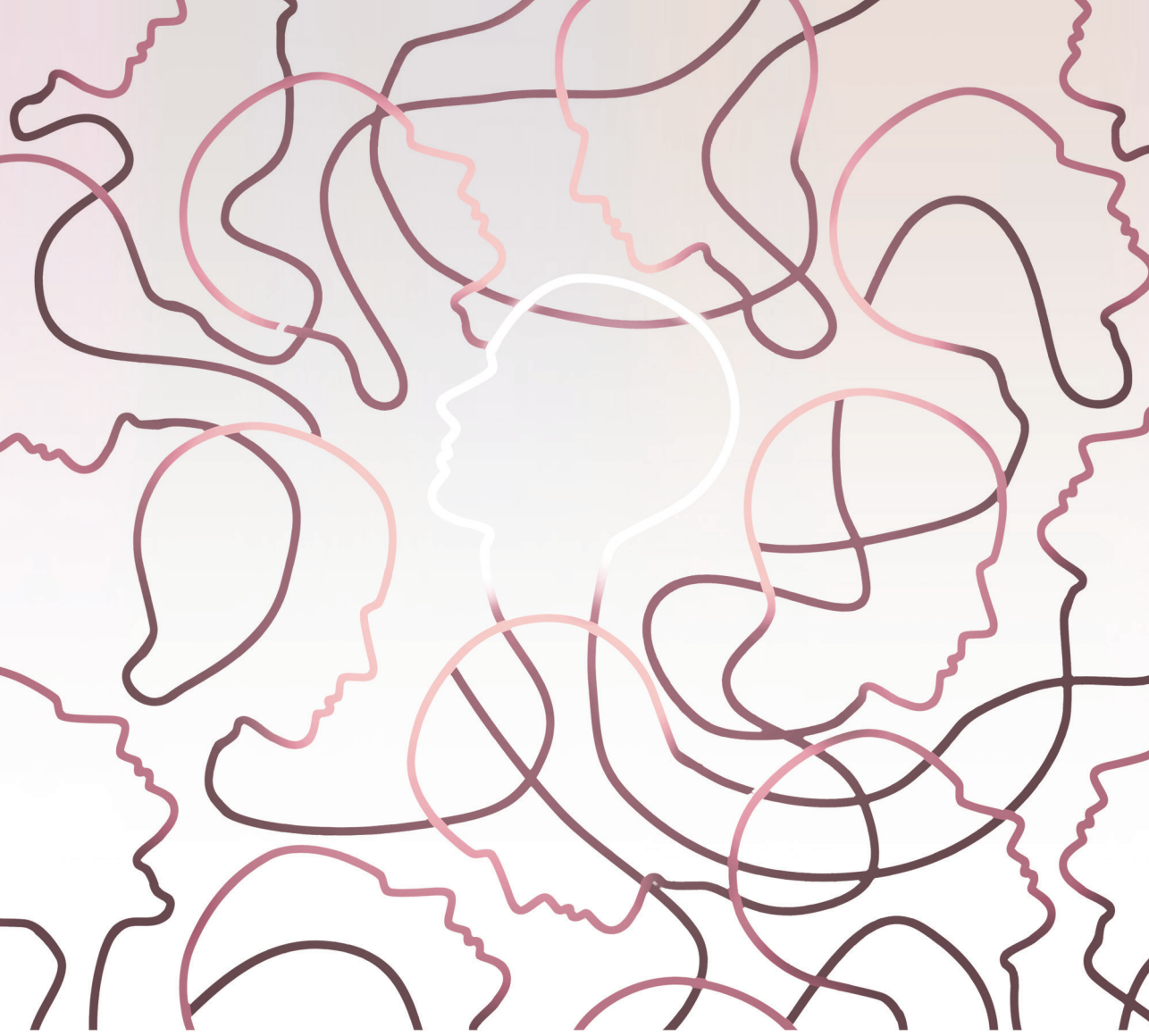

Autism spectrum disorder in people with multiple disabilities

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Chapter 1

General introduction

1. Introduction

In clinical practice, particularly in the field of care for individuals who have combined intellectual and sensory disabilities, the diagnostic process is challenging. Clinicians are often consulted about the behaviour of such multiply disabled individuals. One of their focal questions is whether their behaviour is related to specific disorders, among which Autism Spectrum Disorder (ASD). ASD is a developmental disorder that is characterized by deficits in social communication and interaction and by restricted and repetitive behaviour patterns and resistance to change (American Psychiatric Association, 2013). ASD is said to be more prevalent in persons with intellectual disabilities (Bryson, Bradley, Thompson, & Wainwright, 2008; Matson & Shoemaker, 2009), persons with visual impairments (Brown, Hobson, Lee, & Stevenson, 1997; Mukaddes, Kilincaslan, Kucukyazici, Sevketoglu, & Tuncer, 2007) and in persons with a combination of these disabilities (Carvill, 2001). However, regardless of the actual presence of ASD, symptoms that are typical for ASD are present in people with intellectual disabilities (Matson & Shoemaker, 2009), visual impairments (Brown, et al., 1997; Fraiberg, 1977), hearing loss (Knoors & Vervloed, 2011) or a combination of these disabilities (Evenhuis, Sjoukes, Koot, & Kooijman, 2009; Hoevenaars-van den Boom, Antonissen, Knoors, & Vervloed, 2009).

This overlap in behavioural characteristics of ASD with multiple disabilities raises the question whether the high prevalence of ASD in people with multiple disabilities is accurate or is, in fact, a sign that ASD is over-diagnosed. This could mean that some individuals are unfairly diagnosed with ASD, based on behavioural characteristics that are actually a reflection of their sensory or intellectual disabilities (Andrews & Wyver, 2005; Cass, 1998). However, because symptoms of ASD and characteristics of sensory and intellectual disabilities are so much alike, it is also possible that in some individuals ASD is present but overlooked. ASD can easily be overlooked when clinicians attribute the cause of any behaviour to their sensory or intellectual disabilities, when it could also be a symptom of ASD (Carvill, 2001). This phenomenon where characteristics of one disorder or disability are unfairly attributed to a more notable disability is called diagnostic overshadowing (Reiss, Levitan, & Szyszko, 1982).

A second issue in the field of diagnosing ASD in individuals with multiple disabilities is the lack of diagnostic instruments. When a person shows behaviour that at first glance may be related to ASD, such as stereotyped behaviour or impaired social communication, a first step in the diagnostic process is to screen for ASD symptoms. If a person screens positive for ASD, the next step is a more elaborate diagnostic assessment. The results will then indicate if ASD is present or not. Both screening and diagnosis should be done with valid and reliable instruments, such as checklists, standardized observations and assessment instruments (Nederlandse Vereniging voor Psychiatrie, 2009). This is particularly important for children and adults with

multiple disabilities because of the behavioural overlap between characteristics of sensory and intellectual disabilities and symptoms of ASD. However, as will be shown later in this thesis, for this population no such instruments are available.

The lack of diagnostic instruments can be troubling when it comes to treatment or intervention plans. When clinicians plan treatment, they often take a person's clinical classification into account. A wrong classification can therefore lead to the wrong treatment (Howlin, 2000; Kraijer & Plas, 2006). For example, stereotyped behaviour frequently occurs in individuals with multiple disabilities, but it is also an important diagnostic criterion for ASD. In someone who is blind, stereotyped behaviour may be caused by understimulation, while in persons with ASD stereotyped behaviour is often caused by overstimulation or stress (Frith, 2003; Gense & Gense, 2005; Warren, 1994). If a clinician attributes the stereotyped behaviours to ASD, they might remove external stimuli and place the person in a quiet environment with the purpose of reducing the behaviours. However, if this person does not have ASD and the stereotyped behaviour is caused by understimulation due to blindness, this chosen intervention may even worsen the behaviours. A more appropriate response would then be the opposite, to engage this person with more stimulation.

At this moment, no instruments are available that are specifically designed for assessing symptoms of ASD in people with sensory and intellectual disabilities. A new and valid instrument may help clinicians to correctly attribute behaviours to specific disabilities or disorders in persons with multiple disabilities, in perspective of providing them with optimal treatment. To cope with the challenges that are described above, the aim of the current thesis is to gain more insight into the behavioural characteristics of people with and without ASD who are already known to have sensory and intellectual disabilities. In order to do so, an attempt was made to develop and validate a diagnostic instrument that can assess the presence of ASD in people with sensory and intellectual disabilities.

2. Diagnosing Autism Spectrum Disorder

ASD is a developmental disorder that consists of two main components; see Figure 1 (American Psychiatric Association, 2013). The first component of ASD 'deficits in communication and social interaction' consists of three criteria, all of which have to be present in order to be diagnosed with ASD. The second component of ASD, 'restricted and repetitive patterns of behaviour, interests or activities', consists of four criteria, two of which have to be present to be diagnosed with ASD. Though these main components must be present in each individual with ASD, the disorder is heterogeneous and the symptoms may express themselves differently in each individual, both in quality and quantity (Frith, 2008; Grzadzinski, Huerta, & Lord,

2013; Happé, Ronald, & Plomin, 2006). How ASD expresses itself also depends on the severity of the disorder. The DSM-5 distinguishes three severity levels, based on how strongly the symptoms are expressed and indicating how much support an individual needs in their daily life (American Psychiatric Association, 2013).

ASD often co-occurs with other disorders and disabilities. As mentioned above, the prevalence of ASD is said to be related to the presence of sensory and intellectual disabilities. In addition, persons with ASD are at a higher risk to developing problems in attachment style and they are more sensitive to stress (Corbett, Mendoza, Abdullah, Wegelin, & Levine, 2006; Rutgers, Bakermans-Kranenburg, Van IJzendoorn, & Van Berckelaer-Onnes, 2004; Stewart, Barnard, Pearson, Hasan, & O'Brien, 2006). When more than one of these disabilities co-occurs with another, this may lead to even more ASD typical symptoms.

Research suggests that ASD has a strong genetic component (Devlin & Scherer, 2012; Happé, et al., 2006; Huguet, Ey, & Bourgeron, 2013; Ronald et al., 2006), but a biological marker that can be used in the diagnostic process of ASD has not yet been found. As a result, the diagnosis of ASD remains to be based on the behavioural characteristics (Frith, 2003) as described in Figure 1. The diagnosis of ASD requires an elaborate assessment procedure. In this process, multiple screening- and diagnostic instruments are used and their results are combined with observations and a person's medical and psychological history. It is important to use a multi-disciplinary and multi-method approach (National Institute for Health and Clinical Excellence, 2012; Nederlandse Vereniging voor Psychiatrie, 2009; Risi et al., 2006; Volkmar et al., 2014) in order to find appropriate treatments for each individual (Rutter, 2006) preventing behavioural problems as a consequence (Howlin, 2000).

3. Study sample and definition of multiple disabilities

This thesis focuses on people with multiple disabilities. The expression 'multiple disabilities' is used to refer to a wide range of persons with more than one disability, ranging from any combination of sensory disabilities, motor disabilities, intellectual disabilities and psychiatric disorders (Nakken & Vlaskamp, 2007). However, with 'multiple disabilities' we do not simply mean two separate disabilities existing together, like the comorbidity of two or more somatic diseases. 'Multiple disabilities' refers to a combination of at least two disabilities that influence each other's effects, or influence the possibilities to cope with the effects of the individual disabilities. This means that the treatment and intervention for coping with one of the present disabilities does not automatically work when a person is multiply disabled. The consequences of one disability may impair a person's ability to compensate for the consequences of the other disability (Gunther & de Jong, 1988; Knoors & Vervloed,

A. Persistent deficits in social communication and social interaction across multiple contexts, as manifested by the following, currently or by history (examples are illustrative, not exhaustive):
   1. Deficits in social-emotional reciprocity, ranging, for example, from abnormal social approach and failure of normal back-and-forth conversation; to reduced sharing of interests, emotions, or affect; to failure to initiate or respond to social interactions.
   2. Deficits in nonverbal communicative behaviors used for social interaction, ranging, for example, from poorly integrated verbal and nonverbal communication; to abnormalities in eye contact and body language or deficits in understanding and use of gestures; to a total lack of facial expressions and nonverbal communication.
   3. Deficits in developing, maintaining, and understanding relationships, ranging, for example, from difficulties adjusting behavior to suit various social contexts; to difficulties in sharing imaginative play or in making friends; to absence of interest in peers.

B. Restricted, repetitive patterns of behavior, interests, or activities, as manifested by at least two of the following, currently or by history (examples are illustrative, not exhaustive):
   1. Stereotyped or repetitive motor movements, use of objects, or speech (e.g., simple motor stereotypies, lining up toys or flipping objects, echolalia, idiosyncratic phrases).
   2. Insistence on sameness, inflexible adherence to routines, or ritualized patterns of verbal or nonverbal behavior (e.g., extreme distress at small changes, difficulties with transitions, rigid thinking patterns, greeting rituals, need to take same route or eat same food every day).
   3. Highly restricted, fixated interests that are abnormal in intensity or focus (e.g., strong attachment to or preoccupation with unusual objects, excessively circumscribed or perseverative interests)
   4. Hyper- or hyporeactivity to sensory input or unusual interest 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).

C. Symptoms must be present in the early developmental period (but may not become fully manifest until social demands exceed limited capacities, or may be masked by learned strategies in later life).

D. Symptoms cause clinically significant impairment in social, occupational, or other important areas of current functioning.

E. These disturbances are not better explained by intellectual disability (intellectual developmental disorder) or global developmental delay. Intellectual disability and autism spectrum disorder frequently co-occur; to make comorbid diagnoses of autism spectrum disorder and intellectual disability, social communication should be below that expected for general developmental level.

Figure 1 Criteria for ASD as described in DSM-5 (cited from APA, 2013)

2011; Nakken, 1993). For example, a deaf individual can compensate for the limitations in spoken language communication by using sign language. However, when this person also has a visual impairment, the options for visual compensation become limited to non-existent. As a result, communication is severely impaired, unless a new option to access communication is explored, for example by using the tactile modality.

Within the group of persons with multiple disabilities, this thesis deals with a specific subgroup of the population: Individuals with a moderate to profound intellectual disability, combined with a visual impairment and/or deafblindness. With regard to the intellectual disability, the definitions and classification of the American Association on Intellectual and Developmental Disabilities (2013) and the American Psychiatric Association (2013) were followed. An intellectual disability is defined by impairments in both intellectual and adaptive functioning. The intellectual impairments may refer to deficits in learning, problem solving and reasoning, and can be indicated by an IQ score below 70. The deficits in adaptive functioning can be recognised as a failure to meet standards in various aspects of daily life, including communication, social participation and independent living (American Association on Intellectual and Developmental Disabilities, 2013; American Psychiatric Association, 2013). Four severity levels of intellectual disability are commonly distinguished, namely: mild (IQ score between 50 - 70), moderate (IQ between 35 – 50), severe (IQ between 20 – 35) and profound intellectual disability (IQ score below 20) (World Health Organization, 2016). In the current study, individuals with a moderate, severe and profound intellectual disability were included.

A visual impairment is described by the World Health Organization (2016) using visual acuity and visual field after correction. Five categories of severity of visual impairment are distinguished. *Category 1* is a moderate visual impairment that is defined by a visual acuity between 6/18 and 6/60. When a person has a visual acuity that is better than 6/18, this person has a mild or no visual impairment. *Category 2* is a severe visual impairment and is defined by a visual acuity between 6/60 an 3/60. When visual acuity is below 3/60 or the visual field of a person is less than 10 degrees in the better eye, this is considered *category 3* blindness. *Category 4* blindness ranges between a visual acuity of less than 1/60 to no visual acuity at all but with light perception. When a person has no light perception, this person has *category 5* blindness (World Health Organization, 2016). In this study, persons with visual impairments from categories 1 through 5 have been included.

Hearing loss or deafness can be described as hearing loss in one or both ears (World Health Organization, 2016). One speaks of disabling hearing loss when the hearing loss is greater than 40 dB in the better ear for adults, and 30 dB in the better ear for children (World Health Organization, 2017). Having hearing loss was not an inclusion criterion for our study. However, persons with disabling hearing loss have

been included when they were deafblind. Many different definitions of deafblindness have been used in the literature, based on the level of sensory impairment, level of functioning, the aetiology of the sensory disabilities and even communicative abilities and level of mobility (Ask Larsen & Damen, 2014). Any given combination of a visual impairment with hearing loss can be referred to as deafblindness. Within the current study, we followed the Dutch convention for classification of deafblindness. A person was regarded as deafblind when they had an average hearing loss of at least 35 decibel in addition to any type of visual impairment (Doofblind.nl, 2017).

In sum, the current study focused on people with a moderate to profound intellectual disability combined with a visual impairment or deafblindness. We will refer to this target group as people with multiple disabilities or people with a combination of sensory and intellectual disabilities. Both adults and children were included as participants, with an age range of 6-60 years. Children younger than 6 years of age were not included because certain behavioural characteristics that were assessed may be not have been fully developed at that age. Adults older than 60 years of age were not included in order to prevent participation of adults with early dementia or other behaviours related to old age that could influence the behaviours that were assessed. See Table 1 for an overview of inclusion and exclusion criteria.

Table 1 Inclusion and exclusion criteria for participants in this thesis

Inclusion criteria	
Age	Between 6 – 60 years old
Intellectual Disability	A moderate, severe or profound intellectual disability (IQ < 50), following the criteria described by the AAIDD (2013), APA (2013) and WHO (2016).
Sensory Disability	A visual impairment from categories 1 through 5, following the criteria of the WHO (2016) *OR;* Deafblindess: A visual impairment combined with hearing loss of 35 decibel or more, following the definition of doofblind.nl (2017)
Exclusion criteria	
Motor Abilities	Severe motor disabilities in arms or complete paralysis from neck down

4. ASD symptoms and mental health problems in multiple disabilities

Both an intellectual disability and a sensory disability, whether auditory, visual or a combination of these two, may impair a person's behaviour or daily functioning. However, when these disabilities are present together, the overall impact may be even more severe (Evenhuis, et al., 2009). An example is the deficit in communication mentioned earlier. Deaf individuals often have communicative difficulties, but they can compensate these difficulties by using sign language or speech reading. A visual impairment in addition to deafness rules out these solutions for communication. This situation becomes even more complicated when an additional intellectual disability is present, because intellectual disabilities cause problems with understanding the environment and communicating needs and desires.

The interpretation of the behaviour of individuals with combined sensory and intellectual disabilities is very complex. There is a large overlap between behavioural characteristics caused by their sensory and intellectual disabilities and symptoms of ASD, such as communicative difficulties, stereotyped behaviour and adherence to routines. Moreover, persons with multiple disabilities are also more sensitive to developing mental health problems, such as attachment problems, stress and mood disorders (Bloeming-Wolbrink et al., 2012; Hurley, 2006).

Persons with combined sensory and intellectual disabilities often have challenges in communication and sometimes their communicative abilities may appear absent. The combination of disabilities may also lead to increased impairments in independent living and adaptive behaviour skills and to challenging or problematic behaviour (Carvill, 2001; Evenhuis, et al., 2009). These impairments can also affect other developmental domains or areas of daily living. For example, parents or caregivers can easily miss communicative behaviours because of the atypical nature of these behaviours in persons with multiple disabilities. This can make individuals with disabilities more prone to attachment problems (Schuengel & Janssen, 2006; Sterkenburg, 2008; Warren, 1994).

Mental health problems in persons with multiple disabilities can result in the expression of more ASD typical symptoms. For example, stress or anxiety can also lead to more stereotyped and repetitive behaviours (Kraijer, 2004; Leekam, Prior, & Uljarevic, 2011; Rubin, Coplan, & Bowker, 2013) which are core characteristics of ASD. On the other hand, it appears that not only persons with multiple disabilities, but also persons with ASD are more sensitive to developing mental health problems such as attachment problems, stress and mood disorders (Corbett, et al., 2006; Rutgers, et al., 2004; Stewart, et al., 2006).

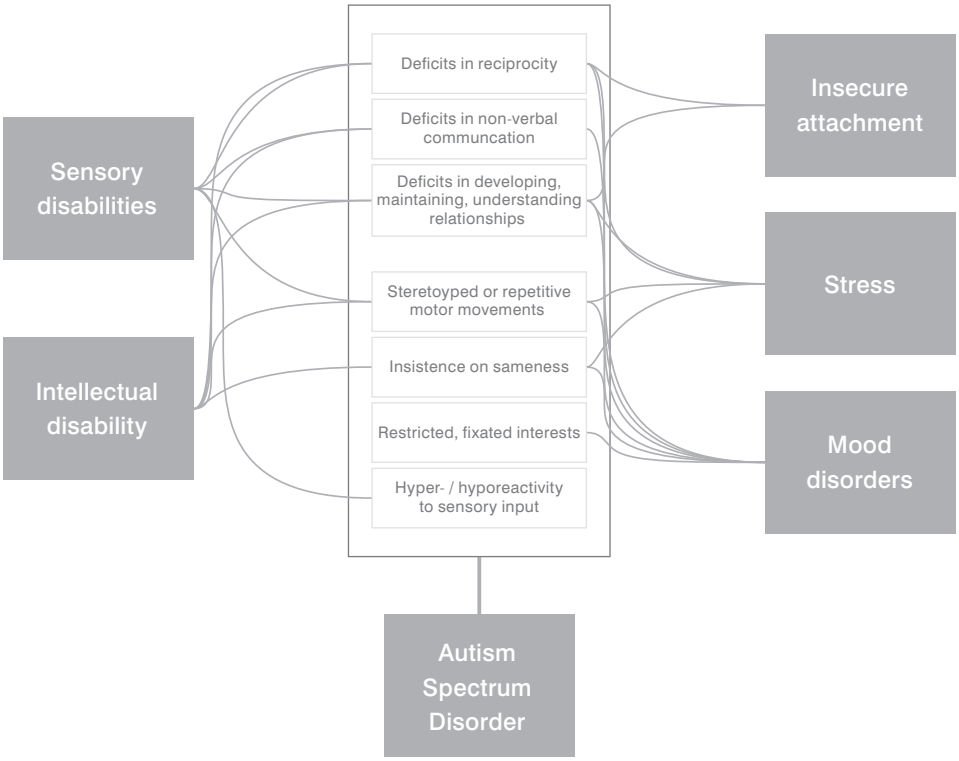

Figure 2 Behavioural overlap between ASD, sensory and intellectual disabilities and mental health problems.

Overall, the behaviours that can be seen in individuals with intellectual disabilities combined with sensory disabilities are very similar to ASD (Carvill, 2001), but can be caused by a variety of disabilities and disorders. Figure 1 shows the complexity of this problem. Persons with sensory and intellectual disabilities show certain behaviours, such as deficits in reciprocity, communication and stereotyped behaviour. These could be the result of their sensory or intellectual disabilities. However, when observed together, these behavioural characteristics fit the diagnostic criteria for ASD. Once a clinician observes the presence of these behaviours combined, they may diagnose ASD, while in fact the behaviours may be caused by one of the other present disabilities. At the same time, the presence of these behavioural impairments may be signs that a mental health disorder is present, such as an insecure attachment, stress or mood disorders.

This large overlap in behaviour creates uncertainty about what the underlying cause is for the observed behaviour patterns, making the diagnostic process difficult. Additionally, there is a lack of diagnostic instruments that take both the intellectual and sensory disability into account. This further complicates the diagnostic process (Bodsworth, Clare, Simblett, & Deafblind UK, 2011; Jure, Rapin, & Tuchman, 1991; Tobin & Hill, 2011). These problems in the diagnostic process may affect the treatment of individuals with multiple disabilities.

Treatments are often based on the diagnostic classifications of a person, therefore a correct diagnosis is necessary to ensure correct treatment (Howlin, 2000; Rutter, 2006). This can be problematic in some cases. As mentioned earlier, the intervention for stereotyped behaviour within ASD is quite the opposite from the intervention for the same behaviour when there is no ASD but only a visual impairment. In these cases, the chosen intervention can have counterproductive effects. The same can be found in other areas. Both multiply disabled adults with and without ASD show impairments in communication. If ASD is present, one might react by reducing the number of communicative attempts and with this, reducing stress related to over-asking the individual. However, if ASD is not present, reducing communicative attempts may result in more stress because the person does not get the social attention they desire.

In order to solve these diagnostic difficulties, it should be clarified when certain behaviours are a normal part of having multiple disabilities and when they are a part of ASD. To do this, more instruments are needed that take into account the sensory disabilities and that can differentiate the behaviours of sensory and intellectual disabilities from symptoms of ASD. Additionally, the symptoms that are described may be signs of ASD, signs of sensory and intellectual disabilities, but can also be signs of mental health problems. These mental health problems are said to be related to the presence of multiple disabilities and ASD. Therefore more insights should be gained in the overlapping behaviours and in the presence of these mental health problems so that any overlap can be unravelled.

5. The current study

In summary, people with combined sensory and intellectual disabilities often present ASD symptoms or behaviours that are similar to ASD symptoms (e.g. Dalby et al., 2009; Dammeyer, 2013; Evenhuis, et al., 2009; Hoevenaars-van den Boom, et al., 2009). These behaviours are not necessarily caused by ASD but can also be attributed to other present disabilities or mental health problems. This makes it very difficult to assess these behaviours, to diagnose disorders and impairments and to create appropriate treatment plans.

In this thesis, two ways to deal with these problems are pursued. Firstly, an instrument was developed to diagnose ASD within persons with multiple disabilities. This instrument was designed to take into account the presence of both intellectual and sensory disabilities. Secondly, symptoms of ASD and symptoms of mental health problems were studied in an attempt to clarify the overlap in behaviours. The current study continues the work on an earlier developed instrument to diagnose ASD in people with a profound intellectual disability and deafblindness (Hoevenaars-van den Boom, et al., 2009). This instrument, Observation of Autism in Deafblindness (O-ADB) is a semi-structured observational instrument designed to differentiate people with and without ASD when they have a profound intellectual disability and deafblindness. The O-ADB appeared successful in its purpose, but not very practical in its use. Administration was labour-intensive and time consuming. Moreover, it was tested on only ten participants, half of them showing very clear signs of ASD and the other half of not having ASD. The O-ADB and was specifically designed for the most severely impaired individuals with regard to cognition and sensory disabilities. As such, this sample was not representative for the population of multiply disabled people and it was not suitable to diagnose the larger group with less severe levels of intellectual disabilities and persons without hearing impairments.

The present study adds to this previous research by further developing the assessment tool, making it suitable for a broader population and more usable in clinical practice. In addition, the overlap between ASD, disabilities and mental health problems were studied. The aims of the current thesis are as follows:

1. To develop an instrument to assess the presence of ASD symptoms, specifically designed to diagnose people with a combination of intellectual disabilities and sensory impairments. This instrument should be appropriate for people with moderate to profound intellectual disabilities combined with a visual impairment, with or without additional hearing impairments (deafblindness). Importantly, the instrument should not be stressful for participants, contain scoring criteria for ASD and show adequate psychometric properties.
2. To describe the overlapping and differentiating characteristics of ASD in people with a combination of intellectual disabilities and sensory impairments. An overview of overlapping behaviours and differentiating characteristics for disabilities, ASD and other common mental health problems should make it easier to determine the aetiologies of specific behaviours and behaviour problems and could make clinicians aware of common combinations of behavioural problems in persons with multiple disabilities.

6. Outline of this thesis

This thesis consists of two parts. The first part addresses the development of a valid and reliable diagnostic instrument for ASD in people with multiple disabilities. The second part addressed the behavioural characteristics of this population and the overlap with ASD and other mental health problems.

The first part of this thesis comprises four chapters. Chapter 2 "Autism spectrum disorders in people with sensory and intellectual disabilities: Symptom overlap and differentiating characteristics" is a review that describes the overlapping characteristics of individuals with intellectual disabilities, visual impairments, auditory impairments and ASD. This chapter served as the basis for designing the diagnostic instrument for ASD in this population.

Chapter 3 "A critical review of screening and diagnostic instruments for autism spectrum disorders in people with sensory impairments in addition to intellectual disabilities" gives an overview of screening and diagnostic instruments for ASD that are currently used in clinical practice. These instruments were reviewed on their psychometric properties and applicability for people with intellectual disabilities and sensory impairments.

Chapter 4 "Behavioural assessment of autism spectrum disorders in people with multiple disabilities" describes the development of an instrument that can diagnose ASD in people with intellectual disabilities and sensory impairments. The pilot study of this instrument, Observation of Autism in people with Sensory and Intellectual Disabilities (OASID), is presented, describing the results of a study with 18 participants.

Chapter 5 "Assessing autism spectrum disorders in people with sensory impairments combined with intellectual disabilities" tests OASID and its psychometric properties on a larger group of 60 participants with multiple disabilities. This chapter will also present the scoring procedure, reference norms and the interpretation of test scores.

The second part of this thesis consists of three chapters studying the characteristics of ASD in people with sensory and/or intellectual disabilities. This part describes which behaviours are differentiating characteristics of ASD from behaviours more typical for individuals with sensory and intellectual disabilities.

Chapter 6 "Mental health profiles of people with sensory and intellectual disabilities" focuses on mental problems that are prevalent in people with a combination of sensory impairments and intellectual disabilities. The presence of these mental health problems, such as stress, attachment problems, anxiety and mood disorders can further complicate the diagnostic process of ASD because of overlapping characteristics. In addition, the presence of ASD can influence the presence of these mental health problems. This chapter describes the prevalence of mental health problems in people with multiple disabilities and the effect ASD has on these problems.

Chapter 7 “Profiles of stereotyped behaviour in people with combined sensory impairments and intellectual disabilities” looks specifically at stereotyped and repetitive behaviours. Stereotyped behaviours appear frequently in people with multiple disabilities, regardless of ASD. This chapter zooms in on specific expressions of stereotyped behaviour and how they may differ between participants with and without ASD.

Chapter 8 “The relationship between stress and autism spectrum disorder in people with sensory and intellectual disabilities: Evidence from cortisol levels” studies stress reactions. Stress can influence the severity and expression of ASD symptoms such as stereotyped behaviour and social withdrawal. This study focuses on a biological marker of stress, the stress hormone cortisol, and how this is related to autistic behaviour in individuals with multiple disabilities.

Finally, our findings will be summarized and discussed in Chapter 9 “General discussion”. This chapter describes the main conclusions of this research project, its limitations and its implications for theory and clinical practice.

