarević, Hedley, Alvares, Varcin, & Whitehouse, 2017), as noted, some forms of RRB can be maladaptive and persistent in ASD and other clinical disorders. Thus, we will argue that a developmental and transdiagnostic approach to the identification of mechanisms involved in the emergence, increase and eventual decrease of RRB during normative development can inform our understanding of RRB in clinical conditions, particularly in ASD.

In this chapter we will first provide a brief overview of the current understanding of the conceptualization and classification of RRB across normative and atypical development. We will then chart the developmental trajectory of these behaviors in typically developing children and contrast this trajectory with the age-related patterns shown in ASD. This summary will be followed by an analysis of cognitive and affective processes whose trajectory is concomitant to the rise and fall of RRB seen in typically developing children. We will summarize current ASD literature suggesting that these mechanisms might serve a crucial role in the development and maintenance of specific RRB subtypes. We conclude the chapter by suggesting necessary steps that future research will need to take in order to enable better understanding and design of effective treatment options for these clinically impactful behaviors.

Conceptualization and Classification of RRB

RRBI is an umbrella term for a heterogeneous set of behaviors ranging from simple motor stereotypies and sensory-related behaviors, to compulsions, routines, rituals and intense preoccupations with particular interests. Considering this complexity and diversity, it is clear that RRBI are not a unitary construct but rather encompass several distinct domains. Given that different domains are likely to be subserved by at least partially distinct mechanisms, a considerable effort has been put into addressing the question of how best to classify this behavioral domain.

Based on clinical observations and a developmental approach, Prior and Macmillan (1973) and later Turner (1999) argued that RRBI can be classified into lower level behaviors such as dyskinesias, tics, stereotyped movements, repetitive manipulation of objects and self-injurious behaviors thought to be more characteristic for younger and lower functioning children (also found in children with intellectual disabilities and brain-based impairments), and higher level repetitive behaviors such as object attachments, repetitive language, insistence on sameness and circumscribed interests thought to be present in older and more able children.

Over the last two decades, a number of factor analytic studies have been conducted in an attempt to clarify the structure of RRBI. Studies conducted in ASD populations using both clinical interview methods such as the Autism Diagnostic Interview-R (ADI-R; Bishop et al., 2013; Richler, Huerta, Bishop, & Lord, 2010) and questionnaire measures including the Repetitive Behavior Questionnaire-2 and the Repetitive Behaviors Scale-Revised (Barrett, Uljarević, Jones, & Leekam, 2018; Georgiades, Papageorgiou, & Anagnostou, 2010; Honey, McConachie, Turner, & Rodgers, 2012; Lidstone et al., 2014) have most consistently identified the following two factors: (i) Repetitive (Sensory) Motor Behaviors (RMB) encompassing hand and finger mannerisms, repetitive use of objects or parts of objects, stereotyped body movements (e.g., rocking) and in some instances sensory-related behaviors, and (ii) Insistence on Sameness (IS) including difficulties with and resistance to minor changes in routine or personal environment, compulsions and rituals, and occasionally restricted, inflexible and all-encompassing focus on specific topics or objects. A very similar two-factor structure has also been replicated across normative development (Barrett et al., 2015; Evans et al., 2017; Leekam et al., 2007; Uljarević, Arnott, et al., 2017) and a range of neuropsychiatric and neurodevelopmental disorders including ASD, obsessive-compulsive disorders (OCD), Tourette syndrome, anxiety disorders, and schizophrenia (Evans et al., 2017).

A number of studies addressing RRB have established a distinct pattern of associations among RMB and IS domains with chronological age (CA) and cognitive ability, as well as preliminary evidence for distinct neurobiological underpinnings, as a further step in establishing the validity of this two-type typology. Although there is currently a lack of long-term longitudinal studies in ASD, based on the findings from short-term longitudinal and cross-sectional studies it is possible to extrapolate that the RMB domain is more prevalent and intense during the early years, with subsequent waning throughout childhood (Esbensen, Seltzer, Lam, & Bodfish, 2009; Harrop et al., 2014; Murphy et al., 2005; Richler et al., 2010). The

IS domain, rare at age two, increases gradually throughout early childhood, staying relatively stable throughout the subsequent development (Esbensen et al., 2009; Murphy et al., 2005; Richler et al., 2010; South et al., 2005). It is important to note that intellectual quotient (IQ) level can moderate the developmental trajectory of RMB and IS in distinctive ways. Richler et al. (2010) who followed children's RRB across four age periods (ages 2, 3, 5, and 9) reported the developmental pattern matching the one described above; however, while having a higher nonverbal IQ at age two was associated with milder concurrent RMB behaviors and reduction in these behaviors across time, IQ had no such effects on the trajectory of IS.

It is helpful to consider the changes seen in children with ASD against those seen in typical development. Cross-sectional studies in normative development have suggested that RMB are very common in the first 12–15 months of development, followed by the relatively sharp decline. For example, Leekam et al. (2007) used the Repetitive Behavior Questionnare-2 (RBQ-2) to explore repetitive sensory and motor behaviors in 675 two-year old TD children and found that, at that age, these behaviors were common, with every item endorsed for 18 to 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). On the other hand, when behaviors such as kicking and banging begin to decrease, it becomes apparent that rigid types of behaviors such as a need for sameness start to increase. Gessell and colleagues (1974) and Gesell (1928) observed that between 2 and 3 years of age, typically developing children show compulsive behaviors including preference for sameness, repetitive and ritualised behaviors, rigidity in likes and dislikes and acute sensory perception for minute details. Evans et al. (1997) found that insistence on sameness behaviors were more common and intense in children aged 24 to 48 months, but also that their endorsement was less common in children aged 48 to 72 months. These cross-sectional findings were extended in a longitudinal study by our group (Uljarević, Arnott, et al., 2017) which collected RBQ-2 data from a sample of 208 typically developing (TD) children when they were 15, 24 and 77 months old. We first explored the trajectory of IS and RMB domains. RMB scores at 15 months were higher than at both 24 and 77 months; RMB score at 24 months was higher than at 77 months. IS scores at 24 months were higher than at both 15 and 77 months; IS at 15 and 77 months did not differ significantly. Hierarchical regression models further demonstrated that RMB and IS developed independently. More specifically, RMB domain at 77 months was predicted only by RMB but not IS at previous time points. Similarly, IS domain at 77 months was predicted by IS but not RMB at both 15 and 24 months.

Neuroimaging studies suggest that RMB can be linked to the dysfunction in motor and premotor cortex, and that IS are related to cognitive/associative loop and anterior cingulate-orbitofrontal cortex loop (for comprehensive overviews please see Langen, Durston, Kas, Van Engeland, & Staal, 2011 and Yerys, 2015). Findings from several familial studies provided evidence for the familiality of IS (Lam, Bodfish, & Piven, 2008; Shao et al., 2003; Silverman et al., 2002; Szatmari

et al., 2006; Uljarević, Evans, Alvares, & Whitehouse, 2016) but not for the RMB subtype. Genetics studies have linked these two subtypes to largely non-overlapping chromosomal regions. For example, Cannon et al. (2010) showed that IS were linked with 2q37.1-q37.3 and RMB with 15q13.1-q14. Shao et al. (2003) have demonstrated that while high IS scores increase the linkage evidence for the 15q11-q13 region at the GABRB3 locus, this was not the case for RMB. Using chromosomal microarray analysis, the potentially important role of the dopamine 3 receptor gene (DRD3), which is highly expressed in the basal ganglia, and specifically in the caudate (Staal, de Krom, & de Jonge, 2012) was identified. More specifically, using the ADI-R to capture RMB and IS domains in ASD individuals aged four to 31 years, Staal et al. (2012) found that those carrying the AA variant of the rs167771 SNP showed more IS behaviors than individuals carrying one or two copies of the minor G-allele, indicating that this polymorphism decreases the risk for IS. No effects were found according to allele variant for the RMB ADI-R factor nor for the social interaction scores.

The reviewed evidence from factor analytic work therefore indicates a stable explanation of the RRB structure across ASD, other clinical populations and normative development (Bishop et al., 2013; Leekam et al., 2007; Lidstone et al., 2014). Findings from factor analyses are further supported by findings from the cross-sectional correlational studies (Bishop et al., 2013; South et al., 2005), longitudinal studies (Richler et al., 2010), and neurobiological findings (Langen et al., 2011), suggesting strong evidence for the distinctiveness of RMB and IS domains. Therefore, in this chapter we adopt the RMB/IS dichotomy.

Integrating Findings across Typical and Atypical Development to Inform the Identification of the Mechanism Involved in the Development and Maintenance of RRB in ASD

Insistence on Sameness

The described pattern of emergence, increase and decrease of IS behaviors over the first 6–7 years of normative development is paralleled by the trajectory of normative fears such as fear of the dark and separation/stranger anxiety which also occurs in the context of typical development and are transitory in nature (Brooker et al., 2014; Evans et al., 1999; Gullone, 2000). Importantly, a range of ritualistic and rigid behavioral patterns are particularly likely to occur at times of transition (such as bedtime), that are accompanied by these normative fears (Evans et al., 1999). The similar developmental course for IS and fear has led to suggestions that IS serve to constrain the unpredictability of the environment and to ward off fear and anxiety, thus serving as an early and rudimentary form of self-regulation (Evans et al., 1999; Gesell et al., 1974; Leekam et al., 2011; Zohar & Felz, 2001). As noted, unlike in normative development, where high levels of IS (and other RRB) are transitionary

and serve adaptive purpose, in ASD these behaviors are persistent and significantly negatively impact all aspects of functioning of affected individuals. However, the potential continuities and discontinuities in mechanisms underpinning typical into atypical RRB have not, until relatively recently, been directly explored.

The normative development period when both IS and normative fears begin to wane (around and after 3–4 years of life), is concomitant to the development of different facets of self-regulation. This includes attentional control, inhibition of dominant and activation of subdominant responses, ability to shift between multiple tasks and mental sets and working memory and the ability to regulate the experience and expression of emotions. These abilities develop gradually, becoming progressively more advanced and complex from early toddlerhood to the school years and beyond (Bridgett, Oddi, Laake, Murdock, & Bachmann, 2013; Zelazo et al., 2003; Zhou, Chen, & Main, 2011) and are supported by the maturation of the orbitofrontal cortex (OFC), the consolidation of pathways between the OFC, the anterior cingulate cortex (ACC), and striatal and limbic regions (in particular the caudate and amygdala, respectively) (see Judge, Evans, Schroepfer, & Gross, 2011 for an overview). Based on these developmental trends, we have suggested (Evans, Lewis, & Iobst, 2004; Uljarevic & Evans, 2017; Uljarević, Hedley, et al., 2017) that as more sophisticated and flexible forms of self-regulation during childhood develop, children rely less on IS behaviors for managing fears. Furthermore, we have hypothesised that IS behaviors, if persistent beyond the developmental period when they are adaptive, may negatively impact subsequent development by limiting children's exposure to situations conducive to developing more elaborate, complex and flexible patterns of self-regulation, as well as other aspects of social, cognitive and emotional development (see also Larkin, Meins, Centifanti, Fernyhough, & Leekam, 2017; Leekam et al., 2011). A range of indirect evidence across normative and atypical development provide support for this model.

Research indicates that individual variation in different aspects of self-regulation abilities during this normative developmental period is associated with IS behaviors (Peleg-Popko & Dar, 2003; Pietrefesa & Evans, 2007; Tregay, Gilmour, & Charman, 2009) and also predicts later internalizing problems (Eisenberg et al., 2009; Hughes & Ensor, 2011; Murray & Kochanska, 2002). As noted, several investigations have found evidence for the link between normative fears and IS in typically developing children. For example, Evans et al. (1999) reported that behaviors including bedtime rituals and hoarding objects were significantly related to overall fears and fear of strangers in a group of 61 children ranging from 1 to 7 years of age.

Anxiety is very prevalent in individuals with ASD, across the life span, with approximately 40% of children and adolescents (Uljarević, Nuske, & Vivanti, 2016; van Steensel, Bogels, & Perrin, 2011) and 53% of adults (Uljarević et al., 2020) with ASD presenting with clinically significant anxiety or anxiety disorders, a rate that is significantly higher than in the general population (Gadow, DeVincent, Pomeroy, & Azizian, 2005). Anxiety is also highly prevalent across other neurodevelopmental and neuropsychiatric disorders that reveal elevated and persistent RRB, such as Down Syndrome (Evans & Gray, 2000; Glenn,

Cunningham, Nananidou, Prasher, & Glenholmes, 2015; Uljarevic & Evans, 2017) and Williams Syndrome (Rodgers, Riby, Janes, Connolly, & McConachie, 2012; Uljarević, Labuschagne, Bobin, Atkinson, & Hocking, 2018) and is an essential component of obsessive-compulsive disorder (OCD). Among people with ASD and other disorders, elevated anxiety levels have been reported to be associated with increased levels of IS behavior (Evans & Gray, 2000; Glenn et al., 2015; Lidstone et al., 2014; Rodgers et al., 2012; Uljarevic & Evans, 2017). For example, in a sample of 120 individuals with ASD (Mean age = 10.7 years, SD = 3.10), Lidstone et al. (2014) found that while higher anxiety was associated with increased severity of IS, it was not significantly associated with RMB. Furthermore, the clinically anxious subgroup exhibited significantly higher rates of IS but not RMB. Similarly, in a sample of 38 children and adolescents with Down Syndrome (Mean age = 10.45 years, SD = 3.81) Uljarević and Evans (2017) found that IS behaviors were associated with more severe fears and anxieties. Delays and impairments across different facets of self-regulation abilities have been consistently found in ASD (Cai, Richdale, Uljarević, Dissanayake, & Samson, 2018; Hill, 2004) and among other neurodevelopmental disorders, including Down Syndrome and OCD (Carney, Brown, & Henry, 2013; Linkovski, Kalanthroff, Henik, & Anholt, 2013, 2016; Memisevic & Sinanovic, 2014). Importantly, these impairments have been found to be associated with both anxiety (Adamek, Nichols, Tetenbaum, Ponzio, & Carr, 2010; De Pauw, Mervielde, Van Leeuwen, & De Clercq, 2011; Hollocks et al., 2014; Wallace et al., 2016) and IS behaviors (see Leekam et al., 2011 for critical overview) in ASD.

Despite the reviewed indirect evidence, only one study to date has attempted to test the model we proposed here directly. In a cross-sectional study from our group (Uljarević, Hedley, et al., 2017) we aimed to characterise the IS-self-regulation-anxiety association by investigating the potential contribution made by self-regulation, assessed via effortful control, to the IS-anxiety relations in a cross-sectional sample of adolescents and young adults with ASD. Our results srevealed that the association between effortful control and anxiety was mediated by IS and that, in turn, the relation between IS behaviors and anxiety was mediated by effortful control. These results provide preliminary, if limited support, for the suggestion that in ASD IS behaviors persist as the primary means of self-regulation and that due to their inflexibility, these behaviors likely reinforce anxiety in the long term. Elevated and persistent IS behaviors can therefore impede the emergence of more developmentally appropriate modes of self-regulation in ASD, either because of the development of positive beliefs about the utility of IS (the mechanisms that would predominantly apply to adolescents and adults with higher IQ) or due to reduced exposure to situations that are conducive to developing more sophisticated self-regulatory strategies. However, elements of this theory warrant stringent future testing using longitudinal designs, and samples spanning normative and atypical development.

Repetitive Motor Behaviors (RMB)

Both observational and questionnaire-based studies suggest that RMB occur early in development and begins to decrease by 2 years of age. For instance, in a large-scale longitudinal observational study, Fyfield (2014), reported an increase in RMB from 6 to 12 months of age and further reported that RMB were associated with more immature locomotor development. This finding is linear with earlier theoretical work by Thelen (1979, 1981) who described RMB in infancy in relation to the development of the motor system. In further analysis of a group of children in their third year, Fyfield also found a decline in the frequency of RMB into toddlerhood, but importantly toddlers who still engaged in RRB in their third year did not have poorer inhibitory control nor higher ratings of Attention Deficit Hyperactivity Disorder (ADHD) symptoms. These findings are the first to support the idea that increased RMB may be linked to maturity of gross motor skills in a community sample and on the other hand to also show that poor self-regulation is not related to RMB.

Findings by Fyfield (2014) on the link between motor development and RMB during the normative development are important to consider in light of the fact that a range of motor atypicalities are frequently observed in ASD. It is estimated that at least 80% of individuals with ASD present with some form of motor impairment (Green et al., 2002; Ming, Brimacombe, & Wagner, 2007; Miyahara et al., 1997; Whyatt & Craig, 2012). Interestingly, although several researchers have demonstrated that motor delays can negatively impact the development and severity of social and communicative symptoms (Gowen & Hamilton, 2013; Leonard & Hill, 2014) and that early motor atypicalities are predictive of a subsequent ASD diagnoses (Brian et al., 2008; Nickel, Thatcher, Keller, Wozniak, & Iverson, 2013), their relation with RRB is yet to be systematically explored. However, several researchers strongly speak to the importance of this link. For example, Bodfish et al. (2000) found that impairments in motor control were associated with increased rates of motor RRB in a sample of individuals with intellectual disability. Furthermore, atypicalities in motor and premotor cortices have been found to be associated with levels of RMB (Estes et al., 2011; Hollander et al., 2005).

Several community sample longitudinal studies by our group highlight language and cognitive ability, the presence of earlier RRB, and parent variables, as further key candidates to focus on when considering mechanisms underlying RMB. For example, Larkin et al., 2017, examined concurrent variables of language and play at 26 months. Results revealed that while children's RMB were related to lower receptive verbal ability and imaginative play at 26 months of age, IS behaviors at this age were unrelated to concurent language and play variables. The previously summarized study by Uljarević, Hedley, et al. (2017), in addition to charting the trajectory of IS and RMB domains across the first 6 years of life, also found that although language may correlate with RRB, it is the RRB itself which is most predictive for later outcome. Importantly, previous research with an ASD sample has also found that gains in language from 2 to 11 years of age were associated with a reduction in repetitive behaviors (Paul, Chawarska, Cicchetti, & Volkmar, 2008; Ray-Subramanian & Ellis Weismer, 2012).

Summary and Future Directions

In this chapter we attempted to provide evidence on how studying development of RRB in samples other tan children with ASD, can provide important clues to uncovering mechanisms underlying the emergence and persistence of this clinically impairing group of symptoms. We consider that findings relating to IS are particularly encouraging suggesting that interplay between delayed self-regulation and anxiety plays a key role in sustaining IS behaviors in individuals with ASD. These results have important clinical implications suggesting that self-regulation is an important intervention target in interventions aimed at decreasing IS. A number of interventions targeting different facets of self-regulation, for example, Tools of Mind (Diamond, Barnett, Thomas, & Munro, 2007), have been shown to increase executive attention in children aged 3–7 years (Rueda, Posner, & Rothbart, 2005). Similar interventions have been shown to be effective in improving executive functioning in children with ADHD (Thorell, Lindqvist, Nutley, Bohlin, & Klingberg, 2009). In addition, Unstuck on Target (Cannon, Kenworthy, Alexander, Werner, & Anthony, 2011), a behavioral approach to enhance developing flexibility and compensatory strategies for impairments in executive functioning, has been shown to increase problem-solving, flexibility, and planning/ organizing aspects of executive functioning based on experimental assessments and parental reports, as well as enabling easier transitions and improving flexibility within classrooms for children with ASD (Kenworthy et al., 2014). Therefore, it will be important in future work to explore the effects of these types of interventions on IS and anxiety.

Findings regarding the RMB domain are less clear and further work is needed. Although the reviewed studies suggest the importance of both motor delays and impairments, as well as language and cognitive delays, unfortunately, to date, RRB studies, in both community and ASD samples have not been large enough nor spanned the period from infancy to late childhood. In addition to focusing on individual factors, it will also be crucial to explore parental and environmental factors as illustrated by Larkin et al. (2017) who reported that maternal depressive symptoms predicted levels of both RSM and IS behaviors. Lower socioeconomic status also predicted independent variance in children's RMB. Further exploration of these relations showed that they were not likely to be attributed to mother's behavior (maternal sensitivity, mind-mindedness, attachment measures). Instead, the results are discussed in terms of stress regulation, self-stimulation, and genetic susceptibility which may link the between maternal depressive symptoms and RRB in the child.

In conclusion, when trying to understand why RRB occur and persist in clinical conditions, it is important to go beyond the simple case-control designs and explore continuities and discontinuities of the mechanisms underlying RRB across normative and atypical development. In order to achieve this, long term trans diagnostic studies are needed, employing comprehensive multimodal measurement instruments that are sensitive to change and to subtle behavioral expressions.

Conflict of Interest Statement Mirko Uljarević is currently supported by the Australian Research Council Discovery Early Career Researcher Award (DE180100632). Darren Hedley is currently supported by a Suicide Prevention Australia National Suicide Prevention Research Fellowship. Omer Linkovski and Susan Leekam declare no conflict of interest.

References

Adamek, L., Nichols, S., Tetenbaum, J. B., Ponzio, C., & Carr, E. G. (2010). Individual temperament and problem behavior in children with autism spectrum disorders. *Focus on Autism and Other Developmental Disabilities, 26*, 173–184.

Arnott, B., McConachie, H., Meins, E., Fernyhough, C., Le Couteur, A., Turner, M., et al. (2010). The frequency of restricted and repetitive behaviors in a community sample of 15-month-old infants. *Journal of Developmental and Behavioral Pediatrics, 31*, 223–229.

Barrett, S. L., Uljarević, M., Baker, E. K., Richdale, A. L., Jones, C. R. G., & Leekam, S. R. (2015). The Adult Repetitive Behaviors Questionnaire-2 (RBQ-2A): A self-report measure of restricted and repetitive behaviors. *Journal of Autism and Developmental Disorders, 45*(11), 3680–3692.

Barrett, S. L., Uljarević, M., Jones, C. R. G., & Leekam, S. R. (2018). Assessing subtypes of restricted and repetitive behavior using the Adult Repetitive Behavior Questionnaire-2 in autistic adults. *Molecular Autism, 9*(1), 58.

Bishop, S. L., Hus, V., Duncan, A., Huerta, M., Gotham, K., Pickles, A., & Lord, C. (2013). Subcategories of restricted and repetitive behaviors in children with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 43*(6), 1287–1297.

Bodfish, J. W., Symons, F. J., Parker, D. E., & Lewis, M. H. (2000). Varieties of repetitive behavior in autism: Comparisons to mental retardation. *Journal of Autism and Developmental Disorders, 30*(3), 237–243.

Brian, J., Bryson, S. E., Garon, N., Roberts, W., Smith, I. M., Szatmari, P., & Zwaigenbaum, L. (2008). Clinical assessment of autism in high-risk 18-month-olds. *Autism, 12*, 433–456.

Bridgett, D. J., Oddi, K. B., Laake, L. M., Murdock, K. W., & Bachmann, M. N. (2013). Integrating and differentiating aspects of self-regulation: Effortful control, executive functioning, and links to negative affectivity. *Emotion, 13*, 47–63.

Brooker, R. J., Buss, K. A., Lemery-Chalfant, K., Aksan, N., Davidson, R. J., & Goldsmith, H. H. (2014). The development of stranger fear in infancy and toddlerhood: Normative development, individual differences, antecedents, and outcomes. *Developmental Science, 16*, 864–878.

Cai, R. Y., Richdale, A., Uljarević, M., Dissanayake, C., & Samson, A. C. (2018). Emotion regulation in autism spectrum disorder: Where we are and where we need to go. *Autism Research, 11*(7), 962–978.

Cannon, D. S., Miller, J. S., Robinson, R. J., Villalobos, M. E., Wahmhoff, N. K., Allen-Brady, K., et al. (2010). Genome-wide linkage analyses of two repetitive behaviors phenotypes in Utah pedigrees with autism spectrum disorders. *Molecular Autism, 1*(3), 1–13.

Cannon, L., Kenworthy, L., Alexander, K. C., Werner, M. A., & Anthony, L. G. (2011). *Unstuck and on target!: An executive function curriculum to improve flexibility for children with autism spectrum disorders*. Baltimore: Paul H. Brookes Pub.

Carney, D. P. J., Brown, J. H., & Henry, L. (2013). Executive function in Williams and Down syndromes. *Research in Developmental Disabilities, 34*, 46–55.

De Pauw, S. S. Q., Mervielde, I., Van Leeuwen, K. G., & De Clercq, B. J. (2011). How temperament and personality contribute to the maladjustment of children with autism. *Journal of Autism and Developmental Disorders, 41*, 196–212.

Diamond, A., Barnett, W. S., Thomas, J., & Munro, S. (2007). Preschool program improves cognitive control. *Science, 318*, 1387–1388.

Eisenberg, N., Valiente, C., Spinrad, T. L., Cumberland, A. J., Liew, J., Reiser, M., & Cumberland, A. J. (2009). Longitudinal relations of children's effortful control, impulsivity, and nega-

tive emotionality to their externalizing, internalizing, and co-occurring behavior problems. *Developmental Psychology, 45*(4), 988–1008.

Esbensen, A. J., Seltzer, M. M., Lam, K. S. L., & Bodfish, J. W. (2009). Age-related differences in restricted repetitive behaviors in autism spectrum disorders. *Journal of Autism and Developmental Disorders, 39*(1), 57–66.

Estes, A., Shaw, D. W., Sparks, B. F., Friedman, S., Giedd, J. N., Dawson, G., et al. (2011). Basal ganglia morphometry and repetitive behavior in young children with autism spectrum disorder. *Autism Research, 4*, 212–220.

Evans, D. W., & Gray, F. L. (2000). Compulsive-like behavior in individuals with down syndrome: Its relation to MA level, adaptive and maladaptive behavior. *Child Development, 71*, 288–300.

Evans, D. W., Gray, F. L., & Leckman, J. F. (1999). The rituals, fears and phobias of young children: Insights from development, psychopathology and neurobiology. *Child Psychiatry and Human Development, 29*, 261–276.

Evans, D. W., Leckman, J. F., Carter, A., Reznick, J. S., Henshaw, D., King, R. A., & Pauls, D. (1997). Ritual, habit, and perfectionism: The prevalence and development of compulsive-like behavior in normal young children. *Child Development, 68*, 58–68.

Evans, D. W., Lewis, M. D., & Iobst, E. (2004). The role of the orbitofrontal cortex in normally developing compulsive-like behaviors and obsessive–compulsive disorder. *Brain and Cognition, 55*(1), 220–234.

Evans, D. W., Uljarević, M., Lusk, L., Loth, E., & Frazier, T. W. (2017). Development of two-dimensional measures of restricted and repetitive behavior in parents and children. *Journal of American Academy of Child and Adolescent Psychiatry, 56*(1), 51–58.

Fyfield, R. (2014). *The rise and fall of repetitive behaviors in a community sample of infants and toddlers.* PhD Thesis, Cardiff University.

Gadow, K. D., DeVincent, C. J., Pomeroy, J., & Azizian, A. (2005). Comparison of DSM-IV symptoms in elementary school-age children with PDD versus clinic and community samples. *Autism, 9*, 392–415.

Georgiades, S., Papageorgiou, V., & Anagnostou, E. (2010). Brief report: Repetitive behaviors in Greek individuals with autism spectrum disorder. *Journal of Autism and Developmental Disorders, 40*(7), 903–906.

Gesell, A. (1928). *Infancy and human growth*. New York: Macmillan.

Gesell, A., Ames, L. B., & Ilg, F. L. (1974). *Infant and the child in culture today*. New York: Harper and Row.

Glenn, S., Cunningham, C., Nananidou, A., Prasher, V., & Glenholmes, P. (2015). Routinised and compulsive-like behaviors in individuals with Down syndrome. *Journal of Intellectual Disability Research, 59*, 1061–1070.

Gowen, E., & Hamilton, A. (2013). Motor abilities in autism: A review using a computational context. *Journal of Autism and Developmental Disorders, 43*, 323–344.

Grahame, V., Brett, D., Dixon, L., McConachie, H., Lowry, J., Rodgers, J., et al. (2015). Managing repetitive behaviors in Young children with Autism Spectrum Disorder (ASD): Pilot randomised controlled trial of a new parent group intervention. *Journal of Autism and Developmental Disorders, 45*(10), 3168–3182.

Green, D., Baird, G., Barnett, A. L., Henderson, L., Huber, J., & Henderson, S. E. (2002). The severity and nature of motor impairment in Asperger's syndrome: A comparison with specific developmental disorder of motor function. *Journal of Child Psychology and Psychiatry, 43*, 655–668.

Gullone, E. (2000). The development of normal fear: A century of research. *Clinical Psychology Review, 20*, 429–451.

Harrop, C., McBee, M., & Boyd, B. A. (2016). How are child restricted and repetitive behaviors associated with caregiver stress over time? A parallel process multilevel growth model. *Journal of Autism and Developmental Disorders, 46*(5), 1773–1783.

Harrop, C., McConachie, H., Emsley, R., Leadbitter, K., Green, J., & The PACT Consortium. (2014). Restricted and repetitive behaviors in autism spectrum disorders and typical develop-

ment: Cross-sectional and longitudinal comparisons. *Journal of Autism and Developmental Disorders, 44*(5), 1207–1219.

Hill, E. L. (2004). Evaluating the theory of executive dysfunction in autism. *Developmental Reviews, 24*(2), 189–233.

Hollander, E., Anagnostou, E., Chaplin, W., Esposito, K., Haznedar, M., Licalzi, E., et al. (2005). Striatal volume on magnetic resonance imaging and repetitive behaviors in autism. *Biological Psychiatry, 58*, 226–232.

Hollocks, M., Jones, C., Pickles, A., Baird, G., Happe, F., & Simonoff, E. (2014). The association between social cognition and executive functioning and symptoms of anxiety and depression in adolescents with autism spectrum disorders. *Autism Research, 7*(2), 216–228.

Honey, E., McConachie, H., Turner, M., & Rodgers, J. (2012). Validation of the repetitive behavior questionnaire for use with children with autism spectrum disorder. *Research in Autism Spectrum Disorders, 6*, 355–364.

Hughes, C., & Ensor, R. (2011). Individual differences in growth in executive function across the transition to school predict externalizing and internalizing behaviors and children's self-perceived academic success at age 6. *Journal of Experimental Child Psychology, 108*, 663–676.

Insel, T. R. (2010). The challenge of translation in social neuroscience: A review of oxytocin, vasopressin, and affiliative behavior. *Neuron, 65*, 768–779.

Judge, P. G., Evans, D. W., Schroepfer, K. K., & Gross, A. C. (2011). Perseveration on a reversal-learning task correlates with rates of self-directed behavior in nonhuman primates. *Behavioral Brain Research, 222*(1), 57–65.

Kanner, L. (1943). Autistic disturbances of affective contact. *Nervous Child, 2*, 217–250.

Kenworthy, L., Anthony, L. G., Naiman, D. Q., Cannon, L., Wills, M. C., Luong-Tran, C., et al. (2014). Randomized controlled effectiveness trial of executive function intervention for children on the autism spectrum. *Journal of Child Psychology and Psychiatry, 55*, 374–383.

Lam, K. S. L., Bodfish, J. W., & Piven, J. (2008). Evidence for three subtypes of repetitive behavior in autism that differ in familiality and association with other symptoms. *Journal of Child Psychology and Psychiatry, 49*(11), 1193–1200.

Langen, M., Durston, S., Kas, M. J. H., Van Engeland, H., & Staal, W. G. (2011). The neurobiology of repetitive behavior and men. *Neuroscience and Biobehavioral Reviews, 35*, 356–365.

Larkin, F., Meins, E., Centifanti, L. C. M., Fernyhough, C., & Leekam, S. R. (2017). How does restricted and repetitive behavior relate to language and cognition in typical development? *Development and Psychopathology, 29*(3), 863–874.

Leekam, S. R., Prior, M. R., & Uljarević, M. (2011). Restricted and repetitive behaviors in autism spectrum disorders: A review of research in the last decade. *Psychological Bulletin, 137*(4), 562–593.

Leekam, S. R., Tandos, J., McConachie, H., Meins, E., Parkinson, K., Wright, C., et al. (2007). Repetitive behaviors in typically developing 2 year olds. *Journal of Child Psychology and Psychiatry, 48*(11), 1131–1138.

Leonard, H. C., & Hill, E. L. (2014). The impact of motor1 development on typical and atypical social cognition and language: A systematic review. *Child and Adolescent Mental Health, 19*, 163–170.

Lidstone, J., Uljarević, M., Sullivan, J., Rodgers, J., McConachie, H., Freeston, M., et al. (2014). Relations among restricted and repetitive behaviors, anxiety and sensory features in children with autism spectrum disorders. *Research in Autism Spectrum Disorders, 8*(2), 82–92.

Linkovski, O., Kalanthroff, E., Henik, A., & Anholt, G. E. (2013). Did I turn off the stove? Good inhibitory control can protect from influences of repeated checking. *Journal of Behavior Therapy and Experimental Psychiatry, 44*(1), 30–36.

Linkovski, O., Kalanthroff, E., Henik, A., & Anholt, G. E. (2016). Stop checking: Repeated checking and its effects on response inhibition and doubt. *Journal of Behavior Therapy and Experimental Psychiatry, 53*, 84–91.

Memisevic, H., & Sinanovic, O. (2014). Executive function in children with intellectual disability–the effects of sex, level and aetiology of intellectual disability. *Journal of Intellectual Disability Research, 58*(9), 830–837.

Ming, X., Brimacombe, M., & Wagner, G. C. (2007). Prevalence of motor impairment in autism spectrum disorders. *Brain Development, 29*, 565–570.

Miyahara, M., Tsujii, M., Hori, M., Nakanishi, K., Kageyama, H., & Sugiyama, T. (1997). Brief report: Motor incoordination in children with Asperger syndrome and learning disabilities. *Journal of Autism and Developmental Disorders, 27*, 595–603.

Murphy, G. H., Beadle-Brown, J., Wing, L., Gould, J., Shah, A., & Holmes, N. (2005). Chronicity of challenging behaviors in people with severe intellectual disabilities and/or autism: A total population sample. *Journal of Autism and Developmental Disorders, 35*, 405–418.

Murray, K. T., & Kochanska, G. (2002). Effortful control: Relation to externalizing and internalizing behaviors and factor structure. *Journal of Abnormal Child Psychology, 30*, 503–514.

Nickel, L. R., Thatcher, A. R., Keller, F., Wozniak, R. H., & Iverson, J. M. (2013). Posture development in infants at heightened vs. low risk for autism spectrum disorders. *Infancy, 18*, 639–661.

Ozonoff, S., Macari, S., Young, G. S., Goldring, S., Thompson, M., & Rogers, S. J. (2008). Atypical object exploration at 12 months of age is associated with autism in a prospective sample. *Autism, 12*, 457–472.

Paul, R., Chawarska, K., Cicchetti, D., & Volkmar, F. (2008). Language outcomes of toddlers with autism spectrum disorders: A two- year follow-up. *Autism Research, 1*, 97–107.

Peleg-Popko, O., & Dar, R. (2003). Ritual behavior in children and mothers' perceptions of family patterns. *Journal of Anxiety Disorders, 17*(6), 667–681.

Pietrefesa, A., & Evans, D. W. (2007). Affective and neuropsychological correlates of children's rituals and compulsive-like behaviors: Continuities and discontinuities with obsessive compulsive disorder. *Brain and Cognition, 65*, 36–46.

Prior, M., & Macmillan, M. B. (1973). Maintenance of sameness in children with Kanner's syndrome. *Journal of Autism and Developmental Disorders, 3*, 154–167.

Ray-Subramanian, C. E., & Ellis Weismer, S. (2012). Receptive and expressive language as predictors of restricted and repetitive behaviors in young children with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 42*, 2113–2120.

Richler, J., Huerta, M., Bishop, S. L., & Lord, C. (2010). Developmental trajectories of restricted and repetitive behaviors and interests in children with autism spectrum disorders. *Development and Psychopathology, 22*(1), 55–69.

Rodgers, J., Riby, D. M., Janes, E., Connolly, B., & McConachie, H. (2012). Anxiety and repetitive behaviors in autism spectrum disorders and Williams syndrome: A cross syndrome comparison. *Journal of Autism and Developmental Disorders, 42*, 175–180.

Rueda, M. R., Posner, M. I., & Rothbart, M. K. (2005). The development of executive attention: Contributions to the emergence of self-regulation. *Developmental Neuropsychology, 28*, 573–594.

Shao, Y., Cuccaro, M. L., Hauser, E. R., Raiford, K. L., Menold, M. M., Wolpert, C. M., et al. (2003). Fine mapping of autistic disorder to chromosome 15q11-q13 by use of phenotypic subtypes. *American Journal of Human Genetics, 72*(3), 539–548.

Silverman, J., Smith, C., Schmeidler, J., Hollander, E., Lawlor, B., Fitzgerald, M., Galvin, P., et al. (2002). Symptom domains in autism and related conditions: Evidence for familiality. *American Journal of Medical Genetics, 114*, 64–73.

South, M., Ozonoff, S., & McMahon, W. M. (2005). Repetitive behavior profiles in Asperger syndrome and high functioning autism. *Journal of Autism and Developmental Disorders, 35*, 145–158.

Sprague, R. L., & Newell, K. M. (1996). *Stereotypies: Brain-behavior relationships*. Washington, DC: American Psychological Association.

Staal, W. G., de Krom, M., & de Jonge, M. V. (2012). Brief report: The dopamine-3-receptor gene (DRD3) is associated with specific repetitive behavior in autism spectrum disorder (ASD). *Journal of Autism and Developmental Disorders, 42*(5), 885–888.

Szatmari, P., Georgiades, S., Bryson, S., Zwaigenbaum, L., Roberts, W., Mahoney, W., Tuff, L., et al. (2006). Investigating the structure of the restricted, repetitive behaviors and interests domain of autism. *Journal of Child Psychology and Psychiatry, 47*, 582–590.

Thelen, E. (1979). Rhythmical stereotypies in normal human infants. *Animal Behavior, 27*, 699–715.

Thelen, E. (1981). Rhythmical behavior in infancy: An ethological perspective. *Developmental Psychology, 17*, 237–257.

Thorell, L. B., Lindqvist, S., Nutley, S. B., Bohlin, G., & Klingberg, T. (2009). Training and transfer effects of executive functions in preschool children. *Developmental Science, 12*, 106–113.

Tregay, J., Gilmour, J., & Charman, T. (2009). Childhood rituals and executive functions. *British Journal of Developmental Psychology, 27*(2), 283–296.

Turner, M. (1999). Annotation: Repetitive behavior in autism: A review of psychological research. *Journal of Child Psychology and Psychiatry, 40*, 839–849.

Uljarević, M., Arnott, B., Carrington, S. J., Meins, E., Fernyhough, C., McConachie, H., et al. (2017). Development of restricted and repetitive behaviors from 15 to 77 months: Stability of two distinct subtypes? *Developmental Psychology, 53*(10), 1859–1868.

Uljarevic, M., & Evans, D. W. (2017). Relationship between repetitive behavior and fear across normative development, autism spectrum disorder, and down syndrome. *Autism Research, 10*(3), 502–507.

Uljarević, M., Evans, D. W., Alvares, G. A., & Whitehouse, A. O. J. (2016). Relationship between restricted and repetitive behaviors in children with autism spectrum disorder and their parents. *Molecular Autism, 7*, 29.

Uljarević, M., Hedley, D., Alvares, G. A., Varcin, K. J., & Whitehouse, A. J. O. (2017). Relationship between early motor milestones and severity of restricted and repetitive behaviors in children and adolescents with autism spectrum disorder (ASD). *Autism Research, 10*(6), 1163–1168.

Uljarević, M., Hedley, D., Rose-Foley, K., Magiati, I., Cai, R.-Y., Dissanayake, C., Richdale, A., & Trollor, J. (2020). Anxiety and depression from adolescence to old age in autism Spectrum disorder. *Journal of Autism and Developmental Disorders, 50*(9), 3155–3165.

Uljarević, M., Labuschagne, I., Bobin, R., Atkinson, A., & Hocking, D. R. (2018). Brief report: The impact of sensory hypersensitivity and intolerance of uncertainty on anxiety in Williams syndrome. *Journal of Autism and Developmental Disorders, 48*(11), 3958–3964.

Uljarević, M., Nuske, H., & Vivanti, G. (2016). Anxiety in autism spectrum disorder. In L. Mazzone & B. Vitiello (Eds.), *Psychiatric symptoms and comorbidities in autism spectrum disorder* (pp. 21–38). Cham: Springer International Publishing.

Uljarević, M., Richdale, A., Cai, R.-Y., Evans, D. W., & Leekam, S. R. (2017). Inter-relationship between insistence on sameness, effortful control and anxiety in adolescents and young adults with autism spectrum disorder (ASD). *Molecular Autism, 8*, 36.

van Steensel, F. J., Bogels, S. M., & Perrin, S. (2011). Anxiety disorders in children and adolescents with autistic spectrum disorders: A meta-analysis. *Clinical Child and Family Psychology, 14*, 302–317.

Wallace, G. L., Kenworthy, L., Pugliese, C. E., Popal, H. S., White, E. I., Brodsky, E., & Martin, A. (2016). Real-world executive functions in adults with autism spectrum disorder: Profiles of impairment and associations with adaptive functioning and co-morbid anxiety and depression. *Journal of Autism and Developmental Disorders, 46*(3), 1071–1083.

Whyatt, C. P., & Craig, C. M. (2012). Motor skills in children aged 7–10 years, diagnosed with autism spectrum disorder. *Journal of Autism and Developmental Disorders, 42*, 1799–1809.

Wolff, J. J., Botterton, K. N., Dager, S. R., Elison, J. T., Estes, A. M., Gu, H., et al. (2014). Longitudinal patterns of repetitive behavior in toddlers with autism. *Journal of Child Psychology and Psychiatry, 55*(8), 945–953.

Wolff, P. H. (1968). Stereotypic behavior and development. *Canadian Psychologist/Psychologie Canadienne, 9*, 474–484.

Yerys, B. E. (2015). An update on the neurobiology of repetitive behaviors in autism. In R. M. Hodapp & D. J. Fidler (Eds.), *International review of research in developmental disabilities* (pp. 91–150). Amsterdam: Academic Press.

Zelazo, P. D., Muller, U., Frye, D., Marcovitch, S., Argitis, S., Boseovski, J., et al. (2003). The development of executive function in early childhood. *Monographs of the Society for Research in Child Development, 68*(3), vii–137.

Zhou, Q., Chen, S. H., & Main, A. (2011). Commonalities and differences in the research on children's effortful control and executive function: A call for an integrated model of self-regulation. *Child Development Perspectives, 6*(2), 112–121.

Zohar, A. H., & Felz, L. (2001). Ritualistic behavior in young children. *Journal of Abnormal Child Psychology, 29*, 121–128.

Chapter 6
Sensory Subtypes in Autism Spectrum Disorder

Alison E. Lane

Sensory Features in Autism Spectrum Disorder

Sensory features have been observed and documented since the earliest reports of Autism Spectrum Disorder (ASD). Kanner (1943) described some sensory features in his initial descriptions of childhood ASD including extreme fear of noisy household appliances, fixation on sensory-stimulating activities e.g. spinning, humming to self and rejection of social touch. His case examples also detail evidence of enhanced sensory abilities such as noticing small changes in the physical arrangement of objects in the room. After initial inclusion in the original diagnostic criteria for ASD, sensory features were dropped from subsequent diagnostic manuals until 2013 when they were re-included in the DSM-5 (American Psychiatric Association, 2013). Current estimates of the prevalence of sensory features in ASD range from 60–95% (Lane, Molloy, & Bishop, 2014; Tomchek & Dunn, 2007). In the more than 60 years that have passed since Kanner's initial observations, sensory features have become a commonly observed aspect of the behavioural presentation of ASD and studies regarding the characterisation, mechanisms and treatment of sensory features have increased exponentially (Cascio, Woynaroski, Baranek, & Wallace, 2016; Uljarević et al., 2017).

Sensory features refer to patterns of behaviour that are suggestive of differences in the way daily sensory stimuli are processed, e.g., covering ears in response to an unexpected sound or failure to respond to a painful stimulus (Schaaf & Lane, 2015). In general, sensory features are considered functionally limiting, with individuals with ASD and their families attributing significant restrictions in participation in daily life activities to sensory symptoms (Dunn, Little, Dean, Robertson, & Evans, 2016; Schaaf, Toth-Cohen, Johnson, Outten, & Benevides, 2011). First-hand

A. E. Lane (✉)
School of Health Sciences, The University of Newcastle, Callaghan, NSW, Australia
e-mail: alison.lane@newcastle.edu.au

E. Gal, N. Yirmiya (eds.), *Repetitive and Restricted Behaviors and Interests in Autism Spectrum Disorders*, Autism and Child Psychopathology Series,
https://doi.org/10.1007/978-3-030-66445-9_6

accounts of the impact of sensory features on daily living indicate that sensory sensitivities (e.g. sensitivity to unexpected sounds like a phone ringing, food tastes or smells) can lead to avoidance behaviours and strong emotional reactions to changes in routine or environments (Ashburner, Bennett, Rodger, & Ziviani, 2013; Dickie, Baranek, Schultz, Watson, & McComish, 2009). Further, some daily sensory experiences are reported as distracting (e.g. visual stimulation of moving ceiling fan) resulting in loss of attention and focus, failure to notice more salient stimuli and social difficulties (Ashburner et al., 2013). There are some reports, however, that sensory features may also enhance function, such as a heightened level of awareness to visual detail that may assist in the performance of some learning tasks.

The definition and characterisation of sensory symptoms has been an issue of some debate and controversy in the literature. Discrepancies can be found between descriptions of sensory features found in clinically-oriented versus more experimental literature. In clinical fields, the emphasis in definition has been on behaviours that limit function and some attempts have been made to characterise specific sensory 'sub-disorders' based on the combination of clinically meaningful symptom sets. Several examples of this are found in the occupational therapy literature (Ayres, 1979; Dunn, 2001; Miller, Anzalone, Lane, Cermak, & Osten, 2007). Ayres' work laid the foundation for the recognition of sensory features as clinically important for children with a variety of developmental disorders, including ASD. Ayres proposed a theoretical framework for the understanding of how impairments in the integration of daily environmental sensory stimuli may lead to identifiable patterns of maladaptive behaviour and learning difficulties. These patterns were further described as 'sensory integration disorders' and a model of treatment for each was developed (Ayres, 1979; Bundy & Murray, 2002). Central to Ayres' theory was a distinction between sensory features based on impairments in 'sensory modulation' versus those related to difficulties in processing somatosensory stimuli (vis a vis, tactile, proprioceptive and vestibular) for the purposes of coordinated, goal-directed movement (Bundy & Murray, 2002). More recently, sensory difficulties associated with impairments in sensory modulation have received the greater attention in the clinical literature.

'Sensory modulation' is defined as the ability of the central nervous system to regulate its responses to sensory input (Bundy & Murray, 2002). Dunn (1997) proposed that impairments in sensory modulation present as symptom sets that fall into one of four sensory quadrants – poor registration, sensory sensitivity, sensory avoiding and sensory seeking. Classification into one of the four quadrants is determined by both a hypothesised 'neurological threshold' indicating the level of stimulation (high or low) needed to elicit a behavioural response and the behavioural 'style' of the individual (either passive or active; Dunn, 1997). Individuals with a high neurological threshold and a passive behavioural style are classified as 'poor registration' demonstrated by behaviours that indicate an attenuated or absent response to a sensory stimulus. 'Sensory seeking' encapsulates individuals with a high neurological threshold but an active behavioural style suggesting that behaviours which appear to increase the level of stimulation gained, are the result of inadequate registration of the available sensory stimuli in the environment. The final two quadrants relate to

individuals with a low neurological threshold. Sensory avoiders use their active behavioural style to remove themselves from sensory stimuli that may become overwhelming or be perceived as highly intense. Individuals classified in the sensory sensitivity quadrant, however, have a passive behavioural style and may demonstrate less overt signs of distress to sensory stimuli such as withdrawal, anxiety or other internalising symptoms (Dunn, 1997).

In contrast to the clinical models described above, researchers in more experimental disciplines have attempted to define sensory features in terms of underlying structures, mechanism and impairment (Marco, Hinkley, Hill, & Nagarajan, 2011). This body of work focuses on biological processes related to sensory features and encapsulates studies in neuroscience and cognitive psychology. Psychophysiological and brain imaging techniques have been used to quantify neurophysiologic responses to sensory stimuli, usually under controlled conditions (see Marco et al., 2011 for a review of this work). Until recently, clinical and experimental inquiries into sensory features have been largely conducted independently of each other (Cascio et al., 2016). This has led to some confusion in the terminology used and understanding of sensory features (Cascio et al., 2016). Schaaf and Lane (2015) have attempted to clarify some of this confusion and provided guidance for terminology usage. These authors suggested that the terms *sensory reactivity*, *sensory perception* and *sensory integration* be used to characterise the full extent of sensory symptoms. *Sensory reactivity* refers to behaviours termed as hyper-, over-, hypo- or under-responsivity or sensitivity. Behaviours indicative of sensory reactivity difficulties might include responses to stimuli that are either too strong (e.g. extreme distress to the sound of a vacuum cleaner turning on) or insufficient (e.g. no response to a painful stimulus). In this chapter, we will use 'sensory reactivity' synonymously with 'sensory modulation'. *Sensory perception* refers to the ability to perceive and interpret sensory stimuli (Schaaf & Lane, 2015). In general, sensory perception refers to cognitive and physiological functions that are measured through standardised behavioural tests or psychophysiological procedures. Finally, *sensory integration* refers to the organisation, binding or assimilation of multiple sensory inputs for the purpose of more detailed understanding of the sensory context (Schaaf & Lane, 2015). Sensory integration is considered synonymous with multisensory integration.

Despite this growing understanding of the breadth of sensory symptoms, sensory features as defined in the DSM-5 diagnostic criteria for ASD (Association, 2013) refer only to difficulties in sensory modulation and are based on clinical conventions. The DSM-5 defines sensory features as:

> *Hyper- or hypo-reactivity to sensory input or unusual interests in sensory aspects of the environment (e.g., apparent indifference to pain/temperature, adverse response to specific sounds or textures, excessive smelling or touching of objects, visual fascination with lights or movement).*

Under current diagnostic guidelines for ASD, sensory features are considered a sufficient but not necessary element of the 'restricted, repetitive patterns of behaviour, interests, or activities (RRBI)' criterion. As such, the observation of sensory features (as described above) in an individual presenting for ASD diagnosis can be included as one of the two elements required to meet the RRBI criterion.

Relation of Sensory Features to Other RRBI

Investigations into the relation of sensory features to other RRBI in ASD are premised on the assumption that one possible function of RRBI is as a mechanism to manage adverse responses to daily sensory stimuli. In the 'over-arousal' theory, it has been postulated that RRBI including repetitive motor behaviours, adherence to routines, and preoccupations may serve to block sensory input that is perceived as threatening or too intense by individuals who experience sensory hyper-reactivity (Schulz & Stevenson, 2019). Evidence to support this theory is found in the literature reporting that increased sensory hyper-reactivity is associated with increased frequency and intensity of repetitive behaviours of all types (Chen, Rodgers, & McConachie, 2008; Schulz & Stevenson, 2019; Wigham, Rodgers, South, McConachie, & Freeston, 2015; Wolff et al., 2019). Further, this relation has been reported to hold across ASD and typically developing groups, regardless of gender, chronological age and IQ (Schulz & Stevenson, 2019).

In a second theory, authors propose that engagement in RRBI by individuals with ASD may serve to provide additional sensory input to individuals who experience sensory hypo-reactivity and are less able to use sensory stimuli in the environment (Joosten & Bundy, 2010). Clinical sensory theorists such as Dunn (1997) and Miller et al. (2007) propose that sensory seeking behaviours, such as repetitive motor behaviours, provide a hypo-reactive individual with an opportunity to generate sensory experiences, which aid in self-regulation, adaptive behaviour and learning. In support of this proposition, Wigham et al. (2015) observed that increased sensory hypo-reactivity but not hyper-reactivity was significantly associated with increased repetitive motor behaviours in ASD. Further, Gal, Dyck, and Passmore (2010) reported that sensory hypo-reactivity was the strongest correlate of stereotyped movements in children with ASD. Other study findings, however, contradict the 'over-arousal'/'seeking' theories. For example, Wigham et al. (2015) also observed that sensory hypo-reactivity was significantly associated with 'insistence on sameness' behaviours. In the context of sensory features, 'insistence on sameness' behaviours are generally considered to be efforts to control or reduce the level of sensory input in the environment and as such, would be more logically related to sensory hyper-reactivity (Black et al., 2017).

It is likely, therefore, that the relations between sensory features and other RRBI cannot be completely explained by the 'over-arousal' and 'seeking' theories and additional factors might be at play (Wolff et al., 2019). For example, several commentators have postulated that there may be an important role for 'intolerance of uncertainty' and anxiety in the interplay between RRBI and sensory features (Joosten & Bundy, 2010; Neil, Olsson, & Pellicano, 2016; Wigham et al., 2015). Wigham reported that these factors in combination at least partially mediated the relations among sensory hypo- and hyper-reactivity and both repetitive motor behaviours and insistence on sameness. Neil et al. (2016) reported that 'intolerance of uncertainty' explained half the variance in sensory sensitivities in children with ASD but that a portion of this was mediated by anxiety. A further study observed that sensory avoiding (thought to be one manifestation of sensory hyper-reactivity)

mediated the relationship between 'insistence on sameness' and anxiety (Lidstone et al., 2014) and Black et al. (2017) observed that sensory hyper-reactivity mediated the relation between 'insistence on sameness' and specific phobias and separation anxiety in ASD only. There are also emerging reports of a role for sensory perception in the manifestation of RRBI. Kargas et al. (2015) found that auditory discrimination impairments (vis a vis intensity and frequency discrimination) in adults with ASD were associated with more severe RRBI as measured by the Autism Diagnostic Observation Schedule and including preoccupations in play, restricted interests, adherence to routines and repetitive motor patterns. Similarly, Kanakri et al. (2017) observed that increased ambient noise levels in classrooms were associated with increased repetitive motor and speech behaviours in ASD.

The findings from the available literature suggest that the relation between sensory features and RRBI is complex and likely to be multifactorial. Interventions targeting sensory features, however, could be hypothesised to reduce the frequency and severity of other RRBI although this has yet to be confirmed in controlled trials. The mechanism by which sensory directed therapies may impact other RRBI is still unknown although a potential common neural circuity between RRBI and sensory features has been identified (Wolff et al., 2017). Further complicating our understanding of this relationship is the fact that many individuals with ASD present with concurrent sensory hyper-, hypo- and seeking behaviours. Further exploration, therefore, of the relation between RRBI and sensory features is warranted considering patterns of sensory features and RRBI within individuals rather than an exclusive focus on specific sensory behaviours and their RRBI correlates in isolation.

Sensory Subtyping

In further efforts to understand the manifestation and impact of sensory features in ASD, recent investigations have attempted to identify specific patterns of sensory symptoms within individuals with ASD. These patterns or 'subtypes' identify homogenous sub-groups of individuals with ASD with similar sensory features. This approach varies substantially from previous sensory research which has focussed more on the identification of discrete sensory behaviours or features but less on the pattern of co-existence of those behaviours within individuals (Hand, Dennis, & Lane, 2017). Efforts to identify subgroups of individuals with ASD with similar sensory features have implications for our understanding of the basis of sensory disturbance in ASD, and also provide a framework for the provision of customised and targeted therapies. To date, there have been seven proposed sensory subtype models in ASD. All subtype models have focussed on identifying distinct patterns of sensory features within toddlers, children and adolescents. There are no current sensory subtype models for adults with ASD. Further, all subtype models are based on observations made by parents or caregivers of individuals with ASD. Objective measures of sensory features (vis a vis neurophysiological data) have not yet been included in subtype models.

Toddler Models

To date, there have been two published reports of sensory subtype studies in toddlers with or with risk for ASD (Ben-Sasson et al., 2008; Philpott-Robinson, Lane, & Harpster, 2016). Utilising hierarchical cluster analysis, Ben-Sasson et al. (2008) reported that toddlers with confirmed diagnoses of ASD were rated by their parents on the Infant-Toddler Sensory Profile (Dunn & Daniels, 2002), to fall into one of three sensory clusters – low frequency of sensory symptoms (26%), high frequency of sensory symptoms (29%) and mixed (45%). Toddlers in the low frequency cluster displayed few sensory symptoms whereas those in the high frequency cluster showed a high number of sensory hyper-, hypo- and seeking behaviours. The mixed sensory cluster demonstrated high levels of both sensory hyper- and hypo-reactivity but less sensory seeking. Further, members of the high frequency cluster showed the highest levels of depression/withdrawal, whereas the high frequency and mixed clusters displayed more negative emotionality than the low frequency group (Ben-Sasson et al., 2008).

Philpott-Robinson et al. investigated sensory features in 12–24 month old toddlers with risk factors for ASD (n = 46). Sensory features were measured using the Infant-Toddler Sensory Profile (Dunn & Daniels, 2002) completed by parents or caregivers. Model-based cluster analysis was used to interrogate responses and identify homogenous subsets of toddlers. Philpott-Robinson et al. identified two primary sensory subtypes in this group: (1) Sensory Adaptive (59%) and (2) Sensory Reactive (41%). The sensory features of members of the Sensory Adaptive subtype were characterised by typical function across sensory domains. Members of the Sensory Reactive subtype, however, displayed symptoms of sensory hyper-reactivity across sensory domains. Whereas sensory subtype membership in this sample was not associated with early ASD risk, toddlers in the Sensory Reactive subtype demonstrated less mature expressive and receptive language abilities.

Childhood Models

Lane Model

One of the first sensory subtype models was proposed by Lane and colleagues (Lane et al., 2014; Lane, Dennis, & Geraghty, 2011; Lane, Young, Baker, & Angley, 2010). This model is based on parent observations of sensory features in children with ASD aged 2–10 years (n = 312 across 3 studies) using the Short Sensory Profile (McIntosh et al. 1999). Model-based cluster analysis was used to identify homogenous subgroups of children with ASD based on their sensory features. On the basis of their findings, Lane and colleagues proposed that children with ASD can be classified into one of four sensory subtypes – Sensory Adaptive, Taste Smell Sensitive, Postural Inattentive and Generalised Sensory Difference (Lane et al., 2014). Subtypes differ from each other on the basis of the *severity* (mild to severe) and *focus* (auditory, taste, smell, proprioceptive and vestibular) of the sensory

symptoms. It is further hypothesised by the authors, that subtype classifications can be understood as relating to difficulties in *sensory reactivity* and/or *multisensory integration* (Hand et al., 2017). In this context, sensory reactivity is considered synonymous with sensory modulation. Difficulties in sensory reactivity manifest as behaviours that are either too intense (hyper-reactive) or insufficiently intense (hypo-reactive) for a given stimulus. For example, crying and extreme upset during hair-cutting may be indicative of sensory hyper-reactivity to tactile stimuli whereas failure to respond to name may be indicative of sensory hypo-reactivity to speech stimuli. Multisensory integration difficulties in Lane's model refer to higher level behaviours that are indicative of potential failures in the assimilation of multiple, concurrent sensory inputs. Such behaviours could include postural and motor coordination difficulties (Hand et al., 2017; Lane et al., 2014). Figure 1 outlines the relation of Lane's four sensory subtypes with their proposed underlying mechanisms.

As can be seen, children with ASD who are classified as Sensory Adaptive, experience no clinically significant difficulties with either sensory reactivity or multisensory integration. Their responses to daily sensory stimuli are reported by their parents to fall within normal limits. Children with ASD who are classified as Taste/Smell Sensitive, however, display behaviours suggestive of difficulties with sensory reactivity. These children do not, however, appear to experience impairment in multisensory integration. Those children with ASD classified as Postural Inattentive display difficulties in postural control, maintenance of body positions against gravity and filtering salient from less salient auditory stimuli. These behaviours are suggestive of impairment in multisensory integration. These children do not, however, appear to experience difficulties in sensory reactivity. Finally, children with ASD classified as Generalised Sensory Difference are reported by their parents to experience difficulties in both sensory reactivity and multisensory integration.

Lane et al. (2014) observed that in a large group of children with ASD presenting for diagnosis of ASD, most were classified into either Sensory Adaptive (37.5%) or Taste/Smell Sensitive (40.2%) subtypes. Patterns of sensory features indicated by Postural Inattentive (10.3%) and Generalised Sensory Difference (12.1%) were less common. As such, this subtype model concludes that significant numbers of

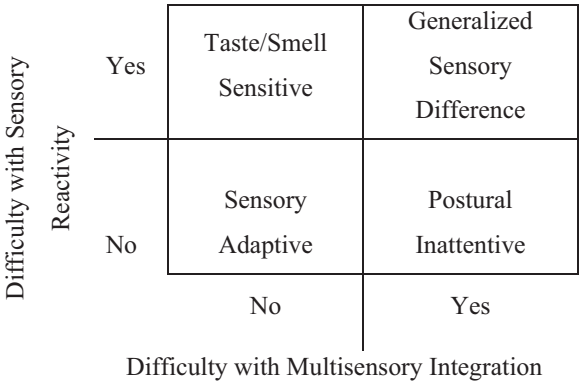

Fig. 1 Lane's sensory subtype model. (From Hand et al., 2017. Reprinted with permission)

children with ASD do not experience clinically significant sensory features. Further, subtype membership was not found to be strongly associated with non-sensory features such as ASD symptom severity, gender or IQ (Lane et al., 2014). Differences between subtypes have been reported, however, in adaptive behaviour with members of subtypes experiencing greatest difficulties with sensory reactivity, being reported to experience the highest levels of challenging behaviours (Lane et al., 2010). Further, members of the Taste/Smell Sensitive subtype displayed the highest levels of communication difficulty and picky eating (Lane et al., 2010, 2011).

Ausderau Model

A second sensory subtype model was proposed by Ausderau et al. (2014). As for the Lane model, this model is based on parent-reported sensory features of children with ASD (2–12 years). In this model, however, the Sensory Experiences Questionnaire (SEQ; (Baranek, Boyd, Poe, David, & Watson, 2007) was utilised. Ausderau et al. (2014) applied Latent Profile Analysis to the SEQ responses of a large sample of participants (n = 1294) to identify homogenous subgroups of children with ASD based on their sensory features. These authors also identified four distinct sensory subtype groupings. The four subtypes were described as: Mild, Extreme-Mixed, Sensitive-Distressed and Attenuated-Preoccupied. As for the Lane model, the subtypes proposed by Ausderau and colleagues differ from each other in terms of the *frequency and intensity* and the *focus* of sensory symptoms. Individuals classified in the Mild subtypes experienced very few sensory symptoms whereas those in the Extreme-Mixed subtype were reported to experience high levels of symptoms across all sensory domains. Individuals classified as Sensitive-Distressed reported more sensory symptoms related to hyper-reactivity and enhanced sensory perception whereas those in Attenuated-Preoccupied reported more symptoms related to hypo-reactivity and sensory interests, repetitions and seeking (Ausderau et al., 2014). Ausderau et al. found that most participants were classified into either the Mild (29%) or Sensitive-Distressed (28%) subtypes with fewer participants in the Extreme-Mixed (17%) or Attenuated-Preoccupied (17%) subtypes.

In follow up work, Ausderau and colleagues have reported that subtype membership was stable after one year (91%) and ASD symptom severity was greater in the Extreme-Mixed subtype relative to the Mild subtype Ausderau et al. (2014). Further, the Attenuated-Preoccupied subtype presented with the lowest proxy IQ and youngest age. Functional outcomes for each subtype also differ. Membership in the Attenuated-Preoccupied subtype was associated with lowest levels of adaptive behaviour whilst Extreme-Mixed was associated with the highest levels of parenting stress (Ausderau et al., 2016). Ausderau et al. (2014) further observe that their subtype groupings provide additional insights into the relations between sensory features and RRBI. In their model, sensory interests, repetitions and seeking behaviours co-occurred with both hyper- (Extreme-Mixed) and hypo-reactive (Attenuated-Preoccupied) symptom sets suggesting the RRBI may serve differing purposes for different subtypes.

Tomchek Model

Recently, a third childhood sensory subtype model has been proposed (Tomchek, Little, Myers, & Dunn, 2018). As for the Lane Model, this model is based on parent reports of sensory features utilising the Short Sensory Profile (McIntosh et al., 1999). Tomchek's model, however, differs from the earlier subtype models in that it is focused only on younger children with ASD (n = 400; aged 3–6 years) and includes developmental features (adaptive and social behaviour, receptive and expressive language and gross and fine motor skills) alongside sensory features within the grouping analysis. Resulting subtypes, therefore, are based on both sensory and developmental features rather than sensory features alone. Further, Tomchek et al. applied an updated factor structure for the Short Sensory Profile to the analysis, based on new data from an ASD-only sample. As in Ausderau et al. (2014), Tomchek et al. utilised Latent Profile Analysis to identify the best subtype model fit to the data.

Tomchek et al. (2018) described four sensory subtypes: Sensorimotor (51%), Selective Complex (15%), Perceptive-Adaptable (25%) and Vigilant-Engaged (10%). Subtypes differed from each other based on age, developmental functioning and sensory features. Specifically, members of the Sensorimotor group were younger, had the lowest developmental functioning and presented with a broad range of sensory symptoms including taste-smell sensitivity, sensory seeking and hypo-responsivity. This contrasted with members of the Perceptive-Adaptable subtype who were also younger but had relatively higher developmental skills particularly in motor, adaptive and social areas, and fewer sensory features. Members of the Selective Complex group were older, showed good motor skills but decreased social and language skills, and demonstrated high levels of sensory hypo-reactivity and sensory seeking. Finally, members of the Vigilant-Engaged subtype were older, had the highest developmental functioning and showed elevated sensory hyper-reactivity and seeking.

Simpson Model

A fourth childhood sensory subtype model was proposed by Simpson, Adams, Alston-Knox, Heussler, and Keen (2019). This model is the first to use the updated Short Sensory Profile-2 (Dunn, 2014) as the basis for subtyping. The Short Sensory Profile-2 (SSP-2) is a substantial revision of the original Short Sensory Profile. Simpson et al. (2019) note that less than 30% of the items between the two measures match. Further, the newer SSP-2 organises items according to Dunn's (1997) quadrant model – sensory sensitivity, sensory avoiding, sensory seeking and poor registration. The original Short Sensory Profile used a seven domain organising structure that incorporated both sensory modality and quadrant descriptor – i.e. tactile sensitivity, taste/smell sensitivity, movement sensitivity, under-responsive/seeking, auditory filtering, low energy/weak and visual/auditory sensitivity. Simpson et al. (2019) conducted their subtyping analysis on SSP-2 reports from caregivers of children with autism (n = 271) aged 4–11 years utilising Dirichelet Process Mixture Modelling (Liverani, Hastie, Papathomas, & Richardson, 2015).

Simpson et al. (2019) identified a two-cluster model as the best solution in their analysis. Clusters were described as: (1) Uniformly Elevated (67%) – indicating elevated sensory scores across all sensory quadrants on the SSP-2, and (2) Raised Avoiding and Sensitivity (33%) – indicating elevated scores in the avoiding and sensitivity quadrants. The authors found no differences between the subtypes on the basis of age or autism-related social communication characteristics.

Adolescent Model

Uljarević, Lane, Kelly, and Leekam (2016) described a sensory subtype model for adolescents with ASD (n = 57; aged 11–17 years). Using an identical methodology to that of Lane et al. (2014), these authors identified three sensory subtypes: Sensory Adaptive (33%), Sensory Moderate (51%) and Sensory Severe (16%). Unlike the previous models, subtypes differed from each other only regarding the frequency and intensity of sensory symptoms rather than the sensory modality or specific sensory behaviours. No differences were observed between subtypes relative to sensory foci in taste/smell, vestibular, proprioceptive, auditory, movement and/or hyper- versus hypo-reactivity as has been reported in childhood sensory subtype models. Further, no clear evidence emerged in this study of specific patterns of sensory difference between groups beyond the overall number of sensory symptoms reported. Similar to previous findings by Lane et al. (2014), however, adolescent subtypes were not different from each other in terms of age, expressive language function or social communication. Differences were identified between sensory subtypes, however, in levels of anxiety with anxiety increasing in adolescents reporting more sensory symptoms.

Summary

Overall, the work completed to date on sensory subtypes in ASD demonstrates a high degree of congruence despite the varying samples, measures and analytic approaches utilised. In school-aged children with ASD, variation in sensory features appears to be best explained by four subtypes that differ from each other on the basis of the severity (frequency and number of sensory symptoms reported), and on the focus of the sensory symptoms. In adolescence, variation in sensory features appears limited to the severity of sensory symptoms only. Only preliminary subtyping results are available for toddlers with or with risk for ASD, however, initial results indicate that at this age, sensory subtypes are characterised by either adaptive sensory functioning (no sensory symptoms) or generalised sensory difficulties (sensory symptoms across domains). Taken together, these findings suggest that:

1. Coherent patterns of sensory features can be identified within children with ASD and not all children with ASD share the same sensory profile. The implications of this finding are that sensory features should be carefully assessed to identify:

(a) the presence of sensory symptoms for purposes of diagnosis, and (b) the type of sensory features experienced by the individual with ASD for purposes of customised treatment planning;
2. Many children with ASD have mild or no clinically significant sensory symptoms as evidenced by the majority of sensory subtype models identifying a 'Mild' or 'Sensory Adaptive' cluster. This finding supports the current diagnostic approach in which sensory symptoms are a 'sufficient' but not 'necessary' subcriterion within the 'restricted, repetitive patterns of behaviour, interests, or activities' domain.
3. Sensory features emerge early in ASD but their pattern of presentation changes with maturation. Whilst severity of sensory symptoms is a consistent source of variation in sensory features in ASD, the focus of sensory symptoms appears to only be a significant contributor to subtype differences during middle childhood.

The evidence presented in this chapter supports the utility of a subtyping approach to the understanding of function and behaviour in individuals with ASD. Current subtype models, however, are limited by the exclusive use of proxy-report measures to identify and characterise subtype features. Further, the measures used differed between subtype models, no doubt contributing to the variations in results achieved. In particular, the scope of the sensory domains addressed by each measure is reflective of conceptual differences between sensory theorists regarding the construct of sensory features in ASD. As written, the DSM-5 only includes clinical sensory features that can be broadly described as related to sensory modulation difficulties. It is evident, however, that sensory features in ASD also include difficulties in sensory perception and sensory integration. The tools used in the generation of sensory subtype models so far, are comprised largely of items representing sensory modulation difficulties. Lane et al. (2014) propose that some items of the Short Sensory Profile are representative of sensory integration, but this theory requires further testing. Before additional progress can be made to understand the nature of sensory features in ASD, a consensus model of the latent constructs underlying sensory function needs to be developed. In doing so, new measures can be aligned to a single construct framework that will assist in the identification of the source of differences between sensory subtypes, generate intervention models targeted to known sensory targets and provide a platform for the study of the emergence of these features in early childhood.

References

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders-Fifth Edition (DSM-5®)*. Arlington, VA: American Psychiatric Association.

Ashburner, J., Bennett, L., Rodger, S., & Ziviani, J. J. (2013). Understanding the sensory experiences of young people with autism spectrum disorder: A preliminary investigation. *Australian Occupational Therapy Journal, 60*(3), 171–180.

Ausderau, K. K., Furlong, M., Sideris, J., Bulluck, J., Little, L. M., Watson, L. R., . . . Baranek, G. T. (2014). Sensory subtypes in children with autism spectrum disorder: Latent profile tran-

sition analysis using a national survey of sensory features. Journal of Child Psychology and Psychiatry, 55(8), 935–944. doi:https://doi.org/10.1111/jcpp.12219.

Ausderau, K. K., Sideris, J., Little, L. M., Furlong, M., Bulluck, J. C., & Baranek, G. T. (2016). Sensory subtypes and associated outcomes in children with autism spectrum disorders. *Autism Research, 9*(12), 1316–1327. https://doi.org/10.1002/aur.1626.

Ayres, A. (1979). *Sensory integration and the child*. Los Angeles: Western Psychological Services.

Baranek, G. T., Boyd, B. A., Poe, M. D., David, F. J., & Watson, L. R. (2007). Hyperresponsive sensory patterns in young children with autism, developmental delay, and typical development. *American Journal on Mental Retardation, 112*(4), 233–245.

Ben-Sasson, A., Cermak, S. A., Orsmond, G. I., Tager-Flusberg, H., Kadlec, M. B., & Carter, A. S. (2008). Sensory clusters of toddlers with autism spectrum disorders: Differences in affective symptoms. *Journal of Child Psychology and Psychiatry, 49*(8), 817–825. https://doi.org/10.1111/j.1469-7610.2008.01899.x.

Black, K. R., Stevenson, R. A., Segers, M., Ncube, B. L., Sun, S. Z., Philipp-Muller, A., et al. (2017). Linking anxiety and insistence on sameness in autistic children: The role of sensory hypersensitivity. *Journal of Autism and Developmental Disorders, 47*(8), 2459–2470. https://doi.org/10.1007/s10803-017-3161-x.