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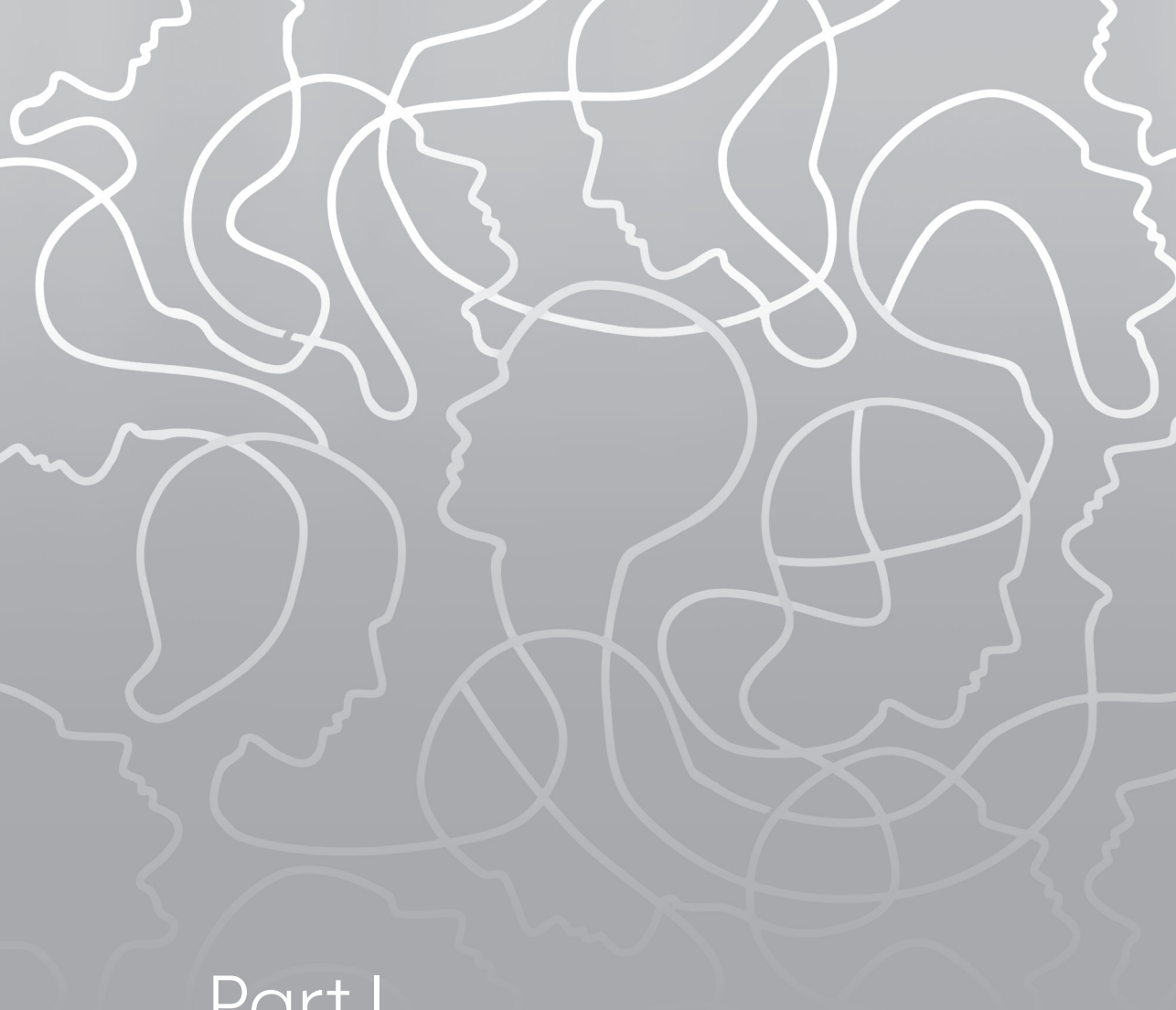
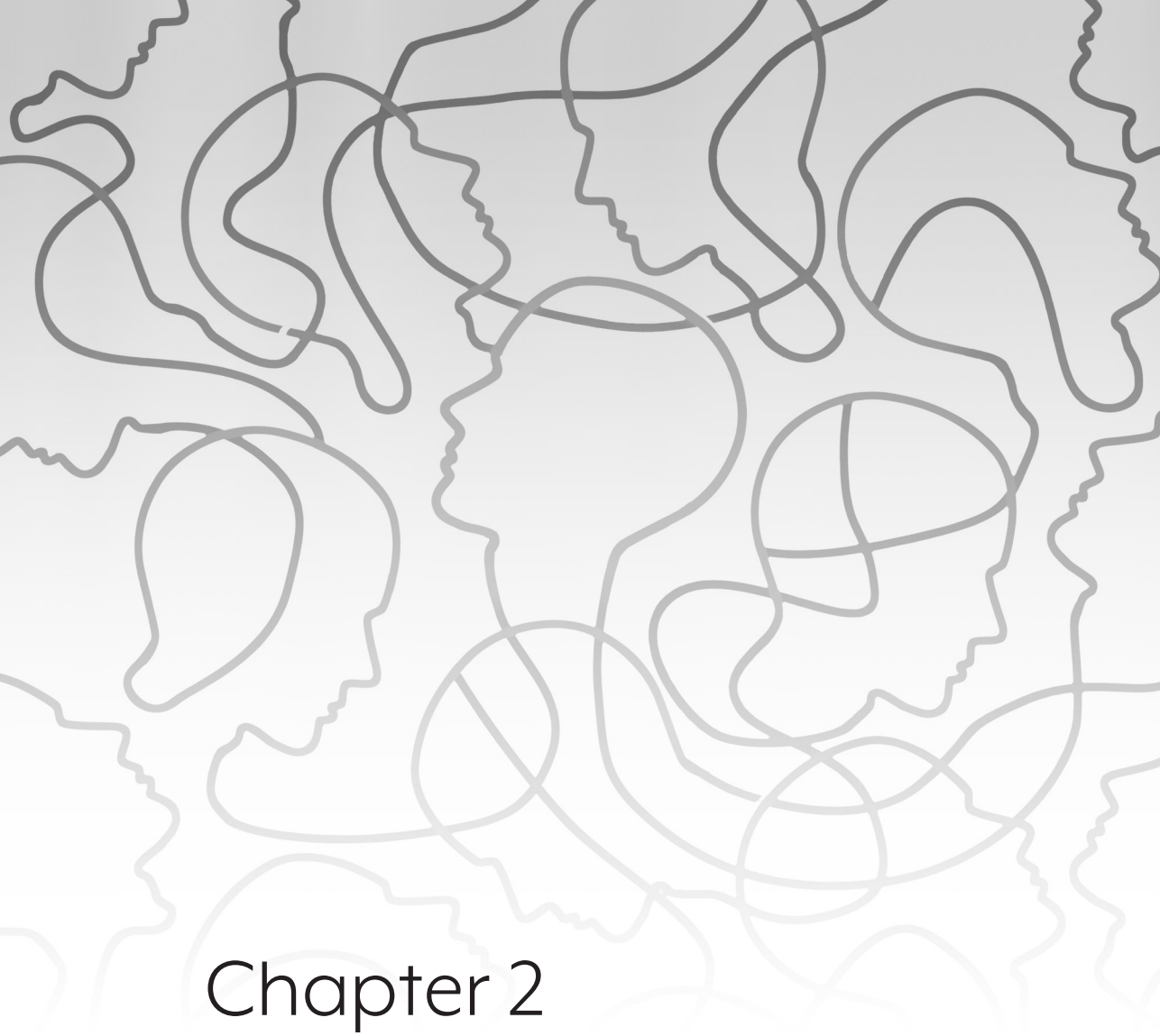


Part I

Assessing autism spectrum disorder in multiple disabilities



Chapter 2

Autism spectrum disorders in people with sensory and intellectual disabilities: Symptom overlap and differentiating characteristics

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1. Introduction

Autism Spectrum Disorders (ASD) are developmental disorders that people are burdened with for their whole life. They origin in childhood and are featured by restrictions in social and emotional development, communication, interests and motor skills (Nevid, Rathus & Greene, 2008). People with autism are characterized by three major deficits as defined by the most recent version of diagnostic and statistical manual of mental disorders (DSM-IV-TR). These deficits include qualitative impairments in social interaction, qualitative impairments in communication and restricted, repetitive and stereotyped patterns of behaviour (American Psychiatric Association, 2000). Behaviours within these main components of ASD may differ per individual because they are expressed in unique ways for each individual. Variations can be found in the way, the intensity and the perseverance with which the symptoms are expressed. Also the core characteristics may vary per individual. Where skills, interests and intellectual levels differ between people, so do the characteristics of autism, only the main problem areas remain the same (Frith, 2008). In the current chapter, not only autism as defined by DSM-IV-TR, but also all variations within the autistic spectrum will be included.

Several symptoms of ASD are not unique but also found in other groups of people with disabilities. Similar behaviours, overlapping symptoms, or even the exact same behavioural characteristics can be found in people with hearing disabilities (Knoors & Vervloed, 2011), visual impairments (Cass, 1998), intellectual disabilities (De Bildt, Sytema, Kraijer & Minderaa, 2005) and combinations of these impairments, such as deafblindness (Hoevenaars-van den Boom, Antonissen, Knoors & Vervloed, 2009). All three of the main components of autism that the DSM-IV-TR describes, are also found in non-autistic people with sensory and intellectual disabilities. Furthermore, the prevalence of ASD seems to be much higher in people with one or more of these disabilities. In the entire population ASD is estimated to occur in at least between 0,1 and 0,6 percent (Fombonne, 2003a, 2003b) and at most 2,64 percent (Kim et al., 2011). In people with intellectual disabilities reported prevalences are much higher, ranging from 4 up to 60 percent (Matson & Shoemaker, 2009). Without giving exact rates the prevalence of ASD and autistic features in people with sensory disabilities is reported to be much higher than in typically developing people (Brown, Hobson, Lee & Stevenston, 1997; Hobson, Lee & Brown, 1999; Jure, Rapin & Tuchman, 1991). It is an interesting question what cause this increase in prevalence when other impairments are involved. An obvious explanation could be a relationship between ASD and sensory or intellectual disabilities. An alternative explanation is an overlap of symptoms, but not of the underlying mechanisms, between autistic people without other disabilities and people with sensory and intellectual impairments. If the latter is the case, some people might be unfairly diagnosed as autistic when in fact

they are not. False positive diagnoses then causes the increase in prevalence of ASD in sensory, intellectually and multiply impaired people.

The overlap in symptoms between people with ASD and people with sensory, intellectual and multiple impairments interferes with the right classification of the behaviour of people with sensory and intellectual disabilities. Several authors stress that even though the symptoms are similar, the processes that underlie these symptoms are different for autistic versus non autistic people (Andrews & Wyvers, 2005; Hobson, 2005; Knoors & Vervloed, 2011). Nevertheless, when behaviours are the same, there is the risk that ASD is either missed or unjustly diagnosed. A wrong classification may lead to a wrong treatment plan, which is especially problematic if the treatment plan is counterproductive for the true underlying cause. A treatment is most effective if it tackles the cause of the behaviours. An example is the stopping of stereotyped movements. Whereas in the blind these are usually caused by a lack of stimulation from the environment (Van Dijk & Janssen, 1993; Warren, 1994) in people with ASD stereotyped movements can occur to get away from too much stimulation from the environment (Frith, 2003; Gense & Gense, 2005).

The current chapter will give a comprehensive overview of the overlapping symptoms between autistic and non-autistic people; it will elaborate on the categories that the DSM-IV-TR distinguishes as well as on the overlap within these categories for autistic and non-autistic people, it will describe the differences between the two groups and finally explain why a better differentiation is necessary.

2. Qualitative impairments in social interaction

The first characteristic of autism, according to DSM-IV-TR is defined as qualitative impairments in social interaction. These impairments can express through a variety of symptoms: problems in reciprocity and sharing of interests and emotions; impairments in non-verbal behaviours and impairments in joint attention, either in sharing, following or directing (American Psychiatric Association, 2000). All of these problems in social behaviours contribute to problems in the development of proper peer relations.

2.1 Reciprocity and peer relationships

Some children with ASD prefer doing things alone and might avoid all kinds of social play (American Psychiatric Association, 2000). Lack of reciprocity is also shown in an aversion to social touch and in problems with responding to your own name (Baranek, 1999). In young children impairments in this area are often expressed as inappropriate responses towards other people and being more interested in objects than people (Frith, 2003).

Autistic people may find it difficult to engage in peer relationships. However, they are not the only ones that have trouble in this area. A recent study about the popularity of deaf children showed that deaf children were less accepted and less popular than their hearing peers. This was explained by them being, amongst others variables, more withdrawn, less prosocial and worse at monitoring a conversation (Wolters, Knoors, Cillessen & Verhoeven, 2011), behaviours also typical for ASD in a hearing population.

People with intellectual disabilities show problems in the area of reciprocity and relationships too. Often, intellectual disabilities are caused by abnormalities in the brain. It is not surprising to find that these abnormalities cause problems in people's emotional and social behaviours. However, not everyone with serious intellectual disabilities has social or emotional problems; some of them are even overly interested in social contact. Reciprocity and engagement are definitely present while communicating with them (Frith, 2008). According to Wing & Gould (1979) one can spot the difference between impaired social behaviour in intellectually impaired people with ASD versus intellectually impaired people without ASD by looking at the severity of the social impairments.

The problems in reciprocity and developing relationships are not limited to people with ASD, and auditory or mental disabilities. In 1977 Selma Fraiberg described the development of blind children. She noticed that blind children do not reach out to their parents as much as their sighted peers do. This may appear as a lack of reciprocity, when in fact seeing a parent makes sighted children reach out. Blind children obviously lack this ability (Fraiberg, 1977). This explains their less frequent attempts in reaching out, without any relationship with reciprocity. Moreover, according to Fraiberg, the absence of reaching out could make parents less responsive to their children, restraining them in their development of relationships. She explains that in the sighted, the smallest amount of eye contact with a baby can make an adult talk or play with them (Fraiberg, 1977). When signals such as reaching out and making eye contact are absent, the development of reciprocity and relationships could be impaired because of this. In fact, because the care for a blind child is so much more challenging and reciprocal signals are easily missed, lack of vision may increase the risk of problems in attachment (Warren, 1994). However, Warren stressed that despite an increased risk, attachment problems can be avoided if the parents of a blind child respond appropriately. Assessing attachment highlights another problem, that is the reliability and validity of assessment instruments and procedures in children with disabilities. Attachment in sighted children is often tested by the strange situation method (Ainsworth & Bell, 1970) where a child's reaction upon reunion with its mother is assessed after it has been left alone or in the presence of a stranger. Children with visual impairments, especially blind children, may not notice the departure and reappearance of their mother and may therefore fail to respond like sighted children would do (Warren, 1994). In this case the perception

problems interfere with possible affirmations of attachment problems. The same problems occur when observing people whilst looking for signals of reciprocity or interest in other people. Because of a loss of sight children with visual impairment or blindness may not notice other people or other people's behaviour. In extreme cases they do not show any interest in their surroundings because of poor vision and direct all their attention to objects within arm's reach or to their own body. This is especially the case in deafblind children who have not only problems in vision but also hearing, the two distant senses. Their remaining senses (touch, smell, taste and proprioception) only function in nearby space, giving the impression that deafblind children are ego-centred. This ego-centeredness is however of a different origin than it is in ASD (Knoors & Vervloed, 2011).

2.2 Verbal and non-verbal social behaviours

In people with ASD, much verbal and non-verbal behaviour is impaired. This can express itself in to unnatural eye-to-eye gaze, a failure to correctly understand and execute facial expressions, atypical body postures and gestures to regulate social interaction. People with ASD often show less eye contact and fewer social smiles to others. They may also show problems in understanding facial expressions and the underlying emotions (Frith, 2003).

Non-verbal behaviours are very important in social communication and are used to make messages more clear. It's hard to imagine communicating without facial expressions, gestures, posture or understanding gaze direction. People with impairments miss a lot of these signals while communicating. In a visually impaired group it may be hard to distinguish autistic people from non-autistic people based on non-verbal behaviours. Non-verbal skills that come natural to people without impairments need to be taught specifically to people with visual impairments (Gense & Gense, 2005), for example by explaining gestures in a tactile way and in natural situations. So even though people with sensory impairments show problems in expressing themselves non-verbally, Gense and Gense (2005) do believe that many behaviours can be taught. On the other hand, in visually impaired people some behaviours may be impossible to teach. Making eye contact and following gaze direction are simply infeasible for people with visual impairments. One cannot expect them to show these behaviours. Since their impairments make some social behaviours impossible to execute, they may use other signs to show their social skills. A blind person will not look someone in the eye when interested in what they have to say, but they may aim their ears towards this person and will thus aim their face in another direction. This behaviour is inappropriate for someone with adequate visual abilities, but the visually impaired will orient with their ears more than with their eyes and it may even point to social interest in another person.

Another complication is that it is important to take into account the severity of intellectual disability when analysing a person's social behaviours. If mental and chronological age do not match, age inappropriate social behaviours might be seen. An example is that people with intellectual disabilities show few gestures and joint attention signs (Osterling, Dawson & Munson, 2002). On the other hand, people with mental retardation and autism responded to their name much less frequently than did people with mental retardation alone (Osterling, et al., 2002), making orientation after hearing ones name a characteristic that may help in differentiating autistic from non-autistic people.

When trying to differentiate autistic behaviours from behaviours due to multiple impairments, Hoevenaars-van den Boom et al. (2009) showed that even though social behaviours appear similar it is possible to differentiate autistic from non-autistic behaviours. They have found a significant difference between autistic and non-autistic deafblind children with profound intellectual disabilities in the areas of social and communicative behaviours in that these children showed and openness for contact and pleasure while in social contact (Hoevenaars-van den Boom et al., 2009).

2.3 Joint Attention and Theory of Mind

Autistic people have trouble sharing interests, emotions and activities (American Psychiatric Association, 2000). Related to this are problems in joint attention. Joint attention refers to the ability to share your attention, by looking where someone else is looking at and by sharing your own interests through pointing, gazing, or other non-verbal behaviour (Frith, 2003). People with ASD may fail to share their emotions, feelings and thoughts but they also can have problems in sharing attention, which is expressed in their inability to follow a pointing finger or the direction of a gaze. This is interesting, because in non-autistic children, both pointing and following a finger or gaze not only relates to the object itself, but also to the other person's feelings and interests for this object. Autistic people fail to point or gaze and follow somebody else's pointing or gazing because they fail to understand other people's interests in the objects (Frith, 2003).

Joint attention is often said to be a precursor of theory of mind (ToM) (Charman et al., 2000). Someone has a ToM when they are capable of attributing a mental state to themselves and to others (Premack & Woodruff, 1978). ToM is one of the most important constructs regarding a deeper understanding of ASD (Noens & Van Berckelaer-Onnes, 2004) and can explain many of the symptoms of ASD. Not only social behaviours as joint attention, but also symbolic play and language problems such as echolalia and reversal of pronouns can be attributed to not having a ToM (Brown, et al., 1997; Pérez-Pereira & Conti-Ramsden, 1999). In simple terms, it refers to being able to realize what people think, feel and want (Frith, 2008). Having a ToM also entails understanding irony and non-literal language, and can therefore also explain

some of the deficits in communication. Another aspect of ToM is being able to take someone else's point of view or perspective. Perspective taking is often measured with false belief tasks, such as the Sally-Anne-task (Hill & Frith, 2003). Baron-Cohen and colleagues used this task to measure false belief in autistic children by showing them two dolls, one called Sally and the other called Anne. They played out a story where Sally had a marble in her basket. Sally left and Anne put the marble in her own basket. By asking children questions on where the marble really is and where Sally would think the marble is, perspective taking can be measured (Baron-Cohen, Leslie & Frith, 1985) and give an indication of the development of a ToM. This is a typical false belief task, but many variations have been used since then. Where in sighted children ToM is tested with a false belief task such as the Sally-Anne task (Baron-Cohen, et al., 1985) or joint attention tasks, these tasks may not be applicable sufficiently enough for children with visual impairment. In addition, joint attention is often measured with gaze direction or pointing, something that blind children are for obvious reasons incapable of showing and is limited in visually impaired children. Peréz-Pereira and Conti-Ramsden (1999) do point out that it is not the pointing or gazing what matters, it is the function of this pointing that is of interest. To measure this, things need to be seen from a blind person's perspective.

Seeing things from a blind person's perspective is difficult when it comes to ToM tasks. Conventional ToM tasks have been carried out on people with impaired vision, showing that visually impaired children invariably performed worse than sighted children. McAlpine and Moore did a false belief task using containers with unexpected contents and asked what another person would think was in it. Many of the blind children failed this task, even though sighted children are able to do this at a younger age (McAlpine & Moore, 1995; Pérez-Pereira & Conti-Ramsden, 2005). A similar study by Minter, Hobson and Bishop (1998) compared visually impaired with sighted children of the same verbal intelligence, and showed similar results. In their first experiment, they did a similar task as the container task McAlpine and Moore used. They used a warm teapot, filled with sand instead of tea. Whereas almost all sighted children were able to pass this task, almost half of the visually impaired children failed to answer false belief questions such as: "What did you think was in here?" and "What would he/she think is in here?" The authors note that blind people may have less experience with hot teapots because of the extra danger their lack of vision provides. Their second experiment was done with three boxes, where the participants helped the experimenter hide a pencil for another experimenter and false belief questions were asked. Again, the visually impaired children performed worse than the sighted, but much better than on the previous task. The authors think this was because they were more involved in this task, because they helped with the hiding (Minter, Hobson & Bishop, 1998). These findings show that children with visual impairments do worse on conventional ToM tasks than do their hearing peers. One could assume that blind

children do not have a ToM, or develop it slower. However, other findings indicate that visually impaired children can pass a ToM task, given an adapted task. In line with the notion that things need to be seen more from a blind person's perspective, it could be possible that visually impaired people have just as much a ToM as sighted people do; it's only measured in the wrong way. Peterson and her colleagues confirmed this. They state that blind people may very well rely on completely different features of an object than sighted people do in order to decide what another person thinks about an object (Peterson, Peterson & Webb, 2000). They tested if this was true by adapting frequently used false belief tasks. For example, they have changed the famous Sally-Anne task to a Sally-Bill task. In this task, there were no dolls or pictures of children with baskets and marbles, but it was a purely narrative story. The experimenters performed four ToM tasks, including similar tasks to the container tasks, a location change task and a story. On average, the children performed best on the Sally-Bill task, 73% of the children passed this task. Despite this result and the careful adaptation of test methods, test methods were not found to be a factor influencing ToM development. Degree of visual impairment was also not found to be of influence in developing a ToM, age was the only significant factor these authors found (Peterson, et al., 2000). These are some interesting findings, firstly because they indicate that visually impaired people can show signs of having a ToM, secondly, because the question is raised where the difference lies between visually impaired and sighted people. According to Minter et al. (1998) tasks need to be adapted to the qualities of visually impaired but Peterson & Siegal (2000) did not find a difference between tasks they used. Brambring and Ashbrock (2010) elaborated on this question. They used a large variety of different tasks that did not require vision and found that performance was better than with traditional tasks but the blind children were on average 19 months older when they were able to perform the same tasks as sighted children. A more recent study (Pijnacker, Vervloed & Steenbergen, 2012) found that children with varying levels of congenital visual impairment when compared with sighted children matched on age and verbal intelligence, had a similar performance on advanced ToM stories (second order false belief, that is beliefs about beliefs) and non-literal stories. Despite a limited access to visual information during interactions, children with congenital visual impairment can develop an effective ToM.

Peterson has not only studied ToM in visually impaired children, but also in deaf children (Peterson & Siegal, 1995, 2000). It looks as if deaf children are strongly delayed or even impaired in their ability to have a ToM. In their 1995 study, Peterson and Siegal tested the Sally-Anne paradigm on several deaf children who were able to communicate in sign language. Even though hearing children with or without intellectual disabilities can pass this task around a mental age of four, only 35% of these deaf children were able to pass at a mental age of 8. Furthermore, these results were similar to results of people with ASD, but worse than the performance of children

with Down syndrome. Notwithstanding the lack of ToM, these deaf children were not autistic as they did not show any of the other characteristics of ASD (Peterson & Siegal, 1995). According to Peterson and Siegel deaf children lack a ToM, because of the lack of understanding the communicative signals of others. It also appears that deaf children, especially those with hearing parents, communicate less at home than hearing children. On the one hand this is because a deaf child does not hear nor understand spoken language and on the other hand because their parents are not very fluent in sign language as an alternative for spoken language (Vaccari & Marschark, 1997). A direct consequence of the lower frequency of communication is that deaf children also communicate less about mental states, feelings and thoughts, which hinders the development of a ToM (Peterson & Siegal, 1995, 2000). This idea was supported in a more recent study that assessed the amount of communication in play sessions for pairs of hearing mothers with their deaf children and compared them to hearing mothers with hearing children. They found that these signing mothers of deaf children do not necessarily communicate less than mothers of hearing children, but they do communicate less about mental states. Additionally, a relationship was found between the amount of communication about mental states of mothers of deaf children and the performance on false belief tasks of their children (Moeller & Schick, 2006). Despite the similar way in which the lack of ToM expresses itself in people with ASD and in deaf, the cause is different. In children who are deaf it is often attributed to a lack of communication about mental states, thoughts and feelings, whereas in ASD it is caused by inability to take someone else's perspective.

Another possibility for why hearing children outperform deaf children on ToM tasks could be that deaf children do have a ToM but only fail on certain aspects related to ToM and conventional tasks fail to test these aspects. Where normally false belief tests and variations of this are undertaken, a recent study addressed other aspects of ToM as well. Ketelaar, Rieffe, Wiefferink and Frijns (2012) assessed deaf children that have received a cochlear implant (CI) at a young age, and compared them to hearing children. They tested other aspects of ToM than false belief, which are the understanding of other's intentions and others desires. The tasks were similar to false belief tasks, only instead of asking what someone would think or believe, it was asked what another person intended to do with an object (after failing this action) or what someone would want to eat (after showing them pictures of food they liked). It appeared that the deaf children and hearing children performed equally well on the intention tasks, but the hearing children outperformed the deaf on false belief tasks and on the desire tasks (Ketelaar, et al., 2012).This study indicates that deaf children may possess some abilities related to a theory of mind. It should be noted, however, that this study only included children with a CI. These children thus had some hearing abilities, though different from hearing children. The study did not include a group that was completely deaf and so conclusions about completely deaf children cannot be drawn.

When children are completely deaf there is, however, still the possibility that, as seen in the visually impaired group, testing methods are not adequate for them. Peterson and Siegal (1995) tried to make their intentions more clear in their false belief questions. They reasoned that someone with limited experience in conversation might expect that the experimenter just wants them to tell the location of Sally's marble, when they ask "Where will Sally look for her marble?" For this reason they altered the question to "Where will Sally first look for her marble?" By adding the word "first" they more clearly imply that they are looking for what sally thinks instead of where the marble really is. This slight alteration improved the deaf children's performance slightly, but not enough to overcome differences in ToM development (Peterson & Siegal, 1995) as the different tasks in the study by Ketalaar et al. (2012) did. Peterson and Siegal only investigated false belief, though, whereas Ketelaar et al. addressed other aspects of ToM and tested children with a CI who do have some hearing abilities, instead of children who are completely unable to hear. The question still remains whether a more appropriate methodology for deaf children could increase their scores on conventional ToM tasks and more research has to be done in order to clarify this.

Finally, people with intellectual disabilities often show ToM impairments as well. Typical developing children start to solve ToM tasks around the age of four to five years of age. A general characteristic of people with intellectual disabilities is that they have mental ages not corresponding to their chronological ages. If mental age is below five, which is the case in profoundly and severely intellectually disabled people, and sometimes also in moderately intellectually impaired people they will probably fail ToM tasks irrespective of their chronological age (Kraijer, 2004). Interpretations of ToM tasks should be done cautiously, when intellectually disabled people likely fail this task unrelated to the presence of ASD, to prevent unnecessary suspicion of ASD.

3. Qualitative impairments in communication

Qualitative impairments in communication form the second criterion that is defined in DSM-IV-TR, and this can refer to the use of language but also to problems in make belief or imitative play. When it comes to language one can find a lack of or delay in language, but also use of repetitive or idiosyncratic language. Autistic people may also find it troubling to initiate and maintain a conversation with others (American Psychiatric Association, 2000).

3.1 Making Conversation

Language is something people use for communication, and so the willingness to communicate is related to their use of language (Frith, 2003). Despite possible technical problems in language the low desire for communication is one of the aspects of ASD

that is mentioned in the DSM-IV-TR, that is not only problems in initiating and maintaining a conversation with others but also a lack of an internal willingness or desire to communicate (American Psychiatric Association, 2000). If people with ASD are simply uninterested in communication, they will not put effort in initiating a social conversation spontaneously. This lack in willingness to communicate also contributes to the language problems found in ASD.

Initiating and maintaining a conversation can be difficult for people with sensory and intellectual disabilities too. The presence of others may go unnoticed for people with visual impairments, and communicative signs may be missed because of blindness or deafness. It has been found that deaf children communicate less with their hearing parents because of their poor skills in spoken language and their parents' poor sign language skills (Vaccari & Marschark, 1997). In people with intellectual disabilities conversational skills may be worse than expected based on their chronological age, moreover, their initiations to communicate may be different, inadequate or even awkward.

Even though all of these impaired groups may show impaired conversation making skills, there are differences between autistic and non-autistic people. An example derived from a deaf population shows that despite other problems in the field of communication, such as monitoring a conversation and pragmatic use of language, non-autistic deaf children are not different from their hearing peers in initiating and maintaining a conversation (Wolters, et al., 2011). But even though deaf children without ASD don't seem to have problems in initiating and maintaining a conversation, they still differ from their hearing peers in pragmatics and monitoring, hampering their conversational skills nevertheless. On the contrary, the impaired conversational skills in autistic people lie in the area of the initiation and maintenance of a conversation (American Psychiatric Association, 2000). It also appeared that one of the areas in which the autistic and non-autistic children with deafblindness and profound intellectual disability differed significantly from each other was the openness and willingness to take initiatives for contact (Hoevenaars-van den Boom, et al., 2009). It is evident that conversation looks different for people with sensory or intellectual impairments versus people without impairments, and conversation skills are hampered by their lack of sensory and intellectual abilities. The difference with autistic people shows itself in the interest for this contact. Non-autistic sensory and/or intellectually impaired people still look for opportunities to make this contact or respond to other people's efforts to make contact, while people with autism lack the interest for this contact.

3.2 Language

Besides a lower interest in communication than people without ASD, people with ASD show some technical language impairments as well. Some autistic people do not speak at all and in others the development of language can be seriously delayed or

altered (Frith, 2003). Furthermore, it appears that joint attention and imitation behaviours, which are known to be impaired in ASD, can predict language abilities (Charman, et al., 2000), which raises the question whether language is directly or indirectly related to ASD. In addition, ToM can be involved as well; one needs to know that one can influence others with their language and how to do so. Typical ASD language problems include direct or delayed echolalia, reversal of pronouns and lack of understanding of emotional meaning in language. People often describe it as 'robot-like' (Nederlandse Vereniging voor Psychiatrie, 2009). People with ASD often interpret the meaning of words literally. The literal meaning of a word does not change over contexts, but the figural meaning does. This is especially vivid in jokes, metaphors and irony. This may also be due to the previously mentioned problems in ToM. Being unable to understand what people mean, people with ASD interpret the words incorrectly (Frith, 2003). A review about language and communication in ASD confirmed this idea by concluding that the language and communication problems are caused by processing problems when interacting with other people (Tager-Flusberg, Paul & Lord, 2005).

People with intellectual disabilities show delays in language as well as atypical language skills that can easily be confused with ASD. A study about the language abilities of a group of autistic children showed that there was a relationship between language abilities and IQ (Tager-Flusberg, et al., 2005). This study was done on autistic people only, but it is a rather expectable finding, even within people without ASD. It makes sense that the language abilities of someone with an intellectual disability are delayed as compared to peers with the same chronological age. This may be confused with the language deficits found in ASD, when in fact they are due to their intellectual disability. For this reason, we should not immediately attribute language issues in people with intellectual disabilities to ASD.

Deaf and people with hearing disabilities often show delays in acquiring language, but can also show peculiar uses of words (Knoors & Vervloed, 2011). Even delays in developing sign language are found for this is often not fully learned until children go to a school for the deaf. Parents are not fluent signers and fail to teach children the full scope of signs they could learn from a signer that is fluent (Vaccari & Marschark, 1997). Atypical language development can also be found in the blind. Without seeing things to potentially talk about, language is centred around other experiences in the blind compared to sighted people (Warren, 1994). Children with congenital visual impairment have been shown to have difficulties with the use of language for pragmatic and social purposes, while structural language (e.g. articulation, grammar, vocabulary) was good or even superior (James & Stojanovik, 2007; Tadić, Pring & Dale, 2010). This delay or odd language use can be confused with what is found in autistic individuals. However, this language delay may be corrected if it is taught in the right way. It's important to realise that when a child misses its vision, they need to get stimulation through the other senses which affects their understanding of the meaning of words (Warren, 1994).

Several language problems that are found in autistic individuals are also found in people with other impairments. A typical example is echolalia, which is also found in visually and intellectually impaired people (Wing & Gould, 1979). Echolalia is the apparently useless repeating of words or phrases, either immediately after they were spoken or after some time. Even in typically developing children, echolalia is sometimes used to learn language (Gense & Gense, 2005), so it's not surprising to find this in people with intellectual disabilities who may have a mental age comparable to when it is normal to use this type of speech. According to Schlesinger, it can be expected for a typically developing 20 month year old to repeat words to indicate more than one (e.g. "apple, apple" for "two apples") (Schlesinger, 1982). Another author described a child of 15 – 18 months old who often repeated her mother's words to learn the names of objects, but also to practice these words (Dore, 1974). It can therefore be expected that a person with a mental age below two years of age to still show signs of echolalia. These examples consist of people with typically developing vision, but blind children use echolalia even more than typically developing children. In part echolalia serves as a means to stay in contact with people that cannot be seen, but it is also suggested that blind children practice their language by using echolalic speech. In this way they try to get a grip on the meaning of words in the absence of vision (Pérez-Pereira & Conti-Ramsden, 1999). Extra practicing of words and phrases also results in more imitations and use of routines in speech. In the blind, one will also find egocentric speech and reversal of personal pronouns(I, you, he etc.), and improper use of deictic terms (e.g. here, there) which could be mistaken for autistic language, because of its atypical nature. Reversal of personal pronouns, which is found in about a third of the speech of blind children and egocentric speech may be caused by a lack of ToM, resulting in these impairments (Brown, et al., 1997). However, a logical explanation can also be based on the visual impairment. The direction of speech and who is speaking to whom determines which personal pronoun is used. Absence of vision makes it difficult to understand that the "I" who is speaking about the self is suddenly referred to as "you" by a person who became the "I" instead. 'Here' and 'there' are relative terms depending on ones spatial position. Without sight it is hard to adopt an allocentric position, most blind people use an egocentric position in processing spatial information. For instance, in way finding one cannot use landmark information to guide people who are blind, because they cannot see these landmarks. Instead one has to give route information related to the blind person's body position in space (Pérez-Pereira & Conti-Ramsden, 1999).

3.3 Imitation and make-belief or symbolic play

Finally, imitative and make-belief play are impaired in people with ASD according to the DSM-IV-TR. People with intellectually disabilities normally show delays or absence of imitation too. In one study, the experimenters showed intellectually disabled

participants an action that could be done with an object, afterwards they asked the participants what could be done with the object. All participants with intellectual disabilities had trouble recalling what could be done with the object. Participants with intellectual disability and ASD performed the worst (Charman, et al., 1997)

Symbolic play can be troubled in people with intellectual disabilities as well. Wing, Gould, Yeats & Brierly (1977) showed that even though only two people of their sample of intellectually disabled people showed the full autistic syndrome, more than half of their participants showed problems in symbolic play. These problems were either characterized as stereotyped play that was a persevering repetitive copy of other's play or no symbolic play at all, but just repetitive manipulations of a part of an object. Despite the fact that only two of their participants had an ASD diagnosis, many showed autistic features. In the group that was able to show symbolic play (43 of 108 participants), only two participants had slight autistic features (Wing, et al., 1977). This finding shows that many intellectual disabled people show impairments in symbolic or make-belief play, and this can therefore not be used as a differentiating characteristic of ASD versus no ASD in this group.

When these people with intellectual disabilities have an additional sensory impairment, problems in symbolic play and imitation can become more evident. It is reasonable to think that people with impaired vision or hearing have more difficulties in imitating because they are less able to perceive actions of others, than people without these impairments. Similarly, symbolic play can be affected. People have less modalities to perceive a toy with, and therefore also see less ways in which they may use it. Combined with an intellectual impairment they can also have troubles in understanding the function the object is intended to have.

Lack of symbolic play was demonstrated to be related to abnormalities in language development that are typical of ASD, such as repetitive speech (Wing, et al., 1977). Similar to many of the impairments in ASD that were discussed, this too can be attributed to a lack of ToM. According to Brown et al. (1997) ASD is characterized by problems in ToM, symbolic play, and context dependent language. Shared features of these three skills in childhood are: 1) there has to be a communication pattern between parent and child regarding feelings and thoughts; 2) one has to see and understand the direction of someone else's attitudes towards a shared world; and 3) feel inclined to identify oneself with this shared world. People with ASD have problems with all three features. Children who are deaf encounter problems with the first feature. They are offered less ToM related language. Children who are blind have trouble with the second feature and subsequently children who are deafblind have trouble with the first and second feature.

4. Restricted, repetitive and stereotyped patterns of behaviour

As the last of three important characteristics, the DSM-IV-TR mentions restricted, repetitive and stereotyped patterns of behaviour, interests and activities. This can refer to motoric stereotypies or mannerisms, preoccupations with objects, parts of objects or interests, or their inflexibility in deviating from routines (American Psychiatric Association, 2000).

4.1 Stereotyped use of objects

Uta Frith confirms that autistic people are often very interested in details, which may appear as restricted interests to others and that routines and repetitions are also of importance for them (Frith, 2003, 2008).These behaviour can be explained by the central coherence theory. This theory poses that autistic people have a weak central coherence, meaning that they have the tendency perceive objects and situations in parts rather than perceiving the whole picture or combine information to holistic patterns (Frith, 2008). As a consequence information is often processed out of context (Hill & Frith, 2003). This theory explains the focus on details, but possibly also the need for repetition and routines shown by people with ASD. The ability to generalize parts to the whole keeps situations similar and predictable, and therefore less frightening. If one misses this ability then a coping mechanism is to stick to routines in order to keep situations predictable and safe. If preformed to the extreme these routines become stereotyped behaviours.

Repetitive and stereotyped use of objects is not only seen in autistic people but also in people with intellectual disabilities. In a study where 108 children with severe and profound mental disabilities were included less than two percent suffered from ASD. However, repetitive routines and stereotyped play were found in 60 percent of this group with a mental age below 20 months (Wing, et al., 1977). Also in children who are blind strong interest in parts of objects and repetitive use of objects can be seen. Mainly this is the result of the blindness-specific constraints on the use of play material that require visual-manual skills. Blind children, when playing alone, prefer toys and materials that produce distinctive tactile or auditory effects (Tröster & Brambring, 1994). Toys are often articles of daily living and objects in their surroundings such as spoons, walls and furniture. Activities are often aimed at making noise (Preisler, 1993; Tröster & Brambring, 1994).This behaviour is thought to be a way of getting hold on the function of an object and in contrast to children with ASD this behaviour can be relatively easily stopped or interrupted.

4.2 Self stimulation

Finally, autistic people show stereotyped movements with their own bodies or parts of their body. These are often thought to be self-stimulatory. Stereotyped movements can be performed with every body part but often involve the hands or walking (Goldman et al., 2009; Militerni, Bravaccio, Falco, Fico & Palermo, 2002) and sometimes become self-injurious (Bodfish, Symons, Parker & Lewis, 2000; Van Hasselt, Hersen, Egan, McKelvey & Sisson, 1989). These movements occur in other developmental disorders as well (Goldman, et al., 2009; Militerni, et al., 2002), but are especially common in ASD. According to Kraijer (2004) self-stimulatory behaviours are often caused by lack of stimulation from the environment. In these situations people use their own bodies to provide themselves with the stimulation they need at that moment. He adds to this that the amount of self-stimulatory behaviour and also intensity and severity, that is whether it is self-injurious, is related to the level of functioning. The lower the functional level of the person, the more the self-stimulatory behaviour increases in amount and severity (Kraijer, 2004).

Stereotyped behaviours occur in people with visual impairments as well. Typical stereotyped behaviours in people who are blind are body rocking, head shaking, eye poking and hand flapping Because these behaviours often occur in the blind, they are sometimes referred to as 'blindisms' (Gense & Gense, 2005; Warren, 1994). Actually this term is not entirely correct, because these stereotyped behaviours are not unique for people who are blind; mannerisms would be a better term. Body rocking and head movements, for instance, are typical examples of behaviours that can be seen in people with visual impairment, intellectual disabilities and ASD (Fraiberg, 1977; Gense & Gense, 2005; Warren, 1994). Stereotyped behaviours were seen in nearly all (Jan, Freeman & Scott, 1977) and in all (Tröster, Brambring & Beelman, 1991b) blind children, but in children with visual impairment the prevalence is still 10-45% (Jan, et al., 1977). There also seems to be an age dependency in stereotyped behaviours in blind children. In the first two years stereotyped behaviours increase in frequency to decline thereafter (Tröster, Brambring & Beelman, 1991a). Stereotyped movements are also found in people with multiple disabilities. Heather Murdoch (1997) suggests that stereotyped behaviours may be a part of normal motor development but that in people with multiple disabilities, these behaviours do not develop further. In a typically developing child, repetitive behaviours appear as well but develop into conscious movements later on, whereas in people with multiple disabilities they may remain repetitive movements. Trying to stop these behaviours may hamper the development of other motor activities or communicative signs (Murdoch, 1997).

Whereas stereotyped movements in people without ASD are part of a normal development, in people with ASD they are part of their syndrome. Gense and Gense (2005) believe that the differences between these behaviours in visually impaired

people with or without ASD can be found in the severity and perseverance of this behaviour. People with ASD show higher intensities and stronger persistence in stereotypical behaviours (Bodfish, et al., 2000; Gense & Gense, 2005). Similar to the behaviours in the intellectually disabled, this could be due to a lack of external stimulation. Especially in the blind, where stimulation from visual input is missing, self-stimulatory stereotyped movements could provide the necessary sensory stimulation (Warren, 1994). Another difference between people with ASD and people without, is that stereotyped behaviour can more easily be interrupted or stopped in people with visual impairments alone (Gense & Gense, 2005). Sometimes not much more has to be undertaken than making the blind person conscious of these unconsciously executed stereotyped behaviour patterns.

5. Differentiation: Why and how?

5.1 Overlap and differences

The overlap in symptoms between autistic and non-autistic people with sensory and intellectual disabilities must be clear after reading this chapter. The diagnosis of ASD is usually based on behavioural characteristics and these can be similar in autistic and non-autistic people with additional impairments. An additional problem is that, although instruments are available for people with intellectual disabilities (Kraijer & De Bildt, 2005; Matson & Boisjoli, 2008), most of the current test instruments do not have separate norms for people with sensory and/or intellectual disabilities. No valid instruments are available for deaf people according to Jure and colleagues (1991), nor for visually impaired people (Hoevenaars-van den Boom, et al., 2009). The overlap in symptoms and trouble in diagnosis cause a distorted representation of ASD in people with sensory, intellectual and multiple impairments. Some people are diagnosed as autistic when they are not, while others do not get the autistic label when they should. So there is both an overdiagnosis (Andrews & Wyver, 2005; Cass, 1998) of ASD in this group, meaning that more people are diagnosed as autistic than necessary because of these overlapping symptoms, as well as an underdiagnosis (Jure, et al., 1991; Roper, Arnold & Monteiro, 2003). In a group of deaf children, for example, the diagnosis of ASD was established significantly later than in a group of hearing children. Autistic behaviours were probably missed because of an earlier diagnosis of hearing impairments or other developmental disabilities (Roper, et al., 2003). The main problem in assessment of ASD can be attributed to a diagnostic overshadowing bias. The diagnostic overshadowing bias was first described for people with intellectual disabilities and is the tendency of clinicians to overlook symptoms of mental health problems in this group and attribute them to being part of "having an intellectual disability" (Mason & Scior, 2004). In the presence of mental

retardation it seems that the diagnostic importance of abnormal behaviour decreases. Blindness, deafness or deafblindness all might add an extra overshadowing bias next to intellectual disability, leading to either false positive or false negative diagnoses of ASD in people with these disabilities.

Despite the obvious similarities between autistic and non-autistic people with sensory and intellectual disabilities, this chapter also outlines that even though the symptoms appear the same, sometimes subtle difference can still be found. This may be due to the possibility that underlying processes of the behaviours are different for autistic and non-autistic individuals (Andrews & Wyver, 2005; Cass, 1998, Knoors & Vervloed, 2011). If attempted, a differentiation can thus be made by studying the subtle differences and underlying causes. A couple of years ago, this was done by making a valid instrument to diagnose ASD in people one of the most challenging combination of disabilities, namely deafblindness and profound intellectual disabilities. Hoevenaars-van den Boom and colleagues (2009) were able to confirm the huge overlap in behavioural symptoms between autistic and non-autistic people, but were also able to successfully distinguish the autistic from non-autistic people with their approach that was suited to the developmental level of the participants. They found that differences in this group can be found in the social communicative field, mostly in openness for contact, reciprocity and joint attention and communicative functions. It is clear that when using a careful and sophisticated approach, a distinction can be made between autistic and non-autistic people with sensory and intellectual disabilities.

5.2 Interaction, treatment and teaching

A fair diagnosis of ASD, or no ASD, is very important for the treatment and interaction with people with sensory and intellectual disabilities. An ASD diagnosis or a lack thereof will affect how a person will be treated, as autistic or not. If a child with ASD is placed in a setting where his or her ASD goes unrecognized, the clinicians and care takers might fail to respond to the needs of this person (Roper, et al., 2003). An important example of why recognition of ASD is so important is the treatment of stereotyped behaviour. Stereotyped movements can be a way to reduce stress (Frith, 2003; Gense & Gense, 2005). In someone with no ASD but with blindness or deafblindness, this behaviour is usually caused when the person does not get enough stimulation from their environment (Van Dijk & Janssen, 1993; Warren, 1994), whereas in persons with ASD stereotyped behaviours can be a way to escape from overstimulation or as a way to ensure the optimal level of arousal. In both cases the way to treat stereotyped behaviour will be different, give extra stimulation or reduce overstimulation, respectively. A valid diagnosis would be very helpful in cases where clinicians or parents have to decide what kind of intervention to give. If it is clear whether someone has ASD or not treatment and interaction can be adjusted. Someone with ASD needs a more structured environment, and needs clear

instructions when something needs to be done. In someone with ASD, things need to be re-explained in new situations, because of their difficulties in generalizing (De Bildt, et al., 2005). It also seems that the earlier we are aware of ASD the better. People with ASD need to be approached in way that is accommodated to their needs (Roper, et al., 2003), and for the wellbeing of the child, it is best if this is done as soon as possible. A recent meta-analysis on intensive early intervention programs for ASD shows that programs that intervene early are most effective and can produce changes in the area of language and adaptive behaviour (Peters-Scheffer, Didden, Korzilius & Sturmey, 2011). Adaptive behaviour was also found to increase as well when additional behavioural treatments were given to children with ASD and intellectual disabilities (Peters-Scheffer et al., 2008). These studies showed that if ASD is treated, successful results can be achieved.

As can be seen throughout this chapter, people with visual impairments show many behaviours that are similar to ASD, such as the lack of understanding of social situations, ego-centeredness, and lack of understanding gestures and facial expressions. But, according to Gense and Gense (2005), these behaviours may still be taught. Teaching appropriate behaviours is especially important, because inappropriate behaviours may interfere with regular social interactions (Warren, 1994) depriving disabled children of these otherwise valuable experiences. And whereas for non-autistic people without visual impairments these behaviours are implicitly learned, in non-autistic visually impaired people, they need to be explicitly taught. With the right type of education, visually impaired people may still learn to interpret social situations, read and understand gestures and facial expressions and learn to play with others (Gense & Gense, 2005). This was also found for two severely mentally disabled deafblind young men, of whom the social interaction became significantly better after tailored training sessions (Van Hasselt, et al., 1989). Although this was only a small study with two participants, it does indicate what a specialized training can mean for children that are not restrained by ASD. The same applies to language. When a delay in language is caused by a lack of seeing things to talk about, parents need to offer more tactile or auditory stimuli (Warren, 1994). Basically, it is important to take into account everything that singular or multiple disabled people lack. When sensory and intellectual impairments are involved, one needs to try and substitute the missing modality for others as much as possible.

6. Summary and Conclusion

Many characteristics of ASD seem to overlap with characteristics that are naturally present in people with sensory disabilities, intellectual impairments or a combination of disabilities. The characteristics appear the same whether ASD is present or not, which makes it difficult to make a valid diagnosis of ASD in this group. All of the criteria that are used in DSM-IV-TR to define ASD are, to some extent, also present in people with one or more of these disabilities. However, if one would look closer to these criteria, and the way they are expressed within people with sensory and intellectual impairments, slight and subtle differences can be found. There are differences in the way the symptoms express themselves, the severity of the symptoms and the underlying causes for the behaviours. Problems also occur in methodology. Paradigms that are used to assess problems that are related to ASD, such as ToM tasks, fail to be successful in differentiating people with sensory or multiple impairments. This overlap and these problems in methodology make it a major challenge to diagnose ASD within people with sensory and intellectual disabilities.

The slight differences in the way symptoms are expressed show that a distinction between autistic behaviours and non-autistic behaviours can be made. Making this distinction is very important to do, because the needs of people with ASD differ very much from people without ASD. To make sure the needs of every individual are met, people should be diagnosed in the right way. This is especially important for those groups with problems in communicating their wants and needs. In order to do this, subtle differences need to be taken into account. Up until this day, no instrument is suited to diagnose ASD or assess autistic behaviours within multiply impaired people. Ideally, a new way to assess autistic behaviours in sensory and intellectually disabled people that takes into account all the difficulties that assessing this group brings forth will be developed. An instrument that can make accurate diagnosis in people with multiple disabilities should account for all the overlapping symptoms and differences that have been described. First of all, intellectual disabilities should be taken into account. Some behaviours that are typical for ASD in people without intellectual disabilities can be simply explained by a person's mental age or shortcomings in intellectual abilities. An example of this is theory of mind, and related to that joint attention, symbolic play and language abilities, that do not develop until a certain age. If an intellectually disabled person has not reached a sufficient mental age, these behaviours should not be used to assess ASD. Secondly, it's important to realise that sensory disabilities withhold a person from perceiving objects and situations the same way a person without sensory disabilities would and may follow a completely different path. When someone is visually impaired or blind, eye contact, following gaze and sharing attention through pointing cannot be used as differentiat-

ing characteristics. Furthermore, it's important to take into account that a person may not always be aware of the presence of objects or people, so failures to respond like a person without ASD can be caused by being unaware of their presence in the first place. Similar precautions should be made for deaf people, who are unable to respond to calling their names, other sounds, and may not even notice the arrival or departure of a person. Finally, a combination of these disabilities can make it more challenging to make diagnostic evaluations of a person. People with multiple disabilities may need more time to process their surroundings and to realise what is expected of them. Furthermore, unexpected and sudden movements or actions, or giving too much information at once may cause a lot of stress that interferes with their performance. Many characteristics that normally differentiate people with ASD from people without ASD should not be assessed or assessed differently in people with multiple impairments. Still, some characteristics of the autistic spectrum are left that can be included in an assessment. Examples that cannot be forgotten include interest in, response to and looking for contact, resistance to change and interest in new items or situations. Sharing of feelings or interests may not occur through pointing or gaze, but may show itself in a more tactile way. It is important to be aware of the different way in which multiply disabled people express themselves. Finally, to account for intellectual disabilities, it is important to assess everything on a level that is suitable for the participants. Do not use complicated questionnaires, but simple toys as much as possible. Only if all of this can be done successfully, autistic people can be differentiated from non-autistic people and personal needs can be met.

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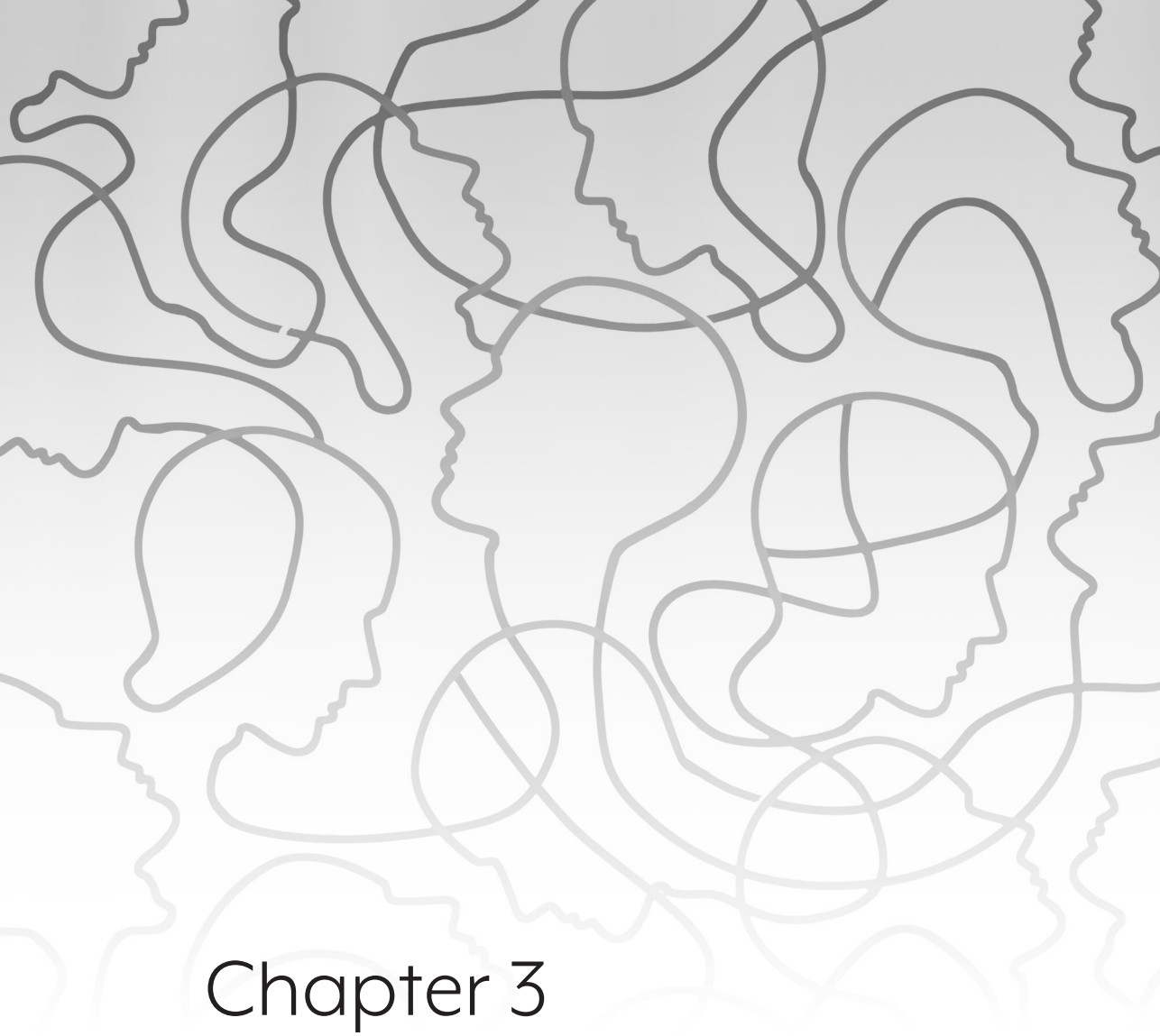

Chapter 3

A critical review of screening and diagnostic instruments for autism spectrum disorders in people with sensory impairments in addition to intellectual disabilities

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Abstract

Instruments that are used for diagnosing of, or screening for Autism Spectrum Disorder (ASD) may not be applicable to people with sensory disabilities in addition to intellectual disabilities. Firstly, because they do not account for equifinality, the possibility that different conditions may lead to the same outcome. Secondly, because they do not have appropriate norms for this target population. The current study reviewed 20 instruments commonly used in the assessment of screening for and diagnosing ASD. Reviewed were: purpose, number of items, psychometric properties (norms, reliability and validity), test availability, and item applicability for people with sensory and intellectual disabilities. Most instruments did not have norms for the target population and all instruments consisted of a quarter or more of invalid items. When using current instruments, caution is required in interpreting test results. For proper assessment of ASD in people with sensory and intellectual disabilities, more instruments are needed that are adapted to the sensory and intellectual disabilities of this population.

1. Introduction

According to the American Psychiatric Association (2013), autism spectrum disorder (ASD) is a developmental disorder characterized by two major components: deficits in communication and social interaction, and repetitive and stereotyped patterns of behavior. The process of diagnosing ASD roughly consists of four general steps (National Institute for Health and Clinical Excellence, 2012; Nederlandse Vereniging voor Psychiatrie, 2009; Volkmar et al., 2014). The first step is to identify any problems or concerns. When there is reason to believe a person might have ASD, the second step is screening for the presence of ASD. Screening is done by talking to parents or caretakers, by studying medical and psychological information and history, by making observations, and through using specific screening instruments. When a person screens positive for ASD, the third step is the application of diagnostic instruments. The final step is to make an individual profile to guide treatment (National Institute for Health and Clinical Excellence, 2012; Nederlandse Vereniging voor Psychiatrie, 2009). Important in this diagnostic process is to combine multiple instruments (Risi et al., 2006) and to incorporate multidisciplinary clinical judgments (Rutter, 2006; Volkmar et al., 2014). Steps two and three may be difficult to conduct in people with both sensory and intellectual disabilities. In this critical review we focus on these steps, the screening and diagnostic instruments commonly used in the assessment of ASD.

Although there is a broad range of instruments that can be used for screening for and diagnosing of ASD, these instruments may not be very valid and/or useful when people develop atypically because of motor, sensory, or intellectual disabilities. For instance, ASD typical behaviors are not only seen in people with ASD but also in people with visual impairments (Cass, 1998; Hobson, Lee, & Brown, 1999), auditory impairments (Knoors & Vervloed, 2011), intellectual disabilities (De Bildt, Sytema, Kraijer, & Minderaa, 2005; Matson & Shoemaker, 2009; Matson, Dempsey, LoVullo, & Wilkins, 2008; Vig & Jedrysek, 1999) and also in people with a combination of these impairments (Dammeyer, 2011, 2013; De Vaan, Vervloed, Knoors, & Verhoeven,2013; Hoevenaars-Van Den Boom, Antonissen, Knoors, & Vervloed, 2009; Rødbroe & Janssen, 2006). It is the latter group that is the focus of this review, people with an intellectual disability combined with a visual impairment or deafblindness. Deafblindness is broadly defined as any combination of both a visual and auditory impairment, and may be congenital or acquired. In this article, no boundaries are set for the severity of visual and auditory impairments (Hoevenaars-Van Den Boom et al., 2009; Larsen & Damen, 2014). Especially for this group, clinicians are often asked to assess the presence of ASD. The reason for this is that people with motor, sensory, and intellectual disabilities show many behaviors that topographically look the same as ASD symptoms, but reflect other underlying causes because they may be caused by the respective

disabilities instead of by ASD. This is an example of equifinality, the possibility that different conditions may lead to the same outcome. As a result, ASD is both over- as well as under-diagnosed in people with multiple disabilities (Andrews & Wyver,2005; Cass, 1998; Jure, Rapin, & Tuchman, 1991; Roper, Arnold, & Monteiro, 2003). This can be either because of diagnostic overshadowing, where symptoms are attributed to the most prominent disability (Carvill, 2001; Hoevenaars-Van Den Boom et al., 2009; Mason & Scior, 2004; Reiss, Levitan, & Szyszko, 1982), or because of diagnostic underrepresentation, which refers to falsely missing relevant behaviors. In the current case, some behaviors do not occur and are therefore not measurable in people with disabilities. For example, eye contact is absent in blind individuals but these people can still be aware of and interested in other people; one only has to measure it in another way (Hoevenaars-Van Den Boom et al., 2009; Kraijer & De Bildt, 2005; Livesley & Jackson, 1992).

Since screening and diagnostic instruments are based on criteria for ASD and ASD-typical behaviors, behavioral overlap with disabilities can cause a decreased usability of ASD instruments in persons with sensory and intellectual disabilities. Despite the ample availability of screening and diagnostic instruments for people with intellectual disabilities alone (Matson & Williams, 2014), there is a lack of instruments that are adjusted to the behaviors of persons with the combination of intellectual and sensory disabilities. Many test instruments, not only for ASD but also for other pathologies, assume that the person under study is able to see and hear (Tobin & Hill, 2011). As was noted by (Bodsworth, Clare, Simblett, & Deafblind UK,2011), this leads to a lack of suitable instruments to assess people with multiple disabilities. This is why many unsuitable instruments are still used in clinical practice.

As screening and diagnostic instruments play such an important part in the process of diagnosing ASD, the current critical review focuses on the question how valid existing instruments are for the assessment of ASD in people with sensory and intellectual disabilities. This review took into account not only commonly used screening and diagnostic instruments, but also instruments with another focus that partly assess ASD or behaviors that are typical for ASD (see American Psychiatric Association, 2013), as these are often used in ASD assessments. We looked at the quality of the assessment material, ease of use, the psychometric properties (reliability and content validity), the presence of norms for people with sensory and intellectual disabilities, and the applicability of all individual items. This review will provide insights for practitioners and researchers into which instruments are suitable for the diagnosis and assessments of ASD in people who have combined sensory and intellectual disabilities.

2. Method

2.1 Materials

Screening and diagnostic instruments for ASD were selected, as well as assessment instruments related to characteristics of ASD. Possible instruments were gathered based on a review of existing instruments for the detection and assessment of ASD (O'Brien, Pearson, Berney, & Barnard, 2001) and a literature search using the following keywords: diagnosis, assessment, instruments, screening, autism, autism spectrum disorders, intellectual disabilities, visual impairments, and multiple disabilities. Instruments were only included if they were available through purchase or online download by the authors in 2014 and 2015 and were available in English or Dutch. Only instruments for which psychometric data were available were included. Of the 14 instruments described in the aforementioned review, 10 were included in the current review in the same or different version. Our search led to a selection of 13 screening and diagnostic instruments and 7 instruments assessing ASD-typical characteristics.

2.2 Procedure

The characteristics of the instruments were assessed according to the guidelines of the Dutch committee on tests and testing (COTAN) (Evers, Sijtsma, Lucassen, & Meijer, 2010) and the BUROs center for testing (see: www.buros.org). The BUROs center is a large testing review center that is part of the University of Nebraska-Lincoln that evaluates tests on general characteristics, development and psychometric properties (BUROs, 2015). The COTAN evaluates tests on their theoretical soundness, quality of materials and manual, norms, and psychometric properties (COTAN, 2015). COTAN reviews of tests as well as reviews of the BUROs center for testing were added if they were available for our selected instruments. For every instrument the following characteristics were assessed: (1) availability and quality of the manual; and (2) the scientific foundations of the development, reliability, and validity of the instrument.