Bundy, A. C., & Murray, E. A. (2002). Sensory integration: A. Jean Ayres' theory revisited. In A. C. Bundy, S. J. Lane, & E. A. Murray (Eds.), *Sensory integration: Theory and practice* (2nd ed.). Philadelphia: F.A. Davis Company.

Cascio, C. J., Woynaroski, T., Baranek, G. T., & Wallace, M. T. (2016). Toward an interdisciplinary approach to understanding sensory function in autism spectrum disorder. *Autism Research, 9*(9), 920–925. https://doi.org/10.1002/aur.1612.

Chen, Y.-H., Rodgers, J., & McConachie, H. (2008). Restricted and repetitive behaviours, sensory processing and cognitive style in children with Autism Spectrum disorders. *Journal of Autism and Developmental Disorders, 39*(4), 635–642. https://doi.org/10.1007/s10803-008-0663-6.

Dickie, V. A., Baranek, G. T., Schultz, B., Watson, L. R., & McComish, C. S. (2009). Parent reports of sensory experiences of preschool children with and without autism: A qualitative study. *American Journal of Occupational Therapy, 63*(2), 172–181. https://doi.org/10.5014/ajot.63.2.172.

Dunn, W. (1997). The impact of sensory processing abilities on the daily lives of young children and their families: A conceptual model. *Infants & Young Children: An Interdisciplinary Journal of Special Care Practices, 9*(4), 23–35.

Dunn, W. (2001). The sensations of everyday life: Empirical, theoretical, and pragmatic considerations. *The American Journal of Occupational Therapy, 55*(6), 608–620.

Dunn, W. (2014). *Sensory Profile 2*. Pearson: San Antonio, TX.

Dunn, W., & Daniels, D. B. (2002). Initial development of the infant/toddler sensory profile. *Journal of Early Intervention, 25*(1), 27–41. https://doi.org/10.1177/105381510202500104.

Dunn, W., Little, L., Dean, E., Robertson, S., & Evans, B. (2016). The State of the Science on sensory factors and their impact on daily life for children: A scoping review. *OTJR: Occupation, Participation and Health, 36*(2_suppl), 3S–26S. https://doi.org/10.1177/1539449215617923.

Gal, E., Dyck, M. J., & Passmore, A. (2010). Relationships between stereotyped movements and sensory processing disorders in children with and without developmental or sensory disorders. *American Journal of Occupational Therapy, 64*, 453–461. https://doi.org/10.5014/ajot.2010.09075.

Hand, B. N., Dennis, S., & Lane, A. E. (2017). Latent constructs underlying sensory subtypes in children with autism: A preliminary study. *Autism Research, 10*(8), 1364–1371. https://doi.org/10.1002/aur.1787.

Joosten, A. V., & Bundy, A. C. (2010). Sensory processing and stereotypical and repetitive behaviour in children with autism and intellectual disability. *American Journal of Occupational Therapy, 57*(6), 366–372.

Kanner, L. J. (1943). Autistic disturbances of affective contact. *The Nervous Child, 2*(3), 217–250.

Kanakri, S. M., Shepley, M., Tassinary, L. G., Varni, J. W., & Fawaz, H. M. (2017). An observational study of classroom acoustical design and repetitive behaviors in children with autism. *Environment and Behavior, 49*(8), 847–873.

Kargas, N., López, B., Reddy, V., & Morris, P. (2015). The relationship between auditory processing and restricted, repetitive behaviors in adults with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 45*(3), 658–668.

Lane, A., Dennis, S., & Geraghty, M. (2011). Brief report: Further evidence of sensory subtypes in autism. *Journal of Autism and Developmental Disorders, 41*(6), 826–831. https://doi.org/10.1007/s10803-010-1103-y.

Lane, A. E., Molloy, C. A., & Bishop, S. L. (2014). Classification of children with autism Spectrum disorder by sensory subtype: A case for sensory-based phenotypes. *Autism Research, 7*(3), 322–333. https://doi.org/10.1002/aur.1368.

Lane, A. E., Young, R. L., Baker, A. E. Z., & Angley, M. T. (2010). Sensory processing subtypes in autism: Association with adaptive behavior. *Journal of Autism and Developmental Disorders, 40*(1), 112–122. https://doi.org/10.1007/s10803-009-0840-2.

Lidstone, J., Uljarević, M., Sullivan, J., Rodgers, J., McConachie, H., Freeston, M., . . . Leekam, S. (2014). Relations among restricted and repetitive behaviors, anxiety and sensory features in children with autism spectrum disorders. *Research in Autism Spectrum Disorders, 8*(2), 82–92. https://doi.org/10.1016/j.rasd.2013.10.001.

Liverani, S., Hastie, D. I., Papathomas, M., & Richardson, S. (2015). PREeMiuM: An R package for profile regression mixture models using Dirichlet processes. *Journal of Statistical Software, 64*(7), 1–30. https://doi.org/10.18637/jss.v064.i07.

Marco, E. J., Hinkley, L. B. N., Hill, S. S., & Nagarajan, S. S. (2011). Sensory processing in autism: A review of neurophysiologic findings. *Pediatric Research, 69*(5 Pt 2), 48R–54R. https://doi.org/10.1203/PDR.0b013e3182130c54.

McIntosh, D. N., Miller, L. J., & Shyu, V. (1999). Development and validation of the Short Sensory Profile. . In W. Dunn (Ed.), *Sensory Profile*. San Antonio, TX: The Psychological Corporation.

Miller, L. J., Anzalone, M. E., Lane, S. J., Cermak, S. A., & Osten, E. T. (2007). Concept evolution in sensory integration: A proposed nosology for diagnosis. *The American Journal of Occupational Therapy, 61*(2), 135–140. https://doi.org/10.5014/ajot.61.2.135.

Neil, L., Olsson, N. C., & Pellicano, E. J. (2016). The relationship between intolerance of uncertainty, sensory sensitivities, and anxiety in autistic and typically developing children. *Journal of Autism and Developmental Disorders, 46*(6), 1962–1973. https://doi.org/10.1007/s10803-016-2721-9.

Philpott-Robinson, K., Lane, A. E., & Harpster, K. (2016). Sensory features of toddlers at risk for autism Spectrum disorder. *American Journal of Occupational Therapy, 70*(4), 1–8. https://doi.org/10.5014/ajot.2016.019497.

Schaaf, R. C., & Lane, A. E. (2015). Toward a best-practice protocol for assessment of sensory features in ASD. *Journal of Autism and Developmental Disorders, 45*(5), 1380–1395. https://doi.org/10.1007/s10803-014-2299-z.

Schaaf, R. C., Toth-Cohen, S., Johnson, S. L., Outten, G., & Benevides, T. W. (2011). The everyday routines of families of children with autism: Examining the impact of sensory processing difficulties on the family. *Autism, 15*(3), 373–389. https://doi.org/10.1177/1362361310386505.

Schulz, S. E., & Stevenson, R. A. J. A. (2019). Sensory hypersensitivity predicts repetitive behaviours in autistic and typically-developing children. *Autism, 23*(4), 1028–1041. https://doi.org/10.1177/1362361318774559.

Simpson, K., Adams, D., Alston-Knox, C., Heussler, H. S., & Keen, D. (2019). Exploring the sensory profiles of children on the autism spectrum using the Short Sensory Profile-2 (SSP-2). *Journal of Autism and Developmental Disorders, 49*, 2069–2079.

Tomchek, S. D., & Dunn, W. (2007). Sensory processing in children with and without autism: A comparative study using the short sensory profile. *The American Journal of Occupational Therapy, 61*(2), 190–200. https://doi.org/10.5014/ajot.61.2.190.

Tomchek, S. D., Little, L. M., Myers, J., & Dunn, W. (2018). Sensory subtypes in preschool aged children with autism Spectrum disorder. *Journal of Autism and Developmental Disorders, 48*(6), 2139–2147.

Uljarević, M., Baranek, G., Vivanti, G., Hedley, D., Hudry, K., & Lane, A. (2017). Heterogeneity of sensory features in autism spectrum disorder: Challenges and perspectives for future research. *Autism Research, 10*(5), 703–710. https://doi.org/10.1002/aur.1747.

Uljarević, M., Lane, A., Kelly, A., & Leekam, S. (2016). Sensory subtypes and anxiety in older children and adolescents with autism spectrum disorder. *Autism Research, 9*(10), 1073–1078. https://doi.org/10.1002/aur.1602.

Wigham, S., Rodgers, J., South, M., McConachie, H., & Freeston, M. J. (2015). The interplay between sensory processing abnormalities, intolerance of uncertainty, anxiety and restricted and repetitive behaviours in autism spectrum disorder. *Journal of Autism and Developmental Disorders, 45*(4), 943–952.

Wolff, J. J., Dimian, A. F., Botteron, K. N., Dager, S. R., Elison, J. T., Estes, A. M., et al. (2019). A longitudinal study of parent-reported sensory responsiveness in toddlers at-risk for autism. *Journal of Child Psychology and Psychiatry, 60*(3), 314–324. https://doi.org/10.1111/jcpp.12978.

Wolff, J. J., Swanson, M. R., Elison, J. T., Gerig, G., Pruett, J. R., Styner, M. A., et al. (2017). Neural circuitry at age 6 months associated with later repetitive behavior and sensory responsiveness in autism. *Molecular Autism, 8*(1), 8. https://doi.org/10.1186/s13229-017-0126-z.

Chapter 7
Sex/Gender and Repetitive and Restrictive Behaviors in Autism Spectrum Disorder

Cory Shulman and Omri Bing

Replication of the observation that autism spectrum disorder (ASD) appears four times as often in males as in females has remained strikingly stable despite evolving diagnostic criteria (Baio et al., 2018; Christensen et al., 2016; Fombonne, 2002; Halladay et al., 2015; Lai, Lombardo, Auyeung, Chakrabarti, & Baron-Cohen, 2015). Males are particularly overrepresented among ASD individuals with average to above-average cognitive ability, among whom estimates of the male to female ratio range from 5.7 to 11:1 (Baird et al., 2006; Fombonne, 2005). Conversely, among those individuals with ASD with moderate to severe intellectual disability (ID), the ratio is closer to 2:1 (Fombonne, 1999; Werling & Geschwind, 2013). As a result of the higher prevalence of ASD in males, females with a clinical diagnosis of ASD and average to above-average intellectual ability tend to be underrepresented both clinically and in research (Halladay et al., 2015). Given the underrepresentation of females with ASD, the clarification of the nature of sex/gender-specific differences in ASD carries with it one of the most compelling prospects for understanding this heterogeneous condition. In this chapter, we will be following Lai et al. (2015) by using the term 'sex/gender' to reflect the awareness that the effects of biological 'sex' and socially constructed 'gender' cannot be easily separated. The present chapter addresses sex/gender differences in restrictive and repetitive behaviors and interests (RRBI), one of the two domains for establishing a diagnosis of ASD. First, we present a review of sex/gender differences in ASD in general,

C. Shulman (✉)
The Autism Center of Hebrew University, The Paul Baerwald School of Social Work and Social Welfare, The Hebrew University of Jerusalem, Jerusalem, Israel
e-mail: cory.shulman@mail.huji.ac.il

O. Bing
The Paul Baerwald School of Social Work and Social Welfare, The Hebrew University of Jerusalem, Jerusalem, Israel

Bob Shapell School of Social Work, Tel Aviv University, Ramat Aviv, Israel

E. Gal, N. Yirmiya (eds.), *Repetitive and Restricted Behaviors and Interests in Autism Spectrum Disorders*, Autism and Child Psychopathology Series,
https://doi.org/10.1007/978-3-030-66445-9_7

followed by a survey of the sex/gender differential in RRBI over the lifespan and in co-occurring conditions, highlighting the effect of age and IQ. We continue with a survey of differences between males and females in diagnosis ascertainment in ASD, ending with recommendations for future research in the area of sex/gender differences in ASD and repetitive and restrictive behaviors and interests.

Sex/Gender Differences in ASD

Since autism emerged as a diagnostic category (Asperger, 1944/1991; Kanner, 1943), the disorder has been mostly observed and depicted in males. Kanner (1943) based his characterization of the disorder on eleven cases, eight of whom were male, while Asperger (1944/1991) based his characterization on four cases all of whom were male. Asperger was fascinated by the male prevalence he observed and tried to explain the sex/gender gap:

> *"How can this be explained? There is certainly a strong hint at a sex-linked or at least sex-limited mode of inheritance. The autistic personality is an extreme variant of male intelligence. Even within the normal variation, we find typical sex differences in intelligence. In general, girls are the better learners. They are more gifted for the concrete and the practical, and for tidy, methodical work. Boys, on the other hand, tend to have a gift for logical ability, abstraction, precise thinking and formulating, and for independent scientific investigation.... In the autistic individual the male pattern is exaggerated to the extreme.... It may be only chance that there are no autistic girls among our cases, or it could be that autistic traits in the female become evident only after puberty. We just do not know." (pp. 84–85)*

Today, 80 years later, the sex/gender gap in the prevalence of ASD is still not well understood.

Over the years a 4:1 male-to-female ratio has been reported consistently in individuals with ASD (Christensen et al., 2016; Fombonne, 1999, 2005; Halladay et al., 2015; Werling & Geschwind, 2013). Some researchers report a lower ratio of approximately 3:1 males to females (Baio, 2014; Kim et al., 2011; Loomes, Hull, & Mandy, 2017), with a surprising 2.3:1 male-to-female ratio reported in a broad population epidemiological review (Mattila et al., 2011). Others report even higher male-to-female ratios of 5.7:1 (Fombonne, 2002) and 9:1 (Brugha et al., 2016). These discrepancies in the sex/gender gap in the prevalence of ASD, which may be partially explained by sampling differences in clinical and epidemiological studies, have led to research attempting to discover potential reasons for the sex/gender gap in the prevalence of ASD. The main areas of research involve investigating the influence of biological factors, ascertainment bias, diagnostic criteria and the instruments used to measure sex/gender differences, in order to understnad the female phenotype which is not captured in the male-based diagnostic criteria of ASD. It may be that autism traits in females and males are distributed differently, manifest differently and at different ages, are concealed by gender specific cultural expectations and/or are more often behaviorally camouflaged in females than in males.

These possibilities and others may contribute to under-referral of females for a clinical diagnosis or a misdiagnosis of females with ASD (Halladay et al., 2015; Lai et al., 2015; Lai, Lombardo, & Baron-Cohen, 2014).

Despite the challenges associated with identifying ASD in females, ongoing research attempts to identify sex/gender differences in ASD symptoms in order to enable clinicians and researchers to diagnose ASD in females. The empirical findings have been equivocal. Some researchers failed to detect sex/gender differences in autism symptomatology (Carter et al., 2007; Holtmann, Bölte, & Poustka, 2007; Mandic-Maravic et al., 2015; Reinhardt, Wetherby, Schatschneider, & Lord, 2015), whereas Hartley and Sikora (2009) and Nicholas et al. (2008) found that females demonstrate fewer repetitive behaviors, while Zwaigenbaum's et al. (2012) research yielded findings which showed that females exhibit fewer impairments in social communication. It has also been reported that females aged three to eighteen years old (Antezana et al., 2019) and adult females (Cohen et al., 2010) demonstrate higher levels of self-injurious behavior (SIB), which has been conceptualized as reflecting stereotypic movements and as such as a manifestation of RRBI in ASD (Bishop et al., 2013; Boyd, McDonough, & Bodfish, 2012; Georgiades, Papageorgiou, & Anagnostou, 2010). The results from other studies indicate that phenotypic differences which are not present at earlier ages emerge later (Halladay et al., 2015; McFayden, Antezana, Albright, Muskett, & Scarpa, 2019; Schroeder et al., 2014; van Wijngaarden-Cremers et al., 2014).

Different methodologies have been employed in these studies, including quantitative and qualitative paradigms, clinical and epidemiological studies, and methodologies including various informants using different instruments. One example of informant research revealed differences in the descriptions by parents of their male children who were being evaluated for ASD and those of parents of females (Hiller, Young, & Weber, 2014). Parents of girls reported that their daughters were more likely to engage in complex imitation of others, had a strong desire to be liked by peers, and had a more advanced vocabulary than boys, whereas parents of boys reported that their sons tended to have more restricted interests and were more isolated or withdrawn in social settings. These findings suggest that the instruments used to measure autism symptomatology affect the sex/gender differences which emerge (Ratto et al., 2018).

The issues relating to prevalence disproportions between males and females include diverse biological processes which may lead to phenotypic sex/gender differences and an ascertainment bias which may result in an artificially low prevalence in females compared with males. We will also consider sampling bias, which may lead to under-referral and misdiagnosis of females with ASD. Furthermore, because the investigation into sex/gender differences in ASD symptoms has not yielded entirely consistent and clear results, we will address the diagnostic process and the instruments used to identify sex/gender similarities and differences, focusing on repetitive and restricted behaviors and interests.

Biological Factors

Several researchers have examined the genetic, hormonal, and neuro-immune processes which lead to phenotypic sex differences in ASD (see Lai et al., 2015; Werling & Geschwind, 2013, for reviews). A specific attempt to address the differences in the prevalence of ASD between males and females was formulated as the "extreme male brain" theory (Baron-Cohen, 2002). According to this theory, ASD is in fact the radicalization of the normative masculine tendency to systemize. Support for this theory may be found in the findings of genetic research. Zhang et al. (2020) revealed that females diagnosed with ASD have more genetic mutations associated with ASD than males with the equivalent level of autism symptoms. These results, together with those of previous studies, suggest that females who carry a genetic predisposition for ASD need higher environmental impact and/or heredity loading compared to males in order for the disorder to manifest itself (Hallmayer et al., 2011; Lai et al., 2015; Werling & Geschwind, 2013; Zhang et al., 2020).

Diagnostic Ascertainment Bias

Evidence from Shattuck's et al. (2009) epidemiological study indicates that females are diagnosed later than males, and females with average and above-average IQ are diagnosed significantly later than both females with below-average IQ and males with average and above-average IQ. It is possible that an inherent diagnostic ascertainment bias may lead to an artificially low prevalence of ASD diagnosed in females compared with males. For example, because young boys tend to display more repetitive and restrictive behaviors than girls, they may be referred for a diagnostic evaluation earlier than girls (Solomon, Miller, Taylor, Hinshaw, & Carter, 2012; Werling & Geschwind, 2013). It is possible that some of the differences which are noted between young boys and girls diagnosed with ASD are similar to sex/gender patterns seen in typically developing children. Current diagnostic criteria were developed primarily from male-biased samples and may not address differential developmental patterns that are commonly seen in neurotypical boys and girls (Rivet & Matson, 2011), and this may be one of the underpinnings of the under-diagnosis of ASD in females.

Most children in the United States who receive an ASD diagnosis do so around the age of 4 years and 8 months (Center for Disease Control and Prevention, 2018). Kopp and Gillberg (1992) investigated the later diagnosis of girls by examining the profiles of girls who received an ASD diagnosis around the age of ten despite the fact that they were evaluated before the age of six because of developmental or behavioral concerns. The clinicians who saw these girls before the age of six did not identify the behavioral patterns they saw in the assessment as possible early signs of ASD in the female phenotype. Since these girls then went on to receive an ASD

diagnosis by the age of ten, these findings raise the possibility that ASD is not recognized in younger girls referred for neurodevelopmental assessment before the age of six, supporting the hypothesis that some of the differences in prevalence rates of males and females are related to the age of initial diagnosis. Finally, differences in social expectations and requirements of males and females in different cultures may contribute to the diagnostic ascertainment bias between males and females (Nazim & Khalid, 2018). Some of these differences may result in parents and clinicians attributing behavior difficulties of females to shyness more than they do for males (Dean, Harwood, & Kasari, 2017; Hull, Mandy, & Petrides, 2017).

Another factor contributing to differences in diagnostic ascertainment may be the generally greater ability of females to engage in socially appropriate behaviors, as typically developing girls attain early socio-communicative milestones before chronologically age-matched boys (Rose & Rudolph, 2006). This developmental divergence may lead to sex/gender-based differences in the manifestation of ASD and place females at risk for under-diagnosis, particularly among those without co-occurring ID. Recently, this ability to blend in socially has been called the "camouflage effect" (Cage & Burton, 2019; Hull et al., 2017; Lai et al., 2011, 2015), denoting the ability that females may "mask" or "camouflage" their ASD symptoms more successfully than males do (Dworzynski, Ronald, Bolton, & Happé, 2012; Gould & Ashton-Smith, 2011), particularly for the short time frame of a diagnostic session.

Restrictive and Repetitive Behaviors and Interests as a Diagnostic Criterion in ASD

Over the years the criteria for establishing a diagnosis of ASD have changed, but the core symptoms have remained largely the same. The symptoms of ASD have been grouped into two domains (i.e., social communication and repetitive and restrictive behaviors and interests), which are reflected in the fifth edition of the *Diagnostic and Statistical Manual for Mental Disorders* (DSM-5: APA, 2013). The criteria for restrictive and repetitive behaviors and interests include (1) repetitive or stereotyped motor movements or use of objects (e.g., lining up objects, repetitive interest in parts of objects), and/or speech (e.g., echolalia, idiosyncratic speech); (2) insistence on sameness and difficulties with minor changes in routines or rigid patterns of verbal or nonverbal behavior; (3) fixated, narrow interests that are unusual in intensity (e.g., perseverative interest in dinosaurs or anime) or focus (e.g., strong attachment to or preoccupation with unusual objects); and (4) unusual responses to sensory aspects of the environment, including both unusual interest in and seeking of sensory input (e.g., visual fascination with moving objects, excessive smelling or touching of objects) and being highly sensitive or aversive to sensory input (e.g., unable to tolerate ordinary noises or textures). Co-occurring conditions such as attention deficit/hyperactivity disorder (ADHD), anxiety, sleep disorder, and disruptive behavior are to be indicated in the diagnostic formulation as well.

The two domains incorporated into an ASD diagnosis have been found to be independent in the general population as well as in ASD (Happé & Ronald, 2008), yet the conceptualization of ASD as a social disorder resulted in neglecting the investigation of RRBI as central to the understanding of ASD. Recent data suggest that RRBI can help explain the sex/gender differences in ASD (Duvekot et al., 2017; Frazier, Georgiades, Bishop, & Hardan, 2014; McFayden et al., 2019). The symptom variation in RRBI reflects the heterogeneity of ASD presentation, and by focusing on RRBI it may be possible to understand sex/gender differences in the identification and diagnosis of ASD. For example, Duvekot et al. (2017) and Hiller et al. (2014) found that RRBI are less predictive of a diagnosis for females than for males. In fact, it seems possible that females with ASD demonstrate overall lower RRBI, in terms of both severity and frequency (Rutherford et al., 2016; van Wijngaarden-Cremers et al., 2014). The study of RRBI in the identification and diagnosis of ASD in males and females may illuminate similarities as well as differences in the female ASD phenotype, which may help to address the under-diagnosis of ASD in females.

Gould (2017) studied sex/gender differences in the diagnostic process and found that females received an ASD diagnosis at an older average age and that higher levels of restricted and repetitive behaviors, which is highly correlated with an ASD diagnosis in boys, were not significantly correlated with an ASD diagnosis in girls. Girls were more likely to be diagnosed with ASD when they had higher total levels of behavioral problems. Boys tended to be more active and exhibit more interests in technical hobbies and facts, whereas girls were more passive and collected information about people rather than 'projects'. The interests of girls with ASD were often similar to those of typically developing girls (e.g., animals, horses and classical literature). It was not the special interests per se that differentiated them from their peers but rather the quality and intensity of their interests and the time spent on them (Gould & Ashton-Smith, 2011). From these findings it is clear that RRBI which are expressed among girls with ASD are different from those which appear in boys and therefore are not adequately captured by most of the current diagnostic instruments which were standardized with male-biased samples. As a result of the differences in the behavioral manifestations of RRBI in girls, clinicians may be less likely to recognize autism characteristics in girls.

Developmental Trajectories of Restrictive and Repetitive Behaviors and Interests

The manifestations of RRBI have been shown to evolve throughout development, so that they may vary in type and intensity at different ages (Seltzer, Shattuck, Abbeduto, & Greenberg, 2004). Longitudinal research has identified common trajectories in the development of children with ASD and sex/gender differences emerge during different developmental time periods. For example, Halladay et al.

(2015) revealed no sex/gender differences at early ages, with significant sex/gender differences in repetitive behavior appearing only after age 6 years (van Wijngaarden-Cremers et al., 2014). These findings must be interpreted cautiously, taking into consideration sample size and composition, as well as the diagnostic instruments employed.

One explanation for the finding that RRBI symptoms are less evident in young girls is that some of these behaviors overlap with ordinary, somewhat repetitive play in toddlers (Barton, Robins, Jashar, Brennan, & Fein, 2013). This similarity between typical behavior and repetitive patterns also appears in verbally-skilled older children of average or above-average intelligence, in which more subtle deficits and behaviors may be missed if the only information considered concerns the child's interactions with a parent, clinician, or other knowledgeable adult, such as a teacher. While a subset of females may in fact evidence symptoms within the first year of life, a more common pattern of symptom expression involves an early course of rather typical development or mild delays followed by the emergence of ASD-related atypical behaviors, including repetitive behaviors and atypical object exploration (Kim & Lord, 2010; Nadig, Vivanti, & Ozonoff, 2009; Paul, Fuerst, Ramsay, Chawarska, & Klin, 2011). Even though toddlers with ASD may demonstrate more severe and/or frequent repetitive actions with objects, motor mannerisms, sensory interests, and non-speech vocalizations compared with children with developmental delays and typically developing children (Kim & Lord, 2010; Schoen, Paul, & Chawarska, 2011; Watt, Wetherby, Barber, & Morgan, 2008), these behaviors are also sometimes observed in other disorders (Lord, Luyster, Guthrie, & Pickles, 2012). Unfortunately, the reduced frequency and quality or complete absence of these behaviors are commonly more challenging for parents and practitioners to identify than the presence of aberrant behaviors that may be seen as more atypical, disruptive, or interfering.

However, more recent findings indicate that high-risk 12-month-olds who are later diagnosed with ASD show more stereotyped motor mannerisms and repetitive manipulation of objects than their low- and high-risk counterparts who do not go on to develop ASD (Elison et al., 2014). Accordingly, more research is needed to better understand the developmental expression of RRBI in children with and without ASD and how this relates to diagnostic criteria and to sex/gender disproportionalities. Whereas social communication skills are more severely impacted in preschool children with significant language and cognitive delays, it appears that the presence of RRBI is relatively independent of developmental level in early childhood. Bishop, Richler, and Lord (2006) found no significant correlations between restricted and repetitive behaviors and interests and nonverbal cognitive skills in children between 2 and 3 years of age, with the exception of sensory interests. A higher percentage of children with moderate impairment in nonverbal IQ showed unusual sensory interests compared to children with nonverbal IQs over 70. However, after age 3 years relations do emerge between nonverbal IQ and several repetitive behaviors. A negative relation with nonverbal IQ was observed for repetitive uses of objects, unusual sensory interests, hand and finger mannerisms, and complex/full-body mannerisms, while a positive relationship was found for fixated interests and insistence on

sameness in male-dominant samples (Bishop et al., 2013; Kim, Thurm, Shumway, & Lord, 2013).

No sex/gender differences in RRBI among adolescents with ASD (Bölte, Duketis, Poustka, & Holtmann, 2011; Frazier & Hardan, 2017; Holtmann et al., 2007; Solomon et al., 2012) emerged when measured by RRB domain scores on the ADOS-2 (Lord et al., 2012) and the ADI-R (Rutter, Le Couteur, & Lord, 2003). In a recent multisite study of 282 adolescents and adults with ASD without intellectual disability (68 females), Pugliese et al. (2016) reported that females were rated as less impaired on the following ADOS RRB items: use of immediate echolalia and stereotyped and idiosyncratic words or phrases. In contrast, adult females tend to self-report more ASD behaviors on the SRS-2 (Lai et al., 2015; Lehnhardt et al., 2016), underscoring the importance of using self-report measures to evaluate sex/gender differences in ASD symptomatology in higher functioning adults.

Co-Occurring Conditions and Differential Diagnosis in RRBI in ASD

The co-occurrence of two or more clinical diagnoses, known as "comorbidity," is particularly salient in individuals with ASD, since an ASD diagnosis can co-exist with a number of other conditions. These additional problems have a substantial negative impact on functioning (Lecavalier, Leone, & Wiltz, 2006) and must be addressed as a crucial part of the diagnostic evaluation. Several large studies, most based on clinically referred samples, have reported that over 70% of children with ASD were above diagnostic thresholds for another developmental, emotional or behavioral disorder and that over 40% may have two or more comorbid mental health conditions (Gjevik, Eldevik, Fjæran-Granum, & Sponheim, 2011; Joshi et al., 2013; Kaat, Gadow, & Lecavalier, 2013; Simonoff et al., 2008), including intellectual impairment (Rydzewska et al., 2019). In the Repetitive Behavior domain, the child with low IQ may present with multiple motor stereotypies and unusual visual behaviors, while the child with higher IQ may manifest this characteristic more in resistance to changes in routines and preoccupations with unusual topics. Specifically, RRBI overlap with other conditions which must be addressed in order to understand their expression in males and females. Individuals with intellectual disabilities, even those without co-occurring ASD, exhibit motor stereotypies as well as repetitive behavior and sometimes echolalia, all of which are included in the diagnostic criteria of RRBI for ASD. In addition, the insistence on sameness, one of the RRBI in ASD, is a central manifestation of obsessive-compulsive disorder (OCD) as well as a manifestation of anxiety. These three conditions will be addressed in order to emphasize the need for multiple diagnoses as well as the need for differential diagnosis.

Most very young children who undergo an ASD assessment also have some general developmental delay (DD). The complexity of understanding the overlap

between ASD and DD is more pronounced in younger children, among whom distinguishing social-communication limitations above and beyond the global developmental delays is difficult even for the most experienced clinicians (Thurm, Farmer, Salzman, Lord, & Bishop, 2019). The common features of individuals with DD and those with both DD and ASD, above and beyond cognitive and language delays, include difficulties in behavioral regulation, aggressive outbursts, and/or self-injurious behavior, considered to be RRBI. Some children may demonstrate overall delays in reaching play milestones. If play appears developmentally appropriate given age and developmental functioning, the presence of nonfunctional use of objects, repetitive play schemes, and inflexibility in play, all expressions of RRBI, may signify ASD.

Differential diagnosis among school-age children is complicated, as many of the conditions that commonly co-occur with ASD at this age also have substantial symptom overlap. Teasing out the individual's interaction style from shyness to phobia to ASD can often be difficult without assessing all environments in which he or she interacts with others. Generalized anxiety disorder (GAD), social phobia, obsessive-compulsive disorder (OCD), depression, and eating disorders have all been found to be related to ASD (Oldershaw, Treasure, Hambrook, Tchanturia, & Schmidt, 2011; Westwood et al., 2016; Westwood, Mandy, & Tchanturia, 2017), although all these have been reported with higher prevalence in females than in males (Solomon et al., 2012). Research has shown that throughout adolescence anxiety problems increase, especially in females with ASD, whereas males tend to have increased levels of depression (Gotham, Brunwasser, & Lord, 2015). A significant proportion of individuals with ASD will develop an anxiety disorder during adolescence. This is more frequently observed in females than in males (Solomon et al., 2012). Restricted and repetitive behavior severity in ASD correlates positively with anxiety severity in cross-sectional surveys (Baribeau et al., 2019). Someone with ASD and anxiety may develop routines that seem rigid; however, it is often in order to gain a sense of control and/or to avoid triggers. Alternatively, the anxiety may manifest itself as obsessive questioning or insistence on sameness rather than in rumination or somatic complaints (Kerns et al., 2016), and this may be misinterpreted by professionals who are unfamiliar with ASD presentation, particularly in higher functioning, more capable individuals with ASD. Like older children, adults with ASD have been found to have co-occurring psychiatric conditions at higher rates than the non-ASD population (Deprey & Ozonoff, 2018). Although males and females with ASD reveal a similar pattern of behavior problems to that of typically developing males and females in childhood, with the boys exhibiting more externalizing behaviors and the girls more internalizing behaviors, by adolescence the sex/gender difference no longer exists, with both males and females with ASD manifesting more internalizing behavior problems, specifically anxiety and depression (Solomon et al., 2012; Werling & Geschwind, 2013).

Behaviors necessary for an ASD diagnosis may overlap with characteristics of other developmental and behavioral conditions and it is imperative to address the issue of differential diagnosis in order to understand whether an ASD diagnosis is appropriate or whether another condition better accounts for the profile revealed

during the diagnostic evaluation. The first step in understanding the overlap among symptoms is to obtain an extensive developmental history in order to examine the consistency of symptoms over time and their pervasiveness over contexts (for a review see Mazefsky, Pelphrey, & Dahl, 2012). If ASD is indeed the primary diagnosis, the core challenges in social communication and interaction and the presence of restricted, repetitive, or unusual behaviors would appear to be the major issues to be addressed. This is particularly important for females, for whom the expression of ASD symptomatology may suggest another disorder.

Lidstone et al. (2014) explored the manner in which atypical reactions to sensory stimuli contribute to the association between restricted and repetitive behaviors and anxiety in children with ASD and found that insistence on sameness was significantly associated with anxiety whereas repetitive motor behaviors were not. The relation between anxiety and insistence on sameness was mediated by sensory aversion. No sex/gender differences were found, possibly as a result of sampling bias, as 110 boys and only ten girls were included in this study.

Interest in a potential link between ASD and eating disorders (ED) has increased recently as eating disturbances and/or abnormal eating behaviors such as eating non-foods, selective eating, avoiding new foods, feeding problems, overeating, and polydipsia are commonly noted among individuals with ASD and they are among the diagnostic criteria for some ED (Kahathuduwa et al., 2019; Keen, 2008; Marshall, Hill, Ziviani, & Dodrill, 2014; Råstam, 2008; Williams, Darlymple, & Neal, 2000; Zucker et al., 2007). High levels of autism traits among women with ED have been reported (Courty et al., 2013; Dell'Osso et al., 2018; Gesi et al., 2017), with more cases of ED co-occurring among females with ASD than in males (Courty et al., 2013; Westwood et al., 2016, 2017). Furthermore, a significant proportion of individuals with a clinical ED diagnosis, with no previous diagnosis of ASD, met diagnostic criteria for ASD on the ADOS-2 when tested (Mandy & Tchanturia, 2015; Westwood & Tchanturia, 2017).

The investigation of the overlap between the behavioral phenotypes observed in eating patterns as expressed in ASD and in ED can shed light on the sex/gender differences in RRBI (Råstam, 2008; Zucker et al., 2007). Wallace, Llewellyn, Fildes, and Ronald (2018) found that higher RRBI scores in individuals in the general population were associated with higher rates of eating disturbances. In another population study Van't Hof et al. (2020) documented that autism traits were associated with different types of eating disturbances in males and in females. These findings suggest the possibility of an underlying mechanism for eating disturbances among people with ASD which involve unusual responses to sensory input, fixated interests and insistence on routines, all expressions of RRBI (Bitsika & Sharpley, 2018; Chistol et al., 2018; Marshall et al., 2014; Råstam, 2008). For example, both avoidance of the sensory stimulation associated with certain foods and insistence on sameness may elicit selective eating.

Since ED are much more common among females than among males (National Institute of Mental Health [NIH], n.d.; Kjelsås, Bjørnstrøm, & Götestam, 2004; Makino, Tsuboi, & Dennerstein, 2004), an ascertainment bias may exist. As previously stated, parents and clinicians may attribute difficulties associated with ASD for females to personality traits or to other disorders more than they do for males

(Dean et al., 2017; Hull, Petrides, et al., 2017). Parallel to the under-diagnosis and misdiagnosis of ASD in females, it may be that males suffering from ED are less likely to be diagnosed as a result of significant differences in the way ED expresses itself in males and females, and the fact that ED are most commonly attributed to females (Recio-Barbero et al., 2019; Strother, Lemberg, Stanford, & Turberville, 2012). In order to understand the striking over-representation of ASD symptomatology in individuals with ED, female-specific diagnostic tools must be developed because, at this point, it is not clear whether ASD and ED are co-occurring conditions or whether a differential diagnosis is necessary. Investigating eating disorders as a possible RRBI in females with ASD may help elucidate specific aspects of the female ASD behavioral phenotype.

Evaluation of RRBI

Assessing RRBI as part of the comprehensive evaluation for diagnostic ascertainment of ASD is complex. It is essential to collect information from parents, caregivers, service providers, teachers and physicians, as RRBI may not express themselves in a clinical evaluation although they may be present in other contexts (Adamou, Johnson, & Alty, 2018). Since RRBI may cause significant impairment to individuals with ASD, this information is crucial. Rituals can consume the majority of the waking hours of an individual and interfere with daily family activities. Affected individuals may become anxious, agitated, or disruptive if such behaviors are interrupted (Gordon, 2000). These behaviors can be socially inappropriate and stigmatizing. RRBI have also been shown to interfere with observational learning (Varni, Lovaas, Koegel, & Everett, 1979), attempts to teach play skills (Koegel, Firestone, Kramme, & Dunlap, 1974), responses to auditory stimuli (Lovaas, Litrownik, & Mann, 1971), performance of discrimination tasks (Koegel & Covert, 1972), and environmental exploration (Fornasari et al., 2013; Pierce & Courchesne, 2001). RRBI are most commonly measured by parent report and/or clinician-informed ratings, but can also be measured by self-report in older adolescents and adults. The findings regarding sex/gender differences in RRBI which have emerged from clinical practice and research are presented next.

Autism Diagnostic Observation Schedule – Second Edition (ADOS-2)

The ADOS-2 (Lord, Rutter, et al., 2012) is a semi-structured, standardized assessment in which the quality of social interaction and communication are evaluated in set "presses", activities which encourage interaction and communication to observe those behaviors associated with an ASD diagnosis. In addition, the presence of repetitive and restrictive behaviors and interests including insistence on sameness is assessed during these tasks. The repetitive and restrictive behavior domain on the

diagnostic algorithm is comprised of clinician codes from restrictive interests, sensory behaviors, verbal rituals, compulsions, repetitive behaviors, finger and hand mannerisms, and stereotyped speech that are observed during the 30–75-minute interactive session. When investigating RRBI empirically, the repetitive and restrictive behavior domain scale of the ADOS is commonly used.

Different RRBI items were most sensitive at identifying ASD in each of the five modules. For example, for younger, lower functioning, less verbal children, "unusually repetitive interests or stereotyped behaviors" and "unusual sensory interest in play material/person" were most indicative of an ASD diagnosis. These are exactly the items in which girls have been found to be less impaired than boys (Hiller et al., 2014). For children and young adolescents, "stereotyped/idiosyncratic use of words or phrases" was more sensitive, again an area in which females were found to be less impaired than males. Finally "excessive interest in or references to unusual or highly specific topics or objects or repetitive behavior" was the most sensitive of the RRBI items for older adolescents and adults, and as reported above the areas of interest of females with ASD have been found to be similar to the areas of interest of females without ASD, with the intensity and quality of the interest diagnostically salient, whereas for males the actual area of interest is often different for males with ASD and for those without. When the ADOS-2 was used to investigate sex/gender differences in ASD, females showed significantly fewer ASD symptoms in the repetitive behavior domain (Frazier et al., 2014; Lai et al., 2011). Only a handful of researchers have examined autism symptomatology using the ADOS-2 in female adults with average cognitive abilities. Whereas Wilson et al. (2016) reported no differences between males and females on the restricted and repetitive behavior domain on the ADOS, Pugliese et al. (2016) found that females had fewer RRBI than males when assessed using the ADOS-2.

Autism Diagnostic Interview – Revised (ADI-R)

The ADI-R (Rutter et al., 2003) is a standardized, semi-structured clinical interview for caregivers of children and adults, which contains 93 items and focuses on behaviors associated with ASD. RRBI have been investigated using the ADI-R either by using the RRBI domain of the algorithm or by analyzing specific items to identify specific sex/gender differences. The algorithm items in RRBI include restricted interest, compulsive adherence to routine and rituals, stereotypes/repetitive motor mannerisms, preoccupation with non-functional manipulation of objects and abnormal sensory interest or aversion. ADI-R item analyses revealed that females with ASD showed fewer autism symptoms related to imaginary play, limited interests, and unusual occupations, all of which are associated with RRBI (Beggiato et al., 2017). Despite the finding that in childhood boys had significantly more RRBI on the ADI-R than did girls (Wilson et al., 2016), no significant sex/gender differences in RRBI were revealed in older adolescents and adults with average or

above-average cognitive abilities on the ADI-R items (Lai et al., 2011; Park et al., 2012), which may be the result of the retrospective nature of the interview.

The Social Responsiveness Scale, Second Edition (SRS-2)

The SRS-2 (Constantino & Gruber, 2012) is divided by age and by informant and is comprised of four forms that allow for evaluation from age 2 years, 5 months, through adulthood (i.e., preschool form, school-age form, adult self-report and adult other report). The SRS-2 yields five treatment subscales, only one of which specifically addresses RRBI. The SRS-2 RRBI subscale has been used to investigate sex/gender differences in ASD. Specifically, Ratto et al. (2018) reported that females 6–16 years old with ASD received higher scores on the RRBI subscale of the SRS-2 than males. Similarly, Torske, Nærland, Øie, Stenberg, and Andreassen (2018) reported a tendency for higher RRBI scores on the SRS-2 for females but the differences were not significant, which may have been due to the smaller sample size in their study. Lai et al. (2015) and Lehnhardt et al. (2016) found that adult females tended to report more RRBI than males. These results reveal the utility of the SRS-2 in distinguishing between gender/sex differences in RRBI's and the importance of employing self-report measures alongside other sources of information when investigating sex/gender differences in ASD.

Autism Spectrum Rating Scale (ASRS)

The Autism Spectrum Rating Scale (ASRS; Goldstein & Naglieri, 2009) is an observer-rated scale completed by parents (or similar caregivers) or teachers (or similar professionals) who rate behavioral characteristics of children aged 2–5 years (early childhood form) and older children aged 7–18 years (school-age form). Both forms require the rater to consider behaviors during the past month. The items measure behaviors characteristic of ASD, which are delineated into eleven scales relating to self-regulation, social/communication, adult socialization, attention, emotionality, peer socialization, language, sensory sensitivity, behavioral rigidity, unusual behaviors, and unusual interests. In addition, a short screening version of the ASRS is provided, consisting of 15 items. Scores on the ASRS are particularly salient when examining sex/gender differences in ASD as it is one of the only questionnaires which was standardized on the same numbers of males and females. Empirical findings from the ASRS indicate that ASD repetitive behaviors are significantly more prevalent among males than females throughout childhood. Comparisons of scores on the ASRS indicate that males have significantly higher scores in stereotypical behavior and sensory sensitivity than females (Camodeca, 2019). Surprisingly, the Chinese version of the ASRS revealed significantly higher scores for males on all RRBI subscales, despite having representative sex/gender

standardization samples (Zhou et al., 2017, 2019), reflecting the importance of taking social norms into account when examining RRBI.

Repetitive Behavior Questionnaire 2 (RBQ-2)

The RBQ-2 (Leekam et al., 2007) is a 20-item questionnaire which provides a list of many specific repetitive behaviors observed in the last month. It can be self- or informant-reported, which has the added benefit of providing information from several perspectives. May (2012, 2014) has used this measure to show that significantly fewer RRBI appear in female children and adults than in males matched by chronological and mental ages.

Repetitive Behavior Survey-R (RBS-R)

The RBS-R was developed to assess complex behaviors (Bodfish, Symons, Parker, & Lewis, 2000; Lam & Aman, 2007). The 43 items of the RBS-R are grouped into six subscales, which include (1) stereotyped behavior (apparently purposeless movements or actions that are repeated in a similar manner); (2) self-injurious behavior (movements or actions that cause or have the potential to cause redness, bruising, or other injury to the body, and that are repeated in a similar manner); (3) compulsive behavior (behavior that is repeated and performed according to a rule or involves things being done "just so"); (4) ritualistic behavior (performing activities of daily living in a particular order or manner); (5) sameness behavior (resistance to change, insisting that things stay the same); and (6) restricted behavior (limited range of focus, interest, or activity). The RBS-R is unique in its ability to assess such a variety of RRBI, providing the most detailed information about specific RRBI, and has been used in many studies to evaluate RRBI (Bishop et al., 2013; Lam & Aman, 2007; Mirenda et al., 2010; Schertz, Odom, Baggett, & Sideris, 2016). Solomon et al. (2012) included typically developing boys and girls matched with boys and girls with ASD in their study, and found that boys and girls with ASD scored higher on all subscales of the RBS-R than typically developing children except for the compulsive behavior scale, in which no differences emerged between girls with and without ASD. The results from this study did not reveal any significant sex/gender differences on any of the subscales of the RBS-R, although the boys tended to show higher scores on the restrictive interests scale. In contrast, Antezana et al. (2019) investigated sex/gender differences in RRBI among males and females 3–18 years old and found that females with ASD manifested more RRBI related to insistence on sameness and restricted, compulsive, and self-injurious behaviors, and fewer stereotyped, restricted behaviors, and circumscribed interests than males.

Summary and Directions for Future Research

As presented in this chapter, clear sex/gender differences emerge in RRBI, revealing that females exhibit fewer of the diagnostic RRBI criteria than males do, and yet may show more self-injurious behaviors, compulsive behaviors and insistence on sameness than males. These differences emerged from self- and parent-report measures of RRBI. Identifying sex/gender differences in RRBI depends on the measures used and it is critical to be aware of the biases inherent in different instruments. ASD presentation is evaluated based on a male-dominated clinical phenotype which may lead to a diagnostic bias, resulting in under-diagnosis of females (e.g., Lidstone et al., 2014). Until the female presentation is better appreciated and better reflected by diagnostic measures, this bias may continue. Therefore, when considering sex/gender differences in RRBI it is important to consider the normed sample used in the development of the specific measure.

Nesting research into sex/gender differences in ASD in a developmental perspective can help to understand similarities and differences among males and females with ASD as they develop, as well as comparing the manifestation of RRBI in ASD and in typically developing males and females, as RRBI are mediated by age. The sex/gender differences in RRBI expression reported in this chapter change over the course of development, with relatively few differences appearing in toddlerhood, more significant differences emerging in early childhood, school-age and early adolescence, with changes again occurring in later adolescence and adulthood, when there seems to be a reduction in the sex/gender differences of RRBI. These findings suggest the importance of comparing sex/gender differences across all ages, as there may be age-related variation in the similarities and/or differences among ASD and typical development groups which need further study. Many of these changes are associated with co-occurring conditions which affect the manifestation of RRBI in males and females with ASD.

Diagnosing ASD among females in a timely manner in order to provide them with the understanding and support that can stem from receiving an autism diagnosis is crucial, but it may not happen until widely used diagnostic and screening tools include female normative and clinical samples. Existing instruments for capturing the sex/gender differences in RRBI have not been sex/gender-normed or evaluated with respect to their sex/gender bias and thus may not be adequately evaluating the presentation of autism in females. This is particularly important in the assessment of RRBI, in that both quantitative and qualitative differences have emerged. To better understand the unique profile of RRBI in females, it will be necessary to identify potential diagnostic markers that clinicians and researchers may not be currently assessing. This could be achieved through cognitive interviewing of affected females and their families to determine whether current measures adequately capture symptom presentation. Based on the literature presented in this chapter, it is imperative that clinicians not rely solely on standardized measures when evaluating females, but obtain a thorough and nuanced developmental history addressing the development and expression of RRBI across the lifespan.

The association between sex/gender and ASD raises many questions that still need to be answered. Further research into sex/gender differences in RRBI has the potential to improve understanding of the sex/gender prevalence differential and may grant new insight into the female phenotype. Most of the studies reviewed in this chapter based their findings on research focusing on people clinically diagnosed with ASD. As discussed above, factors such as the variance in ASD prevalence between males and females across different cognitive abilities, ages, ascertainment bias and diagnostic bias may result in many females (especially those with average and above-average cognitive abilities) being overlooked (McFayden et al., 2019; van Wijngaarden-Cremers et al., 2014). Co-occurring conditions such as anxiety, depression, intellectual disability, and more recently eating disorders may also affect our understanding of female RRBI presentation. Until such time as more female-specific instruments are developed, the RRBI female and male profiles may be suffering from confounding information.

Some of the studies reviewed in this chapter had a majority of male participants, and although a male majority is representative of the ASD epidemiology, such sampling reduces the applicability of the results to females (Antezana et al., 2019; Fulceri et al., 2016; Kim & Lord, 2010; Lidstone et al., 2014). Therefore, future research should include large-scale community based and/or epidemiological studies which are comprised of large samples of females with ASD. The research should also include comparison groups of males and females with typical development and comparison groups of people with other disorders, specifically focusing on anxiety, eating disorders and obsessive-compulsive disorders. Such research may shed light on sex/gender differences in ASD in general and specifically in RRBI presentation.

Some researchers (e.g., Matheis, Matson, Hong, & Cervantes, 2019; Rubenstein et al., 2017; Ventola et al., 2006) have pointed out discrepancies between information obtained from parent- and self-reports and information obtained from clinical, observational measures when evaluating differences in RRBI among males and females. Although these discrepancies may be due to parental biases, they may also stem from an inherent bias in the observational procedures, instruments and scoring when assessing RRBI. Furthermore, when measured globally RRBI have been shown to be inadequate in predicting an ASD diagnosis in males and females, but on a phenomenological level, some RRBI sub-domains highlight key phenotypic differences between males and females with ASD (McFayden et al., 2019). This chapter highlighted the differences in assessment of RRBI using measures designed for autism symptomatology (e.g., ADOS-2, ADI-R) and measures specifically designed to assess RRBI (e.g., RBS-R, RBQ-2). It appears that measures that target a broad range of RRBI are better at distinguishing between males and females with ASD. Therefore, future research should examine sex/gender differences in RRBI using informant- and self-report measures, clinical observation and comprehensive RRBI measures.

It is important to note that sex/gender differences with respect to RRBI presentation can be attributed in part to genetic liability (Szatmari et al., 2012) and to neurological and hormonal variations (Antezana et al., 2019). The relations between

certain RRB subtypes and biological disparities in males and females must be investigated more precisely, by including neurological and hormonal measurements in future research. Advances in understanding the biological mechanisms involved in the presentation of RRBI and the sex/gender differences in RRBI will advance diagnostic precision and will help to develop more specific and reliable treatments.

References

Adamou, M., Johnson, M., & Alty, B. (2018). Autism Diagnostic Observation Schedule (ADOS) scores in males and females diagnosed with autism: A naturalistic study. *Advances in Autism, 4*(2), 49–55.

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). Washington, DC: Author.

Antezana, L., Factor, R. S., Condy, E. E., Strege, M. V., Scarpa, A., & Richey, J. A. (2019). Gender differences in restricted and repetitive behaviors and interests in youth with autism. *Autism Research, 12*(2), 274–283.

Asperger, H. (1944/1991). 'Autistic psychopathy' in childhood (U. Frith, Trans.). In U. Frith (Ed.), *Autism and Asperger syndrome* (pp. 37–92). (This chapter is an annotated translation of a German article by Hans Asperger that was published in "Archiv für Psychiatrie und Nervenkrankenheiten," 1944, 117, 76–136. The original also appeared in "Heilpädagogik," Vienna: Springer-Verlag, 1952) Cambridge, MA: Cambridge University Press. https://doi.org/10.1017/CBO9780511526770.002.

Baio, J. (2014). Prevalence of autism spectrum disorder among children aged 8 years—Autism and developmental disabilities monitoring network, 11 Sites, United States, 2010. *CDC MMWR Surveillance Summaries, 63*, 1–21. https://www.cdc.gov/mmwr/preview/mmwrhtml/ss6302a1.htm.

Baio, J., Wiggins, L., Christensen, D. L., Maenner, M. J., Daniels, J., Warren, Z., et al. (2018). Prevalence of autism spectrum disorder among children aged 8 years—autism and developmental disabilities monitoring network, 11 sites, United States, 2014. *MMWR Surveillance Summaries, 67*(6), 1–23.

Baird, G., Simonoff, E., Pickles, A., Chandler, S., Loucas, T., Meldrum, D., & Charman, T. (2006). Prevalence of disorders of the autism spectrum in a population cohort of children in South Thames: The Special Needs and Autism Project (SNAP). *The Lancet, 368*(9531), 210–215.

Baribeau, D. A., Dupuis, A., Paton, T. A., Hammill, C., Scherer, S. W., Schachar, R. J., et al. (2019). Structural neuroimaging correlates of social deficits are similar in autism spectrum disorder and attention-deficit/hyperactivity disorder: Analysis from the POND Network. *Translational Psychiatry, 9*(1), 1–14.

Baron-Cohen, S. (2002). The extreme male brain theory of autism. *Trends in Cognitive Sciences, 6*(6), 248–254. https://doi.org/10.1016/S1364-6613(02)01904-6.

Barton, M. L., Robins, D. L., Jashar, D., Brennan, L., & Fein, D. (2013). Sensitivity and specificity of proposed DSM-5 criteria for autism spectrum disorder in toddlers. *Journal of Autism and Developmental Disorders, 43*(5), 1184–1195.

Beggiato, A., Peyre, H., Maruani, A., Scheid, I., Råstam, M., Amsellem, F., & Delorme, R. (2017). Gender differences in autism spectrum disorders: Divergence among specific core symptoms. *Autism Research, 10*(4), 680–689. https://doi.org/10.1002/aur.1715.

Bishop, S. L., Hus, V., Duncan, A., Huerta, M., Gotham, K., Pickles, A., et al. (2013). Subcategories of restricted and repetitive behaviors in children with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 43*(6), 1287–1297.

Bishop, S. L., Richler, J., & Lord, C. (2006). Association between restricted and repetitive behaviors and nonverbal IQ in children with autism spectrum disorders. *Child Neuropsychology, 12*(4–5), 247–267.

Bitsika, V., & Sharpley, C. F. (2018). Using parent-and self-reports to evaluate eating disturbances in young girls with Autism Spectrum Disorder. *International Journal of Developmental Neuroscience, 65*, 91–98.

Bodfish, J. W., Symons, F. J., Parker, D. E., & Lewis, M. H. (2000). Varieties of repetitive behavior in autism: Comparisons to mental retardation. *Journal of Autism and Developmental Disorders, 30*(3), 237–243.

Bölte, S., Duketis, E., Poustka, F., & Holtmann, M. (2011). Sex differences in cognitive domains and their clinical correlates in higher-functioning autism spectrum disorders. *Autism, 15*(4), 497–511.

Boyd, B. A., McDonough, S. G., & Bodfish, J. W. (2012). Evidence-based behavioral interventions for repetitive behaviors in autism. *Journal of Autism and Developmental Disorders, 42*(6), 1236–1248.

Brugha, T. S., Spiers, N., Bankart, J., Cooper, S. A., McManus, S., Scott, F. J., et al. (2016). Epidemiology of autism in adults across age groups and ability levels. *The British Journal of Psychiatry, 209*(6), 498–503.

Cage, E., & Burton, H. (2019). Gender differences in the first impressions of autistic adults. *Autism Research, 12*(10), 1495–1504.

Camodeca, A. (2019). Description of criterion validity of the Autism Spectrum Rating Scales 6–18 Parent Report: Initial exploration in a large community sample. *Child Psychiatry and Human Development, 50*(6), 987–1001.

Carter, A. S., Black, D. O., Tewani, S., Connolly, C. E., Kadlec, M. B., & Tager-Flusberg, H. (2007). Sex differences in toddlers with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 37*(1), 86–97.

Center for Disease Control and Prevention. (2018). *Autism spectrum disorder (ASD), identified prevalence of autism spectrum disorder ADDM Network 2000–2014 combining data from all sites*. Retrieved December 17, 2018, from https://www.cdc.gov/ncbddd/autism/data.html

Chistol, L. T., Bandini, L. G., Must, A., Phillips, S., Cermak, S. A., & Curtin, C. (2018). Sensory sensitivity and food selectivity in children with autism spectrum disorder. *Journal of Autism and Developmental Disorders, 48*(2), 583–591.

Christensen, D. L., Baio, J., Braun, K. V., Bilder, D., Charles, J., Constantino, J. N., & Yeargin-Allsopp, M. (2016). Prevalence and characteristics of autism spectrum disorder among 4-year-old children in the autism and developmental disabilities monitoring network, 11 Sites, United States, 2012. *MMWR Surveillance Summaries, 65*(3), 1–23. https://doi.org/10.15585/mmwr.ss6503a1.

Cohen, I. L., Tsiouris, J. A., Flory, M. J., Kim, S. Y., Freedland, R., Heaney, G., et al. (2010). A large scale study of the psychometric characteristics of the IBR modified overt aggression scale: Findings and evidence for increased self-destructive behaviors in adult females with autism spectrum disorder. *Journal of Autism and Developmental Disorders, 40*(5), 599–609.

Constantino, J. N., & Gruber, C. P. (2012). *Social responsiveness scale: SRS-2*. Torrance, CA: Western Psychological Services.

Courty, A., Maria, A. S., Lalanne, C., Ringuenet, D., Vindreau, C., Chevallier, C., et al. (2013). Levels of autistic traits in anorexia nervosa: A comparative psychometric study. *BMC Psychiatry, 13*(1), 222–231.

Dean, M., Harwood, R., & Kasari, C. (2017). The art of camouflage: Gender differences in the social behaviors of girls and boys with autism spectrum disorder. *Autism, 21*(6), 678–689. https://doi.org/10.1177/1362361316671845.

Dell'Osso, L., Carpita, B., Gesi, C., Cremone, I. M., Corsi, M., Massimetti, E., et al. (2018). Subthreshold autism spectrum disorder in patients with eating disorders. *Comprehensive Psychiatry, 81*, 66–72.

Deprey, L., & Ozonoff, S. (2018). Assessment of comorbid psychiatric conditions in autism spectrum disorder. In S. Goldstein, J. A. Naglieri, & S. Ozonoff (Eds.), *Assessment of autism spectrum disorders* (pp. 290–317). New York, NY: Guilford Press.

Duvekot, J., van der Ende, J., Verhulst, F. C., Slappendel, G., van Daalen, E., Maras, A., & Greaves-Lord, K. (2017). Factors influencing the probability of a diagnosis of autism spectrum disorder in girls versus boys. *Autism, 21*(6), 646–658. https://doi.org/10.1177/1362361316672178.

Dworzynski, K., Ronald, A., Bolton, P., & Happé, F. (2012). How different are girls and boys above and below the diagnostic threshold for autism spectrum disorders? *Journal of the American Academy of Child & Adolescent Psychiatry, 51*(8), 788–797.

Elison, J. T., Wolff, J. J., Reznick, J. S., Botteron, K. N., Estes, A. M., Gu, H., et al. (2014). Repetitive behavior in 12-month-olds later classified with autism spectrum disorder. *Journal of the American Academy of Child & Adolescent Psychiatry, 53*(11), 1216–1224.

Fombonne, E. (1999). The epidemiology of autism: A review. *Psychological Medicine, 29*(4), 769–786.

Fombonne, E. (2002). Epidemiological trends in rates of autism. *Molecular Psychiatry, 7*(2), S4–S6.

Fombonne, E. (2005). The changing epidemiology of autism. *Journal of Applied Research in Intellectual Disabilities, 18*(4), 281–294.

Fornasari, L., Chittaro, L., Ieronutti, L., Cottini, L., Dassi, S., Cremaschi, S., et al. (2013). Navigation and exploration of an urban virtual environment by children with autism spectrum disorder compared to children with typical development. *Research in Autism Spectrum Disorders, 7*(8), 956–965.

Frazier, T. W., Georgiades, S., Bishop, S. L., & Hardan, A. Y. (2014). Behavioral and cognitive characteristics of females and males with autism in the Simons Simplex Collection. *Journal of the American Academy of Child & Adolescent Psychiatry, 53*(3), 329–340. https://doi.org/10.1016/j.jaac.2013.12.004.

Frazier, T. W., & Hardan, A. Y. (2017). Equivalence of symptom dimensions in females and males with autism. *Autism, 21*(6), 749–759.

Fulceri, F., Narzisi, A., Apicella, F., Balboni, G., Baldini, S., Brocchini, J., et al. (2016). Application of the Repetitive Behavior Scale-Revised–Italian version–in preschoolers with autism spectrum disorder. *Research in Developmental Disabilities, 48*, 43–52.

Georgiades, S., Papageorgiou, V., & Anagnostou, E. (2010). Brief report: Repetitive behaviours in Greek individuals with autism spectrum disorder. *Journal of Autism and Developmental Disorders, 40*(7), 903–906.

Gesi, C., Carmassi, C., Luciano, M., Bossini, L., Ricca, V., Fagiolini, A., & Dell'Osso, L. (2017). Autistic traits in patients with anorexia nervosa, bulimia nervosa or binge eating disorder: A pilot study. *European Psychiatry, 41*(S1), S100–S100.

Gjevik, E., Eldevik, S., Fjæran-Granum, T., & Sponheim, E. (2011). Kiddie-SADS reveals high rates of DSM-IV disorders in children and adolescents with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 41*(6), 761–769.

Goldstein, S., & Naglieri, J. A. (2009). *Autism spectrum rating scales (ASRS)*. North Tonawanda, NY: Multi-Health System.

Gordon, C. T. (2000). Commentary: Considerations on the pharmacological treatment of compulsions and stereotypies with serotonin reuptake inhibitors in pervasive developmental disorders. *Journal of Autism and Developmental Disorders, 30*(5), 437–438.

Gotham, K., Brunwasser, S. M., & Lord, C. (2015). Depressive and anxiety symptom trajectories from school age through young adulthood in samples with autism spectrum disorder and developmental delay. *Journal of the Academy of Child & Adolescent Psychiatry, 54*(5), 369–376.

Gould, J. (2017). Towards understanding the under-recognition of girls and women on the autism spectrum. *Autism, 21*(6), 703–705.

Gould, J., & Ashton-Smith, J. (2011). Missed diagnosis or misdiagnosis? Girls and women on the autism spectrum. *Good Autism Practice (GAP), 12*(1), 34–41.

Halladay, A. K., Bishop, S., Constantino, J. N., Daniels, A. M., Koenig, K., Palmer, K., & Taylor, J. L. (2015). Sex and gender differences in autism spectrum disorder: Summarizing evidence

gaps and identifying emerging areas of priority. *Molecular Autism, 6*(1), 36.1–36.5. https://doi.org/10.1186/s13229-015-0019-y.

Hallmayer, J., Cleveland, S., Torres, A., Phillips, J., Cohen, B., Torigoe, T., & Lotspeich, L. (2011). Genetic heritability and shared environmental factors among twin pairs with autism. *Archives of General Psychiatry, 68*(11), 1095–1102. https://doi.org/10.1001/archgenpsychiatry.2011.76.

Happé, F., & Ronald, A. (2008). The 'fractionable autism triad': A review of evidence from behavioural, genetic, cognitive and neural research. *Neuropsychology Review, 18*(4), 287–304.

Hartley, S. L., & Sikora, D. M. (2009). Sex differences in autism spectrum disorder: An examination of developmental functioning, autistic symptoms, and coexisting behavior problems in toddlers. *Journal of Autism and Developmental Disorders, 39*(12), 1715–1722. https://doi.org/10.1007/s10803-009-0810-8.

Hiller, R. M., Young, R. L., & Weber, N. (2014). Sex differences in autism spectrum disorder based on DSM-5 criteria: Evidence from clinician and teacher reporting. *Journal of Abnormal Child Psychology, 42*(8), 1381–1393.

Holtmann, M., Bölte, S., & Poustka, F. (2007). Autism spectrum disorders: Sex differences in autistic behavior domains and coexisting psychopathology. *Developmental Medicine & Child Neurology, 49*(5), 361–366. https://doi.org/10.1111/j.1469-8749.2007.00361.x.

Hull, L., Mandy, W., & Petrides, K. V. (2017). Behavioural and cognitive sex/gender differences in autism spectrum condition and typically developing males and females. *Autism, 21*(6), 706–727.

Hull, L., Petrides, K. V., Allison, C., Smith, P., Baron-Cohen, S., Lai, M. C., & Mandy, W. (2017). "Putting on my best normal": Social camouflaging in adults with autism spectrum conditions. *Journal of Autism and Developmental Disorders, 47*, 1–16. https://doi.org/10.1007/s10803-017-3166-5.

Joshi, G., Wozniak, J., Petty, C., Martelon, M. K., Fried, R., Bolfek, A., et al. (2013). Psychiatric comorbidity and functioning in a clinically referred population of adults with autism spectrum disorders: A comparative study. *Journal of Autism and Developmental Disorders, 43*(6), 1314–1325.

Kaat, A. J., Gadow, K. D., & Lecavalier, L. (2013). Psychiatric symptom impairment in children with autism spectrum disorders. *Journal of Abnormal Child Psychology, 41*(6), 959–969.

Kahathuduwa, C. N., West, B. D., Blume, J., Dharavath, N., Moustaid-Moussa, N., & Mastergeorge, A. (2019). The risk of overweight and obesity in children with autism spectrum disorders: A systematic review and meta-analysis. *Obesity Reviews, 20*(12), 1667–1679.

Kanner, L. (1943). Autistic disturbances of affective contact. *Nervous Child, 2*(3), 217–250. Retrieved December 11, 2018, from https://www.rescuepost.com/files/library_kanner_1943.pdf.

Keen, D. V. (2008). Childhood autism, feeding problems and failure to thrive in early infancy. *European Child & Adolescent Psychiatry, 17*(4), 209–216.

Kerns, C. M., Rump, K., Worley, J., Kratz, H., McVey, A., Herrington, J., & Miller, J. (2016). The differential diagnosis of anxiety disorders in cognitively-able youth with autism. *Cognitive and Behavioral Practice, 23*(4), 530–547.

Kim, S. H., & Lord, C. (2010). Restricted and repetitive behaviors in toddlers and preschoolers with autism spectrum disorders based on the Autism Diagnostic Observation Schedule (ADOS). *Autism Research, 3*(4), 162–173.

Kim, S. H., Thurm, A., Shumway, S., & Lord, C. (2013). Multisite study of new autism diagnostic interview-revised (ADI-R) algorithms for toddlers and young preschoolers. *Journal of Autism and Developmental Disorders, 43*(7), 1527–1538.

Kim, Y. S., Leventhal, B. L., Koh, Y. J., Fombonne, E., Laska, E., Lim, E. C., & Song, D. H. (2011). Prevalence of autism spectrum disorders in a total population sample. *American Journal of Psychiatry, 168*(9), 904–912. https://doi.org/10.1176/appi.ajp.2011.10101532.

Kjelsås, E., Bjørnstrøm, C., & Götestam, K. G. (2004). Prevalence of eating disorders in female and male adolescents (14–15 years). *Eating Behaviors, 5*(1), 13–25.

Koegel, R. L., & Covert, A. (1972). The relationship pf self-stimulation to learning in autistic children. *Journal of Applied Behavior Analysis, 5*(4), 381–387.

Koegel, R. L., Firestone, P. B., Kramme, K. W., & Dunlap, G. (1974). Increasing spontaneous play by suppressing self-stimulation in autistic children. *Journal of Applied Behavior Analysis, 7*(4), 521–528.

Kopp, S., & Gillberg, C. (1992). Girls with social deficits and learning problems: Autism, atypical Asperger syndrome or a variant of these conditions. *European Child and Adolescent Psychiatry, 1*(2), 89–99.

Lai, M. C., Lombardo, M. V., Auyeung, B., Chakrabarti, B., & Baron-Cohen, S. (2015). Sex/gender differences and autism: Setting the scene for future research. *Journal of the American Academy of Child and Adolescent Psychiatry, 54*(1), 11–24. https://doi.org/10.1016/j.jaac.2014.10.003.

Lai, M. C., Lombardo, M. V., & Baron-Cohen, S. (2014). Autism. *The Lancet, 383*(9920), 896–910.

Lai, M. C., Lombardo, M. V., Pasco, G., Ruigrok, A. N., Wheelwright, S. J., Sadek, S. A., & MRC AIMS Consortium. (2011). A behavioural comparison of male and female adults with high functioning autism spectrum conditions. *PLoS One, 6*(6), e20835. https://doi.org/10.1371/journal.pone.0020835.

Lam, K. S., & Aman, M. G. (2007). The Repetitive Behavior Scale-Revised: Independent validation in individuals with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 37*(5), 855–866.

Lecavalier, L., Leone, S., & Wiltz, J. (2006). The impact of behaviour problems on caregiver stress in young people with autism spectrum disorders. *Journal of Intellectual Disability Research, 50*(3), 172–183.

Leekam, S., Tandos, J., McConachie, H., Meins, E., Parkinson, K., Wright, C., et al. (2007). Repetitive behaviours in typically developing 2-year-olds. *Journal of Child Psychology and Psychiatry, 48*(11), 1131–1138.

Lehnhardt, F. G., Falter, C. M., Gawronski, A., Pfeiffer, K., Tepest, R., Franklin, J., & Vogeley, K. (2016). Sex-related cognitive profile in autism spectrum disorders diagnosed late in life: Implications for the female autistic phenotype. *Journal of Autism and Developmental Disorders, 46*(1), 139–154.

Lidstone, J., Uljarević, M., Sullivan, J., Rodgers, J., McConachie, H., Freeston, M., et al. (2014). Relations among restricted and repetitive behaviors, anxiety and sensory features in children with autism spectrum disorders. *Research in Autism Spectrum Disorders, 8*(2), 82–92.

Loomes, R., Hull, L., & Mandy, W. P. L. (2017). What is the male-to-female ratio in autism spectrum disorder? A systematic review and meta-analysis. *Journal of the American Academy of Child and Adolescent Psychiatry, 56*(6), 466–474. https://doi.org/10.1016/j.jaac.2017.03.013.

Lord, C., Luyster, R., Guthrie, W., & Pickles, A. (2012). Patterns of developmental trajectories in toddlers with autism spectrum disorder. *Journal of Consulting and Clinical Psychology, 80*(3), 477–489.

Lord, C., Rutter, M., DiLavore, P., Risi, S., Gotham, K., & Bishop, S. (2012). *Autism diagnostic observation schedule–2nd edition (ADOS-2)*. Los Angeles, CA: Western Psychological Corporation.

Lovaas, O. I., Litrownik, A., & Mann, R. (1971). Response latencies to auditory stimuli in autistic children engaged in self-stimulatory behavior. *Behaviour Research and Therapy, 9*(1), 39–49.

Makino, M., Tsuboi, K., & Dennerstein, L. (2004). Prevalence of eating disorders: A comparison of Western and non-Western countries. *Medscape General Medicine, 6*(3), 49–73.

Mandic-Maravic, V., Pejovic-Milovancevic, M., Mitkovic-Voncina, M., Kostic, M., Aleksic-Hil, O., Radosavljev-Kircanski, J., et al. (2015). Sex differences in autism spectrum disorders: Does sex moderate the pathway from clinical symptoms to adaptive behavior? *Scientific Reports, 5*(1), 1–8.

Mandy, W., & Tchanturia, K. (2015). Do women with eating disorders who have social and flexibility difficulties really have autism? A case series. *Molecular Autism, 6*(1), 6–16.

Marshall, J., Hill, R. J., Ziviani, J., & Dodrill, P. (2014). Features of feeding difficulty in children with Autism Spectrum Disorder. *International Journal of Speech-Language Pathology, 16*(2), 151–158.

Matheis, M., Matson, J. L., Hong, E., & Cervantes, P. E. (2019). Gender differences and similarities: Autism symptomatology and developmental functioning in young children. *Journal of Autism and Developmental Disorders, 49*(3), 1219–1231.

Mattila, M. L., Kielinen, M., Linna, S. L., Jussila, K., Ebeling, H., Bloigu, R., & Moilanen, I. (2011). Autism spectrum disorders according to DSM-IV-TR and comparison with DSM-5 draft criteria: An epidemiological study. *Journal of the American Academy of Child and Adolescent Psychiatry, 50*(6), 583–592. https://doi.org/10.1016/j.jaac.2011.04.001.

Mazefsky, C. A., Pelphrey, K. A., & Dahl, R. E. (2012). The need for a broader approach to emotion regulation research in autism. *Child Development Perspectives, 6*(1), 92–97.

McFayden, T. C., Antezana, L., Albright, J., Muskett, A., & Scarpa, A. (2019). Sex differences in an autism spectrum disorder diagnosis: Are restricted repetitive behaviors and interests the key? *Review Journal of Autism and Developmental Disorders, 23*(4), 858–868. https://doi.org/10.1007/s40489-019-00183-w.

Mirenda, P., Smith, I. M., Vaillancourt, T., Georgiades, S., Duku, E., Szatmari, P., et al. (2010). Validating the repetitive behavior scale-revised in young children with autism spectrum disorder. *Journal of Autism and Developmental Disorders, 40*(12), 1521–1530.

Nadig, A., Vivanti, G., & Ozonoff, S. (2009). Adaptation of object descriptions to a partner under increasing communicative demands: A comparison of children with and without autism. *Autism Research, 2*(6), 334–347.

National Institute of Mental Health [NIH]. (n.d.). *Eating disorders*. Retrieved April 19, 2020, from https://www.nimh.nih.gov/health/statistics/eating-disorders.shtml

Nazim, A., & Khalid, R. (2018). Assessment of adaptive functioning of children with typical development attending primary schools in Lahore. *International Journal of Social Sciences & Educational Studies, 4*(4), 124–134. https://doi.org/10.23918/ijsses.v4i4p124.