For the current study, the following information was collected from manuals, scientific literature, and judgments by COTAN or Buros: (1) name and abbreviation of the instrument, authors, and year of publication; (2) purpose of the instrument: screening, diagnosis, evaluation, or which characteristic it evaluates; (3) number of items; (4) duration of the assessment; (5) whether a manual is available or whether the test can be downloaded online; (6) training requirements to use this instrument and which professionals can use it; (7) which method the instrument uses: checklist, interview, or observation; (8) in case of the screening and diagnostic instruments, the source of the ASD criteria, for example DSM-IV, DSM-5, ICD-10 and/or scientific literature; (9) for which target group the instrument was developed; (10) languages in which this instrument is available (however, we are aware that it is possible that more

translations may exist that we don't know of); (11) reliability, and (12) test validity (for reliability and validity multiple sources were used, and in cases of contradictory results all results were reported along with references to the original source; (13) the availability of norms for people with both sensory and intellectual disabilities; and (14) the number of inappropriate items for people with both sensory and intellectual disabilities.

Item appropriateness for people with both sensory and intellectual disabilities was rated by the first author, in collaboration with the other authors, who all have expertise and clinical experience in the field of sensory and intellectual disabilities. These are individuals who besides an intellectual disability (as defined by the American Association on Intellectual and Developmental Disabilities, 2013) also have a visual impairment or deafblindness.

Topographically the same behaviors can have different causes or functions for people with sensory impairments or intellectual disabilities and people with ASD. Instruments for ASD assessment normally check only for symptoms and not the underlying cause. As a result, test items can sometimes be invalid for assessing ASD in children with sensory impairments and intellectual disabilities. Items were rated as inappropriate or sensory biased if at least one of the five following criteria applied to them. Criteria were based upon generally known behaviors of children with sensory impairments (e.g. Knoors & Marschark,2014; Pérez-Pereira & Conti-Ramsden, 2005; Pring, 2005; Warren, 1994). First, the absence of a behavior is an obvious direct consequence of the sensory impairment. For example, gaze following is impossible for blind people and showing a reaction to speech or sound for deaf people. Second, the behavior is caused by indirect or long-term effects of the sensory impairment, such as language impairments in deaf people or odd or clumsy motor behaviors (e.g., to prevent collisions) in blind people. Third, the behavior is a characteristic that develops differently or more slowly in people with sensory impairments. An example is "theory of mind" (ToM), which develops later in children with blindness or deafness. Fourth, the behavior is more likely to be adaptive for people with sensory impairments than typical for ASD. Examples are odd body postures to hear someone better or to focus vision, and head nodding to counteract eye movements caused by nystagmus. Fifth, the behavior is used for compensatory purposes, an example is echolalia in children who are blind. Echolalia, for a blind child, is an expression of practicing language by repeating over and over pieces of speech or to check for the presence of an unseen conversation partner (Pérez-Pereira & Conti-Ramsden, 1999).

3. Results

3.1 Screening and diagnostic instruments for ASD

Of the reviewed screening and diagnostic instruments (see Table 1), nine were designed for screening purposes and three for diagnostic purposes. Instruments were created (or revised) between 1978 and 2009. Number of items ranged from 12 to 206, with the screening instruments having the lowest number of items. No training is required for screening instruments according to their manuals, but at least some training or experience with ASD is required for diagnostic instruments. The screening instruments are either checklists or interviews. Of the diagnostic instruments, the ADI-R (Rutter, Le Couteur, & Lord,2003) and DISCO (Wing, 2003) are interviews and the ADOS is an observation (Lord, Rutter, DiLavore, & Risi, 1999). When it comes to the ASD criteria on which these instruments are based, the screening instruments were all based on scientific literature, while the diagnostic instruments took into account the ASD criteria from the DSM-IV (American Psychiatric Association, 2000) and ICD-10 (World Health Organization, 1992).

The reliability of the instruments ranged from poor to excellent. The diagnostic instruments showed moderate to excellent reliability, whereas reliability was poor to excellent for the screening instruments. In case of the ABC, (Krug, Arick, & Almond,1978) the reliability alone ranged from poor to excellent. This wide range of quality is the result of the fact that different sources reported different types of reliability. Their first research paper (Krug, Arick, & Almond, 1980) reported excellent intrarater reliability and good interrater reliability, but later research found low split-half reliability on scales of language and social or self-help (Volkmar et al., 1988). The other instruments showed more straightforward results when it came to validity and reliability. The results ranged from good and very good for the ADOS (Lord et al., 1999) to insufficient for the ESAT (Buitelaar et al., 2009).

Concerning their applicability for people with both sensory and intellectual disabilities, none of the instruments has norms for this target group except the ABC (Krug et al., 1978). However, though the ABC has norms for people with deafblindness, these norms are relatively old because they stem from 1978 and the sensory disabilities were not taken into account during the development of the instrument. The latter is also seen in the large number of inappropriate items for people with both sensory and intellectual disabilities. Of the instruments that do not have norms for people with sensory and intellectual disabilities, at least one third of the items of were not applicable for use in people with sensory and intellectual disabilities according to one or more of the five validity criteria described in the method section.

Table 1 Instruments for screening and diagnosis of ASD

Name of instrument, authors and sources	Purpose	Number of items	Administration (in minutes)	Manual	Training / expertise	Method
ABC Autism Behavior Checklist Krug, Arick & Almond (1978); Krug, Arick & Almond (1980); Eaves & Milner (1993); Volkmar et al. (1988); Olmi (1998); O'Brien, et al. (2001)	Screening	57	10-20	Checklist available online + part of larger assessment tool (ASIEP)	Experience or using training tapes is recommended	Checklist
ADI-R Autism Diagnostic Interview-Revised Rutter, Le Couteur & Lord (2003); De Jonge & De Bildt (2003); Cicchetti, Lord, Koenig, Klin & Volkmar (2008); De Bildt et al.(2004); Hill et al. (2001); Lord, Rutter & Le Couteur (1994); O'Brien, et al. (2001)	Diagnosis	93	90 – 150 (including scoring)	Available	Experience with ASD and interviewing. Need to study manual, no additional training required.	Interview
ADOS Autism Diagnostic Observation Schedule, module 1 Lord, Rutter, DiLavore, Risi (1999); De Bildt, De Jonge, Lord, Rutter, DiLavore & Risi (2008); O'Brien, et al. (2001)	Diagnosis	37	30 – 60	Available	Training is necessary, at least 2 days of training + additional practice	Assessment +Observation
ASAS Australian Scale for Asperger Syndrome Garnett & Attwood (1993); O'Brien, et al. (2001); Campbell (2005)	Screening	25	10-15	Available	Not reported	Questionnaire
ASQ Autism Screening Questionnaire Also known as SCQ, Social Communication Questionnaire Rutter, Bailey, Lord (2003); Berument, RUtter, Lord, Pickles & Bailey (1999); O'Brien, et al. (2001); Servatius-Oosterling (2010)	Screening	40	10	Available	No	Questionnaire

Criteria based on	Target population	Languages	Reliability	Validity	Norms for people with sensory and intellectual disabilities	Non-applicable items†
Checklists and characteristics of people with ASD.	18 months - Adulthood	English	Interrater reliability – Good (Olmi, 1998). Internal consistency – Good on total score, poor on subscales (O'Brien, et al., 2001)	Content validity, Concurrent validity & Criterion validity – Well established (Olmi, 1998)	Yes	1/3
ICD-10 & DSM-IV	Early child – adulthood, mental age above 2 years.	Over 15 languages including English, French, Spanish and Dutch	Interrater reliability, test-retest reliability, internal consistency – High (O'Brien, et al., 2001); Interrater-and test-retest reliability – Good (Rutter, Le Couteur, et al., 2003)	Construct validity with ICD-10 & Convergent validity – Good (O'Brien, et al., 2001); Discriminative validity – Good (Rutter, Le Couteur, et al., 2003)	No	2/3
DSM-IV & ICD-10	Children and Adults; 15 months and older (module 1 = preverbal)	Over 15 languages including English, French, Spanish and Dutch	Interrater reliability, test retest reliability – high (O'Brien, et al., 2001)	Construct validity with ICD-10 & Convergent validity – Good (O'Brien, et al., 2001)	No	1/2
Not reported	Primary school children	English, Dutch, German	Not reported (O'Brien, et al., 2001)	Not reported (O'Brien, et al., 2001)	No	2/3
Based on the ADI-R	Children 4 years and older	English, Dutch	Not reported (O'Brien, et al., 2001)	Discriminant validity – good (O'Brien, et al., 2001)	No	2/3

Table 1 Continued

Name of instrument, authors and sources	Purpose	Number of items	Administration (in minutes)	Manual	Training / expertise	Method
AUTI-R Van Berckelaer-Onnes & Hoekman (1991); Hoekman (1992); Evers, et al. (2010);	Screening	51	-	Available	-	Interview / Questionnaire
CARS Childhood Autism Rating Scales Schopler, Reichler & Renner (1988); Schopler, Reichler, DeVellis & Daly (1980); Malcolm (2014); O'Brien, et al. (2001); McLellan (2014)	Screening	15	30-60	Available	For psychologist or therapists, experience in assessments required	Observational assessment
DISCO Diagnostic Interview for Social and Communication Disorders Wing (2003); Kent (2014); Wing, Leekam, Libby, Gould & Larcombe (2002); O'Brien, et al. (2001)	Diagnosis	105 items assessing history 206 items assessing current behavior	3 hours	-	Needs adequate clinical experience, training is possible	Interview
ESAT Early Screening for Autistic Traits Buitelaar et al.(2009); Evers, et al. (2010); Swinkels et al. (2006)	Screening	14	-	Available	Experience with ASD screening needed. Additional training in manual	Interview / Checklist
M-CHAT-R/F Modified – Checklist for Autism in Toddlers – Revised with Follow Up Robins, Fein & Barton (2009); Robins et al. (2013); Robins, Fein, Barton & Green (2001); O'Brien, et al. (2001)	Screening	20	5	Available online	Little to no training necessary for health care professionals	Checklist

Criteria based on	Target population	Languages	Reliability	Validity	Norms for people with sensory and intellectual disabilities	Non-applicable items†
Literature, construct of 'early childhood autism'	Verbal and non-verbal children from 10-155 months.	Dutch	Internal consistency – Good (Evers, et al., 2010),Good to very good (Hoekman, 1992); Test-retest reliability – Good & Interrater reliability – Good (Evers, et al., 2010; Van den Berckelaer-Onnes & Hoekman, 1991)	Content validity & Criterion validity – good (Evers, et al., 2010); Construct validity – Good (Hoekman, 1992)	No	1/2
ASD criteria described in literature	Children, two years and older.	English, Dutch, Swedish, Japanese and more	Internal consistency – Good (Malcolm, 2014; O'Brien, et al., 2001), Acceptable (McLellan, 2014); Interrater reliability – Acceptable (McLellan, 2014); Adequate to High (O'Brien, et al., 2001)	Construct validity – Moderate to Good (Malcolm, 2014); Construct and Convergent validity – established (O'Brien, et al., 2001)	No	2/3
Earlier instruments and ASD criteria	Children and Adults	English, Dutch	Not reported (O'Brien, et al., 2001)	Not reported (O'Brien, et al., 2001)	No	History: 1/5 Current: 1/3
Literature on predictors of ASD	Children under 20 months	Dutch, English	Test-retest reliability – Insufficient (Evers, et al., 2010), Very good (Buitelaar, et al., 2009)	Content validity – Insufficient (Evers, et al., 2010); Criterion validity – Insufficient (Evers, et al., 2010); Good predictive validity (Buitelaar, et al., 2009)	No	1/3
Literature, clinical instruments, clinical experience	Young children	Over 15 languages including English, French, Spanish and Dutch	Test-retest reliability - Good (O'Brien, et al., 2001)	Sensitivity and Specificity – Good (O'Brien, et al., 2001)	No	3/4

Table 1 Continued

Name of instrument, authors and sources	Purpose	Number of items	Administration (in minutes)	Manual	Training / expertise	Method
PDD-MRS (AVZ-R) Pervasive Developmental Disorder in Mental Retardation Scale Kraijer (1999); Kraijer & De Bildt (2005); Evers, et al. (2010); Meadows (2007); O'Brien, et al. (2001)	Screening	12	10-30	Available	Experience with ASD, no additional training.	Checklist
PDD-ST-II Pervasive Developmental Disorder Screening Test (second edition) Siegel (2004); Siegel & Van Berckelaer-Onnes (2006); Chittooran (2007); Johnstone (2007)	Screening	12-22 items	10-20	Available	For professionals in clinical practice. No additional training required.	Checklist / Interview
SRS Social Responsiveness Scale Constantino & Gruber (2005); Roeyers, Thys, Druart, De Schryver & Schittekatte (2011) Hoff & Doepke (2014) Evers, et al. (2010)	Screening	65	15-20	Available	For psychologist or therapists, experience in assessments required	Checklist

Note. † Proportion of non-applicable items for people with both sensory and intellectual disabilities as judged by the authors according to the criteria stated in the method section.

3.2 Instruments assessing characteristics of ASD

The instruments assessing characteristics of ASD, shown in Table 2, assess a variety of characteristics, such as: communication, social behavior, repetitive behavior, social functioning, ToM, and adaptive skills. Their purposes are mostly to assess the skill level or the severity level of the target behaviors. Only the CCC-2 is a screening instrument, screening for language impairment (Bishop & Geurts, 2007), and the ComFor, which not only assess level of understanding but also what kind of augmentative communication a person requires (Verpoorten, Noens, & Van Berckelaer-Onnes, 2004). The number of items ranges from 14 to 225, and test administration

Criteria based on	Target population	Languages	Reliability	Validity	Norms for people with sensory and intellectual disabilities	Non-applicable items†
DSM-III, other screening tools, literature	Children and adults with intellectual disabilities; aged 2-70 years	Dutch, English, German, Italian	Interrater reliability, internal consistency & test-retest reliability – Good (Evers, et al., 2010; Meadows, 2007)	Content validity – Good & Criterion validity – sufficient (Evers, et al., 2010); Construct validity – Satisfactory (Meadows, 2007); Sensitivity and specificity – Good (Kraijer, 1999; O'Brien, et al., 2001)	No	1/3 . This does not influence total score according to manual
Research on development of ASD and typical children; DSM-IV-TR	Children between 12-48 months	English, Dutch	Not studied (Johnstone, 2007)	Sensitivity & specificity – Insufficient to good (Chittooran, 2007; Johnstone, 2007; Siegel, 2004)	No	1/2
DSM-5	Children 4-17	English, Dutch, German	Internal consistency –(Hoff & Doepke, 2014); Good (Evers, et al., 2010); Test-retest reliability – Moderate to strong (Hoff & Doepke, 2014)	Criterion validity – Good (Evers, et al., 2010); Sensitivity & Specificity – Excellent & Convergent validity – Low to moderate (Hoff & Doepke, 2014)	No	2/5

varies between 10 and 60 minutes. The CCC-2, CSBQ (Hartman, Luteijn, Moorlag, De Bildt, & Minderaa, 2007) and ToM-Test-R (Steerneman & Meesters, 2009) require an experienced clinician for the assessment and interpretation of the results, but additional training for administrators is only necessary for the ComFor and the Vineland-Z (De Bildt, Kraijer, Sparrow, Balla, & Cicchetti, 2003). The methods of administration are: assessment (1x), checklists (2x), interviews (2x), questionnaire (1x), and combined-interview questionnaire (1x) . The instruments are typically designed for (young) children, though the SRZ (Kraijer, Kema, & De Bildt, 2004), the ComFor, and Vineland- Z are also designed for people with intellectual disabilities.

Table 2 Instruments for assessing characteristics of ASD

Name of instrument, authors and sources	Purpose	Number of items	Duration (in minutes)	Manual	Training / expertise
CCC-2 Children's Communication Checklist – 2 (-NL, Dutch translation) Bishop & Geurts (2007); Evers, et al. (2010); McCauley (2010)	Screen for language impairments, pragmatic problems &, assist in ASD assessment	70	5-10	Available	General knowledge about tests and familiarity with the CCC-2 is required
ComFor Forerunners in Communication Verpoorten, Noens & Berckelaer-Onnes (2004); Noens, Van Berckelaer-Onnes, Verpoorten & Van Duijn (2006); Evers, et al. (2010)	Assess the most suitable form of augmentative communication and assess level of sense-making	36	45	Available	Psychologist, speach therapists with assessment qualification. Additional ComFor training is required.
CSBQ (VISK) Children's Social Behavior Questionnaire Hartman, Luteijn, Moorlag, De Bildt & Minderaa (2007); Evers, et al. (2010); Luteijn, Luteijn, Jackson, Volkmar & Minderaa (2000); De Bildt et al. (2009)	Assess social behavior in child with a pervasive developmental disorder	49	10	Available	Interpretation needs to be done by trained psychologist, pedagogue or psychiatrist
RBQ Repetitive Behaviors Questionnaire Honey, McConachie, Turner & Rodgers (2012); Van Kempen, De Vaan & Vervloed (2013)	Assessment of repetitive or stereotyped behaviors	33	-	Questionnaire only, available online	-
SRZ / SRZ-*i* Social Functioning Scale for the Mentally Retarded (-interview) Kraijer, Kema & De Bildt (2004); Evers, et al. (2010)	Assess social functioning in individuals with intellectual disabilities	31	10 - 25	Available	-

Method	Target population	Languages	Reliability	Validity	Norms for people with sensory and intellectual disabilities	Non-applicable items†
Checklist	Children between 4-15 years, speak in full sentences, normal hearing	English, Dutch	Internal consistency – Sufficient (Bishop & Geurts, 2007; Evers, et al., 2010), Strong (McCauley, 2010); Test-retest reliability – Good to Excellent (McCauley, 2010), sufficient to Good (Bishop & Geurts, 2007)	Construct validity & predictive validity – Insufficient (Evers, et al., 2010); Convergent validity - Sufficient & Divergent validity – good (Bishop & Geurts, 2007); Content validity – Good (McCauley, 2010)	No	3/4
Assessment and observation	People with ID and ASD without or with few verbal communication skills	English, French, Italian, Dutch	Internal consistency – Good, Inter-rater reliability – very high, Test-retest reliability – good (Noens, et al., 2006), Overall reliability – Sufficient (Evers, et al., 2010)	Construct validity – Good (Noens, et al., 2006); Content validity – Insufficient (Evers, et al., 2010)	No	All
Checklist	Children 4- 18 years; children with intellectual disabilities 4-18 years, children with PDD-NOS, ADHD, high functioning ASD.	English, Dutch	Inter-rater reliability – Satisfactory & Internal Consistency – High (Hartman, et al., 2007; Luteijn, et al., 2000), Sufficient (Evers, et al., 2010); Test-retest reliability – High (Luteijn, et al., 2000)	Construct Validity – Established (Hartman, et al., 2007; Luteijn, et al., 2000); Content Validity – Sufficient (Evers, et al., 2010)	No	2/5
Questionnaire	Children with autism, 4-16 years	English, Dutch, Hebrew	Internal consistency – Good (Honey, et al., 2012)	Construct validity – Insufficient & Concurrent validity – Good (Honey, et al., 2012)	No	1/4
Questionnaire / Interview	Children and adults with intellectual disabilities, 4 years and older	Dutch	Internal consistency – Good, interrater reliability – good, Test-retest reliability – good (Evers, et al., 2010; Kraijer, et al., 2004)	Content validity – & Criterion validity – Good (Evers, et al., 2010); Construct validity – Good (Kraijer, et al., 2004)	No	1/4

Table 2 Continued

Name of instrument, authors and sources	Purpose	Number of items	Duration (in minutes)	Manual	Training / expertise
TOM-test-R Theory of Mind test Revised, 2009 Steerneman & Meesters (2009); Evers, et al. (2010)	Assess Theory of Mind and precursors	14	20	Available	Needs to be assessed by clinicians
Vineland-Z Vineland-Z, for children and youth with an intellectual disability (Dutch manual) De Bildt, Kraijer, Sparrow, Balla & Cicchetti (2003); Evers, et al. (2010)	Assess level of adaptive functioning	225	20 – 60	Available	Necessary

Note. † Proportion of non-applicable items for people with both sensory and intellectual disabilities as judged by the authors according to the criteria stated in the method section.

Similarly to the screening and diagnostic instruments, the psychometric properties range from insufficient to good. However, none of the instruments in Table 2 have norms for people with both intellectual and sensory disabilities. Similarly to the screening and diagnostic instruments, at least a quarter the items are inappropriate for people with sensory and intellectual disabilities.

4. Discussion

All instruments show at least adequate psychometric properties on some aspects, though mixed results have been found for the ABC (Krug et al., 1978), ESAT (Buitelaar et al., 2009), CARS (Schopler, Reichler, & Renner, 1988), PDD-MRS (Kraijer & De

Method	Target population	Languages	Reliability	Validity	Norms for people with sensory and intellectual disabilities	Non-applicable items†
Interview	Children 4 – 12 years old	Dutch	Overall reliability insufficiently studied (Evers, et al., 2010); internal consistency – Good (Steerneman & Meesters, 2009), acceptable (Evers, et al., 2010); test-retest reliability – Satisfactory (Steerneman & Meesters, 2009), good (Evers, et al., 2010); interrater reliability – Good (Steerneman & Meesters, 2009)	Construct validity (Steerneman & Meesters, 2009); Content validity – Insufficient (Evers, et al., 2010) Criterion validity – not studied (Evers, et al., 2010)	No	All
Interview	Children and youth with an intellectual disability, 4-18 years	English, Dutch, Spanish	Internal consistency – Good (Evers, et al., 2010)	Content validity – Good; Criterion validity – Good; Construct validity – Good (Evers, et al., 2010)	No	1/2

Bildt, 2005), PDDST-II (Siegel, 2004), SRS (Constantino & Gruber, 2005), CCC-2 (Bishop & Geurts, 2007), ComFor (Verpoorten et al., 2004), CSBQ (Hartman et al., 2007), and the ToM-test-R (Steerneman & Meesters, 2009), and no psychometric properties were reported for the ASAS (Garnett & Attwood, 1993) and DISCO (Wing, 2003). However, the remaining instruments appear to be suitable for the intended purpose and target group. The only instrument with norms for people with deafblindness is the ABC (Krug et al., 1978); however, these norms are the result of their 1978 study and are likely to be outdated. None of the other instruments have norms for people with both sensory and intellectual disabilities. More importantly than the lack of norms, however, is the sensory bias in all of the instruments. In cases of people with sensory impairments, test items either cannot be assessed or cannot be interpreted as signs of ASD. In all instruments, at least one quarter of items show

this bias and are therefore inappropriate for assessing ASD or ASD behaviors in people with sensory and intellectual disabilities. The manual of the AVZ-R, an instrument designed for people with intellectual disabilities, acknowledges that some items are inappropriate for people with additional sensory impairments, but claims that this does not affect the total score (Kraijer, 1999). This conclusion seems illogical; taking into account the number of inappropriate items we think it is impossible to make a valid diagnosis. For several instruments, especially the screening instruments such as the ESAT (Buitelaar et al., 2009) and M-CHAT (Robins, Fein, & Barton, 2009), the cutoff score for ASD is rather low. Without taking into account possible invalid items, a person with sensory and intellectual disabilities would easily score within the clinical range of ASD on these instruments regardless of the actual presence of ASD. The large number of false positive scores reduces test specificity, but at the same time proper ASD symptoms in people with both sensory and intellectual disabilities are also missed. The sensory bias reduces specificity of the reviewed instruments, but also their sensitivity for true ASD symptoms.

If one looks at the five criteria for inappropriate items, the first conclusion to be drawn is that they cannot be used because the behaviors result not from ASD but are consequences of sensory impairments. This problem was found in all of the instruments. Items such as "Does the person make eye contact?" or "Does the person respond to calling their name?" measure abilities that cannot be measured in all people with sensory impairments. These examples are very straightforward, but this problem also occurs more subtly in items that measure gaze following, pointing, showing, and making conversation. Not only should the direct consequences of sensory impairments be taken into account; the indirect consequences of sensory impairments are also important. Many people with congenital deafblindness do not speak but communicate with sign language or gestures (Dalby et al., 2009), pictures or objects (Noens, Berckelaer-Onnes, Verpoorten, & Van Duijn, 2006). Especially when there is an additional intellectual disability, the typical language impairments of ASD such as echolalia (Lin,2014; Roberts, 2014) do not occur in this target group, simply because most of them do not use speech to communicate. Not taking this into account would lead to diagnostic underrepresentation of ASD (Kraijer & De Bildt, 2005; Livesley & Jackson,1992). When a behavior cannot occur one should not attempt to measure this for diagnostic purposes, and subsequently norms or cutoff points should be adjusted.

A construct such as ToM can be present in people with visual impairments, but cannot be tested in the same way as in people without visual impairments. The ToM test that was included in this review (Steerneman & Meesters, 2009) and other ToM tasks, such as false belief tasks such as the Sally Ann task (Baron-Cohen, Leslie, & Frith, 1985), all use visual stimuli to assess the presence of ToM. These stimuli cannot be properly perceived by visually impaired and blind people. The problem for deaf

people is that test instructions are verbal and require good speech and language skills. Deaf children often fail ToM tasks not because they do not have ToM skills but because of inappropriate language skills (Peterson, Wellman, & Liu, 2005). Also, when it comes to communication, people with hearing impairments often use sign language and people with deafblindness communicate through tactile signing (Miles, 2003), pictures, or objects (Noens et al., 2006), communication forms not taken into account by the reviewed instruments which largely rely on oral language to assess communication.

An important issue to consider is the level of intellectual disability and the developmental age of the person assessed in combination with the nature and severity of their sensory impairments. Some behaviors, such as pretend play or ToM, depend on cognitive skills that typically develop after a certain developmental stage is reached. The performance of persons functioning below this level cannot be interpreted in the same way as one would do for someone who does function at or above this developmental level (De Jonge & De Bildt, 2003). With respect to ToM, we now know that blind children are capable of having a ToM, but the development of ToM in children who are blind without intellectual disabilities takes about two years extra compared to sighted children (Brambring & Asbrock, 2010). With additional intellectual disabilities, this delay will surely be longer or ToM may not even develop at all. Developmental delays are also seen for play behavior. Blind children engage more in solitary play than children without visual impairments (Tröster & Brambring, 1994) and show less symbolic play at the same ages as sighted children (Hughes, Dote-Kwan, & Dolendo, 1998), and again, intellectual disabilities will increase these delays.

Adherence to routines is often seen as characteristic of ASD. However, for people with sensory impairments routines are important to get a grip on life, especially with limited options for communication. People who cannot see or hear need routines to understand where they are going, what they are doing, or what they can expect. Therefore adherence to routines cannot be a differentiating factor in itself if one does not check for the perseverance or ability to stop the routine or repetitive behavior (Gense & Gense, 2005). Finally, communication develops differently in people with sensory and intellectual disabilities. They use other modes of communication than spoken language. In addition, social skills such as showing empathy, expressing moral emotions, and supporting peers are shown less frequently in deaf children, likely because they have fewer opportunities to incidentally learn these behaviors (Ketelaar, Wiefferink, Frijns, Broekhof, & Rieffe, 2015; Netten et al., 2015). The social partner in communication also plays a key role in the quality of social interactions with someone who is deafblind and has an intellectual disability (Damen, 2015). When a person's social partners do not adjust their way and mode of communication properly, test scores could easily underrate a person's true ability.

5. Implications

The fact that many items are inappropriate and there are no norms does not mean an ASD instrument is completely worthless. In fact, with care, instruments could potentially still be valuable in the assessment of people with sensory and intellectual disabilities. Their use for screening diagnostic purposes is however limited. The ADI-R, for example, can be used in people with deafblindness to assess their clinical needs (Rutter et al., 2003). Some instruments, especially the ones assessing characteristics of ASD, could also be helpful to assess progress over time, and to evaluate skill levels or whether treatment goals are reached. Though a number of test items are definitely not suitable, many are. It can still be clinically relevant to see how someone's social or adaptive skills have changed over time. As long as the main goal of the instrument is not to decide on a diagnosis of ASD it can still be used to assess someone's level of functioning. Instruments that partly assess ASD, such as the CSBQ (Hartman et al., 2007), the RBQ (Honey, McConachie, Turner, & Rodgers, 2012), the SRZ (Kraijer et al., 2004), and the Vineland-Z (De Bildt et al., 2003) are applicable for use in people with intellectual and sensory disabilities for their intended use but not for diagnosing ASD in this group because the norms no not apply. In our opinion, the clinician involved in the assessment must in all cases have expertise on and experience with people with sensory and intellectual disabilities, whether or not instruments are used. Keep in mind, however, that the clinical opinion can be biased too, especially when instruments cannot give a clear answer (De Bruyn, 2006). We therefore recommend multidisciplinary assessments and the use of multiple instruments if possible (Risi et al., 2006; Rutter, 2006; Volkmar et al., 2014).

A solution to the validity problem might be to adjust test items or instruments. For example, the ComFor (Verpoorten et al.,2004) was not usable at all in people with sensory impairments, so the authors recently developed the ComFor-V, an adaptation suitable for people with both intellectual disabilities and visual impairments (KU Leuven, 2015). For other instruments, test items can be adjusted to make them more appropriate for people with sensory impairments and intellectual disabilities. One can replace spoken words by sign language to assess communication, for example, in people who are deaf. Items assessing joint attention can also be adjusted for people who are blind, for example by changing gaze following to a more appropriate form of inferring attention such as freezing or motor movements, a change in breathing, signs of concentrated listening, or tactile cues given by the blind person in tactile signing (Miles, 2003). The obvious downside of adjusting items is that norms are no longer valid and existing research regarding validity and reliability no longer applies to the adjusted version. Furthermore, as these constructs often develop differently in people with sensory and intellectual disabilities, the interpretation of the results should always be done with care. Extra care is necessary since in the

assessment of people with multiple disabilities, adjustments are often required on an individual level as not everyone has the same level of visual impairment, auditory impairment, or communication skills (Boers, Janssen, Minnaert, & Ruijssenaars, 2013). In these cases, individual progress can be measured but one cannot compare between individuals.

6. Conclusion

Commonly used instruments that were designed to assess the presence of ASD or characteristics of ASD were reviewed for their use in people with sensory and intellectual disabilities. The validity and reliability of these instruments have in most cases been scientifically supported for people without disabilities; the ADOS (Lord et al., 1999) and ADI-R (Rutter et al., 2003) are considered to be the preferred instruments in ASD assessment (De Bildt et al., 2004; Reaven, Hepburn, & Ross, 2008). However, this does not make them applicable for use in people with sensory and intellectual disabilities. The instruments typically used to assess ASD or ASD characteristics are in general not valid for use in people with sensory impairments in addition to intellectual disabilities. For this specific population new instruments are urgently needed. When more information about certain behaviors is required, the reviewed assessment tools can be helpful, only it is important to keep their limitations in mind, to use multiple tools and a multidisciplinary team, and most of all, to take into account a person's individual characteristics, limitations, and possibilities.

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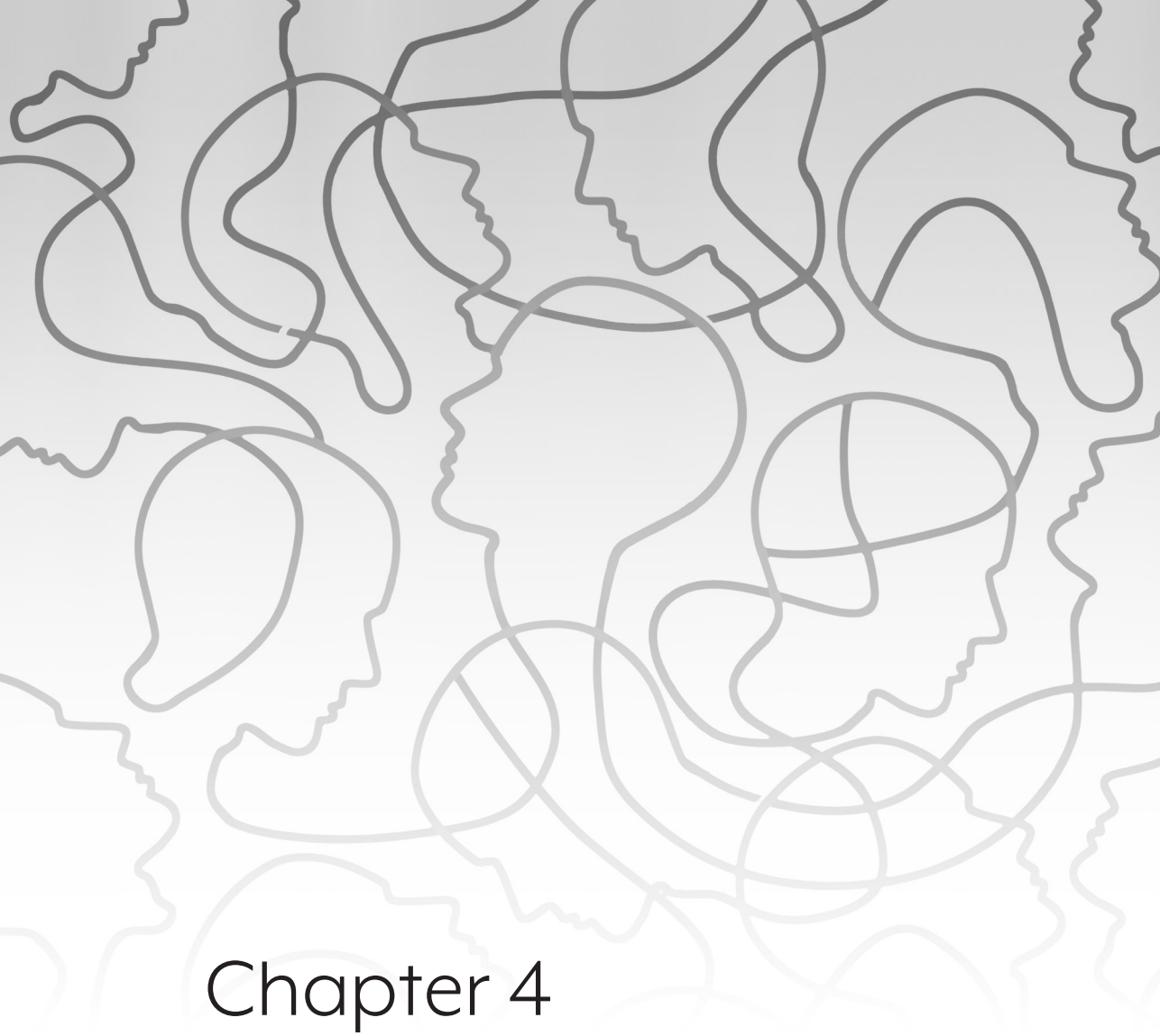

Chapter 4

Behavioural assessment of autism spectrum disorder in people with multiple disabilities

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Abstract

It is difficult to diagnose autism spectrum disorder (ASD) in people with a combination of intellectual and sensory disabilities because of overlap in behaviour. The ASD typical behaviours of people with combined intellectual and sensory disabilities are often caused by their disabilities and not by ASD. Current diagnostic tools are inadequate to differentiate between people with and without ASD when they have these combined disabilities, because tools lack norms for this population or are subjective, indirect or unable to adapt to the variety of disabilities that these people may have. Because giving a correct diagnosis is necessary for treatment and support, a new observational tool was developed to diagnose ASD in people with multiple disabilities, observation of autism in people with sensory and intellectual disabilities (OASID).

Observation of autism in people with sensory and intellectual disabilities was tested on 18 participants with moderate to profound intellectual disabilities, one or dual sensory impairment, with and without ASD. Two independent experts diagnosed these participants as well in order to test the psychometric properties and differentiating abilities of OASID.

Observation of autism in people with sensory and intellectual disabilities showed high inter-rater reliability, internal consistency of scales and content and construct validity. OASID could differentiate people with and without ASD without overlap.

Observation of autism in people with sensory and intellectual disabilities could differentiate people with intellectual disabilities combined with sensory impairments, who clearly had or did not have signs of ASD. People with unclear signs of ADS scored in between those two groups with regard to their OASID scores. Psychometric properties of OASID are promising.

1. Introduction

People with sensory disabilities, intellectual disabilities (IDs) or a combination of these disabilities often show behaviours that may be symptoms of autism spectrum disorder (ASD) that in fact may be caused by their disabilities (De Vaan, Vervloed, Knoors & Verhoeven, 2013a; Hobson, 2005; Hoevenaars-van den Boom, Antonissen, Knoors & Vervloed, 2009; Knoors & Vervloed, 2011; Van Gent, 2012). If it is not taken into account whether behaviours are caused by ASD or another impairment, this may lead to both underdiagnosis and overdiagnosis of ASD in this group (Cass, 1998; Jure, Rapin & Tuchmann, 1991; Roper, Arnold & Monteiro, 2003). The right diagnosis and proper case formulation can improve treatment, education and care (De Vaan, et al., 2013a). The current study focuses on how to diagnose ASD correctly in people who have a combination of sensory and IDs (in this paper called 'multiple disabilities').

Intellectual disabilities as well as sensory impairments can both cause behaviours that are similar to ASD. There is overlap between ASD and ID (Matson & Schoemaker, 2009) and this overlap becomes larger as the level of ID increases (Matson, Dempsey, LoVullo & Wilkins, 2008). This overlap is, for example, seen in stereotyped play or movements (Matson, et al., 2008; Medeiros, Rojahn, Moore & Van Ingen, 2014; Wing, Gould, Yeates & Brierly, 1977).

People with visual impairments show autistic features too (Cass, 1998; Hobson, 2005; Pérez-Pereira & Conti-Ramsden, 1999). They may show a lack of reciprocity in social interaction (Dale, Tadić & Sonksen, 2014; Fraiberg, 1977), poor use of language for social purposes and awkward pragmatic language use (Tadić, Pring, & Dale, 2010), problems in understanding and using non-verbal communication (Gense & Gense, 2005) and stereotyped behaviours (Tröster, Brambring, & Beelmann, 1991; Warren, 1994). These behaviours do not necessarily originate from ASD but are a direct result of the visual impairment.

Communication skills are affected in people with ASD but also in people with hearing impairments. Deafness leads to delays in language development, an absence of spoken language or the use of atypical language (Knoors & Vervloed, 2011). Communication at home is less frequent (Vaccari & Marschark, 1997), and people who are deaf can show impairments in the monitoring of conversations (Wolters, Knoors, Cillessen & Verhoeven, 2011). Delays in of theory of mind are also quite common (Knoors & Marschark. 2014; Peterson & Siegal, 1995, 2000). Other social impairments include deficient contact with others, disordered social imitation and impaired joint attention (Van Gent, 2012; Vig & Jedrysek, 1999) and a preference for objects and physical attributes over social contact (Rogers & Ozonoff, 2005).

People with an ID and additional visual impairments show even more problems. Among others, one finds problems in communication, social and daily living skills (Evenhuis, Sjoukes, Koot & Kooijman 2009), such as initiating social contact or

activities (Munde & Vlaskamp, 2014). Maybe, the largest overlap is found in people with dual sensory disabilities (Dammeyer, 2013; Hoevenaars-van den Boom, et al., 2009). A more complete list of symptom overlap between people with ASD and people without ASD when they have sensory or IDs or a combination of both can be found in the review of De Vaan et al. (2013a).

Despite the presence of instruments to screen for or diagnose ASD in people with IDs (see Matson & Shoemaker 2009; Matson & Williams, 2014), there are still no instruments that can validly diagnose ASD in people with multiple disabilities. The available tools lack norms for people with sensory and/or IDs, and often, items are inappropriate because they require sight or hearing to pass them (Carnaby, 2007; Hoevenaars-van den Boom, et al., 2009; Jure, et al., 1991; Matson & Shoemaker, 2009).

To circumvent the aforementioned problems, Hoevenaars-van den Boom et al. (2009) designed a new instrument, observation of autism in deafblindness (O-ADB), for people with profound IDs and deafblindness and subsequently studied in a pilot study with 10 individuals with profound IDs and deafblindness with or without ASD. Although it differentiated ASD from no ASD successfully, only the most severe cases of deafblindness and ID were included, and administration was lengthy and rather stressful to undergo. The current study elaborated on the O-ADB, taking into account the previously mentioned limitations. The administration was made more practical by decreasing the number of tasks and thus the administration time. Lastly, the items were adapted in such a way that the people with lesser degrees of ID could also be tested. This paper describes the development of this instrument, whether it can differentiate between behaviours of people with ASD and behaviours of people without ASD and whether it can do so in a valid and reliable way.

2. Method

2.1 Participants

Participants were 20 clients recruited from four institutions throughout the Netherlands specialised in providing care or education to people with IDs, visual impairments, hearing impairments and deafblindness. Because of confidentiality, a contact person from every institution selected eligible clients from residential units or schools and approached their legal representatives with written information about the study and a consent form. Staff of the institutional settings recruited participants. To warrant privacy, the exact number of people approached and reasons not to participate were not recorded. As a result, the exact response rate is unknown.

Participants qualified if the following criteria were met: (1) a moderate to profound ID (an IQ below 50), (2) a visual field of less than 20° and/or a visual acuity of less than 6/20 or complete blindness and (3) a chronological age between 5 and 55 years.

Additionally, half the participants could have a hearing loss of at least 35 dB. It was requested that half of our participants were diagnosed with ASD and the other half did not. The institutional contact persons checked the institute's record for suitable participants. Participants were only considered for inclusion when assessments and diagnoses were performed by trained and licensed psychologists, psychiatrists or physicians. Because no accurate criteria existed to diagnose ASD in people with sensory and IDs, a consensus between two experts about the presence of ASD served as the gold standard. After participants were enrolled, groups were formed based on the experts' opinions whether participants had ASD or not. One of the experts was a child psychiatrist who works with deafblind children, whereas the other expert is specialised in dealing with ASD in children with IDs. Both experts made their judgements independent of each other, based on videos of the participants made for this study and a summary of the information in the participant's record. The experts also received a list of ASD typical behaviour as defined by DSM-5.

Two of the 20 participants, participants 9 and 16, were excluded from further participation because of personal circumstances. One of them had such severe motor difficulties that he was unable to perform the tasks that were expected of him. Another participant felt extremely stressed at the day of the experiment, keeping him from participating in four out of five tasks. For 9 of the remaining 18 participants (see Table 1 for an overview of participants), complete consensus regarding ASD diagnosis could be reached. For eight participants nearly reached consensus (one or both experts were uncertain about the diagnosis), for just one participant, the experts totally disagreed. So based on expert judgement, we have three groups: ASD, no ASD and doubtful.

2.2 Materials

2.2.1 Observation of autism in people with sensory and intellectual disabilities (OASID)

For the purpose of this study, a new instrument was developed, named 'Observation of Autism in people with Sensory and Intellectual Disabilities' (OASID), to diagnose ASD in people with sensory and IDs. OASID was designed to provide a more adaptive approach in diagnosing ASD in people with multiple disabilities. OASID is a semi-structured observational instrument.

2.2.1.1 Development

After reviewing the literature and existing instruments, observations of the target population, conversations with caregivers and advice from experts in science and clinicians from the field, the differentiating characteristics for people with sensory and IDs with and without ASD were selected. These findings were compared with the diagnostic criteria from the DSM-5 (American Psychiatric Association, 2013).

Table 1 Participants (excluding participants 9 and 16)

Number	Age	Sex	Intellectual disability	Visual Acuity (in the better eye or both eyes)	Hearing loss (in the better ear or both ears)	Syndromes or relevant medical conditions
1	11	Male	Profound	13/20	Profound hearing loss	Disabilities caused by meningitis 4 days after birth
2	24	Female	Severe	4/10	> 110 dB	Congenital Rubella Syndrome
3	56	Female	Moderate	Blindness	90 dB	Congenital Rubella Syndrome
4	55	Male	Severe	3/100	105 dB	Cerebral Visual Impairment.
5	27	Female	Profound	3/100	85 dB	Retinopathy of Prematurity
6	26	Female	Severe	Severe visual impairment	112 dB	Congenital Rubella Syndrome
7	39	Male	Severe	1/10	Sensitive to sudden and loud noises	-
8	38	Male	Severe	Blindness, no light perception	No hearing loss	-
10	31	Female	Severe	3/20	No hearing loss	-
11	19	Female	Profound	8/100	No hearing loss	De Morsier Syndrome
12	17	Female	Severe	4/10	Reactions to > 45 dB	Wolf-Hirschhorn Syndrome
13	18	Male	Profound	3/10	30 dB	Down Syndrome
14	25	Female	Severe	3/20	No hearing loss	Near Sudden Death Syndrome
15	19	Female	Severe	Blindness, no light perception	No hearing loss	-
17	29	Female	Moderate	Blindness, no light perception	Moderate hearing loss	-
18	23	Female	Moderate	Moderate visual impairment	Deafness	-
19	32	Male	Severe	Blindness, mild light perception.	81 dB	Congenital Rubella Syndrome
20	50	Male	Moderate	2/10	Reactions to > 108 dB	Marschall-Stickler Syndrome

Transformation of these criteria into testable items was carried out by reviewing the literature and items of other instruments, especially of the O-ADB (Hoevenaars-van den Boom, et al., 2009) and the autism diagnostic observation schedule (ADOS) (Lord, Rutter, DiLavore & Risi, 1999). Preliminary versions of OASID were reviewed by all authors and authors of the O-ADB and were discussed with caregivers and other researchers, before the final version was administered to participants.

The first version OASID differentiated between participants with and participants without ASD. However, inter-rater reliability was too low, and internal consistency for one sub-scale was insufficient (de Vaan, Vervloed, Knoors, & Verhoeven, 2013b). Subsequently, OASID item descriptions and the divisions of items across scales were carefully and critically reviewed and revised, aiming for an inter-rater reliability that was at least substantial (Cohen's kappa = 0.6) in the typology of Landis and Koch (1977) and an internal consistency of at least 0.7 (Nunnally & Bernstein, 1994). These revisions resulted in the current version.

2.2.1.2 Procedure of administration

Participants are invited with a familiar caregiver to an assessment room, where the researcher awaits them with several tasks. While the researcher is administrating the tasks, the familiar caregiver is present the whole time for comfort and to assist the researcher if necessary. This was carried out to prevent stress, discomfort and communication problems. Administrating the tasks lasted between 25 min and 1 hour, and this was recorded on video. No scoring occurred during administration of the tasks. Video recordings were scored and observed afterwards.

2.2.1.3 The tasks

Observation of autism in people with sensory and intellectual disabilities consists of five tasks of which an example of which can be found in Table 2. Tasks were partly inspired by the O-ADB (Hoevenaars-van den Boom, et al., 2009) and the ADOS (Lord, et al., 1999). All tasks were intended to elicit specific behaviours, using play materials and toys, that are expected to differentiate between people with and without ASD. All items were based on literature and clinical experience with children with and without ASD and multiple disabilities. An example is given in Table 2. The item described in Table 2 is based on the ASD criteria that people with ASD often show severe preoccupations and have difficulties in breaking routines or small changes (American Psychiatric Association, 2013; Turner, 1999). This is why a familiar play object was brought to the test to be later on removed to further continue playing with other objects. In people with ASD, this object could evoke preoccupied playing or no playing at all (because it is not part of their routine at that time), and removal of the object could induce stress (American Psychiatric Association, 2013). The OASID scoring options take these behaviours into account.

Table 2 Example of observation of autism in people with sensory and intellectual disabilities task and corresponding question and answering possibilities

Task	Example question	Behavior resulting in score of 0	Behavior resulting in score of 1	Behavior resulting in score of 2
The participant has played with their own toy or object and after a while the experimenter took away the object.	How does the person respond to the removal of their own object?	The person does not show anger, sadness or frustration as a response to the removal of their object	The person shows some anger, sadness or frustration as a response to the removal of the object, or briefly clings to it, but gives the object away within three attempts of the experimenter	The person shows anger, sadness or frustration as a response to the removal of their object, he/she may cling to the object and refuses to give the object away even after three attempts of the experimenter

Within one task of OASID, several ASD typical behaviours can be elicited. As a result, only a few tasks with a limited set of materials were included in OASID, avoiding over-stimulation of participants. A distinguishing feature of OASID is the adaptive approach that is used in the administration. Administration of the tasks is adjusted to the abilities, severity of intellectual and sensory disabilities and communication style of individual participants. For example, different prompts are used to elicit joint attention, such as eye gaze and physical direction towards an object. The difficulty level of some test items can also be adapted. For participants with a moderate ID and a slight visual impairment, a difficult puzzle can be given, while for people with profound IDs and blindness or severe motoric difficulties, the task is simplified to putting puzzle pieces in a bucket. The presentation of the puzzle pieces is also adjusted to the participant's abilities. For instance, someone who is blind is presented with the pieces by giving them in their hands or bringing their hands to the pieces, while sighted persons are shown the pieces. Because the objective of the task was to judge the interaction of the person with the experimenter instead of solving the puzzle that was not the objective of the task, but to see how the person interacts with the researcher or asks for help, these adjustments in difficulty could be made. Mode of communication is adjusted to each individual's possibilities. Verbal participants were spoken to, while for others, simple hand gestures and signs will be used, and for other participants, communication consists of handing over objects. Finally, an important principle in administrating OASID is that the researcher did not only attempt to elicit behaviour in the participants but also wait for initiatives of the participant for

social contact and responds to those. This adaptive approach made administration suitable for a broad range of participants.

2.2.1.4 The scoring form

After administration of the tasks, recordings of the session were used to score ASD typical behaviours. Of the 40 items, 29 items scored ASD typical behaviours seen in the five tasks. The remaining 11 items were holistic items scoring behaviours that occurred during the entire administration.

The 40 items were scored on a 3-point Likert scale (0, 1, 2) where a higher score indicated more ASD typical behaviour. Elaborate descriptions for each answering possibility were provided. An example question can be found in Table 2. A final score is obtained by adding all item scores and by calculating two scales and seven sub-scales that are in line with DSM-5 criteria; see Table 3.

Table 3 Scales and sub-scales of OASID along DSM-5 criteria

Scale	Number of items
A. Social behavior and communication	21
1. Reciprocity	9
2. Communication	3
3. Relationships	9
B. Repetitive and stereotyped behavior	19
1. Stereotyped behavior	7
2. Insistence on sameness	6
3. Restricted interests	3
4. Reactivity to sensory input	3

2.2.2 Other measures

In addition to OASID, two questionnaires were filled out by parents or caregivers to study convergent and divergent validity. The Autisme-en verwachte stoornissen-schaal-Z-revisie (AVZ-R) is the Dutch version of the Pervasive Developmental Disorder in Mental Retardation Scale that is a short questionnaire often used to diagnose ASD in children with IDs (Kraijer & De Bildt, 2005). This scale does not take into account possible sensory impairments. Also, parents or caregivers filled out the Dutch translation of the list of behavioural signs of disturbed attachment in young children (Stor & Storsbergen, 2006), originally described by Boris and Zeanah (2005). This list was chosen because behaviours of disturbed attachment may appear similar to ASD typical behaviour but are not equal and differentiation between the two should be

made (Rutgers, Bakermans-Kranenburg, van Ijzendoorn & van Berckelaer-Onnes, 2004; Zeanah, Smyke, Koga, Carlson & The Bucharest Early Intervention Project Core, 2005).

2.3 Procedure

This study was approved by the region's medical ethical review board and legal representatives, or parents signed the informed consent form prior to participation. After this, OASID was administered in a room without distractions, other clients and familiar toys and recorded on video as described previously. The caregiver that was present did not interact with the participant unless necessary and filled in the other questionnaires, while the experimenter performed the tasks with the participant. The recorded sessions were independently scored afterwards by the first author and two trained raters, all of them naive to the participant's background and possible ASD diagnosis.

2.4 Statistical analyses

Because of the ordinal nature of the scoring system, non-parametric tests were used for analyses. For reliability and validity measures, all 18 participants were included. In case of multiple comparisons, a significance level of .01 was used to avoid chance capitalization.

Item distributions for OASID were checked to make sure that all items had discriminating power. Items with identical scores for 90% or more of the participants were excluded. Only for two items, 87% of participants received the same score, and all of the other items had a lower percentage of identical scores. Therefore, no items were excluded.

For the inter-rater reliability, Cohen's kappa (Cohen, 1960) the intraclass correlation coefficient and weighted kappa (Fleiss &Cohen, 1973) were assessed. Because OASID scores are ordinal, a two-point difference between raters is worse than a one-point difference, and weighted kappa takes this into account (Fleiss & Cohen, 1973). Cronbach's alpha was not calculated for individual sub-scales but only for the total scale and the scales 'Social behaviour and communication' and 'Repetitive and stereotyped behaviour', as the small number of items on each sub-scale might negatively influence Cronbach's alpha and underestimate reliability (Cortina, 1993).

3. Results

3.1 Reliability

3.1.1 Inter-rater reliability

Videos of all participants were rated by the first author, and a random selection of seven videos was rated as well by two undergraduate students in pedagogical and educational sciences after they had received training in scoring OASID. First, between student raters, percentage of agreement was 77.8%, the corresponding Cohen's kappa was .65 and weighted kappa was .75. The intraclass correlation coefficient, taking into account scores of all three raters, was .69. All values indicated a substantial to good inter-rater reliability (see Altman, 1991; Landis & Koch, 1977).

3.1.2 Internal consistency

Table 3 shows that a priori OASID consists of two scales and seven sub-scales. To measure the internal consistency of items on these scales, Cronbach's alpha was calculated. Cronbach's alpha for OASID completely was .94, for the first scale 'social communication and interaction', it was .94 and for the second scale 'repetitive and stereotyped behaviour', it was .79. These values indicated good to excellent internal consistency of items (Kline, 1993; Nunnally & Bernstein, 1994).

3.2 Validity

3.2.1 Content validity

A high content validity was pursued by ascertaining that all testable characteristics of ASD were included in OASID. This was carried out in multiple steps. First, a theoretical framework was built based on recent literature about characteristics of ASD in people with sensory and IDs. Second, existing instruments to diagnose ASD such as the O-ADB (Hoevenaars-van den Boom, et al., 2009) and the ADOS (Lord, et al., 1999) were studied to transform the autistic characteristics into testable items. Third, these items and tasks were discussed with experts and clinicians from the field of multiple disabilities and adjusted according to their advice. Finally, we compared our items with criteria in DSM-5 (American Psychiatric Association, 2013), resulting in scales and sub-scales that correspond to these criteria. All DSM-5 criteria for ASD were included. Preliminary versions of OASID were reviewed by all authors and the authors of the O-ADB and discussed with caretakers and clinicians. The two experts who assessed the participants for ASD were not involved in the development of OASID.

3.2.2 Construct validity

OASID was compared with the AVZ-R for convergent validity and with the list of behavioural signs of disturbed attachment for divergent validity. There was no significant correlation between OASID scores and the list of disturbed attachment, $r = .46$

($P = .57$). Although disturbed attachment behaviour may appear similar to ASD symptoms on the surface, disturbed attachment behaviours are not equal to ASD signs, so this lack of correlation indicated good divergent validity.

The AVZ-R is designed for people with IDs alone. It also measures ASD symptoms, and a significant positive correlation was expected; therefore, one-tailed correlation was calculated. The correlation between OASID and the AVZ-R was significant and moderately strong, $r = .40$ ($P = .049$), indicating a moderate divergent validity.

3.3 Differentiation

In this study, the gold standard for ASD was based on consensus of expert judgements. Of the 18 participants, the experts reached consensus in nine participants. For the remaining nine participants, experts either disagreed (one scored yes and one scored no), both doubted the diagnoses or one of the experts doubted, while the other was certain (see Figure 1).

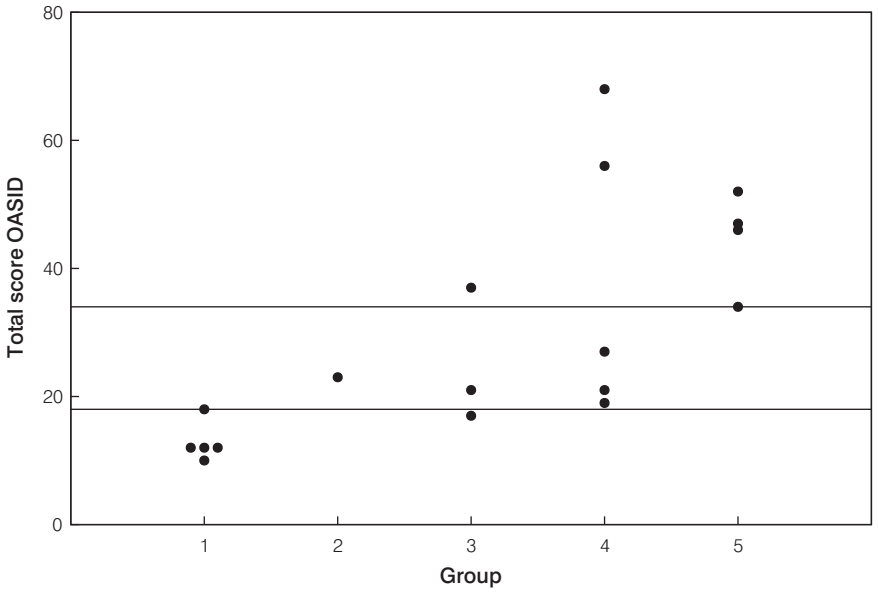

Figure 1 Groups are (1) consensus by two experts of no ASD, (2) no ASD according to one expert, the other doubts, (3) both experts doubt ASD or one was certain of ASD and the other was certain of no ASD, (4) ASD according to one expert and the other doubts, (5) consensus by two experts of ASD.

For Figure 1, five groups were made, with increasing certainty regarding ASD diagnosis. Group 1 contains all the participants without ASD, according to both experts, followed by group 2 where one expert doubted and the other was certain of no ASD. Participants in group 3 consist of participants where both experts doubted or they disagreed. In group 4, one expert doubted, and the other was certain that they had ASD. Finally, participants in group 5 were people with ASD (according to both experts). Two horizontal lines were drawn, corresponding to the lowest score of someone with ASD and the highest score of someone without ASD (groups 5 and 1). As can be seen, there is a large gap between people with and without ASD, and most participants with doubt regarding their diagnosis (groups 2–4) scored in-between these lines. The differentiation between ASD and no ASD is further confirmed with the results from non-parametric tests; see Table 4, which showed significant differences between the ASD and no ASD group for total score, both scales and three of the sub-scales as well.

Table 4 Differences on OASID between people with and without ASD

Scale	ASD M	no ASD M	P
A. Social behavior and communication	28.3	6.2	.008*
1. Reciprocity	10.8	1.2	.008*
2. Communication	4.8	1.4	.008*
3. Relationships	12.8	3.6	.008*
B. Repetitive and stereotyped behavior	16.5	6.6	.008*
1. Stereotyped behavior	7.3	1.2	.008*
2. Insistence on sameness	6.0	4.0	.143
3. Restricted interests	1.8	0.6	.365
4. Reactivity to sensory input	1.5	0.8	.143
OASIS Total SCORE	44.7	12.8	.008*

Note. Diagnosis ASD given by expert consensus. *Significant at .01 level, one tailed.

Of the nine participants that could not receive a definitive diagnosis of ASD or no ASD, five scored between the two horizontal lines. This is not surprising; as experts doubt someone's diagnosis, OASID scores are less conclusive as well. However, there are four cases that caused doubt among experts but scored below the lower or above the upper horizontal line, meaning that despite expert doubt, OASID could diagnose them. One expert doubted the ASD diagnoses of the two participants that received the highest OASID score of all participants. The other expert was certain

that they had ASD. It appeared that the expert in deafblindness, a psychiatrist, was the expert who had some doubts because he believed that their impairments were not caused by ASD but by their ID. The other expert, who has expertise in ASD in people with IDs, however, was certain that they had ASD. The other two participants that score outside the horizontal lines only did so with a few points that may be coincidental. These cases were the two participants that the experts disagreed on, one expert was certain of ASD and the other was certain that there was no ASD.

4. Discussion

Currently, no valid instruments are available to diagnose ASD in people with a combination of sensory and IDs in a wide range. Diagnosing people correctly is important and necessary to contribute to people's needs in living conditions, support and treatment. Because of a large amount of variability in communicative abilities and severity of disabilities, a more adaptive diagnostic approach is needed to adapt to the heterogeneous skills and abilities of people with multiple disabilities. For example, sharing of attention is often carried out in a visual manner, whereas a tactile approach would be more suitable for some people with multiple disabilities (Neerinckx, Vos, Van Den Noortgate & Maes, 2014), and thus, an individual approach is necessary. OASID was designed in this way, and the preliminary findings indicate that it can successfully differentiate between people with multiple disabilities with and without ASD. Not only was the average total score of participants with ASD significantly different from participants without ASD, there were no overlapping scores between these groups.

Consensus among two experts was used as a gold standard for ASD diagnoses. In the cases where an expert doubted whether the participants had ASD, OASID scores pointed in either direction. In one case, doubt seemed to be related to the experts' background. To make sure of giving the right label, only if both experts agreed, participants were included in analysing differences between groups. A limitation of this method is that the two experts used OASID video material, in addition to information from the participant's records, to base their decisions on. Because the OASID raters and the experts watched the same videos, their judgements were not completely independent. However, the experts did not see the OASID questionnaire and scoring rules, making contamination unlikely. At the moment, this is the most appropriate method for diagnosis, as only participants for whom complete consensus was reached were taken into account and no valid diagnostic instruments existed yet for this group.

The total score on OASID, as well as both the Social Behaviour and Communication Scale and the Repetitive and Stereotyped Behaviour Scale, can differentiate people with and without ASD, when the diagnosis is based on expert consensus. On the first

scale, all sub-scales do too. Three out of the four sub-scales in the Repetitive and Stereotyped Behaviour Scale showed no significant differences, but the total for this scale did.

Especially the sub-scales on the social communicative domain showed large differences for people with these disabilities, which is consistent with the earlier research findings with the O-ADB (Hoevenaars-van den Boom, et al., 2009). The lack of differences on sub-scales of repetitive and stereotyped behaviour may be caused by the fact that in people with sensory disabilities, IDs or a combination of these disabilities, these factors are quite common aspects of their behaviour (e.g. Andrews & Wyver, 2005; Fraiberg, 1977; Jan, Freeman, & Scott, 1977; Murdoch, 1997) and may therefore not be differentiating factors in this population, as opposed to in people with IDs alone (Matson et al., 2008). Nevertheless, the total score is differentiating for the two groups; perhaps, if a larger sample had been included, the differences on these sub-scales could have been larger as well. Additionally, some of the sub-scales consist of only three items. Perhaps, clear differences between people with and without ASD on these small sub-scales are not necessary, and a diagnosis can better be based on the total score. The sub-scales may potentially be used for individual treatment purposes, for it can easily be seen in which area the person is impaired the most and needs additional support. After all, the most important goal is to help everyone to obtain the treatment and support that are optimal for each individual.