Nicholas, J. S., Charles, J. M., Carpenter, L. A., King, L. B., Jenner, W., & Spratt, E. G. (2008). Prevalence and characteristics of children with autism-spectrum disorders. *Annals of Epidemiology, 18*(2), 130–136.

Oldershaw, A., Treasure, J., Hambrook, D., Tchanturia, K., & Schmidt, U. (2011). Is anorexia nervosa a version of autism spectrum disorders? *European Eating Disorders Review, 19*(6), 462–474.

Park, S., Cho, S. C., Cho, I. H., Kim, B. N., Kim, J. W., Shin, M. S., et al. (2012). Sex differences in children with autism spectrum disorders compared with their unaffected siblings and typically developing children. *Research in Autism Spectrum Disorders, 6*(2), 861–870.

Paul, R., Fuerst, Y., Ramsay, G., Chawarska, K., & Klin, A. (2011). Out of the mouths of babes: Vocal production in infant siblings of children with ASD. *Journal of Child Psychology and Psychiatry, 52*(5), 588–598.

Pierce, K., & Courchesne, E. (2001). Evidence for a cerebellar role in reduced exploration and stereotyped behavior in autism. *Biological Psychiatry, 49*(8), 655–664.

Pugliese, C. E., Anthony, L. G., Strang, J. F., Dudley, K., Wallace, G. L., Naiman, D. Q., & Kenworthy, L. (2016). Longitudinal examination of adaptive behavior in autism spectrum disorders: Influence of executive function. *Journal of Autism and Developmental Disorders, 46*(2), 467–477. https://doi.org/10.1007/s10803-015-2584-5.

Råstam, M. (2008). Eating disturbances in autism spectrum disorders with focus on adolescent and adult years. *Clinical Neuropsychiatry: Journal of Treatment Evaluation, 5*, 31–42.

Ratto, A. B., Kenworthy, L., Yerys, B. E., Bascom, J., Wieckowski, A. T., White, S. W., & Scarpa, A. (2018). What about the girls? Sex-based differences in autistic traits and adaptive skills. *Journal of Autism and Developmental Disorders, 48*(5), 1698–1711.

Recio-Barbero, M., Fuertes-Soriano, S., Cabezas-Garduño, J., López-Atanes, M., Peña-Rotella, A., & Sáenz-Herrero, M. (2019). Delayed diagnosis of an eating disorder in a male patient with superior mesenteric artery syndrome: Results from a case study. *Frontiers in Psychiatry, 10*, 731–749.

Reinhardt, V. P., Wetherby, A. M., Schatschneider, C., & Lord, C. (2015). Examination of sex differences in a large sample of young children with autism spectrum disorder and typical development. *Journal of Autism and Developmental Disorders, 45*(3), 697–706.

Rivet, T. T., & Matson, J. L. (2011). Review of gender differences in core symptomatology in autism spectrum disorders. *Research in Autism Spectrum Disorders, 5*(3), 957–976.

Rose, A. J., & Rudolph, K. D. (2006). A review of sex differences in peer relationship processes: Potential trade-offs for the emotional and behavioral development of girls and boys. *Psychological Bulletin, 132*(1), 98–131.

Rubenstein, E., Pretzel, R. E., Windham, G. C., Schieve, L. A., Wiggins, L. D., DiGuiseppi, C., et al. (2017). The broader autism phenotype in mothers is associated with increased discordance between maternal-reported and clinician-observed instruments that measure child autism spectrum disorder. *Journal of Autism and Developmental Disorders, 47*(10), 3253–3266.

Rutherford, M., McKenzie, K., Johnson, T., Catchpole, C., O'Hare, A., McClure, I., & Murray, A. (2016). Gender ratio in a clinical population sample, age of diagnosis and duration of assessment in children and adults with autism spectrum disorder. *Autism, 20*(5), 628–634. https://doi.org/10.1177/1362361315617879.

Rutter, M., Le Couteur, A., & Lord, C. (2003). *Autism diagnostic interview-revised*. Los Angeles, CA: Western Psychological Services.

Rydzewska, E., Hughes-McCormack, L. A., Gillberg, C., Henderson, A., MacIntyre, C., Rintoul, J., & Cooper, S. A. (2019). Prevalence of sensory impairments, physical and intellectual disabilities, and mental health in children and young people with self/proxy-reported autism: Observational study of a whole country population. *Autism, 23*(5), 1201–1209.

Schertz, H. H., Odom, S. L., Baggett, K. M., & Sideris, J. H. (2016). Parent-reported repetitive behavior in toddlers on the autism spectrum. *Journal of Autism and Developmental Disorders, 46*(10), 3308–3316.

Schoen, E., Paul, R., & Chawarska, K. (2011). Phonology and vocal behavior in toddlers with autism spectrum disorders. *Autism Research, 4*(3), 177–188.

Schroeder, S. R., Marquis, J. G., Reese, R. M., Richman, D. M., Mayo-Ortega, L., Oyama-Ganiko, R., et al. (2014). Risk factors for self-injury, aggression, and stereotyped behavior among young children at risk for intellectual and developmental disabilities. *American Journal on Intellectual and Developmental Disabilities, 119*, 351–370. https://doi.org/10.1352/1944-7558-119.4.351.

Seltzer, M. M., Shattuck, P., Abbeduto, L., & Greenberg, J. S. (2004). Trajectory of development in adolescents and adults with autism. *Mental Retardation and Developmental Disabilities Research Reviews, 10*(4), 234–247.

Shattuck, P. T., Durkin, M., Maenner, M., Newschaffer, C., Mandell, D. S., Wiggins, L., et al. (2009). Timing of identification among children with an autism spectrum disorder: Findings from a population-based surveillance study. *Journal of the American Academy of Child & Adolescent Psychiatry, 48*(5), 474–483.

Simonoff, E., Pickles, A., Charman, T., Chandler, S., Loucas, T., & Baird, G. (2008). Psychiatric disorders in children with autism spectrum disorders: Prevalence, co-morbidity, and associated factors in a population-derived sample. *Journal of the American Academy of Child and Adolescent Psychiatry, 47*(8), 921–929.

Solomon, M., Miller, M., Taylor, S. L., Hinshaw, S. P., & Carter, C. S. (2012). Autism symptoms and internalizing psychopathology in girls and boys with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 42*(1), 48–59.

Strother, E., Lemberg, R., Stanford, S. C., & Turberville, D. (2012). Eating disorders in men: Underdiagnosed, undertreated, and misunderstood. *Eating Disorders, 20*(5), 346–355.

Szatmari, P., Liu, X. Q., Goldberg, J., Zwaigenbaum, L., Paterson, A. D., Woodbury-Smith, M., et al. (2012). Sex differences in repetitive stereotyped behaviors in autism: Implications for genetic liability. *American Journal of Medical Genetics Part B: Neuropsychiatric Genetics, 159*(1), 5–12.

Thurm, A., Farmer, C., Salzman, E., Lord, C., & Bishop, S. (2019). State of the field: Autism versus Intellectual Disability. *Frontiers in Psychiatry, 10*, 526–536.

Torske, T., Nærland, T., Øie, M. G., Stenberg, N., & Andreassen, O. A. (2018). Meta-cognitive aspects of executive function are highly associated with social functioning on parent-rated measures in children with autism spectrum disorder. *Frontiers in Behavioral Neuroscience, 11*, 258. https://doi.org/10.3389/fnbeh.2017.00258.

van Wijngaarden-Cremers, P. J., van Eeten, E., Groen, W. B., van Deurzen, P. A., Oosterling, I. J., & van der Gaag, R. J. (2014). Gender and age differences in the core triad of impairments in autism spectrum disorders: A systematic review and meta-analysis. *Journal of Autism and Developmental Disorders, 44*(3), 627–635. https://doi.org/10.1007/s10803-013-1913-9.

Van't Hof, M., Ester, W. A., Serdarevic, F., van Berckelaer-Onnes, I., Hillegers, M. H., Tiemeier, H., & Jansen, P. W. (2020). The sex-specific association between autistic traits and eating behavior in childhood: An exploratory study in the general population. *Appetite, 147*(147), 104519. https://doi.org/10.1016/j.appet.2019.104519.

Varni, J. W., Lovaas, O. I., Koegel, R. L., & Everett, N. L. (1979). An analysis of observational learning in autistic and normal children. *Journal of Abnormal Child Psychology, 7*(1), 31–43.

Ventola, P. E., Kleinman, J., Pandey, J., Barton, M., Allen, S., Green, J., & Fein, D. (2006). Agreement among four diagnostic instruments for autism spectrum disorders in toddlers. *Journal of Autism and Developmental Disorders, 36*(7), 839–847.

Wallace, G. L., Llewellyn, C., Fildes, A., & Ronald, A. (2018). Autism spectrum disorder and food neophobia: Clinical and subclinical links. *The American Journal of Clinical Nutrition, 108*(4), 701–707.

Watt, N., Wetherby, A. M., Barber, A., & Morgan, L. (2008). Repetitive and stereotyped behaviors in children with autism spectrum disorders in the second year of life. *Journal of Autism and Developmental Disorders, 38*(8), 1518–1533.

Werling, D. M., & Geschwind, D. H. (2013). Sex differences in autism spectrum disorders. *Current Opinion in Neurology, 26*(2), 146–153. https://doi.org/10.1097/WCO.0b013e32835ee548.

Westwood, H., Eisler, I., Mandy, W., Leppanen, J., Treasure, J., & Tchanturia, K. (2016). Using the autism-spectrum quotient to measure autistic traits in anorexia nervosa: A systematic review and meta-analysis. *Journal of Autism and Developmental Disorders, 46*(3), 964–977.

Westwood, H., Mandy, W., & Tchanturia, K. (2017). Clinical evaluation of autistic symptoms in women with anorexia nervosa. *Molecular Autism, 8*(1), 12–21.

Westwood, H., & Tchanturia, K. (2017). Autism spectrum disorder in anorexia nervosa: An updated literature review. *Current Psychiatry Reports, 19*(7), 41–51.

Williams, P. G., Darlymple, N., & Neal, J. (2000). Eating habits of children with autism. *Pediatric Nursing, 26*, 259–264.

Wilson, C. E., Murphy, C. M., McAlonan, G., Robertson, D. M., Spain, D., Hayward, H., et al. (2016). Does sex influence the diagnostic evaluation of autism spectrum disorder in adults? *Autism, 20*(7), 808–819.

Zhang, Y., Li, N., Li, C., Zhang, Z., Teng, H., Wang, Y., ... & Sun, Z. (2020). Genetic evidence of gender difference in autism spectrum disorder supports the female-protective effect. *Translational Psychiatry, 10*(1), 1–10.

Zhou, H., Zhang, L., Zou, X., Luo, X., Xia, K., Wu, L., & Fombonne, E. (2017). Chinese norms for the Autism Spectrum Rating Scale. *Neuroscience Bulletin, 33*(2), 161–167.

Zhou, H., Li, C. P., Huang, Y., Zou, X. B., Luo, X. R., Wu, L. J., ... & Wang, Y. (2019). Reliability and validity of the translated Chinese version of Autism Spectrum Rating Scale (2–5 years). *World Journal of Pediatrics, 15*(1), 49–56.

Zucker, N. L., Losh, M., Bulik, C. M., LaBar, K. S., Piven, J., & Pelphrey, K. A. (2007). Anorexia nervosa and autism spectrum disorders: guided investigation of social cognitive endophenotypes. *Psychological Bulletin, 133*(6), 976–1006.

Zwaigenbaum, L., Bryson, S. E., Szatmari, P., Brian, J., Smith, I. M., Roberts, W., ... & Roncadin, C. (2012). Sex differences in children with autism spectrum disorder identified within a high-risk infant cohort. *Journal of Autism and Developmental Disorders, 42*(12), 2585–2596.

Chapter 8
The Measurement of Restricted and Repetitive Behaviors in Autism Spectrum Disorder

Robyn L. Young and Alliyza Lim

Introduction

The image of the non-verbal child, flapping and spinning, totally absorbed in their own obsessive interests, as described by Kanner (1943), represents the more traditional view of Autism Spectrum Disorder (ASD) and the stereotypical behaviors that accompany this condition. However, with the revision of the Diagnostic and Statistical Manual of Mental Disorders (DSM) in 1994 (4th edition; DSM-IV; American Psychiatric Association, 1994), the spectrum was expanded to include persons with a milder variant of the disorder (i.e. Asperger's syndrome). With this came a broader interpretation of the presentation of these restricted and repetitive behaviors and interests (RRBI) in the current edition of the DSM, the DSM-5 (American Psychiatric Association, 2013). As a result, the DSM-5 now includes heterogenic behaviors such as motor stereotypies, sensory-related behaviors, circumscribed interests, rituals, excessive sensitivity to change and echolalic speech. While it is agreed that these behaviors are pervasive in this condition, and form part of the ASD diagnostic criteria, there remains a lack of consensus regarding a definition of RRBI (Leekam, Prior, & Uljarevic, 2011) and how pervasive these behaviors must be to be considered deviant and of diagnostic significance. This, therefore, creates challenges for researchers and clinicians in designing valid and reliable assessments of RRBI that are sensitive to this disorder, yet specific to ASD. The purpose of this chapter is to operationalize these behaviors and review the currently available tools so that we may determine whether these tools are valid measures of these behaviors.

R. L. Young (✉) · A. Lim
College of Education, Psychology and Social Work, Flinders University, Adelaide, SA, Australia
e-mail: Robyn.young@flinders.edu.au

E. Gal, N. Yirmiya (eds.), *Repetitive and Restricted Behaviors and Interests in Autism Spectrum Disorders*, Autism and Child Psychopathology Series,
https://doi.org/10.1007/978-3-030-66445-9_8

The Identification and Classification of RRBI

The Diagnostic and Statistical Manual of Mental Disorders: Fifth Edition (DSM-5) describes four areas of RRBI including: (a) stereotypical behaviors (Criterion B1); (b) rigidity in thinking, inflexible adherence to routines, or ritualized patterns of behavior (Criterion B2); (c) perseverative interests (Criterion B3); and (d) hyper- or hypo-activity to sensory input or unusual interest in sensory aspects of the environment (Criterion B4; American Psychiatric Association, 2013). The inclusion of (d) above reflects the seminal views of both Kanner (1943) and Asperger (1944) who identified unusual sensory behaviors, such as aversion to loud noises and food sensitivity, as being an integral part of the disorder. Indeed, bizarre responses to the environment formed a diagnostic criterion when Infantile Autism was introduced to the DSM-3 (American Psychiatric Association, 1980) but was removed in subsequent editions until it was reintroduced in its most recent revision in 2013 (5th edition; DSM-5; American Psychiatric Association, 2013).

While this delineation of RRBI seems reasonable, further delineation within each of the criterion is required in order to develop a better operational definition of these behaviors. Factor analyses have divided these behaviors into two core sub-groups based on the requisite level of cognitive skills (Turner, 1999): lower-order behaviors such as repetitive sensory and motor behavior (RSMB) and higher-order behaviors such as rigidity in thinking, interests and routines, collectively referred to as insistence on sameness (IS; Barrett et al., 2015), with the former being more common in younger children and those with intellectual disabilities (Bishop, Richler, & Lord, 2006; Boyd, McDonough, Rupp, Khan, & Bodfish, 2011; Gilchrist et al., 2018; Morgan, Wetherby, & Barber, 2008). For example, referring to DSM-5 criteria, we may see lower-order behaviors such as flapping, spinning, rocking, head-banging and/or toe-walking. Conversely, B1 behavior may manifest as idiosyncratic or repetitive speech. Similarly, B2 behaviors may present as rigidity in daily activities such as eating from the same bowl or distress if the same route is not followed, or alternatively having a strong set of rigid beliefs around issues of social justice.

Another distinction can be based on whether the behaviors are *with* or *without* objects. For example, B1 lower-order behaviors without objects may include pacing, rocking, jumping, spinning, skipping, hand movements, clapping, finger movements (e.g. tapping, twirling and shaking), nail biting, shoulder movements, back arching, toe walking, feet stamping and odd movements of the mouth, eyes, nose or tongue (Goldman et al., 2009). Others involving objects that could be classified within B1 criteria include using an object repeatedly by either swiping, squeezing, spinning, touching, mouthing, licking, pressing, flipping, rubbing, rolling or dropping it or looking at it from an unusual angle (c.f. Allison et al., 2008). Somewhat confusingly, some of these behaviors may also be seen as sensory seeking or avoiding behaviors, and if so, they would be better positioned under B4 criteria. This adds to the difficulty in operationalizing these behaviors if we also address the purpose or function of such behaviors. The repetitive use of speech also permeates each criterion within section B of the DSM-5 (American Psychiatric Association, 2013). For example, one may say

phrases, sounds or words, repeatedly ask the same question (B1; Wing, Leekam, Libby, Gould, & Larcombe, 2002), be rigid in their conversations (B2), continually talk on the same topic (B3; Bangerter et al., 2017), or make odd noises such as whispers, whistling, and growling sounds repeatedly (B4; Enloe & Rapp, 2014).

Using the RRBI elicited from the Autism Diagnostic Interview-Revised (ADI-R; Le Couteur, Lord, & Rutter, 2003; Lord, Rutter, & Le Couteur, 1994), Turner's (1999) categorization of 'higher' and 'lower' level behaviors has been supported. However, these factor structures are broad and heterogenic and may minimize the differences in these repetitive behaviors and whether they are a response to outside stimuli or triggers (e.g. Honey, Leekam, Turner, & McConachie, 2007; Turner, 1999). Although this dichotomous approach considers circumscribed interests as higher-level behaviors, "unusual preoccupations", which are atypical and specific and one of the most commonly identified RRBI particularly in young children, are not considered within this categorization (Young, Brewer, & Pattinson, 2003). To date, there is no consensus as to a clear factor structure of RRBI and thus it is not surprising that no valid measure has been developed (Honey, Rodgers, & McConachie, 2012).

Deviance of RRBI

Once we have identified what the behavior looks like, and which criterion it reflects, we need to determine if it is significantly deviant. Some repetitive behaviors may be so atypical that deviance is assumed (e.g. head banging), but for others the deviance is determined by the intensity, frequency, quantity of individual behaviors, or response to interruption. By definition, deviation refers to deviance from normality, but this may also include its uniqueness and how it might deviate from behavior found in other disorders.

Determining if the behaviors deviate from what might be considered typical requires a broader understanding of these RRBI, how they present, whether they are found in the typical population and an understanding of how they transition across the lifespan. As yet, there are no tools developed that address the range and severity required to reliably determine whether an individual meets any "deviation" criteria and at what age the behavior might be considered to be of clinical significance. Further, although people with ASD, in general, show more of these behaviors than typically developing people (Kern et al., 2007; Klintwall et al., 2011; Leekam, Nieto, Libby, Wing, & Gould, 2007; Talay-Ongan & Wood, 2010; Tomchek & Dunn, 2007), it is not clear if these behaviors are unique to ASD and contribute to the sensitivity and specificity of the diagnosis for individuals with ASD across the spectrum regardless of age and level of functioning. This poses significant difficulty for the development of tools designed to measure these behaviors. Although RRBI are considered to be core diagnostic features of ASD (Kim & Lord, 2010), they are also seen in persons without ASD who are diagnosed with other developmental

disorders and neuropsychological conditions (for reviews see Langen, Durston, Kas, van Engeland, & Staal, 2011; Leekam et al., 2011).

Added to the confusion, there may be differences in our perception of deviance based on the sex of the person presenting the behavior. Empirical findings suggest that males with ASD tend to be more severely affected by RRBI compared to females (Van Wijngaarden-Cremers et al., 2014) and that the presence of certain stereotypies may be predictive of an individual's sex. For example, numerous researchers have suggested that, generally, males with ASD may be more severely affected by stereotypical movement and interaction with objects (e.g. Hiller, Young, & Weber, 2014; Kumazaki et al., 2015; Mandy et al., 2012), while behaviors such as rubbing, scratching or hair pulling may be more prevalent amongst girls with their restricted interests more gender and developmentally appropriate (Attwood et al., 2006; Hiller et al., 2014). Given that current assessment tools were designed based on literature with predominately male samples, it is possible that these do not adequately reflect the subtly different features and patterns of ASD characteristics with which women and girls present (Lai & Baron-Cohen, 2015). The development of future assessment tools should consider this when selecting relevant items.

Measurement

In order to consider the assessment required to measure these behaviors, we must first operationalize these behaviors. Factor analytic studies have not always produced the binary outcome described above (i.e. lower- versus higher-order behaviors) with some proposing up to five different factors (e.g. stereotypy, self-injury, compulsive, ritualistic, and sameness; Bishop et al., 2013). It is not surprising then that the terminology is lacking consistency. Even when describing the same behaviors, the nomenclature is inconsistent. For example, obsessive preoccupations may be referred to as circumscribed interests, abnormal object attachments, or intense interests. The behavioral referents associated with the terminology is also lacking (Bodfish, Symons, Parker, & Lewis, 2000) which causes confusion not only recognizing these behaviors but also how they should be classified. Further, the qualitative and quantitative difference that should be exhibited to consider these behaviors to be of clinical significance is unclear. Few studies have operationalized each of the behaviors and commented on what constitutes deviation from normality. For example, is it unusual to have read Harry Potter more than 10 times? Morgan et al. (2008) state that somehow three appears to be the magical number for deviance when collecting objects in a hand or lining up or stacking objects. Similarly, Stronach and Wetherby (2014) suggest 10 seconds is the amount of time required to not attend to a novel object for the behavior to be considered atypical. In their study, Goldman et al. (2009) noted the lack of validated stereotypy instruments and thus chose to score as stereotypy any apparently purposeless repetitive movement seen at least twice non-contiguously. Figure 8.1 below is an attempt to categorize behaviors that have been mentioned repeatedly in the literature, but the quantity required for deviation remains unknown.

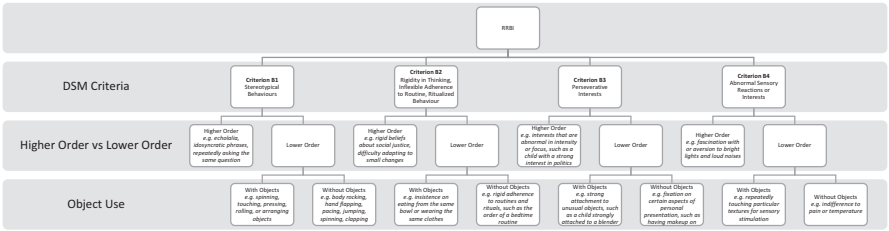

Fig. 8.1 Examples of RRBI by DSM-5 criteria

RRBI Assessment Methods

Several standardized instruments were developed to assess restricted interests and repetitive behaviors. In their systematic review of RRBI in children with ASD, Honey, Rodgers, and McConachie (2012) identified the three most commonly used measures: Repetitive Behavior Scale – Revised (RBS-R), Autism Diagnostic Interview – Revised (ADI-R), and Repetitive Behavior Questionnaire (RBQ). Despite their wide use, all have limitations, including a paucity of psychometric data, limited range of behaviors addressed, limited consideration of deviation from normality in terms of duration and frequency, and limited information relating to the factor structures underpinning them. To date there has been no measure specifically developed and validated against the DSM-5 criteria for ASD. The following section will provide a summary of RRBI assessment methods used in empirical literature, which can be broadly categorized into three main groups: questionnaires, interviews, and direct observation scales. A general overview of the advantages and disadvantages of each method will be discussed first, followed by a review of the various assessment tools available.

Questionnaires

A significant advantage of using a questionnaire over an interview or behavioral observation is that questionnaires are easy, quick, and inexpensive to administer. Because they are completed by the client (or the client's caregiver) and scored according to a published algorithm, no additional training is required to administer a questionnaire. This may be particularly useful in areas where there is limited access to mental health resources. In addition, in contrast to direct observation, self-report or caregiver-report questionnaires are more representative of behaviors that occur in the client's natural environment (Karabekiroglu & Aman, 2009). However, a disadvantage of questionnaires is that they may not be capable of capturing the full range of RRBI present in ASD populations. Given the vast number of behaviors that can be classified as RRBI – from higher-order behaviors such as insistence on sameness to lower-order behaviors such as hand-flapping – it is unlikely that any one questionnaire will be able to adequately measure all possible RRBI. Self-report measures are also susceptible to over-reporting or under-reporting of symptoms by the client.

Interviews

An advantage of using interviews over questionnaires is that interviews allow for the attainment of more qualitative data that can aid in understanding the individual's unique presentation. Furthermore, ratings on semi-structured interviews are often based on clinician judgment, which reduces the impact of over-reporting or under-reporting of symptoms by clients. The biggest disadvantage of clinical interviews, however, is that they are time-consuming and can only be administered by individuals with appropriate levels of training. This may make interviews impractical if the individual needs to be assessed regularly (e.g. to monitor treatment progress) or in areas where mental health resources are limited. The high time commitment also causes interviews to generally cost more per assessment than questionnaires.

Direct Observation Scales

Direct observation scales are structured assessment tools that allow information to be collected on observable behaviors (Newman, 2013). In contrast to questionnaires and interviews that provide indirect accounts of the behavior of an individual from a third party, the strength of direct observation scales is that they allow the clinician to obtain first-hand data about specific behaviors of interest (Newman, 2013). Therefore, an advantage of direct observation scales over questionnaires or interviews is that it reduces the likelihood of error due to misinterpretation of items or responses. Direct observation scales are also less dependent on verbal abilities and may be a more suitable alternative to questionnaires in populations with low rates of literacy. The downside of direct observation scales, however, is that similar to interviews, they are time consuming and require high levels of training to administer, which reduces their practicality and cost-effectiveness. Furthermore, data obtained from direct observation scales may not be an accurate representation of the individual's behavior in the home or school environment, as observation of the individual is limited to only a certain setting (usually the clinician's office) for a certain time frame. Finally, as the name implies, direct observation scales are, by default, only capable of measuring observable behaviors. Thus, higher-order RRBI such as restricted interests and insistence on sameness may not be adequately captured through a direct observation scale.

Emerging Methods of Assessment

With the rapid advancement of technology, it is important to acknowledge new and emerging methods of assessing RRBI. One such method that is being developed is automated detection. A recent study by Gilchrist et al. (2018) evaluated the use of

accelerometers attached to the individual's wrist and torso to detect movement and an algorithm to classify and measure repetitive movements. Only two repetitive behaviors (body rocking and hand flapping) were examined in this study, but it was found that the generalized algorithms were able to achieve 80% sensitivity for body rocking and 93% sensitivity for hand flapping. Although such methods would not be able to measure higher-order RRBI, they allow certain motor stereotypies to be monitored in real-world settings with increased ease and accuracy, and further research in the area is warranted.

RRBI Assessment Tools

The following section will provide an overview of the assessment tools currently available to measure RRBI in infancy, childhood, adulthood, and across the lifespan.

Infancy

Questionnaires

Early Screening for Autistic Traits (ESTAT; Swinkels et al., 2006)

The Early Screening for Autistic Traits (ESTAT) is a 14-item screening measure for children aged over 14 months old. It contains four items assessing RRBI such as motor stereotypies, preoccupations, and reaction to sensory stimuli. While the ESTAT has been found to be able to distinguish children with ASD from typically developing children, it is less sensitive to the differences between children with ASD and children with developmental delays. It is also possible that the ESTAT failed to detect milder forms of ASD, as it was found to have low sensitivity when compared to estimates of ASD prevalence (Dietz, Swinkels, van Daalen, van Engeland, & Buitelaar, 2006).

First Year Inventory (FYI; Baranek, Watson, Crais, & Reznick, 2003)

The First Year Inventory (FYI) is a caregiver-report measure designed to identify children in community samples who are at risk of an eventual ASD diagnosis based on behaviors in the first 12 months of life. The instrument contains 65 items in two overarching domains (Social-Communication and Sensory-Regulatory Functioning) consisting of eight subdomains (Social-Affective Engagement, Imitation, Expressive Communication, Sensory Processing, Regulatory Patterns, Reactivity, and Repetitive Motor Behavior). A longitudinal study by Turner-Brown, Baranek, Reznick, Watson, and Crais (2013) found that 31% of children who met cut-off

scores on the FYI at 12 months old had a diagnosis of ASD at age three, while 85% had a developmental disability or concern. This suggests that the FYI is a promising screening tool for ASD in infants. However, further independent validation of its clinical utility and psychometric properties is required.

Modified Checklist for Autism in Toddlers – Revised, with Follow-Up (M-CHAT-R/F; Robins, Fein, & Barton, 2009)

The Modified Checklist for Autism in Toddlers – Revised, with Follow-Up (M-CHAT-R/F) is a caregiver-report ASD screener for children between 16 and 30 months old. It consists of 20 yes/no questions about the child's behavior and requires parents to complete follow-up questions if the child's score indicates that they are at 'medium risk' of ASD. With regard to the assessment of RRBI, the M-CHAT-R/F includes two items on sensory sensitivities and one item on stereotypical motor behavior. The tool has been validated in a large standardization sample consisting of more than 16,000 toddlers and has been shown to have good predictive validity for ASD diagnosis; children who score over three have a 47.5% chance of being diagnosed with ASD and 94.6% chance of being diagnosed with a developmental delay (Robins et al., 2014).

Repetitive Behavior Scale for Early Childhood (RBS-EC; Wolff, Boyd, & Elison, 2016)

The Repetitive Behavior Scale for Early Childhood (RBS-EC) is a 34-item caregiver-report questionnaire adapted from the Repetitive Behavior Scale – Revised that is designed for use in children aged 17–25 months. It demonstrates good psychometric properties; however, its clinical utility remains unknown.

Quantitative Checklist for Autism in Toddlers (Q-CHAT; Allison et al., 2008)

The Quantitative Checklist for Autism in Toddlers (Q-CHAT) is a 25-item caregiver-report screening measure for ASD, designed for children aged 18–24 months old. The Q-CHAT is a revision of the original CHAT that was designed to improve its sensitivity in detecting children at risk of ASD. Instead of binary yes/no answers, items on the Q-CHAT are rated on a five-point Likert scale to allow for a wider range of responses, in recognition that ASD behaviors occur on a continuum. RRBI assessed by the Q-CHAT include sensory sensitivities, ritualistic behavior, difficulty with change, repetitive motor behavior, and repetitive behavior towards objects. The Q-CHAT has been shown to demonstrate good test-re-test reliability ($r = .82$), moderate internal consistency ($\alpha = .67$), and to discriminate between ASD and non-ASD groups with a large effect size (Allison et al., 2008).

Interviews

Baby and Infant Scale for Children with Autism Traits (BISCUIT Part 1; Matson, Wilkins, & Fodstad, 2011)

The Baby and Infant Scale for Children with Autism Traits (BISCUIT Part 1) is a clinician-rated interview designed to assess for symptoms of ASD in children aged 17–37 months. In an interview with the child's caregiver, 62 items on three factors (Repetitive Behavior/Restricted Interests, Socialization/Nonverbal Communication, and Communication) are rated on a 3-point Likert scale. Of the 62 items, 30 items relate to Repetitive Behavior/Restricted Interests. A significant strength of the BISCUIT-Part 1 is that it is one of the few assessment tools validated for use in very young children.

Direct Observation Scales

Autism Detection in Early Childhood (ADEC; Young, 2007)

The Autism Detection in Early Childhood (ADEC) is a play-based 16-item observation checklist developed to identify ASD in young children between the ages of 12 and 36 months. Of the 16 items, only four reflect behaviors that might identify RRBI. These are ritualistic play, functional play, reaction to common sounds, and task switching. All items were identified from retrospective parental reports (Young et al., 2003) and video analysis (Clifford, Young, & Williamson, 2007) and have been clearly operationalized and thus can be measured reliably. The ADEC's scoring approach helps to capture the finer nuances of the behavior of children with ASD, allowing for the possibility that children at risk of ASD may present with a reduced rate of key behaviors as well as a wide spectrum of autistic behaviors (e.g. Allison et al., 2008). The ADEC shows high concurrent validity, sensitivity, specificity and predictive values. The psychometric properties of the RRBI items in isolation have not, however, been formally assessed.

Autism Observation Scale for Infants (AOSI; Bryson, Zwaigenbaum, McDermott, Rombough, & Brian, 2008)

The Autism Observation Scale for Infants (AOSI) is an 18-item observational measure designed to detect ASD in children aged 6–18 months. Target behaviors are observed and rated by a trained clinician while the child is engaged in a standard set of activities using various toys and objects. RRBI are assessed on two items: Atypical Motor Behaviors (e.g. repetitive motor behaviors) and Atypical Sensory Behaviors (e.g. smelling of toys). The AOSI demonstrated modest test-retest (r = .61 for total scores) and inter-rater (r = .60–.91, depending on the item) reliability (Bryson et al., 2008). Scores on the AOSI were found to correlate with scores

on the Autism Diagnostic Observation Schedule (ADOS) only for children over the age of 12 months, suggesting that the tool is not useful for predicting ASD diagnoses in children under 12 months (Gammer et al., 2015; Zwaigenbaum et al., 2005).

Childhood and Adolescence

Questionnaires

Autism Behavior Checklist (ABC; Krug, Arick, & Almond, 1980)

The Autism Behavior Checklist (ABC) is a 57-item caregiver-report questionnaire for use among school-aged children. It comprises five domains, two of which relate to repetitive behaviors: Body and Object Use, and Sensory Behaviors. The Body and Object Use subscale assesses stereotypical motor movements (e.g. flapping, rocking, walking on toes, twirling), rituals and insistence on sameness (e.g. insistence in keeping certain objects with him/her, and having complicated rituals like lining things up) as well as self-injurious behavior (e.g. head banging, hand biting). The Sensory Behaviors subscale assesses unusual sensory activity such as insensitivity or oversensitivity to noises and light, insensitivity to pain, and non-reactivity to visual stimuli. Items relate to both historical information (e.g. the age at which a symptom was first identified) and current behavioral functioning. When the recommended cut-off scores were used for ASD screening, a significant number of individuals were found to be misclassified, with a high proportion of false negatives (Rellini, Tortolani, Trillo, Carbone, & Montecchi, 2004; Volkmar et al., 1988). Inter-rater reliability of the ABC was also found to be weak (Volkmar et al., 1988).

Autism Spectrum Screening Questionnaire (ASSQ; Ehlers, Gillberg, & Wing, 1999)

The Autism Spectrum Screening Questionnaire (ASSQ) is a 27-item caregiver-report questionnaire designed for use in children and adolescents aged 6–16 years with high-functioning ASD. The ASSQ comprises four subscales: Social Interaction, Communication, Restricted and Repetitive Behavior, and Motor Clumsiness and Other Associated Symptoms. A significant weakness of the ASSQ is that the Restricted and Repetitive Behavior subscale only comprises five items, and thus, does not capture the full range of RRBI demonstrated in ASD populations.

Behavior Flexibility Rating Scale – Revised (BFRS-R; Green et al., 2007)

The Behavior Flexibility Rating Scale – Revised (BFRS-R) is a 16-item caregiver-report questionnaire designed to measure behavioral flexibility and insistence on sameness in children with ASD and other developmental disorders. The extent to

which individuals are able to cope with changes in three types of situations is examined across the three subscales of the BFRS-R: Flexibility Toward Objects (e.g. a commonly used object is misplaced and cannot be found), Flexibility Toward the Environment (e.g. a planned event is delayed or cancelled because of unforeseen circumstances), and Flexibility Toward Persons (e.g. the person becomes separated from his/her family or group). The BFRS-R has been found to demonstrate good internal consistency, inter-rater reliability, and intra-rater reliability when used among children aged 2–17 years (Peters-Scheffer et al., 2007). However, other psychometric properties remain unknown.

Cambridge University Obsessions Questionnaire (Baron-Cohen & Wheelwright, 1999)

The Cambridge University Obsessions Questionnaire is an open-ended caregiver-report questionnaire aimed to obtain information regarding obsessional interests and behaviors in children with ASD. It asks caregivers to indicate and describe whether their child has ever had an obsession in 19 different categories (e.g. collecting things, animals, vehicles). It also provides the opportunity for caregivers to indicate any other obsession displayed by the child that does not fall into one of these categories. A significant strength of the Cambridge University Obsessions Questionnaire is that it allows for the collection of qualitative data from the caregiver, which most questionnaires do not accommodate. However, it was designed as a purely descriptive tool and does not provide any information about the severity, frequency, or functional impact of the obsession. In addition, although the measure includes one category called 'Sensory Experiences,' it largely focuses on restricted interests and does not address other forms of RRBI such as motor stereotypies and insistence on sameness.

Childhood Asperger Syndrome Test (CAST; Scott, Baron-Cohen, Bolton, & Brayne, 2002)

The Childhood Asperger Syndrome Test (CAST) is a 37-item parent-report questionnaire designed to screen for high functioning autism conditions in children aged between 4 and 11 years old. It contains six items that assess RRBI of repetitive motor behavior, restricted or unusual interests, and routines and rituals.

Gilliam Autism Rating Scale – Third Edition (GARS-3; Gilliam, 2014)

The Gilliam Autism Rating Scale – Third Edition (GARS-3) is a 58-item norm-referenced tool designed to screen for ASD in children and adolescents between the ages of 3 and 22 years. It was developed to be completed by the individual's caregiver, teacher, or other professional with high levels of contact with the individual.

Items on the GARS-3 are divided into six subscales: Restricted/Repetitive Behaviors, Social Interaction, Social Communication, Emotional Responses, Cognitive Style, and Maladaptive Speech. Of the 58 items on the GARS-3, 13 are related to Restricted/Repetitive Behaviors. All individuals in the normative sample were diagnosed with ASD, and thus, the scaled score on the Restricted/Repetitive Behavior subscale provides an indication of the severity of the individual's RRBI in relation to other individuals with ASD.

Repetitive Behavior Questionnaire (RBQ; Turner, 1995)

The Repetitive Behavior Questionnaire (RBQ) is a widely used 33-item caregiver-report questionnaire that examines a range of specific repetitive behaviors seen in both typical and atypical populations. When used in a sample of children with ASD aged between 3 and 16 years, the RBQ was found to comprise two factors (insistence on sameness/circumscribed interests and sensory/motor behaviors) and demonstrated good internal consistency and convergent validity (Honey, McConachie, Turner, & Rodgers, 2012). It was also deemed to be suitable for use as an outcome measure of RRBI in children and adolescents with ASD (Scahill et al., 2015). However, the test-retest reliability and sensitivity to change of the RBQ remains unknown (Scahill et al., 2015).

Repetitive Behavior Questionnaire – 2 (RBQ-2; Leekam et al., 2007)

The Repetitive Behavior Questionnaire – 2 (RBQ-2) is a 20-item caregiver-report questionnaire developed to measure repetitive behaviors in children. It was derived from items from the Repetitive Behaviors Questionnaire (RBQ) and the Diagnostic Interview for Social and Communication Disorders (DISCO). The RBQ-2 comprises four subscales: Unusual Sensory Interests, Repetitive Motor Movements, Rigidity/Adherence to Routine, and Preoccupations with Restricted Patterns of Interests, and thus, a strength of the RBQ-2 is that it captures both higher-order and lower-order RRBI. In addition, a notable difference between the RBQ and the RBQ-2 is that the RBQ-2 was designed to be suitable for use in very young children (as young as 24 months). However, a weakness of the RBQ-2 is that its clinical utility remains unknown.

Sameness Questionnaire (Prior & MacMillan, 1973)

The Sameness Questionnaire is a 28-item caregiver-report questionnaire that assesses insistence on sameness in children. Items assess preference for sameness in areas such as placement of furniture, food, and use of cutlery, as well as ritualistic behaviors (e.g. verbal rituals, lining objects up, rituals in bathing and dressing). In a sample of 32 children aged 3–11 years, Prior and MacMillan (1973) found that

children with ASD scored significantly higher on the Sameness Questionnaire than children without ASD. However, little is known about its other psychometric properties.

Sensory Experiences Questionnaire (SEQ; Baranek, David, Poe, Stone, & Watson, 2006)

The Sensory Experiences Questionnaire (SEQ) is a 21-item caregiver-report questionnaire designed to examine the sensory interests of children aged 5 months to 6 years. Items assess hyper- and hypo-sensory response patterns across auditory, visual, vestibular, gustatory/olfactory, and tactile sensory stimuli and whether these sensory experiences occur in a predominantly social context (e.g. physical or eye contact with others) or non-social context (e.g. dislike of textured objects), resulting in four subscales: Hypo-Social, Hyper-Social, Hypo-Non Social, Hyper-Non Social. The SEQ has been found to accurately discriminate the sensory patterns of children with ASD from children with developmental delays and typically developing children (Baranek et al., 2006).

Survey of Favorite Interests and Activities (Smerbeck, 2017)

The Survey of Favorite Interests and Activities is a 53-item caregiver-report questionnaire that aims to assess the functional impact of engaging in restricted interests among children and adolescents with ASD aged between 6 and 17 years. A unique feature of the Survey of Favorite Interests and Activities is that it takes into consideration positive impacts of restricted interests, in addition to negative ones, recognizing that for many individuals with ASD, restricted interests provide substantial benefit. Thus, although this measure does not directly assess the prevalence of RRBI, it may be a useful tool in helping clinicians identify areas of concern associated with restricted interests. A weakness of this measure is that it is limited only to restricted interests and does not provide information on the functional impact of other RRBI, such as motor stereotypies.

Interviews

Children's Yale-Brown Obsessive-Compulsive Scale for ASD (CYBOCS-ASD; Scahill et al., 2014)

The Children's Yale-Brown Obsessive-Compulsive Scale for ASD (CYBOCS-ASD) is a semi-structured clinician-rated interview with the caregiver of a child or adolescent with ASD. It contains a checklist of 39 possible RRBI grouped into eight categories: Washing/Cleaning, Checking, Repeating, Counting, Ordering/Arranging, Hoarding/Saving, Excessive Games/Superstitious Behavior, Rituals

Involving Another Person, and Miscellaneous. A strength of the CYBOCS-ASD is that the severity of the RRBI is rated on five dimensions (Time Spent, Interference, Distress, Resistance, and Control), thereby capturing both the intensity and level of functional impairment associated with the behavior. A significant weakness, however, is that the CYBOCS-ASD was adapted from the original CYBOCS (a measure for OCD), and thus, may not include the full range of RRBI relevant to ASD.

Direct Observation Scales

Childhood Autism Rating Scale – Second Edition (CARS-2; Schopler, Reichler, DeVellis, & Daly, 1980)

The Childhood Autism Rating Scale – Second Edition (CARS-2) is a 15-item clinician-administered observational measure of ASD symptomology for use in individuals aged 2 years or older. RRBI assessed include stereotypical behaviors, sensory sensitivities, resistance to change, and repetitive speech. In a sample of 183 individuals aged 1–62 years old, the CARS-2 was found to demonstrate high sensitivity (84%) and specificity (100%) when used against DSM-5 criteria for ASD (Dawkins, Meyer, & Van Bourgondien, 2016).

Adulthood

Questionnaires

Adult Repetitive Behaviors Questionnaire – 2 (RBQ-2A; Barrett et al., 2015)

The Adult Repetitive Behaviors Questionnaire – 2 (RBQ-2A) is a 20-item self-report questionnaire that examines repetitive motor behaviors and insistence on sameness in adults. It was derived from the Repetitive Behavior Questionnaire – 2, due to the lack of self-report measures of RRBI that are suitable for use in an adult population (Barrett et al., 2015). This is more suitable for adults with high-functioning ASD who are living independently and do not have a caregiver who can accurately report their RRBI (Barrett et al., 2015). Strengths of the RBQ-2A include good psychometric properties when used in an ASD population, and the ability to capture both higher-order and lower-order RRBI. A weakness of the RBQ-2A is that because it was adapted from the RBQ-2 measure for children, it may not contain items that are solely applicable to an adult population (Barrett et al., 2015). In addition, it demonstrates poor psychometric properties when used in a neurotypical population, and its clinical utility in the diagnostic process remains unknown (Barrett et al., 2015).

Special Interest Motivation Scale (SIMS; Grove, Roth, & Hoekstra, 2016)

The Special Interest Motivation Scale (SIMS) is a 20-item self-report measure that examines motivation to engage in special interests among adults with ASD. It comprises five dimensions of motivation, including Personal Life Values and Goals, Intrinsic Interest and Knowledge, Prestige, Engagement, and "Flow" and Achievement. Although the SIMS does not assess RRBI directly, it aids in understanding the reasons why individuals with ASD engage in certain RRBI, which may be useful in the development of intervention plans.

Across the Lifespan

Questionnaires

Aberrant Behavior Checklist (ABC; Aman, Singh, Stewart, & Field, 1985)

The Aberrant Behavior Checklist (ABC) is a 58-item caregiver-report questionnaire that examines five categories of behavior: Irritability, Lethargy/Social Withdrawal, Stereotypic Behavior, Hyperactivity/Noncompliance, and Inappropriate Speech (Kaat, Lecavalier, & Aman, 2014). Although it was originally developed for use among individuals with developmental disabilities in residential care (Aman et al., 1985), the ABC has since been validated for use in toddlers and children with ASD aged 2 years or older (Kaat et al., 2014; Karabekiroglu & Aman, 2009). Since its publication, it has also been translated into over 30 different languages and has been widely used in over 325 studies (Aman, 2012). A strength of the ABC is that the Stereotypic Behavior subscale not only assesses the frequency of RRBI, but also the extent to which the behavior interferes with daily functioning. This, along with its good psychometric properties, sensitivity to change, and low burden on respondents, has led the Stereotypic Behavior subscale to be deemed a suitable outcome measure for RRBI (Scahill et al., 2015). A weakness of the ABC, however, is that the Stereotypic Behavior subscale only examines motor stereotypies and does not include items relating to higher-order RRBI, such as restricted interests or insistence on sameness.

Adult Routines Inventory (ARI) and Childhood Routines Inventory – Revised (CRI-R; Evans, Uljarevic, Lusk, Loth, & Frazier, 2017)

The Adult Routines Inventory (ARI) and the Childhood Routines Inventory – Revised (CRI-R) are a 55-item self-report questionnaire and 62-item caregiver-report questionnaire, respectively, each with two subscales: Repetitive Sensory Motor Behaviors/Compulsions (RSMBC) and Rigidity/Insistence on Sameness (RIS). When used together, the ARI and CRI-R were developed to measure a broad

range of RRBI across the lifespan, from 1 year of age to adulthood (Evans et al., 2017). They were designed for use in both typical and atypical development, and thus, a strength of the ARI and CRI-R is that they can be used across various populations without exhibiting the floor effect. Although they demonstrate good psychometric properties, a weakness of the ARI and CRI-R is that data were validated against self-reported diagnoses rather than formal screening measures.

Autism Behavior Inventory (ABI; Bangerter et al., 2017)

The Autism Behavior Inventory (ABI) is a 93-item caregiver-report web-based questionnaire designed to measure changes in core symptoms of ASD in individuals aged 3 years or older. The ABI consists of five subscales: Social Communication, Restrictive and Repetitive Behaviors, Mental Health, Self-Regulation, and Challenging Behavior. A strength of the ABI is that it was designed to detect changes in symptoms, and therefore, it is useful for the monitoring of treatment outcomes (Bangerter et al., 2017). In addition, the ABI is unique in that it was specifically designed for and validated in the context of online administration. The low burden on respondents and the capacity of the system to send reminders when a rating is due are likely to reduce attrition rates and prevent backdating of questionnaire completion (Bangerter et al., 2017). However, at this time, the process of validation of the ABI is still ongoing and certain psychometric properties of the measure remain unknown.

Repetitive and Restricted Behavior Scale (RRB; Bourreau, Roux, Gomot, Bonnet-Brilhault, & Barthelemy, 2009)

The Repetitive and Restricted Behavior Scale (RRB) is a 35-item caregiver-report questionnaire that aims to measure the full range of RRBI in an ASD population. Thus, a strength of the RRB is that it includes items pertaining to both higher-order RRBI, such as rituals, and lower-order RRBI, such as motor stereotypies. In addition, it does not reference specific behavioral manifestations of RRBI (e.g. "insists on sitting in the same place"); instead, items on the RRB scale reference broader categories of behavior (e.g. "rituals for daily living activities"), thus capturing a broader range of RRBI. However, despite this, it appears that the RRB scale is not sufficient to account for the full complexity of RRBI in the ASD population. In the validation study of the RRB, involving 145 individuals aged 3–33 years, the factor structure of the measure only explained 43% of variance in the data.

Repetitive Behavior Scale – Revised (RBS-R; Bodfish et al., 2000)

The Repetitive Behavior Scale – Revised (RBS-R) is a 43-item caregiver-report questionnaire that examines the frequency and severity of RRBI in six subscales: Stereotyped Behavior, Self-Injurious Behavior, Compulsive Behavior, Routine

Behavior, Sameness Behavior, and Restricted Behavior. Rather than providing categorical diagnostic information, the RBS-R was designed as a quantitative measure of the spectrum of RRBI symptoms (Mirenda et al., 2010). Since its development, it has been validated for use among ASD individuals aged 2–48 years (Lam & Aman, 2007; Mirenda et al., 2010) and has also been deemed appropriate for use as an outcome measure of RRBI in children and adolescents with ASD (Scahill et al., 2015). A strength of the RBS-R is its consistent rating scale across different types of RRBI, in which all behaviors are rated with regard to both frequency and functional impact (Honey, Rodgers, & McConachie, 2012). This is in contrast to certain measures, such as the RBI and RBQ, whereby some behaviors are rated according to frequency, while others are rated according to functional impact (Honey, Rodgers, & McConachie, 2012). However, a weakness of the RBS-R is the lack of statistical evidence to support the proposed six subscales (Honey, Rodgers, & McConachie, 2012).

Social Communication Questionnaire (SCQ; Rutter, Bailey, & Lord, 2008)

The Social Communication Questionnaire (SCQ; formerly known as the Autism Screening Questionnaire) is a 40-item caregiver-report questionnaire that is widely used as an ASD screening tool in individuals aged 4 years and over. The items on the SCQ yield a Total Score and three sub-scores that correspond to the domains of Reciprocal Social Interaction, Communication, and Restricted, Repetitive and Stereotyped Patterns of Behavior. Although the score on the Restricted, Repetitive and Stereotyped Patterns of Behavior subscale can provide some information on the level of severity of RRBI, it should be noted that the main purpose for which the SCQ has been validated is the identification of individuals who may qualify for an ASD diagnosis based on their Total Score on the SCQ. At present, there has been insufficient research on the individual sub-scores of the SCQ, and thus, the value of the SCQ in assessing RRBI independent of other ASD symptoms is unknown.

Social Responsiveness Scale – Second Edition (SRS-2; Constantino & Gruber, 2012)

The Social Responsiveness Scale – Second Edition (SRS-2) is a 65-item self-report or caregiver-report questionnaire designed to screen for ASD in individuals aged 4 years and over. The SRS-2 comes in four forms: Preschool Form (to be completed by a parent or teacher for children aged 2.5–4.5 years), School-Age Form (to be completed by a parent or teacher for children aged 4–18 years), Adult Form (to be completed by a relative or friend for adults aged 19 years and over), and Adult Self-Report Form (a self-report form for adults aged 19 years and over). In addition to the Total Score, items on the SRS-2 can also be grouped into five subscales: Social Awareness, Social Cognition, Social Communication, Social Motivation, and Restricted Interests and Repetitive Behavior. The Restricted Interests and Repetitive

Behavior subscale comprises 12 items that assess stereotypy and circumscribed interests. The SRS-2 demonstrated good psychometric properties and is widely used as a brief screening tool for ASD. However, the psychometric properties and clinical utility of the Restricted Interests and Repetitive Behavior subscale specifically have not been evaluated.

Stereotyped Behavior Scale (SBS; Rojahn, Matlock, & Tasse, 2000)

The Stereotyped Behavior Scale (SBS) is a 24-item caregiver-report questionnaire that examines the frequency and severity of 24 stereotyped behaviors. Although the SBS showed good psychometric properties for use among individuals with intellectual disability, it has not been validated for use specifically among an ASD population. In addition, a weakness of the SBS in assessing RRBI is that it does not include any items pertaining to restricted interests or insistence on sameness.

Interviews

Autism Diagnostic Interview – Revised (ADI-R; Le Couteur et al., 2003)

The Autism Diagnostic Interview – Revised (ADI-R) is a semi-structured interview with an individual's caregiver regarding their symptoms of ASD, and can be used to assess both children and adults with a mental age of above 2 years. The ADI-R consists of 93 items on three domains (Language/Communication, Reciprocal Social Interactions, and Restricted, Repetitive and Stereotyped Behaviors and Interests) and requires approximately 90 to 150 minutes to administer. Of the 93 items, 14 items pertain to RRBI and scores from eight of these items are included in the RRB algorithm. The ADI-R has been widely used in empirical research and has been shown to have good psychometric properties. However, because the ADI-R was developed as a diagnostic tool, it may not be able to detect subtle changes in RRBI (Honey, Rodgers, & McConachie, 2012), thus making it inappropriate for use as a measure of treatment progress. Another weakness of the ADI-R is that it is only scored in relation to caregiver ratings of severity, and not frequency, which may limit its utility (Honey, Rodgers, & McConachie, 2012). Finally, because prior training is required in order to administer the ADI-R, it may not be accessible or affordable for all populations.

Behavior Problems Inventory (BPI-01; Rojahn, Matson, Lott, Esbensen, & Smalls, 2001)

The Behavior Problems Inventory (BPI-01) is a semi-structured clinician-rated interview designed to measure behavior problems in individuals with intellectual and developmental disabilities. The BPI-01 consists of 52 items relating to three

domains (Self-Injurious Behavior, Stereotypic Behavior, and Aggressive/Destructive Behavior) that are rated with regard to frequency and severity of the behavior. The BPI-01 was validated for use in a wide age range (14–91 years) and demonstrated good psychometric properties. However, a limitation of the BPI-01 is that it does not contain items pertaining to restricted interests, and its suitability for use specifically in an ASD population remains unknown.

Diagnostic Interview for Social and Communication Disorders (DISCO; Wing et al., 2002)

The Diagnostic Interview for Social and Communication Disorders (DISCO) is a semi-structured caregiver interview designed to obtain information about an individual's developmental history from birth to current age (it can be used for individuals of all ages). RRBI assessed include stereotypical motor behaviors, repetitive routines, insistence on sameness in the environment, attachment to objects, and sensory sensitivities. All items are rated with regard to level of impairment over the lifetime and current level of impairment. Inter-rater reliability was found to be good with inter-rater agreement of over .75 on over 80% of items (Wing et al., 2002). The DISCO was found to demonstrate 89% agreement when compared against the ADI-R, suggesting good convergent validity (Nygren et al., 2009). It also showed good discriminant validity with age and verbal IQ (Leekam, Libby, Wing, Gould, & Taylor, 2002). However, a notable limitation of the DISCO is that it is highly time consuming, consisting of over 300 questions and requiring up to two to three hours to administer.

Yale Special Interests Interview (South, Klin, & Ozonoff, 1999)

The Yale Special Interests Interview is a semi-structured clinician-rated caregiver interview aimed at examining circumscribed interests of individuals diagnosed with high-functioning ASD (South, Ozonoff, & McMahon, 2005). The interview explores both historical and current circumscribed interests across four developmental stages (depending on the age of the individual) – preschool age, school age, adolescence, and adulthood – and examines the extent to which they interfere with personal, family, and social functioning. A strength of the Yale Special Interest Interview is that RRBI are identified by the caregiver in relation to the specific child being assessed, thus allowing a wide variety of RRBI to be examined. In addition, it also enables the assessment of lifetime restricted interests, which can provide useful clinical information with regard to long-term changes in RRBI. This measure has been adapted into a questionnaire called the Yale Special Interests Survey (Klin, Danovitch, Merz, & Volkmar, 2007), which is identical in content but presented in written form.

Direct Observation Scales

Autism Diagnostic Observation Schedule – Second Edition (ADOS-2; Lord et al., 2012)

The Autism Diagnostic Observation Schedule – Second Edition (ADOS-2) is a semi-structured assessment tool that measures communication, social interaction, and restricted and repetitive behaviors in both children and adults suspected of having ASD. Through the use of specific tasks and activities, the ADOS-2 elicits a variety of behaviors consistent with a diagnosis of ASD which are then observed and coded by the examiner. The ADOS-2 has good psychometric properties and has been recognized to be the gold standard assessment tool for the diagnosis of ASD. However, the RRBI domain demonstrated lower internal consistency and test-retest reliability values compared to the other domains, which the authors attribute to the limited opportunity to observe such behaviors during the 40–60-minute administration period (McCrimmon & Rostad, 2014).

Summary

As illustrated above, many tools are available that purport to measure RRBI across a range of different age groups. However, despite this, there is no tool designed specifically for the assessment of RRBI in individuals with ASD that is validated against current DSM-5 diagnostic criteria. Furthermore, the clinical utility of many measures of RRBI have yet to be established.

The sheer diversity of RRBI within the ASD population suggests that it is unlikely that any one measure will be suitable for use for all individuals and all purposes. For example, a particular tool may be excellent at screening for RRBI but not at measuring response to treatment. It may be, therefore, that the reason why limited information is available about the clinical utility of current assessment tools is that the standards by which they are to be evaluated have not been clearly defined. Thus, when developing an assessment tool, it is important for researchers to be clear on the target demographic of the measure and the specific purpose for which it is being developed. These factors must then be taken into consideration during the process of psychometric validation and when selecting a measurement tool for research or clinical practice.

Selecting a Measurement Tool

Given that there is currently no measure of RRBI that has been specifically developed and validated against the DSM-5 criteria for ASD, we propose that in order to answer the question, "*How* do we measure RRBI?" we must first consider the *Why, What, Who, When*, and *Where* of the intended assessment:

- *Why?* Consider the purpose of the assessment. The measurement of RRBI can provide useful clinical information to aid in screening for ASD, diagnosis of ASD, intervention planning, and evaluation of intervention outcomes. However, each of these purposes may require a slightly different approach to the measurement of RRBI in order to obtain the most relevant information. For example, screening for ASD necessitates the use of a measure that is able to sufficiently capture a broad range of RRBI, whereas the evaluation of intervention outcomes would only require the measurement of the specific RRBI that was targeted in the intervention. In addition, a clear understanding of the purpose of the assessment would also inform the choice between a lifetime measure of RRBI and a current measure of RRBI.
- *What?* Consider the operationalization of RRBI for the purpose of the assessment. Given the broad range of behaviors that constitute RRBI, it is no surprise that the way in which these behaviors are quantified also varies substantially, with behaviors being measured in terms of frequency, severity, duration, or level of functional impairment. Thus, a clear operationalization of the target behavior is essential in selecting an appropriate measurement tool. For example, a measure of frequency may be appropriate for motor stereotypies but not for inflexible adherence to routines which may be more suitably assessed in terms of associated functional impairment. In situations where the operationalization of RRBI encompasses a wide range of behaviors, the simultaneous use of several measures may be most appropriate (e.g. Rojahn et al., 2013).
- *Who?* Consider the target population with respect to age, diagnosis, and level of functioning. This is important in determining the practical aspects of the assessment (e.g. self-report versus caregiver-report), but is also essential to ensure that the selected measure has been validated for use in the target demographic. Consider also the level of expertise of the professional administering the assessment. As interviews and direct observation scales typically require higher levels of training to administer than questionnaires, these methods of assessment may not be practical in areas with limited access to mental health resources.
- *When?* Consider when the behavior occurs. If known, information on when the behavior occurs may be useful to consider when selecting a measurement tool. This may include information such as whether the behavior is ongoing or historical, and whether the behavior varies under different environmental conditions. This is important not only to inform the choice of assessment tool (e.g. lifetime measure versus current measure, questionnaire versus direct observation) but also the selection of the most appropriate informant (e.g. self-report versus caregiver-report).
- *Where?* Consider where the assessment will be held and its possible impact on the demonstration of the target RRBI. RRBI may be displayed at different frequencies or intensities in different situational contexts (e.g. at home versus at school) and this possible variation must be taken into account when selecting an appropriate measurement tool. For example, data obtained from the use of a direct observation scale may yield a more accurate representation of the individual's behavior when conducted in the home or school environment as compared to a clinician's office. In contrast, data from a questionnaire or interview are less susceptible to fluctuations associated with the assessment venue.

Future Considerations

The literature contains a plethora of examples as to what we might consider to be RRBI, yet a consensus has, to date, not been reached. Our review of the literature has led to the following operational understanding of RRBI that should be considered during test selection and/or development. For an RRBI to be considered of diagnostic value, the behavior must:

- be atypical (for that age) in either kind, intensity, duration or frequency
- significantly impact everyday life
- be unresponsive to social reinforcers
- cause distress or anxiety if interrupted or changed
- be irrelevant to the context
- be motivated by intrinsic needs

Although there is a gluttony of tools to measure these behaviors, there is no tool designed specifically for the assessment of RRBI in persons with ASD that is validated against the current diagnostic criteria that addresses all of the aforementioned criteria. This is not surprising given the scope of this definition. However, it is argued here that if RRBI are going to form part of diagnostic criteria, the aforementioned criteria must be addressed. Further, and as suggested earlier, some behaviors are atypical by default, but others need to be understood in terms of their deviation from normality. There are no normative data that enables these judgments to be made. For atypical behavior to be identified, we must have a better understanding of the normality of RRBI. There is a need for validated tool(s) to assess RRBI in the ASD population, contrasted with the typical population, across the lifespan validated against the DSM-5 criteria. It is hoped that when developing new tools or refining existing ones, consideration will be given to the conceptualization of RRBI provided above.

References

Allison, C., Baron-Cohen, S., Wheelwright, S., Charman, T., Richler, J., Pasco, G., & Brayne, C. (2008). The Q-CHAT (Quantitative Checklist for Autism in Toddlers): A normally distributed quantitative measure of autistic traits at 18-24 months of age: Preliminary report. *Journal of Autism and Developmental Disorders, 38*, 1414–1425. https://doi.org/10.1007/s10803-007-0509-7.

Aman, M. G. (2012). Aberrant Behavior Checklist: Current identity and future developments. *Clinical and Experimental Pharmacology, 2*, e114. https://doi.org/10.4172/2161-1459.1000e114.

Aman, M. G., Singh, N. N., Stewart, A. W., & Field, C. J. (1985). The Aberrant Behaviour Checklist: A behaviour rating scale for the assessment of treatment effects. *American Journal of Mental Deficiency, 89*, 485–491.

American Psychiatric Association. (1980). *Diagnostic and statistical manual of mental disorders* (3rd ed.). Washington, DC: Author.

American Psychiatric Association. (1994). *Diagnostic and statistical manual of mental disorders* (4th ed.). Washington, DC: Author.

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). Washington, DC: Author.

Asperger, H. (1944). Die autistisehen psychopathen im kindesalter. *Archives of Psychiatry Nervenkrankh, 117*, 76–136. https://doi.org/10.1007/BF01837709.

Attwood, T., Grandin, T., Bolick, T., Faherty, C., Iland, L., Myers, J. M., & Wroble, M. (2006). *Aspergers and girls*. Arlington, TX: Future Horizons.

Bangerter, A., Ness, S., Aman, M. G., Esbensen, A. J., Goodwin, M. S., Dawson, G., . . . Pandina, G. (2017). Autism Behavior Inventory: A novel tool for assessing core and associated symptoms of autism spectrum disorder. *Journal of Child and Adolescent Psychopharmacology, 27*, 814–822. https://doi.org/10.1089/cap.2017.0018.

Baranek, G. T., David, F. J., Poe, M. D., Stone, W. L., & Watson, L. R. (2006). Sensory Experiences Questionnaire: Discriminating sensory features in young children with autism, developmental delays, and typical development. *Journal of Child Psychology and Psychiatry, 47*, 591–601. https://doi.org/10.1111/j.1469-7610.2005.01546.x.

Baranek, G. T., Watson, L. R., Crais, E., & Reznick, S. (2003). *First-year inventory (FYI) 2.0*. Chapel Hill, NC: University of North Carolina.

Baron-Cohen, S., & Wheelwright, S. (1999). 'Obsessions' in children with autism or Asperger syndrome: Content analysis in terms of core domains of cognition. *British Journal of Psychiatry, 175*, 484–490. https://doi.org/10.1192/bjp.175.5.484.

Barrett, S. L., Uljarevic, M., Baker, E. K., Richdale, A. L., Jones, C. R., & Leekam, S. R. (2015). The Adult Repetitive Behaviours Questionnaire-2 (RBQ-2A): A self-report measure of restricted and repetitive behaviours. *Journal of Autism and Developmental Disorders, 45*, 3680–3692. https://doi.org/10.1007/s10803-015-2514-6.

Bishop, S. L., Hus, V., Duncan, A., Huerta, M., Gotham, K., Pickles, A., . . . Lord, C. (2013). Subcategories of restricted and repetitive behaviors in children with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 43*, 1287–1297. https://doi.org/10.1007/s10803-012-1671-0.

Bishop, S. L., Richler, J., & Lord, C. (2006). Association between restricted and repetitive behaviors and nonverbal IQ in children with autism spectrum disorders. *Child Neuropsychology, 12*, 247–267. https://doi.org/10.1080/09297040600630288.

Bodfish, J. W., Symons, F. J., Parker, D. E., & Lewis, M. H. (2000). Varieties of repetitive behavior in autism: Comparisons to mental retardation. *Journal of Autism and Developmental Disorders, 30*, 237–243. https://doi.org/10.1023/a:1005596502855.

Bourreau, Y., Roux, S., Gomot, M., Bonnet-Brilhault, F., & Barthelemy, C. (2009). Validation of the Repetitive and Restricted Behaviour Scale in autism spectrum disorders. *European Child and Adolescent Psychiatry, 18*, 675–682. https://doi.org/10.1007/s00787-009-0028-5.

Boyd, B. A., McDonough, S. G., Rupp, B., Khan, F., & Bodfish, J. W. (2011). Effects of a family-implemented treatment on the repetitive behaviors of children with autism. *Journal of Autism and Developmental Disorders, 41*, 1330–1341. https://doi.org/10.1007/s10803-010-1156-y.

Bryson, S. E., Zwaigenbaum, L., McDermott, C., Rombough, V., & Brian, J. (2008). The Autism Observation Scale for Infants: Scale development and reliability data. *Journal of Autism and Developmental Disorders, 38*, 731–738. https://doi.org/10.1007/s10803-007-0440-y.

Clifford, S., Young, R., & Williamson, P. (2007). Assessing the early characteristics of autistic disorder using video analysis. *Journal of Autism and Developmental Disorders, 37*, 301–313. https://doi.org/10.1007/s10803-006-0160-8.

Constantino, J. N., & Gruber, C. P. (2012). *Social Responsiveness Scale – Second edition (SRS-2)*. Torrance, CA: Western Psychological Services.

Dawkins, T., Meyer, A. T., & Van Bourgondien, M. E. (2016). The relationship between the Childhood Autism Rating Scale: Second edition and clinical diagnosis utilizing the DSM-IV-TR and the DSM-5. *Journal of Autism and Developmental Disorders, 46*, 3361–3368. https://doi.org/10.1007/s10803-016-2860-z.

Dietz, C., Swinkels, S., van Daalen, E., van Engeland, H., & Buitelaar, J. K. (2006). Screening for autistic spectrum disorder in children aged 14-15 months. II: Population screening with the Early Screening of Autistic Traits Questionnaire (ESAT). Design and general findings. *Journal of Autism and Developmental Disorders, 36*, 713–722. https://doi.org/10.1007/s10803-006-0114-1.

Ehlers, S., Gillberg, C., & Wing, L. (1999). A screening questionnaire for Asperger syndrome and other high-functioning autism spectrum disorders in school age children. *Journal of Autism and Developmental Disorders, 29*, 129–141. https://doi.org/10.1023/a:1023040610384.

Enloe, K. A., & Rapp, J. T. (2014). Effects of noncontingent social interaction on immediate and subsequent engagement in vocal and motor stereotypy in children with autism. *Behavior Modification, 38*, 374–391. https://doi.org/10.1177/0145445513514081.

Evans, D. W., Uljarevic, M., Lusk, L. G., Loth, E., & Frazier, T. (2017). Development of two dimensional measures of restricted and repetitive behavior in parents and children. *Journal of the American Academy of Child and Adolescent Psychiatry, 56*, 51–58. https://doi.org/10.1016/j.jaac.2016.10.014.

Gammer, I., Bedford, R., Elsabbagh, M., Garwood, H., Pasco, G., Tucker, L., . . . Team, B. (2015). Behavioural markers for autism in infancy: Scores on the Autism Observational Scale for Infants in a prospective study of at-risk siblings. *Infant Behavior Development, 38,* 107–115. https://doi.org/10.1016/j.infbeh.2014.12.017.

Gilchrist, K. H., Hegarty-Craver, M., Christian, R. B., Grego, S., Kies, A. C., & Wheeler, A. C. (2018). Automated detection of repetitive motor behaviors as an outcome measurement in intellectual and developmental disabilities. *Journal of Autism and Developmental Disorders, 48*, 1458–1466. https://doi.org/10.1007/s10803-017-3408-6.

Gilliam, J. E. (2014). *Gilliam Autism Rating Scale – Third edition (GARS-3)*. Austin, TX: Pro-Ed.

Goldman, S., Wang, C., Salgado, M. W., Greene, P. E., Kim, M., & Rapin, I. (2009). Motor stereotypies in children with autism and other develompental disorders. *Developmental Medicine and Child Neurology, 51*, 30–38. https://doi.org/10.1111/j.1469-8749.2008.03178.

Green, V. A., Sigafoos, J., O'Reilly, M., Pituch, K. A., Didden, R., Lancioni, G. E., & Singh, N. N. (2007). Behavioral flexibility in individuals with autism: Theory, assessment, and intervention. In L. Zhao (Ed.), *Autism research and advances* (pp. 63-77). New York: Nova Science Publishers.

Grove, R., Roth, I., & Hoekstra, R. A. (2016). The motivation for special interests in individuals with autism and controls: Development and validation of the Special Interest Motivation Scale. *Autism Research, 9*, 677–688. https://doi.org/10.1002/aur.1560.

Hiller, R. M., Young, R. L., & Weber, N. (2014). Sex differences in autism spectrum disorder based on DSM-5 criteria: Evidence from clinician and teacher reporting. *Journal of Abnormal Child Psychology, 42*, 1381–1393. https://doi.org/10.1007/s10802-014-9881-x.

Honey, E., Leekam, S., Turner, M., & McConachie, H. (2007). Repetitive behaviour and play in typically developing children and children with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 37*, 1107–1115. https://doi.org/10.1007/s10803-006-0253-4.

Honey, E., McConachie, H., Turner, M., & Rodgers, J. (2012). Validation of the Repetitive Behaviour Questionnaire for use with children with autism spectrum disorder. *Research in Autism Spectrum Disorders, 6*, 355–364. https://doi.org/10.1016/j.rasd.2011.06.009.

Honey, E., Rodgers, J., & McConachie, H. (2012). Measurement of restricted and repetitive behaviour in children with autism spectrum disorder: Selecting a questionnaire or interview. *Research in Autism Spectrum Disorders, 6*, 757–776. https://doi.org/10.1016/j.rasd.2011.10.011.

Kaat, A. J., Lecavalier, L., & Aman, M. G. (2014). Validity of the Aberrant Behavior Checklist in children with autism spectrum disorder. *Journal of Autism and Developmental Disorders, 44*, 1103–1116. https://doi.org/10.1007/s10803-013-1970-0.

Kanner, L. (1943). Autistic disturbances of affective contact. *Nervous Child, 2*, 217–250.

Karabekiroglu, K., & Aman, M. G. (2009). Validity of the Aberrant Behavior Checklist in a clinical sample of toddlers. *Child Psychiatry and Human Development, 40*, 99–110. https://doi.org/10.1007/s10578-008-0108-7.

Kern, J. K., Trivedi, M. H., Grannemann, B. D., Garver, C. R., Johnson, D. G., Andrews, A. A., . . . Schroeder, J. L. (2007). Sensory correlations in autism. *Autism, 11*, 123–134. https://doi.org/10.1177/1362361307075702.

Kim, S. H., & Lord, C. (2010). Restricted and repetitive behaviors in toddlers and preschoolers with autism spectrum disorders based on the Autism Diagnostic Observation Schedule (ADOS). *Autism Research, 3*, 162–173. https://doi.org/10.1002/aur.142.

Klin, A., Danovitch, J. H., Merz, A. B., & Volkmar, F. R. (2007). Circumscribed interests in higher functioning individuals with autism spectrum disorder: An exploratory study. *Research and Practice for Persons with Severe Disabilities, 32*, 89–100. https://doi.org/10.2511/rpsd.32.2.89.

Klintwall, L., Holm, A., Eriksson, M., Carlsson, L. H., Olsson, M. B., Hedvall, A., . . . Fernell, E. (2011). Sensory abnormalities in autism: A brief report. *Research in Developmental Disabilities, 32*, 795–800. https://doi.org/10.1016/j.ridd.2010.10.021.

Krug, D. A., Arick, J. R., & Almond, P. (1980). Behavior checklist for identifying severely handicapped individuals with high levels of autistic behavior. *Journal of Child Psychology and Psychiatry, 21*, 221–229. https://doi.org/10.1111/j.1469-7610.1980.tb01797.x.