Two of the 20 participants were unable to finish OASID, and for nine others, the diagnosis was unclear, but after applying our tentative thresholds, this number was reduced to five. These five participants would receive a diagnosis of mild ASD vs. severe ASD in the people above the threshold. With the help of OASID, the number of ASD signs can be placed on a continuum from no via mild to severe ASD. Probably, this provides a more appropriate way of categorising ASD symptoms as opposed to dichotomising in ASD or no ASD, for this spectrum is also found in DSM-5 (American Psychiatric Association, 2013). Despite two outliers, OASID scores were consistent with expert consensus. All participants that received an expert consensus of ASD or no ASD received the same diagnosis with OASID. Future studies should bring down the number of persons for whom OASID and experts disagree.

The lack of diagnostic means for people with multiple disabilities to compare OASID with was solved by using expert consensus as a gold standard for the ASD diagnosis. However, in many cases, only one of the experts was unsure, while the other was confident of his or her diagnosis. This is not surprising as the current target population is difficult to diagnose, because of the aforementioned overlap in behaviour between people with and without ASD (e.g. Cass, 1998; De Vaan, et al., 2013a, Hoevenaars-van den Boom et al., 2009; Knoors & Vervloed, 2011). Additionally, the fact that ASD occurs on a spectrum (American Psychiatric Association, 2013) and is not a dichotomous label can explain the cases in which experts were uncertain

about several of the participants. Because of these difficulties and that they did not consult each other; reaching consensus in half of the participants is an asset and confirms reliability of their diagnoses.

The fact that OASID can differentiate people with and without ASD would be meaningless without evidence of good reliability and validity. Despite the small sample and the fact that this measure is only semi-structured, evidence for substantial to good reliability was found. Unfortunately, content validity could not be supported by statistical tests because of the small sample size, but other steps were taken to assume good content validity. The measures used for construct validity preliminary showed that OASID is a valid tool, as both convergent and divergent validity were indicated. A low correlation was found between OASID and the Pervasive Developmental Disorder in Mental Retardation Scale (Kraijer & De Bildt, 2005), which was expected as both instruments aim to measure ASD, yet in a different group. Divergent validity was confirmed by finding no correlation with the list of disturbed attachment behaviours (Boris & Zeanah, 2005). This was also expected because despite ASD typical behaviours in people without a secure attachment (Rutgers, et al., 2004; Zeanah et al., 2005), people with ASD can still show signs of a secure attachment (Rutgers et al., 2004), so a correlation had to be absent to indicate divergent validity. Inter-rater reliability was substantial, which is especially promising considering that the observations were carried out by two undergraduate students with a minimum amount of experience with ASD and the target population.

A limitation of the current study is the small sample size of 18 participants. Given that OASID is in the early stages of development and the potential target group is small altogether, the low number is acceptable, because we had to reserve potential participants for future research with OASID. All participants were recruited from institutional settings or schools, potentially harming representativeness of the sample. In the Netherlands, however, people with these types and combinations of disabilities primarily live in institutional settings (Evenhuis, Theunissen, Denkers, Verschuure & Kemme, 2001), and as such, the sample is representative of the study population. Representativeness was further enhanced by recruiting participants from different settings throughout the country.

The O-ADB was a successful tool in differentiating people with and without autism when participants were deafblind and had profound IDs (Hoevenaars-van den Boom et al., 2009). Our goal of broadening the target population to people with less severe intellectual and sensory disabilities was reached by including also people without auditory impairments and with a moderate and severe ID. OASID consisted of a fewer number of tasks than the O-ADB, making the administration also less tiresome and stressful for participants.

Benefits of OASID as opposed to other diagnostic tools are that it is specifically designed for this complex group of multiply disabled persons and that it uses

observations of a situation in which ASD typical behaviours are evoked from the participants as opposed to observations performed in natural surroundings or to questionnaires. The benefit of an observational tool as opposed to a questionnaire is that actual behaviour of a participant is tested vs. the interpretations of the behaviour by only one rater. Where testing in a natural setting complicates the potential recognition of ASD typical behaviour, this study showed that testing in an experimental setting with OASID makes it possible to recognise ASD. Additionally, existing questionnaires may not be adaptive enough to account for the variability often found in this population of multiply disabled people, while administration of OASID is adjusted to the individual participant. Another advantage of OASID is that its scales are consistent with DSM-5 criteria (American Psychiatric Association, 2013), and its scores reflect the continuum or spectrum that DSM-5 also proposes. Regardless of these positive results, it remains important that OASID is integrated in a broader diagnostic assessment, including multiple tests and using more than one informant (Carnaby, 2007).

It must be noted that this study was only the first step in the development of OASID and more research is necessary. Our results show that in a small group of participants, OASID could differentiate between people with and without ASD when they have IDs and sensory impairments and potentially reduce the group of people for whom there are doubts.

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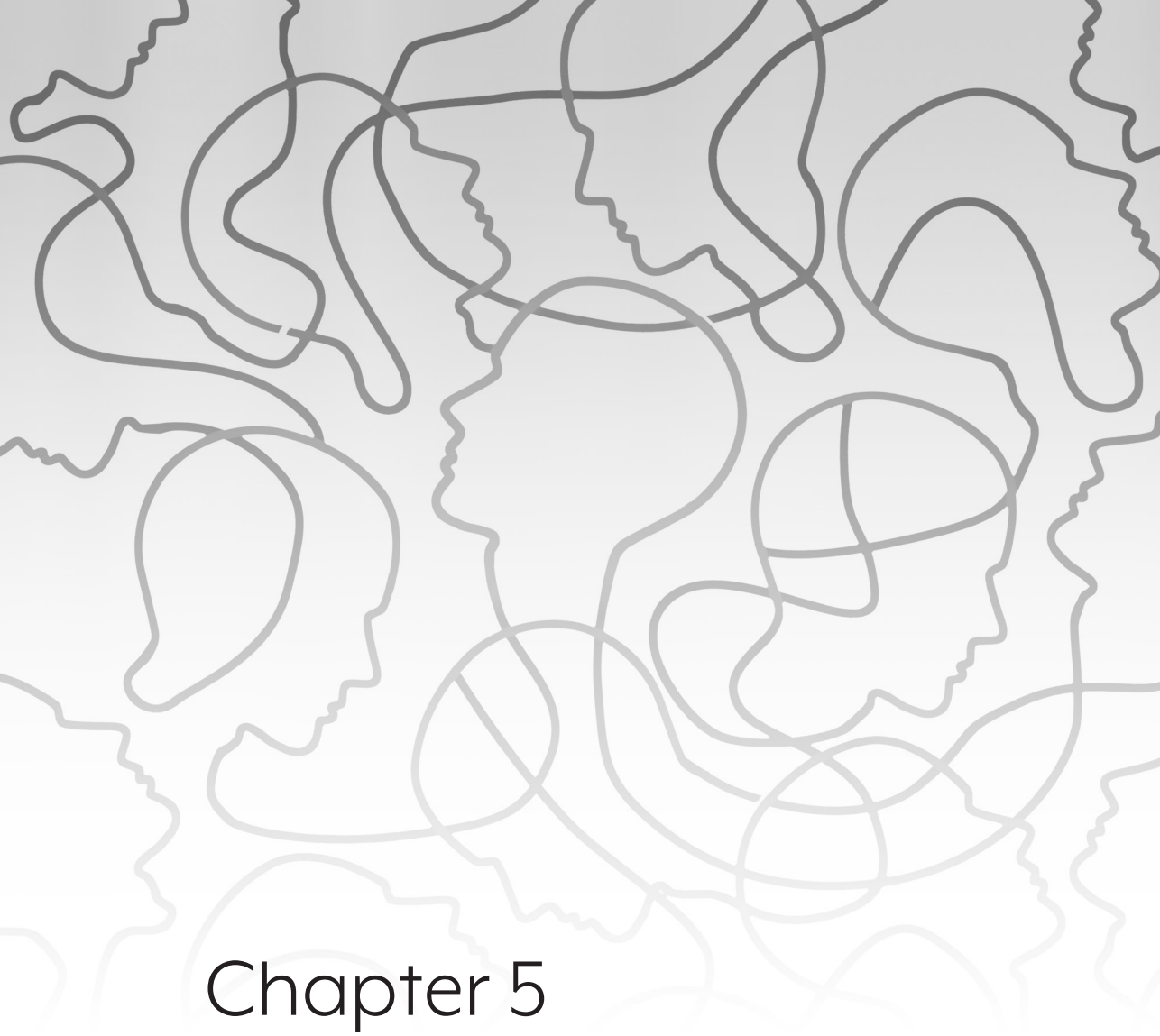

Chapter 5

Assessing autism spectrum disorder in people with sensory impairments combined with intellectual disabilities

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Abstract

People with sensory impairments combined with intellectual disabilities show behaviours that are similar to Autism Spectrum Disorder (ASD). The instrument Observation of Autism in people with Sensory and Intellectual Disabilities (OASID) was developed to diagnose ASD in this target group. The current study focuses on the psychometric properties of OASID.

Sixty individuals with intellectual disabilities in combination with visual impairments and/or deafblindness participated in this study. The OASID assessment was administered and rated by three independent observers. By means of expert consensus cut-off scores for OASID were created. To determine the concurrent validity OASID was compared with the Pervasive Developmental Disorder for People with Mental Retardation (PDD-MRS) and the Childhood Autism Rating Scale second edition (CARS-2).

The intra-rater reliability, the inter-rater reliability, internal consistency and concurrent validity of OASID were good to excellent. Cut-off scores were established based on criteria from the DSM-5. OASID was able to differentiate between four severity levels of ASD.

1. Introduction

People who have intellectual disabilities combined with visual impairments or deafblindness show impairments that may also occur in autism spectrum disorder (ASD) (de Vaan et al. 2013; Hoevenaars-van den Boom et al. 2009; van Gent 2012). ASD is characterised by impairments in communication and social interaction, repetitive and stereotyped behaviour and resistance to change (American Psychiatric Association 2013). These impairments, however, are not exclusive to ASD. Some of these impairments are, for example, also seen in people with social communication disorders (American Psychiatric Association 2013), insecure attachment styles (Zeanah et al. 2005; Rutgers et al. 2004), combined sensory and intellectual disabilities (de Vaan et al. 2016b) and even in typically developing children (Frith 2003). In order to diagnose and classify atypical behaviour correctly it is important to establish what causes a person's behaviour. Determining the aetiology of behaviour is required for choosing an appropriate intervention strategy. To this end the diagnostic instrument 'Observation of Autism in people with Sensory and Intellectual Disabilities' (OASID) was developed in a pilot study (de Vaan et al. 2016b). The aim of OASID is to correctly diagnose the presence of ASD in individuals with a moderate to profound intellectual disability, combined with a visual impairment or deafblindness. The pilot study showed good inter-observer agreement, excellent internal consistency and adequate content and construct validity (de Vaan et al. 2016b). The pilot, however, studied only 18 people making it hard to calculate cut-off scores. The current paper focuses on improving the calculations of the psychometric properties: firstly by including more participants, secondly by adding an extra instrument for calculating the construct validity, and thirdly by proposing cut-off scores.

It is difficult to correctly interpret ASD symptoms in persons with combined sensory and intellectual disabilities because symptoms are often not unique to ASD alone. Persons with hearing loss and visual impairments can show drawbacks and peculiarities in language and communication (Knoors and Vervloed 2011; Wolters et al. 2011; Cass 1998; Dale et al. 2014; Fraiberg 1977; Gense and Gense 2005) and a delay in the development of Theory of Mind (Peterson and Siegal 2000; Peterson et al. 2000). These impairments are similar to some diagnostic criteria for ASD that were mentioned earlier. In addition, people with visual impairments also show frequently stereotyped behaviours (Tröster et al. 1991). Finally, an intellectual disability can also be the cause of behaviours that are often found in ASD, with symptom overlap becoming larger as the intellectual disability is more severe (Matson et al. 2008; Matson and Shoemaker 2009). The combination of sensory and intellectual disabilities make it even more likely that these people show behaviour patterns often seen in ASD (Carvill 2001). Especially in the domains of social and communicative development and daily living skills people with both sensory and intellectual

disabilities show many typical features of ASD (Dalby et al. 2009; Evenhuis et al. 2009; Hoevenaars-van den Boom et al. 2009; Munde and Vlaskamp 2014). Examples of these behaviours include: stereotyped movements, such as hand waving and body rocking (Gense and Gense 2005; Medeiros et al. 2014), lack of reciprocity in social interaction (Dale et al. 2014), poor use of language for social purposes and awkward pragmatic language use (Tadić et al 2010). Prevalence of ASD in people with sensory or intellectual disabilities is much higher than the 1-2% normally found in the total population (CDC 2017). Almost half (about 56%) of children identified with ASD have below average intellectual ability (CDC 2017). According to Jure et al. (2016) ASD is 30 times more frequent in children who are blind than in the sighted population and Cass (1998) mentioned that about one third of totally blind children show symptoms of ASD. ASD is also common in deaf children although this seems to be related more to comorbid intellectual disabilities than to deafness per se (Jure et al. 1991).

In addition to the overlap in behaviour, there is a lack of diagnostic and assessment instruments that are suitable for people with the above mentioned disabilities (Bodsworth et al. 2011; Hoevenaars-van den Boom et al. 2009; Jure et al. 1991; de Vaan et al. 2016a). In most cases diagnostic instruments do not have norms for people with combined sensory and intellectual disabilities; see for instance the manuals of the ADOS (Lord et al. 1999), ADI-R (Rutter et al. 2003) and a validity study on the Autism Behavior Checklist (Krug et al 1980). Furthermore, testing procedures do not often take sensory impairments into account (Tobin and Hill 2011). Sometimes researchers and clinicians adjust diagnostic instruments for people with sensory impairments, but these adjustments are usually not validated or are found inappropriate by Williams et al. (2014). Because of the high prevalence of symptoms of ASD in people with multiple disabilities, even persons without ASD can score above the ASD cut-off on diagnostic instruments (Dammeyer 2014). For these reasons, there is an urgent need for the development of new assessment procedures for people with sensor or intellectual disabilities, or a combination of both (Nakken and Vlaskamp 2007; Tobin 1994).

ASD or ASD-related features among congenitally blind, deaf and deafblind children have been reported in clinical studies (see Dammeyer 2014). The main problem with these studies is the use of instruments that are invalid for these populations (de Vaan et al. 2016a). Despite the difficulties in diagnosing ASD in people with combined sensory and intellectual disabilities, research has shown it is possible to differentiate people with and without ASD (Dammeyer 2014; de Vaan et al. 2016b; Hoevenaars-van den Boom et al. 2009). The development of the OASID fits into this line of research. OASID was developed to help to diagnose ASD in people with combined sensory and intellectual disabilities. OASID is a semi-structured observational instrument for diagnosing ASD in this target population. In an earlier

study with 18 participants, OASID proved to be a reliable and valid diagnostic instrument (de Vaan et al. 2016b). Given the small size of the study sample, replication with a larger group of participants was deemed necessary. Note that OASID is not designed to replace a full psychological assessment, including interviews, patient history, observations and testing. OASID is intended as a useful addition to existing assessment methods.

In the current study, people are included with moderate to profound intellectual disabilities, combined with visual impairment or deafblindness. Though the term deafblindness might imply complete lack of both sight and hearing, it can also be defined as any given combination of visual and auditory impairments (Dammeyer 2012; Ask Larsen and Damen 2014). The latter definition was used in this study. This paper consists of a description of some psychometric properties of OASID and a proposed heuristic to classify people with ASD that is in line with the current classification of ASD in the DSM-5 (American Psychiatric Association 2013).

2. Methods

2.1 Participants

Participants were 60 individuals (42 male, 18 female) with ages ranging between 6-55 (M = 31.6, SD = 14.9), 17 of which were children (18 or younger). The broad age range was used to ensure ample potential participants. Earlier studies have indicated that in persons with intellectual disabilities, especially when over 65, the prevalence of dementia is significantly higher than in the typical population (Cooper 1997; Strydom et al. 2009). To prevent possible interference of behaviours that are due to dementia or old age, a conservative maximum age, 60 instead of 65, was used as exclusion criterion. Participants had moderate (n=11), severe (n=24) or profound (n= 25) intellectual disabilities. A total of 34 participants used verbal language to communicate, ranging from a few words to full sentences, whereas two used sign language, two used tactile sign language, and 22 used no language or communication system at all. OASID administrators spoke verbally to participants or with a combination of verbal and sign language. All participants had a visual impairment, 30 of them were blind with or without light perception. Of all participants 16 were deafblind. Information regarding intellectual disability and sensory impairments was collected from the records of the participants and were established in the past by licenced psychologists, physicians, ophthalmologists and audiologists independent of the current study. According to the medical records the aetiologies of the disabilities were: prematurity (n=8), brain damage (n=6), congenital rubella syndrome (n=5), Down syndrome (n=5), Leber's amaurosis (n=4), Goldenhar syndrome (n=2), Angelman syndrome (n= 1), consanguineous parents (n=1), Bardet-Biedl syndrome

(*n*=1), other birth deficits (*n*=11). For 16 participants the aetiology was unreported or unknown. Participants were recruited in collaboration with four residential institutions and three schools for people with intellectual and sensory disabilities throughout the Netherlands. To maintain privacy and anonymity until participation was confirmed, recruitment was performed entirely by institutional staff, and thus not the research team. The selection criteria were: a moderate to profound intellectual disability combined with a visual impairment or blindness according to the criteria of the ICD-10 (World Health Organization 2016), or deafblindness, which was defined as any combination of a visual and auditory impairment.

Within our study, no subgroups were made based on age, level of intellectual disability or level of sensory impairments. This was done since, with the current number of participants and the number of potential subgroups, the number of participants per group would be very small, resulting in limited statistical power.

2.2 Materials

2.2.1 Observation of Autism in people with Sensory and Intellectual Disabilities

OASID is a semi-structured observational assessment for ASD in people with combined sensory and intellectual disabilities (de Vaan et al. 2016b). An experimenter conducted five tasks with each participant in a playful manner, adjusting communication and play level to their abilities and impairments. For example, one task consisted of a puzzle with four different degrees of difficulty that could be adjusted to the participant's cognitive and motor abilities. Communication about the puzzle was achieved through spoken language, sign language, tactile sign language or by simply presenting puzzle pieces, depending on the participant's communication style.

The play session was recorded on video and scored offline to a 40-item checklist. Items had three possible scores, ranging from 0 to 2, reflecting absent, intermediate or full presentation of features of autism.

All items were accommodated to the participant's level and type of impairment. For example, it was asked if the participant responded to initiations for contact or sought the researcher's attention, whether by eye contact or alternative means if the participant was blind. Additional examples of seeking contact in an alternative way were given, for example 'taking the researcher's hand' or 'talking to the researcher'.

Scores on individual items were added to obtain a total score and a score on two scales, namely: 'Social Behaviour and Communication' and 'Repetitive and Stereotyped Behaviour'. These two scales were based on the domains of ASD described in the DSM-5 (American Psychiatric Association 2013). In line with the criteria described in the DSM-5, seven subscales were defined. The first three subscales are part of the first scale, namely 'reciprocity', 'non-verbal communication' and 'relationships'. The following four subscales are part of the second scale 'stereotyped and repetitive behaviours', 'insistence on sameness', 'restricted and

fixated interests' and 'hyper- or hypo reactivity to sensory input'. High scores indicated more ASD typical behaviours. These diagnostic criteria were transformed into testable items based on existing items in the O-ADB (Hoevenaars-van den Boom et al. 2009) and the ADOS (Lord et al. 1999), in addition to expert experiences and observations of how diagnostic criteria may express themselves in the current target populations. An earlier study found the reliability and validity of OASID to be good (de Vaan et al. 2016b). The inter-rater reliability was demonstrated by a weighted kappa of 0.75 and an intraclass correlation coefficient of 0.69. The internal consistency showed a Cronbach's alpha of .94 for both scales. Construct validity in the pilot study was established by looking at divergent and convergent validity. A lack of a significant correlation with the list for disturbed attachment was found , $r = .46$, $p = .57$ and a positive correlation with the PDD-MRS (as described below), $r = .40$, $p = .049$ respectively.

2.2.2 Pervasive Developmental Disorders in Mental Retardation Scale

In the current study the original Dutch version of the Pervasive Developmental Disorders in Mental Retardation Scale (PDD-MRS; Kraijer and de Bildt 2005; Kraijer 1999) was used to determine concurrent validity. The PDD-MRS is a 12-item questionnaire designed to diagnose ASD or Pervasive Developmental Disorders (PDD) in people with intellectual disabilities. Questions can be answered with a positive or a negative score; all the positive scores were counted and weighed to result in a total score. Scores of 10 and above indicated ASD, scores of 6 and below indicated no ASD; scores that were in between gave uncertain results. The PDD-MRS is found to have good inter-rater reliability, test-retest reliability and internal consistency (Evers et al. 2010; Meadows 2007). Content validity was also good, criterion validity was sufficient, and sensitivity and specificity were good (Evers et al. 2010; Kraijer 1999; Meadows 2007; O'Brien et al. 2001).

The PDD-MRS was chosen as a measure of concurrent validity within this study because it is one of the few instruments that was specifically designed for ASD in people with intellectual disabilities (Kraijer and de Bildt 2005). Additionally, it is an originally Dutch measurement and its interpretations are based on a Dutch sample, increasing the validity for use in our Dutch sample.

2.2.3 Childhood Autism Rating Scale 2

The Childhood Autism Rating Scale, Second Edition (CARS2; Schopler et al. 2010) was used to determine the concurrent validity of OASID. The CARS-2 is a screening tool for ASD in children, consisting of 15 items that can each be scored on a scale ranging from 1 point for normal behaviour to 4 points for severely abnormal behaviour. Half points can also be given, making it a 7-point Likert scale. A total raw score is calculated by adding scores on all 15 items. Higher scores indicate more severe

symptoms of autism (Schopler et al. 2010). Reliability of the CARS-2 is reported to be satisfactory (McLellan 2014; O'Brien et al. 2001) and validity is moderate to good (Malcolm 2014).

The CARS-2 was chosen as an additional measure for validity within this study because of its psychometric properties. Additionally, it could easily be conducted using the tasks of OASID, without further burdening participants with additional testing.

2.3 Procedure

Ethical approval for the study was gained from the Committee on Research Involving Human Subjects Arnhem-Nijmegen and conforms to World Medical Association declaration of Helsinki on the Ethical Principles for Medical Research Involving Human Subjects (World Medical Association 2013). As participants were deemed incapacitated, legal representatives of participants were asked for informed consent. Legal representatives were informed that the assessment would stop if there were reasons to believe the participant was unwilling to continue the assessment. This was never necessary.

Participants were tested within their own institution or school by one of three trained administrators. Assessments were done in quiet rooms with few if any stimuli to avoid distraction, with a familiar caregiver but without other clients present. Administration of OASID took between 20 and 55 minutes (36 minutes on average). After administration of OASID, caregivers were asked to fill out the PDD-MRS and provide record information regarding background, intellectual disability, and visual and auditory impairment.

The OASID assessment was recorded on video and was scored afterwards. The first author scored all videos. The second and third observers (two Master's students in Educational and Pedagogical Science, both with experience with this target population and OASID assessments) scored 42 and 43 videos respectively, in order to assess inter-rater reliability. These second and third observer also scored 10 videos twice (after at least one month) to assess intra-rater reliability. Two additional Master's students in Educational and Pedagogical Science administered the CARS-2 by observing the OASID video material.

Two experts in the field of ASD and intellectual disabilities and/or deafblindness independently observed videos of 14 randomly selected participants. One expert is a child psychiatrist with expertise in the field of deafblindness. The other expert has a PhD in special education and ample experience in diagnosing ASD in children with an intellectual disability. In addition to OASID video material, both experts received brief information from the participants' records and a list of ASD criteria based on the DSM-5. The experts were blind to the protocol of OASID and method of scoring to prevent contamination between OASID scoring and their judgments. The expert

judgments served as the gold standard for determining cut-off scores in this study. Because of the scope of the project and available funds we were able to have both experts observe 14 extra random cases. In addition to these 14 randomly selected participants from the current study, we added the 18 expert judgments from our previous study (de Vaan et al. 2016b), because these judgments were done in exactly the same manner by the same two experts, which enabled data pooling. As a result we had a larger number of expert judgments on which to base cut-off scores within the current study.

2.4 Statistical Analyses

Intra- and inter-rater reliability was determined with weighted Cohen´s Kappa (Fleiss and Cohen 1973). Weighted kappa gives a more adequate estimation of reliability than unweighted kappa, since absolute differences between scores are taken into account. An intra-class correlation coefficient (ICC) with consistency in a two-way mixed model was used to assess reliability between three raters, as this corresponds best to weighted kappa (see Hallgren 2012). Reliability is deemed substantial when the kappa value is above 0.60, and almost perfect when it is above 0.80 (Landis and Koch 1977). ICC is good above 0.60 and excellent above 0.75 (see Barret 2001).

Internal consistency of scales was assessed with Cronbach's alpha, with a required minimum of 0.7, but ideally near 0.9 (Kline 1993). However, in scales with a low number of items a lower Cronbach's alpha is acceptable, as Cronbach's alpha is very sensitive to number of items and will underestimate reliability when there are few items (Cortina 1993).

Concurrent validity was assessed by calculating Spearman-rank correlations between OASID scores and the PDD-MRS and CARS-2 scores as all these instruments aim to measure the same construct, which is ASD. In addition, expert judgments were also used to assess concurrent validity of OASID scores. Based on expert judgments the participants were split up into five groups: (1) both experts agreed on no ASD, (2) one expert was certain of no ASD, one expert doubted, (3) both experts doubted or they disagreed, (4) one expert was certain of ASD, one expert doubted, and (5) both experts agreed on the presence of ASD. To establish concurrent validity and cut-off scores, the 18 judgments from an earlier study (de Vaan et al. 2016b) were added to the 14 expert judgments in the current study

3. Results

3.1 Intra-rater reliability

Two observers scored 10 videos they had scored before for a second time. The first observer had 89.3% exact agreement, corresponding to a weighted kappa of 0.89. The second observer had 89.4% exact agreement, corresponding to a weighted kappa of 0.90. According to the criteria of Landis and Koch (1977) the intra-rater reliability was almost perfect.

3.2 Inter-rater reliability

All combinations of data from observers resulted in a weighted kappa of 0.63. The ICC over all three observers was also 0.63. The ICC for scale A 'Social behaviour and communication' was 0.64 and for scale B 'Repetitive and Stereotyped behaviour' 0.60. The subscale levels' ICC are depicted in Table 1. According to the guidelines of Cicchetti (1994) these ICC's are rated as 'good'. The total and scale scores of OASID were highly correlated between all possible observer pairs; see Table 2.

3.3 Internal consistency

Cronbach's alpha for scale A 'social Behaviour and Communication' was 0.91 and for scale B 'Repetitive and Stereotyped Behaviour' 0.85. These values can be interpreted as excellent and good, respectively (DeVellis 2012). The internal consistencies for the subscales are depicted in Table 1.

Table 1 Reliabilities of Scales and Subscales of OASID

	Number of items	Cronbach's Alpha	ICC
Scale A "Social Behavior and Communication"	**21**	**0.91**	**0.64**
Reciprocity	9	0.80	0.66
Non-verbal Communication	3	0.59	0.65
Relationships	9	0.85	0.60
Scale B "Repetitive and Stereotyped Behavior"	**19**	**0.85**	**0.60**
Stereotyped and repetitive movements	7	0.73	0.70
Insistence on sameness	6	0.72	0.51
Restricted and fixated interests	3	0.43	0.60
Hyper- or hypo reactivity to sensory input	3	0.32	0.51
Total	**40**	**0.94**	**0.63**

Table 2 Correlations between OASID Scores

	Rater 1 and rater 2 (n=43)	Rater 1 and rater 3 (n =42)	Rater 2 and rater 3 (n=42)
OASID total score	0.93*	0.92*	0.93*
Scale A "Social Behavior and Communication"	0.93*	0.90*	0.91*
Scale B "Repetitive and Stereotyped Behavior"	0.83*	0.82*	0.87*

Note. * $p < 0.01$

3.4 Concurrent validity

To assess concurrent validity, a one-tailed Spearman's rank correlation was calculated between the total scores on OASID and the total scores on the PDD-MRS and the CARS-2. A small significant correlation was found between total scores on OASID and the PDD-MRS, $\rho = .243$, $p = .038$. The CARS-2 was rated by two observers. The total scores on OASID were moderately to highly significantly correlated with the total scores on the CARS-2 for both observers, $\rho = .652$, $p < .001$ and $\rho = .801$, $p < .001$, respectively.

A moderately strong Spearman-rank correlation was found between OASID scores and the ranks formed by the combined expert judgements regarding the presence of ASD, $\rho = 0.67$, $p < 0.001$.

3.5 Cut-off scores

Most diagnostic and screening instruments for ASD provide one total score on which the final classification is based. Scores above the cut-off score are indicative of ASD. However, according to the definition of ASD in the DSM-5 (American Psychiatric Association 2013) this scoring method has two problems. Firstly, ASD occurs on a spectrum and is not merely a dichotomous label of ASD and no ASD. After one receives the diagnosis of ASD, that person can be classified within different levels of severity, to which different levels of required support correspond. Working with one cut-off score does not take into account these severity levels implicated in the DSM-5. Secondly, the DSM-5 clearly states that impairments in both domains of ASD must be present in order to diagnose ASD. A single total score could indicate impairments in both domains, but not necessarily, because it could also indicate the presence of many impairments in only one domain. In the latter case, when persons are diagnosed with ASD based on a high total score, they may not have all the required symptoms to assign the label ASD.

Because of these two problems with current scoring protocols, we propose a new protocol for scoring OASID, one that takes into account the spectrum of impairments in both domains. Firstly, a person must score high enough in both

domains of ASD to receive an ASD classification (scale A 'social behaviour and communication' and scale B 'repetitive and stereotyped behaviour'). Secondly, the resulting scores must contain information on ASD severity, namely more or fewer symptoms on the continuum of ASD.

The judgments by the two independent experts were used as the gold standard for determining cut-off scores. Cut-off scores were made for both scales separately. The experts reached exact agreement about the presence or absence of ASD in 13 cases. In 12 cases one expert doubted the presence of ASD and in 7 cases both experts doubted or they disagreed on the presence of ASD. For determining cut-off scores, only participants for whom the experts reached complete agreement were taken into account.