Kumazaki, H., Muramatsu, T., Kosaka, H., Fujisawa, T. X., Iwata, K., Tomoda, A., . . . Mimura, M. (2015). Sex differences in cognitive and symptom profiles in children with high functioning autism spectrum disorders. *Research in Autism Spectrum Disorders, 13-14*, 1–7. https://doi.org/10.1016/j.rasd.2014.12.011.

Lai, M. C., & Baron-Cohen, S. (2015). Identifying the lost generation of adults with autism spectrum conditions. *The Lancet Psychiatry, 2*, 1013–1027. https://doi.org/10.1016/s2215-0366(15)00277-1.

Lam, K. S., & Aman, M. G. (2007). The Repetitive Behavior Scale-Revised: Independent validation in individuals with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 37*, 855–866. https://doi.org/10.1007/s10803-006-0213-z.

Langen, M., Durston, S., Kas, M. J., van Engeland, H., & Staal, W. G. (2011). The neurobiology of repetitive behavior: ...And men. *Neuroscience and Biobehavioral Reviews, 35*, 356–365. https://doi.org/10.1016/j.neubiorev.2010.02.005.

Le Couteur, A., Lord, C., & Rutter, M. (2003). *The Autism Diagnostic Interview – Revised (ADI-R)*. Los Angeles: Western Psychological Services.

Leekam, S., Tandos, J., McConachie, H., Meins, E., Parkinson, K., Wright, C., . . . Le Couteur, A. (2007). Repetitive behaviours in typically developing 2-year-olds. *Journal of Child Psychology and Psychiatry, 48,* 1131–1138. https://doi.org/10.1111/j.1469-7610.2007.01778.x.

Leekam, S. R., Libby, S. J., Wing, L., Gould, J., & Taylor, C. (2002). The Diagnostic Interview for Social and Communication Disorders: Algorithms for ICD-10 childhood autism and Wing and Gould autistic spectrum disorder. *Journal of Child Psychology and Psychiatry, 43*, 327–342. https://doi.org/10.1111/1469-7610.00024.

Leekam, S. R., Nieto, C., Libby, S. J., Wing, L., & Gould, J. (2007). Describing the sensory abnormalities of children and adults with autism. *Journal of Autism and Developmental Disorders, 37*, 894–910. https://doi.org/10.1007/s10803-006-0218-7.

Leekam, S. R., Prior, M. R., & Uljarevic, M. (2011). Restricted and repetitive behaviors in autism spectrum disorders: A review of research in the last decade. *Psychological Bulletin, 137*, 562–593. https://doi.org/10.1037/a0023341.

Lord, C., Rutter, M., DiLavore, P. C., Risi, S., Gotham, K., & Bishop, S. (2012). *Autism Diagnostic Observation Schedule, Second edition (ADOS-2) manual (part I): Modules 1–4*. Torrance, CA: Western Psychological Services.

Lord, C., Rutter, M., & Le Couteur, A. (1994). Autism Diagnostic Interview – Revised: A revised version of a diagnostic interview for caregivers of individuals with possible pervasive developmental disorders. *Journal of Autism and Developmental Disorders, 24*, 659–685. https://doi.org/10.1007/BF02172145.

Mandy, W., Chilvers, R., Chowdhury, U., Salter, G., Seigal, A., & Skuse, D. (2012). Sex differences in autism spectrum disorder: Evidence from a large sample of children and adolescents.

Journal of Autism and Developmental Disorders, 42, 1304–1313. https://doi.org/10.1007/s10803-011-1356-0.

Matson, J. L., Wilkins, J., & Fodstad, J. C. (2011). The validity of the Baby and Infant Screen for Children with Autism Traits: Part 1 (BISCUIT: Part 1). *Journal of Autism and Developmental Disorders, 41*, 1139–1146. https://doi.org/10.1007/s10803-010-0973-3.

McCrimmon, A., & Rostad, K. (2014). Test review: Autism Diagnostic Observation Schedule, Second edition (ADOS-2) manual (part II): Toddler module. *Journal of Psychoeducational Assessment, 32*, 88–92. https://doi.org/10.1177/0734282913490916.

Mirenda, P., Smith, I. M., Vaillancourt, T., Georgiades, S., Duku, E., Szatmari, P., . . . Pathways in, A. S. D. S. T. (2010). Validating the Repetitive Behavior Scale – Revised in young children with autism spectrum disorder. *Journal of Autism and Developmental Disorders, 40*, 1521–1530. https://doi.org/10.1007/s10803-010-1012-0.

Morgan, L., Wetherby, A. M., & Barber, A. (2008). Repetitive and stereotyped movements in children with autism spectrum disorders late in the second year of life. *Journal of Child Psychology and Psychiatry, 49*, 826–837. https://doi.org/10.1111/j.1469-7610.2008.01904.x.

Newman, T. (2013). Direct observations scales. In F. R. Volkmar (Ed.), *Encyclopedia of autism spectrum disorders*. New York: Springer. https://doi.org/10.1007/978-1-4419-1698-3_1027.

Nygren, G., Hagberg, B., Billstedt, E., Skoglund, A., Gillberg, C., & Johansson, M. (2009). The Swedish version of the Diagnostic Interview for Social and Communication Disorders (DISCO-10). Psychometric properties. *Journal of Autism and Developmental Disorders, 39*, 730–741. https://doi.org/10.1007/s10803-008-0678-z.

Peters-Scheffer, N., Didden, R., Green, V. A., Sigafoos, J., Korzilius, H., Pituch, K.,... & Lancioni, G. (2007). The behavior flexibility rating scale-revised (BFRS-R): Factor analysis, internal consistency, inter-rater and intra-rater reliability, and convergent validity. *Research in Developmental Disabilities, 29*, 398–407. https://doi.org/10.1016/j.ridd.2007.07.004.

Prior, M., & MacMillan, M. B. (1973). Maintenance of sameness in children with Kanner's syndrome. *Journal of Autism and Childhood Schizophrenia, 3*, 154–167. https://doi.org/10.1007/BF01537990.

Rellini, E., Tortolani, D., Trillo, S., Carbone, S., & Montecchi, F. (2004). Childhood Autism Rating Scale (CARS) and Autism Behavior Checklist (ABC) correspondence and conflicts with DSM-IV criteria in diagnosis of autism. *Journal of Autism and Developmental Disorders, 34*, 703–708. https://doi.org/10.1007/s10803-004-5290-2.

Robins, D. L., Casagrande, K., Barton, M., Chen, C. M., Dumont-Matthieu, T., & Fein, D. (2014). Validation of the Modified Checklist for Autism in Toddlers, Revised with Follow-up (M-CHAT-R/F). *Pediatrics, 133*, 37–45. https://doi.org/10.1542/peds.2013-1813.

Robins, D. L., Fein, D., & Barton, M. (2009). *Modified Checklist for Autism in Toddlers, Revised, with Follow-up (M-CHAT-R/F)*. Self-published.

Rojahn, J., Matlock, S. T., & Tasse, M. J. (2000). The Stereotyped Behavior Scale: Psychometric properties and norms. *Research in Developmental Disabilities, 21*, 437–454. https://doi.org/10.1016/S0891-4222(00)00057-3.

Rojahn, J., Matson, J. L., Lott, D., Esbensen, A. J., & Smalls, Y. (2001). The Behavior Problems Inventory: An instrument for the assessment of self-injury, stereotyped behavior, and aggression/destruction in individuals with developmental disabilities. *Journal of Autism and Developmental Disorders, 31*, 577–588. https://doi.org/10.1023/a:1013299028321.

Rojahn, J., Schroeder, S. R., Mayo-Ortega, L., Oyama-Ganiko, R., LeBlanc, J., Marquis, J., & Berke, E. (2013). Validity and reliability of the Behavior Problems Inventory, the Aberrant Behavior Checklist, and the Repetitive Behavior Scale – Revised among infants and toddlers at risk for intellectual or developmental disabilities: A multi-method assessment approach. *Research in Developmental Disabilities, 34*, 1804–1814. https://doi.org/10.1016/j.ridd.2013.02.024.

Rutter, M., Bailey, A., & Lord, C. (2008). *Social Communication Questionnaire*. Los Angeles: Western Psychological Services.

Scahill, L., Aman, M. G., Lecavalier, L., Halladay, A. K., Bishop, S. L., Bodfish, J. W., . . . Dawson, G. (2015). Measuring repetitive behaviors as a treatment endpoint in youth with autism spectrum disorder. *Autism, 19*, 38–52. https://doi.org/10.1177/1362361313510069.

Scahill, L., Dimitropoulos, A., McDougle, C. J., Aman, M. G., Feurer, I. D., McCracken, J. T., . . . Vitiello, B. (2014). Children's Yale-Brown Obsessive Compulsive Scale in Autism Spectrum Disorder: Component structure and correlates of symptom checklist. *Journal of the American Academy of Child and Adolescent Psychiatry, 53*, 97–107. https://doi.org/10.1016/j.jaac.2013.09.018.

Schopler, E., Reichler, R. J., DeVellis, R. F., & Daly, K. (1980). Toward objective classification of childhood autism: Childhood Autism Rating Scale (CARS). *Journal of Autism and Developmental Disorders, 10*, 91–103. https://doi.org/10.1007/BF02408436.

Scott, F. J., Baron-Cohen, S., Bolton, P., & Brayne, C. (2002). The CAST (Childhood Asperger Syndrome Test). *Autism, 6*, 9–31. https://doi.org/10.1177/1362361302006001003.

Smerbeck, A. (2017). The Survey of Favorite Interests and Activities: Assessing and understanding restricted interests in children with autism spectrum disorder. *Autism, 23*, 247–259. https://doi.org/10.1177/1362361317742140.

South, M., Klin, A., & Ozonoff, S. (1999). *The Yale Special Interests Interview*. Unpublished measure.

South, M., Ozonoff, S., & McMahon, W. M. (2005). Repetitive behavior profiles in Asperger syndrome and high-functioning autism. *Journal of Autism and Developmental Disorders, 35*, 145–158. https://doi.org/10.1007/s10803-004-1992-8.

Stronach, S., & Wetherby, A. M. (2014). Examining restricted and repetitive behaviors in young children with autism spectrum disorder during two observational contexts. *Autism, 18*, 127–136. https://doi.org/10.1177/1362361312463616.

Swinkels, S. H., Dietz, C., van Daalen, E., Kerkhof, I. H., van Engeland, H., & Buitelaar, J. K. (2006). Screening for autistic spectrum in children aged 14 to 15 months. I: The development of the Early Screening of Autistic Traits Questionnaire (ESAT). *Journal of Autism and Developmental Disorders, 36*, 723–732. https://doi.org/10.1007/s10803-006-0115-0.

Talay-Ongan, A., & Wood, K. (2010). Unusual sensory sensitivities in autism: A possible crossroads. *International Journal of Disability, Development and Education, 47*, 201–212. https://doi.org/10.1080/713671112.

Tomchek, S. D., & Dunn, W. (2007). Sensory processing in children with and without autism: A comparative study using the Short Sensory Profile. *American Journal of Occupational Therapy, 61*, 190–200. https://doi.org/10.5014/ajot.61.2.190.

Turner, M. (1995). *Repetitive behaviour and cognitive functioning in autism* (Unpublished doctoral thesis). University of Cambridge, UK.

Turner, M. (1999). Annotation: Repetitive behaviour in autism: A review of psychological research. *Journal of Child Psychology and Psychiatry, 40*, 839–849. https://doi.org/10.1111/1469-7610.00502.

Turner-Brown, L. M., Baranek, G. T., Reznick, J. S., Watson, L. R., & Crais, E. R. (2013). The First Year Inventory: A longitudinal follow-up of 12-month-old to 3-year-old children. *Autism, 17*, 527–540. https://doi.org/10.1177/1362361312439633.

Van Wijngaarden-Cremers, P. J. M., van Eeten, E., Groen, W. B., Van Deurzen, P. A., Oosterling, I. J., & Van der Gaag, R. J. (2014). Gender and age differences in the core triad of impairments in autism spectrum disorders: A systematic review and meta-analysis. *Journal of Autism and Developmental Disorders, 44*, 627–635. https://doi.org/10.1007/s10803-013-1913-9.

Volkmar, F. R., Cicchetti, D. V., Dykens, E., Sparrow, S. S., Leckman, J. F., & Cohen, D. J. (1988). An evaluation of the Autism Behavior Checklist. *Journal of Autism and Developmental Disorders, 18*, 81–97. https://doi.org/10.1007/BF02211820.

Wing, L., Leekam, S. R., Libby, S. J., Gould, J., & Larcombe, M. (2002). The Diagnostic Interview for Social and Communication Disorders: Background, inter-rater reliability and clinical use. *Journal of Child Psychology and Psychiatry, 43*, 307–325. https://doi.org/10.1111/1469-7610.00023.

Wolff, J. J., Boyd, B. A., & Elison, J. T. (2016). A quantitative measure of restricted and repetitive behaviors for early childhood. *Journal of Neurodevelopmental Disorders, 8*, 27. https://doi.org/10.1186/s11689-016-9161-x.

Young, R. L. (2007). *Autism detection in early childhood: ADEC*. Victoria, Australia: Australian Council of Educational Research.

Young, R. L., Brewer, N., & Pattinson, C. (2003). Parental identification of early behavioural abnormalities in children with autistic disorder. *Autism, 7*, 125–143. https://doi.org/10.1177/1362361303007002002.

Zwaigenbaum, L., Bryson, S., Rogers, T., Roberts, W., Brian, J., & Szatmari, P. (2005). Behavioral manifestations of autism in the first year of life. *International Journal of Developmental Neuroscience, 23*, 143–152. https://doi.org/10.1016/j.ijdevneu.2004.05.001.

Chapter 9
Language Comprehension and Speech Production in Young Children with Autism Spectrum Disorder: Psycho-Linguistic Insights on Restricted, Repetitive Behaviors and Interests

Esther Dromi, Alona Oren, and Aviva Mimouni-Bloch

Introduction

Over the last decade, research findings revealed that the great variability that is characteristic of Autism spectrum disorder (ASD) is also manifested in the areas of language comprehension and speech production. According to published estimates, the population of adults with ASD is divided into three subgroups: 15–20% fail to use speech for communication, approximately 30% attain only limited speech and use it mainly to fulfill basic needs, and the remaining 50% obtain good command of the structural aspects of grammar, but still exhibit mild to moderate communicative difficulties (Dromi, 2018; Luyster & Lord, 2009).

While phonology and syntax are generally viewed as less impaired in individuals with ASD, semantics and pragmatics are areas of salient difficulty, that are considered a core deficit in this condition. This impairment seems to persist even in individuals with "optimal outcomes" following language intervention (Suh et al., 2014). A delay in the emergence of first words is among the earliest signs of ASD in young children and the most frequent reason given by parents seeking a diagnosis during the second year of life (Adamson, Romski, & Barton-Hulsey, 2014; Davidson & Ellis Weismer, 2017).

E. Dromi (✉)
Constantiner School of Education, Tel Aviv University, Ramat Aviv, Israel

A. Oren
The Pediatric Neurology and Developmental Unit, Loewenstein Rehabilitation Medical Center, Raanana, Israel

A. Mimouni-Bloch
The Pediatric Neurology and Developmental Unit, Loewenstein Rehabilitation Medical Center, Raanana, Israel

Sackler Faculty of Medicine, Tel Aviv University, Tel-Aviv, Israel

E. Gal, N. Yirmiya (eds.), *Repetitive and Restricted Behaviors and Interests in Autism Spectrum Disorders*, Autism and Child Psychopathology Series,
https://doi.org/10.1007/978-3-030-66445-9_9

Language impairments in individuals with ASD are attributed to a number of reasons: (A) reduced social interest and limited adult-child interaction during infancy, (B) a lack of social learning from adults throughout early childhood (Tomblin, 2011), (C) deficits in Theory of Mind (Astington & Jenkins, 1999; Peterson, Wellman, & Liu, 2005), (D) Weak Central Coherence (Happé & Frith, 2006) and (E) Executive dysfunction (e.g., limitations in working memory, inhibition, goal-oriented behaviors, planning, and self-monitoring). Each of the above factors may contribute to delays in the process of language learning, resulting in an atypical and prolonged language acquisition trajectory (Davidson & Ellis Weismer, 2017; Norbury, 2013).

Research indicates that the prognosis of individuals with ASD with respect to educational, adaptive, and emotional outcomes is closely linked to their linguistic accomplishments during the preschool years (Kuhl et al., 2013; Norbury, 2013). Thus, over the past decade, enormous efforts have been devoted to the study of language comprehension and speech production, to the development of clinical assessment tools for the evaluation of linguistic skills, and to the development of effective early language intervention programs for children with ASD (Dawson et al., 2010; Dromi, 2018).

Co-morbid Language Impairments with ASD

ASD and developmental language disorders (DLD) are seen today as co-morbid conditions. Kjelgaard and Tager-Flusberg (2001) were the first to argue for the need to differentiate between individuals with ASD who have language impairments (ALI-Autism Language Impaired) and those who develop typical language (ALN-Autism Language Normal). The publication of the *Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition* (DSM-5) in 2013, highlighted the need to independently evaluate language abilities, since it is no longer regarded as a defining domain for the diagnosis of ASD. The recognition that not all individuals with ASD show overt language difficulties led to the decision of combining the two communicative categories, both verbal and nonverbal, and the category of social deficits into one broader category, termed Social-Communication Impairments. Post- hoc statistical analyses supported this decision and indicated that items previously belonging to the two separate domains in DSM-IV are, indeed, included under a single factor. Furthermore, the agreement among clinicians has improved due to the use of broader categories and past confusion concerning the existence of verbal and nonverbal specific symptoms has now been eliminated (Lord & Bishop, 2015).

ASD is currently essentially characterized by two broad areas of impairment:(1) Persistent deficits in communication and in social interaction across multiple contexts; and (2) restricted and repetitive patterns of behaviors, interests, or activities (RRBI). Verbal and nonverbal specific items appear in the DSM-V in these two defining domains of impairment. Difficulties in initiating and sustaining eye contact, gaze aversion, restricted gesturing, and difficulties in interpreting facial

expressions and other nonverbal cues appear as item examples in the domain of social –communication impairments. The display of non-functional use of language, such as echolalia, verbal rituals, stereotyped language and memorized speech are now included under the domain of RRBI. The justification for including this last set of linguistic symptoms under RRBI was that these behaviors are not commonly seen in children with typical language development. Moreover, they are often repetitive and distinct from nonverbal gestures and oral productions that serve social and communication purposes.

In the remaining sections of this chapter, we will present research findings on language comprehension and speech production in children with ASD. The following review indicates the need to refer children who are suspected or who have been diagnosed with ASD for a detailed assessment of language comprehension as well as speech production. We argue that the distinction between communication, language and speech is essential for describing the linguistic profile of children with ASD. In the last section of this chapter, the relations between RRBI and the language abilities of children with ASD are discussed.

Language Comprehension: The Foundation for Grammar

During the pre-linguistic period infants utilize bi-directional social mechanisms of communication with parents, or with other main caretakers, that lead to typical strategies for language development (Kuhl et al., 2013; Meltzoff, 2013). The interactive experiences of children with ASD during the pre-linguistic period are radically different from those of typically developing (TD) children, due to their innate biological impairments. Therefore, the question of how children with ASD go about learning language is intriguing.

TD children construct grammatical rules via active processing of the language addressed to them. This process begins very early in infancy and progresses throughout childhood. Controlled experiments show that towards the end of the first year of life, TD infants identify the phonotactic attributes of their native language, identify syllabic units in the input, differentiate between possible words and atypical syllabic combinations and show a preference for listening to their native language over a foreign language (Jusczyk, 1997). Shortly following their first birthday, toddlers begin to connect words with their underlying meanings and respond to simple instructions such as “where is daddy?”, “give me a hand”, etc. (Kuhl et al., 2008).

Comprehending language involves the establishment of a connection between consistent phonological units and cues perceived through visual and other sensory channels. Thus, the child must link the linguistic symbols s/he hears with the designated object, event, relation, or complete experience in the real world. The scientific term for such designations is “reference” or “extension” (Dromi, 1988). Relations between linguistic terms and real-world entities are random and, therefore, require processing, organization, and memorization. TD children, Late Talkers and children with other developmental disorders usually show better comprehension than

production. Children with ASD exhibit a different profile as a group, young children with ASD are more likely to display weakness in comprehension even relative to their existing production abilities.

Davidson and Ellis Weismer (2017) examined the hypothesis that the discrepancy between comprehension and production may serve as a clinical marker of ASD. A group of 32 children previously-diagnosed with ASD and 32 children diagnosed as Late Talkers (with a high risk for DLD) participated in their longitudinal study. A battery of several language tests was administered at the ages of 30, 44 and 66 months. Results indicated that between the ages of 24–30 months, an overwhelming majority of participants in the ASD group (91%) exhibited weakness in comprehension relative to their production scores, whereas none of the other groups displayed such a discrepancy. The production - comprehension gap steadily diminished with age and disappeared when the participants with ASD were 66 months old. Based on these results, the authors concluded that lower comprehension than production may be an age-specific marker of ASD with high sensitivity and very high specificity.

In a series of innovative experimental studies, Naigles and colleagues utilized The Intermodal Preferential Looking (IPL) Paradigm for testing language comprehension in children with ASD. By utilizing this objective measure, the researchers avoided the inherent difficulty of administering language tests to participants with ASD who often refuse to cooperate in formal testing situations. The percentage of time looking at competing video scenes, as well as the latency of the first look at the scene that matches a verbal instruction, generated conclusions on the comprehension of target words and verbal instructions.

Naigles and Fein (2016) reported that 2–3-year-old participants with ASD demonstrated an understanding of both simple (e.g., Subject-Verb order) and more complex (e.g., subject and object WH-questions) sentences in English. They also found that four-year-old children diagnosed with ASD could correctly distinguish between simple past tense and progressive past forms (Tovar, Fein, & Naigles, 2015). However, they reported difficulties of young children with ASD to relate words to their referents and speculated that the word biases that direct TD children to meaning of new words operate differently in children with ASD (Tovar et al., 2015).

Preissler and Carey (2005) also found that children with ASD are less likely to follow the speaker's referential intent while mapping new words into referents, that is, they seldom rely on social cues such as the speaker's gaze or pointing in order to successfully connect words with real-world experiences.

Rote Learning as a Symptom in ASD

Difficulties in processing linguistic input explain why individuals with ASD rely on rote learning, use echolalia, and often reverse pronouns when they speak. Children with ASD compensate for their difficulty of internalizing grammatical rules by reciting what they hear. A common clinical manifestation is the production of

memorized sets of words and sentences from television shows or books that the children do not fully comprehend. For example, a girl who wanted to open a box approached her mother and asked, "Do you want me to open it for you?", a phrase often addressed to her by the mother in similar situations. Another example is of a boy who was afraid of a popping balloon and therefore quoted a sentence from the Israeli book "*A Tale of Five Balloons"*: "Boom, bang, what happened? The balloon exploded, the balloon was torn" (Dromi & Shteiman, 2007). Parents also frequently report that their children are able to recite entire books verbatim, having heard them only a few times. Such children may recount entire stories by heart but seem unable to respond to simple requests or straight forwards questions, such as "give me the ball", "where is the car?", or "take the bottle". The use of memorized sequences, including numbers, letters, and geometric forms, is an indication of weakness in language comprehension. As such, the behavior of reciting texts and lists (numbers, color names, and alphabet) should not be encouraged by parents and clinicians. In line with the DSM-5 revision, all reciting forms including those that are taught to young children by parents are considered memorized expressions and hence are categorized under RRBI.

Echolalia as a Symptom in ASD

Echolalia is defined as an immediate or delayed repetition of utterances that includes segmental and supra-segmental aspects of the model sentence. For many years, it was regarded as one of the most salient abnormal speech behaviors of individuals with ASD. Today it is considered to be an example of RRBI, since it is judged by naïve observers as non-functional for communication.

McEvoy, Loveland, and Landry (1988) reported that echolalia was recorded in children with ASD who had minimal spontaneous expressive language. They argued that this behavior is not reflective of participants' chronological age or nonverbal intelligence, rather, it is a clinical marker for ASD. Tager-Flusberg and Calkins (1990) compared echolalic and non-echolalic speech productions of participants with ASD. They found that echoed utterances were significantly shorter and less grammatically-advanced than the spontaneous utterances of the same speakers. These findings encourage further exploration.

Over the years, the theoretical framework has evolved from echolalia as a meaningless repetition of others' speech to echolalia as an attempt to accomplish communication without having the underlying grammatical rules for generating productive speech (Charlop, 1983). Prizant and Duchan (1981) proposed that echolalic productions may serve a wide range of pragmatic purposes such as turn-taking, assertions, affirmatives, answers, requests, rehearsals (assisting linguistic processing by earning time), and self-regulation.

This claim led clinicians to consider cases in which echolalic utterances serve as means for requesting activities or objects, as well as a means for self-regulation and invitation for communication. Our clinical experience is that sometimes echolalic

utterances serve as means to retrieve a missing word from the mental lexicon. This explanation should be regarded and investigated in future studies, as it may serve as a productive strategy for eliciting productive speech (Dromi, 2018).

Pronoun Reversals as a Symptom in ASD

Pronoun reversals, in which the speaker fails to shift reference between the first and second person, are directly connected to language comprehension and are very common in individuals with ASD. Kim, Paul, Tager-Flusberg, and Lord (2014) argued that most young children with ASD go through a prolonged period of reversing pronouns and that they stop making these errors at older ages as their receptive language skills develop.

Pronoun reversals are not exclusive to ASD and are often recorded in blind children, in children with DLD, and may even be briefly recorded in young TD children. Rice, Oetting, Marquis, Bode, and Pae (1994) explained that the information processing demands of having to shift and make reference between the speaker and the listener make pronoun acquisition a highly-demanding task for all speakers. Other researchers attributed pronoun errors to the difficulty that individuals with ASD have in conceptualizing the social notions of "self" and "other", as these are embedded in discourse (Lee, Hobson, & Chiat, 1994).

Difficulties in Linguistic Inferences, Metaphors and Idioms

An area which poses a great challenge for individuals with ASD, is integrating information from various sources in order to fully comprehend complex grammatical structures and especially connected discourses. Deficits in understanding ambiguities, jokes, figurative expressions, idioms, and metaphors in oral or in written language, are routinely reported in this population. Such structures require inferences and therefore, they cause confusion and misunderstanding in individuals with ASD (Norbury, 2013).

For example, while bathing, a 3-year-old anxiously asked his mother whether he needs to wash his hair today and failed to infer from her expression "we washed it yesterday" the meaning of "no". In a series of studies, Norbury (2005a, 2005b) showed that ALN and ALI children varied significantly in their ability to process and comprehend jokes, metaphors, and idioms. She argued that this difference is associated with the overall deficit in language abilities in the two groups. Her hypothesis was that difficulties observed in narrative comprehension result from shifting between literal and figurative meanings, as well as from lack of inhibition of interpretations that are irrelevant to the communication context at hand. These attainments are developmentally complex and may not be achieved by many individuals with ASD even by adulthood.

Speech Production: Babbling, Words and Sentences

Absence of Canonical Babbling as a Red Flag for ASD

A few months prior to the emergence of first words, TD infants begin to produce sequences of syllabic constructions that are termed "Canonical Babbling". Oller and colleagues maintained that the production of syllabic constructions is the earliest indication that the baby advances in a typical trajectory of language development. Repeated syllables such as "babababa" or "dedede" imply that the baby is listening to the language input and is already segmenting it into basic units. A strong link has been identified between the emergence of canonical babbling (or lack of it) and subsequent cognitive impairments, sensory disorders and linguistic disabilities (Oller & Eilers, 1988; Oller, Eilers, Neal, & Schwartz, 1999).

Indeed, toddlers with ASD show a lack of canonical babbling or a significant delay in its emergence. Patten et al. (2014) conducted a retrospective analysis of video- recordings by examining the vocalizations produced by 37 infants at ages 9–12 months, and again at 15–18 months. 23 out of the 37 participants were later diagnosed with ASD. A comparison between those two groups revealed that the children who were later diagnosed with ASD were much older when they began to babble. They demonstrated significantly lower amounts of canonical babbling compared to TD and exhibited significantly fewer total vocalizations at both ages. This finding suggests that the absence of canonical babbling should be considered one of ASD's red flags.

The Emergence of First Words by Toddlers with ASD

In order to ascertain the average age for the emergence of first words by children with ASD, Howlin (2003) compared ADI-R retrospective questionnaires of 38 adults diagnosed with High Functioning Autism (HFA) and 42 adults with Asperger's syndrome (AS). On the basis of parental reports, she found that the average timing of first words in individuals with HFA was 38 months, compared to 15 months in the AS group. The rate of single word learning in the HFA group was extremely prolonged and, in some cases, lasted until children were 9 years old. Similar reports on significant delays in the production of first words as well as extended periods of constructing single word vocabularies appear in more recent publications in which parent-questionnaire-data were analyzed (Naigles & Chin, 2017; Tek, Mesite, Fein, & Naigles, 2014). At the same time, there are researchers who claim that early words appear on time in ASD, and in a few cases, accelerated rates of learning new words or nonsense words were reported (Kjelgaard & Tager-Flusberg, 2001; Loucas et al., 2008; Venker, Kover, & Ellis Weismer, 2016).

Rescorla and Safyer (2013) analyzed the first words that toddlers were reported by their parents to have said. The vocabulary composition of the first 100 words was

remarkably similar among the TD and ASD groups, consisting mostly of labels for food, body parts, and people. However, as lexical levels reached more than 100 words, vocabulary composition began to differ by group, with ASD children producing fewer words for actions and household items. In a study of 15 dyads of mothers and TD toddlers and 9 dyads of mothers and toddlers diagnosed with ASD, Oren (2017) directly examined the age at which children in the two groups reached a productive lexicon of 40–70 different words in Hebrew. The expansion of the productive lexicon was measured by a standardized questionnaire (HCDI –WG, Gendler-Shalev & Dromi, 2005) at two subsequent time points: 2 months and 4 months later. A significant delay was found in the average age by which children with ASD reached similar vocabulary sizes. While on average TD children produced 40–70 different words at the age of 17 months, children with ASD reached this level at an average age of 32 months. It is interesting to note that children in both groups had remarkably similar growth curves for accumulating additional new words following the attainment of the first milestone of 40–70 different words.

Word Extensions by Children with ASD

The extension of new words beyond a single exemplar, a single context or a single time and place, requires representational abilities that enable thinking about categories as opposed to single objects, specific events, and particular relationships (Dromi, 2008). Studies on word learning by children with ASD provide unique evidence for qualitative differences in the link formed between words and categories of meanings (Dromi, 2018). Irregular use of words is common among individuals with ASD and signifies idiosyncratic, non-conventional meanings. Even adults with ASD report about their tendency to utter words with unusual connotations. We and others have reported that some participants with ASD self-report that they enjoy using non-existing words or even "gibberish" sounds in conversations or during self-play (Dromi, 2018). Dromi and Shteiman (2007) described a child with ASD who, at the age of four years, began to speak. He was able to pick up photos of objects from a board display, to request the appropriate items, but was otherwise nonverbal. During one language intervention session, he pointed to the board on which there were six photos of everyday objects (e.g., a ball, a book, a doll, a box), and labeled them consecutively. However, he was unable to label real, corresponding objects presented to him. Indeed, difficulties in word extensions are common in ASD. Thus, it is recommended to carefully examine the different contexts in which the same words are uttered rather than merely count the number of words in a child's vocabulary.

Tek, Jaffery, Fein, and Naigles (2008) presented novel objects paired with novel nouns to toddlers with ASD and examined whether the children would extend this label to new objects of the same shape and new objects of the same color. Whereas TD toddlers as young as 24 months of age spontaneously extended the labels to new, same-shaped objects, language-matched children with ASD could not do it, even after multiple presentations over the course of two years of language intervention.

In-Depth Semantic Abilities of Individuals with ASD

The semantic abilities of children with ASD were found to be inferior to those of TD control groups who were matched by language comprehension scores rather than by age. McGregor et al. (2012) found that picture naming, definitions, and word associations were much more sensitive indicators of lexical learning in school-age children with ALI than tests of lexical comprehension. They reported that participants with ALI performed similarly to participants with language impairments without ASD with regards to several measures of semantic knowledge.

Pragmatics- the Core Difficulty of Individuals with ASD

In TD infants a well-established understanding of the communicative functions of words precedes the emergence of early words. Very little is known about the development of communicative functions by young children with ASD. To directly explore this question, Oren (2017) video- recorded dyadic interactions between 9 children (ages 22–40 months) who were previously diagnosed with ASD, and their mothers. She also video-recorded in similar conditions the dyadic interactions of 15 TD children at the same productive levels and their mothers. The TD participants were much younger than the participants with ASD. Oren conducted a frame–by–frame analysis of all the speech productions of each child in context. The aim of this rigorous analysis was to identify the communicative functions that were intended by the speakers. Child utterances were classified into declarative, requests, objections, non-communicative speech and unclassified productions. Results indicated that children in the ASD and the TD groups used words mostly for declarative purposes. In both groups the request category was the second in prevalence. A highly significant group difference was noted regarding the rate of the non- communicative use of speech, which was much higher in the children with ASD. As early as the first video-recording, when participants' lexicon did not exceed 40–70 different words, 90% of TD toddlers' words and only 65% of the ASD toddlers' words were used for communicative purposes. During the subsequent visits, communicative productions in the ASD children gradually rose, but did not reach the level of TD even by the end of the study (75% versus 90%).

Speech Intelligibility - Pitch, Voicing and Intonation in ASD

Paul (2010) was the first to point out the unique speech characteristics of children with ASD. She stated that atypical speech patterns often call unwanted attention to speakers with ASD and contribute to low speech intelligibility. Children with ASD

use flat intonation, often described as “strange”, “excessive”, or “robotic” (Green & Tobin, 2009; Naigles & Chin, 2017).

In a most ambitious attempt to characterize the speech characteristics of individuals with ASD, Parish-Morris and colleagues (2016) generated a Linguistic Data Consortium with the aim of analyzing ADOS audio-recordings of more than 1200 toddlers, children, teens, and adults with ASD. Out of this massive cohort, 100 language samples were analyzed in a computerized fashion to identify specific speech features that are associated with ASD. The following characteristics were found: individuals with ASD show smaller lexical diversity than TD, they often tend to use filler words such as “um” and “uh”, they exhibit more instances of disfluency. Their rate of speech is slower than that expected according to their linguistic age, they make longer pauses before responding during conversation, their prosody (intonation and rate) is deviant and their fundamental frequency is higher and more variable than among TD or children with other developmental disorders (Parish-Morris et al., 2016).

It was also reported that speakers with ASD often fail to take into consideration the addressee, and therefore produce non-comprehensible utterances. Such expressions may be classified as means of self-stimulation or simply self-play. Non-comprehensible productions might also serve for sensory stimulation and they often have characteristics of “gibberish” that is impossible to phonetically transcribe or to imitate. Some individuals with ASD report that they often produce sounds, oral clicks, words, and even sentences for self-regulation. Talking to self is common in individuals with ASD who have established grammar and utilize speech as means of scaffolding their own motor behaviors (Dromi & Shteiman, 2007). We are not aware of research that addressed the acoustics or linguistic structures of such speech productions.

RRBI and Language Development - What Can Be Said to Date?

Studies on the relation between language development and RRBI are scarce. In a recent publication by Whitten, Unruh, Shafer, and Bodfish (2018), the association between the two core areas of impairment in ASD (i.e. Social-Communication and RRBI) was compared in a sample of 222 children and adolescents with ASD and 158 TD children. A significant moderate correlation was found between communication scores and social scores in all participants, but no correlation was identified between the overall social-communication scores and the scores in RRBI. Subgrouping participants according to severity levels revealed that participants with low severity symptoms achieved scores on the two core domain areas of ASD that did not converge. In contrast, the scores of the two core domains did converge among participants who presented profound and severe levels of autism. In this sample, the two groups of participants did not significantly differ in terms of IQ scores. The authors

concluded that evidence for "convergent" or "divergent" causes for ASD depends on the sample characteristics. They proposed that special attention should be paid to the severity subclasses of ASD and to the type of the RRBI that participants exhibit.

In a number of reports, the severity of RRBI symptoms was found to be negatively correlated with cognitive or adaptive skills. Relatively higher levels of sensory and motor self-stimulating behaviors were observed in low functioning individuals with ASD (Berry, 2017; Ray-Subramanian & Weismer, 2012; Troyb et al., 2016). In a longitudinal follow-up investigation of 40 children, Troyb et al. (2016) measured the presence and the severity of RRBI in two age ranges: 1–2 years old and 3–5 years old. They later assessed the same children's cognitive functioning when they were 8–10 years old. Their results indicated that the RRBI measures at the first observation did not predict cognitive skills at age 8–10. At the ages of 3–5, more severe preoccupation with parts of objects predicted lower verbal reasoning abilities at age 8–10. Those children with hypo- and hyper reactivity to sensory stimulation had lower nonverbal abilities at age 8–10. Children with more extensive stereotyped and repetitive motor movements showed lower cognitive abilities, more rigid adaptive skills, and more severe autism symptoms at the third assessment when they were 8–10 years old. The results of this study indicated that the relation between RRBI and high-order cognitive and linguistic skills is complex and should be explored with caution. A differential approach to the various subcategories of RRBI as well as to different time points in the course of development is required before clear conclusions about associations between core domains can be drawn (Berry, 2017).

Ray-Subramanian and Ellis Weismer (2012) examined whether language skills and cognitive levels, as measured by the Preschool Language Scale, Fourth Edition (PLS-4) and the Mullen Scale of Early Learning respectively, were associated with clinically-observed RRBI at ages 2 and 3. Their sample included 115 children who were seen at two time-points. They found that at age 2, the correlation between RRBI scores and linguistic or nonverbal skills were not statistically significant. At age 3, expressive language scores were negatively-correlated with RRBI scores. Most importantly, increases in receptive and expressive language scores on the PLS-4 from age 2 to age 3, were negatively associated with the scores in RRBI over the same period of time. The authors concluded that progress in language comprehension, together with an increase in language use for communication, may lead to a decreased need for self-occupation or soothing with sensory or motor self-stimulating behaviors.

Summary Points

1. The findings on language comprehension and speech production strongly support the conclusion that individual variability is much greater in ASD than in TD and in any other clinical group.

2. Young children with ASD face difficulties in segmenting and processing the language that is addressed to them. Many children show a tendency to utter sentences and phrases that they hear with an incomplete representation of their meanings.
3. The ASD population is divided into two groups, ALI and ALN. Early words emerge considerably later in most children with ASD compared to TD children.
4. Word meaning acquisition is challenging for children with ASD, as manifested by the unusual use of existing words and the use of idiosyncratic oral productions.
5. Echolalic speech is indicative of difficulties in comprehension and/or in retrieving words from the mental lexicon. Yet, it is not exclusively found in ASD and in some cases may be viewed as a means to continue communication.
6. Pronoun reversals occur more often in children with ASD, and for prolonged periods of time, compared to TD children or children with other developmental disorders.
7. Metaphoric language poses a great challenge for children and adults with ASD. Interpretation of idioms, indirect requests, and implications are often impossible.
8. The use of speech for communicative purposes poses one of the greatest challenges in this population and, thus, one of the main therapeutic goals is teaching the children to use their language efficiently, in a flexible manner, for various communicative intentions.
9. The overall intelligibility of speech in children with ASD is often low, due to segmental, prosodic as well as pragmatic reasons. Disfluency, rate of speech, as well as the use of fillers differentiate them as a distinct clinical group.
10. The relations between the severity of RRBI, the level of cognitive functioning, adaptive behaviors, and language skills are very complex. For this reason, it is desirable to test this question with a close look at subgroups of participants with reference to their symptom severity, subtypes of RRBI, as well as different age groups.
11. Future study designs should strive for greater sophistication with the aim of obtaining a deeper understanding of this multi-faceted issue, and generalizations should be proposed with great caution and be followed in meticulous future studies.

References

Adamson, L. B., Romski, M., & Barton-Hulsey, A. (2014). Early language acquisition in autism spectrum disorders: A developmental view. In *Comprehensive guide to autism* (pp. 1061–1080). New York, NY: Springer.

Astington, J. W., & Jenkins, J. M. (1999). A longitudinal study of the relation between language and theory-of-mind development. *Developmental Psychology, 35*(5), 1311–1320.

Berry, K. C. (2017). *From subtypes to phenotypes: Discovering the clinical predictors of RRB profiles in ASD* [UCLA]. https://escholarship.org/uc/item/4tf534qn

Charlop, M. H. (1983). The effects of echolalia on acquisition and generalization of receptive labeling in autistic children. *Journal of Applied Behavior Analysis, 16*(1), 111–126. https://doi.org/10.1901/jaba.1983.16-111.

Davidson, M. M., & Ellis Weismer, S. (2017). A discrepancy in comprehension and production in early language development in ASD: Is it clinically relevant? *Journal of Autism and Developmental Disorders, 47*(7), 2163–2175. https://doi.org/10.1007/s10803-017-3135-z.

Dawson, G., Rogers, S., Munson, J., Smith, M., Winter, J., Greenson, J., Donaldson, A., & Varley, J. (2010). Randomized, controlled trial of an intervention for toddlers with autism: The early start Denver Model. *Pediatrics, 125*(1), e17–e23. https://doi.org/10.1542/peds.2009-0958.

Dromi, E. (1988). *Early lexical development*. Cambridge, UK: Cambridge University Press.

Dromi, E. (2008). Old data – New eyes: Theories of word meaning acquisition. In C. M. Gathercole (Ed.), *Routes to language: Studies in Honor of Melissa Bowerman* (1st ed., pp. 39–59). London, UK: Psychology Press.

Dromi, E. (2018). In Lev-Har & Rachel (Eds.), *Autism: A journey toward understanding the spectrum*. Herzliya, Israel: Niv Books.

Dromi, E., & Shteiman, M. (2007). Communication, language and speech in young children with autism: Assessment and intervention principles. In S. Lowinger (Ed.), *Interactive approaches to the treatment of children with communication disorders*. Haifa, Israel.

Gendler-Shalev, H., & Dromi, E. (2005). *The adaptation of HCDI-WG parent questionnaire to Hebrew*. Tel-Aviv, Israel: Tel-Aviv University.

Green, H., & Tobin, Y. (2009). Prosodic analysis is difficult… but worth it: A study in high functioning autism. *International Journal of Speech-Language Pathology, 11*(4), 308–315.

Happé, F., & Frith, U. (2006). The weak coherence account: Detail-focused cognitive style in autism spectrum disorders. *Journal of Autism and Developmental Disorders, 36*(1), 5–25. https://doi.org/10.1007/s10803-005-0039-0.

Howlin, P. (2003). Outcome in high-functioning adults with autism with and without early language delays: Implications for the differentiation between autism and Asperger syndrome. *Journal of Autism and Developmental Disorders, 33*(1), 3–13.

Jusczyk, P. W. (1997). *The discovery of spoken language*. Cambridge, MA: A Bradford Book.

Kim, S. H., Paul, R., Tager-Flusberg, H., & Lord, C. (2014). Language and communication in autism. In F. R. Volkmar, R. Paul, S. J. Rogers, & K. A. Pelphrey (Eds.), *Handbook of autism and pervasive developmental disorders, assessment, interventions, and policy* (2nd ed., pp. 230–262). Hoboken, NJ: Wiley.

Kjelgaard, M. M., & Tager-Flusberg, H. (2001). An investigation of language impairment in autism: Implications for genetic subgroups. *Language and Cognitive Processes, 16*(2–3), 287–308.

Kuhl, P. K., Coffey-Corina, S., Padden, D., Munson, J., Estes, A., & Dawson, G. (2013). Brain responses to words in 2-year-olds with autism predict developmental outcomes at age 6. *PLoS One, 8*(5), e64967.

Kuhl, P. K., Conboy, B. T., Coffey-Corina, S., Padden, D., Rivera-Gaxiola, M., & Nelson, T. (2008). Phonetic learning as a pathway to language: New data and native language magnet theory expanded (NLM-e). *Philosophical Transactions of the Royal Society of London B: Biological Sciences, 363*(1493), 979–1000.

Lee, A., Hobson, R. P., & Chiat, S. (1994). I, you, me, and autism: An experimental study. *Journal of Autism and Developmental Disorders, 24*(2), 155–176.

Lord, C., & Bishop, S. L. (2015). Recent advances in autism research as reflected in DSM-5 criteria for autism spectrum disorder. *Annual Review of Clinical Psychology, 11*, 53–70.

Loucas, T., Charman, T., Pickles, A., Simonoff, E., Chandler, S., Meldrum, D., & Baird, G. (2008). Autistic symptomatology and language ability in autism spectrum disorder and specific language impairment. *Journal of Child Psychology and Psychiatry, 49*(11), 1184–1192.

Luyster, R., & Lord, C. (2009). The language of children with autism. In E. L. Bavin (Ed.), *The Cambridge handbook of child language* (1st ed.). Cambridge, UK: Cambridge University Press.

McEvoy, R. E., Loveland, K. A., & Landry, S. H. (1988). The functions of immediate echolalia in autistic children: A developmental perspective. *Journal of Autism and Developmental Disorders, 18*(4), 657–668.

McGregor, K. K., Berns, A. J., Owen, A. J., Michels, S. A., Duff, D., Bahnsen, A. J., & Lloyd, M. (2012). Associations between syntax and the lexicon among children with or without ASD and language impairment. *Journal of Autism and Developmental Disorders, 42*(1), 35–47.

Meltzoff, A. N. (2013). Origins of social cognition: Bidirectional self-other mapping and the "Like-Me" hypothesis. In M. Banaji & S. Gelman (Eds.), *Navigating the social world: What infants, children, and other species can teach us (social cognition and social neuroscience)* (1st ed., pp. 139–144). New York, NY: Oxford University Press.

Naigles, L. R., & Chin, I. (2017). Language development in children with autism. In E. L. Bavin & L. R. Naigles (Eds.), *The Cambridge handbook of child language* (2nd ed.). Cambridge, UK: Cambridge University Press.

Naigles, L. R., & Fein, D. (2016). Looking through their eyes: Tracking early language comprehension in ASD. In L. R. Naigles (Ed.), *Innovative investigations of language in autism spectrum disorder* (1st ed., pp. 49–69). Cambridge, UK: De Gruyter Mouton.

Norbury, C. F. (2005a). Barking up the wrong tree? Lexical ambiguity resolution in children with language impairments and autistic spectrum disorders. *Journal of Experimental Child Psychology, 90*(2), 142–171.

Norbury, C. F. (2013). Autism spectrum disorders and communication. In L. Cummings (Ed.), *The Cambridge handbook of communication disorders*. Cambridge, UK: Cambridge University Press.

Norbury, C. F. (2005b). The relationship between theory of mind and metaphor: Evidence from children with language impairment and autistic spectrum disorder. *British Journal of Developmental Psychology, 23*(3), 383–399.

Oller, D. K., & Eilers, R. E. (1988). The role of audition in infant babbling. *Child Development, 59*, 441–449.

Oller, D. K., Eilers, R. E., Neal, A. R., & Schwartz, H. K. (1999). Precursors to speech in infancy: The prediction of speech and language disorders. *Journal of Communication Disorders, 32*(4), 223–245.

Oren, A. (2017). *Pragmatic aspects of early lexicon among children with autistic spectrum disorders and toddlers with typical development*. Tel-Aviv, Israel: Tel-Aviv University.

Parish-Morris, J., Cieri, C., Liberman, M., Bateman, L., Ferguson, E., & Schultz, R. T. (2016). *Building language resources for exploring autism spectrum disorders*. LREC... International conference on language resources & evaluation: [Proceedings]. International conference on language resources and evaluation, 2016, pp. 2100–2107.

Patten, E., Belardi, K., Baranek, G. T., Watson, L. R., Labban, J. D., & Oller, D. K. (2014). Vocal patterns in infants with autism spectrum disorder: Canonical babbling status and vocalization frequency. *Journal of Autism and Developmental Disorders, 44*(10), 2413–2428.

Paul, R. (2010). Communication development and assessment. In K. Chawarska, A. Klin, & F. R. Volkmar (Eds.), *Autism Spectrum disorders in infants and toddlers: Diagnosis, assessment, and treatment* (pp. 76–103). New York, USA: Guilford Press.

Peterson, C. C., Wellman, H. M., & Liu, D. (2005). Steps in theory-of-mind development for children with deafness or autism. *Child Development, 76*(2), 502–517.

Preissler, M. A., & Carey, S. (2005). The role of inferences about referential intent in word learning: Evidence from autism. *Cognition, 97*(1), B13–B23.

Prizant, B. M., & Duchan, J. F. (1981). The functions of immediate echolalia in autistic children. *Journal of Speech and Hearing Disorders, 46*(3), 241–249.

Ray-Subramanian, C. E., & Weismer, S. E. (2012). Receptive and expressive language as predictors of restricted and repetitive behaviors in young children with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 42*(10), 2113–2120.

Rescorla, L., & Safyer, P. (2013). Lexical composition in children with autism spectrum disorder (ASD). *Journal of Child Language, 40*(1), 47–68.

Rice, M. L., Oetting, J. B., Marquis, J., Bode, J., & Pae, S. (1994). Frequency of input effects on word comprehension of children with specific language impairment. *Journal of Speech, Language, and Hearing Research, 37*(1), 106–122.

Suh, J., Eigsti, I.-M., Naigles, L., Barton, M., Kelley, E., & Fein, D. (2014). Narrative performance of optimal outcome children and adolescents with a history of an autism spectrum disorder (ASD). *Journal of Autism and Developmental Disorders, 44*(7), 1681–1694.

Tager-Flusberg, H., & Calkins, S. (1990). Does imitation facilitate the acquisition of grammar? Evidence from a study of autistic, Down's syndrome and normal children. *Journal of Child Language, 17*(3), 591–606.

Tek, S., Jaffery, G., Fein, D., & Naigles, L. R. (2008). Do children with autism spectrum disorders show a shape bias in word learning? *Autism Research, 1*(4), 208–222.

Tek, S., Mesite, L., Fein, D., & Naigles, L. (2014). Longitudinal analyses of expressive language development reveal two distinct language profiles among young children with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 44*(1), 75–89.

Tomblin, B. (2011). Co-morbidity of autism and SLI: Kinds, kin and complexity. *International Journal of Language & Communication Disorders, 46*(2), 127–137.

Tovar, A. T., Fein, D., & Naigles, L. R. (2015). Grammatical aspect is a strength in the language comprehension of young children with autism spectrum disorder. *Journal of Speech, Language, and Hearing Research, 58*(2), 301–310.

Troyb, E., Knoch, K., Herlihy, L., Stevens, M. C., Chen, C.-M., Barton, M., Treadwell, K., & Fein, D. (2016). Restricted and repetitive behaviors as predictors of outcome in autism spectrum disorders. *Journal of Autism and Developmental Disorders, 46*(4), 1282–1296.

Venker, C. E., Kover, S. T., & Ellis Weismer, S. (2016). Brief report: Fast mapping predicts differences in concurrent and later language abilities among children with ASD. *Journal of Autism and Developmental Disorders, 46*(3), 1118–1123. https://doi.org/10.1007/s10803-015-2644-x.

Whitten, A., Unruh, K. E., Shafer, R. L., & Bodfish, J. W. (2018). Subgrouping autism based on symptom severity leads to differences in the degree of convergence between core feature domains. *Journal of Autism and Developmental Disorders, 48*(6), 1908–1919.

Chapter 10
Social Interaction Among Individuals with Autism Spectrum Disorder: Relations with Repetitive and Restricted Behaviors and Interests

Parisa Ghanouni and Tal Jarus

Introduction

Imagine that you watch a movie in a language that you do not know. Although you do not comprehend what the characters say, you might still grasp some social information based on the characters' interactions and the events that take place on the screen. You can easily recognise tone of voices and types of interactions, if it is relaxed, aggressive, or tense. Using your visual and auditory senses to take in information, and comparing it with your previous experiences, you can interpret and predict what happens in the situation. How would you react if you could not interpret those non-verbal interactions or get overwhelmed by the stimuli presented on the screen? You would probably turn off the video as neither you enjoy it nor understand the content. If you were unable to turn off the video or move somewhere else, you might get irritated and stressed. This might be manifested in physiological and behavioural responses to the situation (Notarius & Levenson, 1979).

This might be an example on how individuals with social difficulties such as autism spectrum disorder experience everyday life.

Autism spectrum disorder (ASD) is a neurodevelopmental condition that is manifested by (a) persistent deficits in social interaction and communication as well as (b) restricted and repetitive patterns of behaviour and interest (American Psychiatric Association, 2013). These two core diagnostic features of ASD negatively affect the ability of individuals with ASD to be involved in social activities and to have opportunities to acquire additional skills. The ASD symptoms are present from early in

P. Ghanouni (✉)
Dalhousie University, Halifax, NS, Canada
e-mail: Parisa.Ghanouni@dal.ca

T. Jarus
University of British Columbia, Vancouver, BC, Canada

E. Gal, N. Yirmiya (eds.), *Repetitive and Restricted Behaviors and Interests in Autism Spectrum Disorders*, Autism and Child Psychopathology Series,
https://doi.org/10.1007/978-3-030-66445-9_10

development and may last throughout life (Baron-Cohen, Wheelwright, Hill, Raste, & Plumb, 2001; Kuusikko et al., 2009).

ASD is one of the most prevalent childhood disorders, with about 1 out of 59 children currently being diagnosed with ASD (Baio et al. 2018). The prevalence of the diagnosis is on the rise (Wingate et al., 2014). Although the precision of diagnostic tools and increased awareness may contribute to this growth, the possibility of true increased incidence rates of autism cannot be ruled out (Ouellette-Kuntz et al., 2014). It is expected that this prevalence will continue its upward trend; therefore, a better understanding of its phenotype, appropriate planning, and resource allocation should be considered.

The aim of this chapter is to explain (a) what social interaction looks like in individuals with ASD and (b) how it may be affected by RRBI due to the interplay between the two diagnostic domains.

Social Communication Difficulties

Social impairments are a core element in defining the characteristics of ASD. A wide spectrum of difficulties in maintaining relationships, persistent deficits in verbal and non-verbal social communication, problems in turn taking, challenges in socio-emotional reciprocity, and misinterpretation in socio-emotional contexts are often observed in individuals with ASD (American Psychiatric Association, 2013; Sperry & Mesibov, 2005). These deficits limit their relationships with others and prevent them from maintaining their social networks and friendships. Consequently, this may compromise social participation of individuals with ASD and can make them vulnerable to additional social and psychosocial difficulties (Kawachi & Berkman, 2001).

Problems in communication skills and impaired social interaction, both in quality and quantity, can be observed in children with ASD. These children have fewer social interactions and spend less time interacting with others (Hilton, Crouch, & Israel, 2008). They also show heightened anxiety in social contexts compared to their typically developing (TD) peers (Corbett et al., 2014; Corbett, Schupp, Simon, Ryan, & Mendoza, 2010). This hyper-arousal and social anxiety in children with ASD can be the result of poor adaptive social skills and coping strategies to respond appropriately. Enhanced anxiety can also reflect a greater awareness of their own limited social skills in preparation for appropriate social interactions (Corbett et al., 2010). As children with ASD get older, they gain more insight about their limited social competencies and they experience more stress in social contexts (Corbett et al., 2010; Kuusikko et al., 2008; Lopata, Volker, Putnam, Thomeer, & Nida, 2008).

Children with ASD show delay and difficulty in perspective-taking, that is, the ability to interpret others' mental states and predict social behaviours. According to theory of mind (Baron-Cohen, 1990; Happé & Frith, 1995), individuals with ASD may experience problems in understanding that other people have different viewpoints, leading to dysfunction in inferring others' emotions, desires, and intentions

(Baron-Cohen, 1997; Happé & Frith, 1995). Deficits in intention-reading and not being capable of understanding the desires, feelings, and emotions of others contribute to the challenges children with ASD face with social interactions (Baron-Cohen, 1990; Moran et al., 2011). The ability to attribute behaviours to various intentions and emotions, and understanding what is in the mind of others are parts of empathy. To empathize with others, it is necessary not only to understand others' mental states or emotions (cognitive part), but also to respond to them appropriately (affective part) (Aaan Het Rot & Hogenelst, 2014). Perspective-taking, cognitive empathy, and affective empathy are linked together and difficult to separate (Bensalah, Caillies, & Anduze, 2016). Children with ASD experience difficulties in these aspects, which profoundly affect their abilities to empathize and display socio-emotional reciprocity (American Psychiatric Association, 2013; Baron-Cohen, 2002; Ghanouni et al., 2019; McIntosh, Reichmann-Decker, Winkielman, & Wilbarger, 2006; Minio-Paluello, Baron-Cohen, Avenanti, Walsh, & Aglioti, 2009; Pasalich, Dadds, & Hawes, 2014). Socio-emotional difficulties in individuals with ASD may also be due to problems in attending and orienting to relevant social stimuli. Specifically, researchers believe that difficulties in the rapid shifting of attention between social stimuli and the inability to share attention with others result in profound social problems in individuals with ASD (Dawson et al., 2004; Dawson, Meltzoff, Osterling, Rinaldi, & Brown, 1998). As the nature of social stimuli is variable and unpredictable, individuals with ASD have difficulties processing social stimuli and drawing attention to them (Dawson et al., 2004). Also, individuals with ASD may not find social stimuli intrinsically motivating and rewarding, which result in reduced attention to faces (Chevallier, Kohls, Troiani, Brodkin, & Schultz, 2012). People with ASD tend to focus more on objects rather than faces, and typically avoid eye contact (McPartland, Dawson, Webb, Panagiotides, & Carver, 2004; Swettenham et al., 1998; Wallace, Coleman, & Bailey, 2008). The inability to detect and respond precisely to social stimuli, such as eye gaze and emotional faces, thus hinders the development of socio-emotional competencies in people with ASD (Dawson et al., 2004).

When focusing on the face, individuals with ASD process emotional faces differently than their peers. Some researchers reveal that individuals with ASD tend to focus more on the lower parts of the faces (i.e., the mouth) than the upper parts (i.e., the eyes), which result in difficulty processing negative or complex emotions (Joseph & Tanaka, 2003; Langdell, 1978). Although attending to the mouth may assist in obtaining better verbal information for individuals with ASD, lack of attention to the eyes may result in difficulty in understanding the mental states conveyed through the eyes (Klin, Jones, Schultz, Volkmar, & Cohen, 2002). Individuals with ASD adopt a more feature-based or detail-focused processing approach rather than a whole-based processing approach used by their TD counterparts, which may contribute to difficulties in recognizing emotions (Behrmann et al., 2006). According to the weak central coherence theory, individuals with ASD over-rely on details and extract local information (Happé & Frith, 2006). This atypical visual processing and increased local bias among individuals with ASD interfere with their ability to interpret emotional faces, a task that mostly requires holistic processing of the whole face (Behrmann, Thomas, & Humphreys, 2006).

Nature of Social Interactions

When two or more entities such as humans interact with each other and convey some information, there are some events that occur at the same time. This may include movements of the body, trunk, head, and eyes or producing some sounds and speech while the whole body changes its position and posture in relation to the environment (Sanchez Puerta, Valerio, & Bernal, 2016). These events, along with other numerous events in the environment, occur concurrently or sequentially within, between and among individuals in various ways. Paralinguistic and linguistic cues are conceptual dependents and can affect interpersonal communication. However, these various social information cues are not entirely visible in the context of social interaction. The dynamics of interactions among individuals and environments along with the numerous events are constantly happening in the environment within spatial and temporal contexts. These co-occurring events make the perception of interactions, identification of the meaning based on the context, and determination of the consequences and effects of events extremely complex for all individuals, and particularly for those with socio-emotional difficulties such as ASD (Anolli, Duncan, Magnusson, & Riva, 2005).

Facial expressions and verbal communication are not the only sources for obtaining socially relevant information. It is assumed that the ability to perceive social information heavily rely on observing human motion and inferring from gestures (Blake & Shiffrar, 2007). Visual processing of body movements or biological motion plays a pivotal role in successful daily life interactions, in particular adaptive social and non-verbal behaviours (Allison, Puce, & McCarthy, 2000; Pavlova, 2011). Whereas children with TD can do this effortlessly, children with ASD have difficulties in accurately extracting information based on human movements (Blake, Turner, Smoski, Pozdol, & Stone, 2003; Pavlova, 2011). Previous studies usually used point-light displays of body movements to explore the perception of biological motion among children with and without ASD (Blake et al., 2003; Freitag et al., 2008). Children with TD can implicitly infer physical characteristics such as age and gender as well as higher order characteristic such as affect and intention from biological movements (Clarke, Bradshaw, Field, Hampson, & Rose, 2005; Kozlowski & Cutting, 1977; Mather & Murdoch, 1994; Pollick, Paterson, Bruderlin, & Sanford, 2001; Troje, 2002). However, children with ASD demonstrate some impairments in identifying subjective states of human movements or describing their actions and may even show difficulties in distinguishing animate from inanimate objects (Congiu, Schlottmann, & Ray, 2010). These atypical patterns in the perception of biological motion in children with ASD appear early in life and have cascading consequences for social development and interpersonal communication (Klin, Lin, Gorrindo, Ramsay, & Jones, 2009).

The perception of biological motion and social cognitive functions is tightly related (Pavlova, 2011). Social cognition or the cognitive mechanisms underlying social behaviours such as theory of mind, processing of the eye, head, mouth, and body movements develop over time (Allison et al., 2000; Senju, 2013). Human

infants are attentive to the display of upright but not inverted human point-light images (Yoon & Johnson, 2009). As individuals grow, they may be able to identify social meaning such as deceptive and true intentions of behaviours from body movements (Runeson & Frykholm, 1983). Therefore, the perceptual system for processing biological motion might be functionally related to social competencies (Pavlova, 2011). To perceive social information and biological motion, attending to visual cues plays a pivotal role. However, over-selective attention among individuals with ASD might affect their ability to perceive social situations appropriately (Pierce, Glad, & Schreibman, 1997). In natural environment, where there are multiple cues and events happen at the same time, individuals with ASD perform weaker than in the situations in which there are single cues (Burke & Cerniglia, 1990; Pierce et al., 1997). Individuals with ASD have difficulty in spontaneously attending to relevant social cues though they may process when they are explicitly asked for or when their attention is navigated towards relevant cues (Senju, 2013). This lack of competencies might result in profound impairment in social interaction.

Social Interactions in Individuals with ASD and Its Interplay with RRBI

As described above, social characteristics and RRBI are two diagnostic criteria of ASD. These core features might differ in nature from each other, but they may be related. Below are some examples that may describe its interconnection:

1. Common underlying mechanisms
2. Social withdrawn as a result of RRBI
3. RRBI as a result of lack of social understanding

Common Underlying Mechanisms

Sensory processing deficits, executive functions and differences in information processing due to altered brain neural connectivity were described in the literature as potential underlying mechanisms that stand at the base of various ASD traits.

Repetitive motor behaviours are often associated with sensory processing deficits in individuals with ASD (Boyd et al., 2010; Gabriels et al., 2008). However, not all repetitive behaviours necessarily include sensory feedback components, and likewise not all atypical sensory behaviours are repetitive. It has been assumed that repetitive behaviours may serve as a strategy for individuals with ASD to regulate their arousal level or manage their poor sensory processing either by creating self stimulation or avoiding and restricting it (Baker, Lane, Angley, & Young, 2008; Kientz & Dunn, 1997; Turner, 1999; Wigham, Rodgers, South, McConachie, &

Freeston, 2015). In other words, overload of sensory input, hyper-arousal, and anxiety might also contribute to demonstrating other RRBI. The desire of individuals with ASD for sameness and their intolerance of uncertainty when the environment is not predictable can explain why they may show hyper-arousal and social anxiety in unpredictable environments such as social situations, leading to desire for social isolation and lack of communication (Boulter, Freeston, South, & Rodgers, 2014; Chamberlain et al., 2013; Joyce, Honey, Leekam, Barrett, & Rodgers, 2017).

The pervasive and rigid nature of RRBI in individuals with ASD may be linked to the deficits in "executive functions" (South, Ozonoff, & Mcmahon, 2007). Executive function is an umbrella term for a set of functions including cognitive flexibility, response inhibition, and other mental processes. It has been shown that individuals with ASD may have problems in some domains of executive functions. Although the evidence is mixed, individuals with higher deficits in executive functions may demonstrate persistent production of the same behaviour, motor movement patterns, circumscribed interest, or repetitive speech that affect their communication (Boyd, McBee, Holtzclaw, Baranek, & Bodfish, 2009; Liss et al., 2001; Lopez, Lincoln, Ozonoff, & Lai, 2005; South et al., 2007; Turner, 1999). The inflexibility of individuals with ASD to switch from one activity to the other and difficulty in inhibiting inappropriate behaviour may lead to the repetition of the ongoing line of thought, affecting social participation (Turner, 1999). It is possible that cognitive impairments and executive dysfunctions observed in individuals with ASD may be associated with manifestation of restricted and repetitive behaviors (Lopez et al., 2005).

Individuals with ASD may demonstrate difficulties in information integration and information processing due to "altered brain neural connectivity" (Belmonte et al., 2004; Rippon, Brock, Brown, & Boucher, 2007; Zikopoulos & Barbas, 2013). This atypical synchronisation and imbalance between excitatory and inhibitory brain activities affect how people with ASD function in social situations (Pérez Velázquez & Galán, 2013; Rubenstein & Merzenich, 2003). Most interactions that happen in social environments are hidden to our eyes. However, poor signal-to-noise and excessive generation of information at resting state, regardless of the presence of relevant stimuli or attending to them, might justify how individuals with ASD may show hyper-arousal and be trapped in their limited internal world (Markram & Markram, 2010; Pérez Velázquez & Galán, 2013; Rippon et al., 2007). The weak central coherence observed in individuals with ASD can be associated with demonstration of restricted and repetitive behaviours and difficulties in social interaction (Lewis & Kim, 2009).

Social Withdrawn as a Result of RRBI

The RRBI may cause significant challenges for individuals with ASD and their families, including severe dysfunctions due to resistance to changes that interfere with their daily activities (Bishop, Richler, Cain, & Lord, 2007; Dunlap, Dyer, & Koegel, 1983). These behaviours are socially stigmatized and are considered as

some of the most stressful behaviours for parents (Bishop et al., 2007). When these behaviours are interrupted, individuals with ASD might get extremely anxious, distressed, and agitated. They might also demonstrate further aggressive behaviours in response to this interruption. The RRBI is shown to affect the ability of children with ASD in observational learning, attending to environmental stimuli, and actively exploring the environment (Pierce & Courchesne, 2001). Reduced tendency to explore new objects, topics that are outside of their scope of interests, and failure to engage in varieties of activities have developmental consequences. This can result in missing the learning opportunities that have profound effects on the development of sensory-motor, cognition, and structural and functional neural systems (Pierce & Courchesne, 2001). Such cascading effects can add to the socio-emotional problems that individuals with ASD experience.

In addition, the repetitive vocal behaviors and stereotypical movements, characterizing some people with ASD may reduce opportunities for children with ASD to engage in social activities (Durand & Carr, 1987), interfere with learning, academic skills, and prevent children with ASD from engaging in peer interactions (Durand & Carr, 1987; Koegel & Covert, 1972; Repp, Singh, Karsh, & Deitz, 1991). They might also create social stigma and rejections by others that lead to social exclusion and isolation (Farrugia, 2009; Gray, 1993; Kinnear, Link, Ballan, & Fischbach, 2016). Many parents of typically developing children may be reluctant to let their children interact with a child with disabilities in general and specifically those with ASD, as it may need extra supervision (Geisthardt, Brotherson, & Cook, 2002; Solish, Perry, & Minnes, 2010). These reactions by others might not always be rational responses and may carry assumptions about why the child displays atypical or disruptive behaviours (Gray, 2002; Kinnear et al., 2016). The lack of acceptance by others may results in further distress to children with ASD and their families and thus limiting further social interactions.

In addition to repetitive behaviours, restricted interests and preoccupations can affect the communication of individuals with ASD. Insistence on sameness and inability to adapt to changes in daily schedule impair social functioning and engagement in activities (Gabriels, Cuccaro, Hill, Ivers, & Goldson, 2005; Klin, Danovitch, Merz, & Volkmar, 2007). The restricted interests and preoccupations may range from unusual activities such as memorizing serial numbers to more typical hobbies such as an interest in math. The preoccupations, rigidity, and invariant nature of activities in individuals with ASD may prevent them from the development of peer-relations as they only care about their private interest (Cohen & Volkmar, 1997). Circumscribed interests may provide one-sided conversations and rigidity in switching between activities that can result in peer rejections (Boyd, Conroy, Mancil, Nakao, & Alter, 2007). As new skills can be acquired through observation and peer modelling, not being able to take turns in group activities and inability to inhibit desired tasks among children with ASD limit their learning opportunities (Charlop, Schreibman, & Tryon, 1983). Lack of social support and being excluded from peer groups make individuals with ASD prone for higher risk of coexisting conditions such as anxiety or depression that consequently further limit their social interactions (Bellini, 2004; Kuusikko et al., 2008; Meyer, Mundy, Van Hecke, & Durocher, 2006).

RRBI as a Result of Lack of Social Understanding

Although the evidence is scarce, there might be an association between social impairments and emergence of repetitive behaviours (Schultz, 2005). While social deficits in children with ASD can be evident as early as 6–12 months, repetitive and restricted behaviours usually emerge later in life between 2–4 years old (Lord, 1995; Maestro et al., 2002; Moore & Goodson, 2003; Osterling & Dawson, 1994). Individuals with ASD infrequently engage in interactions with peers and may isolate themselves from social activities. This might be due to their poor adaptive behaviours and social competencies. Dysregulated externalizing behaviours, such as impulsivity and aggression, and dysregulated internalizing behaviours, such as withdrawal and anxiety, can affect the quality of interaction and predict lower peer acceptance (Bauminger, Solomon, & Rogers, 2010; Mesman, Bongers, & Koot, 2001). Children with ASD may have trouble in maintaining friendships and are more likely to be bullied by their peers because of their lack of social skills (Bitsika & Sharpley, 2014; Rowley et al., 2012). Being socially isolated and having limited networks of friends among children with ASD affect their self-confidence in attending social situations (McCauley et al., 2019; Valkenburg, Peter, & Schouten, 2006). Individuals with ASD may find social situations stress-inducing as they may struggle in meeting social expectations. To be able to predict and gain control in a confusing and unpredictable social world, individuals with ASD insist on sameness, restricted interests, and routines. Any changes in predictability of environments may result in feeling irritated. Enhanced anxiety in social situation among individuals with ASD may also reflect an awareness of their limited social skills (Corbett et al., 2010). This is enhanced when children with ASD get older, as they gain more insight about their poor social skills and might experience more stress in social contexts (Corbett et al., 2010; Kuusikko et al., 2008; Lopata et al., 2008). Therefore, insistence on sameness in individuals with ASD might remain stable or even worsen over time (Bishop et al., 2006; Richler et al., 2010).

Furthermore, it has been shown that social deprivations in non-human primates can govern stereotypical and self-injury behaviours (Lewis, Gluck, Beauchamp, Keresztury, & Mailman, 1990; Mc Kinney Jr, 1974). These stereotypical behaviours may function to reduce stress caused by social deprivation (Mason, 1991). It may be assumed that poor social interaction in children with ASD can be stressful and lead to the development of restricted and repetitive behaviours. The repetitive behaviours observed in individuals with ASD may serve as a coping mechanism sustained or changed over time by environmental factors.

Peer support and a circle of friends can protect children from the negative effects of environmental stressors (Dumont & Provost, 1999; Whitaker, Barratt, Joy, Potter, & Thomas, 1998). Peer-mediated strategies not only increase social interactions but also have a collateral effect on reducing disruptive and stereotypical behaviours in children with ASD (Lee, Odom, & Loftin, 2007; Loftin, Odom, & Lantz, 2008; Oke & Schreibman, 1990). Peer training and functional communication strategies can provide opportunities for children with ASD to practice social skills with various

partners to foster their communication and reduce a range of aberrant behaviours (Bird, Dores, Moniz, & Robinson, 1989; Durand & Carr, 1992; Watkins et al., 2015). However, as there is a high variability in the presence of symptoms and phenotypes of ASD (Betancur, 2011; Georgiades et al., 2013), further studies investigating how social progression might affect RRBI and vice versa, and how these clinical manifestations are intensified or faded over time are warranted.

Summary

Although RRBI and impaired social skills are two main diagnostic domains in individuals with ASD, the majority of previous studied are in the social domain area. There is little information on how these two domains reinforce each other and what underlying mechanisms may take part to form these behaviours among children with ASD during development. It should be noted that both core pillars of ASD can create stigma that prevent participation in social activities and therefore, limit environmental learning opportunities for children with ASD. Promoting social engagement and support from peers may reduce stereotypical behaviours in individuals with ASD. However, it is still unclear what comprehensive interventions are best to implement and how to train peers about the typical manifestation of ASD that may seem to be odd from general viewpoints. Further studies should elucidate the extent to which effects of interventions can be achieved and maintained over time to better inform both family members and professionals.