For both scales cut-off scores were determined by taking the highest scale score of the participant for whom there was consensus that ASD was not present and the lowest scale score of the participant with consensus on the presence of ASD. Therefore, both scales consist of two cut-off scores. Participants with scores below the lowest cut-off score were categorised as not showing symptoms of ASD on that scale. Scores between the two cut-off scores depicted mild symptoms of autism and scores above the highest cut-off score depicted true symptoms of autism on that scale. Table 3 shows the cut-off scores on both OASID scales and the corresponding classifications. As mentioned earlier, the DSM-5 states that impairments in both domains need to be present. In line with the DSM-5, a diagnosis of ASD can only be made when scores on OASID above the cut-off score on both scales are reached. Since ASD severity is distributed along a spectrum, the classifications of scores were also made according to this spectrum. Table 4 shows the classification of possible scores.

Table 3 Cut-off scores for OASID Scales

	Score on scale A 'Social Behaviour and Communication'	Score on scale B 'Repetitive and Stereotyped Behaviour'
No autistic symptoms	11 and below	7 and below
Mild autistic symptoms	12 – 17	8 – 11
Severe Autistic symptoms	18 and above	12 and above

Table 4 Severity of ASD symptoms

Score on OASID	Interpretation
No autistic symptoms on both scales	No ASD symptoms
No autistic symptoms on one scale, mild symptoms on other scale	No ASD symptoms
No autistic symptoms on one scale, severe symptoms on other scale	Mild ASD symptoms
Mild autistic symptoms on both scales	Mild ASD symptoms
Mild autistic symptoms on one scale, severe symptoms on other scale	Severe ASD symptoms
Severe autistic symptoms on both scales	Profound ASD symptoms

Note. OASID comprises two scales, which are (A) Social behaviour and communication, and (B) stereotyped and repetitive behaviours. Symptoms of ASD must be present on both scales in order to diagnose ASD. To interpret the severity of ASD, an interpretation of symptom severity is required; this can be derived from Table 3.

3.6 Potentially confounding factors

Afterwards possible confounding factors for the ASD classification were checked. No correlation between age and OASID score was found, $r = -.089$, $p = .51$, suggesting that OASID scores are most likely unrelated to and not confounded by age.

Chi square tests were performed to assess the proportion of the different levels of visual impairments, auditory impairments and intellectual disabilities among the four proposed groups of ASD (see paragraph cut-off scores). Three visual impairment groups were made: (1) visually impaired but uses sight, (2) blind with light perception and (3) blind without light perception. Chi square tests revealed that levels of visual impairments were not associated with ASD groups, χ^2 (6, $n = 60$) = 7.87, $p = .25$. Three auditory impairment groups were made: (1) no auditory impairment, (2) auditory impairment and (3) deaf. Level of auditory impairment was not associated with ASD groups, χ^2 (6, $n = 60$) = 8.48, $p = .21$. For level of intellectual disability three groups were made: (1) moderate intellectual disability, (2) severe intellectual disability and (3) profound intellectual disability. Level of intellectual disability was associated with ASD groups, χ^2 (6, $n = 60$) = 23.27, $p = .001$. Persons with profound intellectual disability were more often classified with profound ASD (60%) than people with a severe intellectual disability (12, 5%). No one with a moderate intellectual disability was in this ASD group.

4. Discussion

Because it is difficult to assess the presence of ASD in people with combined sensory and intellectual disabilities, OASID was developed to assist in this process. The current study tested the reliability and validity of OASID on a group of participants

with a moderate to profound intellectual disability combined with a visual impairment or deafblindness. This study elaborated on a previous study which described the psychometric properties of OASID for a relatively small sample of 18 participants (de Vaan et al. 2016b). The results of the current study with 60 participants showed excellent intra-rater reliability, good inter-rater reliability, good internal consistency of scales and good concurrent validity of OASID with two other instruments and expert judgement. On subscale level, reliability was low only for the subscales with few items (i.e. 3 items), namely 'non-verbal communication', 'restricted and fixated interests' and 'hyper- or hypo reactivity to sensory input'. This does not necessarily mean that the scales are not reliable, since Cronbach's alpha underestimates reliability when there are a small number of items (Cortina 1993). For clinical interpretations of individual OASID scores, we recommend the use of only the scale scores and total scores, not the scores on discrete subscales.

To establish concurrent validity, correlations were calculated between OASID scores and scores on the PDD-MRS, CARS-2 and expert judgments. A moderately strong correlation was found between OASID scores and expert judgments. Partly, the experts based their judgments on video material of the OASID assessment, but they had no insight into how OASID was scored; they only watcged play sessions. Therefore, we believe contamination between OASID and the expert's opinion is kept to a minimum. We found significant correlations between OASID scores and the PDD-MRS and the CARS-2 scores, which points to good concurrent validity. This was expected because both instruments were also developed to assess the presence of ASD. The correlation between the OASID and both expert judgments and the CARS-2 scores were moderately strong to strong. This is in contrast to the correlation with the PDD-MRS, which was significant but also small.

The relatively high correlation between OASID, the expert judgments and the CARS-2 scores may partly be due to the fact that they were all based on the same video recordings. However, this also means that contextual factures are the same and cannot be responsible for variation in outcome of the different instruments. The PDD-MRS was chosen as a measure of concurrent validity because it is one of the few instruments available that is specifically developed for assessing ASD in people with intellectual disabilities (Kraijer and de Bildt 2005). Nevertheless, the PDD-MRS was not developed for use in people with additional sensory impairments, hence explaining the rather low correlation with OASID. To estimate the severity of ASD symptoms, OASID is probably a better fit for people with sensory and intellectual disabilities than the CARS-2 and PDD-MRS.

In the second part of this study, a heuristic was proposed for scoring OASID. The DSM-5 (American Psychiatric Association 2013) states that ASD consists of two behavioural domains and that impairments in both domains need to be present before a classification of ASD can be established. It further acknowledges that when

someone has ASD, the behaviours occur on a severity spectrum, leading to different levels of ASD instead of only a strict distinction between ASD and no ASD. Diagnostic instruments should therefore take these severity levels into account (Mehling and Tassé 2015). Furthermore, behaviours symptomatic of autism need to be scored in both behavioural domains described in the DSM-5. When diagnosing ASD with a total score alone, a high score does not necessarily mean that impairments occur in both domains. In contrast to most of the existing diagnostic instruments, the advantage of our heuristic in establishing ASD severity is that it takes into account the score in both ASD domains and distinguishes between different levels of ASD severity. The latter resulted in four possible levels of severity: no, mild, severe and profound ASD symptoms.

The current study focused on a rather broad age range within our target population. Both children and adults were taken into account in the development of OASID. This is in contrast to most studies, where ASD instruments are usually aimed at children alone or have separate norms for children and adults (de Vaan et al. 2016a). However, since all participants have a moderate to profound intellectual disability and impaired communicative abilities, we do believe they can be grouped together for the purpose of the current study. This belief was strengthened by a lack of a correlation between age and OASID scores. Level of visual impairment and level of auditory impairment were also taken into account as possible confounders. However, chi-square tests indicated that level of visual and auditory impairments were equally distributed across ASD groups; hence we do not believe these impairments severely affected OASID scores. Level of intellectual disability, however, was associated with ASD group classification. Specifically, compared to the other ASD groups, in the group with profound ASD symptoms, a high proportion of people with profound intellectual disabilities were found. This is not uncommon since there is ample evidence that the prevalence of ASD is higher in people with intellectual disabilities than in people without, and that severity of intellectual disability and severity of ASD are related (Matson and Shoemaker 2009; O'Brien and Pearson 2004; de Bildt et al. 2005). The current results are in line with these findings. This study was based on the results of 60 participants, making the use of subgroups based on disabilities or age statistically difficult. For future research, and for the further development of OASID, it is recommended that OASID is tested on larger groups of participants, to fully study the effects of age, level of intellectual disability and level of sensory impairments. Only after these studies would it be possible to determine if different cut-off points are required for specific subpopulations.

A possible limitation of the current study is the gold standard used for determining the cut-off scores, namely using consensus judgements of two experts who used video material and brief anamnestic information on the participants. Though the ADOS (Lord et al. 1999) and ADI-R (Rutter et al. 2003) are often seen as a gold

standard in ASD diagnoses (de Bildt et al. 2004; Reaven et al. 2008), the ADOS was found over-inclusive in individuals with an intellectual disability and many tasks could not be assessed in individuals with severe disabilities (Sappok et al. 2013). The ADI-R is only found suitable for people with a developmental age of above two years (Rutter et al. 2003). Because the ADOS and ADI-R are not suitable for our population, we chose to use expert judgments in addition to two other instruments that we felt came closest to being valid instruments for our population. We readily admit that the Autism Behavior Checklist (ABC) as used by Dammeyer (2014) could have been an alternative, but administering this checklist did not fit our study plan. Moreover, although the ABC was validated on people with intellectual disabilities and deaf-blindness, it is also not validated for people with a combination of these disabilities. Since up to now no valid instruments exist to diagnose ASD in people with combined sensory and intellectual disabilities (de Vaan et al. 2016a), it was not possible to use results of other instruments as the gold standard. Where no gold standard exists, expert consensus is a commonly used method.

Unfortunately, in many participants no consensus was reached because either one or both of the experts doubted the presence of ASD. This could be partly caused by the limited information they received about participants. A formal diagnosis of ASD can only be made by a multidisciplinary team in a multimethod assessment procedure combined with anamnestic information (Risi et al. 2006; Rutter 2006; Volkmar et al. 2014). In addition, the experts' classifications were based on their final decision concerning the presence of ASD, not on their judgments on individual behavioural characteristics or the severity of symptoms. Future research could focus on the criteria that experts use to come to their decisions regarding the presence of ASD, and also on designing guidelines for best practices to come to a clinical diagnosis of ASD.

In conclusion, OASID proved to be a reliable and valid tool that scores ASD in line with DSM-5 criteria, expert judgments and scores on two instruments. It was developed specifically for people with combined intellectual disabilities and sensory impairments, aimed at overcoming the risk of over- diagnosing ASD in this group. The current study elaborated on a pilot study with a larger sample of 60 participants. The results of this study indicate that OASID can be a useful tool in assessing behaviour of individuals with combined sensory and intellectual disabilities. Reference points were established in order to interpret the severity of ASD symptoms, which adds to the clinical usability of OASID.

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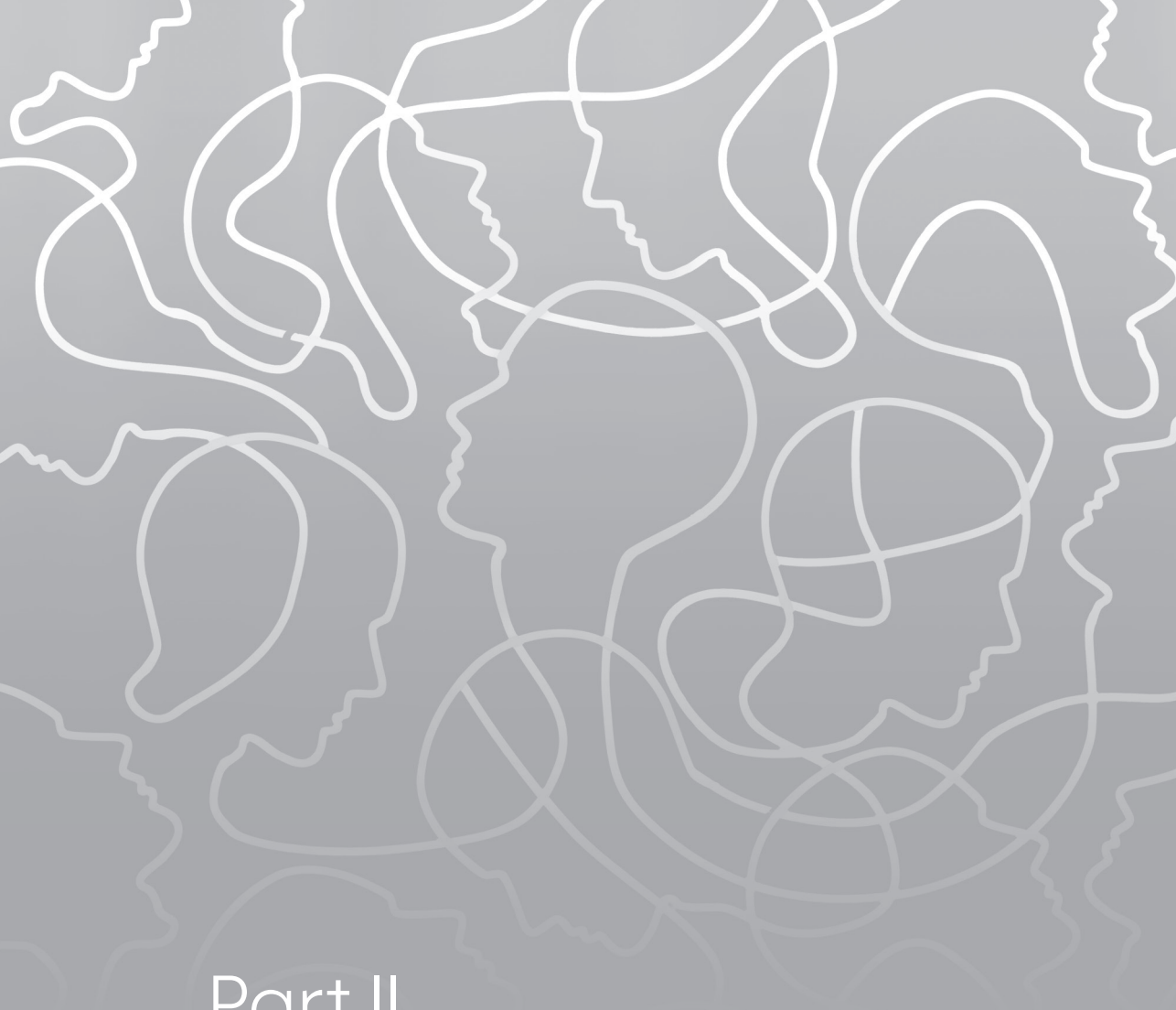
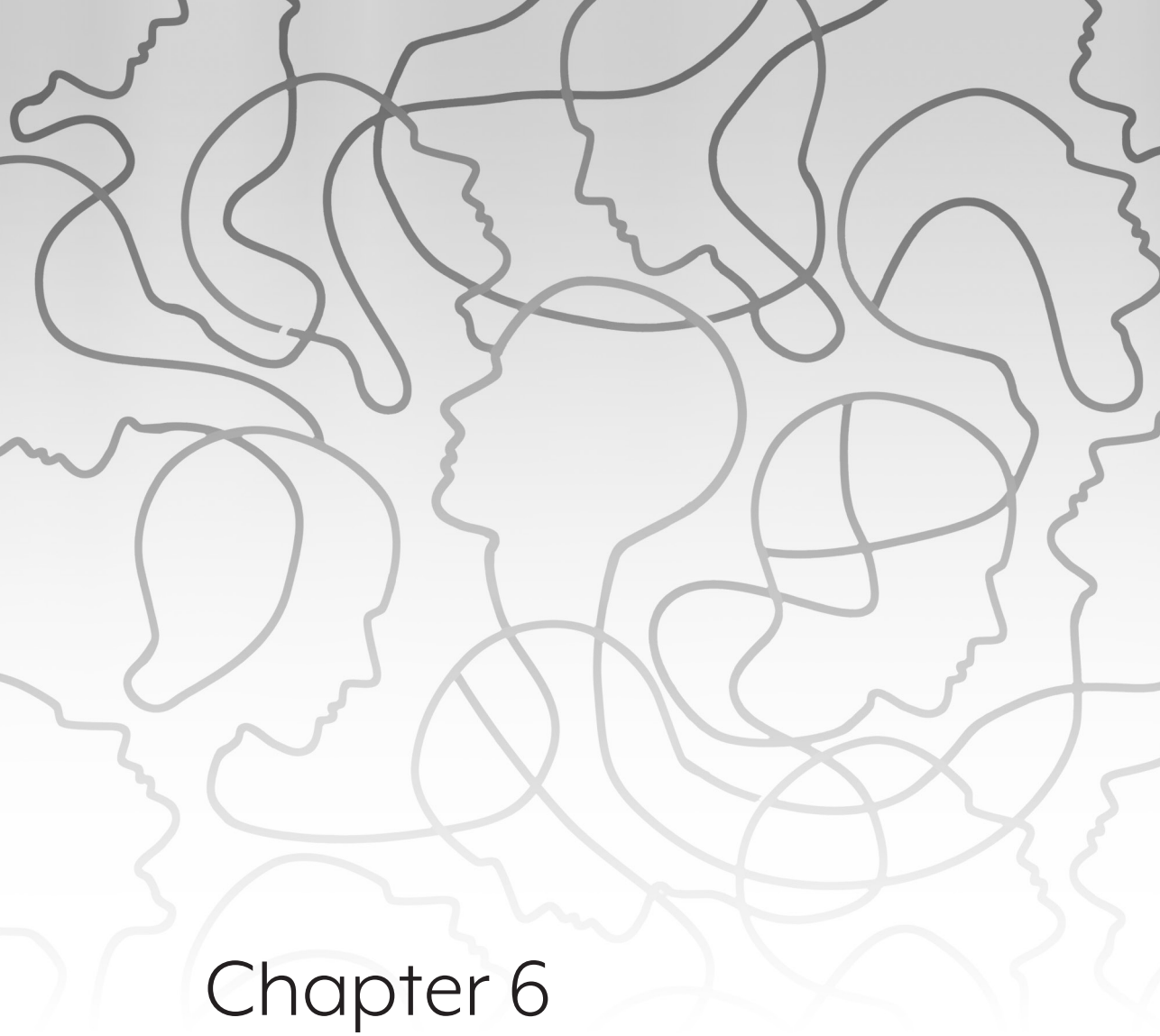


Part II

Mental health problems and autism spectrum disorder in multiple disabilities



Chapter 6

Mental health profiles of people with sensory and intellectual disabilities

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Abstract

Individuals with combined intellectual disabilities and sensory impairments can be more susceptible to developing mental health problems, especially when there is an additional Autism Spectrum Disorder (ASD). Furthermore, symptoms of their disabilities may overlap with symptoms of specific mental health problems, making it difficult to assess the aetiology of behaviours. This study assessed the presence of mental health problems in persons with intellectual and sensory disabilities, specifically attachment style, mood disorders and stress, and the effect of an additional ASD on mental health.

Participants were 60 individuals with an intellectual disability combined with sensory impairments. The presence of ASD was assessed with Observation of Autism in people with Sensory and Intellectual Disabilities (OASID). The list of Disturbed Attachment Behaviours, the Anxiety, Depression and Mood Sale (ADAMS) and Stress Survey Schedule (SSS) were used to assess presence and severity of other mental health problems.

Results showed that almost every participant showed signs of a disturbed attachment. Stress and mood disorders were not very prevalent. An additional ASD resulted in higher scores on disturbed attachment, manic and hyperactive behaviour and social avoidance.

The study found no concerning prevalence of the mental health problems anxiety, mood or stress, but only for attachment style. In some cases the presence of an ASD can lead to a slightly different profile of mental health problems in individuals with an intellectual disability combined with a visual impairment or deafblindness. When these symptoms occur it is recommend to always check the presence of ASD.

1. Introduction

Mental health problems are common in individuals with a combination of sensory and intellectual disabilities. Examples are stress (Bloeming-Wolbrink et al., 2012), an insecure attachment (Janssen, Schuengel, & Stolk, 2002) and depression or mood disorders (Hurley, 2006). They also often show behaviours that could be indicative of these mental health problems, such as stereotyped behaviours or aggressive behaviours (Poppes, van der Putten, & Vlaskamp, 2010). In addition, both people with sensory and intellectual disabilities are at a high risk of developing an Autism Spectrum Disorder (ASD) (Jesper Dammeyer, 2011; De Bildt, Sytema, Kraijer, & Minderaa, 2005). ASD in itself is also associated with more mental health problems (e.g. Corbett, Mendoza, Abdullah, Wegelin, & Levine, 2006; Stewart, Barnard, Pearson, Hasan, & O'Brien, 2006). Therefore, ASD in addition to sensory and intellectual disabilities seems lead to a high prevalence of mental health problems. This makes the diagnosis of mental health problems very complex, since symptoms such as stereotyped behaviour are not only indications of mental health problems, they are also core characteristics of ASD (American Psychiatric Association, 2013) and are also often shown by individuals with multiple disabilities without ASD (De Vaan, Vervloed, et al., 2016b; Hoevenaars-van den Boom, Antonissen, Knoors, & Vervloed, 2009). The current study focuses on individuals who have a moderate to profound intellectual disability combined with a visual impairment or deafblindness, with and without ASD. The goal is to describe the range of mental health problems in this group, including the effect of an additional ASD on the presence of mental health problems.

One of the mental health problems that persons with sensory and intellectual disabilities can possibly have is an insecure attachment style (Fraiberg, 1977; Janssen, et al., 2002; Stor & Storsbergen, 2006). This is similar to children with ASD, who are generally less securely attached to their caregivers, especially when the ASD is combined with an intellectual disability (Rutgers, Bakermans-Kranenburg, van IJzendoorn, & van Berckelaer-Onnes, 2004; Rutgers et al., 2007; Van IJzendoorn et al., 2007). An insecure attachment relationship has severe consequences, such as disturbances in emotional well-being, the occurrence of externalising behaviour problems and stress (Fearon, Bakermans-Kranenburg, Van IJzendoorn, Lapsley, & Roisman, 2010; Stor & Storsbergen, 2006).

Individuals with sensory impairments and intellectual disabilities are also thought to be more susceptible to stress. Because of their impairments, sensory information is often missed, making their everyday lives more unpredictable and thus more stressful (Bloeming-Wolbrink, et al., 2012; Corbett, et al., 2006; Dickerson & Kemeny, 2004). Furthermore, for people with disabilities it may be more difficult to cope with these stressors, especially when they cannot seek comfort with an attachment figure

(Janssen, et al., 2002; Schuengel & Janssen, 2006). When there is an additional diagnosis of ASD, individuals are even more susceptible to stress, for example in new and unfamiliar situations (Corbett, et al., 2006) or in social situations (Dickerson & Kemeny, 2004).

Finally, both persons with disabilities and persons with ASD may be more susceptible to developing mood disorders (Hurley, 2006; Stewart, et al., 2006). In turn, mood disorders, anxiety or stress can lead to social withdrawal and an increase in stereotyped behaviours (Kraijer, 2004; Rubin, Coplan, & Bowker, 2013; Stewart, et al., 2006), which are all also symptoms of ASD (American Psychiatric Association, 2013). Mental health problems such as these could thus increase the severity of ASD symptoms. This is supported by Ghaziuddin, Ghaziuddin & Greden (2002) who stated that curing mood disorders could also reduce the ASD symptoms. Insight in prevalent mental health problems in this population is therefore be helpful for treatment.

The symptoms of ASD and mental health problems overlap in persons with combined sensory and intellectual disabilties. This makes it rather difficult to assess the aetiology of these behaviours. A consequence is that this may lead to an over-diagnosis as well as an underdiagnosis of mental health problems in this target group. Whether the one or the other is the case often depends on which condition is thought to be the most notable according to the treating psychologist or caregivers. These phenomena are known as diagnostic overshadowing and diagnostic under-representation (Mason & Scior, 2004). ASD in addition to the sensory and intellectual disabilities complicates both diagnosis and treatment. To prevent inaccurate diagnoses and treatment it is important to create a complete profile of mental health problems. For the population of people with combined sensory and intellectual disabilities this has not been done yet. Because both the combinations of sensory and intellectual disabilities as well as ASD and intellectual disabilities increase the risk for mental health problems, it is likely that the combination of intellectual disability, sensory impairments and ASD puts people at an even higher risk of developing mental health problems.

Firstly, this study will assess the presence of some of the most common mental health problems that have been described for this population: an insecure attachment, stress and mood disorders. Secondly, this study will assess if an additional ASD in this population will lead to a different profile of mental health problems in people with combined sensory and intellectual disabilities.

2. Method

2.1 Participants

Participants were recruited in four locations of three residential institutions and in three schools for people with sensory and intellectual disabilities within the Netherlands. We believe a representative sample was reached by recruiting in locations of all of the institutions and schools specialized in our target population within the Netherlands. Inclusion criteria were (1) a moderate to profound intellectual disability, (2) a visual impairment, (3) between 6-60 years of age. An additional auditory impairment was allowed. Participants were selected by a contact person from each facility to maintain anonymity until consent for participation was given. Because of this procedure there was no information about response rate.

Participants were 60 people aged between 6 and 55 years old (M = 31.6, SD = 14.9). The sample consisted of 42 males and 18 females. Participants were diagnosed with moderate (n=11), severe (n=24) or profound (n= 25) intellectual disabilities. All participants had a visual impairment (n =30) or were blind with or without light perception (n =30). There were 16 participants with additional auditory impairments. According to the definitions of Dammeyer (2012) and Ask Larsen and Damen (2014) they were deafblind. ASD was diagnosed as part of an earlier study using Observation of Autism in people with Sensory and Intellectual Disabilities (OASID) (De Vaan, Vervloed, et al., 2016a, 2016b). This instrument was designed specifically for this target population. Results showed that 32 participants had ASD and the remaining 28 participants did not (De Vaan, Vervloed, et al., 2016b). Sensory impairments and intellectual disabilities were diagnosed prior to and independent of this study by licenced psychologists or physicians. For this study, this information was retrieved directly from the participants' records of their residential facility or school.

2.2 Materials

2.2.1 List of Disturbed Attachment Behaviours

The list of disturbed attachment behaviours (Boris & Zeanah, 2005) is a screening instrument that gives an indication of how securely attached persons are to their caregivers. The questionnaire consists of 8 descriptions of behaviour on a 5-point Likert scale. A total score is calculated by adding scores on the eight individual questions. Higher scores are indicative of more disturbed attachment behaviours (Stor & Storsbergen, 2006).

2.2.2 Anxiety, Depression and Mood Scale

The Dutch translation (Hermans, Jelluma, & Evenhuis, 2008)of the Anxiety Depression and Mood Scale (ADAMS; Esbensen, Rojahn, Aman, & Ruedrich, 2003a) was used. The ADAMS consists of 28 multiple choice questions, in which the prevalence or

severity of behaviours are rated on a scale from 0 (not a problem) to 3 (severe problem). The 28 items are divided over five scales: 'manic/hyperactive behaviour', 'depressed mood', 'social avoidance', 'general anxiety' and 'compulsive behaviour'. The ADAMS is a psychometrically valid and reliable screening tool for anxiety, depression and mood disorders in individuals with an intellectual disability (Esbensen, Rojahn, Aman, & Ruedrich, 2003b).

2.2.3 Stress Survey Schedule

The Stress Survey Schedule for Autism and Other Developmental Disabilities (SSS; Groden, 2001) is a 62-items questionnaire that can be used to measure stress and identify specific stressors for individuals with ASD and other developmental disabilities (Groden et al., 2001). All items are rated on 5-point scales of stress intensity, ranging from 'none to mild' to 'severe'. A score can be calculated for 10 potential problem areas: 'changes', 'anticipation', 'unpleasant', 'positive', 'sensory/personal', 'food related', 'social/environmental', 'rituals', 'fears' and 'life stressors'. The SSS was found to be valid and reliable for its purpose (Groden, et al., 2001) .

2.3 Procedure

This study was approved by the Committee on Research Involving Human Subjects, Arnhem-Nijmegen, and was in line with the Ethical Principles for Medical Research Involving Human Subjects of the World Medical Association declaration of Helsinki (World Medical Association, 2013). Participants were recruited through their residential facility or school. Because participants were legally incapable, parents or legal representatives were asked for informed consent. After consent was given, a familiar caregiver was asked to fill in the questionnaires.

In order to assess whether the participants had scores within the clinical ranges, we compared scores of our participants to norms or cut-off scores of these questionnaires when available. When cut-off scores or norms were not published, scores of participants were compared to mean scores of similar populations.

3. Results

3.1 Mental health problems

3.1.2 Attachment

For the list of disturbed attachment behaviours, scores ranging from 0-8 indicate no disturbed attachment, scores ranging from 8-24 indicate possible disturbed attachment, and scores of 24 and above indicate a probable disturbed attachment (Stor & Storsbergen, 2006). Table 1 shows how many participants fell into each category.

Table 1 Distribution of participants on attachment categories

	Number of participants	
Score on list of disturbed attachment	*n*	% of total
0-8 no disturbed attachment	2	3.3%
9-24 possible disturbed attachment	38	63.3%
24+ probable disturbed attachment	20	33.3%

3.1.2 Anxiety, Depression and Mood

For the ADAMS, cut-off scores have only been reported for the Depressed Mood and General Anxiety subscale. These cut-off scores are meant for screening purposes (Hermans, Jelluma, van der Pas, & Evenhuis, 2012). For people with moderate to profound intellectual disabilities, the Depressed Mood cut-off is a score of 11. Two participants scored above the cut-off (3.3%). In addition, Esbensen, Rojahn, Aman and Ruedrich (2003b) reported percentile ranks for a large group of participants. Table 2 shows how many participants from the current study scored above the 90th and 95th percentile rank.

Table 2 Number of participants that score within clinical range

		Number of participants	
ADAMS scale	Percentile rank	*n*	% of total
Manic/ Hyperactive Behaviour	90th – 95th	1	1.6%
	Above 95th	2	3.3%
Depressed Mood	90th – 95th	0	0%
	Above 95th	2	3.3%
Social Avoidance	90th – 95th	5	8.3%
	Above 95th	4	6.7%
General Anxiety	90th – 95th	1	1.6%
	Above 95th	1	1.6%
Compulsive Behaviour	90th – 95th	1	1.6%
	Above 95th	0	0%

3.1.3 Stress Survey Schedule

For the Stress Survey Schedule, mean scores were reported as a function of sex, verbal ability or age (Goodwin, Groden, Velicer, & Diller, 2007). Since the larger part of our group consisted of non-verbal participants or participants with very limited

verbal skills, we compared our participants to the non-verbal group that was reported. Table 3 shows the comparison of our groups to the non-verbal group described in Goodwin, et. al (2007). We reported how many participants scored two SD's or more above the mean.

3.2 Differences between ASD and no ASD

Differences between participants with and without ASD were compared using non-parametric Mann-Whitney U tests. Corresponding effect sizes were calculated by the following formula $r = z / \sqrt{N}$ (Pallant, 2010). All medians and significant differences between the two groups on the List of Disturbed Attachment, ADAMS and SSS are presented in Table 4. Participants with ASD scored significantly higher than participants without ASD on the list of disturbed attachment and on the ADAMS scales Manic and Hyperactive Behaviour and Social Avoidance. Participants without ASD score higher on the SSS scale 'Unpleasant' than people with ASD. All effect sizes were medium.

Table 3 Number of participants that scored two SD's or more above the mean compared to non-verbal individuals[1]

	Number of participants	
SSS scale	*n*	% of group total
Changes	2	3.3%
Anticipation	3	5%
Unpleasant	3	5%
Sensory / Personal	25	41.7%
Food related	0	0%
Positive	0	0%
Social / Environmental	5	8.3%
Rituals	0	0%

Note: [1]Retrieved from Goodwin, et. al (2007).