References

Aaan Het Rot, M., & Hogenelst, K. (2014). The influence of affective empathy and autism spectrum traits on empathic accuracy. *PLoS One, 9*, e98436.

Allison, T., Puce, A., & McCarthy, G. (2000). Social perception from visual cues: Role of the STS region. *Trends in Cognitive Sciences, 4*, 267–278.

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders (DSM-5®)*. Arlington, TX: American Psychiatric Pub.

Anolli, L., Duncan, S., Magnusson, M., & Riva, G. (2005). Understanding social interaction: Discovering hidden structure with model and algorithms. In *The hidden structure of interaction: From neurons to culture patterns*. Amsterdam, Netherlands: IOS Press.

Baio, J., et al. (2018). Prevalence of autism spectrum disorder among children aged 8 years—Autism and Developmental Disabilities Monitoring Network, 11 Sites, United States, 2014. *MMWR Surveillance Summaries, 67*, 1–23.

Baker, A. E., Lane, A., Angley, M. T., & Young, R. L. (2008). The relationship between sensory processing patterns and behavioural responsiveness in autistic disorder: A pilot study. *Journal of Autism and Developmental Disorders, 38*, 867–875.

Baron-Cohen, S. (1990). Autism: A specific cognitive disorder of & lsquo; mind-blindness. *International Review of Psychiatry, 2*, 81–90.

Baron-Cohen, S. (1997). *Mindblindness: An essay on autism and theory of mind*. Cambridge, MA: MIT Press.

Baron-Cohen, S. (2002). The extreme male brain theory of autism. *Trends in Cognitive Sciences, 6*, 248–254.

Baron-Cohen, S., Wheelwright, S., Hill, J., Raste, Y., & Plumb, I. (2001). The "Reading the Mind in the Eyes" test revised version: A study with normal adults, and adults with Asperger syndrome or high-functioning autism. *Journal of Child Psychology and Psychiatry, 42*, 241–251.

Bauminger, N., Solomon, M., & Rogers, S. J. (2010). Externalizing and internalizing behaviors in ASD. *Autism Research, 3*, 101–112.

Behrmann, M., Avidan, G., Leonard, G. L., Kimchi, R., Luna, B., Humphreys, K., & Minshew, N. (2006). Configural processing in autism and its relationship to face processing. *Neuropsychologia, 44*, 110–129.

Behrmann, M., Thomas, C., & Humphreys, K. (2006). Seeing it differently: Visual processing in autism. *Trends in Cognitive Sciences, 10*, 258–264.

Bellini, S. (2004). Social skill deficits and anxiety in high-functioning adolescents with autism spectrum disorders. *Focus on Autism and Other Developmental Disabilities, 19*, 78–86.

Belmonte, M. K., Allen, G., Beckel-Mitchener, A., Boulanger, L. M., Carper, R. A., & Webb, S. J. (2004). Autism and abnormal development of brain connectivity. *Journal of Neuroscience, 24*, 9228–9231.

Bensalah, L., Caillies, S., & Anduze, M. (2016). Links among cognitive empathy, theory of mind, and affective perspective taking by young children. *The Journal of Genetic Psychology, 177*, 17–31.

Betancur, C. (2011). Etiological heterogeneity in autism spectrum disorders: More than 100 genetic and genomic disorders and still counting. *Brain Research, 1380*, 42–77.

Bird, F., Dores, P. A., Moniz, D., & Robinson, J. (1989). Reducing severe aggressive and self-injurious behaviors with functional communication training. *American Journal on Mental Retardation, 94*, 37.

Bishop, S. L., Richler, J., Cain, A. C., & Lord, C. (2007). Predictors of perceived negative impact in mothers of children with autism spectrum disorder. *American Journal on Mental Retardation, 112*, 450–461.

Bitsika, V., & Sharpley, C. F. (2014). Understanding, experiences, and reactions to bullying experiences in boys with an autism spectrum disorder. *Journal of Developmental and Physical Disabilities, 26*, 747–761.

Blake, R., & Shiffrar, M. (2007). Perception of human motion. *Annual Review of Psychology, 58*, 47.

Blake, R., Turner, L. M., Smoski, M. J., Pozdol, S. L., & Stone, W. L. (2003). Visual recognition of biological motion is impaired in children with autism. *Psychological Science, 14*, 151–157.

Boulter, C., Freeston, M., South, M., & Rodgers, J. (2014). Intolerance of uncertainty as a framework for understanding anxiety in children and adolescents with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 44*, 1391–1402.

Boyd, B. A., Baranek, G. T., Sideris, J., Poe, M. D., Watson, L. R., Patten, E., & Miller, H. (2010). Sensory features and repetitive behaviors in children with autism and developmental delays. *Autism Research, 3*, 78–87.

Boyd, B. A., Conroy, M. A., Mancil, G. R., Nakao, T., & Alter, P. J. (2007). Effects of circumscribed interests on the social behaviors of children with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 37*, 1550–1561.

Boyd, B. A., McBee, M., Holtzclaw, T., Baranek, G. T., & Bodfish, J. W. (2009). Relationships among repetitive behaviors, sensory features, and executive functions in high functioning autism. *Research in Autism Spectrum Disorders, 3*, 959–966.

Burke, J. C., & Cerniglia, L. (1990). Stimulus complexity and autistic children's responsivity: Assessing and training a pivotal behavior. *Journal of Autism and Developmental Disorders, 20*, 233–253.

Chamberlain, P. D., Rodgers, J., Crowley, M. J., White, S. E., Freeston, M. H., & South, M. (2013). A potentiated startle study of uncertainty and contextual anxiety in adolescents diagnosed with autism spectrum disorder. *Molecular Autism, 4*, 31.

Charlop, M. H., Schreibman, L., & Tryon, A. S. (1983). Learning through observation: The effects of peer modeling on acquisition and generalization in autistic children. *Journal of Abnormal Child Psychology, 11*, 355–366.

Chevallier, C., Kohls, G., Troiani, V., Brodkin, E. S., & Schultz, R. T. (2012). The social motivation theory of autism. *Trends in Cognitive Sciences, 16*, 231–239.

Clarke, T. J., Bradshaw, M. F., Field, D. T., Hampson, S. E., & Rose, D. (2005). The perception of emotion from body movement in point-light displays of interpersonal dialogue. *Perception, 34*, 1171–1180.

Cohen, D. J., & Volkmar, F. R. (1997). *Handbook of autism and pervasive developmental disorders* (2nd ed.). New York, NY: J. Wiley.

Congiu, S., Schlottmann, A., & Ray, E. (2010). Unimpaired perception of social and physical causality, but impaired perception of animacy in high functioning children with autism. *Journal of Autism and Developmental Disorders, 40*, 39–53.

Corbett, B. A., Schupp, C. W., Simon, D., Ryan, N., & Mendoza, S. (2010). Elevated cortisol during play is associated with age and social engagement in children with autism. *Molecular Autism, 1*, 13.

Corbett, B. A., Swain, D. M., Newsom, C., Wang, L., Song, Y., & Edgerton, D. (2014). Biobehavioral profiles of arousal and social motivation in autism spectrum disorders. *Journal of Child Psychology and Psychiatry, 55*, 924–934.

Dawson, G., Meltzoff, A. N., Osterling, J., Rinaldi, J., & Brown, E. (1998). Children with autism fail to orient to naturally occurring social stimuli. *Journal of Autism and Developmental Disorders, 28*, 479–485.

Dawson, G., Toth, K., Abbott, R., Osterling, J., Munson, J., Estes, A., & Liaw, J. (2004). Early social attention impairments in autism: Social orienting, joint attention, and attention to distress. *Developmental Psychology, 40*, 271–283.

Dumont, M., & Provost, M. A. (1999). Resilience in adolescents: Protective role of social support, coping strategies, self-esteem, and social activities on experience of stress and depression. *Journal of Youth and Adolescence, 28*, 343–363.

Dunlap, G., Dyer, K., & Koegel, R. L. (1983). Autistic self-stimulation and intertrial interval duration. *American Journal of Mental Deficiency, 88*, 194.

Durand, V. M., & Carr, E. G. (1987). Social influences on "self-stimulatory" behavior: Analysis and treatment application. *Journal of Applied Behavior Analysis, 20*, 119–132.

Durand, V. M., & Carr, E. G. (1992). An analysis of maintenance following functional communication training. *Journal of Applied Behavior Analysis, 25*, 777–794.

Farrugia, D. (2009). Exploring stigma: Medical knowledge and the stigmatisation of parents of children diagnosed with autism spectrum disorder. *Sociology of Health and Illness, 31*, 1011–1027.

Freitag, C. M., Konrad, C., Häberlen, M., Kleser, C., von Gontard, A., Reith, W., et al. (2008). Perception of biological motion in autism spectrum disorders. *Neuropsychologia, 46*, 1480–1494.

Gabriels, R. L., Agnew, J. A., Miller, L. J., Gralla, J., Pan, Z., Goldson, E., et al. (2008). Is there a relationship between restricted, repetitive, stereotyped behaviors and interests and abnormal sensory response in children with autism spectrum disorders? *Research in Autism Spectrum Disorders, 2*, 660–670.

Gabriels, R. L., Cuccaro, M. L., Hill, D. E., Ivers, B. J., & Goldson, E. (2005). Repetitive behaviors in autism: Relationships with associated clinical features. *Research in Developmental Disabilities, 26*, 169–181.

Geisthardt, C. L., Brotherson, M. J., & Cook, C. C. (2002). Friendships of children with disabilities in the home environment. *Education and Training in Mental Retardation and Developmental Disabilities, 37*, 235–252.

Georgiades, S., Szatmari, P., Boyle, M., Hanna, S., Duku, E., Zwaigenbaum, L., et al. (2013). Investigating phenotypic heterogeneity in children with autism spectrum disorder: A factor mixture modeling approach. *Journal of Child Psychology and Psychiatry, 54*, 206–215.

Ghanouni, P., Jarus, T., Zwicker, J., Lucyshyn, G., Chauhan, J., & Moir, S. (2019). Perceived barriers and existing challenges in participation of children with autism spectrum disorders: "He did not understand and no one else seemed to understand him". *Journal of Autism and Developmental Disorders, 49*, 3136–3145.

Gray, D. E. (1993). Perceptions of stigma: The parents of autistic children. *Sociology of Health and Illness, 15*, 102–120.

Gray, D. E. (2002). 'Everybody just freezes. Everybody is just embarrassed': Felt and enacted stigma among parents of children with high functioning autism. *Sociology of Health and Illness, 24*, 734–749.

Happé, F., & Frith, U. (1995). Theory of mind in autism. In *Learning and cognition in autism* (pp. 177–197). Boston, MA:Springer.

Happé, F., & Frith, U. (2006). The weak coherence account: Detail-focused cognitive style in autism spectrum disorders. *Journal of Autism and Developmental Disorders, 36*, 5–25.

Hilton, C. L., Crouch, M. C., & Israel, H. (2008). Out-of-school participation patterns in children with high-functioning autism spectrum disorders. *American Journal of Occupational Therapy, 62*, 554–563.

Joseph, R. M., & Tanaka, J. (2003). Holistic and part-based face recognition in children with autism. *Journal of Child Psychology and Psychiatry, 44*, 529–542.

Joyce, C., Honey, E., Leekam, S. R., Barrett, S. L., & Rodgers, J. (2017). Anxiety, intolerance of uncertainty and restricted and repetitive behaviour: Insights directly from young people with ASD. *Journal of Autism and Developmental Disorders, 47*, 3789–3802.

Kawachi, I., & Berkman, L. F. (2001). Social ties and mental health. *Journal of Urban Health, 78*, 458–467.

Kientz, M. A., & Dunn, W. (1997). A comparison of the performance of children with and without autism on the Sensory Profile. *American Journal of Occupational Therapy, 51*, 530–537.

Kinnear, S. H., Link, B. G., Ballan, M. S., & Fischbach, R. L. (2016). Understanding the experience of stigma for parents of children with autism spectrum disorder and the role stigma plays in families' lives. *Journal of Autism and Developmental Disorders, 46*, 942–953.

Klin, A., Danovitch, J. H., Merz, A. B., & Volkmar, F. R. (2007). Circumscribed interests in higher functioning individuals with autism spectrum disorders: An exploratory study. *Research and Practice for Persons with Severe Disabilities, 32*, 89–100.

Klin, A., Jones, W., Schultz, R., Volkmar, F., & Cohen, D. (2002). Visual fixation patterns during viewing of naturalistic social situations as predictors of social competence in individuals with autism. *Archives of General Psychiatry, 59*, 809–816.

Klin, A., Lin, D. J., Gorrindo, P., Ramsay, G., & Jones, W. (2009). Two-year-olds with autism orient to non-social contingencies rather than biological motion. *Nature, 459*, 257–261.

Koegel, R. L., & Covert, A. (1972). The relationship of self-stimulation to learning in autistic children. *Journal of Applied Behavior Analysis, 5*, 381–387.

Kozlowski, L. T., & Cutting, J. E. (1977). Recognizing the sex of a walker from a dynamic point-light display. *Perception & Psychophysics, 21*, 575–580.

Kuusikko, S., Haapsamo, H., Jansson-Verkasalo, E., Hurtig, T., Mattila, M.-L., Ebeling, H., et al. (2009). Emotion recognition in children and adolescents with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 39*, 938–945.

Kuusikko, S., Pollock-Wurman, R., Jussila, K., Carter, A. S., Mattila, M.-L., Ebeling, H., et al. (2008). Social anxiety in high-functioning children and adolescents with autism and Asperger syndrome. *Journal of Autism and Developmental Disorders, 38*, 1697–1709.

Langdell, T. (1978). Recognition of faces: An approach to the study of autism. *Journal of Child Psychology and Psychiatry, 19*, 255–268.

Lee, S., Odom, S. L., & Loftin, R. (2007). Social engagement with peers and stereotypic behavior of children with autism. *Journal of Positive Behavior Interventions, 9*, 67–79.

Lewis, M., & Kim, S. (2009). The pathophysiology of restricted repetitive behavior. *Journal of Neurodevelopmental Disorders, 1*, 114–132.

Lewis, M. H., Gluck, J. P., Beauchamp, T. L., Keresztury, M. F., & Mailman, R. B. (1990). Long-term effects of early social isolation in Macaca mulatta: changes in dopamine receptor function following apomorphine challenge. *Brain Research, 513*, 67.

Liss, M., Fein, D., Allen, D., Dunn, M., Feinstein, C., Morris, R., et al. (2001). Executive functioning in high-functioning children with autism. *The Journal of Child Psychology and Psychiatry and Allied Disciplines, 42*, 261–270.

Loftin, R. L., Odom, S. L., & Lantz, J. F. (2008). Social interaction and repetitive motor behaviors. *Journal of Autism and Developmental Disorders, 38*, 1124–1135.

Lopata, C., Volker, M. A., Putnam, S. K., Thomeer, M. L., & Nida, R. E. (2008). Effect of social familiarity on salivary cortisol and self-reports of social anxiety and stress in children with high functioning autism spectrum disorders. *Journal of Autism and Developmental Disorders, 38*, 1866–1877.

Lopez, B. R., Lincoln, A. J., Ozonoff, S., & Lai, Z. (2005). Examining the relationship between executive functions and restricted, repetitive symptoms of autistic disorder. *Journal of Autism and Developmental Disorders, 35*, 445–460.

Lord, C. (1995). Follow-up of two-year-olds referred for possible autism. *Journal of Child Psychology and Psychiatry, 36*, 1365–1382.

Maestro, S., Muratori, F., Cavallaro, M. C., Pei, F., Stern, D., Golse, B., & Palacio-Espasa, F. (2002). Attentional skills during the first 6 months of age in autism spectrum disorder. *Journal of the American Academy of Child & Adolescent Psychiatry, 41*, 1239–1245.

Markram, K., & Markram, H. (2010). The intense world theory–a unifying theory of the neurobiology of autism. *Frontiers in Human Neuroscience, 4*, 224.

Mason, G. J. (1991). Stereotypies: A critical review. *Animal Behaviour, 41*, 1015–1037.

Mather, G., & Murdoch, L. (1994). Gender discrimination in biological motion displays based on dynamic cues. *Proceedings of the Royal Society B, 258*, 273–279.

Mc Kinney, W. T., Jr. (1974). Animal models in psychiatry. *Perspectives in Biology and Medicine, 17*, 529–542.

Mccauley, J., Harris, M., Zajic, M., Swain-Lerro, L., Oswald, T., Mcintyre, N., et al. (2019). Self-esteem, internalizing symptoms, and theory of mind in youth with autism spectrum disorder. *Journal of Clinical Child & Adolescent Psychology, 48*, 400–411.

McIntosh, D. N., Reichmann-Decker, A., Winkielman, P., & Wilbarger, J. L. (2006). When the social mirror breaks: Deficits in automatic, but not voluntary, mimicry of emotional facial expressions in autism. *Developmental Science, 9*, 295–302.

McPartland, J., Dawson, G., Webb, S. J., Panagiotides, H., & Carver, L. J. (2004). Event-related brain potentials reveal anomalies in temporal processing of faces in autism spectrum disorder. *Journal of Child Psychology and Psychiatry, 45*, 1235–1245.

Mesman, J., Bongers, I. L., & Koot, H. M. (2001). Preschool developmental pathways to pre-adolescent internalizing and externalizing problems. *The Journal of Child Psychology and Psychiatry and Allied Disciplines, 42*, 679–689.

Meyer, J. A., Mundy, P. C., Van Hecke, A. V., & Durocher, J. S. (2006). Social attribution processes and comorbid psychiatric symptoms in children with Asperger syndrome. *Autism, 10*, 383–402.

Minio-Paluello, I., Baron-Cohen, S., Avenanti, A., Walsh, V., & Aglioti, S. M. (2009). Absence of embodied empathy during pain observation in Asperger syndrome. *Biological Psychiatry, 65*, 55–62.

Moore, V., & Goodson, S. (2003). How well does early diagnosis of autism stand the test of time? Follow-up study of children assessed for autism at age 2 and development of an early diagnostic service. *Autism, 7*, 47–63.

Moran, J. M., Young, L. L., Saxe, R., Lee, S. M., O'Young, D., Mavros, P. L., & Gabrieli, J. D. (2011). Impaired theory of mind for moral judgment in high-functioning autism. *Proceedings of the National Academy of Sciences, 108*, 2688–2692.

Notarius, C. I., & Levenson, R. W. (1979). Expressive tendencies and physiological response to stress. *Journal of Personality and Social Psychology, 37*(7), 1204–1210. https://doi.org/10.1037/0022-3514.37.7.1204.

Oke, N. J., & Schreibman, L. (1990). Training social initiations to a high-functioning autistic child: Assessment of collateral behavior change and generalization in a case study. *Journal of Autism and Developmental Disorders, 20*, 479–497.

Osterling, J., & Dawson, G. (1994). Early recognition of children with autism: A study of first birthday home videotapes. *Journal of Autism and Developmental Disorders, 24*, 247–257.

Ouellette-Kuntz, H., Coo, H., Lam, M., Breitenbach, M. M., Hennessey, P. E., Jackman, P. D., et al. (2014). The changing prevalence of autism in three regions of Canada. *Journal of Autism and Developmental Disorders, 44*, 120–136.

Pasalich, D. S., Dadds, M. R., & Hawes, D. J. (2014). Cognitive and affective empathy in children with conduct problems: Additive and interactive effects of callous–unemotional traits and autism spectrum disorders symptoms. *Psychiatry Research, 219*, 625–630.

Pavlova, M. A. (2011). Biological motion processing as a hallmark of social cognition. *Cerebral Cortex, 22*, 981–995.

Pérez Velázquez, J. L., & Galán, R. F. (2013). Information gain in the brain's resting state: A new perspective on autism. *Frontiers in Neuroinformatics, 7*, 37.

Pierce, K., & Courchesne, E. (2001). Evidence for a cerebellar role in reduced exploration and stereotyped behavior in autism. *Biological Psychiatry, 49*, 655–664.

Pierce, K., Glad, K. S., & Schreibman, L. (1997). Social perception in children with autism: An attentional deficit? *Journal of Autism and Developmental Disorders, 27*, 265–282.

Pollick, F. E., Paterson, H. M., Bruderlin, A., & Sanford, A. J. (2001). Perceiving affect from arm movement. *Cognition, 82*, B51–B61.

Repp, A., Singh, N. N., Karsh, K., & Deitz, D. (1991). Ecobehavioural analysis of stereotypic and adaptive behaviours: Activities as setting events. *Journal of Intellectual Disability Research, 35*, 413–429.

Rippon, G., Brock, J., Brown, C., & Boucher, J. (2007). Disordered connectivity in the autistic brain: Challenges for the 'new psychophysiology'. *International Journal of Psychophysiology, 63*, 164–172.

Richler, J., Huerta, M., Bishop, S., & Lord, C. (2010). Developmental trajectories of restricted and repetitive behaviors and interests in children with autism spectrum disorders. *Development and Psychopathology, 22*(1), 55–69. https://doi.org/10.1017/S0954579409990265.

Rowley, E., Chandler, S., Baird, G., Simonoff, E., Pickles, A., Loucas, T., & Charman, T. (2012). The experience of friendship, victimization and bullying in children with an autism spectrum disorder: Associations with child characteristics and school placement. *Research in Autism Spectrum Disorders, 6*, 1126–1134.

Rubenstein, J., & Merzenich, M. M. (2003). Model of autism: Increased ratio of excitation/inhibition in key neural systems. *Genes, Brain and Behavior, 2*, 255–267.

Runeson, S., & Frykholm, G. (1983). Kinematic specification of dynamics as an informational basis for person-and-action perception: Expectation, gender recognition, and deceptive intention. *Journal of Experimental Psychology: General, 112*, 585–615.

Sanchez Puerta, M. L., Valerio, A., & Bernal, M. G. (2016). *Taking stock of programs to develop socioemotional skills: A systematic review of program evidence*. Washington D.C: The World Bank.

Schultz, R. T. (2005). Developmental deficits in social perception in autism: The role of the amygdala and fusiform face area. *International Journal of Developmental Neuroscience, 23*, 125–141.

Senju, A. (2013). Atypical development of spontaneous social cognition in autism spectrum disorders. *Brain and Development, 35*, 96–101.

Solish, A., Perry, A., & Minnes, P. (2010). Participation of children with and without disabilities in social, recreational and leisure activities. *Journal of Applied Research in Intellectual Disabilities, 23*, 226–236.

South, M., Ozonoff, S., & Mcmahon, W. M. (2007). The relationship between executive functioning, central coherence, and repetitive behaviors in the high-functioning autism spectrum. *Autism, 11*, 437–451.

Sperry, L. A., & Mesibov, G. B. (2005). Perceptions of social challenges of adults with autism spectrum disorder. *Autism, 9*, 362–376.

Swettenham, J., Baron-Cohen, S., Charman, T., Cox, A., Baird, G., Drew, A., et al. (1998). The frequency and distribution of spontaneous attention shifts between social and nonsocial stimuli in autistic, typically developing, and nonautistic developmentally delayed infants. *The Journal of Child Psychology and Psychiatry and Allied Disciplines, 39*, 747–753.

Troje, N. F. (2002). Decomposing biological motion: A framework for analysis and synthesis of human gait patterns. *Journal of Vision, 2*, 371–387.

Turner, M. (1999). Annotation: Repetitive behaviour in autism: A review of psychological research. *The Journal of Child Psychology and Psychiatry and Allied Disciplines, 40*, 839–849.

Valkenburg, P. M., Peter, J., & Schouten, A. P. (2006). Friend networking sites and their relationship to adolescents' well-being and social self-esteem. *Cyberpsychology & Behavior, 9*, 584–590.

Wallace, S., Coleman, M., & Bailey, A. (2008). Face and object processing in autism spectrum disorders. *Autism Research, 1*, 43–51.

Watkins, L., O'Reilly, M., Kuhn, M., Gevarter, C., Lancioni, G. E., Sigafoos, J., & Lang, R. (2015). A review of peer-mediated social interaction interventions for students with autism in inclusive settings. *Journal of Autism and Developmental Disorders, 45*, 1070–1083.

Whitaker, P., Barratt, P., Joy, H., Potter, M., & Thomas, G. (1998). Children with autism and peer group support: Using 'circles of friends'. *British Journal of Special Education, 25*, 60–64.

Wigham, S., Rodgers, J., South, M., McConachie, H., & Freeston, M. (2015). The interplay between sensory processing abnormalities, intolerance of uncertainty, anxiety and restricted and repetitive behaviours in autism spectrum disorder. *Journal of Autism and Developmental Disorders, 45*, 943–952.

Wingate, M., Kirby, R., Pettygrove, S., Cunniff, C., Schulz, E., Ghosh, T., . . . Centers for Disease Control and Prevention (CDC). (2014). Prevalence of autism spectrum disorder among children aged 8 years — autism and developmental disabilities monitoring network, 11 sites, united states, 2010. *Morbidity and Mortality Weekly Report: Surveillance Summaries, 63*, 1–21.

Yoon, J. M., & Johnson, S. C. (2009). Biological motion displays elicit social behavior in 12-month-olds. *Child Development, 80*, 1069–1075.

Zikopoulos, B., & Barbas, H. (2013). Altered neural connectivity in excitatory and inhibitory cortical circuits in autism. *Frontiers in Human Neuroscience, 7*, 609.

Chapter 11
The Role of Anxiety Symptoms in Understanding Restricted, Repetitive Behaviours and Interests in Autism Spectrum Disorders

Ayelet Ben-Sasson and Kevin Stephenson

While repetitive and restrictive behaviors (RRBI) is a core symptom domain in ASD, anxiety-related disorders (ARDs) are considered to be co-ocurring. About 40% of children and adolescents with ASD meet criteria for at least one ARD (van Steensel, Bögels, & Perrin, 2011), with 63–87% of them having clinical anxiety symptoms and "atypical" anxiety symptoms (Kerns et al., 2014; Muris, Steerneman, Merckelbach, Holdrinet, & Meesters, 1998; Williams, Leader, Mannion, & Chen, 2015). These anxiety rates are higher than those reported among typically developing peers (Gadow, DeVincent, Pomeroy, & Azizian, 2004; Guttmann-Steinmetz, Gadow, DeVincent, & Crowell, 2010) or those with intellectual disabilities (Brereton, Tonge, & Einfeld, 2006). Co-occurring anxiety symptoms in ASD are linked with functional and health impairments, including increased self-injurious behavior (SIB), depressive symptoms, gastrointestinal difficulties, sleep problems, parental/family stress, and additional healthcare needs (Ahmedani & Hock, 2012; Bellini, 2006; Kerns et al., 2015; Williams et al., 2015). Anxiety can lead to greater stress, exacerbation of core ASD symptoms, and disruptive behaviors (Bos, Diamantopoulou, Stockmann, Begeer, & Rieffe, 2018). Anxiety and RRBI have been found to be interrelated in psychopathology, leading scholars to investigate the nature and underlying mechanisms of this association.

Anxiety symptoms and related disorders are often unidentified or misdiagnosed in ASD (Kerns et al., 2014; MacNeil, Lopes, & Minnes, 2009). This can be attributed to common negative affectivity indicators of both ARD and ASD such as distress, avoidance and withdrawal as well as obsessions and compulsions which are characteristics of both.

A. Ben-Sasson (✉)
Department of Occupational Therapy, University of Haifa, Haifa, Israel

K. Stephenson
Department of Pediatrics and Psychology, Nationwide Children's Hospital, Columbus, OH, USA

E. Gal, N. Yirmiya (eds.), *Repetitive and Restricted Behaviors and Interests in Autism Spectrum Disorders*, Autism and Child Psychopathology Series,
https://doi.org/10.1007/978-3-030-66445-9_11

Further challenging differential diagnosis of ASD from ARD is the fact that both children and adults with high anxiety symptoms have elevated scores on autism symptom measures (South, Carr, Stephenson, Maisel, & Cox, 2017; Van Steensel, Bögels, & Wood, 2013). This demonstrates how the interplay between core autism symptoms and anxiety occurs for individuals with ARDs and those with elevated anxiety symptoms and not only for those with ASD. Hence, it can be challenging to differentiate social avoidance associated with ASD versus social anxiety in ARD or to differentiate RRBI from obsessive compulsive disorder (OCD: Gjevik, Eldevik, Fjæran-Granum, & Sponheim, 2011; MacNeil et al., 2009).

RRBI are disruptive and provoke distress and anxiety (Wood & Gadow, 2010), limiting social participation (Ludlow, Skelly, & Rohleder, 2012) and learning (Leekam, Plissrior, & Uljarevic, 2011), and can therefore be extremely challenging for individuals with ASD and their families. Nevertheless, RRBI are addressed much less in ASD intervention programs than are social communication deficits. Understanding the role of anxiety in RRBI can advance our understanding of distinct clinical phenotypes in ASD and guide the search for common etiologies and for interventions that consider the interaction between core ASD symptoms and anxiety.

The specific aims of this chapter are to:

1. Characterize the **relation** between Anxiety and RRBI in ASD.
2. Outline **explanations** for the interplay between anxiety and RRBI in ASD.
3. Describe RRBI in **other ARDs**.
4. Discuss challenges in **identifying and differentiating** anxiety from RRBI in ASD.

RRBI in ASD

The DSM-5 (APA, 2013) classifies the RRBI domain into four types of symptoms: (1) Repetitive and stereotyped speech, movement or use of objects; (2) Routines, rituals and resistance to change; (3) Circumscribed and restricted interests, and (4) Hypo- or hyper-reactivity[1] to sensory input, including unusual sensory interests (hereafter referred to as sensory symptoms). Evidence (Bishop et al., 2013; Lidstone et al., 2014) demonstrates the distinction between two types of RRBI in ASD, Insistence on Sameness (IS, i.e., types (2) and (3) in the DSM-5) and Repetitive Sensory and Motor Behaviors (RSMB, i.e., types (1) and (4) in the DSM-5). IS refers to behavioral rigidity, resistance to change, practicing routines and rituals, and circumscribed and narrow areas of interest. RSMB[2] refer to body mannerisms, atypical sensory responses, and repetitive manipulation of objects.

[1] The term sensory hyper-reactivity will be used throughout the chapter to refer to atypical over-response to sensation. This type of RRBI has different terms in the literature such as sensory over-responsivity, sensory avoidance, sensory defensiveness, and sensory sensitivity.

[2] The RSMB construct in some measures does not include hypo- or hyper-reactivity symptoms rather focuses on repetitive movements and unusual exploration and seeking of stimulation. In

Anxiety Symptoms in ASD

Manifestations of anxiety symptoms in ASD can be classified as traditional or atypical. Traditional anxiety symptoms conform to DSM-5 ARD criteria (e.g., selective mutism, separation anxiety disorder, specific phobia, social anxiety disorder, panic disorder, agoraphobia, general anxiety disorder). These more traditional symptoms are characterized by elevated physiological arousal, which create a predisposition to anxiety (Bellini, 2006), and are associated with higher language abilities, anxious cognitive styles and sensory hyper-reactivity (considered part of the RRBI domain). Atypical anxiety is a maladaptive coping strategy (Hartley, Sikora, & McCoy, 2008; Spiker, Lin, Van Dyke, & Wood, 2012) which is also associated with an anxious cognitive style as well as ASD symptoms (Kerns et al., 2014), such as excessive worry around circumscribed interests (rather than generalized worry), fear related to novelty and change, social anxiety without fear of negative evaluation and unusual phobias (e.g., beards, toilet bowls). Note that in this chapter, in line with the reviewed literature, ARD will also refer to additional disorders which were excluded from the Anxiety Disorder section in the DSM-5, i.e., PTSD and OCD.

Evidence suggests that levels of anxiety symptoms in ASD change over development. Using cross-sectional data with individuals from 17 months to 65 years-of-age, Davis III et al. (2011) found a rise in anxiety from toddlerhood to childhood, reduced anxiety into early adulthood, and rising anxiety into older adulthood. Longitudinal investigations showed no significant changes in anxiety over short-term durations, such as one year (May, Cornish, & Rinehart, 2014), while a long-term follow-up study of individuals ages 6 through 24 years found a linear relation between age and anxiety with a significant interaction with biological sex. Females with ASD initially had less anxiety than males but by age 24 years, there was no significant difference between them (Gotham, Brunwasser, & Lord, 2015). Closer investigation of the course of anxiety in ASD throughout development at the population and individual levels can shed light on the path of their emergence.

The Association Between Symptoms of Anxiety and RRBI in ASD

Increased anxiety symptoms have been associated with more severe ASD symptoms (Wood & Gadow, 2010), and specifically with the RRBI core domain (e.g., Rodgers, Glod, Connolly, & McConachie, 2012). Some researchers show associations with anxiety across types of RRBI (Stratis & Lecavalier, 2013), while others focus on specific types of RRBI in relation with specific types of anxiety (e.g., Rodgers et al., 2012). The following reviewed evidence attests to differences in the nature of the association between anxiety and specific types of RRBI. Among RRBI, IS in

such cases the term repetitive movements will be used.

particular has been consistently associated with higher levels of anxiety (Gotham et al., 2013; Rodgers et al., 2012; Spiker et al., 2012; Stratis & Lecavalier, 2013). Individuals with both ASD and an ARD versus those with ASD but without ARD, had significantly greater severity of IS and repetitive movement symptoms. For the anxious subgroup of ASD, IS was specifically associated with separation anxiety and peer physical injury scores, but not with other types of anxiety symptoms (i.e., panic/agoraphobia, social phobia, OCD, generalized anxiety disorder) (Rodgers et al., 2012).

Another type of RRBI which is associated with anxiety in ASD across age groups is sensory hyper-reactivity (Ben-Sasson et al., 2008; Green, Ben-Sasson, Soto, & Carter, 2012). When comparing individuals with ASD who are anxious from individuals with ASD who are not anxious, for the non-anxious subgroup, more repetitive movements correlated with OCD symptoms but not with other types of anxiety (Rodgers et al., 2012). At the same time, it is important to understand whether the types of anxiety investigated can explain why in other studies repetitive movements were not associated with anxiety (Factor, Condy, Farley, & Scarpa, 2016; Stratis & Lecavalier, 2013).

The interaction between anxiety and RRBI in ASD appears to depend upon the type of anxiety and type of RRBI analyzed hence assessments should clearly define these types of symptoms. Although SIB are not part of the diagnostic criteria for ASD, they are thought of as repetitive behaviors in measures such as the Repetitive Behavior Scale-Revised (Lam & Aman, 2007). SIB occurs in 27% to 30% of individuals diagnosed with ASD and also appears in some cases of ARD (e.g., skin picking, hair pulling; Trepal & Wester, 2007). Evidence suggests that children with ASD who meet clinical cutoffs for ARD have significantly higher rates of SIB compared to children with ASD who do not meet criteria (Muskett, Capriola-Hall, Radtke, Factor, & Scarpa, 2019). Stratis and Lecavalier (2013) found that level of adaptive functioning moderated the association between SIB and anxiety in ASD. Specifically, more frequent SIB was predictive of higher anxiety among individuals with higher adaptive functioning, whereas SIB was predictive of less anxiety in the lower adaptive functioning group. Adaptive functioning is an important dimension to measure for fully understanding the interplay between RRBI and anxiety in ASD and its specific expression.

Explanations for the Association Between Anxiety and RRBI

Although the association between RRBI and anxiety in ASD is well-documented, the mechanisms explaining the direction of the effect are not agreed upon. Wood and Gadow (2010) questioned whether this association is a “true” comorbidity or “false”. “True” comorbidity refers to one of the following options: (1) anxiety is phenotypically and etiologically identical in comorbid (ASD and ARD) and monomorbid conditions (ARDs); (2) anxiety symptoms in ASD are phenotypically altered by ASD pathogenic processes; or (3) anxiety in ASD is a derivative of the

core ASD symptomatology with potentially different etiology than monomorbid ARDs. "False" comorbidity refers to an inaccurate diagnosis, which will be discussed in the Section "Challenges in Identification."

Sensory motor symptoms similar to those included under the RRBI ASD domain are prevalent among several psychopathologies including ASD, ARDs, OCD and ADHD (Fig. 11.1). Scholars have called attention to the nonspecific nature of these symptoms and their prodromal appearance. As such, these symptoms are described as indicators of brain vulnerability that put the individual at risk for developing psychopathology (Levit-Binnun, Davidovitch, & Golland, 2013). The cross-syndromic nature of the relation between anxiety and RRBI clearly raises questions regarding the common mechanism of this relation across conditions, as opposed to specific to ASD.

Next, three possible explanations for the co-occurrence of anxiety and RRBI symptoms in ASD and their supporting evidence will be outlined: **1. Anxiety causes RRBI; 2. RRBI causes anxiety; 3. Common mechanisms** (Fig. 11.2).

1. ***Anxiety causes RRBI***

The first explanation for the co-occurrence of anxiety and RRBI examined is controversial and suggests that anxiety motivates the emergence of RRBI. Within this explanation, the possibility that anxiety exacerbates RRBI will also be discussed. The mechanisms by which anxiety symptoms lead to RRBI also considers RRBI as a means for regulating emotional, social, and sensory experiences as described next.

Indeed, literature suggests that RRBI, particularly circumscribed interests and symbolic reenactment of restricted interests in play in ASD, serve as maladaptive coping mechanisms aimed to reduce anxiety (Rodgers et al., 2012; Spiker et al., 2012). This maladaptive cycle starts with RRBI providing immediate relief of anxiety by controlling the environment and creating predictable behavioral outcomes. As a result, the individual builds positive beliefs about the role and function of

Fig. 11.1 Overlapping disorders sharing anxiety, repetitive behaviors, and dysregulation

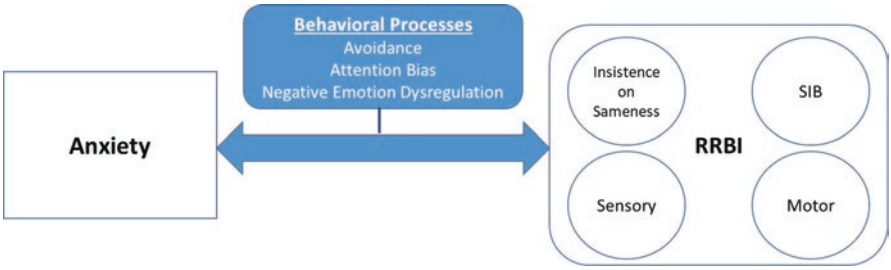

Fig. 11.2 The RRBI Anxiety Interplay Model

RRBI, further expanding and maintaining these behaviors. Consequently, the individual relies more on RRBI, further limiting his/her engagement, creating an anxiety cycle (Rodgers et al., 2012).

Anxiety can also explain the presence of sensory atypicalities that many people with ASD suffer from and are part of the DSM-5 definition of RRBI: Imagine yourself in a frightening situation in which all your senses are ready to fight or flight, you scan the area for potential threat and literally jump at any sound. According to this explanation, anxiety elicits sensory hyper-reactivity through hypervigilance (i.e., scanning the environment for threat-related stimuli) and attention biases. These, in turn, lead to focusing attention on stimuli and showing difficulty disengaging from it (Mobini & Grant, 2007).

In order to fully understand how anxiety can lead to RRBI, the behavioral manifestations of ARD must be examined. ARDs are also characterized by hyperarousal (i.e., elevated levels of autonomic responses) and poor regulation of negative emotions (Craske, 2003) which can lead to a dysregulated sensory reaction through attentional bias. Children who are hyper-aroused and constantly scan the environment for threat-related stimuli are more likely to notice sensory stimuli. This, coupled with poor emotional regulation, will exacerbate hyper-reactivity as the individual is more likely to attribute threat to these stimuli and will have difficulty disengaging from them and inhibiting negative affect (Green & Ben-Sasson, 2010). The path of anxiety leading to RRBI by hyperarousal and elevated attention to detail is supported by cluster analysis research pointing to a distinct subgroup of children with ASD who share sensory hyper-reactivity, excellent memory, over-focused and over-selective attention (Liss, Saulnier, Fein, & Kinsbourne, 2006). Sensory hyper-reactivity in this subgroup is conjectured to reflect increased likelihood of attending and disengaging from bothersome sensory stimuli.

Further support for the causal relation of anxiety leading to sensory hyper-reactivity comes from animal research. Mice, genetically inbred to be anxiety-prone showed poorer balance and postural control compared to non-anxious mice (Lepicard et al., 2003). These capacities are associated with poor proprioceptive and vestibular modulation observed in children with sensory hyper-reactivity (Miller, Anzalone, Lane, Cermak, & Osten, 2007). Examples from the auditory domain have been reported with mice genetically prone to anxiety, having stronger auditory startle reaction than non-anxious mice (Plappert & Pilz, 2002). Animal research

supports a directional relation between anxiety and sensory hyper-reactivity, whether it be through hyper-vigilance, attentional bias, hyper-arousal or dysregulation.

Furthermore, sensory avoidance was identified as mediating the influence of anxiety on RRBI among youth with ASD. Interestingly RSMB were not associated with anxiety, but with sensory avoidance (Lidstone et al., 2014). This finding supports earlier views of RRBI as an ongoing attempt to regulate arousal imbalance in ASD (Zentall & Zentall, 1983). In light of this, IS symptoms are understood as an attempt to minimize incoming sensory stimulation; however, leading to a maladaptive strategy by creating and/or maintaining anxiety. In contrast, RSMBs can be understood as an effective strategy for arousal regulation given their dissociation with anxiety.

C*lassical aversive conditioning* can account for the maintenance and exacerbation of sensory hyper-reactivity as opposed to its initiation. According to this mechanism, the aversive sensory stimulus (e.g., noise is the conditioned stimulus) is associated with a previously perceived neutral stimulus (e.g., bus). For instance, constantly scanning the environment for noise leads to a preference for attending to aversive, unexpected noise and a higher likelihood to over-react to noise (Green & Ben-Sasson, 2010). The conditioning can be strengthened by an individual's anxiety traits, physiological arousal and perceived uncontrollability over aversive events (Craske, 2003). Hence, a child with a pre-existing ARD is more likely to associate a physiological reaction with a sensory stimulus. This occurs when children with ARD regulate negative affectivity through avoidance of fear-eliciting stimuli (Craske, 2003). Such avoidance further decreases development of adaptive regulation of response. Therefore, sensory hyper-reactivity can result from hypervigilance, poor regulation, conditioning and avoidance; thus, maintaining and exacerbating the association.

2. ***RRBI causes anxiety***

This approach views anxiety as a consequence of RRBI and can be demonstrated through various pathways. The pervasive challenges that individuals with ASD experience as a result of their cognitive, sensory and social-communication deficits can lead to RRBI such as resistance to change, insistence on sameness, circumscribed interests (Greenway & Howlin, 2010) and ultimately to anxiety. This explanation views RRBI as a factor of ASD challenges and anxiety as an outcome of both.

A more direct model in line with this explanation was suggested by Wood and Gadow (2010). According to this model, anxiety and mood dysregulation occur as a result of ASD-related stressors, including unpredictability of social encounters, peer rejection/victimization, aversive sensory experiences, and inability to engage in preferred repetitive behaviors. The model further suggests that anxiety may be a consequence of ASD-specific symptoms and act as a moderator in increased ASD symptomatology, including repetitive behaviors.

Among RRBI, sensory hyper-reactivity has been proposed to trigger specific phobia or generalized anxiety through conditioning. The aversive sensory stimuli are associated with certain objects or situations, leading to the conditioning of these

objects or situations. In turn, the conditioned stimuli can cause anxiety in the absence of the feared stimuli (Green & Ben-Sasson, 2010). This *classical conditioning* mechanism may explain the emergence of specific phobia, which are prevalent in ASD (Gadow et al., 2004) and fits the conditioning hypothesis. At the same time, conditioning can explain the relation between sensory hyper-reactivity and generalized anxiety. Generalized fear is likely to occur when the unconditioned stimulus does not consistently occur with the conditioned stimulus, known as *context conditioning* (Grillon, 2008). Context conditioning often leads to behavioral avoidance, as a situation or location triggers the conditional fear as opposed to a specific object. For instance, a child may avoid malls or birthday parties because s/he anticipates noise. The more generalized the anxiety, the greater the impairment (Green & Ben-Sasson, 2010). Various factors contribute to the strength of the conditioning of a particular situation, including the frequency, degree of controllability and predictability of the event (Craske, 2003). In addition, the language and cognitive deficits of people with ASD may challenge one's ability to predict and control stressful situations. Frequent, uncontrolled and unpredicted conditioned stimuli may cause a child to become generally hypervigilant to sensory stimuli and to maintain hyperarousal, leading to a general state of anxiety (Green & Ben-Sasson, 2010). This explanation is supported by longitudinal research of a large sample of toddlers with ASD, which demonstrated that early sensory hyper-reactivity significantly predicted anxiety symptoms 18 months later, while early anxiety did not predict later hyper-reactivity (Green et al., 2012). These findings suggest that atypical sensory responses contribute to the development of anxiety symptoms in ASD, warranting further testing over a longer period. Another longitudinal study also provided evidence of unidirectional influence of repetitive speech and stereotypical behaviors on later anxiety symptoms. However, this effect was completely attenuated once early anxiety symptoms were controlled for (Teh, Chan, Tan, & Magiati, 2017). Furthermore, intervention research in ASD shows that anti-anxiety medications reduce RRBI (Hollander et al., 2012), further supporting the effect of anxiety on RRBI.

While theoretically, the two directional explanations discussed seem to be contradictory, they can also co-exist given their applicability to different types of RRBI. For example, IS dominated the explanation that anxiety causes RRBI. Both explanations clearly do not pertain to all individuals with ASD, as not all individuals with ASD have hyperarousal or sensory hyper-reactivity. In addition, it is possible for the direction of causality to differ among subgroups of ASD. Although directional mechanisms have been presented, the direction of the relation may indeed be circular in that RRBI can lead to distress and promote anxiety, and heightened anxiety can further increase severity of ASD symptoms.

3. ***Shared Mechanisms***

As opposed to one construct being secondary to the other, it is plausible that a third shared mechanism explains the expression of both constructs. Two potential shared mechanisms will be discussed, mechanisms that may explain both anxiety and RRBI, one relates to common mediators and the other to a neurobiological mechanism.

Third Party Factors

Some constructs were identified in the literature as mediators of the impact of RRBI and anxiety. Such a construct is intolerance of uncertainty (IU), which refers to the difficulty to endure uncertain situations. Underlying IU is the perception that uncertainty is stressful and upsetting and unexpected events are negative and should be avoided at all costs (Rodgers et al., 2012). Two key factors underlie IU: *desire for predictability*, which refers to disliking unexpected events and *uncertainty paralysis*, which refers to feeling stuck when experiencing uncertainty (Birrell, Meares, Wilkinson, & Freeston, 2011). Both factors resonate with the nature of ASD: rigidity, difficulty with changes, and need for sameness. Insistence on sameness has been hypothesized as a strategy to reduce distress caused by IU. This can explain findings of increased IS observed over time among individuals with ASD (Richler, Huerta, Bishop, & Lord, 2010). Within ASD, symptoms of worry, IU, and repetitive behaviors have been negatively associated with startle response during an uncertain potentiated startle task (Chamberlain et al., 2013). Furthermore, IU has been shown to mediate the association between broad autism symptoms and degree of anxiety in children and adolescents with ASD (Boulter, Freeston, South, & Rodgers, 2014), as well as in adults with or without ASD (Maisel et al., 2016). However, the exact contribution of IU as a specific mediator between RRBI and anxiety is less clear. Uljarević, Carrington, and Leekam (2016) report that IU partially mediated the association between sensory sensitivity and anxiety in mothers of children with ASD. Neil, Olsson, and Pellicano (2016) investigated these variables in children with and without ASD using a model of anxiety to mediate the link between sensory sensitivity and IU and found that anxiety acted as a partial mediator for the ASD group, but not in the typically developing group. This mediation supports the conjecture that the nature of the interplay between anxiety and RRBI is unique to ASD. Additional research is needed to illuminate the mediating role of IU in explaining the interplay of RRBI and anxiety.

Another ASD related factor which can explain both anxiety and RRBI is social motivation. Low social motivation is associated with elevated anxiety symptoms and emotion dysregulation (Swain, Scarpa, White, & Laugeson, 2015). Findings point that social motivation deficits in children and adolescents with ASD partially mediated the relation between anxiety and IS (Factor et al., 2016). This may occur through low social reward reducing social motivation, leading to seeking non-social rewards in the form of RRBI. This strengthens the differential mechanism of association between anxiety and IS versus RSMB and introduces the contribution of social deficits to the model (Fig. 11.2).

The Neurobiology of Anxiety in ASD

It is thought that individuals with ASD and anxiety symptoms constitute a unique ASD subgroup, which is biologically different from those with ASD but without anxiety (Wood & Gadow, 2010). This position views the comorbidity of anxiety

symptoms and ARDs in ASD as a distinct endophenotype. Herrington and colleagues investigated the role of the amygdala in individuals with ASD with and without significant anxiety symptoms. They found evidence for associations between anxiety and reduced amygdala volume (Herrington et al., 2017) and greater amygdala activation (Herrington et al., 2017). Basal ganglia regions have also been identified in repetitive motor behaviors in both people with OCD and ASD (Estes et al., 2011; Langen et al., 2014). In addition, deep brain stimulation of the nucleus accumbens in the basal ganglia has been shown to decrease anxiety symptoms in individuals with treatment-refractory OCD (Denys et al., 2010). This can explain the similarity between OCD and ASD symptoms and their high rate of co-occurrence.

There is also evidence of disrupted neural circuits in both anxiety and repetitive behaviors. The Pre-Frontal Cortex (PFC) is thought to be involved in down-regulation of amygdala activity during emotional regulation (Wager, Davidson, Hughes, Lindquist, & Ochsner, 2008) and abnormal (often reduced) amygdala-PFC connectivity has been implicated in ARDs (Makovac et al., 2016). Amygdala-PFC connectivity has been found to be reduced in one study (Swartz, Wiggins, Carrasco, Lord, & Monk, 2013) and increased in another (Monk et al., 2010) in ASD, and it is unclear whether this is due to upregulation or to a possible inability to down-regulate. Additional research is needed to better understand how brain systems are related to anxiety and repetitive behaviors in ASD, but the current literature indicates that underlying shared mechanisms are possible.

Genetic research reveals potential overlaps between anxiety and RRBI as well. Various adenosine A_{2A} receptor gene (*ADORA2A*) variants have been associated with anxiety and increased ASD symptoms (including repetitive behaviors; Freitag et al., 2010). Gadow, Roohi, DeVincent, Kirsch, and Hatchwell (2009) reported preliminary findings of a shared association between brain-derived neurotrophic factor (*BDNF*) polymorphisms and both social anxiety and repetitive (tic) behaviors in ASD. Glutamate transporter gene (*SLC1A1*) allelic variation was, however, associated with anxiety, but not with repetitive behaviors, in children with ASD (Gadow, Roohi, DeVincent, Kirsch, & Hatchwell, 2010), although *SLC1A1* polymorphisms have been associated with basal ganglia-mediated activity and repetitive behaviors in OCD (Zike et al., 2017) and may represent a shared risk-factor for anxiety and repetitive behaviors. Oxcytocin is a neuropeptide with hypothesized roles in the development of ASD (Kranz et al., 2016) but its role in RRBI is unclear as on one hand, Oxytocin infusion has been associated with a decrease in repetitive behaviors in adults with ASD (Hollander et al., 2003), but on the other hand, no significant associations were reported between Oxcytocin and repetitive behavior severity in a recent meta-analysis (Kranz et al., 2016).

Repetitive Behaviors in Anxiety Related Disorders

This section reviews RRBI that characterize ARDs (e.g., general anxiety disorder, social phobia, panic disorder, PTSD) and specifically OCD. OCD is a family of disorders defined by obsessive recurrent thoughts and compulsive behaviors. The

compulsions often aim to reduce anxiety and avoid feared situation (APA, 2013). Individuals with ARDs do not typically present with the classical RRBI associated with ASD, rather anxious individuals show other forms of repetitive behaviors and thoughts. These are important to recognize, as their manifestation in ASD may alert professionals to consider the presence of anxiety. For example, abnormal sensory functioning is an RRBI commonly seen outside ASD in disorders such as OCD (Rieke & Anderson, 2009) and other ARDs (Conelea, Carter, & Freeman, 2014). Cognitive symptoms of anxiety can also assist in identifying anxiety in ASD. Cognitive inflexibility, is an example of a symptom which is present in ASD (Leung & Zakzanis, 2014), and plays a role in the development and maintenance of generalized anxiety disorder (Lee & Orsillo, 2014) hence can potentially distinguish those with ASD who also present with anxiety traits. In addition, individuals with ARDs suffer from perseverative negative thoughts, which can lead to negative affectivity and avoidance (Sorg, Vögele, Furka, & Meyer, 2012). Thus, signs of negative affectivity and avoidance in ASD may stem from repetitive thoughts associated with their anxiety.

ASD and OCD both have repetitive all-consuming thoughts that lead to functional impairment, engagement in rituals and repetitive actions, and sensory sensitivities. While 37.2% of individuals with ASD also meet criteria for OCD (Leyfer et al., 2006), 20% of individuals with OCD show autistic traits (Bejerot, Nylander, & Lindström, 2001). The similarity between ASD and OCD can lead to misdiagnosis of OCD among individuals with ASD (Bejerot et al., 2001).

One of the hurdles for accurate evaluation of RRBI which coincides with ARD is use of different terminologies across disciplines and populations to describe similar symptoms. Table 11.1 presents a comparison of RRBI terms used in the field of ASD and OCD versus other ARDs. For instance, while hyper- and hypo-reactivity to sensation as well as abnormal sensory cravings and interests are referred to in ASD (Ben-Sasson et al., 2019), sensory phenomena together with "Not Just Right Experiences" describe sensory abnormalities in those with OCD. Cross-syndromic research and training would advance the field by leading to unified terminology and raise awareness of parallel symptoms when careful differential diagnosis is warranted.

Therefore, it is of importance to identify common and differentiating features of RRBI in these disorders as it can facilitate distinct measurements and guide the discovery of common etiologies. Indeed, various researchers compared ASD, OCD, and anxiety symptoms between individuals with pure OCD or ASD (Cath, Ran, Smit, Van Balkom, & Comijs, 2008; Jiujias, Kelley, & Hall, 2017; McDougle, Kresch, Goodman, & Naylor, 1995; Russell, Mataix-Cols, Anson, & Murphy, 2005; Zandt, Prior, & Kyrios, 2007) and are reviewed in the following section:

ASD and OCD Similarities

1. Level of general anxiety symptoms (Cath et al., 2008).
2. High-level repetitive behaviors: obsessions, insistence on sameness, fixated interests; and low-level repetitive behaviors: compulsions, repetitive sensory and motor behaviors (Jiujias et al., 2017).

Table 11.1 Comparison of RRBI Terms between Disorders

RRBI Term	ASD	OCD	ARDs
Repetitive Thoughts	Insistence on sameness, restricted and fixated interests, rigid thinking patterns	Obsessions	Ruminative/ perseverative negative thoughts, cognitive rigidity
Repetitive Behaviors	Ritualized behaviors, strict adherence to routines, stereotypic movements, echolalia, idiosyncratic speech, nonfunctional play	Compulsions, tics, body-focused repetitive behaviors	Behavioral avoidance
Abnormal Sensory Functioning	Hyper- or hypo-reactivity to or abnormal interest in sensory stimuli	"Just right experiences", Sensory phenomena	Sensory-processing sensitivity; harm avoidance

3. The presentation of repetitive behaviors becomes more complex over time as well as possible decreases in low-level versus high-level behaviors (Zandt et al., 2007).
4. Children with OCD have similar levels of more traditional ASD-related repetitive behaviors as those with ASD, as measured by the RBQ, including sameness behavior and repetitive movements (Zandt et al., 2007).
5. A comorbid ASD and OCD group and a pure OCD group demonstrated similar degrees of increased deficits in social skills and attention to detail on the Autism Quotient (Cath et al., 2008).

ASD and OCD Differences

1. Higher severity of OCD symptoms in a pure OCD group than in a comorbid ASD and OCD group (Cath et al., 2008; Russell et al., 2005). Lower obsession scores in the comorbid group contributed to these findings (Cath et al., 2008).
2. A comorbid ASD and OCD group was differentiated by hoarding, touching, tapping, self-injurious behaviors (McDougle et al., 1995), somatic obsessions and repetitive rituals (Russell et al., 2005), and more sexual obsessions (Russell et al., 2005).
3. Absence of checking, counting, aggressive and symmetry-related repetitive thoughts in an ASD group (McDougle et al., 1995).
4. A comorbid ASD and OCD had lower communication, imagination, and attention switching scores than a pure OCD group did (Cath et al., 2008).

A few observations can assist in differentiating between ASD-based versus OCD-based RRBI. First, the child's associated emotional reactions as opposed to identifying the behavior itself should be examined: While in ASD, obsessive thoughts in a narrow area of interest pose a pleasant experience, in OCD they are usually unpleasant and associated with harm/threat. Second, it is important to

identify whether compulsive behavior is linked to a particular obsession, which is more characteristic of OCD than ASD.

Furthermore, similar to ASD, atypical sensory responses were reported in OCD (Lewin, Wu, Murphy, & Storch, 2015; Rieke & Anderson, 2009), and abnormal sensory phenomena have been specifically associated with repeated behaviors in OCD (Ferrão et al., 2012). However, no study has included a comparison of ASD and OCD samples in terms of their sensory abnormalities. In Fig. 11.3, the percentage of extreme scores on the Sensory Profile sensory symptom questionnaire are compared based on published data from two studies (Clince, Connolly, & Nolan, 2016; Rieke & Anderson, 2009). The largest gaps are in the lower rates of hyporeactivity (called low registration in measure) in OCD versus ASD and the higher rates of sensory seeking in OCD versus ASD. Further research can help determine whether the nature and interference rather than frequency of sensory aversions differ among groups.

There is scarce evidence which relates to comparing ASD and OCD conflicts, potentially due to variations in inclusion criteria for ASD. For example, some researchers focused on higher functioning individuals with ASD (Russell et al., 2005), while others included various IQ levels (McDougle et al., 1995). These methodological disparities between studies can explain the contrasting findings in repetitive behavior and anxiety between groups.

The comorbidity rates of ASD and OCD may be inflated by the similarity between symptoms reflecting RRBI and OCD (Wood & Gadow, 2010; Zandt et al., 2007). As OCD and ASD share features of repetition and compulsion, careful differentiation between these disorders is needed. The behavioral differences listed above can assist in such differentiation.

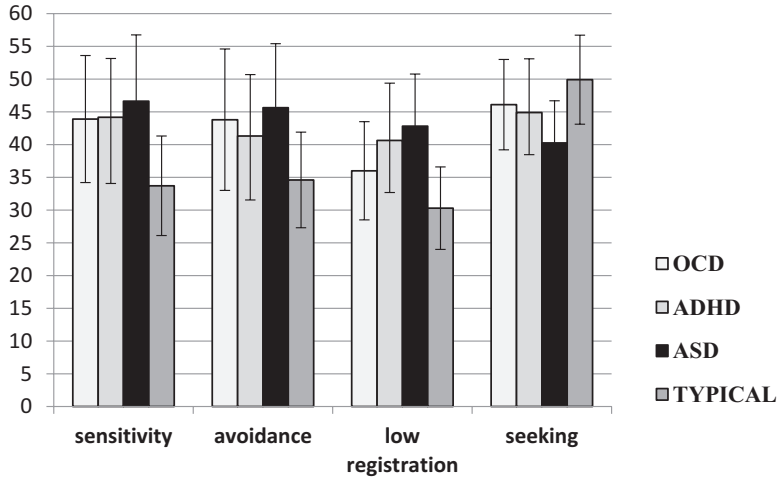

Fig. 11.3 Percentage of Extreme Sensory Profile Questionnaire Scores across Disorders

Challenges in Identifying and Differentiating RRBI from Anxiety in ASD

We next describe the challenges in measuring RRBI and ARDs in order to distinguish RRBI from ARDs in ASD, including: (1) Low language, communication and developmental levels. (2) Overlap between RRBI and anxiety symptoms among various scales. (3) Specificity of RRBI and anxiety measures. And (4) suitability of anxiety measures for evaluation in ASD. These measurement issues led scholars to postulate that the comorbidity of RRBI and ARDs is in some cases 'false' or artifactual due to inaccurate differential or dual diagnoses (Wood & Gadow, 2010). Furthermore, the overlapping symptoms between ASD and other psychiatric disorders reflect discontinuity in classification systems between child and adult psychiatry, leading to lack of awareness in adult psychiatry of the manifestations of psychiatric disorders in childhood (Cath et al., 2008).

1. ***Low language, communication and developmental levels:***

Assessment of anxiety symptoms relies on identifying negative and obsessive thought patterns. Hence, it is difficult to identify cognitive features of anxiety in pre-verbal children or those with severe language and communication impairments. In the absence of the capacity to share their thoughts with the examiner, the assessment must rely on proxy reports and/or interpretation of the source(s) of distressed behavior. Furthermore, developmental delay in ASD hinders the collection of valid diagnostic information from the individual and separating anxiety symptoms from global delay. In addition, many individuals with ASD often experience difficulty in directed expression of emotions and others may find it difficult to interpret their emotional expressions (Davis, Saeed, & Antonacci, 2008). The difficulty parents or other observers have in reading behaviors in ASD may contribute to inaccurate identification of sources of distress.

2. ***Overlap between symptoms of RRBI and anxiety in different scales:***

Evaluating the association between anxiety and RRBI is obscured by overlapping symptoms in measurement tools. Some scholars view anxiety indicators in ASD as a proxy of RRBI core symptoms. In a theoretical model of clinical anxiety in ASD, ASD-related stressors such as restricting engagement in repetitive behavior, social unpredictability, and sensory aversive experiences, are displayed as triggers of elevated anxiety symptoms. As such, specific ASD core symptoms may be misidentified as signs of an ARD rather than part of the ASD phenotype (Wood & Gadow, 2010). General descriptors of distress, avoidance, anger and hyperactivity are examples of behaviors described in both anxiety and RRBI. More specific indicators, such as lining-up objects and self-injury appear in scales of both domains. As a result, some indicators may be misidentified as anxiety symptoms when they are indeed a reflection of core ASD symptomatology and vice versa.

This measurement overlap may reflect conceptual differences among the various professions designing RRBI versus anxiety measures. Evidence indicates the tendency of professionals from different disciplines to interpret similar behaviors as

representing different constructs based on their training and expertise. In a study comparing views of occupational therapists and psychologists, they were asked to rate behaviors from early childhood standardized assessments as representing sensory versus anxiety symptoms. Findings showed that occupational therapists tended to rate items as representing sensory abnormalities, while psychologists tended to rate items as representing anxiety, regardless of the items' original construct (Ben-Sasson, Cermak, Orsmond, Carter, & Fogg, 2007). These findings highlight the need for interdisciplinary assessment to obtain an objective, accurate diagnosis of ARDs in ASD; one that doesn't depend on professional background or the measure used. This also calls for tools with strong divergent validity, demonstrating their utility for identifying 'true' comorbid ARD and RRBI in ASD.

3. ***Specificity of RRBI and anxiety measures:***

The types of anxiety and RRBI measures used in research shape the presence and nature of the association between these constructs. In one study, early sensory hyper-reactivity was found to predict later anxiety symptoms in ASD (Green et al., 2012). Duvekot, van der Ende, Verhulst, and Greaves-Lord (2018) did not find a relation between a broader RRBI score and later anxiety. These results may reflect that the measures of anxiety and RRBI in the latter did not distinguish between subtypes of symptoms within each construct. Research indicating differing patterns of association of anxiety with IS versus RSMB (Rodgers et al., 2012; Spiker et al., 2012) calls for their differentiation in measurement. Multifaceted measures of symptoms (rather than broad symptom measures/screens) in investigations of the relationship between RRBI and anxiety are needed.

4. ***Suitability of anxiety measure for evaluation in ASD:***

Traditional anxiety tools do not sufficiently quantify diffuse anxiety symptoms such as those associated with the atypical anxiety present in ASD. This may explain differences in rates of anxiety reported across ASD studies (Kerns et al., 2014). When selecting an anxiety tool for individuals with ASD, one should be aware of its suitability for quantifying anxiety in ASD as opposed to assessing the diathesis of social-communication ASD symptoms. Empirically using poorly differentiated tools requires understanding that some of the correlation between the constructs is accounted for by measurement overlap. There is a call to select anxiety measures that are suitable for evaluating anxiety in ASD by minimizing overlapping indicators (Wood & Gadow, 2010).

Summary

Looking at the interplay of anxiety and RRBI contributes immensely to understanding the functional impairments people with ASD experience. Recognizing the role of anxiety and RRBI in the emergence, maintenance and/or exacerbation of each other can inform research and clinical attempts to reveal their underlying

mechanisms. For some individuals with ASD anxiety can explain the nature of their RRBI, while for others RRBI causes anxiety. Common neurobiological indicators of anxiety and RRBI as well as shared mediating factors strengthen the notion of a common ground rather than coincidental co-occurrence. Such evidence is also promising when thinking of behavioral and pharmaceutical treatments for targeting both conditions. There is need to ensure that anxiety in young and non-verbal individuals with ASD is not misdiagnosed and to develop tools which can assist professionals' identification of underlying anxiety symptoms in ASD.

References

Ahmedani, B. K., & Hock, R. M. (2012). Health care access and treatment for children with comorbid autism and psychiatric conditions. *Social Psychiatry and Psychiatric Epidemiology, 47*, 1807–1814.

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). Washington, DC: American Psychiatric Association.

Bejerot, S., Nylander, L., & Lindström, E. (2001). Autistic traits in obsessive-compulsive disorder. *Nordic Journal of Psychiatry, 55*(3), 169–176.

Bellini, S. (2006). The development of social anxiety in adolescents with autism spectrum disorders. *Focus on Autism and Other Developmental Disabilities, 21*, 138–145.

Ben-Sasson, A., Cermak, S. A., Orsmond, G. I., Carter, A. S., & Fogg, L. (2007). Can we differentiate sensory over-responsivity from anxiety symptoms in toddlers? Perspectives of occupational therapists and psychologists. *Infant Mental Health Journal, 28*(5), 536–558.

Ben-Sasson, A., Cermak, S. A., Orsmond, G. I., Tager-Flusberg, H., Kadlec, M. B., & Carter, A. S. (2008). Sensory clusters of toddlers with autism spectrum disorders: Differences in affective symptoms. *Journal of Child Psychology and Psychiatry, 49*(8), 817–825.

Ben-Sasson, A., Gal, E., Fluss, R., Katz-Zetler, N., & Cermak, S. A. (2019). Update of a meta-analysis of sensory symptoms in ASD: A new decade of research. *Journal of Autism and Developmental Disorders*, *49*(12), 4974–4996.

Birrell, J., Meares, K., Wilkinson, A., & Freeston, M. (2011). Toward a definition of intolerance of uncertainty: A review of factor analytical studies of the Intolerance of Uncertainty Scale. *Clinical Psychology Review, 31*(7), 1198–1208.

Bishop, S. L., Hus, V., Duncan, A., Huerta, M., Gotham, K., Pickles, A., & Lord, C. (2013). Subcategories of restricted and repetitive behaviors in children with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 43*(6), 1287–1297.

Bos, M. G., Diamantopoulou, S., Stockmann, L., Begeer, S., & Rieffe, C. (2018). Emotion control predicts internalizing and externalizing behavior problems in boys with and without an autism spectrum disorder. *Journal of Autism and Developmental Disorders, 48*, 2727–2739.

Boulter, C., Freeston, M., South, M., & Rodgers, J. (2014). Intolerance of uncertainty as a framework for understanding anxiety in children and adolescents with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 44*(6), 1391–1402.

Brereton, A. V., Tonge, B. J., & Einfeld, S. L. (2006). Psychopathology in children and adolescents with autism compared to young people with intellectual disability. *Journal of Autism and Developmental Disorders, 36*, 863–870.

Cath, D. C., Ran, N., Smit, J. H., Van Balkom, A. J., & Comijs, H. C. (2008). Symptom overlap between autism spectrum disorder, generalized social anxiety disorder and obsessive-compulsive disorder in adults: A preliminary case-controlled study. *Psychopathology, 41*(2), 101–110.

Chamberlain, P. D., Rodgers, J., Crowley, M. J., White, S. E., Freeston, M. H., & South, M. (2013). A potentiated startle study of uncertainty and contextual anxiety in adolescents diagnosed with autism spectrum disorder. *Molecular Autism, 4*(1), 31.

Clince, M., Connolly, L., & Nolan, C. (2016). Comparing and exploring the sensory processing patterns of higher education students with attention deficit hyperactivity disorder and autism spectrum disorder. *American Journal of Occupational Therapy, 70*(2), 7002250010p1-7002250010p9.

Conelea, C. A., Carter, A. C., & Freeman, J. B. (2014). Sensory over-responsivity in a sample of children seeking treatment for anxiety. *Journal of Developmental and Behavioral Pediatrics, 35*(8), 510.

Craske, M. G. (2003). *Origins of phobias and anxiety disorders: Why more women than men?* Oxford, UK: Elsevier.

Davis, E., Saeed, S. A., & Antonacci, D. J. (2008). Anxiety disorders in persons with developmental disabilities: Empirically informed diagnosis and treatment. *Psychiatry Quarterly, 79*, 249–263.

Davis, T. E., III, Hess, J. A., Moree, B. N., Fodstad, J. C., Dempsey, T., Jenkins, W. S., & Matson, J. L. (2011). Anxiety symptoms across the lifespan in people diagnosed with autistic disorder. *Research in Autism Spectrum Disorders, 5*(1), 112–118.

Denys, D., Mantione, M., Figee, M., van den Munckhof, P., Koerselman, F., Westenberg, H., Bosch, A., & Schuurman, R. (2010). Deep brain stimulation of the nucleus accumbens for treatment-refractory obsessive-compulsive disorder. *Archives of General Psychiatry, 67*(10), 1061–1068.

Duvekot, J., van der Ende, J., Verhulst, F. C., & Greaves-Lord, K. (2018). Examining bidirectional effects between the autism spectrum disorder (ASD) core symptom domains and anxiety in children with ASD. *Journal of Child Psychology and Psychiatry, 59*(3), 277–284.

Estes, A., Shaw, D. W., Sparks, B. F., Friedman, S., Giedd, J. N., Dawson, G., Bryan, M., & Dager, S. R. (2011). Basal ganglia morphometry and repetitive behavior in young children with autism spectrum disorder. *Autism Research, 4*(3), 212–220.

Factor, R. S., Condy, E. E., Farley, J. P., & Scarpa, A. (2016). Brief report: Insistence on sameness, anxiety, and social motivation in children with autism spectrum disorder. *Journal of Autism and Developmental Disorders, 46*(7), 2548–2554.

Ferrão, Y. A., Shavitt, R. G., Prado, H., Fontenelle, L. F., Malavazzi, D. M., de Mathis, M. A., Hounie, A. G., Miguiel, E. C., & do Rosário, M. C. (2012). Sensory phenomena associated with repetitive behaviors in obsessive-compulsive disorder: An exploratory study of 1001 patients. *Psychiatry Research, 197*(3), 253–258.

Freitag, C. M., Agelopoulos, K., Huy, E., Rothermundt, M., Krakowitzky, P., Meyer, J., Deckert, J., von Gontard, A., & Hohoff, C. (2010). Adenosine A 2A receptor gene (ADORA2A) variants may increase autistic symptoms and anxiety in autism spectrum disorder. *European Child and Adolescent Psychiatry, 19*(1), 67–74.

Gadow, K. D., DeVincent, C. J., Pomeroy, J., & Azizian, A. (2004). Psychiatric symptoms in preschool children with PDD and clinic and comparison samples. *Journal of Autism and Developmental Disorders, 34*(4), 379–393.

Gadow, K. D., Roohi, J., DeVincent, C. J., Kirsch, S., & Hatchwell, E. (2009). Association of COMT (Val158Met) and BDNF (Val66Met) gene polymorphisms with anxiety, ADHD and tics in children with autism spectrum disorder. *Journal of Autism and Developmental Disorders, 39*(11), 1542–1551.

Gadow, K. D., Roohi, J., DeVincent, C. J., Kirsch, S., & Hatchwell, E. (2010). Brief report: Glutamate transporter gene (SLC1A1) single nucleotide polymorphism (rs301430) and repetitive behaviors and anxiety in children with autism spectrum disorder. *Journal of Autism and Developmental Disorders, 40*(9), 1139–1145.

Gjevik, E., Eldevik, S., Fjæran-Granum, T., & Sponheim, E. (2011). Kiddie-SADS reveals high rates of DSM-IV disorders in children and adolescents with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 41*(6), 761–769.

Gotham, K., Bishop, S. L., Hus, V., Huerta, M., Lund, S., Buja, A., Krieger, A., & Lord, C. (2013). Exploring the relationship between anxiety and insistence on sameness in autism spectrum disorders. *Autism Research, 6*(1), 33–41.

Gotham, K., Brunwasser, S. M., & Lord, C. (2015). Depressive and anxiety symptom trajectories from school age through young adulthood in samples with autism spectrum disorder and developmental delay. *Journal of the American Academy of Child & Adolescent Psychiatry, 54*(5), 369–376.

Green, S. A., & Ben-Sasson, A. (2010). Anxiety disorders and sensory over-responsivity in children with autism spectrum disorders: Is there a causal relationship? *Journal of Autism and Developmental Disorders, 40*(12), 1495–1504.

Green, S. A., Ben-Sasson, A., Soto, T. W., & Carter, A. S. (2012). Anxiety and sensory over-responsivity in toddlers with autism spectrum disorders: Bidirectional effects across time. *Journal of Autism and Developmental Disorders, 42*(6), 1112–1119.

Greenway, R., & Howlin, P. (2010). Dysfunctional attitudes and perfectionism and their relationship to anxious and depressive symptoms in boys with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 40*(10), 1179–1187.

Grillon, C. (2008). Models and mechanisms of anxiety: Evidence from startle studies. *Psychopharmacology, 199*, 421–437.

Guttmann-Steinmetz, S., Gadow, K. D., DeVincent, C. J., & Crowell, J. (2010). Anxiety symptoms in boys with autism spectrum disorder, attention-deficit hyperactivity disorder, or chronic multiple tic disorder and community controls. *Journal of Autism and Developmental Disorders, 40*(8), 1006–1016.

Hartley, S. L., Sikora, D. M., & McCoy, R. (2008). Prevalence and risk factors of maladaptive behaviour in young children with autistic disorder. *Journal of Intellectual Disability Research, 52*(10), 819–829.

Herrington, J. D., Maddox, B. B., Kerns, C. M., Rump, K., Worley, J. A., Bush, J. C., McVey, A. J., Schultz, R. T., & Miller, J. S. (2017). Amygdala volume differences in autism spectrum disorder are related to anxiety. *Journal of Autism and Developmental Disorders, 47*(12), 3682–3691.

Herrington, J. D., Maddox, B. B., McVey, A. J., Franklin, M. E., Yerys, B. E., Miller, J. S., & Schultz, R. T. (2017). Negative valence in autism spectrum disorder: The relationship between amygdala activity, selective attention, and co-occurring anxiety. *Biological Psychiatry: Cognitive Neuroscience and Neuroimaging, 2*(6), 510–517.

Hollander, E., Novotny, S., Hanratty, M., Yaffe, R., DeCaria, C. M., Aronowitz, B. R., & Mosovich, S. (2003). Oxytocin infusion reduces repetitive behaviors in adults with autistic and Asperger's disorders. *Neuropsychopharmacology, 28*(1), 193.

Hollander, E., Soorya, L., Chaplin, W., Anagnostou, E., Taylor, B. P., Ferretti, C. J., Wasserman, S., Swanson, E., & Settipani, C. (2012). A double-blind placebo-controlled trial of fluoxetine for repetitive behaviors and global severity in adult autism spectrum disorders. *American Journal of Psychiatry, 169*(3), 292–299.

Jiujias, M., Kelley, E., & Hall, L. (2017). Restricted, repetitive behaviors in autism spectrum disorder and obsessive–compulsive disorder: A comparative review. *Child Psychiatry and Human Development, 48*(6), 944–959.

Kerns, C. M., Kendall, P. C., Berry, L., Souders, M. C., Franklin, M. E., Schultz, R. T., Miller, J., & Herrington, J. (2014). Traditional and atypical presentations of anxiety in youth with autism spectrum disorder. *Journal of Autism and Developmental Disorders, 44*(11), 2851–2861.

Kerns, C. M., Kendall, P. C., Zickgraf, H., Franklin, M. E., Miller, J., & Herrington, J. (2015). Not to be overshadowed or overlooked: Functional impairments associated with comorbid anxiety disorders in youth with ASD. *Behavior Therapy, 46*, 29–39.

Kranz, T. M., Kopp, M., Waltes, R., Sachse, M., Duketis, E., Jarczok, T. A., Degenhardt, F., Görgen, K., Meyer, J., Freitag, C. M., & Chiocchetti, A. G. (2016). Meta-analysis and association of two common polymorphisms of the human oxytocin receptor gene in autism spectrum disorder. *Autism Research, 9*(10), 1036–1045.

Lam, K. S., & Aman, M. G. (2007). The repetitive behavior scale-revised: Independent validation in individuals with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 37*(5), 855–866.

Langen, M., Bos, D., Noordermeer, S. D., Nederveen, H., van Engeland, H., & Durston, S. (2014). Changes in the development of striatum are involved in repetitive behavior in autism. *Biological Psychiatry, 76*(5), 405–411.

Lee, J. K., & Orsillo, S. M. (2014). Investigating cognitive flexibility as a potential mechanism of mindfulness in generalized anxiety disorder. *Journal of Behavior Therapy and Experimental Psychiatry, 45*(1), 208–216.

Leekam, S. R., Plissrior, M. R., & Uljarevic, M. (2011). Restricted and repetitive behaviors in autism spectrum disorders: A review of research in the last decade. *Psychological Bulletin, 137*(4), 562.

Lepicard, E. M., Venault, P., Negroni, J., Perez-Diaz, F., Joubert, C., Nosten-Bertrand, M., et al. (2003). Posture and balance responses to a sensory challenge are related to anxiety in mice. *Psychiatry Research, 118*, 273–284.

Leung, R. C., & Zakzanis, K. K. (2014). Brief report: Cognitive flexibility in autism spectrum disorders: A quantitative review. *Journal of Autism and Developmental Disorders, 44*(10), 2628–2645.

Levit-Binnun, N., Davidovitch, M., & Golland, Y. (2013). Sensory and motor secondary symptoms as indicators of brain vulnerability. *Journal of Neurodevelopmental Disorders, 5*(1), 26.

Lewin, A. B., Wu, M. S., Murphy, T. K., & Storch, E. A. (2015). Sensory over-responsivity in pediatric obsessive compulsive disorder. *Journal of Psychopathology and Behavioral Assessment, 37*(1), 134–143.

Leyfer, O. T., Folstein, S. E., Bacalman, S., Davis, N. O., Dinh, E., Morgan, J., et al. (2006). Comorbid psychiatric disorders in children with autism: Interview development and rates of disorders. *Journal of Autism and Developmental Disorders, 36*, 849–861.

Lidstone, J., Uljarević, M., Sullivan, J., Rodgers, J., McConachie, H., Freeston, M., Le Couteur, A., Prior, M., & Leekam, S. (2014). Relations among restricted and repetitive behaviors, anxiety and sensory features in children with autism spectrum disorders. *Research in Autism Spectrum Disorders, 8*(2), 82–92.

Liss, M., Saulnier, C., Fein, D., & Kinsbourne, M. (2006). Sensory and attention abnormalities in autistic spectrum disorders. *Autism, 10*(2), 155–172.

Ludlow, A., Skelly, C., & Rohleder, P. (2012). Challenges faced by parents of children diagnosed with autism spectrum disorder. *Journal of Health Psychology, 17*(5), 702–711.

MacNeil, B. M., Lopes, V. A., & Minnes, P. M. (2009). Anxiety in children and adolescents with autism spectrum disorders. *Research in Autism Spectrum Disorders, 3*(1), 1–21.

Maisel, M. E., Stephenson, K. G., South, M., Rodgers, J., Freeston, M. H., & Gaigg, S. B. (2016). Modeling the cognitive mechanisms linking autism symptoms and anxiety in adults. *Journal of Abnormal Psychology, 125*(5), 692.

Makovac, E., Meeten, F., Watson, D. R., Herman, A., Garfinkel, S. N., Critchley, H. D., & Ottaviani, C. (2016). Alterations in amygdala-prefrontal functional connectivity account for excessive worry and autonomic dysregulation in generalized anxiety disorder. *Biological Psychiatry, 80*(10), 786–795.

May, T., Cornish, K., & Rinehart, N. (2014). Does gender matter? A one-year follow-up of autistic, attention and anxiety symptoms in high-functioning children with autism spectrum disorder. *Journal of Autism and Developmental Disorders, 44*(5), 1077–1086.

McDougle, C. J., Kresch, L. E., Goodman, W. K., & Naylor, S. T. (1995). A case-controlled study of repetitive thoughts and behavior in adults with autistic disorder and obsessive-compulsive disorder. *The American Journal of Psychiatry, 152*(5), 772.

Miller, L., Anzalone, M., Lane, S., Cermak, S., & Osten, E. (2007). Concept evolution in sensory integration: A proposed nosology for diagnosis. *American Journal of Occupational Therapy, 61*, 135–140.

Mobini, S., & Grant, A. (2007). Clinical implications of attentional bias in anxiety disorders: An integrative literature review. *Psychotherapy Theory, Research, Practice, Training, 44*(4), 450–462.

Monk, C. S., Weng, S. J., Wiggins, J. L., Kurapati, N., Louro, H. M., Carrasco, M., Maslowsky, J., Risi, S., & Lord, C. (2010). Neural circuitry of emotional face processing in autism spectrum disorders. *Journal of Psychiatry and Neuroscience, 35*(2), 105.

Muris, P., Steerneman, P., Merckelbach, H., Holdrinet, I., & Meesters, C. (1998). Comorbid anxiety symptoms in children with pervasive developmental disorders. *Journal of Anxiety Disorders, 12*(4), 387–393.

Muskett, A., Capriola-Hall, N. N., Radtke, S. R., Factor, R., & Scarpa, A. (2019). Repetitive behaviors in autism spectrum disorder: Associations with depression and anxiety symptoms. *Research in Autism Spectrum Disorders, 68*, 101449.

Neil, L., Olsson, N. C., & Pellicano, E. (2016). The relationship between intolerance of uncertainty, sensory sensitivities, and anxiety in autistic and typically developing children. *Journal of Autism and Developmental Disorders, 46*(6), 1962–1973.

Plappert, C. F., & Pilz, P. K. D. (2002). Difference in anxiety and sensitization of the acoustic startle response between the two inbred mouse strains BALB/cAN and DBA/2N. *Genes, Brain and Behavior, 1*(3), 178–186.

Richler, J., Huerta, M., Bishop, S. L., & Lord, C. (2010). Developmental trajectories of restricted and repetitive behaviors and interests in children with autism spectrum disorders. *Development and Psychopathology, 22*(1), 55–69.

Rieke, E. F., & Anderson, D. (2009). Adolescent/adult sensory profile and obsessive-compulsive disorder. *The American Journal of Occupational Therapy, 63*(2), 138.

Rodgers, J., Glod, M., Connolly, B., & McConachie, H. (2012). The relationship between anxiety and repetitive behaviours in autism spectrum disorder. *Journal of Autism and Developmental Disorders, 42*(11), 2404–2409.

Russell, A. J., Mataix-Cols, D., Anson, M., & Murphy, D. G. (2005). Obsessions and compulsions in asperger syndrome and high-functioning autism. *The British Journal of Psychiatry, 186*(6), 525–528.

Sorg, S., Vögele, C., Furka, N., & Meyer, A. H. (2012). Perseverative thinking in depression and anxiety. *Frontiers in Psychology, 3*, 20.

South, M., Carr, A. W., Stephenson, K. G., Maisel, M. E., & Cox, J. C. (2017). Symptom overlap on the srs-2 adult self-report between adults with ASD and adults with high anxiety. *Autism Research, 10*(7), 1215–1220.

Spiker, M. A., Lin, C. E., Van Dyke, M., & Wood, J. J. (2012). Restricted interests and anxiety in children with autism. *Autism, 16*(3), 306–320.

Stratis, E. A., & Lecavalier, L. (2013). Restricted and repetitive behaviors and psychiatric symptoms in youth with autism spectrum disorders. *Research in Autism Spectrum Disorders, 7*(6), 757–766.

Swain, D., Scarpa, A., White, S., & Laugeson, E. (2015). Emotion dysregulation and anxiety in adults with ASD: Does social motivation play a role? *Journal of Autism and Developmental Disorders, 45*(12), 3971–3977.

Swartz, J. R., Wiggins, J. L., Carrasco, M., Lord, C., & Monk, C. S. (2013). Amygdala habituation and prefrontal functional connectivity in youth with autism spectrum disorders. *Journal of the American Academy of Child and Adolescent Psychiatry, 52*(1), 84–93.

Teh, E. J., Chan, D. M. E., Tan, G. K. J., & Magiati, I. (2017). Continuity and change in, and child predictors of, caregiver reported anxiety symptoms in young people with autism spectrum disorder: A follow-up study. *Journal of Autism and Developmental Disorders, 47*(12), 3857–3871.

Trepal, H., & Wester, K. (2007). Self-injurious behaviors, diagnoses, and treatment methods: What mental health professionals are reporting. *Journal of Mental Health Counseling, 29*(4), 363–375.

Uljarević, M., Carrington, S., & Leekam, S. (2016). Brief report: Effects of sensory sensitivity and intolerance of uncertainty on anxiety in mothers of children with autism spectrum disorder. *Journal of Autism and Developmental Disorders, 46*(1), 315–319.

van Steensel, F. J., Bögels, S. M., & Perrin, S. (2011). Anxiety disorders in children and adolescents with autistic spectrum disorders: A meta-analysis. *Clinical Child and Family Psychology Review, 14*(3), 302.

van Steensel, F. J., Bögels, S. M., & Wood, J. J. (2013). Autism spectrum traits in children with anxiety disorders. *Journal of Autism and Developmental Disorders*, *43*(2), 361–370.

Wager, T. D., Davidson, M. L., Hughes, B. L., Lindquist, M. A., & Ochsner, K. N. (2008). Prefrontal-subcortical pathways mediating successful emotion regulation. *Neuron, 59*(6), 1037–1050.

Williams, S., Leader, G., Mannion, A., & Chen, J. (2015). An investigation of anxiety in children and adolescents with autism spectrum disorder. *Research in Autism Spectrum Disorders, 10*, 30–40.

Wood, J. J., & Gadow, K. D. (2010). Exploring the nature and function of anxiety in youth with autism spectrum disorders. *Clinical Psychology: Science and Practice, 17*(4), 281–292.

Zandt, F., Prior, M., & Kyrios, M. (2007). Repetitive behaviour in children with high functioning autism and obsessive compulsive disorder. *Journal of Autism and Developmental Disorders, 37*(2), 251–259.

Zentall, S. S., & Zentall, T. R. (1983). Optimal stimulation: A model of disordered activity and performance in normal and deviant children. *Psychological Bulletin, 94*(3), 446.

Zike, I. D., Chohan, M. O., Kopelman, J. M., Krasnow, E. N., Flicker, D., Nautiyal, K. M., Bubser, M., Kellendonk, C., Jones, C. J., Stanwood, G., Tanaka, K. F., Moore, H., Ahmari, S. E., & Veenstra-VanderWeele, J. (2017). OCD candidate gene SLC1A1/EAAT3 impacts basal ganglia-mediated activity and stereotypic behavior. *Proceedings of the National Academy of Sciences, 114*(22), 5719–5724.

Chapter 12
The Relationship Between Eating Pathologies and Restricted, Repetitive Behaviours and Interests in Autism Spectrum Disorders

Roni Enten-Vissoker

Introduction

Individuals with autism spectrum disorder frequently suffer from some form of eating pathology. Disordered eating has been reported in the literature to be more prevalent in individuals with ASD than in those with typical development (Rastam & Wentz, 2014). While in the past, eating disorders included mainly anorexia nervosa (AN) and bulimia nervosa (BN), conditions prevalent mainly in women and typically driven by concerns about body shape or weight problems, the current DSM5 criteria for eating disorders is far more inclusive and includes avoidant/restrictive food intake disorder (ARFID), pica and rumination (APA, 2013). According to a recent study, eating disorders affect between 6% and 17% of adults with ASD, although their true prevalence is likely higher (Howlin & Magiati, 2017).

Although "eating pathology" typically refers to an eating disorder such as AN or BN, in the context of ASD, eating and feeding *problems* also fall under this umbrella term. The predominant eating pathology in ASD, eating problems, are the more common comorbidity in this population, and have been found to occur in up to 90% of children with autism spectrum disorders (Kodak & Piazza, 2008). Unlike eating disorders which have specific criteria for diagnosis (APA, 2013), eating problems are less clearly defined and vary in presentation; they include food selectivity, food refusal, chewing and swallowing problems, food rituals and more. Unlike eating disorders, many eating problems are motor and sensory-oriented by nature (Nadon, Feldman, Dunn, & Gisel, 2011).

Selective eating or food selectivity, the most prevalent eating problem among children with ASD, has been found to be strongly related to sensory processing disorders, which are a part of the ASD diagnostic criteria for restricted, and repetitive patterns of behavior, interests, or activities (RRBI) (APA, 2013). RRBI are manifested by hyper or hypo reactivity to sensory input or unusual interest in sensory aspects of

R. Enten-Vissoker (✉)
Individualized Bio-Nutrition Clinic, Israel

E. Gal, N. Yirmiya (eds.), *Repetitive and Restricted Behaviors and Interests in Autism Spectrum Disorders*, Autism and Child Psychopathology Series,
https://doi.org/10.1007/978-3-030-66445-9_12

environment, stereotyped or repetitive speech, motor movements or use of objects, excessive adherence to routines, and rituals, and restricted and fixated interests. Other manifestations of RRBI in the eating setting, include insistence on specific methods of food preparation, food types, and mealtime rules (Ahearn, Castine, Nault, & Green, 2001; Raiten & Massaro, 1986; Schreck, Williams, & Smith, 2004; Williams, Dalrymple, & Neal, 2000; Williams, Gibbons, & Schreck, 2005).

Children with ASD are frequently reported to display behaviors that are manifestations of RRBI in the eating setting, including insistence on specific methods of food preparation, food types, and mealtime rules. Although both RRBI and eating pathologies are highly researched areas with the potential to affect participation and function, to date, few researchers have directly assessed potential connections between the two. In order to shed light on their relation and explore possible shared underlying causes and/or mechanisms, in this chapter the literature and recent studies on the relation between eating pathologies, including eating disorders and eating problems, and RRBI, in individuals with ASD will be reviewed.

RRBI in ASD

The diagnostic criteria for ASD includes socio-communicative deficits and repetitive and restrictive behaviors and interests (RRBI) (APA, 2013). According to the DSM-5, RRBI include at least two of the following:

1. Stereotyped or repetitive speech, motor movements or use of objects
2. Excessive adherence to routines, ritualized patterns of verbal and nonverbal behavior
3. Highly restricted and fixated interests that are abnormal in intensity or focus
4. Hyper or hypo reactivity to sensory input or unusual interest in sensory aspects of environment (APA, 2013).

While RRBI are a heterogeneous group of behaviors, some research suggests they can be divided into two subcategories: repetitive sensory motor (RSM) behaviors, which include motor mannerisms, sensory seeking behaviors, and repetitive use of objects (aka lower order RRBI); and insistence on sameness (IS), behaviors which are characterized by compulsions and rituals and difficulties with changes in routine (aka higher order RRBI) (Cuccaro et al., 2003). Such behavioral subcategories may assist in organizing the larger ASD population into smaller groups of individuals with more similar behavioral profiles, facilitating efforts to uncover ASD etiologies (Bishop et al., 2013).

Eating and Feeding Disorders in ASD

Eating disorders (EDs) are considered to be major diseases of the modern world and are among the most prevalent public health problems in female adolescents and young adults, reaching epidemic proportions in many Western countries and

increasingly being identified among individuals with ASD (Hoek, 2006; Howlin & Magiati, 2017). In recent years, the age of onset of EDs has decreased, beginning in pre-pubescence. The main feeding/eating disorders are AN, BN, and Binge Eating Disorder (BED) and the revised DSM-5 added three additional conditions that fall under feeding disorders: pica, rumination and ARFID. These conditions comprise a new diagnostic category of feeding disorders of infancy and childhood, with the intention of specifically addressing eating/feeding problems in children (Keen, 2008). Pica, defined as the repetitive ingestion of items without nutritional value, such as paint, hair and dirt, is a behavior that can result in serious effects and a high mortality rate (Matson, Hattier, Belva, & Matson, 2013). Rumination is the non-purposeful regurgitation of recently ingested food from the stomach to the mouth, where it is either expelled or re-swallowed (Mousa, Montgomery, & Alioto, 2014).

Unlike AN and BN, which are emotionally and psychologically driven, pica and rumination, which span the eating disorder/problem categories, may originate from emotional, physiological/sensory or mental origins (McLoughlin & Hassanyeh, 1990; Nicholls & Bryant-Waugh, 2009). The ARFID diagnosis describes those whose symptoms do not match the criteria for traditional eating disorder diagnoses, but who experience clinically significant struggles with eating and feeding, failing to consume adequate amounts of food. Symptoms of ARFID typically appear in infancy or childhood, and may also persist into adolescence and adulthood (Fisher et al., 2014).

In contrast to EDs such as AN and BN, the term "feeding disorder" which is a formal diagnosis used in the ICD-10, relates to psychologically driven, emotional disturbances stemming from an organic condition (Bryant-Waugh, Markham, Kreipe, & Walsh, 2010). Different studies have suggested that while up to 80% of the childhood eating/feeding problems have a significant behavioral component, 16–30% of them are organic (Sanders, Patel, Le Grice, & Shepherd, 1993). While physicians and therapists can typically manage mild feeding problems, more severe problems among children can often be life-threatening without adequate intervention. Indeed, EDs remain highly misunderstood disorders and likely reflect complex interdependent multidimensional causalities, including genetic, biological, psychological, familial, and sociocultural factors (Treasure, Claudino, & Zucker, 2010).

Eating Disorders and RRBI in ASD

Though distinctly different in their etiology, studies have begun to highlight the genetic and symptomatic overlap between ASD and eating disorders, making individuals with ASD vulnerable to the aforementioned health risks resulting from eating and feeding disorders. In fact, some estimates hold that as much as 20 percent of people with enduring eating disorders have ASD (Wentz et al., 2005). This growing body of literature which highlights the prevalence of autistic symptoms in individuals with disordered eating, points to shared patterns of behavior and symptomology, including underlying difficulties in cognitive, social and emotional functioning, and specifically higher order RRBI (Westwood, Mandy, & Tchanturia, 2017).

Like ASD, EDs are complex disorders which result from interdependent multidimensional causes. In both conditions, individuals experience difficulties understanding and interpreting social cues, tend to fixate on tiny details that make it difficult to see the big picture, and thrive on rules, routines and rituals. Individuals with AN, defined by severely restricted eating, are known to engage in eating rituals similar to higher level RRBI seen in people with ASD in the eating setting, such as cutting food into very small pieces, separating food on the plate, chewing a certain number of times, excessively chewing before swallowing, eating by food group or meticulously measuring or arranging food (Calugi, Chignola, & Dalle Grave, 2019). It is thought that some individuals with AN may have a genetic tendency toward perfectionism, sensitivity and perseverance (Mayo Clinic, 2018).

Del'Osso et al. found that increased symptoms of ASD were highest among participants with eating disorders with restrictive behaviors. Individuals with anorexia scored significantly higher than participants with binge eating behaviors both on the Adult Autism Subthreshold Spectrum (AdAS) total score, as well as on the inflexibility and adherence to routine and restricted interest/rumination AdAS Spectrum domain scores. In addition, significant correlations were identified between the interpersonal distrust eating disorders inventory-2 sub-scale and the nonverbal communication and the restricted interest and rumination AdAS spectrum domains as well as between the social insecurity EDI-2 sub-scale and the inflexibility and adherence to routine and restricted interest and rumination domains (Dell'Osso et al., 2018).

Pooni, et al. also identified autistic symptoms among adolescents who had eating disorders in infancy and childhood (early onset ED) in their study, which compared them to a control group with typical development (Pooni, Ninteman, Bryant-Waugh, Nicholls, & Mandy, 2012). They reported that more than half the subjects (54.5%) showed repetitive, self-injurious, and compulsive behaviors and insistence on sameness. The group suggested that the similarities between the cognitive profiles of AN and ASD both appear to be associated with weak central coherence (Happe & Frith, 2006; Lopez, Tchanturia, Stahl, & Treasure, 2009) and difficulties with the executive function of set shifting (Roberts, Tchanturia, Stahl, Southgate, & Treasure, 2007). Similarities to ASD such as impairments in social cognition, in theory of mind (Oldershaw, Treasure, Hambrook, Tchanturia, & Schmidt, 2011), avoidance of eye gaze (Watson, Werling, Zucker, & Platt, 2010), abnormal response to social reward (Watson et al., 2010) and difficulties with emotion recognition have also been noted among adults with anorexia nervosa. Future research is warranted to further explore the relation between these two disorders and their relation with RRBI.

Eating Problems

While the role of eating and feeding disorders in ASD is gaining increasing attention by clinicians, eating problems are already recognized as a common co-morbidity in which the aforementioned dimensions of RRBI are evident. Though not clearly

defined in the literature, eating/feeding problems typically refer to a pattern of oral or enteral consumption of nutrients that deviates from the norm enough to lead to negative social and/or health consequences. There are various types of eating problems, and they vary in terms of etiology, behavior and severity (Gal, Hardal-Nasser, & Engel-Yeger, 2011; Laud, Girolami, Boscoe, & Gulotta, 2009; Matson, 2009). Eating problems are common in early childhood, occurring among 25–35% of typically developing children, and in up to 80–90% of children with developmental delays (Kodak & Piazza, 2008). Though these are often prominent problems of children with developmental delays, the lack of universally accepted definitions or current classification system for eating/feeding problems presents a challenge in the identification of such problems in infancy and early childhood (Arts-Rodas & Benoit, 1998).

The common eating/feeding problems include:

1. Food refusal – The inability or refusal to eat certain foods, defined as a child's refusal to eat all or most foods presented, resulting in the failure to meet caloric needs or on a supplemental formula (Williams, Field, & Seiverling, 2010). This eating/feeding problem is considered to be one of the more severe. It may also manifest as decreased appetite, turning of the head, mouth closure upon presentation of foods, spitting out, gagging, and vomiting of food (Williams et al., 2010) and is frequently associated with an underlying medical problem.
2. Food selectivity/restricted food intake – Defined as eating only a narrow variety of foods, and often used to refer to a range of different eating problems, such as selectivity by texture and type, eating a limited repertoire of accepted foods, and high-frequency single food intake and often has a sensory or motor-based origin (Marí-Bauset, Zazpe, Mari-Sanchis, Llopis-González, & Morales-Suárez-Varela, 2014; Matson, 2009).
3. Aggression and tantrums in the eating/feeding setting – Defined as exhibiting aggressive behavior towards self or others during meals such that the mealtime is disturbed, often stemming from sensory or physiological discomfort or behavioral origins (Provost, Crowe, Osbourn, McClain, & Skipper, 2010).
4. Rumination and pocketing – One of the official ICD-9 feeding disorders, defined as the non-purposeful regurgitation of recently ingested food from the stomach to the mouth following consumption, where it is either expelled or re-swallowed and the pocketing of food in the cheeks for extended periods (Nicholls & Bryant-Waugh, 2009; Seiverling, Williams, & Sturmey, 2010).
5. Chewing and swallowing problems – Often caused by impaired oral-motor health, may lead to food aspiration, choking, or life-threatening respiratory infections (Field, Garland, & Williams, 2003; Seiverling et al., 2010).
6. Poor appetite – Poor appetite or a lack of desire to take in adequate amounts of food can result from sensory or physiological origins, such as certain nutritional deficiencies (Beighley, Matson, Rieske, & Adams, 2013).
7. Vomiting/gastro-esophageal reflux (GER) –One of the most common forms of gastrointestinal dysfunction among children with ASD and has a significant influence on eating/feeding (Buie et al., 2010).

8. Pica – One of the ICD-9 feeding disorders, defined as the repetitive ingestion of items without nutritional value, such as paint, hair and dirt, a behavior that results in serious effects and high mortality rate (Matson, Belva, Hattier, & Matson, 2011).
9. Over-or under-eating – Eating too much or too little food as related to physiological needs/requirements, over eating can be a manifestation of RRBI (Broder-Fingert, Brazauskas, Lindgren, Iannuzzi, & Van Cleave, 2014; Williams et al., 2000).
10. Eating rituals – Ritualistic or repetitive patterns of behavior are commonly believed to contribute to food selectivity (Matson, 2009) Children with ASD often display insistence on specific methods of preparation, food types, and mealtime rules, a manifestation of RRBI (Zandt, Prior, & Kyrios, 2007).
11. Eating too quickly (Beighley et al., 2013)
12. Spitting/eating – Defined as chewing food and spitting it out without swallowing any of it

The aforementioned eating/feeding problems occur more frequently among children with ASD than children with other disabilities (Dominick, Davis, Lainhart, Tager-Flusberg, & Folstein, 2007) and typically developing children (Schreck et al., 2004), making eating a recurrent challenge and stressor for many parents. In one of the first literature reviews on the subject, out of seven studies performed between 1994 and 2004, it was estimated that 46% to 89% of children with ASD have atypical feeding habits (Ledford & Gast, 2006), which has since been confirmed by other research (Provost et al., 2010). Though few population studies have been conducted, well over half of children with ASD are reported to be selective eaters (Twachtman-Reilly, Amaral, & Zebrowski, 2008) and have been repeatedly found to display more of other eating/feeding problems, including food refusal, idiosyncratic mealtime behavior, and acceptance of a limited variety and texture of food items, than typically developing children (Schreck et al., 2004). Youth with ASD have specifically been found to be more selective regarding food groups, textures, tastes, and temperatures, and are more likely to refuse foods.

Though there are numerous types, food selectivity is known as the most common eating problem and occurs significantly more in those with ASD than typically developing children (Vissoker, Latzer, & Gal, 2015). Numerous studies have suggested that food selectivity in ASD is, at least in part, a manifestation of RRBI/restricted interests and activities (Ahearn et al., 2001). Ledford and Gast (2006) reported that 89% of parents of children with ASD stated their children followed repetitive patterns of food choice (Ledford & Gast, 2006) and in all studies reviewed, significant feeding difficulties were also reported, primarily in the form of selectivity by type and/or texture. In addition, Cornish et al. (Cornish, 1998) reported that 59% of children with ASD ate fewer than 20 different foods; indeed, selective children with ASD often exhibit a preference for starches, snack foods, and processed foods and display a lack of willingness to eat fruits, vegetables, and proteins.

Provost et al. (2010) also reported on the presence of eating routines or rituals in many young children with ASD. In their study, at least one-third of the children

were considered ritualistic eaters (8 children, 33%), and even more ate the same food in a repetitive manner (10 children, 42%), or had routines or rituals with food or eating (9 children, 37%). In addition, twelve (50%) of the children with ASD required food prepared in a special way, and six (25%) became upset if a mealtime routine was broken, and eight (33%) of the children with ASD were reported to stuff their mouths and cheeks. Though more young children with ASD ate in a repetitive manner and preferred certain food temperatures, food colors, and food packaging, these tendencies were also not significantly different than for the children with typical development. They purported that the reason for this finding may be related to the decreased prevalence of repetitive behaviors or need for sameness in many preschool children with ASD compared to when they are older, and that preschool-aged children with typical development may also exhibit some of these behaviors but they may become less prevalent with increasing age.

Nutritional Implications of Eating/Feeding Problems in ASD

While eating problems can present challenges in daily life and social settings, one of the greater concerns is that chronic eating/feeding problems can put children at risk for medical and developmental problems, including under-nutrition, suboptimal growth, social deficits and poor academic progress, as well as risk of nutrient deficiencies, such as vitamins, minerals and amino acids (Evans et al., 2008). A recent meta-analysis of 17 studies found that children with ASD are subject to lower intake of calcium and protein (Sharp et al., 2013). Other commonly reported nutrients consumed in insufficient amounts include calcium, iron, vitamins A, C, D, B6, B12, E, and K, as well as fiber, folic acid, and zinc (Hyman et al., 2012; Sharp et al., 2013).

Children with certain eating/feeding problems such as food selectivity, compulsive or binge eating may experience increased risk of overweight and obesity. According to the 2003–2004 National Survey of Children's Health, children with ASD are 40% more likely to be obese compared to typically developing children (Curtin, Anderson, Must, & Bandini, 2010); a strong preference for energy dense foods such as chips, cakes, hot dogs, and pizza seen among food selective adolescents may increase the likelihood of developing these conditions (Strahan, 2016). Despite the risks, feeding concerns in ASD are often overlooked in the clinical setting, possibly because such eating patterns have not been shown to be associated with compromised growth.

However, these preferences have been found to influence nutritional status in various ways. In their 2013 review, Sharp et al. identified three studies in which the relation between restricted patterns of intake and nutritional status was specifically investigated (Sharp et al., 2013). Herndon et al. (2009) reported intake of fewer servings of dairy and that this relation remained after excluding children following a gluten and casein free diet. They concluded that nutritional issues associated with ASD may be related to patterns of food selectivity beyond what could be attributed

to parent-mediated dietary manipulations. Zimmer et al. (2012) found, even after excluding children on elimination diets, that selective eaters with ASD had lower intake of calcium, vitamin B12, and vitamin D, compared to non-selective eaters with ASD, as well as lower intake of protein, calcium, vitamin A, and vitamin D, compared with typically developing peers. Thus, further investigating the relation between RRBI and eating problems is of importance.

Eating Problems and RRBI in ASD

There is evidence that ASD and eating disorders possess shared underlying difficulties in cognitive, social and emotional functioning, and higher order RRBI, such as routines and rituals. The lower order RRBI, characterized by sensory-motor oriented behaviors, can have a significant impact on eating problems in ASD. The presence of disordered sensory processing (refers to the ability to receive, organize and interpret stimuli, including oral, visual, tactile, vestibular, and auditory experiences) in some children with autism, and sensory over-responsivity specifically, is generally accepted. Difficulties with sensory processing have been repeatedly identified among children with ASD, with differences found between individuals with ASD and controls (Ben-Sasson et al., 2007; Germani et al., 2014; Kern et al., 2006). Ben-Sasson et al. (2007) found that toddlers with ASD are more likely to be under-responsive, display avoidance, and exhibit a low frequency of sensory-seeking behaviors (Ben-Sasson et al., 2007). In addition, Suarez, Nelson, and Curtis (2012) found that children with both severe food selectivity (i.e. those who accepted less than 10 foods) and moderate food selectivity (i.e. those who accepted 11–20 foods) had significantly higher scores on a measure of sensory over-responsivity than children who accepted 21 or more foods (Suarez et al., 2012).

Both eating problems, specifically food selectivity, and high level RRBI/stereotypical behaviors have been correlated specifically with hyper-sensory responsiveness (Boyd et al., 2010; Boyd, McBee, Holtzclaw, Baranek, & Bodfish, 2009; Johnson et al., 2014; Joosten & Bundy, 2010; Suarez et al., 2012), yet, there is also evidence that both hypo and hyper sensory input may manifest as food selectivity in children with ASD, making it even more challenging to separate the physiological aspects of feeding difficulty from behavioral aspects (Twachtman-Reilly et al., 2008). For example, it is possible that early tactile hyper- sensitivity may contribute to eating/feeding behaviors, such as avoiding certain foods, textures, tastes, smells and temperatures seen in children with ASD. In addition, tactile defensiveness and oral defensiveness may be part of a larger problem in modulating sensory input, which can take different forms, and affect various activities of daily living including eating/feeding (Cermak, Curtin, & Bandini, 2010). Suarez et al. theorized that the discomfort resulting from sensory overload during meals can lead to anxiety, prompting children to adopt repetitive and restricted behaviors and limit food intake to foods perceived as “safe,” with the goal of converting mealtime to a more

predictable experience and reducing levels of anxiety (Suarez et al., 2012). More research is needed to understand more about this relation.

It is important to note that while food selectivity often occurs in children with sensory integration dysfunction, food *refusal* is a more serious condition, often associated with the presence of a medical issue such as gastro-esophageal reflux or some other form of gastrointestinal dysfunction. According to a review by Williams et al. (2010), the most common medical diagnosis found among children with food refusal was gastro-esophageal reflux (69%), followed by other diagnoses including cardiopulmonary conditions (33%), neurological conditions (25%), food allergies (15%), anatomical anomalies (14%), and delayed gastric emptying (6%) (Williams et al., 2010).

Low level of cognitive function has also been found to be characteristic of both the differences between low and high levels of RRBI and a measure related to frequency and severity of eating problems in ASD (Gal et al., 2011; Turner, 1999). Both low order RRBI and eating problems have been found to be related to low cognitive function in individuals with ASD (Gal et al., 2011; Turner, 1999). Gal et al. (2011) found that among those with eating problems and intellectual disability, the frequency of eating problems increased with increased severity of intellectual disability. In addition, Green et al. (2009) reported that in ASD, lower cognition was also linked with greater motor impairment. In light of these findings, it is likely that cognitive level is a mediator which influences motor delays specific to low level RRBI as well as some eating problems, including problems with proper mouth closure, chewing, frequent choking, and problems swallowing.

As previously mentioned, although both RRBI and eating problems have been well studied as independent areas of research, very few have explored the connection between the full range of RRBI and eating problems, or the specific relation between individual eating problems and RRBI among children with ASD. The following section presents some of the key studies which have directly investigated the relationships between the two.

In 2014, Suarez, Nelson, and Curtis (2014) looked at the relation between repetitive behavior and food selectivity among children with ASD, based upon parental report questionnaires. Two different surveys were distributed to participants at two time points; the defining sensory over-responsivity [SOR] scale (included 19 items related to tactile, visual, auditory, and vestibular processing problems) was given to 141 participants and the second questionnaire, the repetitive behavior scale (RBS-R) was completed by 52 participants (Bodfish, Symons, & Lewis, 1999). Significant differences were found in repetitive behavior and impairments between children labeled as severely food selective versus those with moderate food selectivity; the more severely selective children were found to exhibit more repetitive behavior compared to those with moderate food selectivity. In addition, a stable, significant relation between food selectivity and sensory over-responsivity was identified, and restrictive and repetitive behavior (time 2) was found to significantly predict membership in the severe food selectivity group. However, when sensory over-responsivity and both restricted and repetitive behaviors were included in the regression model, only sensory over-responsivity significantly predicted severe

food selectivity. The group concluded that the variance in the RRBI score could be explained by the child's sensory (SOR) score, suggesting a close relation between these two factors, with prominence for the sensory variable. There was no significant unique variance found to be contributed by the RRB to food selectivity classification. A key limitation of this research was that it evaluated a single eating problem, food selectivity, and relied on parental report of child symptoms.

A second study by Johnson et al. (2014) explored the relation between mealtime behavior and other behavioral characteristics among 256 children with ASD, aged 2–11. It evaluated a range of eating symptoms, sensory symptoms and RRBI using the Brief Autism Mealtime Behavior Inventory (BAMBI) (Lukens & Linscheid, 2008), the Repetitive Behavior Checklist Revised (RBSR) (Bodfish & Lewis, 2002) and the Short Sensory Profile (SSP) (Dunn, 1999; McIntosh, Miller, Shyu, & Dunn, 1999). Significant correlations were identified between behavioral problems during mealtime, RRBI (general score only), sensory sensitivity and extroverted and introverted behaviors. The group noted the strong predictive relation of the RBS-R (which suggested an increased likelihood of parent-reported feeding behaviors when repetitive and ritualistic behaviors are rated high) and the support these findings provide for the anecdotally reported relations of repetitive behaviors interfering with feeding and mealtimes (Schreck et al., 2004). The results also indicated that higher BAMBI scores (more feeding/mealtime problems) could be predicted by lower SSP scores (greater sensory impairment), a relation suggested previously in other works, particularly sensory over-responsivity (Bennetto, Zampella, Kuschner, Bender, & Hyman, 2012; Cermak et al., 2010; Lane, Young, Baker, & Angley, 2010; Suarez et al., 2012). The authors suggested that in light of the high correlations between repetitive and ritualistic behaviors and sensory behaviors identified in this, as well as in previous work (Boyd et al., 2010; Chen, Rodgers, & McConachie, 2009), children with ASD who have significant repetitive and ritualistic behaviors as well as sensory sensitivities should be considered as at particular risk for problematic feeding behaviors. Again, this study is limited by the use of parental report which may not fully reflect the child's own sensory experience (Johnson et al., 2014).

Findings by Tanner et al. (2015) regarding repetitive behaviors, sensory reactivity, and challenging behaviors among 35 children with ASD, aged 4–10, differed from those of Johnson et al. (2014). They found no differences in measures of food refusal and acceptance and in challenging behaviors, anxiety, repetitive behaviors, and sensory reactivity between food selective and non-selective groups. However, scores on the RBS–R for repetitive behaviors during mealtime, were significantly different between selective and nonselective children, suggesting that the repetitive behaviors of children with selective eating may not be fully captured by the RBS–R. In addition, participants in the selective eating group had marginally lower scores in the SSP taste/smell sensitivity domain and no difference in the short sensory profile total score. This finding suggests that sensory reactivity for selective eaters may be limited to the gustatory and olfactory systems and less to social-emotional status, as no significant differences were observed between groups for anxiety/depression or somatic complaints (Tanner et al., 2015).

Finally, research from an unpublished Master's thesis (Mansur-Odeh, 2014) expanded upon the aforementioned research by examining a full range of both RRBI and eating problems among 66 children with ASD, compared to a control group (46 boys and 20 girls, aged 3–7 years old). The study utilized the RBS-R, and the eating problems and patterns questionnaire (EPQ) (recently renamed the AutEat) (Gal, Gal-Mishal, & Stolar, 2012) a tool developed for the assessment of eating problems and patterns of food intake in the ASD population. Mansur-Odeh identified the following associations:

- Strong correlations were found between the general scores of the RBS-R and the general score of the eating problems questionnaire (Gal et al., 2012), with greater RBS-R scores correlated with the presence of more eating problems in the ASD group.
- Medium to strong correlations between the general score of the RBS – revised, compulsive and ritualistic behaviors and sameness and difficulty with changes, and restricted interests and rituals and sameness in eating (Bodfish, Symons, Parker, & Lewis, 2000; Turner, 1999)
- A positive, significant correlation was found between stereotypical behavior (low level RRBI) on the RBS-R and the general score of the EPQ.
- A positive correlation was also found between aggression to oneself and others during meals on the RBS-R, and eating delay and avoidance on the EPQ.
- In addition, a weak to moderate, positive correlation was found between the excessive/overeating domain on the RBS-R and pica on the EPQ.

The results reveal that RRBI were positively correlated with eating problems in ASD. Specifically, stereotypical behaviors were correlated with eating problems in general, and aggression was correlated with eating avoidance. Indeed, children with ASD have been found to display extroverted and withdrawn behaviors in different situations as well as in daily activities, including eating (Lecavalier, 2006). Johnson et al. (2014) reported similar findings, and purported that extroverted and disruptive behaviors displayed by children with ASD in various situations may appear also at mealtime and may increase levels of anxiety, and result in repetitive and restricted behaviors in attempt to self-soothe (Ben-Sasson et al., 2007; Johnson et al., 2014; Joosten, Bundy, & Einfeld, 2009).

The differences identified between high and low order RRBI and their manifestations in the eating setting are of value; children with low order RRBI (stereotypical movements and self-injurious behavior) showed aggressive behavior at mealtime to themselves and others, eating avoidance and delay and some also displayed difficulty chewing and swallowing, while those with high order RRBI (aka compulsive and ritualistic behavior and insistence on sameness, difficulty with change and restricted interests) displayed the full range of eating problems assessed on the EPQ. The differences found between low and higher order RRBI and eating problems may be related to any number of mediating variables, including cognitive level and patterns and sensory and/or motor delays; more research is required to better understand them.

As previously mentioned, correlations between high level RRBI with rituals and sameness during eating may be indicative of weak central coherence. This leads to engagement in repetitive behaviors, restricted interests and resistance to change. Chen et al. (2009) purported that weak central coherence is the root cause of repetitive and restricted behaviors such as preference for sameness, performance of rituals and use of the same words. It therefore and may be a root cause of certain eating problems, and lead to strict use of certain dishes or utensils at meals, food rituals and sameness during mealtime.

It is important to mention that although the majority of the research focuses upon eating problems such as food selectivity and avoidance, RRBI can also affect and present as overeating, binge eating and compulsive eating, all of which have symptomatic overlap with RRBI. Individuals with ASD are more likely than neurotypical counterparts to have a body mass index (BMI) within the obesity or overweight range for their ages (Bennetto et al., 2012). In one recent study it was found that male children with ASD, who were overweight or obese, had more problematic mealtime and feeding behaviors than overweight or obese typically developing children, as indicated by the higher scores on a Behavior Pediatrics Feeding Assessment Scale (BPFA) in the ASD group (Castro et al., 2016). However, in another study of younger male and female children, a Nutritional Survey of Children with Autism Spectrum Disorder in Chongqing, China, no differences in feeding behaviors (assessed by questionnaire depicting oral function, eating problems, and others) across weight categories was yielded (Liu et al., 2016). In a study of one adolescent male with ASD, beneficial effects for severe obsessive food craving, binge eating, weight gain and behavioral problems were attained with liraglutide therapy, a glucagon-like peptide-1 (GLP-1) analog. The treatment had the effect of reduced weight and unwanted behavior by preventing food-related repetitive thoughts and compulsions, an area that warrants further exploration of this novel target for treating food-related behavioral problems and aggressive behavior in ASD (Jarvinen, Laine, Tikkanen, & Castren, 2019). Though yet not investigated, increased total energy intake and macronutrient distribution as a result of compulsive overeating or binge eating may also contribute to excess weight gain and its consequent health risks among children with ASD. Further studies are required to shed more light on the relation between RRBI and compulsive and overeating in individuals with ASD.

Summary and Conclusion

Eating problems and RRBI represent behaviors that can pose significant disruption and impair the quality of life of individuals on the spectrum and their families. A growing body of evidence supports the relation between the higher and lower order RRBI and a range of eating problems seen in ASD, with a clear role of sensory impairment on eating problems such as food selectivity as well as cognitive and motor impairment, all of which may represent underlying mechanisms. Since limited studies have explored the range of eating problems and RRBI, more expansive research is surely required to

understand the role of higher and lower order RRBI in eating problems and to gain further insight to understudied areas such as over and compulsive eating.

These findings highlight the importance of further study; future research should focus on exploring the different domains of RRBI and eating problem types and their relationships, among children with ASD with the goal of deepening knowledge and supporting clinicians in tailoring interventions for children and their families, to support quality of and participation in daily life. In addition, use of ASD specific tools such as the AutEat questionnaire (Gal et al., 2012) can be especially helpful in such research.

References

Ahearn, W. H., Castine, T., Nault, K., & Green, G. (2001). An assessment of food acceptance in children with autism or pervasive developmental disorder-not otherwise specified. *Journal of Autism and Developmental Disorders, 31*(5), 505–511.

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders: DSM-5*. Arlington, TX: American Psychiatric Association.

Arts-Rodas, D., & Benoit, D. (1998). Feeding problems in infancy and early childhood: Identification and management. *Paediatrics & Child Health, 3*(1), 21–27.

Beighley, J. S., Matson, J. L., Rieske, R. D., & Adams, H. L. (2013). Food selectivity in children with and without an autism spectrum disorder: Investigation of diagnosis and age. *Research in Developmental Disability, 34*(10), 3497–3503. https://doi.org/10.1016/j.ridd.2013.07.026.

Bennetto, L., Pennington, B. R., & Rogers, S. J. (1996). Intact and impaired memory functions in autism. *Child Development, 67*, 1816–1835.

Bennetto, L., Zampella, C., Kuschner, E., Bender, R., & Hyman, S. (2012). Food preferences in autism spectrum disorders and their relationship to sensory and behavioral symptoms.

Ben-Sasson, A., Cermak, S., Orsmond, G., Tager-Flusberg, H., Carter, A., Beth Kadlec, M., & Dunn, W. (2007). Extreme sensory modulation behaviors in toddlers with autism spectrum disorders. *The American Journal of Occupational Therapy, 61*, 584–592.

Bishop, S. L., Hus, V., Duncan, A., Huerta, M., Gotham, K., Pickles, A., et al. (2013). Subcategories of restricted and repetitive behaviors in children with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 43*(6), 1287–1297. https://doi.org/10.1007/s10803-012-1671-0.

Bodfish, J., Symons, F., Parker, D., & Lewis, M. (2000). Varieties of repetitive behavior in autism: Comparisons to mental retardation. *Journal of Autism and Developmental Disorders, 30*, 237–243. https://doi.org/10.1023/A:1005596502855.

Bodfish, J. W., Lewis, M. H., & Disorder, V. t. R. B. S.-R. i. Y. C. w. A. S. (2002). *Repetitive behavior in autism*. Paper presented at the International Meeting for Autism Research (IMFAR), Orlando, FL.

Bodfish, J. W., Symons, F. J., & Lewis, M. H. (1999). *The repetitive behavior scale*. Western Carolina Center Research Reports.

Boyd, B. A., Baranek, G. T., Sideris, J., Poe, M. D., Watson, L. R., Patten, E., & Miller, H. (2010). Sensory features and repetitive behaviors in children with autism and developmental delays. *Autism Research, 3*(2), 78–87. https://doi.org/10.1002/aur.124.

Boyd, B. A., McBee, M., Holtzclaw, T., Baranek, G. T., & Bodfish, J. W. (2009). Relationships among repetitive behaviors, sensory features, and executive functions in high functioning autism. *Research in Autism Spectrum Disorders, 3*(4), 959–966. https://doi.org/10.1016/j.rasd.2009.05.003.

Broder-Fingert, S., Brazauskas, K., Lindgren, K., Iannuzzi, D., & Van Cleave, J. (2014). Prevalence of overweight and obesity in a large clinical sample of children with autism. *Academic Pediatrics, 14*(4), 408–414. https://doi.org/10.1016/j.acap.2014.04.004.

Bryant-Waugh, R., Markham, L., Kreipe, R. E., & Walsh, B. T. (2010). Feeding and eating disorders in childhood. *International Journal of Eating Disorders, 43*(2), 98–111. https://doi.org/10.1002/eat.20795.

Buie, T., Campbell, D. B., Fuchs, G. J., 3rd, Furuta, G. T., Levy, J., Vandewater, J., et al. (2010). Evaluation, diagnosis, and treatment of gastrointestinal disorders in individuals with ASDs: A consensus report. *Pediatrics, 125*(Suppl 1), S1–S18. https://doi.org/10.1542/peds.2009-1878C.

Calugi, S., Chignola, E., & Dalle Grave, R. (2019). A longitudinal study of eating rituals in patients with anorexia nervosa. *Frontiers in Psychology, 10*, 15. https://doi.org/10.3389/fpsyg.2019.00015.

Castro, K., Faccioli, L. S., Baronio, D., Gottfried, C., Perry, I. S., & Riesgo, R. (2016). Feeding behavior and dietary intake of male children and adolescents with autism spectrum disorder: A case-control study. *International Journal of Developmental Neuroscience, 53*, 68–74. https://doi.org/10.1016/j.ijdevneu.2016.07.003.

Cermak, S. A., Curtin, C., & Bandini, L. G. (2010). Food selectivity and sensory sensitivity in children with autism spectrum disorders. *Journal of the American Dietetic Association, 110*(2), 238–246. https://doi.org/10.1016/j.jada.2009.10.032.

Chen, Y. H., Rodgers, J., & McConachie, H. (2009). Restricted and repetitive behaviours, sensory processing and cognitive style in children with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 39*(4), 635–642. https://doi.org/10.1007/s10803-008-0663-6.

Cornish, E. (1998). A balanced approach towards healthy eating in autism. *Journal of Human Nutrition and Dietetics, 11*(6), 501–509. https://doi.org/10.1046/j.1365-277X.1998.00132.x.

Cuccaro, M. L., Shao, Y., Grubber, J., Slifer, M., Wolpert, C. M., Donnelly, S. L., et al. (2003). Factor analysis of restricted and repetitive behaviors in autism using the Autism Diagnostic Interview-R. *Child Psychiatry and Human Development, 34*(1), 3–17.

Curtin, C., Anderson, S. E., Must, A., & Bandini, L. (2010). The prevalence of obesity in children with autism: A secondary data analysis using nationally representative data from the National Survey of Children's Health. *BMC Pediatrics, 10*, 11. https://doi.org/10.1186/1471-2431-10-11.

Dell'Osso, L., Carpita, B., Gesi, C., Cremone, I. M., Corsi, M., Massimetti, E., et al. (2018). Subthreshold autism spectrum disorder in patients with eating disorders. *Comprehensive Psychiatry, 81*, 66–72. https://doi.org/10.1016/j.comppsych.2017.11.007.

Dominick, K. C., Davis, N. O., Lainhart, J., Tager-Flusberg, H., & Folstein, S. (2007). Atypical behaviors in children with autism and children with a history of language impairment. *Research in Developmental Disability, 28*(2), 145–162. https://doi.org/10.1016/j.ridd.2006.02.003.

Dunn, W. (1999). *The sensory profile: User's manual*. San Antonio, TX: Psychological Corporation.

Evans, C., Dunstan, R. H., Rothkirch, T., Roberts, T. K., Reichelt, K. L., Cosford, R., et al. (2008). Altered amino acid excretion in children with autism. *Nutritional Neuroscience, 11*(1), 9–17. https://doi.org/10.1179/147683008x301360.

Field, D., Garland, M., & Williams, K. (2003). Correlates of specific childhood feeding problems. *Journal of Paediatric and Child Health, 39*(4), 299–304.

Fisher, M. M., Rosen, D. S., Ornstein, R. M., Mammel, K. A., Katzman, D. K., Rome, E. S., et al. (2014). Characteristics of avoidant/restrictive food intake disorder in children and adolescents: A "new disorder" in DSM-5. *Journal of Adolescent Health, 55*(1), 49–52. https://doi.org/10.1016/j.jadohealth.2013.11.013.

Gal, E., Gal-Mishal, R., & Stolar, O. (2012). *AutEat questionnaire*. Haifa, Israel: University of Haifa.

Gal, E., Hardal-Nasser, R., & Engel-Yeger, B. (2011). The relationship between the severity of eating problems and intellectual developmental deficit level. *Research in Developmental Disabilities, 32*(5), 1464–1469. https://doi.org/10.1016/j.ridd.2010.12.003.

Germani, T., Zwaigenbaum, L., Bryson, S., Brian, J., Smith, I., Roberts, W., et al. (2014). Brief report: Assessment of early sensory processing in infants at high-risk of autism spectrum disorder. *Journal of Autism and Developmental Disorders, 44*(12), 3264–3270. https://doi.org/10.1007/s10803-014-2175-x.

Green et al. (2009). https://pubmed.ncbi.nlm.nih.gov/19207298/.

Happe, F., & Frith, U. (2006). The weak coherence account: Detail-focused cognitive style in autism spectrum disorders. *Journal of Autism and Developmental Disorders, 36*, 5–25. https://doi.org/10.1007/s10803-005-0039-0.

Herndon et al. (2009). https://pubmed.ncbi.nlm.nih.gov/18600441/.

Hoek, H. (2006). Incidence, prevalence and mortality of anorexia nervosa and other eating disorders. *Current Opinion in Psychiatry, 19*, 389–394. https://doi.org/10.1097/01.yco.0000228759.95237.78.

Howlin, P., & Magiati, I. (2017). Autism spectrum disorder: Outcomes in adulthood. *Current Opinion in Psychiatry, 30*(2), 69–76. https://doi.org/10.1097/yco.0000000000000308.

Hyman, S. L., Stewart, P. A., Schmidt, B., Cain, U., Lemcke, N., Foley, J. T., et al. (2012). Nutrient intake from food in children with autism. *Pediatrics, 130*(Suppl 2), S145–S153. https://doi.org/10.1542/peds.2012-0900L.

Jarvinen, A., Laine, M. K., Tikkanen, R., & Castren, M. L. (2019). Beneficial effects of GLP-1 agonist in a male with compulsive food-related behavior associated with autism. *Frontiers in Psychiatry, 10*, 97. https://doi.org/10.3389/fpsyt.2019.00097.

Johnson, C. R., Turner, K., Stewart, P. A., Schmidt, B., Shui, A., Macklin, E., et al. (2014). Relationships between feeding problems, behavioral characteristics and nutritional quality in children with ASD. *Journal of Autism and Developmental Disorders, 44*(9), 2175–2184. https://doi.org/10.1007/s10803-014-2095-9.

Joosten, A. V., & Bundy, A. C. (2010). Sensory processing and stereotypical and repetitive behaviour in children with autism and intellectual disability. *Australian Occupational Therapy Journal, 57*(6), 366–372. https://doi.org/10.1111/j.1440-1630.2009.00835.x.

Joosten, A. V., Bundy, A. C., & Einfeld, S. L. (2009). Intrinsic and extrinsic motivation for stereotypic and repetitive behavior. *Journal of Autism and Developmental Disorders, 39*(3), 521–531. https://doi.org/10.1007/s10803-008-0654-7.

Keen, D. V. (2008). Childhood autism, feeding problems and failure to thrive in early infancy. Seven case studies. *European Child and Adolescent Psychiatry, 17*(4), 209–216. https://doi.org/10.1007/s00787-007-0655-7.

Kern, J. K., Trivedi, M. H., Garver, C. R., Grannemann, B. D., Andrews, A. A., Savla, J. S., et al. (2006). The pattern of sensory processing abnormalities in autism. *Autism, 10*(5), 480–494. https://doi.org/10.1177/1362361306066564.

Kodak, T., & Piazza, C. C. (2008). Assessment and behavioral treatment of feeding and sleeping disorders in children with autism spectrum disorders. *Child and Adolescent Psychiatric Clinics of North America, 17*(4), 887–905, x–xi. https://doi.org/10.1016/j.chc.2008.06.005.

Lane, A. E., Young, R. L., Baker, A. E., & Angley, M. T. (2010). Sensory processing subtypes in autism: Association with adaptive behavior. *Journal of Autism and Developmental Disorders, 40*(1), 112–122. https://doi.org/10.1007/s10803-009-0840-2.

Laud, R. B., Girolami, P. A., Boscoe, J. H., & Gulotta, C. S. (2009). Treatment outcomes for severe feeding problems in children with autism spectrum disorder. *Behavior Modification, 33*(5), 520–536. https://doi.org/10.1177/0145445509346729.

Lecavalier, L. (2006). Behavioral and emotional problems in young people with pervasive developmental disorders: Relative prevalence, effects of subject characteristics, and empirical classification. *Journal of Autism and Developmental Disorders, 36*(8), 1101–1114. https://doi.org/10.1007/s10803-006-0147-5.

Ledford, J., & Gast, D. L. (2006). Feeding problems in children with autism spectrum disorders: A review. *Focus on Autism and Other Developmental Disorders, 21*(3), 153–166.

Liu, X., Liu, J., Xiong, X., Yang, T., Hou, N., Liang, X., et al. (2016). Correlation between nutrition and symptoms: Nutritional survey of children with autism spectrum disorder in Chongqing, China. *Nutrients, 8*(5). https://doi.org/10.3390/nu8050294.

Lopez, C., Tchanturia, K., Stahl, D., & Treasure, J. (2009). Weak central coherence in eating disorders: A step towards looking for an endophenotype of eating disorders. *Journal of Clinical and Experimental Neuropsychology, 31*(1), 117–125. https://doi.org/10.1080/13803390802036092.

Lukens, C. T., & Linscheid, T. R. (2008). Development and validation of an inventory to assess mealtime behavior problems in children with autism. *Journal of Autism and Developmental Disorders, 38*(2), 342–352. https://doi.org/10.1007/s10803-007-0401-5.

Mansur-Odeh, R. (2014). *The correlation between repetitive and restricted behaviors and interests and eating problems among children with ASD* (Master's). University of Haifa.

Marí-Bauset, S., Zazpe, I., Mari-Sanchis, A., Llopis-González, A., & Morales-Suárez-Varela, M. (2014). Food selectivity in autism spectrum disorders: A systematic review. *Journal of Child Neurology, 29*(11), 1554–1561. https://doi.org/10.1177/0883073813498821.

Matson, F. (2009). The treatment of food selectivity and other feeding problems in children with autism spectrum disorders. *Research in Autism Spectrum Disorders, 3*(2), 455–461. https://doi.org/10.1016/j.rasd.2008.09.005.

Matson, J., Belva, B., Hattier, M. A., & Matson, M. L. (2011). Pica in persons with developmental disabilities: Characteristics, diagnosis, and assessment. *Research in Autism Spectrum Disorders, 5*(4), 1459–1464. https://doi.org/10.1016/j.rasd.2011.02.006.

Matson, J., Hattier, M. A., Belva, B., & Matson, M. L. (2013). Pica in persons with developmental disabilities: Approaches to treatment. *Research in Developmental Disability, 34*(9), 2564–2571. https://doi.org/10.1016/j.ridd.2013.05.018.

Mayo Clinic. (2018). *Anorexia nervosa*. Retrieved from https://www.mayoclinic.org/diseases-conditions/anorexia-nervosa/symptoms-causes/syc-20353591

McIntosh, D., Miller, L. J., Shyu, V., & Dunn, W. (1999). Overview of the short sensory profile. In W. Dunn (Ed.), *Sensory profile user's manual* (pp. 59–73). San Antonio, TX: Pearsons.

McLoughlin, I. J., & Hassanyeh, F. (1990). Pica in a patient with anorexia nervosa. *The British Journal of Psychiatry, 156*, 568–570. https://doi.org/10.1192/bjp.156.4.568.

Mousa, H. M., Montgomery, M., & Alioto, A. (2014). Adolescent rumination syndrome. *Current Gastroenterology Reports, 16*(8), 398. https://doi.org/10.1007/s11894-014-0398-9.

Nadon, G., Feldman, D. E., Dunn, W., & Gisel, E. (2011). Association of sensory processing and eating problems in children with autism spectrum disorders. *Autism Research and Treatment, 2011*, 541926. https://doi.org/10.1155/2011/541926.

Nicholls, D., & Bryant-Waugh, R. (2009). Eating disorders of infancy and childhood: Definition, symptomatology, epidemiology, and comorbidity. *Child and Adolescent Psychiatric Clinics of North America, 18*(1), 17–30. https://doi.org/10.1016/j.chc.2008.07.008.

Oldershaw, A., Treasure, J., Hambrook, D., Tchanturia, K., & Schmidt, U. (2011). Is anorexia nervosa a version of autism spectrum disorders? *European Eating Disorder Review, 19*(6), 462–474. https://doi.org/10.1002/erv.1069.

Pooni, J., Ninteman, A., Bryant-Waugh, R., Nicholls, D., & Mandy, W. (2012). Investigating autism spectrum disorder and autistic traits in early onset eating disorder. *International Journal of Eating Disorders, 45*(4), 583–591. https://doi.org/10.1002/eat.20980.

Provost, B., Crowe, T. K., Osbourn, P. L., McClain, C., & Skipper, B. J. (2010). Mealtime behaviors of preschool children: Comparison of children with autism spectrum disorder and children with typical development. *Physical and Occupational Therapy in Pediatrics, 30*(3), 220–233. https://doi.org/10.3109/01942631003757669.

Raiten, D. J., & Massaro, T. (1986). Perspectives on the nutritional ecology of autistic children. *Journal of Autism and Developmental Disorders, 16*(2), 133–143.

Rastam, M., & Wentz, E. (2014). ASD, eating problems, and overlap with anorexia and bulimia nervosa. In V. B. Patel & C. R. Martin (Eds.), *Comprehensive guide to autism* (pp. 2015–2034). New York, NY: Springer.

Roberts, M. E., Tchanturia, K., Stahl, D., Southgate, L., & Treasure, J. (2007). A systematic review and meta-analysis of set-shifting ability in eating disorders. *Psychological Medicine, 37*(8), 1075–1084. https://doi.org/10.1017/s0033291707009877.

Sanders, M. R., Patel, R. K., Le Grice, B., & Shepherd, R. W. (1993). Children with persistent feeding difficulties: An observational analysis of the feeding interactions of problem and non-problem eaters. *Health Psychology, 12*(1), 64–73. https://doi.org/10.1037//0278-6133.12.1.64.

Schreck, K., Williams, K., & Smith, A. (2004). A comparison of eating behaviors between children with and without autism. *Journal of Autism and Developmental Disorders, 34*(4), 433–438. https://doi.org/10.1023/B:JADD.0000037419.78531.86.

Seiverling, L., Williams, K., & Sturmey. (2010). Assessment of feeding problems in children with autism spectrum disorders. *Journal of Developmental and Physical Disabilities, 22*, 401–413.

Sharp, W. G., Berry, R. C., McCracken, C., Nuhu, N. N., Marvel, E., Saulnier, C. A., Klin, A., Jones, W., & Jaquess, D. L. (2013). Feeding problems and nutrient intake in children with autism spectrum disorders: A meta-analysis and comprehensive review of the literature. *Journal of Autism and Developmental Disorders, 43*(9), 2159–2173. https://doi.org/10.1007/s10803-013-1771-5.

Strahan, B. E. (2016). Food selectivity in obese adolescents with autism spectrum disorder. *Journal of Child and Adolescent Behavior, 4*, 268.

Suarez, M. A., Nelson, N. W., & Curtis, A. B. (2012). Associations of physiological factors, age, and sensory over-responsivity with food selectivity in children with autism spectrum disorders. *The Open Journal of Occupational Therapy, 1*(1), 2.

Suarez, M. A., Nelson, N. W., & Curtis, A. B. (2014). Longitudinal follow-up of factors associated with food selectivity in children with autism spectrum disorders. *Autism, 18*(8), 924–932. https://doi.org/10.1177/1362361313499457.

Tanner, K., Case-Smith, J., Nahikian-Nelms, M., Ratliff-Schaub, K., Spees, C., & Darragh, A. R. (2015). Behavioral and physiological factors associated with selective eating in children with autism spectrum disorder. *American Journal of Occupational Therapy, 69*(6), 6906180030p1-6906180030p8. https://doi.org/10.5014/ajot.2015.019273.

Treasure, J., Claudino, A. M., & Zucker, N. (2010). Eating disorders. *Lancet, 375*(9714), 583–593. https://doi.org/10.1016/s0140-6736(09)61748-7.

Turner, M. (1999). Annotation: Repetitive behaviour in autism: A review of psychological research. *Journal of Child Psychology and Psychiatry, 40*(6), 839–849.

Twachtman-Reilly, J., Amaral, S. C., & Zebrowski, P. P. (2008). Addressing feeding disorders in children on the autism spectrum in school-based settings: Physiological and behavioral issues. *Language Speech and Hearing Services in Schools, 39*(2), 261–272. https://doi.org/10.1044/0161-1461(2008/025).

Vissoker, R. E., Latzer, Y., & Gal, E. (2015). Eating and feeding problems and gastrointestinal dysfunction in autism spectrum disorders. *Research in Autism Spectrum Disorders, 12*, 10–21. https://doi.org/10.1016/j.rasd.2014.12.010.

Watson, K. K., Werling, D. M., Zucker, N. L., & Platt, M. L. (2010). Altered social reward and attention in anorexia nervosa. *Frontiers in Psychology, 1*, 36. https://doi.org/10.3389/fpsyg.2010.00036.

Wentz, E., Lacey, J. H., Waller, G., Rastam, M., Turk, J., & Gillberg, C. (2005). Childhood onset neuropsychiatric disorders in adult eating disorder patients. A pilot study. *European Child and Adolescent Psychiatry, 14*(8), 431–437. https://doi.org/10.1007/s00787-005-0494-3.

Westwood, H., Mandy, W., & Tchanturia, K. (2017). Clinical evaluation of autistic symptoms in women with anorexia nervosa. *Molecular Autism, 8*. https://doi.org/10.1186/s13229-017-0128-x.

Williams, K. E., Field, D. G., & Seiverling, L. (2010). Food refusal in children: A review of the literature. *Research in Developmental Disability, 31*(3), 625–633. https://doi.org/10.1016/j.ridd.2010.01.001.

Williams, K. E., Gibbons, B. G., & Schreck, K. A. (2005). Comparing selective eaters with and without developmental disabilities. *Journal of Developmental and Physical Disabilities, 17*(3), 299–309. https://doi.org/10.1007/s10882-005-4387-7.

Williams, P. G., Dalrymple, N., & Neal, J. (2000). Eating habits of children with autism. *Pediatric Nursing, 26*(3), 259–264.

Zandt, F., Prior, M., & Kyrios, M. (2007). Repetitive behaviour in children with high functioning autism and obsessive compulsive disorder. *Journal of Autism and Developmental Disorders, 37*(2), 251–259. https://doi.org/10.1007/s10803-006-0158-2.

Zimmer et al. (2012). https://pubmed.ncbi.nlm.nih.gov/21556968/.

Chapter 13
Early Intervention and Restricted, Repetitive Behaviours and Interests

Maya Yaari and Cheryl Dissanayake

Background

Restricted and repetitive behaviours and interests (RRBI) are a core feature in Autism Spectrum Disorders (ASD), and their presence is required for a diagnosis, according to the Diagnostic and Statistical Manual for Mental Disorders (DSM-5: American Psychiatric Association, 2013). This category of behaviours is very broad, defining a wide range of idiosyncratic actions, including:

1. Stereotyped, repetitive movements (e.g., hand-flapping), repetitive use of objects (e.g., spinning wheels, lining up toys), or repetitive language (e.g. echolalia, idiosyncratic language).
2. Insistence on sameness (e.g. inflexible adherence to routines such that the child may become distressed in response to changes in routine or environment), and ritualized patterns of behaviour (e.g., greeting rituals).
3. Highly restricted, fixated interests that are unusual in their intensity or content (e.g., strong attachment to or preoccupation with unusual objects, excessively interest in bus schedules).
4. Hyper- or hypo-reactivity to sensory input (e.g., apparent indifference to pain/temperature, adverse response to specific sounds or textures) or unusual interests in sensory aspects of the environment (e.g. excessive smelling or touching of objects, visual fascination with lights or movement).

In this chapter we discuss RRBI in the context of ASD early intervention (EI) research. RRBI may impede children's learning, decrease social interaction and cause substantial parental distress. However, while EI research in ASD has substantially progressed over the last years, its main focus remains on social-communication

M. Yaari (✉) · C. Dissanayake
Olga Tennison Autism Research Centre, School of Psychology and Public Health, La Trobe University, Melbourne, VIC, Australia

E. Gal, N. Yirmiya (eds.), *Repetitive and Restricted Behaviors and Interests in Autism Spectrum Disorders*, Autism and Child Psychopathology Series,
https://doi.org/10.1007/978-3-030-66445-9_13

difficulties and outcomes, with much less known about intervention effects on RRBI, and how these behaviours are addressed in the context of EI. The neurodiversity movement has spurred an increase in the number of first-person accounts of autism than previously available (Pellicano & Stears, 2011). These testimonies regarding personal experiences, including the use of RRBI have increased our understanding of these behaviours and the functions they may serve for children with ASD (Baron-Cohen, 2017; Kapp, Gillespie-Lynch, Sherman, & Hutman, 2013). Subscribing to a partnership-based approach to intervention in which therapy goals are driven by the family and individuals in collaboration with the therapist informs and questions the approaches to reduce RRBI in ASD.

RRBI in Childhood: Functions in Typical and Atypical Development

RRBI are observed in typically developing infants and toddlers, at an age in which they are considered common and developmentally appropriate (Barber, Wetherby, & Chambers, 2012; Harrop et al., 2014; Leekam et al., 2007). Repetition of movements and actions and ritualistic behaviours are considered part of the process of skill acquisition, usually reducing over time and development, as the child attains mastery of the skill (Leekam et al., 2007; MacDonald et al., 2007; Wolff et al., 2014). These behaviours may also serve other functions such as self-soothing, reducing of anxiety by increasing predictability of routines, regulation of arousal and energy levels, or self-stimulation (Larkin, Meins, Centifanti, Fernyhough, & Leekam, 2017).

The RRBI associated with ASD differ from those observed among typically developing children (and those with other developmental difficulties) in the increased frequency, intensity, variety, and persistence over time of these behaviours, such that they can interfere with learning and daily functioning (Bodfish, Symons, Parker, & Lewis, 2000; Harrop et al., 2014; Matson, Dempsey, & Fodstad, 2009). RRBI in ASD have been documented as a cause of parental concern and distress and perceived as more challenging for parents to manage than social-communication difficulties (Boyd, McDonough, & Bodfish, 2012; Harrop, McBee, & Boyd, 2016).

The ASD literature categorizes RRBI as lower- and higher-order (Leekam, Prior, & Uljarevic, 2011; Turner, 1999). Lower-order RRBI involve motor stereotypy such as hand flapping or rocking, and object stereotypy such repetitively opening and closing a door and spinning or lining objects, with these behaviours considered as more common among younger children and those with developmental delay. Higher-order RRBI, considered more common in older aged children and among those without cognitive impairments, include obsessive engagement in odd interests and hobbies, insistence on sameness and the repetitive use of language (Leekam et al., 2011). Assessment of RRBI is conducted via caregiver questionnaires and

interviews, teacher reports, structured and unstructured observations of children and coding of videotaped material (Leekam et al., 2011; McConachie et al., 2015). RRBI are commonly assessed as part of the diagnostic process, to determine if they are present and meet diagnostic criteria. This is usually conducted via the well-established ASD diagnostic measures, the Autism Diagnostic Interview (ADI) in which caregivers are questioned about their child's current and past behaviours, and the Autism Diagnostic Observation Schedule (ADOS), which is an observational measure. RRBI are also assessed in the context of early intervention, to monitor the progress of children over time, and examine outcomes and efficacy of intervention programs (Leekam et al., 2011).

RRBI and Social Learning

Prospective high-risk infant sibling studies have been useful in charting the presence and early emergence of RRBI, even prior to social-communication impairments (Baranek, 1999; Ozonoff et al., 2008; Rogers, 2009). RRBI in very young children with ASD may impact learning and hence the acquisition of social, cognitive and adaptive skills. Learning in young children occurs within social contexts, via social attention, imitation, and joint engagement with other people (Dawson, 2008). When children preferentially engage with objects, often in repetitive ways, as in the case of ASD, their opportunities to learn are limited. Take, for example, a child with ASD repeatedly flicking a doll's eyes or lining blocks in a particular way instead of engaging in social play by showing the toy to his/her parents, or engaging in turn-taking with them, which can limit his/her ability to learn from others and the relational activity itself. On the other hand, the social-communication difficulties and delayed functional play skills evident in ASD may further contribute to limiting the range of behaviours the child engages in, leading to RRBI becoming increasingly prominent in the child's repertoire. Findings regarding the association between RRBI in early years and poorer later social and cognitive outcomes (Ausderau et al., 2016; Larkin et al., 2017; Ozonoff et al., 2008; Ray-Subramanian & Ellis Weismer, 2012; Troyb et al., 2016), indicate their possible impact on learning. These findings coupled with a lack of understanding of the function of these behaviours for children with ASD has led to approaches aimed at reducing them.

Early Intervention and RRBI

There is a distinction in the intervention literature between comprehensive treatment models (CTM) and focused intervention practices (FIP). CTMs are designed to achieve broad developmental gains across multiple domains, and are usually intensive, and delivered over an extended period of time. The efficacy of these interventions is usually assessed via standardised measures of ASD symptoms and

cognitive and adaptive functioning (Odom, Boyd, Hall, & Hume, 2010). In FIPs, on the other hand, strategies are employed for a limited period of time in order to target specific behavioural symptom/s or to attain a particular skill. Examining the efficacy of such targeted intervention is done usually via case series/studies, with the outcome being child specific - attaining a particular target. These FIPS can be integrated within CTMs or employed individually as targeted interventions. Focusing on young children in the pre-school years, we will first examine the different theoretical and clinical approaches to RRBI within CTMs, followed by evidence on the effects of these interventions on RRBI and how RRBI features may predict the treatment outcome. We will then describe the specific FIPs targeting RRBI, their evidence-base and potential "spill-over" effects on other behavioural domains.

Early Intervention Frameworks

Early interventions (EI) for children with ASD, in general, vary with regards to the theoretical approach, which generally informs the service delivery model and strategies employed. The main EI frameworks are behavioural, developmental, relationship-based, and sensory-motor (Raulston & Machalicek, 2017), with many incorporating more than one framework. Within the behavioural framework and the Applied Behaviour Analysis (ABA) approach, behaviours – including RRBI – are maintained because they serve a function or, in other words, the behaviour is maintained by the consequences that follow it. Reinforcers can be social or non-social, positive or negative, or a combination of different types. Social positive reinforcers can be attention or access to an attractive object or activity; social negative reinforcers can be avoiding a task or activity; non-social (often called automatic) positive reinforcers can be a sensory stimulation which is independent from social mediation; and non-social negative reinforcers can be removal of a distressing sensory stimulus (Rapp & Vollmer, 2005). Using a Functional Behavioural Assessment, the practitioner explores antecedents and reinforcers of the unwanted behaviour, and employs appropriate Positive Behavioural Support strategies, Discrete Trail Training (DTT) or Pivotal Response Treatment (PRT) techniques to reduce behaviours that are considered to interfere with learning and adaptive functioning, and shape and reinforce more adaptive behaviours (Harrop, 2015; Odom, Collet-Klingenberg, Rogers, & Hatton, 2010).

Interventions based on developmental or relationship-based approaches (e.g. the Developmental, Individual-differences, & Relationship-based model, DIR) usually emphasize the child's social-emotional development and skills, and the caregiver's responsivity to the child's cues. Subscribing to "follow the child's lead", the practitioner does not attempt to directly change or shape the child's behaviours but rather joins the child in his/her activity to enhance motivation and facilitate the development of his/her communication skills to express and articulate his/her needs (Harrop, 2015). The approach to RRBI within integrative, Naturalistic Developmental Behavioural Interventions such as the Early Start Denver Model (ESDM) includes both ABA and a relationship-based approaches. Extension of the child's functional

behavioural repertoire, enhancing his/her communication skills and increasing the reinforcing value of social interactions are considered the ways to reduce RRBI. The practitioner applies behavioural strategies to manage behaviours considered destructive or disruptive, emphasizing the replacement of repetitive behaviours with more adaptive, communicative, developmentally mature ones (Rogers & Dawson, 2010).

Other interventions, which focus mainly on parent-child joint attention and communication (e.g., Green et al., 2010; Kasari et al., 2014) do not specify their theoretical and clinical approach to the child's RRBI. In Harrop's (2015) review of 29 evidence-based, parent-mediated EIs in the context of RRBI, none of these parent-mediated interventions focused primarily on RRBI as a primary intervention target or outcome. Additionally, the majority of these interventions did not even include strategies to address RRBI.

Measuring RRBI Outcomes in Comprehensive Early Interventions

Comprehensive EI models attempt to reduce RRBI that interfere with learning by focusing on expanding social and communicative skills and behaviours so that stereotyped and repetitive behaviours within the child's repertoire are reduced as a result of social interactions becoming more rewarding. Thus, as the RRBI are not a direct target of the intervention, they are typically not assessed or reported as outcome measures in intervention studies (Harrop, 2015). The improvements documented among children in early intensive interventions are mainly in social-communication skills, overall ASD symptomatology and change in diagnostic status; other commonly reported outcomes are language and cognition abilities and adaptive behaviours (French & Kennedy, 2018; Harrop, 2015).

A systematic review on measures to assess intervention outcomes (McConachie et al., 2015) allows insights regarding how RRBI related outcomes are assessed in the context of early intervention. Measures of RRBI and Sensory processing were examined separately. The measures to assess outcomes of interventions that met inclusion criteria of the review were the RRB scale of the ADOS, which is an observational measure, the RRB scale of the Autism Diagnostic Interview (ADI) and the Repetitive Behaviours Scale-Revised (RBS-R) to collect parent reports on child's RRBI. Measures to assess sensory-related behaviours included in the review were the Sense and Self-Regulation Checklist, Sensory Profile and Short Sensory Profile. Examining the psychometric properties of these measures, the ADOS was the measure with documented moderate sensitivity to change. There is limited evidence suggesting sensitivity to change of the ADI, and there is no available evidence for the RBS-R or sensory measures regarding sensitivity to change (McConachie et al., 2015). Thus, RRBI are measured in interventions as part of the diagnostic process, or as part of an autism severity outcome; yet, in order to measure RRBI and potential changes in them following intervention, there is a clear need to develop and utilise measures that are more individualised and (more) sensitive to change.

Evidence for changes in RRBI in the context of EI is limited compared to other outcomes. The ADOS total algorithm score is comprised of the Social Affect (SA) and Restricted Repetitive Behaviour (RRB) scales. Interestingly, ASD symptomatology outcomes in EI studies usually include total ADOS algorithm scores or the SA scales while changes in the RRB scales are less commonly reported (French & Kennedy, 2018; Harrop, 2015). Next, we will review the evidence from therapist and parent delivered EI on RRBI.

Therapist-Delivered Early Interventions and RRBI

In the ESDM randomised controlled trial (RCT), 48 toddlers with ASD were randomised to intensive ESDM or to a typical community treatment (e.g. developmental preschool which typically includes special education and related services such as speech and language therapy and occupational therapy). The RRBI outcomes were assessed via parent reports on the RBS. While finding significant improvements in children's cognitive and adaptive functioning and change in their ASD diagnosis, there was little evidence of change in parent reported RRBI (Dawson et al., 2010). In a 2-year follow-up, the SA and RRB scales of the ADOS served as separate outcome measures. Here, demonstrating long-term efficacy, the ESDM intervention groups showed significantly lower ADOS total scores (indicating less symptoms) compared to the treatment as usual group; the intervention group had also lower RRB scores – an unexpected result that was not observed in the short-term follow up. Scores on the parent-reported RRBI, as in the early follow-up study, did not differ between groups in the 2-year follow up (Estes et al., 2015).

Boyd et al. (2011) conducted a multi-site longitudinal study involving 198 children who were participating in three different CTMs. Overall significant gains and improvements in social-communication skills and ASD severity were reported among all children. However, RRBI across all groups remained constant over time, based on both parent and teacher reports on the RBS (Boyd, McDonough, Rupp, Khan, & Bodfish, 2011). Different results were observed in a study involving 86 children who received intensive EI services in Greece (average of 24 hours per week). Makrygianni and Reed (2010) found reductions in RRBI following 9 months of EI, as assessed via parental reports with the RBS (Makrygianni & Reed, 2010).

Effects of PRT on RRBI were documented in an open-trial involving 15 children, who received 16 weeks of PRT. RRBI were assessed with parental report on the RBS-R and the Stereotypy subscale of the Aberrant Behaviour Checklist. Regardless of initial severity, significant reductions in RRBI from baseline to the endpoint were documented for a variety of RRBI. Interestingly, this improvement was independent of the improvements in the social-communication domain - thus suggesting a more direct effect of the PRT on RRBI, although this EI explicitly targets social-communication behaviours (Ventola et al., 2016).

Parent-Mediated Interventions and RRBI

In the Early Social Interaction (ESI) trial (Wetherby et al., 2014), 82 children with ASD and their caregivers were randomised to two different types of intervention delivery – i.e., individual versus group ESI. Improvements from baseline to the end of the 9-month interventions were documented in all children's social-communication behaviours, measured with the ADOS-SA scale. No difference was found between the two interventions on RRBI, measured by the ADOS-RRB scale, which were found to increase over time similarly in both groups (Wetherby et al., 2014). This increase in RRBI, regardless of intervention, resonates with evidence from observational studies, documenting trajectories of increasing RRBI over time among young children with ASD (Richler, Huerta, Bishop, & Lord, 2010; Wolff et al., 2014).

In the pilot study of the parent-mediated communication trial (PACT), 28 preschool children were randomized to the intervention, involving psycho-educational sessions for parents comprising six monthly treatment sessions and six maintenance sessions versus routine care alone (Aldred et al., 2004). The intervention focus is on parental communication behaviour during interaction with their child, aimed at enhancing shared attention and parental responsivity. The results indicated a non-significant ($p = .086$) improvement on the ADOS-RRB scale in the intervention group compared to the control group. However, in the later PACT RCT, ASD severity was assessed with the ADOS-SA scale and total algorithm score, with small intervention effects; results were not reported separately for the ADOS-RRB scale (Green et al., 2010).

In a pilot study of another parent-mediated intervention for one-year-old children at risk for ASD, 16 children were randomised into the intervention group versus referral to community services (Baranek et al., 2015). The Sensory Processing Assessment, a play-based measure, and the parental report on the Sensory Experience Questionnaire were used to measure children's hyper- and hypo-responsiveness to sensory stimuli. Compared to children who were referred to community services, children in the intervention group showed better receptive language skills, their parents showed less directive interaction behaviour and reported better communication and socialisation adaptive skills thus demonstrating positive effects of the intervention with regards to child social communication. Parents in the intervention group reported *higher* levels of their child's hyper-responsiveness and *lower* levels of hypo-responsiveness than parents in the control group thus demonstrating mixed results with regards to parental report on child sensory responsivity. Observed-based child responsivity did not significantly change following the intervention. However, these outcomes were not replicated in a recent RCT, including 87 children, that showed minimal evidence of intervention efficacy on children's outcomes (Watson et al., 2017).

Harrop et al. (2016), Harrop, McBee, et al. (2016) examined the effects of a 10-week caregiver-mediated JASPER (Joint Attention, Symbolic Play, Engagement, and Regulation) intervention on RRBI. This study is one of the first to use a detailed behavioural coding of videotaped parent-child interactions, before, after and 6 months post-intervention to assess RRBI related outcomes. The videos were

coded for three RRBI variables: the occurrence and type of child RRBs; parental response to the child RRB (i.e., did the parent respond to the child behaviour and if so was it a verbal, physical, or redirection response); and success of parental response (i.e., was it followed by the child stopping the behaviour or engaging in a positive, communicative behaviour). As the intervention, targeting social-communication behaviours, has previously shown effects on child joint engagement, play, and parental behaviour, 'spill-over effects' were expected on RRBI. This study involved 86 child-caregiver dyads already receiving intensive intervention, randomised to additional active JASPER coaching or to additional parent-education weekly sessions. Based on coding of the videos, all children, regardless of intervention group, showed stable rates of RRBI during the intervention, and an *increase* in the 6-months follow-up. The authors suggest that this trend is in line with previous studies on trajectories of RRBI in children with ASD, and the relative lack of change in child RRBI is understood in light of the focus of the intervention on social-communication behaviours. Although there was no improvement in the child's observed RRBI, changes in parental behaviour in the parent-child interaction in regards to RRBI was observed. An improvement in caregiver's responses to the child's RRBs was observed for both groups, but was larger for the JASPER group, who responded to more child RRBI - i.e., more of the child's RRBI were followed by a parental response and not ignored or un-noticed. The success rates of parental responses improved for both groups as well, to a slightly larger extent in the JASPER group (Harrop et al., 2016).

To summarise, as apparent from the studies reviewed, there is limited evidence regarding effects of EI on child's RRBI. The results also appear mixed, with initial results differing from follow up studies (e.g. Dawson et al., 2010; Estes et al., 2015) and pilot results not replicated in the main studies (e.g. Baranek et al., 2015; Watson et al., 2017). Some studies show decreases in RRBI following intervention, whilst others show stability or increased RRBI over time. Parent-mediated interventions, which are increasingly common, often do not include strategies for parents to respond to their child's RRBs. Notably, these interventions are designed around parent-child interactions. Child RRBI occur frequently during parent child interactions, and are commonly followed by various parental responses, which are not always successful in stopping or redirecting the child (Harrop et al., 2016; Harrop, Tu, Landa, Kasier, & Kasari, 2018) resulting in increased parental distress (Harrop, McBee, et al., 2016). Thus even if the parent-mediated intervention does not explicitly target reduction of RRBI, it is important to include strategies to support and direct parents regarding how to respond to their child's behaviour, including RRBI, in a way that will enhance communication, joint engagement and learning opportunities (Harrop, 2015).

RRBI as Predictors of Early Intervention Outcomes

Behavioural characteristics related to RRBI were examined in several EI studies as potential predictors of treatment outcomes, identifying behavioural profiles of children who may be more or less responsive to a specific intervention. In a study com-

paring two social-skills interventions (Shih et al., 2016), different 'responder profiles' were identified by conducting assessments at baseline, mid- and end-points of the intervention. These profiles were determined by the initial levels of social engagement and the extent and rate of progress in social engagement during the intervention. These responder-groups differed on several baseline characteristics, including the RRB domain of the ADOS. The group of children who entered the study with low initial engagement and who did not make substantial progress in the intervention had the highest initial levels of RRBI compared to children in the other sub-groups (Shih, Patterson, & Kasari, 2016), again indicating that RRBI may interfere with learning.

Vocal repetitiveness/stereotypy was examined as a potential predictor of response to PRT intervention in two studies, yielding conflicting results. Using a single subject design, Sherer and Schreibman (2005) examined videos of the baseline assessments of six children receiving PRT to identify behavioural differences between responders and non-responders. The results showed that appropriate engagement with toys, less avoidance of people, and more stereotyped and repetitive vocalizations/verbalizations at baseline characterized children who made more gains in the intervention – i.e., 'responders' (Sherer & Schreibman, 2005). Different results with regards to vocal repetitiveness were reported in a later study, with a community sample of 57 children who participated in a 1-year intervention. The children who showed greatest gains in the expressive language domains were characterized at baseline by higher expressive language and cognitive skills, more positive affect and appropriate toy engagement, less social avoidance and **less** stereotyped and repetitive vocalizations (Fossum, Williams, Garon, Bryson, & Smith, 2018). The authors suggest that the discrepancy between the studies may be explained by the different baseline characteristics of the participating children and the outcome measure. In the 2005 study, children had lower cognitive and spoken language skills relative to those in the later study. It may be that among these children, any vocal production, even repetitive, provided interaction and teaching opportunities and thus were associated with improved gains. For the children in the later study, in which outcomes were measured in terms of gains in expressive language, more repetitive vocalizations may have reduced progress in expressive language acquisition (Fossum et al., 2018).

Measuring RRBI Outcomes in Focused Intervention Practices

Several reviews on FIPs addressing RRBI are available (DiGennaro Reed, Hirst, & Hyman, 2012; Odom, Boyd, et al. 2010; Odom et al. 2010; Patterson, Smith, & Jelen, 2010; Rapp & Vollmer, 2005; Raulston & Machalicek, 2017). In general, many of these EIs stem from behavioural science and ABA. Strategies to reduce unwanted behaviours, also called Positive Behavioural Support, are conceptualised as antecedent-based or consequence-based. Antecedent-based strategies change the conditions before the targeted behaviour occurs by modifying the environment and/

or the child's repertoire in a way that will reduce the likelihood of the unwanted behaviour to occur. Consequence-based strategies focus on what happens after the unwanted behaviour occurs, attempting at un-coupling or disrupting the association between the behaviour and the reinforcing consequence.

Antecendent-based strategies for RRBI's include enriching the child's environment with competing alternative reinforcers – i.e., more adaptive toys that he/she likes, removing positive reinforcers of the non-adaptive behaviour, or providing "matched" alternatives such as more adaptive objects for RRBI (environment modification/stimulus control). Notably, it may be insufficient to simply introduce alternative objects and activities, so that the practitioner prompts the child to engage with the alternative objects and to engage in other behaviours. Antecendent-based strategies may also include expanding the child's behavior and play repertoire and teaching him/her alternative ways to communicate his/her needs (skills enrichment, functional communication training). Other antecedent based startegies may include visual cues and schedules or video guided technologies to indicate times when a child is allowed or not to engage in specific behaviours and to guide transitions between activities (Boyd et al., 2012; Odom, Boyd, et al. 2010; Odom et al. 2010). Another strategy that has shown some promise in reducing stereotyped behaviour, yet not fully understood, is physical exercise (Boyd et al., 2012). Bremer, Crozier, and Lloyd (2016) recently conducted a comprehensive systematic review to examine effects of a range of exercise interventions (jogging, horseback riding, martial arts, swimming or yoga/dance) on various outcomes amongst children with ASD. In six studies involving RRBI-related outcomes, children engaged in physical activity prior to an activity in which RRBI commonly occur. A significant decrease in RRBI was documented in five out of the six studies. In one study in which RRBI were assessed at post-intervention and 30 days post-intervention, significant reductions in RRBI were observed from pre to post-intervention, but not at the follow-up. Thus, it seems that while showing immediate effects, there is still a need to maintain these effects. It has been hypothesized that the physical activity may provide the child with a similar, competing intrinsic reinforcer as the stereotypic behaviour, or that it changes the child's arousal levels and thus decreasing the child's need to engage in RRBI as a means of regulating his/her arousal (Boyd et al., 2012).

Consequent-based interventions include stopping the child from engaging in the RRBI by physically or verbally interrupting and redirecting his/her to another behaviour (response interruption/redirection), uncoupling the behaviour-reinforcer association by removing or terminating the reinforcer (extinction), and reinforcing alternative behaviours (differential reinforcement). The intervention may build opon the child's restricted play and expand it. For example, the pratctioner may imitate the child's repetitive behaviour of driving a car back and forth, or lining up cars, and gradually expand the child's repertoire to include more symbolic and social elements – such as two cars crashing or chasing, cars having a driver and passengers, lining up to go into a garage to be fixed, etc. (Koegel & Koegel, 2006; Rogers & Dawson, 2010). Different interventions have been recommended for different types of RRBI as listed below (Boyd et al., 2012; Lanovaz

& Sladeczek, 2012; Odom, Boyd, et al. 2010; Odom et al. 2010; Raulston & Machalicek, 2017).

For *repetitive body movements, vocalisations and object manipulation (stereotypies*), the recommended antecedent-based strategies include environmental modification and enrichment, skill enrichment, functional communication training, visual cues and schedules, and physical exercise. Consequence-based strategies include extinction, response blocking, interrupting or redirecting, and differential reinforcement. For *insistence on sameness,* differential reinforcement is suggested to expand behaviours and encourage novel interests, and visual schedules and video-based technologies are recommended to ease difficulties in tolerating changes and uncertainties in routine. For *circumscribed interests*, as an intense interest in an area may not necessarily interfere with functioning, but could actually be a strength, interventions are generally not deemed as necessary. Rather, the child's motivation in the circumscribed area is commonly built upon and capitalised for teaching new skills and improving social and comunication skills. These intersets can be used in antecedent-based strategies, including the child's interest in an activity to increase motivation or as a consequence; for example using the Premack principle with a child's interest – when teaching a child to sort toys, pack-away or to perform an activity he/she does not want to, the practiotioner can offer the motivating activity/interest after the child has completed the less-desired activity.

Odom and collegues (Odom, Boyd, et al. 2010; Odom et al. 2010) identified 24 practices (FIPs) adressing a range of targets for children with ASD that met predefined criteria for evidence-based practice. The Positive Behavioural Support strategies that were identified as established evidence-based practices included: functional behavioural analysis, stimulus control/environmental modification, response interruption/redirection, functional communication training, extinction, and differential reinforcement. Addional evidence-based practices identified by Boyd et al. (2012) in a review that focused on the higher-order RRBI were cognitive behaviour therapy techniques of cognitive reframing and exposure, and visual schedules, however these are only suitable for older and more able individuals with ASD.

In their review, DiGnennaro and colleagues (2012) provide a descriptive overview of empirical studies using behavioural interventions to treat stereotypy (motor, vocal repetitive behaviours and non-functional manipulation of objects) in 3- to 18-year old children with ASD. Summarising evidence of assessment and intervention practices for a total of 128 individuals, the authors note that the majority of the intervention studies did not include a functional behavioural assessment to identify the function of the behaviour. The common strategies were consequence-based and despite its critical importance, most studies did not include a measurement of treatment integrity (DiGennaro Reed et al., 2012).

Given the potential association between RRBI and social learning, Lanovaz, Robertson, Soerono, and Watkins (2013) conducted a systematic review exploring the 'spill-over' effects of reduction of RRBI on other behaviours (Lanovaz et al., 2013). The review included 60 studies, mostly case studies, in which strategies to reduce RRBI were effective, and another behavioural outcome was measured,

including a total of 218 individuals with ASD and other developmental disabilities. The results of this review suggest that, in general, the reduction of stereotyped behaviours may be associated with an increase in other behaviours. Notably, sometimes these are adaptive behaviours, but sometimes the targeted reduced behaviours are replaced by other non-adaptive behaviours. Thus, it is important, in planning the intervention, to purposefully strengthen alternative adaptive behaviours. Focusing on eliminating the RRBI in itself may not be sufficient without introducing new alternative activities (Lanovaz et al., 2013).

Parent-Mediated Focused Interventions for RRBI

Several parent-delivered interventions have been developed to specifically target RRBI. Boyd et al. (2011) developed the Family-Implemented Treatment for Behavioural Inflexibility (FITBI) co-implemented by a therapist and parents of five children with ASD over 12 weeks. A significant decrease in RRBI was documented for all participants at post-intervention, and maintained for most of them (Boyd et al., 2011). In another single case pilot study involving three young children with ASD, Lin and Koegel (2018) used an intervention based on self-management and PRT principles, specifically designed to address high-order RRBI. The intervention aims at expanding children's interests and improving behavioural flexibility, with gains noted in observed and parent-reported child flexibility and an increased variety of activities engaged in by the children. An increase in positive parent and child affect during interaction was also observed, as well as overall reduction in parent ratings on the Repetitive Behaviour Questionnaire (Lin & Koegel, 2018).

The 'Managing Repetitive Behaviours' program (Grahame et al., 2015) is an 8-week group intervention for parents, specifically targeting RRBI. It is designed to help parents understand RRBI and apply functional analysis and behavioural strategies to effectively address their child's RRBI. The intervention was developed in consultation with parents, incorporating evidence-based practices, video feedback, interactive activities and emphasis on mutual support and knowledge-sharing among parents, to build their confidence and capacity in managing their child's behaviour. A pilot RCT of the program involving 25 families assessing feasibility, acceptability and initial outcomes has shown promising positive results. A larger-scale RCT is now needed to establish the efficacy of the program (Grahame et al., 2015).

Sensory-Based Interventions

Although evidence suggests that most individuals with ASD have sensory related difficulties, which have substantial effect on learning and daily functioning (Lane, Young, Baker, & Angley, 2010; Weitlauf, Sathe, McPheeters, & Warren, 2017), the

evidence base for the diagnosis and intervention of sensory difficulties is still developing (Uljarević et al., 2017), particularly given their recent inclusion as diagnostic criteria (APA, 2013). Sensory -focused strategies commonly target sensory aversions (e.g. sensitivity to light and sounds), and address processing deficiencies (e.g. prolonged visual examination, sensory seeking, hypo-responsivity). Broadly, interventions targeting sensory challenges involve the incorporation of sensory experiences (e.g. sounds, texture, pressure), and are largely dominated by Sensory Integration therapy and Sensory-based approaches. Sensory Integration therapy is a clinic-based approach using combinations of sensory and kinetic stimuli in child-directed activities to improve the child's adaptive responses. Sensory-based interventions are characterized as classroom-based interventions that use single-sensory strategies such as balls, vests, or swings to influence a child's state of arousal (Case-Smith, Weaver, & Fristad, 2015).

There is some promising evidence on the efficacy of sensory-based interventions in goal attainment and the reduction of negative response to sensory activities (e.g., Fazlioğlu & Baran, 2008; Schaaf et al., 2014; see Weitlauf et al., 2017 for a review) but mixed results are also apparent (Barton, Reichow, Schnitz, Smith, & Sherlock, 2015; Watling & Hauer, 2015). The evidence to support the use of sensory-integration therapy is only moderate to date. Despite substantial progress over the last years and initial promising results, considerable heterogeneity in study design and populations, restricted study quality with high risk of bias, limited follow-up periods, and lack of treatment fidelity- limits the evidence base for these interventions. It remains unclear how these interventions work, what the underlying mechanisms targeted are, as well as how generalizable any improvements may be over time to other settings (Barton et al., 2015; Case-Smith et al., 2015; Weitlauf et al., 2017). Larger studies are needed with adequate samples, using fidelity measures, and longer-term follow-ups with carefully operationalized definitions and systematic methods to address the efficacy of sensory integration therapy for children with ASD.

Other interventions to address a range of sensory related difficulties in ASD have been identified in a recent systematic review. These include interventions based on environmental enrichment, auditory integration, music-therapy, massage, tactile-based tasks and weighed blankets (Weitlauf et al., 2017). These strategies and techniques are usually employed in conjunction with other interventions, with mixed evidence so far, from relatively small and potentially biased studies to support efficacy for these approaches. Environmental enrichment strategies involve exposure to the sensory stimuli the child shows aversion to, in order to promote his/her tolerance of them. Evidence from two small RCTs involving the same protocol, suggests efficacy in improving sensory reactivity as well as ASD symptomatology, receptive language and non-verbal IQ following an environment-enrichment protocol (Weitlauf et al., 2017; Woo, Donnelly, Steinberg-Epstein, & Leon, 2015; Woo & Leon, 2013). Interventions incorporating auditory components, such as filtered sound to ameliorate sensory processing challenges show some evidence of improvement in parent-reported hearing sensitivity from several studies, with relatively small samples with potential risk of bias (Weitlauf et al., 2017). Music therapy-

based interventions, involving playing or singing music, or movement to music, show mixed evidence, with improvements in social-communication outcomes documented in several small RCTs, however RRBI or sensory behavioural outcomes were not reported (e.g., Gattino, Riesgo, Longo, Leite, & Faccini, 2011; Srinivasan, Eigsti, Gifford, & Bhat, 2016). As these interventions varied in their techniques and strategies, and sample sizes were relatively small it is difficult to generalise conclusions regarding music-therapy based interventions across the different studies (Geretsegger, Elefant, Mössler, & Gold, 2014; Weitlauf et al., 2017). Massage-based interventions incorporate touch-based approaches by a therapist or caregiver. Studies involving massage compared either massage intervention versus no massage, or massage intervention added to other treatments versus treatment without massage intervention. Results from these studies show promising evidence that massage can improve ASD symptom severity and sensory-related difficulties (Lee, Kim, & Ernst, 2011; Weitlauf et al., 2017). Various alternative and complementary therapies and techniques are in wide use for children with ASD for a variety of difficulties, including RRBI (Höfer, Hoffmann, & Bachmann, 2017; Perrin et al., 2012). However, there is insufficient evidence regarding their efficacy in improving children's outcomes despite their extensive use, and therefore further research is needed.

Summary and Conclusion

While there has been much progress in measuring social, communication, cognitive and adaptive functioning in the context of ASD early intervention, there is relative paucity of evidence on the outcomes of interventions on RRBI. There may be a few reasons accounting for this lack of outcome data. First, social communication deficits are considered primary to ASD while RRBI are also evident in other conditions. The heterogeneity in RRBI in terms of their clinical significance, functions and underlying mechanisms may also contribute to the relative lack of EI research on RRBI. Finally, the availability of standardised tools to assess RRBI, based on behavioural observation and parental report that are sensitive to change is limited.

The evidence from studies in which RRBI related outcomes were reported has yielded mixed results regarding the effects of intensive EI programs on children's RRBI, suggesting the need for more well-designed research in this area. It is also important to consider the association between parental stress and children's RRBI, which indicates the need to support parents so that they can appropriately and effectively respond to and manage their child's RRBI. Indeed, it is important to increase knowledge about RRBI more generally amongst both parents and professionals.

Both prospective observational and intervention studies provide valuable information on how child baseline features may serve as prognostic factors or moderating factors in intervention. With large heterogeneity in intervention outcomes, it is understood that intervention is not a "one size fits all" such that a specific intervention will be more effective for children with specific characteristics (e.g. age, lan-

guage abilities). Hence, the importance of individualised treatment plans is increasingly acknowledged. Given emerging evidence that children's RRBI characteristics may impact intervention outcomes, RRBI characteristics, their context, functions, and interference with learning should be considered as potential moderators/mediators of intervention outcomes. These topics deserve more research, and the assessment of RRBI is thus important in the process of choosing and planning an intervention.

Assessing RRBI as part of planning an intervention differs from assessing the RRBI for diagnostic purposes where the focus is on the presence of these behaviours. Knowing that RRBI are present (as needed for a diagnosis) is not sufficient information for planning an intervention. It is important to assess how the RRBI relate to the other difficulties, which are the particular RRBI behaviours that impede social relatedness, and which should be targeted with the appropriate strategies to reduce, expand, constrict to specific times or replace with more adaptive or socially acceptable behaviours. If behaviours serve a function, alternative ways to address the need should be considered. RRBI can also be identified as potential reinforcers to be used to facilitate learning and skill acquisition, while other behaviours that do not disrupt learning and daily function should be accepted, tolerated, and perhaps even encouraged to facilitate better outcomes for people with ASD.

References

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5th ed.). Washington, DC: Author.

Aldred, C., Green, J., & Adams, C. (2004). A new social communication intervention for children with autism: pilot randomised controlled treatment study suggesting effectiveness. *Journal of Child Psychology and Psychiatry, 45*(8), 1420–1430.

Ausderau, K. K., Sideris, J., Little, L. M., Furlong, M., Bulluck, J. C., & Baranek, G. T. (2016). Sensory subtypes and associated outcomes in children with autism spectrum disorders. *Autism Research, 9*(12), 1316–1327. https://doi.org/10.1002/aur.1626.

Baranek, G. T. (1999). Autism during infancy: A retrospective video analysis of sensory-motor and social behaviors at 9-12 months of age. *Journal of Autism and Developmental Disorders, 29*(3), 213–224. https://doi.org/10.1023/A:1023080005650.

Baranek, G. T., Watson, L. R., Turner-Brown, L., Field, S. H., Crais, E. R., Wakeford, L., et al. (2015). Preliminary efficacy of adapted responsive teaching for infants at risk of autism spectrum disorder in a community sample. *Autism Research and Treatment, 2015*. https://doi.org/10.1155/2015/386951.

Barber, A. B., Wetherby, A. M., & Chambers, N. W. (2012). Brief report: Repetitive behaviors in young children with autism spectrum disorder and developmentally similar peers: A follow up to Watt et al. (2008). *Journal of Autism and Developmental Disorders, 42*(9), 2006–2012. https://doi.org/10.1007/s10803-011-1434-3.

Baron-Cohen, S. (2017). Editorial perspective: Neurodiversity – a revolutionary concept for autism and psychiatry. *Journal of Child Psychology and Psychiatry, 58*(6), 744–747. https://doi.org/10.1111/jcpp.12703.

Barton, E. E., Reichow, B., Schnitz, A., Smith, I. C., & Sherlock, D. (2015). A systematic review of sensory-based treatments for children with disabilities. *Research in Developmental Disabilities, 37*, 64–80. https://doi.org/10.1016/j.ridd.2014.11.006.

Bodfish, J. W., Symons, F. J., Parker, D. E., & Lewis, M. H. (2000). Varieties of repetitive behavior in autism: Comparisons to mental retardation. *Journal of Autism and Developmental Disorders, 30*(3), 237–243. https://doi.org/10.1023/A:1005596502855.

Boyd, B. A., McDonough, S. G., & Bodfish, J. W. (2012). Evidence-based behavioral interventions for repetitive behaviors in autism. *Journal of Autism and Developmental Disorders, 42*(6), 1236–1248. https://doi.org/10.1007/s10803-011-1284-z.

Boyd, B. A., McDonough, S. G., Rupp, B., Khan, F., & Bodfish, J. W. (2011). Effects of a family-implemented treatment on the repetitive behaviors of children with autism. *Journal of Autism and Developmental Disorders, 41*(10), 1330–1341. https://doi.org/10.1007/s10803-010-1156-y.

Bremer, E., Crozier, M., & Lloyd, M. (2016). A systematic review of the behavioural outcomes following exercise interventions for children and youth with autism spectrum disorder. *Autism, 20*(8), 899–915.

Case-Smith, J., Weaver, L. L., & Fristad, M. A. (2015). A systematic review of sensory processing interventions for children with autism spectrum disorders. *Autism, 19*(2), 133–148. https://doi.org/10.1177/1362361313517762.

Dawson, G. (2008). Early behavioral intervention, brain plasticity, and the prevention of autism spectrum disorder. *Development and Psychopathology, 20*(3), 775–803. https://doi.org/10.1017/S0954579408000370.

Dawson, G., Rogers, S., Munson, J., Smith, M., Winter, J., Greenson, J., et al. (2010). Randomized, controlled trial of an intervention for toddlers with autism: The Early Start Denver Model. *Pediatrics, 125*(1), e17–e23. https://doi.org/10.1542/peds.2009-0958.

DiGennaro Reed, F. D., Hirst, J. M., & Hyman, S. R. (2012). Assessment and treatment of stereotypic behavior in children with autism and other developmental disabilities: A thirty ear review. *Research in Autism Spectrum Disorders, 6*(1), 422–430. https://doi.org/10.1016/j.rasd.2011.07.003.

Estes, A., Munson, J., Rogers, S. J., Greenson, J., Winter, J., & Dawson, G. (2015). Long-term outcomes of early intervention in 6-year-old children with autism spectrum disorder. *Journal of the American Academy of Child & Adolescent Psychiatry, 54*(7), 580–587. https://doi.org/10.1016/j.jaac.2015.04.005.

Fazlioğlu, Y., & Baran, G. (2008). A sensory integration therapy program on sensory problems for children with autism. *Perceptual and Motor Skills, 106*(2), 415–422.

Fossum, K. L., Williams, L., Garon, N., Bryson, S. E., & Smith, I. M. (2018). Pivotal response treatment for preschoolers with autism spectrum disorder: Defining a predictor profile. *Autism Research, 11*, 153–165. https://doi.org/10.1002/aur.1859.

French, L., & Kennedy, E. M. M. (2018). Annual research review: Early intervention for infants and young children with, or at-risk of, autism spectrum disorder: A systematic review. *Journal of Child Psychology and Psychiatry, 59*(4), 444–456. https://doi.org/10.1111/jcpp.12828.

Gattino, G. S., Riesgo, R. D. S., Longo, D., Leite, J. C. L., & Faccini, L. S. (2011). Effects of relational music therapy on communication of children with autism: A randomized controlled study. *Nordic Journal of Music Therapy, 20*(2), 142–154.

Geretsegger, M., Elefant, C., Mössler, K. A., & Gold, C. (2014). Music therapy for people with autism spectrum disorder. *Cochrane Database of Systematic Reviews, 6*(6), CD004381.

Grahame, V., Brett, D., Dixon, L., McConachie, H., Lowry, J., Rodgers, J., et al. (2015). Managing repetitive behaviours in young children with autism spectrum disorder (ASD): Pilot randomised controlled trial of a new parent group intervention. *Journal of Autism and Developmental Disorders, 45*, 3168–3182. https://doi.org/10.1007/s10803-015-2474-x.

Green, J., Charman, T., McConachie, H., Aldred, C., Slonims, V., Howlin, P., et al. (2010). Parent-mediated communication-focused treatment in children with autism (PACT): A randomised controlled trial. *The Lancet, 375*(9732), 2152–2160. https://doi.org/10.1016/S0140-6736(10)60587-9.

Harrop, C. (2015). Evidence-based, parent-mediated interventions for young children with autism spectrum disorder: The case of restricted and repetitive behaviors. *Autism, 19*(6), 662–672. https://doi.org/10.1177/1362361314545685.

Harrop, C., Gulsrud, A., Shih, W., Hovsepyan, L., & Kasari, C. (2016). Characterizing caregiver responses to restricted and repetitive behaviors in toddlers with autism spectrum disorder. *Autism, 20*(3), 330–342. https://doi.org/10.1177/1362361315580443.

Harrop, C., McBee, M., & Boyd, B. A. (2016). How are child restricted and repetitive behaviors associated with caregiver stress over time? A parallel process multilevel growth model. *Journal of Autism and Developmental Disorders, 46*(5), 1773–1783. https://doi.org/10.1007/s10803-016-2707-7.

Harrop, C., McConachie, H., Emsley, R., Leadbitter, K., Green, J., & PACT Consortium. (2014). Restricted and repetitive behaviors in autism spectrum disorders and typical development: Cross-sectional and longitudinal comparisons. *Journal of Autism and Developmental Disorders, 44*(5), 1207–1219. https://doi.org/10.1007/s10803-013-1986-5.

Harrop, C., Tu, N., Landa, R., Kasier, A., & Kasari, C. (2018). Sensory behaviors in minimally verbal children with autism spectrum disorder: How and when do caregivers respond? *American Journal on Intellectual and Developmental Disabilities, 123*(1), 1–16. https://doi.org/10.1352/1944-7558-123.1.1.

Höfer, J., Hoffmann, F., & Bachmann, C. (2017). Use of complementary and alternative medicine in children and adolescents with autism spectrum disorder: A systematic review. *Autism, 21*(4), 387–402.

Kapp, S. K., Gillespie-Lynch, K., Sherman, L. E., & Hutman, T. (2013). Deficit, difference, or both? Autism and neurodiversity. *Developmental Psychology, 49*(1), 59–71. https://doi.org/10.1037/a0028353.

Kasari, C., Siller, M., Huynh, L. N., Shih, W., Swanson, M., Hellemann, G. S., & Sugar, C. A. (2014). Randomized controlled trial of parental responsiveness intervention for toddlers at high risk for autism. *Infant Behavior and Development, 37*(4), 711–721. https://doi.org/10.1016/j.infbeh.2014.08.007.

Koegel, R. L., & Koegel, L. K. (2006). *Pivotal response treatments for autism: Communication, social, and academic development*. Baltimore, MD: Paul H. Brookes.

Lane, A. E., Young, R. L., Baker, A. E., & Angley, M. T. (2010). Sensory processing subtypes in autism: Association with adaptive behavior. *Journal of Autism and Developmental Disorders, 40*(1), 112–122. https://doi.org/10.1007/s10803-009-0840-2.

Lanovaz, M. J., Robertson, K. M., Soerono, K., & Watkins, N. (2013). Effects of reducing stereotypy on other behaviors: A systematic review. *Research in Autism Spectrum Disorders, 7*(10), 1234–1243. https://doi.org/10.1016/j.rasd.2013.07.009.

Lanovaz, M. J., & Sladeczek, I. E. (2012). Vocal stereotypy in individuals with autism spectrum disorders: A review of behavioral interventions. *Behavior Modification, 36*(2), 146–164. https://doi.org/10.1177/0145445511427192.

Larkin, F., Meins, E., Centifanti, L. C. M., Fernyhough, C., & Leekam, S. R. (2017). How does restricted and repetitive behavior relate to language and cognition in typical development? *Development and Psychopathology, 29*(3), 863–874. https://doi.org/10.1017/S0954579416000535.

Lee, M. S., Kim, J. I., & Ernst, E. (2011). Massage therapy for children with autism spectrum disorders: A systematic review. *The Journal of Clinical Psychiatry, 72*(3), 406–411.

Leekam, S., Tandos, J., McConachie, H., Meins, E., Parkinson, K., Wright, C., et al. (2007). Repetitive behaviours in typically developing 2-year-olds. *Journal of Child Psychology and Psychiatry, 48*(11), 1131–1138. https://doi.org/10.1111/j.1469-7610.2007.01778.x.

Leekam, S. R., Prior, M. R., & Uljarevic, M. (2011). Restricted and repetitive behaviors in autism spectrum disorders: A review of research in the last decade. *Psychological Bulletin, 137*(4), 562–593. https://doi.org/10.1037/a0023341.

Lin, C. E., & Koegel, R. (2018). Treatment for higher-order restricted repetitive behaviors (H-RRB) in children with autism Spectrum disorder. *Journal of autism and developmental disorders, 48*(11), 3831–3845.

MacDonald, R., Green, G., Mansfield, R., Geckeler, A., Gardenier, N., Anderson, J., et al. (2007). Stereotypy in young children with autism and typically developing children. *Research in Developmental Disabilities, 28*(3), 266–277. https://doi.org/10.1016/j.ridd.2006.01.004.

Makrygianni, M. K., & Reed, P. (2010). Factors impacting on the outcomes of Greek intervention programmes for children with autistic spectrum disorders. *Research in Autism Spectrum Disorders, 4*(4), 697–708. https://doi.org/10.1016/j.rasd.2010.01.008.

Matson, J. L., Dempsey, T., & Fodstad, J. C. (2009). Stereotypies and repetitive/restrictive behaviours in infants with autism and pervasive developmental disorder. *Developmental Neurorehabilitation, 12*(3), 122–127. https://doi.org/10.1080/17518420902936730.

McConachie, H., Parr, J. R., Glod, M., Hanratty, J., Livingstone, N., Oono, I. P., Robalino, S., Baird, G., Beresford, B., Charman, T., Garland, D., Green, J., Gringras, P., Jones, G., Law, J., Le Couteur, A. S., Macdonald, G., McColl, E. M., Morris, C., Rodgers, J., Simonoff, E., Terwee, C. B., & Williams, K. (2015). Systematic review of tools to measure outcomes for young children with autism spectrum disorder. *Health Technology Assessment, 19*(41), 1–506.

Odom, S. L., Boyd, B. A., Hall, L. J., & Hume, K. (2010). Evaluation of comprehensive treatment models for individuals with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 40*(4), 425–436. https://doi.org/10.1007/s10803-009-0825-1.

Odom, S. L., Collet-Klingenberg, L., Rogers, S. J., & Hatton, D. D. (2010). Evidence-based practices in interventions for children and youth with autism spectrum disorders. *Preventing School Failure: Alternative Education for Children and Youth, 54*(4), 275–282. https://doi.org/10.1080/10459881003785506.

Ozonoff, S., Macari, S., Young, G. S., Goldring, S., Thompson, M., & Rogers, S. J. (2008). Atypical object exploration at 12 months of age is associated with autism in a prospective sample. *Autism, 12*(5), 457–472. https://doi.org/10.1177/1362361308096402.

Patterson, S. Y., Smith, V., & Jelen, M. (2010). Behavioural intervention practices for stereotypic and repetitive behaviour in individuals with autism spectrum disorder: A systematic review. *Developmental Medicine and Child Neurology, 52*(4), 318–327. https://doi.org/10.1111/j.1469-8749.2009.03597.x.

Pellicano, E., & Stears, M. (2011). Bridging autism, science and society: Moving toward an ethically informed approach to autism research. *Autism Research, 4*(4), 271–282. https://doi.org/10.1002/aur.201.

Perrin, J. M., Coury, D. L., Hyman, S. L., Cole, L., Reynolds, A. M., & Clemons, T. (2012). Complementary and alternative medicine use in a large pediatric autism sample. *Pediatrics, 130*(Supplement 2), S77–S82.

Rapp, J. T., & Vollmer, T. R. (2005). Stereotypy I: A review of behavioral assessment and treatment. *Research in Developmental Disabilities, 26*(6), 527–547. https://doi.org/10.1016/j.ridd.2004.11.005.

Raulston, T. J., & Machalicek, W. (2017). Early intervention for repetitive behavior in autism spectrum disorder: A conceptual model. *Journal of Developmental and Physical Disabilities, 30*(1), 89–109. https://doi.org/10.1007/s10882-017-9566-9.

Ray-Subramanian, C. E., & Ellis Weismer, S. (2012). Receptive and expressive language as predictors of restricted and repetitive behaviors in young children with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 42*(10), 2113–2120. https://doi.org/10.1007/s10803-012-1463-6.

Richler, J., Huerta, M., Bishop, S. L., & Lord, C. (2010). Developmental trajectories of restricted and repetitive behaviors and interests in children with autism spectrum disorders. *Development and Psychopathology, 22*(1), 55–69.

Rogers, S. J. (2009). What are infant siblings teaching us about autism in infancy?. *Autism Research, 2*(3), 125–137.

Rogers, S. J., & Dawson, G. (2010). *Early Start Denver Model for young children with autism: Promoting language, learning, and engagement*. New York, NY: Guildford Press.

Schaaf, R. C., Benevides, T., Mailloux, Z., Faller, P., Hunt, J., Van Hooydonk, E., et al. (2014). An intervention for sensory difficulties in children with autism: A randomized trial. *Journal of Autism and Developmental Disorders, 44*(7), 1493–1506.

Shih, W., Patterson, S. Y., & Kasari, C. (2016). Developing an adaptive treatment strategy for peer-related social skills for children with autism spectrum disorders. *Journal of Clinical Child & Adolescent Psychology, 45*(4), 469–479.

Sherer, M. R., & Schreibman, L. (2005). Individual behavioral profiles and predictors of treatment effectiveness for children with autism. *Journal of Consulting and Clinical Psychology, 73*(3), 525.

Srinivasan, S. M., Eigsti, I.-M., Gifford, T., & Bhat, A. N. (2016). The effects of embodied rhythm and robotic interventions on the spontaneous and responsive verbal communication skills of children with Autism Spectrum Disorder (ASD): A further outcome of a pilot randomized controlled trial. *Research in Autism Spectrum Disorders, 27*, 73–87. https://doi.org/10.1016/j.rasd.2016.04.001.

The Boyd (2001) citation is an error, it should be the same reference as above from 2011.

Troyb, E., Knoch, K., Herlihy, L., Stevens, M. C., Chen, C.-M., Barton, M., et al. (2016). Restricted and repetitive behaviors as predictors of outcome in autism spectrum disorders. *Journal of Autism and Developmental Disorders, 46*(4), 1282–1296. https://doi.org/10.1007/s10803-015-2668-2.

Turner, M. (1999). Annotation: Repetitive behaviour in autism: A review of psychological research. *The Journal of Child Psychology and Psychiatry and Allied Disciplines, 40*(6), 839–849.

Uljarević, M., Baranek, G., Vivanti, G., Hedley, D., Hudry, K., & Lane, A. (2017). Heterogeneity of sensory features in autism spectrum disorder: Challenges and perspectives for future research. *Autism Research, 10*(5), 703–710. https://doi.org/10.1002/aur.1747.

Ventola, P. E., Yang, D., Abdullahi, S. M., Paisley, C. A., Braconnier, M. L., & Sukhodolsky, D. G. (2016). Brief report: Reduced restricted and repetitive behaviors after pivotal response treatment. *Journal of Autism and Developmental Disorders, 46*(8), 2813–2820. https://doi.org/10.1007/s10803-016-2813-6.

Watling, R., & Hauer, S. (2015). Effectiveness of Ayres Sensory Integration® and sensory-based interventions for people with autism spectrum disorder: A systematic review. *American Journal of Occupational Therapy, 69*(5), 6905180030p1–6905180030p12.

Watson, L. R., Crais, E. R., Baranek, G. T., Turner-Brown, L., Sideris, J., Wakeford, L., et al. (2017). Parent-mediated intervention for one-year-olds screened as at-risk for autism spectrum disorder: A randomized controlled trial. *Journal of Autism and Developmental Disorders, 47*(11), 3520–3540. https://doi.org/10.1007/s10803-017-3268-0.

Weitlauf, A. S., Sathe, N., McPheeters, M. L., & Warren, Z. E. (2017). Interventions targeting sensory challenges in autism spectrum disorder: A systematic review. *Pediatrics, 139*(6), e20170347. https://doi.org/10.1542/peds.2017-0347.

Wetherby, A. M., Guthrie, W., Woods, J., Schatschneider, C., Holland, R. D., Morgan, L., & Lord, C. (2014). Parent-implemented social intervention for toddlers with autism: An RCT. *Pediatrics, 134*(6), 1084–1093. https://doi.org/10.1542/peds.2014-0757.

Wolff, J. J., Botteron, K. N., Dager, S. R., Elison, J. T., Estes, A. M., Gu, H., et al. (2014). Longitudinal patterns of repetitive behavior in toddlers with autism. *Journal of Child Psychology and Psychiatry, 55*(8), 945–953. https://doi.org/10.1111/jcpp.12207.

Woo, C. C., Donnelly, J. H., Steinberg-Epstein, R., & Leon, M. (2015). Environmental enrichment as a therapy for autism: A clinical trial replication and extension. *Behavioral Neuroscience, 129*(4), 412.

Woo, C. C., & Leon, M. (2013). Environmental enrichment as an effective treatment for autism: A randomized controlled trial. *Behavioral Neuroscience, 127*(4), 487.

Chapter 14
"Rep-Mod": An Intervention Model for Restricted, Repetitive Behaviours and Interests

Eynat Gal and Ayelet Ben-Sasson

Introduction

Repetitive and restricted behaviors and interests (RRBI) are a defining characteristic of autism spectrum disorders (ASD) that exist across all levels of the spectrum (American Psychiatric Association, 2013). The umbrella term RRBI denotes a broad and heterogeneous class of behaviors linked by repetition, rigidity, invariance, and inappropriateness to the place and context in which they arise (Bodfish, Symons, Parker, & Lewis, 2000; Leekam, Prior, & Uljarevic, 2011; Yerys, 2015); however, they can vary in their frequency, intensity, variability, and severity (Gal, 2011; Wilkes & Lewis, 2018).

RRBI occur among individuals with different levels of intellectual ability, including those with high functioning ASD (Bodfish et al., 2000; South, Ozonoff, & McMahon, 2005). Researchers tend to classify RRBI into two categories: (a) lower-level RRBI, consisting of repetitive motor and sensory behaviors such as repetitive hand or finger movements, and (b) higher-level RRBI, consisting of insistence on sameness, narrow interests, rigid routines, and rituals (Cuccaro et al., 2003; Richler, Bishop, Kleinke, & Lord, 2007; Turner, 1999). Although research findings indicate that RRBI are less severe among older than younger individuals with ASD (Esbensen, Seltzer, Lam, & Bodfish, 2009; Shattuck et al., 2007), they remain a diagnostic criterion of ASD across the lifespan and continue to present challenges as these individuals age.

Society often looks negatively upon RRBI, considering them to be nonfunctional, nonadaptive, and involving socially isolating behaviors. Consequently, many interventions addressing RRBI focus on eliminating or reducing them to a minimum.

However, clinical behavioral observations in the 1970s (Rincover, Cook, Peoples, & Packard, 1979) underscored the potential of harnessing the motivation to engage

E. Gal (✉) · A. Ben-Sasson
Department of Occupational Therapy, University of Haifa, Mount Carmel, Haifa, Israel

E. Gal, N. Yirmiya (eds.), *Repetitive and Restricted Behaviors and Interests in Autism Spectrum Disorders*, Autism and Child Psychopathology Series,
https://doi.org/10.1007/978-3-030-66445-9_14

in RRBI to promote and advance learning skills and adaptive behaviors. In addition, autobiographical accounts, as well as various studies, indicate that these behaviors fulfill a particular need/purpose and have the potential to evolve into an occupation (Chamak, Bonniau, Jaunay, & Cohen, 2008; Jones, Quigney, & Huws, 2003). Therefore, to increase participation in daily activities, it is important to consider ways of integrating the individual's intense drive to engage in RRBI and, for certain individuals, their high skill level in their "restricted" area of interest. In this chapter, we describe a clinical model that guides individualized assessment of the function and fit between the person, the environment, and the occupation as related to RRBI to inform different types of interventions. It further presents practical ways of using specific types of RRBI in a manner that enhances the individual's participation in daily life activities. These include adding empowering behaviors that potentially assist in participation and reducing those that restrict participation. Lastly, the model presents reassessment and new goal setting. The presentation of the model is followed by case studies that demonstrate the model's implementation.

RRBI: Implications for the Intervention Model

Both low-level and high-level RRBI are generally considered as a serious concern for people with ASD, as well as for their family members, age peers, educators, and clinicians who work with them. Stereotyped movements (SM) and stereotyped manipulation of objects, which are low-level RRBI, often appear bizarre and grotesque, differ significantly from normal "play behavior," and challenge engagement in age-appropriate occupations such as social play, self-care, and academic learning (Cunningham & Schreibman, 2008; Joosten & Bundy, 2010). These types of RRBI tend to remain relatively stable over time (Esbensen et al., 2009), limiting the opportunity to develop more flexible or age-appropriate forms of regulation. Moreover, even adults with ASD who are cognitively able and aware of the inappropriateness of such behaviors within social contexts often feel compelled to perform them and may choose to isolate themselves to be able to perform these movements in privacy. As a result, a cycle of movements–social isolation–movements is created.

Early literature described low-level RRBI as a crucial factor in predicting outcomes across the lifespan. It listed SM among the maladaptive behaviors that contribute to institutional placement, as opposed to placement in less restrictive, community residential facilities (Eyman, Borthwick, & Miller, 1981). In addition, SM were identified as a prominent factor for increasing stress in families of infants with disabilities, leading to the possibility of dysfunctional family environments and eventually more restrictive residential placement (Beckman, 1983).

Like SM, sensory hyper- and hypo-responsivity, as well as unusual sensory interests, which are currently a part of the ASD diagnostic criteria for RRBI (APA, 2013), may challenge the functioning both of individuals with ASD and of those who support them (Harrop, McBee, & Boyd, 2016; Leekam et al., 2011). Hyper- or hypo-reactivity to sensory input is characterized by extreme or indifferent responses

to sensory information (e.g., tactile, vestibular, and proprioceptive; APA, 2013). Stimuli such as loud noises, strong lighting, and excessive sensory stimulation can cause distractions, make the task hard to perform, or become a source of stress (Hendricks, 2010).

The so called "high-level" RRBI, such as insistence on sameness or restricted interests, also may have negative implications for the individuals who perform them. They often may appear to be "odd" and inappropriate to the person's age or situation, and therefore can be socially stigmatizing (Cunningham & Schreibman, 2008). In addition, the person's restrictive and intense focus on interests may reduce opportunities for situations conducive to developing more flexible, functional, and elaborate cognitive and social abilities (Leekam et al., 2011). These symptoms can dramatically limit an individual's social repertoire or interaction (Attwood, 2003; Klin, Danovitch, Merz, & Volkmar, 2007).

The emotional aspects of RRBI are important to understand when considering the challenging nature of these behaviors. The stress provoked by the restriction and/or interruption in the performance of RRBI can increase the manifestation and, in turn, may make the disability (Wood & Gadow, 2010) and anxiety (Uljarević, Richdale, Evans, Cai, & Leekam, 2017) more pronounced. The negative emotionality involved in these behaviors also affects daily family routines and level of family impairment (Bagby, Dickie, & Baranek, 2012; Ben-Sasson, Soto, Martínez-Pedraza, & Carter, 2013).

In sum, the various RRBI that people with ASD present can negatively influence development and may interfere with daily functioning along their lifespan. A child's absorption in RRBI can limit participation in typical growth-promoting sensory-motor and social experiences and affect the typical expansion of learning (Gal, 2011). As adults, RRBI may pose many difficulties in the process of finding and maintaining employment and may present challenges in interactions with employers and co-workers (Baldwin, Costley, & Warren, 2014; Weissman-Nitsan, Schreuer, & Gal, 2019).

Despite these various challenges, the literature also suggested their various advantages for the participation and well-being of the person with ASD. By definition, RRBI in general, and SM specifically, lack an obvious goal or function; however, there is evidence that they in fact are rewarding because they compensate for hyper- or hypo-responsivity to sensory stimuli (Gal & Dyck, 2009). Possibly, the individual invokes them as a coping mechanism to modulate levels of arousal and thus maintain homeostasis (Gal, Dyck, & Passmore, 2002). Indeed, first-hand accounts of people with ASD who have high cognitive abilities suggest that RRBI function to regulate strong emotions (e.g., anxiety or anger; Joyce, Honey, Leekam, Barrett, & Rodgers, 2017; Rodgers, Glod, Connolly, & McConachie, 2012) and as a coping mechanism to deal with unpleasant sensations or to calm the anxiety the experience of a new environment provoked (Jones et al., 2003).

High-order RRBI, such as insistence on sameness and special restricted interests, may have great advantages as well. The insistence on sameness in ASD often is associated with special abilities such as a strong sense for detail (Austin, Wareham, & Busquets, 2008; Gal, Selanikyo, Erez, & Katz, 2015). In some children on the autism

spectrum, the special abilities—if directed, guided, and expanded—could evolve into a meaningful occupation and even a career. For example, a child who obsesses about weather patterns may mature into a meteorologist as an adult; one interested in books or fascinated by dates and other numbers may find a future career working in a library (Gal, 2011). The RRBI also can serve as a means for communication and as an attention-seeking strategy. As such, there are individuals with ASD for which engagement in RRBI provides means to convey a challenge in learning or to escape a situation or boredom. Close observation of the RRBI antecedents in context may help determine which specific function they serve (Murray-Slutsky & Paris, 2005).

"Rep-Mod": Intervention Model (See Fig. 14.1)

Interventions related to RRBI usually aim to enhance the participation of the individual with ASD, whether that person is a child, adolescent, or adult. Review of the literature showed how most available interventions were developed for lower-order, rather than higher-order, RRBI (Boyd, McDonough, & Bodfish, 2012). Such interventions can use the proposed model to consider the need for decreasing the disadvantages and increasing the advantages of these behaviors for the specific individual. Our proposed model is based on two rehabilitation-related theoretical frameworks: the international classification of functioning, disability, and health (ICF; World Health Organization [WHO], 2001) and the person-environment-occupation model (PEO; Law et al., 1996). The ICF introduced two critical concepts relevant to this model: (a) disability is a result of environmental (i.e., physical, social, cultural) demands and opportunities and not only a matter of limitations in body functions and structures, and (b) there is a need to evaluate the person's participation in daily activities occupations and personal roles (WHO, 2001).

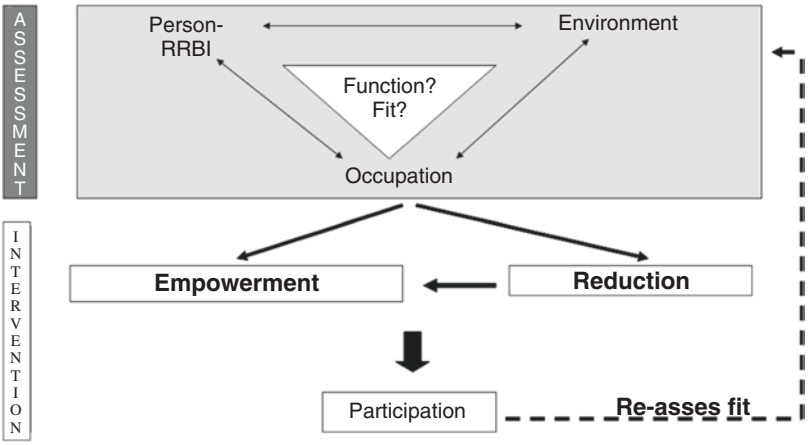

Fig. 14.1 *The "Rep-Mod" intervention model*

The PEO framework depicts occupational performance as an outcome of the fit between the person, environment, and occupation. Disability and malfunction occur when there is no good match between these three components (Law et al., 1996). In the proposed model, the assessment of RRBI involves evaluating the fit among the specific behavior, its environmental context and consequences, and the occupational demands and goals. Change in fit is also reassessed following the intervention and is considered an outcome measure of the intervention.

The working hypothesis is that there are internal and external factors that originate, maintain, and regulate RRBI (Berkson, Gutermuth, & Baranek, 1995). These factors are viewed as the underlying mechanisms that can explain the presentation of a particular RRBI. Thus, if one could decipher them, the road to addressing them would be smoother. Underlying mechanisms can be internal and/or external to the person. Internal mechanisms refer to organismic-biological aspects within the individual, such as sensory hyper-responsivity, sensory hypo-responsivity, poor communication skills, and anxiety. External factors are those aspects in the social or physical environment that fuel a behavior. Examples of external factors are an under- or over-challenging activity, an overwhelming physical setting from a sensory perspective, a stressful situation, and lack of attention from caretakers.

"Rep-Mod", the proposed intervention model, aims to decrease the RRBI that are identified as disadvantageous for the individual and to enhance/develop those identified as potential facilitators of the individual's participation in age-appropriate activities, social interactions, hobbies, or work. The model begins with assessing the target behavior by analyzing the context in which it occurs and then assessing the fit between the target behavior, the person's occupation and environment. It continues with an intervention plan for reducing and/or empowering the identified RRBI to increase the person's participation in daily occupations. After the intervention has been implemented, the fit between the person's RRBI, environment, and occupation is reassessed and the intervention is completed or new goals are established (Fig. 14.1).

Assessment Process

The assessment process includes identifying the behavior; its environment; the individual's activities, occupations, roles, and goals; and their interplay. The steps of the assessment are:

1. Identify the various RRBI that the individual with ASD performs
2. Identify which behaviors are concerns for the individual/parents and caregivers/therapists
3. Identify characteristics of the behavior(s)
4. Identify the underlying mechanisms of the behavior (e.g., sensory or social)
5. Evaluate the fit between the target behavior, the environment (i.e., social and physical), and the occupation (i.e., the occupation's demands, roles, personal goals)

These goals can be achieved by gathering detailed information about the behavior through multiple sources, such as formal assessments and interviews. Understanding what actions and responses are involved and where, with whom, and when the behavior occurs is important in guiding the search for the target behavior's underlying mechanism. To identify these mechanisms, it is recommended to conduct a functional behavioral analysis or, in the case of individuals who are cognitively able, to use in-depth interviews. Such an analysis/interview may document the events that occur before and after the behavior. In the case of verbal individuals, this process involves asking the person what function the behavior serves for them, why they perform it, and how it affects their life. When such questioning is impossible, caregivers may try to provide answers from their own point of view; thus, conducting a functional behavioral analysis is recommended. This method originates from behavioral intervention methods (Iwata, Dorsey, Slifer, Bauman, & Richman, 1982), which guide the identification of the behavior's antecedents and contingencies. For instance, it may allow one to determine the social consequences and rewards, including social attention or escape from demands, that maintain the behavior.

During the assessment, several factors may be detected as explanations for an RRBI; similarly, there are cases in which no mechanism is identified. Nonetheless, a lack of findings does not necessarily imply that this behavior has no explanatory factor. Rather, it may indicate a limitation in the assessment tool, particularly when more typical forms of behavioral rewards, such as primary and social rewards, are not driving the behavior. Example underlying mechanisms of an RRBI may include communication, attention-seeking, sensory-motor imbalance such as sensory hyper- or hypo-reactivity, or the need to balance anxiety and gain a sense of control (Murray-Slutsky & Paris, 2005).

The fit aspect of the assessment focuses on the match between the particular RRBI addressed and the person's environment and occupation. It aims to define how the target behavior affects the person within the specific environment and if it negatively affects the occupation or, rather, has the potential to positively affect or integrate within the occupation or even develop into an occupation. That is, specific behaviors may interfere with participation in one environment but be fully appropriate in another; they may decrease abilities in one occupation but may be functional in another. When there is a poor fit between the RRBI and the person's environment and occupation, the intervention needs to improve the fit. Fit evaluation also allows for weighing the costs versus benefits of the behavior in terms of the behavior's impact on the person's and their caregivers' safety, health, social-emotional state, social participation, and occupational performance.

Intervention Methods

The proposed intervention strategies integrate principles from sensory-based interventions as well as behavioral-based methods. This integration is necessary to address the behaviors, regardless of whether they have an identified mechanism or

not. Such a combination of methods will assist in addressing both the factors that originate from a behavior and those that maintain it, in order to address non-adaptive behaviors. Intervention strategies can be divided into empowering and reducing strategies. Empowering strategies are those that integrate the behavior or its motivating factors within an adaptive activity/occupation. When this is not possible, for instance, due to the behavior's injurious nature, the professional starts by limiting/reducing the behavior to a certain context (e.g., time or place) using behavioral techniques. Alongside reduction efforts, the professional may empower the individual by finding a substitutive adaptive behavior that is a good match for the individual's life.

Empowering Methods

Empowering methods are those that aim to put an RRBI to productive use for the individual through expansion and through its use in functional social-adaptive tasks. Two types of empowering methods, namely *transformation* of a narrow interest to a wider/functional interest and the use of an RRBI as a *reinforcement* are outlined.

Transformation

Expansion of an interest to enhance its functionality or adaptiveness may serve as empowerment. A narrow interest may be transformed into (a) a wider interest, (b) a functional interest, (c) competitive work, or (d) a way to deal with an ASD-related challenge such as expanding social interactions. For example, a person with ASD who throughout their lifespan was fascinated by trucks (e.g., played repetitive games with toy trucks as a child and had an antique truck collection as a teenager) may be guided as an adult to become a truck driver. A person who was attracted to visual shapes as a child and excelled in visual-perception tasks as a teenager may, as an adult, be directed towards work at a quality assurance company. A teenager with ASD who taught herself to play guitar and spends most of her free time in her room playing alone may be encouraged to play with other people with or without ASD a few times a week, using her hobby as a way to create social relationships based on shared special interests.

Reinforcement

The stereotyped behaviors and restricted and repetitive areas of interests of persons with ASD often are perceived as interfering with their ability to take on an integral, active role in daily occupations, such as: self-care, play, learning, vocation, and socializing. Although these behaviors may not fit social norms and expectations, it is important to consider their empowering potential. For instance, these behaviors may serve as motivating, calming, and organizing factors for people with ASD. The

behaviors' characteristics (e.g., type, duration, frequency, and intensity) and the consequences for taking part in daily activities are unique to each individual and require an individualized and creative analysis of their functional potential. Therefore, RRBI such as repetitive manipulation of objects or spending excessive time locked in restricted interests may be used to reinforce participation in a functional activity the person would otherwise avoid. As such, the behavior needs to be associated with a specific task and restricted by time, place, and duration. For example, a child with low functioning abilities who likes repetitive sand play may be permitted to play with sand for 5 minutes after completing a task that meets set goals, such as assisting with activities of daily living. Likewise, a cognitively able child with ASD whose special interest is to read phone books may be permitted to be involved with the activity for 10 minutes following participation in a math lesson.

Reduction Methods

"Reduction" methods apply to behaviors that professionals aim to reduce due to the behaviors' interference with the individual's participation or their negative impact on the person's health. Within the *reduction*, professionals also can aim to *add*. For example, if a child is a picky eater and it affects their health and ability to participate in social and family meals (see section "Case Study 2"), then the aim would be to reduce the selective eating by adding foods to the child's diet. In addition, "reduction" may focus on decreasing an inappropriate behavior while introducing a more functional alternative. To illustrate, people who like to rock in a sitting position—a behavior that distracts their family members—may be guided to rock in a rocking chair, a behavior structured to occur in a specific place and perceived to be more adaptive.

Reassessment

Participation in daily tasks is the goal of both the enhancement and the reduction techniques. In the reassessment stage, the clinician evaluates how enhancing or reducing a behavior affected participation in daily tasks and whether it directly or indirectly related to the RRBI goal. For example, a child who used to spend 80% of their spare time in water play but reduced it to 10% following the intervention plan may respond to the enforced change by spending most of the time crying and avoiding participation in any activity. Alternatively, the child may start to engage in classroom activities previously avoided. In the reassessment, the therapist is encouraged to observe the child's response to inform decisions on whether to cease, continue, or change the intervention. Reassessment also should address the current RRBI the person with ASD performs and their post-intervention fit with the environment and occupation. Which RRBI negatively affect participation? Which benefit the child and in which situations? Which have the potential to facilitate participation following further intervention?

The timeframe of the reassessment phase depends on the set goal. For example, a goal may include stages and appropriate timepoints for reassessment. A target window for reducing repetitive water-play behavior could be a month, whereas acquiring a profession that aligns with special interests may take several months or years.

This model presents practical ways for using RRBI, such as SM and intense special interests, in a manner that enhances the individual's participation in life activities. The application of the intervention model stages is demonstrated through the following four case studies from different age groups and at various levels of the autism spectrum.

Case Studies

Case Study 1

Assessment

A. ***Person.*** John, male, 8 years old, ASD level 3, almost non-verbal: at assessment, he had about 30 spoken words but did not construct sentences. He resided at a "home" with five other children with special needs and four staff members.
   RRBI: SM: rocking, hand flapping
   Repetitive manipulation of objects: sand play, water play, chewing shirts
   Special interests: attracted to letters
B. ***Occupation.*** Student
C. ***Environment.*** Special education school; had an individual educational work plan; received occupational therapy and speech therapy
D. ***Fit/implication.*** Staff members were not concerned with the social implications of rocking and hand flapping because most children in John's classroom performed them and the behaviors did not seem to disturb his participation in school activities.
   Major concern. *Water play*: John had a constant urge to open the tap and play with running water. He spent 70% to 80% of his time next to the sink playing with water. He cried and threw tantrums when prevented from engaging in water play.
   Other concerns: wasting water; avoiding participation in class activities; skin health.

Intervention

A. **Reduction**

   Goal: To reduce/limit water play
   ***Activity*:** Signs on the wall in front of the tap read "Yes" (green) or "No" (red). The sign was normally on the red circle and was turned to green at specific times and for specific durations:

(a) 10 minutes upon arrival to class in the morning

(b) 10 minutes after breakfast

(c) 10 minutes after lunch

B. **Empowerment**

Goal 1: Transform water play into a meaningful and functional task
***Activity*.** John assists in washing his classmates' breakfast dishes.
Goal 2: Use water play for learning, such as to read words rather than letters only
Activity. Written words "yes" and "no" represented time for water play. At first, the words were used together with the green (yes) and red (no) colors; after 2 weeks, the colored background was removed, and only the words represented whether there was permission to engage in water play.

Participation/Reassessment

In the first week of the intervention, John cried whenever he was prevented from water play. After a month, he played with water only when the sign was turned to "yes." He still asked for the "yes" sign and pointed to it but, when denied, left it and participated in other activities. Within 2 weeks, John could read the words "yes" and "no" without their distinctive backgrounds or clues and he played with water only during the permitted times.

Case Study 2

Assessment

A. ***Person*.** Brett, male, 11 years old, ASD level 2, verbal; lived with his parents and two older, typically developing siblings
RRBI. SM: repetitive pacing
Special interests: Photographs, dates, numbers, photos of school trips; preferred to spend most of his time looking at pictures from school trips; memorized correctly every event and all dates of trips since he started school
B. ***Occupation*.** Student
C. ***Environment*.** Special education classroom within a general education school; had an individual work plan; received occupational therapy (once a week) and art therapy (once a week).
D. ***Fit/implication*.** Brett's repetitive pacing was not a major concern because he had learned to limit it to recess time.
Major concern. His teacher was concerned with his restricted interests because he insisted on spending most of his time looking at school-trip pictures in the class photo album and avoided participation in school activities.

Intervention

A. **Reduction**

Goal: Decrease time spent sitting in the corner alone with the class picture book
Activity. Create a constructed schedule that limited this activity to very specific times—three independent blocks of 10 minutes per day and three additional times depending on the schoolwork schedule (reinforcement).
Activity. Create a visual, pictorial daily schedule that includes these times.

B. **Empowerment**

Goal 1. To extend Brett's interest to a meaningful activity
***Activity*.** Brett took part in a photography course, allowing him to become the "official" photographer at school and during fieldtrips.
Goal 2. Transform special interests and abilities into a meaningful occupation
Activity. Brett received a job as an assistant to the school's librarian; he coded and numbered books and created the archives of the fieldtrips.

Participation/Reassessment

Brett loved the photography course and, during 6 months following the goal setting, developed his hobby as a photographer. On the last fieldtrip, he was given responsibility for photography, and he created a section for school-trip photos on the school's website. He also took photos at family events and in everyday life. The librarian appreciated his work, and library work became a part of his schedule. He had a potential hour in the library at the end of each school day, which also was used as a reinforcement for participating in the other school lessons and tasks. However, Brett continued pacing, which presented an issue at home. Therefore, limiting pacing at home was defined as a goal to be addressed in the reassessment and future intervention plan.

Case Study 3

Assessment

A. ***Person.*** Mary, female, 14 years old, ASD level 1, verbal, lived with her parents and two brothers.
RRBI. Extreme sensory hyper-responsivity, especially in taste and smell senses; Mary was a very picky eater, eating only 20 foods (mostly carbohydrates) and avoiding smooth textures; she ate only her mother's food and only from a specific plate.
Special interest. Guitar playing; preferred to play guitar alone in every free minute

B. ***Occupation.*** Student
C. ***Environment***
Home. Mary had two younger brothers and parents who loved to travel overseas with their children. At home, she spent many hours playing the electric guitar (at which she excelled). She had a good relationship with her parents and brothers.
School. Mary studied at a regular middle school. She was a good student and did not require special academic support at school. She had no social life at or after school and refused to go on school excursions and trips due to food selectivity. Mary received a psychotherapy session once a week and an occupational therapy session once a month.
D. ***Fit/implication.*** Mary's schoolteacher was not concerned because she studied well. Although Mary was isolated during recess, it was not perceived as problematic at school because she spent that free time playing guitar in the music room. Her parents and brothers felt very limited because her food selectivity caused her extreme stress in restaurants and affected the family's overseas trips. Her mother was tired of being responsible for all of Mary's meals, and Mary was frustrated by her social isolation. She wanted to have friends but could not seem to create relationships with other teens.

Intervention

A. **Reduction**

Goal: To reduce food selectivity (to increase the variety of foods eaten)
***Activity*:** Taste at least one new food per week.
Principles for intervention:
1. *Graduated exposure (foods)*. Start with foods that are similar to old foods in color, texture, or taste.

2. *Behavioral*. Reinforce each new tasted food with a favorite "old" food.

3. *Graduated exposure (dishes)*. Present preferred foods in a different dish gradually, starting with once a week, going on to a different dish every meal.

B. **Empowerment**

Goal: Expand guitar play so that Mary plays within a group and in front of an audience
***Activity*:** Connect Mary with other music players her age.

Participation/Reassessment

Mary's occupational therapist identified a band of teens with and without ASD and encouraged her to sign up for this band. Six months later, she was a part of that band, practicing two afternoons per week and performed at schools and community

events. With the occupational therapist, an eating program was designed to address sensory issues and use behavioral rewards for exposure to new foods. Mary initially refused to eat on a different plate; it took 3 months until she agreed to try. At the time of reassessment, 6 months later, she ate from a different plate from time to time, and her diet included 10 new foods. This "opened the door" for eating away from home. The main goal that Mary set at the reassessment included eating with her family in a restaurant on their next overseas trip.

Case Study 4

Assessment

A. ***Person.*** Jeff, male, 32 years old, ASD level 1. Jeff was the youngest of four siblings and had always lived with his parents. He completed a Ph.D. in chemistry 5 years ago but had never been employed. He spent his time at home, greatly neglecting self-care.
   RRBI. Sensory hyper-responsivity: Jeff was extremely sensitive to noises and smells and reported that he suffered in the community and at any place in which he could not control the noises and smells. He also had tactile sensitivity, did not like being touched, and wore only short clothes even in winter. Because he hated the feeling of water touching his skin, he seldom showered.
   Special interest. Jeff had been fascinated by wolves his entire life. In the past 3 years, he extended his special interests. According to his report during the assessment, he had 23 "special interests," including chemistry, mathematics, cats, bicycles, visual shapes, electronics, and anthropology.
   Insistence on sameness: Jeff was a "computer freak." He spent most of his time in front of his computer searching for information regarding his special interests.
B. ***Occupation.*** None
C. ***Environment.*** Home, lived with his parents; seldom socialized
D. ***Fit/implication.*** Jeff initially reported that he was happy with his life. According to his parents, he felt fascinated by researching and studying his many interests with the internet providing him an endless source of information. However, his parents reported that they would like him to move out of the house, find an apartment and a job. They were concerned that despite his being bright and having a Ph.D., he would not be accepted to any job due to his poor social skills, poor hygiene, and severe sensory issues.

Intervention

A. **Reduction**

   Goal 1: Participate in environments that present auditory and smell challenges
   ***Activity 1*:** Complete an auditory and smell desensitization program

***Activity 2*:** Put on earplugs or earphones when in crowded and noisy places
Goal 2: Improve hygiene by dealing with tactile sensitivity (according to the model-reducing sensitivity)
***Activity 1*:** Discuss the importance of hygiene and its effect on communication in general and specifically at work
***Activity 2*:** Identify compensations for participation such as pursuing the option of taking a bath rather than showers
***Activity 3*:** Create a new daily routine that included grooming in a very detailed way

B. **Empowerment**

Goal: Expand interest in one specific area that is relevant for a job
***Activity*:** Identify a job of interest; apply to a community program that places people with ASD in jobs; prepare for the job by concentrating on related knowledge

Participation/Reassessment

Six months later, Jeff started to attend a special vocational program for adults with ASD. He went through job interviews for nonprofessional jobs but was rejected due to inappropriate dressing and body odor. A year later, in a group therapy within the vocational program, Jeff confronted the rejection in job interviews and its reasons. He agreed to go through a grooming program and even took a short course in dress codes. He later attended a quality assurance course and started to work 20 hours per week through a community program. He still lived at home with his parents. In the reassessment 2 years after the original assessment, Jeff was ready to explore new residence options.

Summary

Being a spectrum, the heterogeneity within autism is not surprising. Each person with ASD presents a different profile of strengths, difficulties, motivations, and support needs. There is great need for intervention programs that address the individual RRBI of people with ASD across the lifespan. “Rep-Mod,” the intervention model presented in this chapter, offers a roadmap for building an intervention program for these RRBI. It is unique in considering the benefits these behaviors hold for the person with ASD while cautiously addressing personal needs and preferences. This model builds upon the strong intrinsic motivation for performing RRBI in people with ASD and the implications of some of these behaviors and the skill-building potential of others. Careful assessment of the rewarding nature of RRBI, as well as the cognitive, social, emotional, and/or perceptual functions they serve, is warranted.

The intervention begins with assessing the underlying functions of the behavior and the fit between the person and their relevant environments and occupations. To enhance fit, the model suggests a series of reducing and/or empowering strategies. Its goal is to ultimately build the person's capacity to adaptively utilize their RRBI. Although this is not possible for all forms of behavior, we hope this model will lead to a better recognition of the natural tendencies of the individual, thus offering a strong starting point to promote the learning and engagement of people with ASD along the lifespan.

References

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders (DSM-5)*. Arlington, VA: American Psychiatric.

Attwood, T. (2003). Understanding and managing circumscribed interests. In M. Prior (Ed.), *Learning and behavior problems in Asperger syndrome* (pp. 126–147). New York, NY: Guilford Press.

Austin, R. D., Wareham, J., & Busquets, J. (2008). *Specialisterne: Sense and details.* Harvard Business School Case No. 608–109. Boston, MA: Harvard Business School.

Bagby, M. S., Dickie, V. A., & Baranek, G. T. (2012). How sensory experiences of children with and without autism affect family occupations. *American Journal of Occupational Therapy, 66*, 78–86. https://doi.org/10.5014/ajot.2012.000604.

Baldwin, S., Costley, D., & Warren, A. (2014). Employment activities and experiences of adults with high-functioning autism and Asperger's disorder. *Journal of Autism and Developmental Disorders, 44*, 2440–2449. https://doi.org/10.1007/s10803-014-2112-z.

Beckman, P. J. (1983). Influence of selected child characteristics on stress in families of handicapped infants. *American Journal of Mental Deficiency, 88*, 150–156.

Ben-Sasson, A., Soto, T. W., Martínez-Pedraza, F., & Carter, A. S. (2013). Early sensory overresponsivity in toddlers with autism spectrum disorders as a predictor of family impairment and parenting stress. *Journal of Child Psychology and Psychiatry, 54*, 846–853. https://doi.org/10.1111/jcpp.12035.

Berkson, G., Gutermuth, L., & Baranek, G. (1995). Relative prevalence and relations among stereotyped and similar behaviors. *American Journal on Mental Retardation, 100*(2), 137–145.

Bodfish, J. W., Symons, F. J., Parker, D. E., & Lewis, M. H. (2000). Varieties of repetitive behavior in autism: Comparisons to mental retardation. *Journal of Autism and Developmental Disorders, 30*, 237–243. https://doi.org/10.1023/A:1005596502855.

Boyd, B. A., McDonough, S. G., & Bodfish, J. W. (2012). Evidence-based behavioral interventions for repetitive behaviors in autism. *Journal of Autism and Developmental Disorders, 42*, 1236–1248. https://doi.org/10.1007/s10803-011-1284-z.

Chamak, B., Bonniau, B., Jaunay, E., & Cohen, D. (2008). What can we learn about autism from autistic persons? *Psychotherapy and Psychosomatics, 77*(5), 271–279. https://doi.org/10.1159/000140086.

Cuccaro, M. L., Shao, Y., Grubber, J., Slifer, M., Wolpert, C. M., Donnelly, S. L., et al. (2003). Factor analysis of restricted and repetitive behaviors in autism using the Autism Diagnostic Interview-R. *Child Psychiatry and Human Development, 34*, 3–17. https://doi.org/10.1023/A:1025321707947.

Cunningham, A. B., & Schreibman, L. (2008). Stereotypy in autism: The importance of function. *Research in Autism Spectrum Disorders, 2*, 469–479. https://doi.org/10.1016/j.rasd.2007.09.006.

Esbensen, A. J., Seltzer, M. M., Lam, K. S., & Bodfish, J. W. (2009). Age-related differences in restricted repetitive behaviors in autism spectrum disorders. *Journal of Autism and Developmental Disorders, 39*, 57–66. https://doi.org/10.1007/s10803-008-0599-x.

Eyman, R. K., Borthwick, S. A., & Miller, C. (1981). Trends in maladaptive behavior of mentally retarded persons placed in community and institutional settings. *American Journal of Mental Deficiency, 85*, 473–477.

Gal, E. (2011). Nosology and theories of repetitive and restricted behaviours and interests. In J. L. Matson & P. Sturmey (Eds.), *International handbook of autism and pervasive developmental disorders* (pp. 115–125). New York, NY: Springer.

Gal, E., & Dyck, M. (2009). Stereotyped movements among children who are visually impaired or blind. *Journal of Visual Impairment and Blindness, 103*, 754–765. https://doi.org/10.1177/2F0145482X0910301105.

Gal, E., Dyck, M., & Passmore, A. (2002). Sensory differences and stereotyped movements in children with autism. *Behaviour Change, 19*, 207–219. https://doi.org/10.1375/bech.19.4.207.

Gal, E., Selanikyo, E., Erez, A. B. H., & Katz, N. (2015). Integration in the vocational world: How does it affect quality of life and subjective well-being of young adults with ASD. *International Journal of Environmental Research and Public Health, 12*, 10820–10832. https://doi.org/10.3390/ijerph120910820.

Harrop, C., McBee, M., & Boyd, B. A. (2016). How are child restricted and repetitive behaviors associated with caregiver stress over time? A parallel process multilevel growth model. *Journal of Autism and Developmental Disorders, 46*, 1773–1783. https://doi.org/10.1007/s1080.

Hendricks, D. (2010). Employment and adults with autism spectrum disorders: Challenges and strategies for success. *Journal of Vocational Rehabilitation, 32*, 125–134. https://doi.org/10.3233/JVR-2010-0502.

Iwata, B. A., Dorsey, M., Slifer, K., Bauman, K., & Richman, G. (1982). Toward a functional analysis of self-injury. *Analysis and Intervention in Developmental Disabilities, 2*, 3–20. https://doi.org/10.1901/jaba.1994.27-197.

Jones, R. P., Quigney, C., & Huws, J. C. (2003). First-hand accounts of sensory perceptual experiences in autism: A qualitative analysis. *Journal of Intellectual and Developmental Disability, 28*, 112–121. https://doi.org/10.1080/1366825031000147058.

Joosten, A. V., & Bundy, A. C. (2010). Sensory processing and stereotypical and repetitive behaviour in children with autism and intellectual disability. *Australian Occupational Therapy Journal, 57*, 366–372. https://doi.org/10.1111/j.1440-1630.2009.00835.x.

Joyce, C., Honey, E., Leekam, S. R., Barrett, S. L., & Rodgers, J. (2017). Anxiety, intolerance of uncertainty and restricted and repetitive behaviour: Insights directly from young people with ASD. *Journal of Autism and Developmental Disorders, 47*(12), 3789–3802. https://doi.org/10.1007/s10803-017-3027-2.

Klin, A., Danovitch, J. H., Merz, A. B., & Volkmar, F. R. (2007). Circumscribed interests in higher functioning individuals with autism spectrum disorders: An exploratory study. *Research and Practice for Persons with Severe Disabilities, 32*, 89–100. https://doi.org/10.2511/rpsd.32.2.89.

Law, M., Cooper, B., Strong, S., Stewart, D., Rigby, P., & Letts, L. (1996). The person-environment-occupation model: A transactive approach to occupational performance. *Canadian Journal of Occupational Therapy, 63*, 9–23. https://doi.org/10.1177/2F000841749606300103.

Leekam, S. R., Prior, M. R., & Uljarevic, M. (2011). Restricted and repetitive behaviors in autism spectrum disorders: A review of research in the last decade. *Psychological Bulletin, 137*, 562–593. https://doi.org/10.1037/a0023341.

Murray-Slutsky, C., & Paris, B. A. (2005). *Is it sensory or is it behavior? Behavior problem identification, assessment, and intervention*. Austin, TX: Hammill Institute on Disabilities.

Richler, J., Bishop, S. L., Kleinke, J. R., & Lord, C. (2007). Restricted and repetitive behaviors in young children with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 37*, 73–85. https://doi.org/10.1007/s10803-006-0332-6.

Rincover, A., Cook, R., Peoples, A., & Packard, D. (1979). Sensory extinction and sensory reinforcement principles for programming multiple adaptive behavior change. *Journal of Applied Behavior Analysis, 12*, 221–233. https://doi.org/10.1901/jaba.1979.12-221.

Rodgers, J., Glod, M., Connolly, B., & McConachie, H. (2012). The relationship between anxiety and repetitive behaviours in autism spectrum disorder. *Journal of Autism and Developmental Disorders, 42*(11), 2404–2409. https://doi.org/10.1007/s10803-012-1531-y.

Shattuck, P. T., Seltzer, M. M., Greenberg, J. S., Orsmond, G. I., Bolt, D., Kring, S., Lounds, J., & Lord, C. (2007). Change in autism symptoms and maladaptive behaviors in adolescents and adults with an autism spectrum disorder. *Journal of Autism and Developmental Disorders, 37*, 1735–1747. https://doi.org/10.1007/s10803-006-0307-7.

South, M., Ozonoff, S., & McMahon, W. M. (2005). Repetitive behavior profiles in Asperger syndrome and high-functioning autism. *Journal of Autism and Developmental Disorders, 35*, 145–158. https://doi.org/10.1007/s10803-004-1992-8.

Turner, M. (1999). Annotation: Repetitive behaviour in autism; a review of psychological research. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 40*, 839–849. https://doi.org/10.1111/1469-7610.00502.

Uljarević, M., Richdale, A. L., Evans, D. W., Cai, R. Y., & Leekam, S. R. (2017). Interrelationship between insistence on sameness, effortful control and anxiety in adolescents and young adults with autism spectrum disorder (ASD). *Molecular Autism, 8*, 36. https://doi.org/10.1186/s13229-017-0158-4.

Weissman-Nitsan, M., Schreuer, N., & Gal, E. (2019). Employers' perspectives regarding reasonable accommodations for employees with autism spectrum disorder. *Journal of Management & Organization, 25*, 481–498. https://doi.org/10.1017/jmo.2018.59.

Wilkes, B., & Lewis, M. (2018). The neural circuitry of restricted repetitive behavior: Magnetic resonance imaging in neurodevelopmental disorders and animal models. *Neuroscience & Biobehavioral Reviews, 92*, 152–171.

Wood, J. J., & Gadow, K. D. (2010). Exploring the nature and function of anxiety in youth with autism spectrum disorders. *Neuroscience and Biobehavioral Reviews, 92*, 152–171. https://doi.org/10.1016/j.neubiorev.2018.05.022.

World Health Organization. (2001). I*nternational classification of functioning, disability and health (ICF)*. https://apps.who.int/iris/handle/10665/42407

Yerys, B. E. (2015). An update on the neurobiology of repetitive behaviors in autism. In R. M. Hodapp & D. J. Fidler (Eds.), *International review of research in developmental disabilities* (Vol. 49, pp. 91–150). Waltham, MA: Academic Press.

Chapter 15
Restricted, Repetitive Behaviours and Interests in the Workplace: Barriers, Advantages, and an Individual Difference Approach to Autism Employment

Simon M. Bury, Darren Hedley, and Mirko Uljarević

Introduction

Employment represents an important component of human experience, a rite of passage into adulthood, the offer of financial security, and the potential to give life meaning. However, for people with ASD, it can also represent a seemingly impossible minefield of social and structural rules and requirements. It is well established that certain aspects of the traditional recruitment processes (e.g., interviews) that place a high importance on social performance can function as a barrier to obtaining employment for those with ASD. Furthermore, the broad array of unwritten social rules and conventions present challenges for employees with ASD in maintaining employment (Bury, Flower, Zulla, Nicholas, & Hedley, 2020; Mawhood & Howlin, 1999). Thus, it is widely accepted that a range of modifications to the recruitment procedures along with work-place adjustments may be required to enable individuals with ASD to more readily gain and sustain employment. However, even if the social aspects of employment are managed, diagnostic factors associated with the restricted and repetitive behaviours and interests (RRBI), such as insistence on sameness and rigid routines and behaviours, pose an additional layer of limitations and thus can present significant barriers to obtaining and maintaining employment.

Given the challenges described above, it is not surprising that individuals with ASD are significantly underrepresented in the work force, with high unemployment rates reported worldwide (Australian Bureau of Statistics, 2019; Baldwin, Costley,

S. M. Bury (✉) · D. Hedley
Olga Tennison Autism Research Centre, School of Psychology and Public Health, La Trobe University, Melbourne, VIC, Australia
e-mail: S.Bury@latrobe.edu.au

M. Uljarević
Melbourne School of Psychological Sciences, University of Melbourne, Melbourne, VIC, Australia

E. Gal, N. Yirmiya (eds.), *Repetitive and Restricted Behaviors and Interests in Autism Spectrum Disorders*, Autism and Child Psychopathology Series,
https://doi.org/10.1007/978-3-030-66445-9_15

& Warren, 2014; Holwerda, van der Klink, Groothoff, & Brouwer, 2012; Howlin, Goode, Hutton, & Rutter, 2004; Shattuck et al., 2012), leading to a world-wide "call to action" by the United Nations (Ki-moon, 2015). In Australia, only 27.3% of individuals with ASD are employed, which is lower than people without disabilities (80.3%), and also lower than all disabilities groups combined (47.8%; Australian Bureau of Statistics, 2019). Similarly, for young adults in the US, fewer individuals with ASD reported working outside the home (58%) than young adults with emotional, speech, or learning disabilities (90%), and those with intellectual disability (74%; Roux, Shattuck, Rast, Rava, & Anderson, 2015). Furthermore, when individuals with ASD are employed, they are more likely to be underemployed, or working in positions that are unsuited or misaligned with their skills and vocational education (Hedley et al., 2017; Shattuck et al., 2012).

As employment in the general population is consistently linked to better mental health and well-being outcomes (Backhans & Hemmingsson, 2012; Feather & O'Brien, 1986; Heinz et al., 2018; McKee-Ryan, Song, Wanberg, & Kinicki, 2005; Voßemer et al., 2018; Wanberg, 2012), especially when associated with greater job satisfaction (Faragher, Cass, & Cooper, 2005), employment represents an important avenue for improved well-being outcomes in individuals with ASD. While some research has shown well-being to remain relatively stable over the initial 12 months of employment for individuals with ASD (Hedley, Uljarević, Bury, & Dissanayake, 2019), other research has shown employment amongst this population to be linked with greater well-being outcomes (e.g., self-esteem, sense of purpose, financial independence; Hedley et al., 2018) and aspects of quality of life (Gal, Landes, & Katz, 2015), highlighting the importance of improving employment prospects amongst individuals with ASD.

There have been numerous efforts aimed at reducing the under-representation of individuals with ASD in the workplace, with a range of support programs developed to assist individuals with ASD in obtaining and maintaining employment showing somewhat encouraging results (Flower, Hedley, Spoor, & Dissanayake, 2019; Hedley, Uljarević, Cameron, et al., 2017; Hedley, Uljarević, & Hedley, 2017). Importantly, parallel streams of emerging research have started to identify factors that (i) present challenges to maintaining employment and can therefore be addressed through the provision of targeted support or by modifying human resource processes that otherwise can present a barrier to people with ASD (Hedley et al., 2018), and (ii) can serve as enablers, strengths, and areas of expertise, and therefore counteract negative perceptions surrounding employment expectations and outcomes (Holwerda et al., 2012). A recent movement within the media (Cook, 2012), non-government organisations (United Nations Regional Information Centre for Western Europe, 2015), and academia (Austin & Pisano, 2017), has focused on promoting the second group of factors highlighted above. These factors have been further argued to bring a competitive advantage to the employment space (c.f., Bury, Hedley, Uljarević, Dissanayake, & Gal, 2019).

The nature of the 'autism advantage', as it is often termed, is typically centred on the restrictive and repetitive behaviours and interests (RRBI) diagnostic criteria for

ASD (American Psychiatric Association, 2013).[1] Strengths thought to be somewhat unique to the autism profile, and associated with RRBI, include increased tolerance for, or liking of repetitive tasks, and greater attention to detail. This chapter provides critical appraisal of the existing evidence regarding RRBI as they pertain to talent, firstly focusing on the research conducted on the autism advantage more generally, followed by a review of the evidence supporting the 'autism advantage' in employment. We then provide a discussion of the benefits of adopting an individualised approach to the identification of both strengths and support needs for people with ASD in the workplace.

RBBIs in Adulthood: Potential for Barrier and Strength?

Together with social interaction and communication deficits, RRBI represent the core diagnostic features of autism (American Psychiatric Association, 2013). While longitudinal and cross-sectional research findings indicate that RRBI are somewhat less severe among older than younger individuals with ASD (Esbensen, Seltzer, Lam, & Bodfish, 2009; Shattuck et al., 2007), they are still present across adolescence and adulthood, and may therefore exert negative influence on educational and employment outcomes.

RRBI represent a heterogeneous group of behaviours that are characterised by repetition, rigidity, and invariance, that may often be situationally inappropriate (Bodfish, Symons, Parker, & Lewis, 2000; Leekam, Prior, & Uljarevic, 2011). Regardless of their presentation complexity, a range of factor analytic studies have consistently identified that RRBI can be best described by the following two overarching domains: Repetitive Sensory Motor Behaviours (RSMB) and Insistence on Sameness (IS; Barrett et al., 2015; Georgiades, Papageorgiou, & Anagnostou, 2010; Honey, McConachie, Randle, Shearer, & Le Couteur, 2008; Lam, Bodfish, & Piven, 2008; Lidstone et al., 2014), with some research providing evidence for the Circumscribed Interests as an additional domain (Honey et al., 2008; Lam et al., 2008). This factor structure appears to be invariant across age and gender.

It is important to note that these behaviours are part of normative development as they are transient and serve particular adaptive functions (e.g., motor development and maturation; Sprague & Newell, 1996; Thelen, 1979; Wolff, 1968). However, in autism, and some other classified disorders (e.g., Obsessive-Compulsive Disorder,

[1] Autism Spectrum Disorder (ASD) is a cluster or neurodevelopmental disorders, which are characterised by impairments in social interaction, communication and restricted and repetitive behaviours and interests (DSM-5; American Psychiatric Association, 2013). Heterogeneity not only characterises the presentation of autism traits across the spectrum, but also the level of impact autism can have on functioning, and subsequent supports. While the discussion that follows on restricted and repetitive behaviours and interests can be applicable across the spectrum, our analysis has a greater focus on strengths and supports associated with autism traits more generally, without addressing any additional support required for individuals with a co-occurring intellectual disability.

Attention Deficit Hyperactivity Disorder; Langen, Durston, Kas, van Engeland, & Staal, 2011), these behaviours persist beyond the developmental stage when they are adaptive and can exert negative influence on other aspects of development (e.g., self-regulation, learning, social development). For example, insistence on sameness has been argued to function as a form of self-regulation in normative development (Evans et al., 1997; Evans, Gray, & Leckman, 1999; Uljarević, Richdale, Evans, Cai, & Leekam, 2017), as a way to control an uncertain evironment, and by doing so reduce normative fears and anxiety. Normatively, these behaviours are replaced by more sophisticated forms of self-regulation as children develop (Evans et al., 1999). However, autism by its very nature is a developmental disorder, with delays in a range of cognitive abilities (e.g., executive functioning), that have strong links with insistence on sameness (Leekam et al., 2011; South, Ozonoff, & McMahon, 2005; Tregay, Gilmour, & Charman, 2009; Uljarević, Richdale, et al., 2017). Consequently, the ability to develop more age appropriate and flexible forms of regulation are limited, and these less developed forms of self-regulation remain relatively stable across the lifespan (Esbensen et al., 2009).

Taken together, RRBI can remain maladaptive throughout development for individuals with ASD, significantly impeding social, cognitive and emotional development; representing significant challenges both to the individuals with ASD and to those who provide them with support (Harrop, McBee, & Boyd, 2016; Leekam et al., 2011). Furthermore, while RRBI can vary in presentation, intensity, variability and severity (Gal, 2011; Wilkes & Lewis, 2018; Yerys, 2015), the limited scope of interests can negatively impact development of social repertoire and inclusion (Attwood, 2003; Cunningham & Schreibman, 2008; Klin, Danovitch, Merz, & Volkmar, 2007), and the restrictive nature and compulsion to perform RRBI can interfere with formal education and employment, thus reducing opportunities wherein more sophisticated, flexible and functional cognitive and social abilities can develop (Leekam et al., 2011).

In addition to repetitive sensory motor behaviours, insistence on sameness and circumscribed interests; an extreme or indifferent response to sensory information (e.g., aural, tactile, proprioceptive) represents the fourth domain of the RRBI diagnostic criteria. Although originally noted by Kanner (1943) in his early description of the disorder, hyper- and hypo-reactivity to sensory input has only been included as a diagnostic criterion in the most recent iteration of the DSM (DSM-5; American Psychiatric Association, 2013). The presentation of sensory concerns is marked by significant variations in the presence and severity of sensory sensitivity across domains both within and between individuals (Ben-Sasson et al., 2007; Leekam, Nieto, Libby, Wing, & Gould, 2007; Uljarević et al., 2017). Although not necessarily repetitive in nature, sensory symptoms may engender RRBI directly (Gal, Dyck, & Passmore, 2010), or through anxiety (Lidstone et al., 2014), and can have significant impact on daily functioning (Smith & Sharp, 2013).

Despite the reviewed evidence that RRBI present a significant barrier to development, they can also be viewed as a benefit to the individual. However, the extent to which positive aspects are beneficial in one area must be weighed against support needs in other areas. For example, while some individuals with ASD and their family members view circumscribed interests as a positive aspect of the condition,

associating it with well-being, personal validation, and a motivation for personal growth (Mercier, Mottron, & Belleville, 2000), circumscribed interests may also restrict social interaction, involvement in other activities, difficulties switching to other activities, and require a higher amount of accommodation from those that support them (Turner-Brown, Lam, Holtzclaw, Dichter, & Bodfish, 2011). Additionally, some people with ASD and their family members believe RRBI function to regulate strong emotions (e.g., anxiety, anger; Joyce, Honey, Leekam, Barrett, & Rodgers, 2017; Rodgers, Glod, Connolly, & McConachie, 2012); however, as noted above, although such self-regulatory behaviours may be helpful in the short-term, they may also limit the opportunity to develop more adaptive behaviours, and consequently reinforce anxiety (Uljarević, Richdale, et al., 2017).

Beyond well-being and self-regulation, some have argued that the non-social aspects of the autism diagnosis (i.e., RRBI) can lead to talent and superior performance in various cognitive areas (e.g., attention to detail) when compared to people without autism (Baron-Cohen, Ashwin, Ashwin, Tavassoli, & Chakrabarti, 2009). Evidence from experimental studies in support of this position have shown superior autism performance in areas such as enhanced visual search abilities (Kaldy, Giserman, Carter, & Blaser, 2016; O'Riordan, Plaisted, Driver, & Baron-Cohen, 2001; Plaisted, O'Riordan, & Baron-Cohen, 1998), greater performance on hidden figures tasks (Shah & Frith, 1983), or abilities recognising patterns (Stevenson & Gernsbacher, 2013). While Happé and Frith (2006) attribute superior performance to a bias in information processing, Baron-Cohen et al. (2009) suggest it represents an advantageous 'style of thinking' unique to autism. Known as 'hyper-systemising', this style of thinking, which includes a search for rules and consistency, is suggested to underlie talent in "recognizing repeating patterns in stimuli" (p. 1377). Baron-Cohen and colleagues propose that superior performance in attention to detail stems from hypersensitivity, which leads to greater visual acuity (e.g., Ashwin, Ashwin, Rhydderch, Howells, & Baron-Cohen, 2009; but see Bach & Dakin, 2009 for an alternate interpretation).

Nonetheless, while superior attention to detail can clearly be an advantage, it is also important to consider other possible mechanisms which may underpin RRBI, and how this may affect behaviour more holistically. For example, as mentioned above, executive functioning has been suggested as an alternative mechanism that might subserve RRBI (South et al., 2005). This could mean that the qualities that allow individuals to demonstrate greater talent in attention to detail, could also lead to behaviours such as inflexibility and difficulties adjusting to change, which can be problematic in certain environments (e.g., education, employment).

Evidence summarized in this section is largely consistent with the diagnostic requirement that symptomology must cause "clinically significant impairment" (American Psychiatric Association, 2013), and in the case of RRBI, they can indeed present significant challenges to individual development and functioning in diagnosed individuals. However, despite a weaker evidence base, and as outlined above, arguments have been put forward that RRBI can present a strength, potentially leading to superior performance on tasks that could be beneficial or advantageous in the workplace. The next section examines the interaction between RRBI and the workplace environment, in order to better assess the evidence for an 'autism advantage'.

RBBIs in the Workplace

Looking for and starting work brings with it a varied set of social challenges, from job-interviews which place great importance on personality and interpersonal interactions, to joining a new social structure with new or different unwritten social rules, conventions and hierarchy. Managing these employment- related social challenges can present a significant barrier to people with ASD (Bury, Flower et al., 2020; Mawhood & Howlin, 1999; Müller, Schuler, Burton, & Yates, 2003). Beyond the social aspects of employment, the structure, processes and physical environment of a workplace can also present barriers to maintaining employment and managing workloads, particularly regarding aspects of RRBI (e.g., insistence on sameness, rigid routines and rituals). Work in and of itself requires a significant change to one's daily routine and lifestyle, which people with ASD may find challenging. Once employed, research shows that employees with ASD may experience difficulties managing changes to work settings or tasks, adjusting to set routines and rules, and moving from specifics to a more general scope of work tasks (Hillier et al., 2007; Mawhood & Howlin, 1999; Müller et al., 2003). In addition to the structured nature of modern workplaces, the interaction between the physical work environment and sensory sensitivities (e.g., fluorescent lights, open work environment, noise; Kirchner & Dziobek, 2014) can impact on individual stress levels, job satisfaction, and performance (Krieger, Kinébanian, Prodinger, & Heigl, 2012; Pfeiffer, Braun, Kinnealey, Derstine Matczak, & Polatajko, 2017).

In order to manage and support the challenges individuals with ASD face in the workplace, vocational placement and employment programs have utilised various measures such as job coaches, job search assistance, assessment and placement, as well as on the job training and accommodations (Flower et al., 2019; Hedley, Uljarević, Cameron, et al., 2017; Hedley, Uljarević, & Hedley, 2017; Hillier et al., 2007). In evaluating the efficacy of such programs, researchers tend to focus on the broader measures of success (e.g., actual employment, improved wages and work-hours), with such programs reporting some success in supporting individuals with ASD to achieve many of these goals (Hedley, Uljarević, Cameron, et al., 2017). Although this research does not include focused measures of job performance within these positions, particularly in regards to RRBI, descriptions of the job roles are often formulated in a way that highlight particular skills (e.g., "repetitive tasks that require a high attention to detail and an intensive focus"; Wehman et al., 2014, p. 496). In a similar way, in describing admirable qualities of their employees with ASD, employers report positive qualities that could fit under the RRBI umbrella including adherence to rules and attention to detail (Hedley et al., 2018; Hillier et al., 2007), which could underlie other strengths such as reliability and honesty. Thus, when appropriate supports are available, individuals with ASD can not only succeed in the workplace, but can potentially bring qualities and skills related to their autism diagnosis (i.e., RRBI) that are advantageous to employers. However, the degree to which these skills are present amongst all employees with ASD, or whether they are comparatively different from employees without autism, is not clear.

Based on clinical experience, practitioners have long suggested that attributes of the autism diagnosis may be advantageous in the work environment (Smith, Belcer, & Juhrs, 1995). For example, it has been suggested that individuals with ASD may flourish in jobs "others find unpleasant" (Van Bourgondien & Woods, 1992, p. 229). These might include jobs with limited social interaction or that are repetitive in nature. Indeed, some people with ASD reported that they prefer repetitive tasks at a higher rate than individuals without autism (Gal et al., 2015), and that they can perform these tasks more accurately and without becoming bored (Müller et al., 2003). Thus, *some* employees with ASD may be able to fill positions or be assigned work tasks that others do not enjoy, or are able to sustain, with the potential to reduce errors that arise due to lapses in concentration.

Beyond performing tasks that others find monotonous or are hard to staff, RRBI in autism have also been suggested to contribute to superior task performance above and beyond that of non-autistic peers. These claims draw from the experimental research described above, highlighting superior performance in hyper-systemised thinking and attention to detail (e.g., visual search tasks). Indeed, such skills could prove beneficial to specific work tasks (e.g., reviewing satellite images for troop movement, software testing), which has led companies and even governments to seek out individuals with ASD where such skills are valued. Specific industries that have been targeted include the information technology sector and cyber security (Austin & Pisano, 2017). Similarly, the intensity of focus and preoccupation some individuals with ASD give to their circumscribed interests, and the extended knowledge they can develop in these areas, have led to suggestions that aligning these interests to employment could be beneficial (Attwood, 2003; Bross & Travers, 2017; Müller et al., 2003; Olney, 2000).

Circumscribed interests may also be linked to preference for mechanical and non-social aspects of the world (e.g., physical systems, understanding the functioning of machines; Baron-Cohen & Wheelwright, 1999), leading to extended knowledge in specific professional areas (e.g., mathematics; Baron-Cohen, Wheelwright, Skinner, Martin, & Clubley, 2001). However, others have found that preferences may actually be more evenly distributed between natural science or technical fields, social sciences and creative areas (Kirchner & Dziobek, 2014). Similarly, aligning savant or special skills (e.g., mathematics, music, art, memory), that are suggested to be present in between 28% to 42% of the autism population (Bennett & Heaton, 2012; Howlin, Goode, Hutton, & Rutter, 2009), could engender an 'autism advantage' in a suitable workplace that is able to capitalise on these special skills.

Although the literature described above suggests that individuals with ASD have a great potential to show strengths in the work place and, given a supportive environment, they could potentially outperform their non-autistic peers on specific work tasks, close inspection of the research itself reveals the evidence stems primarily from opinion pieces, qualitative or small studies (e.g., Austin & Pisano, 2017; Müller et al., 2003). Furthermore, research reporting superior performance in controlled laboratory settings (e.g., Kaldy et al., 2016; Stevenson & Gernsbacher, 2013) does not directly speak to actual performance within the workplace. While advocates emphasise the 'autism advantage' in employment, particularly in regards to

RRBI, specifically attention to detail, tolerance for repetitive tasks, and circumscribed interests, it is important to examine the scientific evidence supporting this stance. Below we provide a detailed overview of the empirical research on this topic.

What Evidence Is There for an 'Autism Advantage' at Work?

In order to evaluate the current state of evidence regarding the autism advantage, particularly as it relates to RRBI in the workplace, we conducted a systematic review of the available research literature (for the full PRISMA procedure please see Bury, Hedley, Uljarević, & Gal, 2020). Papers included in the review were empirical peer-reviewed studies that measured work performance of people with ASD, and when possible, a non-autistic comparison group, in the workplace or when performing naturalistic approximations of work tasks. Of interest were studies in which actual work-based performance of individuals with ASD was measured. Surprisingly, only four qualitative and two quantitative studies met criteria for inclusion. Careful analysis of these studies revealed four themes characterising aspects of the RRBI domain: attention to detail; tolerance for repetitive tasks; special or circumscribed interests; and other RRBI related advantages and concerns. We summarise the results of the review below.

Attention to Detail

Aspects of attention to detail were addressed in four studies, offering mixed findings. Scott et al. (2017) surveyed employers of individuals with ASD, and asked them to rate their performance (i.e., below standard, meets standard, above standard), and when possible, to also rate employees without autism who worked in similar job roles (e.g., tasks, skills, hours). Using a single item measure ("How would you describe your employee's attention to detail?"), employees with ASD were significantly more likely to be rated above standard (55%) on attention to detail than their colleagues without autism (19%). In a qualitative study conducted with nine individuals with ASD, two of Smith and Sharp's (2013) participants believed their sensory sensitivity led to greater attention to detail and workplace performance. For example, one said that their hypersensitivity to taste was beneficial to developing a more refined palate as a chef. Similarly, while not focused on employment, attention to detail was perceived to be an advantage at work (i.e., shop assistant, gardener) by employed participants in Russell et al.'s (2019) qualitative study investigating the broader concept of 'autistic advantage'. Participants in this study also perceived that these advantages could also present challenges in certain contexts, for example when under time constraints. In one of the few experimental studies on the topic, Gonzalez, Martin, Minshew, and Behrmann (2013) used a naturalistic baggage search task to assess performance advantage associated with visual

search (i.e., attention to detail). Participants included individuals with and without autism, who were required to scan x-rays of luggage and identify contraband or reject bags without contraband. Although the predicted superiority in visual search of individuals with ASD versus those without was not immediately apparent, with no difference between the correct identification and rejection of contraband in the first 160 trials, in the second set of 160 trials, the performance of participants with ASD in correctly rejecting bags without contraband significantly improved, while the performance of the group without autism decreased, although the difference did not reach statistical significance. Taken together, while there was some evidence of greater attention to detail as reported by employers of individuals with ASD, and employees with ASD themselves, objective analysis of visual search skills failed to identify a clear advantage in this area.

Tolerance for Repetitive Tasks

Individuals with ASD are often suggested to prefer and perform well at repetitive tasks. Although no study directly tested tolerance for repetitive tasks, Gonzalez et al. (2013) may have inadvertently demonstrated tolerance for repetition in their participants with ASD. In their study, the performance of non-autistic participants on one of the highly repetitive tasks deteriorated over time. However, participants with ASD improved in their level of performance in the second set of trials, suggesting potentially higher tolerance for repetitive tasks, at least over the trial period. In two qualitative studies (Krieger et al., 2012; Pfeiffer et al., 2017), participants reported that their work performance improved when work was clearly structured and defined, and that they benefitted from having consistent schedules and routines. However, other participants indicated that too much structure can lead to increased stress, thereby detrimentally affecting performance (Pfeiffer et al., 2017).

Special/Circumscribed Interests

Similarly, no study included a direct investigation of whether alignment of special interests with employment would lead to superior work performance. However, in the qualitative study by Pfeiffer et al. (2017), participants with ASD reported that their job satisfaction and performance improved when their employment matched their special skills and interests. Furthermore, one participant stated that the alignment of interest to employment led to early career success as a computer scientist; however, others were found to take a more circuitous path to career success that was not as evidently linked to the alignment of their special interests and their career (Krieger et al., 2012).

Other RRBI Related Advantages and Concerns

Reviewed studies also highlighted challenges and obstacles created by RRBI. For example, sensory hypersensitivity (e.g., fluorescent lights, noise, movement, temperature, unexpected human touch) was identified as a barrier impacting work performance if not addressed or managed appropriately (Krieger et al., 2012; Pfeiffer et al., 2017). Furthermore, employees with ASD in the study by Scott et al. (2017) were more likely to be rated by their employer as below standard for 'flexibility' (28%) when compared to their colleagues without autism (8%). However, possibly reflecting greater rigid adherence to rules, employees with ASD were more likely to be rated as above standard for "work ethic", with 71% of employees with ASD rated above standard, compared to only 30% of those without autism (assessed with one item: "This employee's work ethic is best described as:").

Overall, despite some evidence of superior performance in areas associated with RRBI, due to the paucity of studies and reliance on subjective accounts, there currently is no strong empirical support or evidence either for or against the existence of an 'autism advantage' in the work place. More attention in defining and operationalising key variables, for example, providing a clear definition of attention to detail (or work ethic) as well as increased clarity and conceptual distinction between the circumscribed interest construct and expertise is needed in order to more clearly link the hypothesised autism advantage with the empirical evidence. Furthermore, there was very little research directly comparing workplace skills and performance between employees with ASD and suitably matched non-autistic controls. It is thus difficult to establish whether reported improvements in performance were associated with individual improvements in task performance resulting from practice, or learning the task, or truly represent an 'autism advantage' associated with autistic traits and underlying cognitive mechanisms.

An Individualised Approach to Autism Strength in the Workplace

Although we identified very limited research evidence of an 'autism advantage' in the workplace, an approach to autism that highlights strengths provides some important balance to a condition more commonly characterised by its 'deficits'. Nonetheless, as we have argued previously (Bury et al., 2019), there is potential for a disconnect between how autism is discussed as a potential advantage in employment—an almost stereotyped view of autism—and how autism is discussed more generally. On the one hand, positive, strength-based accounts of autism forge the path for broader and clearer advocacy that have the potential to counteract the negative narrative around autism generally, and employment specifically (Holwerda et al., 2012). However, stereotyped accounts of autism such as those sometimes portrayed in the media, even when positive, may inadvertently pressure those with

ASD to outperform their peers despite the challenges they often face in the work environment. Indeed, Happé and Frith (2009) argue that public fascination with the savant skills of some with ASD may lead to the "dangerous consequence" of expecting these skills from *everyone* on the autism spectrum (p. 1346). Unfortunately, this warning seems to have been somewhat overlooked over the last decade. To better support people with ASD in the workplace we reiterate the call for an individual difference perspective in determining both strengths and support needs (Bury et al., 2019, Bury, Hedley, et al., 2020).

An individual differences approach to autism employment must recognise strengths associated with ASD as well as acknowledging that autism is characterised by significant heterogeneity, both in terms of autistic traits and in their severity. Although there is some evidence of superior performance in attention to detail within controlled laboratory settings (e.g., Kaldy et al., 2016; Plaisted et al., 1998; Shah & Frith, 1983; Stevenson & Gernsbacher, 2013), superior performance in association with the presence of autism is not always evident (Happé & Frith, 2006). Heterogeneity across the autism presentation, along with a multitude of other factors (many of which we have discussed here), is therefore likely to impact individual performance and is likely to be environmentally dependent. Furthermore, given the broad and imprecise diagnostic criteria which requires the contribution of only two of four RRBI domains for a formal diagnosis (American Psychiatric Association, 2013), RRBI presentation is similarly likely to vary significantly on an individual basis. Put simply, whereas some individuals may prefer work that is repetitive in nature, others may prefer a structured work environment that provides an opportunity to express their creativity, or is intellectually challenging (Müller et al., 2003). Indeed, many individuals with ASD exhibit a creative flair, expressed in outstanding works of art (although it could also be argued that attention to detail underlies the expression of artistic talent in autism; Happé & Frith, 2009; Happé & Vital, 2009). Placing someone with ASD in a repetitive role based solely on their autism diagnosis could therefore undermine these strengths, further impacting job satisfaction and ultimately, potential for success and sustained employment.

An individual differences approach acknowledges that the mechanism underlying strengths in one area of functioning could also lead to the need for greater supports in other areas (Russell et al., 2019). As mentioned above, while attention to detail or a singular focus on an area of interest could represent a significant strength in certain work tasks, these abilities could manifest due to difficulties with executive function (South et al., 2005), which can also present as common workplace challenges such as adjusting to changing routines or environments (Müller et al., 2003; Scott et al., 2017). Understanding that the superior performance in these areas may also require some additional strategies and workplace adaptations to assist with executive functioning (e.g., task switching and priority planning), could lead to greater sustainability of employment, and utilisation of individual strengths.

To better support individual strengths within the workplace it will be important to develop carefully designed, ecologically valid experimental studies that account for the different mechanisms suggested to underpin RRBI (e.g., hyper-systemising, Baron-Cohen et al., 2009; executive dysfunction and cognitive control, Demetriou

et al., 2018; anxiety and intolerance of uncertainty management, Joyce et al., 2017; altered reward processing, Kohls, Antezana, Mosner, Schultz, & Yerys, 2018; predictive coding, Pellicano & Burr, 2012; Uljarević, Richdale, et al., 2017), so as to better understand individual strengths, and identify their limitations. It is also important to develop and utilise assessment tools that generate individual work profiles designed specifically for autism (e.g., Gal et al., 2015; Gal, Meir, & Katz, 2013; Royo-León et al., 2018) that will capture individual strengths and challenges, to best identify individual strengths and abilities, and to identify work adaptations that can support them. Assessing the workplace, not only for environmental (e.g., sensory) and social challenges, but also what the workplace can offer individuals with ASD (Nicholas et al., 2019; Nicholas & Klag, 2020; Vogus & Taylor, 2018), may best assist matching individualised work profiles with the workplace.

There is no doubt that people with ASD bring unique perspectives to the workplace as a result of their diagnosis, often involving overcoming, or continuing to manage significant challenges. It is both imperative and timely to recognise the contributions of the autism community to the broader society. At the same time, it is also important to recognise the heterogeneity within autism such that, in addition to their individual strengths and perspectives, each individual brings a unique set of support needs. While stereotyped views of an 'autism advantage' may help reduce overall stigma, there is need for caution that individual differences and needs are not overlooked. We hope that this will lead to a better fit between the work environment and the individual, capitalising on their strengths and engendering their success in the workplace.

Conflict of Interest Statement Simon M Bury has received research funding from DXC Technology and ANZ Bank. Darren Hedley has received research funding from DXC Technology, the Australian Government Department of Human Services, and the Australian Government Department of Defence. Darren Hedley is currently supported by a Suicide Prevention Australia National Suicide Prevention Research Fellowship. Mirko Uljarević is currently supported by the Australian Research Council Discovery Early Career Researcher Award (DE180100632).

References

American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders (DSM-5®)*. Washington: American Psychiatric Association Publishing.

Ashwin, E., Ashwin, C., Rhydderch, D., Howells, J., & Baron-Cohen, S. (2009). Eagle-eyed visual acuity: An experimental investigation of enhanced perception in autism. *Biological Psychiatry, 65*, 17–21. https://doi.org/10.1016/j.biopsych.2008.06.012.

Attwood, T. (2003). Understanding and managing circumscribed interests. In M. Prior (Ed.), *Learning and behavior problems in asperger syndrome* (pp. 126–147). New York: The Guilford Press.

Austin, R. D., & Pisano, G. P. (2017). Neurodiversity as a competitive advantage. *Harvard Business Review, 95*, 1–9.

Australian Bureau of Statistics. (2019). 44300DO060_2018 Survey of Disability, Ageing and Carers: Autism in Australia. Retrieved from https://www.abs.gov.au/AUSSTATS/subscriber.

nsf/log?openagent&44300do060.xls&4430.0&Data%20Cubes&5C2149D4F6353996CA2584C0000C3B7F&0&2018&29.11.2019&Latest

Bach, M., & Dakin, S. C. (2009). Regarding "eagle-eyed visual acuity: An experimental investigation of enhanced perception in autism". *Biological Psychiatry, 66*, 19–20. https://doi.org/10.1016/j.biopsych.2009.02.035.

Backhans, M. C., & Hemmingsson, T. (2012). Unemployment and mental health--who is (not) affected? *European Journal of Public Health, 22*, 429–433.

Baldwin, S., Costley, D., & Warren, A. (2014). Employment activities and experiences of adults with high-functioning autism and Asperger's disorder. *Journal of Autism and Developmental Disorders, 44*, 2440–2449. https://doi.org/10.1007/s10803-014-2112-z.

Baron-Cohen, S., Ashwin, E., Ashwin, C., Tavassoli, T., & Chakrabarti, B. (2009). Talent in autism: Hyper-systemizing, hyper-attention to detail and sensory hypersensitivity. *Philosophical Transactions of the Royal Society B: Biological Sciences, 364*, 1377–1383. https://doi.org/10.1098/rstb.2008.0337.

Baron-Cohen, S., & Wheelwright, S. (1999). 'Obsessions' in children with autism or Asperger syndrome: Content analysis in terms of core domains of cognition. *The British Journal of Psychiatry, 175*, 484–490. https://doi.org/10.1192/bjp.175.5.484.

Baron-Cohen, S., Wheelwright, S., Skinner, R., Martin, J., & Clubley, E. (2001). The autism-spectrum quotient (AQ): Evidence from asperger syndrome/high-functioning autism, males and females, scientists and mathematicians. *Journal of Autism and Developmental Disorders, 31*, 5–17. https://doi.org/10.1023/A:1005653411471.

Barrett, S. L., Uljarević, M., Baker, E. K., Richdale, A. L., Jones, C. R., & Leekam, S. R. (2015). The Adult Repetitive Behaviours Questionnaire-2 (RBQ-2A): A self-report measure of restricted and repetitive behaviours. *Journal of Autism and Developmental Disorders, 45*, 3680–3692. https://doi.org/10.1007/s10803-015-2514-6.

Bennett, E., & Heaton, P. (2012). Is talent in autism spectrum disorders associated with a specific cognitive and behavioural phenotype? *Journal of Autism and Developmental Disorders, 42*, 2739–2753. https://doi.org/10.1007/s10803-012-1533-9.

Ben-Sasson, A., Cermak, S. A., Orsmond, G. I., Tager-Flusberg, H., Carter, A. S., Kadlec, M. B., & Dunn, W. (2007). Extreme sensory modulation behaviors in toddlers with autism spectrum disorders. *The American Journal of Occupational Therapy, 61*, 584–592. https://doi.org/10.5014/ajot.61.5.584.

Bodfish, J. W., Symons, F. J., Parker, D. E., & Lewis, M. H. (2000). Varieties of repetitive behavior in autism: Comparisons to mental retardation. *Journal of Autism and Developmental Disorders, 30*, 237–243. https://doi.org/10.1023/A:1005596502855.

Bross, L. A., & Travers, J. C. (2017). Special interest areas and employment skills programming for secondary students with autism. *Teaching Exceptional Children, 50*, 74–83. https://doi.org/10.1177/0040059917730846.

Bury, S. M., Hedley, D., Uljarević, M., Dissanayake, C., & Gal, E. (2019). If you've employed one person with autism…: An individual difference approach to the autism advantage at work. *Autism, 23*, 1607–1608. https://doi.org/10.1177/1362361318794937.

Bury, S. M., Hedley, D., Uljarević, M., & Gal, E. (2020). The autism advantage at work: A critical and systematic review of current evidence. *Research in Developmental Disabilities, 105*, 13750. https://doi.org/10.1016/j.ridd.2020.103750

Bury, S. M., Flower, R. L., Zulla, R., Nicholas, D. B., & Hedley, D. (2020). Workplace Social Challenges Experienced by Employees on the Autism Spectrum: An International Exploratory Study Examining Employee and Supervisor Perspectives. *Journal of Autism and Developmental Disorders*. https://doi.org/10.1007/s10803-020-04662-6

Cook, G. (2012). The Autism Advantage. *The New Your Times Magazine*. Retrieved from https://www.nytimes.com/2012/12/02/magazine/the-autism-advantage.html

Cunningham, A. B., & Schreibman, L. (2008). Stereotypy in autism: The importance of function. *Research in Autism Spectrum Disorders, 2*, 469–479. https://doi.org/10.1016/j.rasd.2007.09.006.

Demetriou, E., Lampit, A., Quintana, D., Naismith, S., Song, Y., Pye, J., . . . Guastella, A. (2018). Autism spectrum disorders: A meta-analysis of executive function. Molecular Psychiatry, 23, 1198–1204. doi:https://doi.org/10.1038/mp.2017.75.

Esbensen, A. J., Seltzer, M. M., Lam, K. S., & Bodfish, J. W. (2009). Age-related differences in restricted repetitive behaviors in autism spectrum disorders. *Journal of Autism and Developmental Disorders, 39*, 57–66. https://doi.org/10.1007/s10803-008-0599-x.

Evans, D. W., Gray, F. L., & Leckman, J. F. (1999). The rituals, fears and phobias of young children: Insights from development, psychopathology and neurobiology. *Child Psychiatry and Human Development, 29*, 261–276. https://doi.org/10.1023/A:102139293.

Evans, D. W., Leckman, J. F., Carter, A., Reznick, J. S., Henshaw, D., King, R. A., & Pauls, D. (1997). Ritual, habit, and perfectionism: The prevalence and development of compulsive-like behavior in normal young children. *Child Development, 68*, 58–68. https://doi.org/10.1111/j.1467-8624.1997.tb01925.x.

Faragher, E. B., Cass, M., & Cooper, C. L. (2005). The relationship between job satisfaction and health: A meta-analysis. *Occupational and Environmental Medicine, 62*(2), 105–112.

Feather, N. T., & O'Brien, G. E. (1986). A longitudinal study of the effects of employment and unemployment on school-leavers. *Journal of Occupational Psychology, 59*, 121–144.

Flower, R. L., Hedley, D., Spoor, J. R., & Dissanayake, C. (2019). An alternative pathway to employment for autistic job-seekers: A case study of a training and assessment program targeted to autistic job candidates. *Journal of Vocational Education & Training, 71*(3), 407–428. https://doi.org/10.1080/13636820.2019.1636846.

Gal, E. (2011). Nosology and theories of repetitive and restricted behaviours and interests. In J. L. Matson & P. Sturmey (Eds), *International handbook of autism and pervasive developmental disorders* (pp. 115–125). New York: Springer.

Gal, E., Dyck, M. J., & Passmore, A. (2010). Relationships between stereotyped movements and sensory processing disorders in children with and without developmental or sensory disorders. *American Journal of Occupational Therapy, 64*, 453–461. https://doi.org/10.5014/ajot.2010.09075.

Gal, E., Landes, E., & Katz, N. (2015). Work performance skills in adults with and without high functioning autism spectrum disorders (HFASD). *Research in Autism Spectrum Disorders, 10*, 71–77. https://doi.org/10.1016/j.rasd.2014.10.011.

Gal, E., Meir, A. B., & Katz, N. (2013). Development and reliability of the autism work skills questionnaire (AWSQ). *American Journal of Occupational Therapy, 67*, e1–e5. https://doi.org/10.5014/ajot.2013.005066.

Georgiades, S., Papageorgiou, V., & Anagnostou, E. (2010). Brief report: Repetitive behaviours in Greek individuals with autism spectrum disorder. *Journal of Autism and Developmental Disorders, 40*, 903–906. https://doi.org/10.1007/s10803-009-0927-9.

Gonzalez, C., Martin, J. M., Minshew, N. J., & Behrmann, M. (2013). Practice makes improvement: How adults with autism out-perform others in a naturalistic visual search task. *Journal of Autism and Developmental Disorders, 43*, 2259–2268. https://doi.org/10.1007/s10803-013-1772-4.

Happé, F., & Frith, U. (2006). The weak coherence account: Detail-focused cognitive style in autism spectrum disorders. *Journal of Autism and Developmental Disorders, 36*, 5–25. https://doi.org/10.1007/s10803-005-0039-0.

Happé, F., & Frith, U. (2009). The beautiful otherness of the autistic mind. *Philosophical Transactions of the Royal Society B, 364*, 1345–1350. https://doi.org/10.1098/rstb.2009.0009.

Happé, F., & Vital, P. (2009). What aspects of autism predispose to talent? *Philosophical Transactions of the Royal Society of London B: Biological Sciences, 364*, 1369–1375. https://doi.org/10.1098/rstb.2008.0332.

Harrop, C., McBee, M., & Boyd, B. A. (2016). How are child restricted and repetitive behaviors associated with caregiver stress over time? A parallel process multilevel growth model. *Journal of Autism and Developmental Disorders, 46*, 1773–1783. https://doi.org/10.1007/s10803-016-2707-7.

Hedley, D., Cai, R., Uljarevic, M., Wilmot, M., Spoor, J. R., Richdale, A., & Dissanayake, C. (2018). Transition to work: Perspectives from the autism spectrum. *Autism, 22*, 528–541. https://doi.org/10.1177/1362361316687697.

Hedley, D., Uljarević, M., Bury, S. M., & Dissanayake, C. (2019). Predictors of mental health and well-being in employed adults with Autism Spectrum disorder at 12-month follow-up. *Autism Research, 12*, 482–494. https://doi.org/10.1002/aur.2064.

Hedley, D., Uljarević, M., Cameron, L., Halder, S., Richdale, A., & Dissanayake, C. (2017). Employment programmes and interventions targeting adults with autism spectrum disorder: A systematic review of the literature. *Autism, 21*, 929–941. https://doi.org/10.1177/1362361316661855.

Hedley, D., Uljarević, M., & Hedley, D. F. (2017). Employment and living with Autism: Personal, social and economic impact. In S. Halder & L. C. Assaf (Eds), *Inclusion, disability and culture* (pp. 295–311). Switzerland: Springer.

Heinz, A. J., Meffert, B. N., Halvorson, M. A., Blonigen, D., Timko, C., & Cronkite, R. (2018). Employment characteristics, work environment, and the course of depression over 23 years: Does employment help foster resilience? *Depression and Anxiety, 35*(9), 861–867. https://doi.org/10.1002/da.22782.

Hillier, A., Campbell, H., Mastriani, K., Izzo, M. V., Kool-Tucker, A. K., Cherry, L., & Beversdorf, D. Q. (2007). Two-year evaluation of a vocational support program for adults on the autism spectrum. *Career Development for Exceptional Individuals, 30*, 35–47. https://doi.org/10.1177/08857288070300010501.

Holwerda, A., van der Klink, J. J., Groothoff, J. W., & Brouwer, S. (2012). Predictors for work participation in individuals with an autism spectrum disorder: A systematic review. *Journal of Occupational Rehabilitation, 22*, 333–352. https://doi.org/10.1007/s10926-011-9347-8.

Honey, E., McConachie, H., Randle, V., Shearer, H., & Le Couteur, A. S. (2008). One-year change in repetitive behaviours in young children with communication disorders including autism. *Journal of Autism and Developmental Disorders, 38*, 1439–1450. https://doi.org/10.1007/s10803-006-0191-1.

Howlin, P., Goode, S., Hutton, J., & Rutter, M. (2004). Adult outcome for children with autism. *Journal of Child Psychology and Psychiatry, 45*, 212–229. https://doi.org/10.1111/j.1469-7610.2004.00215.x.

Howlin, P., Goode, S., Hutton, J., & Rutter, M. (2009). Savant skills in autism: Psychometric approaches and parental reports. *Philosophical Transactions of the Royal Society B: Biological Sciences, 364*, 1359–1367. https://doi.org/10.1098/rstb.2008.0328.

Joyce, C., Honey, E., Leekam, S. R., Barrett, S. L., & Rodgers, J. (2017). Anxiety, intolerance of uncertainty and restricted and repetitive behaviour: Insights directly from young people with ASD. *Journal of Autism and Developmental Disorders, 47*, 3789–3802. https://doi.org/10.1007/s10803-017-3027-2.

Kaldy, Z., Giserman, I., Carter, A. S., & Blaser, E. (2016). The mechanisms underlying the ASD advantage in visual search. *Journal of Autism and Developmental Disorders, 46*, 1513–1527. https://doi.org/10.1007/s10803-013-1957-x.

Kanner, L. (1943). Autistic disturbances of affective contact. *Nervous child, 2*(3), 217–250.

Ki-moon, B. (2015). Secretary-General Invites Business to Commit to Hiring People with Autism, as He Launches 'Call to Action' Initiative on World Day. Retrieved from https://www.un.org/press/en/2015/sgsm16639.doc.htm

Kirchner, J. C., & Dziobek, I. (2014). Towards successful employment of adults with autism: A first analysis of special interests and factors deemed important for vocational performance. *Scandinavian Journal of Child and Adolescent Psychiatry and Psychology, 2*, 77–85. https://doi.org/10.21307/sjcapp-2014-011.

Klin, A., Danovitch, J. H., Merz, A. B., & Volkmar, F. R. (2007). Circumscribed interests in higher functioning individuals with autism spectrum disorders: An exploratory study. *Research and Practice for Persons with Severe Disabilities, 32*, 89–100. https://doi.org/10.2511/rpsd.32.2.89.

Kohls, G., Antezana, L., Mosner, M. G., Schultz, R. T., & Yerys, B. E. (2018). Altered reward system reactivity for personalized circumscribed interests in autism. *Molecular Autism, 9*, *9*. https://doi.org/10.1186/s13229-018-0195-7.

Krieger, B., Kinébanian, A., Prodinger, B., & Heigl, F. (2012). Becoming a member of the work force: Perceptions of adults with Asperger syndrome. *Work, 43*, 141–157. https://doi.org/10.3233/WOR-2012-1392.

Lam, K. S., Bodfish, J. W., & Piven, J. (2008). Evidence for three subtypes of repetitive behavior in autism that differ in familiality and association with other symptoms. *Journal of Child Psychology and Psychiatry, 49*, 1193–1200. https://doi.org/10.1111/j.1469-7610.2008.01944.x.

Langen, M., Durston, S., Kas, M. J., van Engeland, H., & Staal, W. G. (2011). The neurobiology of repetitive behavior:… and men. *Neuroscience and Biobehavioral Reviews, 35*, 356–365. https://doi.org/10.1016/j.neubiorev.2010.02.005.

Leekam, S. R., Nieto, C., Libby, S. J., Wing, L., & Gould, J. (2007). Describing the sensory abnormalities of children and adults with autism. *Journal of Autism and Developmental Disorders, 37*, 894–910. https://doi.org/10.1007/s10803-006-0218-7.

Leekam, S. R., Prior, M. R., & Uljarevic, M. (2011). Restricted and repetitive behaviors in autism spectrum disorders: A review of research in the last decade. *Psychological Bulletin, 137*, 562–593. https://doi.org/10.1037/a0023341.

Lidstone, J., Uljarević, M., Sullivan, J., Rodgers, J., McConachie, H., Freeston, M., . . . Leekam, S. (2014). Relations among restricted and repetitive behaviors, anxiety and sensory features in children with autism spectrum disorders. Research in Autism Spectrum Disorders*,* 8, 82–92. doi:https://doi.org/10.1016/j.rasd.2013.10.001.

Mawhood, L., & Howlin, P. (1999). The outcome of a supported employment scheme for high-functioning adults with autism or Asperger syndrome. *Autism, 3*, 229–254. https://doi.org/10.1177/1362361399003003003.

McKee-Ryan, F., Song, Z., Wanberg, C. R., & Kinicki, A. J. (2005). Psychological and physical well-being during unemployment: A meta-analytic study. *The Journal of Applied Psychology, 90*, 53–76.

Mercier, C., Mottron, L., & Belleville, S. (2000). A psychosocial study on restricted interests in high functioning persons with pervasive developmental disorders. *Autism, 4*, 406–425. https://doi.org/10.1177/1362361300004004006.

Müller, E., Schuler, A., Burton, B. A., & Yates, G. B. (2003). Meeting the vocational support needs of individuals with Asperger syndrome and other autism spectrum disabilities. *Journal of Vocational Rehabilitation, 18*, 163–175.

Nicholas, D. B., Hedley, D., Randolph, J. K., Raymaker, D. M., Robertson, S. M., & Vincent, J. (2019). An expert discussion on employment in autism. *Autism in Adulthood, 1*(3), 162–169. https://doi.org/10.1089/aut.2019.29003.djn.

Nicholas, D. B., & Klag, M. (2020). Critical reflections on employment among autistic adults. *Autism in Adulthood*. https://doi.org/10.1089/aut.2020.0006.

Olney, M. F. (2000). Working with autism and other social-communication disorders. *Journal of Rehabilitation, 66*, 51–56.

O'Riordan, M. A., Plaisted, K. C., Driver, J., & Baron-Cohen, S. (2001). Superior visual search in autism. *Journal of Experimental Psychology: Human Perception and Performance, 27*, 719–730. https://doi.org/10.1177/1362361304045219.

Pellicano, E., & Burr, D. (2012). When the world becomes 'too real': A Bayesian explanation of autistic perception. *Trends in Cognitive Sciences, 16*, 504–510. https://doi.org/10.1016/j.tics.2012.08.009.

Pfeiffer, B., Braun, K., Kinnealey, M., Derstine Matczak, M., & Polatajko, H. (2017). Environmental factors impacting work satisfaction and performance for adults with autism spectrum disorders. *Journal of Vocational Rehabilitation, 47*, 1–12. https://doi.org/10.3233/Jvr-170878.

Plaisted, K., O'Riordan, M., & Baron-Cohen, S. (1998). Enhanced discrimination of novel, highly similar stimuli by adults with autism during a perceptual learning task. *The Journal*

of Child Psychology and Psychiatry and Allied Disciplines, 39, 765–775. https://doi.org/10.1111/1469-7610.00375.

Rodgers, J., Glod, M., Connolly, B., & McConachie, H. (2012). The relationship between anxiety and repetitive behaviours in autism spectrum disorder. *Journal of Autism and Developmental Disorders, 42*, 2404–2409. https://doi.org/10.1007/s10803-012-1531-y.

Roux, A. M., Shattuck, P. T., Rast, J. E., Rava, J. A., & Anderson, K. A. (2015). *National autism indicators report: Transition into young adulthood*. Philadelphia: Life Course Outcomes Research Program, A.J. Drexel Autism Institute, Drexel University.

Royo-León, M., Hedley, D., Gal, E., Katz, N., Waisman, N., Schreuer, N., Uljarević, M., Spoor, J., Masters, C., Bury, S., Juárez-Gallegos, M. A., Ramirez-Ochoa, D. D., & Barroso-Barajas, A. J. (2018). eWPQ: Development of a computer-based work performance review application for persons with autism spectrum disorder. In *Congreso Internacional de Ingeniería Electrónica Memoria Electro* (Vol. 40, pp. 101–107). Mexico: Instituto Tecnologico de Chihuahua.

Russell, G., Kapp, S. K., Elliott, D., Elphick, C., Gwernan-Jones, R., & Owens, C. (2019). Mapping the autistic advantage from the accounts of adults diagnosed with autism: A qualitative study. *Autism in Adulthood, 1*(2), 124–133. https://doi.org/10.1089/aut.2018.0035.

Scott, M., Jacob, A., Hendrie, D., Parsons, R., Girdler, S., Falkmer, T., & Falkmer, M. (2017). Employers' perception of the costs and the benefits of hiring individuals with autism spectrum disorder in open employment in Australia. *PLoS One, 12*, 1–16. https://doi.org/10.1371/journal.pone.0177607.

Shah, A., & Frith, U. (1983). An islet of ability in autistic children: A research note. *Journal of Child Psychology and Psychiatry, 24*, 613–620. https://doi.org/10.1111/j.1469-7610.1983.tb00137.x.

Shattuck, P. T., Narendorf, S. C., Cooper, B., Sterzing, P. R., Wagner, M., & Taylor, J. L. (2012). Postsecondary education and employment among youth with an autism spectrum disorder. *Pediatrics, 129*, 1042–1049. https://doi.org/10.1542/peds.2011-2864.

Shattuck, P. T., Seltzer, M. M., Greenberg, J. S., Orsmond, G. I., Bolt, D., Kring, S., . . . Lord, C. (2007). Change in autism symptoms and maladaptive behaviors in adolescents and adults with an autism spectrum disorder. Journal of Autism and Developmental Disorders, 37, 1735–1747. doi:https://doi.org/10.1007/s10803-006-0307-7.

Smith, M. D., Belcer, R. G., & Juhrs, P. D. (1995). *A guide to successful employment for individuals with autism*. Baltimore: P. H. Brookes.

Smith, R. S., & Sharp, J. (2013). Fascination and isolation: A grounded theory exploration of unusual sensory experiences in adults with Asperger syndrome. *Journal of Autism and Developmental Disorders, 43*, 891–910. https://doi.org/10.1007/s10803-012-1633-6.

South, M., Ozonoff, S., & McMahon, W. M. (2005). Repetitive behavior profiles in Asperger syndrome and high-functioning autism. *Journal of Autism and Developmental Disorders, 35*, 145–158. https://doi.org/10.1007/s10803-004-1992-8.

Sprague, R. L., & Newell, K. M. (1996). *Stereotyped movements: Brain and behavior relationships.* Washington, DC: American Psychological Association.

Stevenson, J. L., & Gernsbacher, M. A. (2013). Abstract spatial reasoning as an autistic strength. *PLoS One, 8*, e59329. https://doi.org/10.1371/journal.pone.0059329.

Thelen, E. (1979). Rhythmical stereotypies in normal human infants. *Animal Behaviour, 27*, 699–715. https://doi.org/10.1016/0003-3472(79)90006-x.

Tregay, J., Gilmour, J., & Charman, T. (2009). Childhood rituals and executive functions. *British Journal of Developmental Psychology, 27*, 283–296. https://doi.org/10.1348/026151008X299737.

Turner-Brown, L. M., Lam, K. S., Holtzclaw, T. N., Dichter, G. S., & Bodfish, J. W. (2011). Phenomenology and measurement of circumscribed interests in autism spectrum disorders. *Autism, 15*, 437–456. https://doi.org/10.1177/1362361310386507.

Uljarević, M., Baranek, G., Vivanti, G., Hedley, D., Hudry, K., & Lane, A. (2017). Heterogeneity of sensory features in autism spectrum disorder: Challenges and perspectives for future research. *Autism Research, 10*, 703–710. https://doi.org/10.1002/aur.1747.

Uljarević, M., Richdale, A. L., Evans, D. W., Cai, R. Y., & Leekam, S. R. (2017). Interrelationship between insistence on sameness, effortful control and anxiety in adolescents and young adults with autism spectrum disorder (ASD). *Molecular Autism, 8*, 1–6. https://doi.org/10.1186/s13229-017-0158-4.

United Nations Regional Information Centre for Western Europe. (2015). Employment: The Autism Advantage! Retrieved from https://www.unric.org/en/latest-un-buzz/29720-employment-the-autism-advantage

Van Bourgondien, M. E., & Woods, A. V. (1992). Vocational possibilities for high-functioning adults with autism. In E. Schopler & G. B. Mesibov (Eds), *High-functioning individuals with autism* (pp. 227–239). Boston: Springer.

Vogus, T. J., & Taylor, J. L. (2018). Flipping the script: Bringing an organizational perspective to the study of autism at work. *Autism, 22*, 514–516. https://doi.org/10.1177/1362361318776103.

Voßemer, J., Gebel, M., Täht, K., Unt, M., Högberg, B., & Strandh, M. (2018). The effects of unemployment and insecure jobs on well-being and health: The moderating role of labor market policies. *Social Indicators Research, 138*, 1229–1257.

Wanberg, C. R. (2012). The individual experience of unemployment. *Annual Review of Psychology, 63*, 369–396.

Wehman, P. H., Schall, C. M., McDonough, J., Kregel, J., Brooke, V., Molinelli, A., . . . Collins, H. T. (2014). Competitive employment for youth with autism spectrum disorders: Early results from a randomized clinical trial. Journal of Autism and Developmental Disorders, 44, 487–500. doi:https://doi.org/10.1007/s10803-013-1892-x.

Wilkes, B., & Lewis, M. (2018). The neural circuitry of restricted repetitive behavior: Magnetic resonance imaging in neurodevelopmental disorders and animal models. *Neuroscience & Biobehavioral Reviews, 92*, 152–171. https://doi.org/10.1016/j.neubiorev.2018.05.022.

Wolff, P. H. (1968). Stereotypic behavior and development. *Canadian Psychologist/Psychologie Canadienne, 9*, 474–484. https://doi.org/10.1037/h0082674.

Yerys, B. E. (2015). An update on the neurobiology of repetitive behaviors in autism. *International Review of Research in Developmental Disabilities, 49*, 91–150.

Index

E. Gal, N. Yirmiya (eds.), *Repetitive and Restricted Behaviors and Interests in Autism Spectrum Disorders*, Autism and Child Psychopathology Series,
https://doi.org/10.1007/978-3-030-66445-9