Table 4 Differences between ASD and no ASD on the attachment list, ADAMS and SSS

	No ASD (n=28)	ASD (n=32)			
Instrument / Scale	*Mdn*	*Mdn*	*U*	*p*	Effect Size *r*
Attachment list	18.0	24.0	260.000	.005**	.36
ADAMS					
Manic / Hyperactive behaviour	2.0	4.5	295.000	.022*	.29
Depressed Mood	2.0	2.5	386.000	.353	
Social Avoidance	2.0	5.0	288.000	.017*	.31
General Anxiety	5.0	5.0	434.000	.835	
Compulsive behaviour	2.0	1.5	418.000	.651	
SSS					
Changes	25.0	26.0	438.000	.882	
Anticipation	13.0	13.0	446.000	.976	
Unpleasant	19.0	14.5	293.500	.022*	.30
Sensory / Personal	9.0	9.0	420.500	.682	
Food related	5.0	5.0	420.000	.674	
Positive	11.0	10.5	445.000	.964	
Social / Environmental	4.0	3.0	360.500	.170	
Rituals	10.5	7.5	344.000	.121	
Fears	9.0	9.5	439.500	.899	
Life stressors	15.0	12.5	362.000	.201	

Note * $p < .05$, ** $p < .01$.

4. Discussion

The current study investigated the presence of mental health problems individuals with a combination of sensory and intellectual disabilities. Firstly, we studied whether attachment style, anxiety, depression and mood disorders, and stress were present in our target population by comparing them to norm scores or cut off scores. Secondly, we assessed the effect of an additional ASD on these mental health problems by looking at the differences in scores between participants with and without ASD.

On the list of disturbed attachment behaviour, we found that only a very small proportion of our sample did not show signs of a disturbed attachment. The larger part of our participants scored within the range of a 'possible disturbed attachment'. A quarter of our sample had a score that indicated a 'probable disturbed attachment'.

Though the literature suggests that disabilities as well as ASD can both contribute to the development of a less secure attachment style (e.g. Janssen, et al., 2002; Rutgers, et al., 2004; Stor & Storsbergen, 2006), these numbers are quite high. Sterkenburg (2008) has shown that attachment based treatment in individuals with sensory and intellectual disabilities can help to regulate stress and reduce challenging behaviour. It is therefore recommended to always screen for attachment related problems in individuals from this population and that treatment is adjusted in the case that a disturbed attachment is present. However, there is a possibility that the prevalence of attachment problems that we found is slightly overestimated. The list of disturbed attachment behaviours is not specifically developed for this population and some signals may have been unfairly attributed to attachment problems when they're in fact part of their intellectual or sensory disability. Nevertheless, it is important remain vigilant for signs of an attachment style within this population.

Our results on the ADAMS revealed that only a few participants scored above the 90th percentile on any of the scales. Based on the ADAMS there is no reason to believe that our participants show clinically concerning manic or hyperactive behaviour, general anxiety, depressed mood or compulsive behaviour. On social avoidance, high scores were found. Fifteen percent of our participants scored above the 90th percentile. This may be due to the high prevalence of ASD in this group, as social avoidance is one of the key characteristics of ASD (American Psychiatric Association, 2013).

With the SSS we identified stressful situations for our participants. The norm group we used for comparison were non-verbal participants from an earlier study by Groden et al. (2001). Since the larger part of our participants were non-verbal, this was the most appropriate group to compare our participants to. Norms for our exact population were not available. On most of the scales of the SSS less than ten percent of our group scored more than two SD's above the mean of the non-verbal control group. However, on the sensory / personal scale, over 40% of our participants scored more than two SD's above the mean of non-verbal controls, meaning that sensory and personal stressors affected these individuals strongly. Because they are sensory impaired, this may not seem surprising. Missing visual or auditory cues from the environment can make situations unpredictable and therefore more stressful for these participants (Bloeming-Wolbrink, et al., 2012). Items on this scale include 'being touched' and 'being hugged'. For individuals with visual impairments, who rely on touch and tactile information, being touched could indeed be stressful, especially when unexpected.

In the second part of our study we assessed the differences between participants with and without ASD. It was found that participants with ASD scored higher on the disturbed attachment list and the ADAMS scales 'manic / hyperactive behaviour' and 'social avoidance'. It is in line with earlier research that persons with ASD show less

secure attachment styles than individuals without ASD (Rutgers, et al., 2004; Rutgers, et al., 2007; Van IJzendoorn, et al., 2007). Also, participants with ASD show more social avoidance. In itself, this finding makes sense, as social avoidance is a symptom of ASD (American Psychiatric Association, 2013; Richer, 1976). However, these two findings could also be connected as additional impairments in social skills could be related to an insecure attachment style (Rutgers, et al., 2007). Finally, participants with ASD scored higher on manic and hyperactive behaviours, this is also in line with expectations because hyperactivity is frequently observed in individuals with ASD (Aman & Langworthy, 2000; American Psychiatric Association, 2013).

Participants without ASD scored higher on the SSS scale 'unpleasant' than participants with ASD. This indicates that persons without ASD show a higher stress reaction to unpleasant events than individuals with ASD. Items on this scale include questions related to receiving criticism from others (Groden, 2001). Perhaps their impairments in the social domain and their level of cognitive impairment make it difficult for persons with ASD to correctly interpret criticism and thus they experience it as less stressful. On the other scales no significant differences were found between participants with and without ASD.

The mental health profiles of participants with ASD were slightly different from the profiles of participants without ASD. Some of this could be due to wrongly attributing ASD symptoms to attachment, stress, anxiety and mood disorders. It is therefore recommended to always also assess the presence of ASD when overlapping symptoms occur, especially when there are problems related to an insecure attachment style, manic and hyperactive behaviours, or social avoidance. When ASD is present, these symptoms could very well be caused by ASD and treatment should be focused on ASD. However, when ASD is absent, these same symptoms may be caused by another mental health problem such as a mood disorder and treatment should be focused on reducing these problems first.

This study has some limitations. First, there are only a few instruments that are specifically developed for the population of people with combined sensory and intellectual disabilities. A recent study has shown that on instruments that were not specifically developed for this populations items are not always valid (De Vaan, Vervloed, Hoevenaars-van den Boom, et al., 2016). In addition, because of the specific target population, norms or cut-off scores for this population are often not available and it is difficult to decide which group is most appropriate for comparison. In this study, the list of disturbed attachment behaviours did not have separate norms for specific populations and the ADAMS only for individuals with intellectual disabilities without sensory impairments. For the SSS our best comparison group were non-verbal individuals. More research is needed to develop new instruments or at least norms for this specific target population so that mental health problems can be assessed validly.

Secondly, the diagnosis of ASD distinguishes multiple severity levels (American Psychiatric Association, 2013). The diagnosis of ASD for this study was done with the OASID that distinguishes three severity levels of ASD (De Vaan, Vervloed, et al., 2016a). However, because of small numbers of participants in each ASD severity group, all participants with ASD were combined to form one group. For future studies, it would be interesting to see the severity of ASD is correlated to having more mental health problems.

Persons with multiple disabilities show behaviours that could be interpreted as symptoms of mental health problems, including ASD, depression, anxiety, stress or insecure attachment style. Depending on the diagnostic instruments that are used, one or more of these problems could be diagnosed. Traditionally, this is seen as comorbidity, with each symptom having its own underlying pathological process, similar to somatic diseases. An alternative view is presented in more recent network analyses. Network analyses have shown that symptoms interact and influence each other, creating a new network of symptoms that are not necessarily linked to one specific diagnosis but may be activated by other symptoms (Borsboom & Cramer, 2013; Boschloo et al., 2015; Bringmann et al., 2013). It is argued that in mental health, symptoms can be the result of many different processes, causes and even symptoms of other disorders. Disorders may often be diagnosed together because of the overlap in symptoms (Fried et al., 2017). This could explain the large prevalence of ASD and attachment disorder in the current population. For this population it is especially important that treatment is not focused on addressing the symptoms of specific disorders, but on improving a person's overall mental health and wellbeing (Do-en, 2007).

The current study explored the presence of mental health problems in individuals with combined sensory and intellectual disabilities. It showed that most participants with sensory and intellectual disabilities showed signs of a disturbed attachment and these problems are largest in the ASD group. This finding is important for clinical practice and we recommend that in this population everyone is screened for attachment related problems and treatment is adjusted to these findings. In addition, we found that participants with ASD show a less secure attachment style, more manic and hyperactive behaviour and more social avoidance than individuals without ASD. Persons with ASD showed a slightly different profile of mental health problems than persons without ASD. When these mental health problems occur in individuals with sensory and intellectual disabilities, it is always recommended to assess the presence of an ASD and adjust treatment to the presence or absence of ASD.

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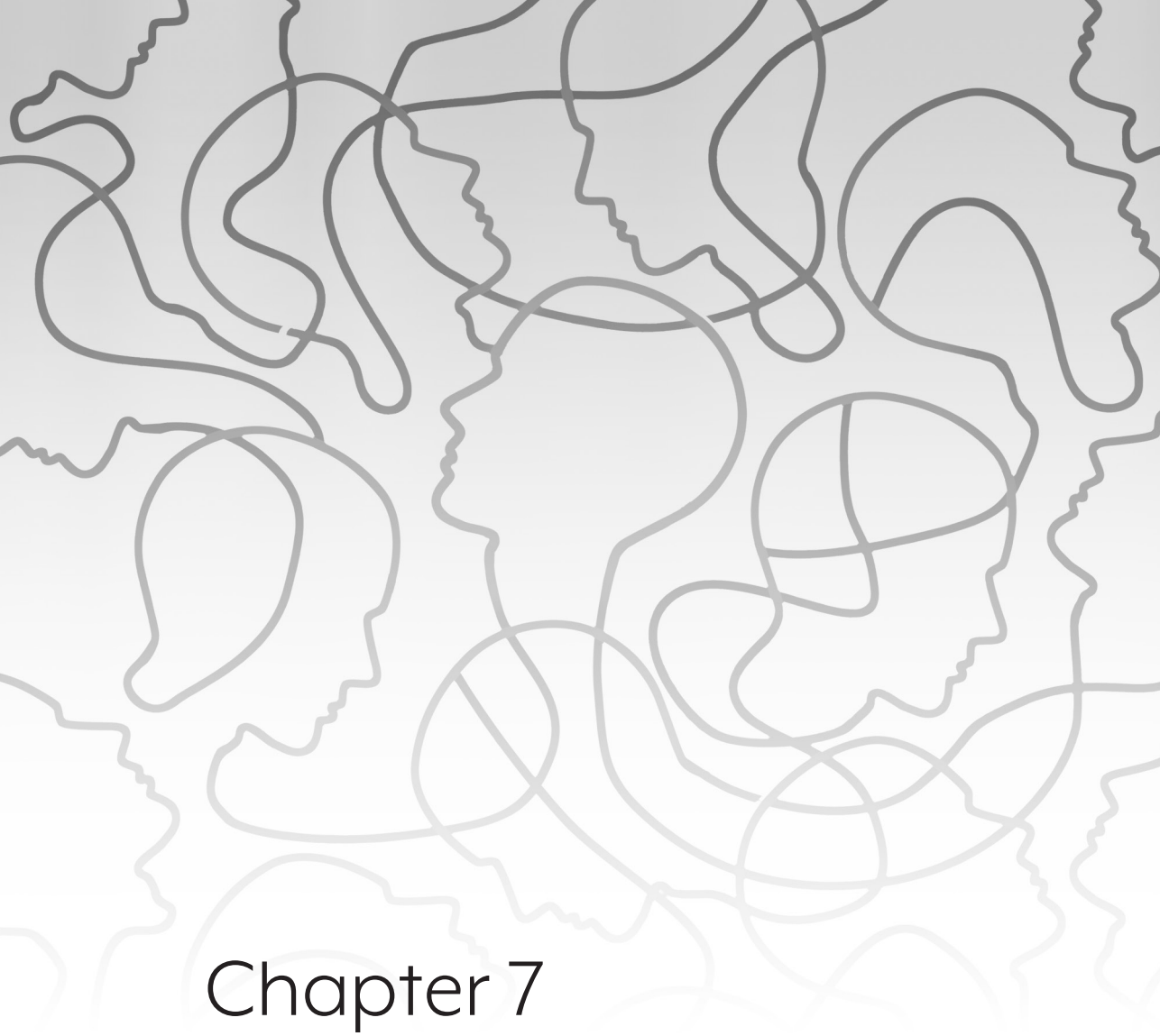

Chapter 7

Profiles of stereotyped behaviour in people with combined sensory impairments and intellectual disabilities

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Abstract

Stereotyped and repetitive behaviours are characteristics of Autism Spectrum Disorder (ASD) but also occur in individuals with combined intellectual and sensory disabilities. This paper looked at the differences in type, frequency and duration of stereotyped behaviours between individuals with and without ASD in this population.

The study included 59 individuals with intellectual disabilities and sensory impairments. The presence of ASD was assessed using Observation of Autism in people with Sensory and Intellectual Disabilities (OASID). Separate from these assessments video recordings were scored by observers naive to the ASD status of participants for stereotyped and repetitive behaviours.

Stereotyped and repetitive behaviours were more prevalent in participants with ASD, though a large proportion of participants without ASD showed them too. Participants with ASD showed, on average, more frequent and in duration longer stereotyped and repetitive behaviours, especially self-injurious behaviours. No differences were found for vocal, motoric and stereotyped behaviours with objects. The mean duration of each episode of stereotyped behaviour did not differ between groups. Cluster analysis revealed a distinct group of individuals without ASD who showed a high number of stereotyped behaviours.

1. Introduction

Autism Spectrum Disorder (ASD) is characterized by two major components, (1) deficits in social communication and social interaction, and (2) restricted and repetitive behaviour patterns, interests or activities (American Psychiatric Association, 2013). In people with a combination of sensory and intellectual disabilities the same deficits and atypical behaviours are also present (de Vaan, Vervloed, Knoors, & Verhoeven, 2013; Hobson, 2005; Hoevenaars-van den Boom, Antonissen, Knoors, & Vervloed, 2009). Both people with ASD and people who have a combination of sensory and intellectual disabilities show autistic features. The biggest overlap may be found in the domain of repetitive and stereotyped behaviours. This is not surprising, because stereotyped behaviours occur very frequently in individuals with both sensory and intellectual disabilities (Murdoch, 1997). The question presents itself whether the presence of repetitive and stereotyped behaviours is a differentiating characteristic, of ASD in people with a combination of intellectual and sensory disabilities. The current study will focus on this question by looking at the occurrence and severity of stereotyped and repetitive behaviours in people with a combination of sensory and intellectual disabilities with and without ASD.

The DSM-5 describes four different expressions of stereotyped behaviour that are part of ASD, namely: (1) stereotyped and repetitive movements, objects or speech, (2) insistence on sameness and adherence to routines, (3) restricted and fixated interests and (4) different reactivity to sensory input (American Psychiatric Association, 2013). Though these behaviours are described as being typical for ASD, they are also often seen in people with sensory or intellectual disabilities. As such, a relationship has been found between stereotyped behaviours and developmental level (Militerni, Bravaccio, Falco, Fico, & Palermo, 2002). Research also showed that individuals with ASD and an intellectual disability perform more stereotyped behaviours than individuals with an intellectual disability without ASD (Bodfish, Symons, Parker, & Lewis, 2000; Goldman et al., 2009). Research by Richards, Oliver, Nelson, and Moss (2012) further suggested that for people with intellectual disabilities the prevalence of self-injurious behaviours is high when someone has ASD as well and the amount of stereotyped behaviours is correlated with ASD severity (Militerni, et al., 2002).

Though there seems to be a relationship between ASD and stereotyped and repetitive behaviours, they underlying cause of the behaviours may be different, depending on a person's impairments. Andrews and Wyver (2005) suggest that the stereotyped behaviours found in people with sensory impairments may be have completely different causes and serve different functions than the ones found in people with ASD, even though the behaviours may express themselves in an identical way. Turner (1999) suggests that some specific behaviours may be due to ASD while

others are part of another. Certain behaviours may indeed be specifically correlated to a person's visual impairment, such as eye poking or eye pressing which occurs frequently in blind children (Tröster, Brambring, & Beelmann, 1991). Another option is that intellectual disabilities, sensory impairments and stereotyped behaviours are due to the same cause. Mukaddes and colleagues (2007) found that persons with blindness and ASD often have greater neurological impairments, such as lower intellectual levels or cerebral palsy. They suggest the possibility that the neurological damage that is responsible for the visual impairment or intellectual disability is also the cause of ASD typical behaviours such as repetitive and stereotyped behaviours.

Regardless of the cause of stereotyped and repetitive behaviours, earlier research findings indicated that stereotyped and repetitive behaviours are indeed frequently seen in people with sensory and/or intellectual disabilities, but also in people with ASD. The prevalence, frequency and severity of stereotyped and repetitive behaviours are known to be a differentiating factor between people with and without ASD (Lord, 1995; MacDonald et al., 2007; Szatmari, Bartolucci, & Bremner, 1989; Turner, 1999; Watt, Wetherby, Barber, & Morgan, 2008). At the same time, people with both sensory and intellectual disabilities show a high prevalence of stereotyped and repetitive behaviours, caused by their sensory disabilities and not necessarily by ASD (Gal, Dyck, & Passmore, 2010; Poppes, van der Putten, & Vlaskamp, 2010; Tröster, et al., 1991). This raises the question whether stereotyped and repetitive behaviours are characteristic factors for ASD and can serve as differentiating behaviours or clinical markers for the assessment of ASD in people with a combination of intellectual and sensory disabilities. In fact earlier studies have indicated that in people with both sensory and intellectual disabilities the persons with ASD versus the ones without ASD show more impairments in the social domain and less in the domain of stereotyped and repetitive behaviours (de Vaan, Vervloed, Peters-Scheffer, et al., 2016; Hoevenaars-van den Boom, et al., 2009). This could mean that stereotyped and repetitive behaviours are of no or limited use in diagnosing ASD in people with both sensory and intellectual disabilities.

The present study takes a closer look at stereotyped and repetitive behaviours in people with a combination of sensory and intellectual disabilities with and without ASD. What types of stereotyped behaviours are seen and how do they differ between people with and without ASD? First, a possible underlying structure of stereotyped and repetitive behaviours is examined. Secondly, differences between people with and without ASD within this target population are assessed. The focus will be on the type, duration and frequency of stereotyped behaviours. Finally, we will cluster participants with comparable stereotyped and repetitive behaviours, to see if behavioural profiles can be made regardless of the combination of disabilities.

2. Method

2.1 Participants

Participants were recruited from four residential institutions and three schools for people with multiple disabilities. Information about disabilities and impairments was obtained from personal files kept at their school or institution, and were determined by licensed psychologists, psychiatrists, physicians, ophthalmologists and audiologists in the past independently of this study. A contact person from each institution recruited potential participants and asked legal representatives for written consent. For privacy reasons the authors did not receive information about clients that did not what to participate, so the non-response rate is unknown.

Participants were 59 individuals (41 male, 18 female) with a mean age of 31.32 years (SD = 14.92, range = 6 – 55 years). All participants had a moderate to profound intellectual disability and a visual impairment, ranging from moderate visual impairment to blindness without light perception. A total of 16 individuals had an additional auditory impairment and were thus deafblind (following the definitions of Ask Larsen & Damen, 2014; Dammeyer, 2012). The presence of ASD was assessed using Observation of Autism in people with Sensory and Intellectual Disabilities (OASID; see de Vaan, Vervloed, Peters-Scheffer, et al., 2016). OASID scores normally lead to an interpretation of No ASD, Mild ASD, Severe ASD or Profound ASD symptoms (De Vaan et al., 2018), but for the purpose of this study two groups were created: No ASD (n=27) and ASD (n=32). The latter group consisted of people with Mild, Severe and Profound ASD symptoms. See Table 1 for an overview of participant characteristics in both groups. An independent samples t-test showed that age did not differ between groups. Chi-square tests showed that the distribution of levels of visual and auditory impairment were equal between groups. Only the distribution of level of intellectual disability was not the same between groups, χ^2 (2, n = 59) = 20.48, p < .001. The level of intellectual disability was more severe in people with ASD. This is not uncommon, because level of intellectual disability and autism were frequently found to be related (e.g.Matson & Shoemaker, 2009; O'Brien & Pearson, 2004).

2.2 Materials

2.2.1 OASID

Observation of Autism in people with Sensory and Intellectual Disabilities **(**OASID) is a semi-structured observational assessment tool, designed to diagnose ASD in people with intellectual disabilities combined with sensory impairments (de Vaan, Vervloed, Peters-Scheffer, et al., 2016). The first author and two experimenters, trained in the assessment of OASID by the first author, played five tasks with the participants in order to trigger ASD typical behaviour, using toys and interaction

Table 1 Participant Characteristics

Characteristics		No ASD (n=27)	ASD (n=32)
Age *M (SD)*		33.0 (14.35) range = 8-55	29.8 (15.49) range = 6-53
Sex *n*	Male	20	10
	Female	7	22
Intellectual disability *n*	Moderate	8	2
	Severe	16	9
	Profound	3	22
Visual impairment *n*	Impaired	15	15
	Blind (with light perception)	1	4
	Blind (no light perception	11	13
Auditory impairment *n*	No	16	27
	Impaired	9	3
	Deaf	2	2

games adjusted to the participants' abilities. This play session lasted between 20 and 55 minutes, and was recorded on video. The video of each session was observed afterwards and scored for autistic behaviours using 40 items scored on a three point Likert scale. A total score was calculated for two scales, namely: 'Social Interaction and Communication' and 'Stereotyped and Repetitive Behaviours'. Both scales conform to the DSM-5 criteria for ASD. The combination of the two scales leads to a final score indicating ASD severity: no ASD, mild ASD, severe ASD or profound ASD. OASID was found to be both a valid and reliable measure of ASD in people with sensory and intellectual disabilities (de Vaan, Vervloed, Peters-Scheffer, et al., 2016; De Vaan, et al., 2018).

2.2.2 Observation

In order to assess the presence, frequency and duration of stereotyped behaviour, four behavioural categories were created based on the literature: Motor, self-injurious, and vocal stereotyped behaviour and repetitive behaviours with objects (e.g. Moore & Magyar, 2012; Singer, 2009; Weiss, 2002). Each category was comprised of one or more stereotyped or repetitive behaviours (see Table 2). Behaviours within each category were mutually exclusive, but behaviours from different categories could overlap.

For this study, we observed a ten-minute session from the OASID assessment. As described above and more elaborately in De Vaan, et al., (2016), this assessment consisted of a play session that was similar for each participant. Examples of tasks

Table 2 Observed behaviours and definitions

	Description
Motor Stereotyped Behaviours	
Cradle	Move back and forth with upper body, sitting down or standing up
Spinning	Spin around with entire body
Pacing	Step feet back and forth, while sitting down or standing up / walking
Head-rocking	Rock head from shoulder to shoulder or up and down
Hands clapping	Slap hands together
Hands flapping	Move hands up and down or back and forth in the air
Finger flicking	Push fingers together, may create sound
Mouthing / Sucking	Use mouth as if sucking
Hair pulling	Pull or fidget own hair
Teeth grinding	Moving teeth over each other
Self-injurious Stereotyped Behaviours	
Violent head banging	Hit head against floor, walls, furniture or objects
Slapping	Hit their own body hard, may cause wounds
Eye pressing	Push on eye with entire hand
Eye poking	Press eye with one or multiple fingers
Self biting	Use teeth to bite in own body / skin
Self scratching	Use fingernails to scratch skin, may cause wounds or irritation
Self pinching	Pinch oneself so hard it may cause skin damage
Orifice poking	Use objects to poke orifices such as mouth, nose, ears
Nail picking	Break or rip nails
Vocal Stereotyped Behaviours	
Yelling / Screaming / Squealing	Raise voice
Repetitive Behaviours with Objects	
Tick	Tick an object against another object, the floor, furniture, other objects or own body
Wiggle	Wiggle an object within hands
Bounce	Bounce an object on the floor or furniture
Balance	Balance an object back and forth on the floor, furniture or other object
Classify / sort objects	Sort objects based on colour, shape, size or purpose
Preoccupations / repetitive movements	Move parts of an object repetitively instead of playing with entire object

that were played are physical interaction games with song and movements and solving a puzzle. Because the assessment was unfamiliar to the participants the first ten minutes served as a warming up for the participant to get acquainted with the situation and experimenter. Stereotyped behaviours during these first ten minutes were therefore disregarded. Observations were performed with The Observer XT software (Noldus., 2015). The Observer software allows for the design of a coding scheme and to code behaviour accordingly. While watching the video a rater coded on- and offset times of behaviours. The time in between start and stop defined an episode of stereotyped behaviour. This provided information about which behaviours occurred, how many episodes there were (frequencies) and how long each episode lasted (duration).The coding scheme is shown in Table 2. When a behaviour appeared to be starting but ended within less than three bouts , the raters were instructed not to take this behaviour into account as repetitive, attempts were not scored.

A ten-minute fragment of the OASID assessment was analysed, starting after ten minutes in the assessment and ending at the end of the twentieth minute. Videos were scored independently by two student assistants, who had previously received training by the first author in the use of the Observer XT and the coding scheme. Both raters completed all video fragments. The Observer XT interrater reliability analysis revealed an overall Kappa of 0.98, indicating excellent inter-rater reliability.

2.3 Procedure

This study was approved by the local Committee on Research Involving Human Subjects and conformed to the Ethical Principles for Medical Research Involving Human Subjects of the World Medical Association declaration of Helsinki (World Medical Association, 2013). Parents or legal representatives gave informed consent prior to participation. Firstly, the OASID assessment was performed, which lasted between 20 and 55 minutes. This session was recorded on video and scored afterwards for the whole session by means of the OASID questionnaire. After the assessment four behavioural categories were created to assess the presence of stereotyped and repetitive behaviour. These categories and corresponding behaviours were the result of a literature study on stereotyped and repetitive behaviours . Secondly, a ten minute fragment of each OASID video recording was selected. For each participant the 11^{th} until the 20^{th} minute were analysed with the Observer XT software. Since categories could overlap each video had to be scored in four separate runs, one run for each behavioural category, in order to guarantee observation accuracy. Both OASID and the ten minute fragments were scored for stereotyped behaviour using observations. However, the OASID assessment only included questions about the mere presence of stereotyped behaviour, whereas the observations with The Observer XT zoomed in on the types, frequency and duration of these behaviours.

2.4 Statistical Analyses

First, a principal component analysis using oblique rotation was performed to check if our a priori categories of stereotyped behaviour could be confirmed based on our data. Secondly, groups (ASD versus no ASD) were compared. To compare proportions we used Chi-square tests and to compare differences in duration, frequency and duration per episode we used non-parametric Mann-Whitney U tests, because assumptions regarding normality and equal variance were violated. Corresponding effect size *r* was calculated using the formula of Pallant (2010).

We made comparisons between groups on the types and total number of stereotyped behaviour, and compared duration, frequency and duration per episode. Duration was defined as the total duration in seconds that participants engaged in stereotyped behaviour. Frequency was the number of separate episodes of stereotyped behaviour that were observed. Duration per episode was the average duration of each episode of stereotyped behaviour, calculated by dividing duration by frequency. In case of planned multiple comparisons, a more conservative statistical significance level of .01 was used, to avoid capitalization on chance and control for familywise error rate. Finally clusters of participants were made using the durations of the different stereotyped behaviour categories. This was done using a hierarchical cluster analysis according to the procedure described by Yim & Ramdeen (2015).

3. Results

3.1 Principal component analysis on categories of stereotyped behaviour

Prior to this study we created four categories of stereotyped and repetitive behaviour based on the literature, see Table 2. An exploratory principal component analysis was performed. This was done to see if our data led to the same categories that were created a priori. The variables that we inserted were the total duration of each separate behaviour. For six of these 26 variables there was no variance, so these were excluded from this analysis. These variables were: Spinning, Mouthing/sucking, Teeth grinding, Violent head banging, Nail picking and Classify/sort objects. The principal component analyses was performed on the remaining 20 variables. We chose to use eigenvalues of 1.0 as a cut off for number of factors. Ten factors were extracted accordingly, explaining 78.6% of the total variance. This is a large number of factors considering the number of variables (an average of two variables per category) and does not comply with the categories that were found in the literature. The results showed that based on the data stereotyped and repetitive behaviours could not be reduced to clear latent structures. This is why we will perform our calculations on individual variables and the a priori categories.

3.2 Proportion of people with stereotyped behaviour

The first column for each group in Table 3 shows the number and percentage of people in this group that showed each type of stereotyped behaviour. Within the ASD group, 100% of the participants showed stereotyped behaviour as opposed to 85.2% of people without ASD, a significantly higher proportion, χ^2 (1, n = 59) = 5.07, p = .024.

For the different categories of stereotyped behaviours, differences in proportions of people were found for injurious stereotyped behaviour, χ^2 (1, n = 59) = 7.31, p = .007, where 84.4% of participants with ASD showed self-injurious behaviour, versus 51.9% of participants without ASD. For the other categories no significant differences in proportions were found.

3.3 Types of stereotyped behaviour

As can be seen in Table 3, some types of stereotyped and repetitive behaviour did not occur at all in our sample, including spinning, mouthing/sucking, teeth grinding, nail picking and sorting or classifying objects. Furthermore, pacing and bouncing and balancing of objects only occurred in the no ASD group, whereas finger flicking and violent head banging was only observed in people with ASD.

In addition, persons with ASD showed significantly more types of stereotyped behaviour than persons without ASD, respectively 4.00 (*SD* = 2.08, *Md* = 3.5) types of stereotyped behaviour versus 2.56 (*SD* = 2.23, *Md* = 2.0) types, *U* = 250.000, *p* = .006, *r* =0.36.

3.4 Differences in duration of stereotyped behaviour

Differences in duration of stereotyped behaviour (see Table 3) between people with and without ASD were tested with Mann-Whitney U tests. Differences were found for the total duration of stereotyped behaviour, *U* = 232.000, *p*= .002, *r* = .40, (ASD *Md* = 120.13, No ASD *Md* = 29.66) and for the duration of the category injurious stereotyped behaviours. *U* = 225.500, *p* = .001, *r* = .41 (ASD *Md* = 23.57, No ASD *Md* = 1.67). In general, people with ASD showed longer mean durations of stereotyped behaviour than people without ASD. Differences in duration were not found for the categories motor and vocal stereotyped behaviours or repetitive behaviours with objects, and any of the discrete stereotyped behaviours.

As can be seen in Table 3, a large proportion of participants did not show certain types of stereotyped behaviour at all. This affected the group mean, and possibly caused differences as the proportion of people without stereotyped behaviour is larger in the no ASD group. The analyses were repeated with participants with no stereotyped behaviour treated as missing, so only participants with stereotyped behaviours were taken into account. Mann-Whitney U tests revealed no differences on any of the categories of stereotyped behaviour between people with and without ASD, only the total score differed significantly between ASD (*M* = 196.68, *SD* = 185.52,

Md = 120.13) and no ASD (*M* = 107.77, *SD* = 142.0, *Md* = 37.07), *U* = 232.000, *p* = .02, *r* = .31. There is no longer a difference for self-injurious behaviours. On the discrete stereotyped behaviours, no significant differences were found.

3.5 Differences in frequency and duration per episode of stereotyped behaviour

Differences between groups in frequency (see Table 3) of stereotyped behaviour episodes were assessed with Mann-Whitney U tests. This revealed differences between the ASD and no ASD groups for the total frequency of stereotyped behaviour. The ASD group showed a higher frequency (*Md* = 21.5) than the no ASD group (*Md* = 6.0), *U* = 198.500, *p* < .001, *r* = .46. Also on self-injurious stereotyped behaviour people with ASD (*Md* = 6.00) showed a higher frequency of stereotyped behaviour than people without ASD (*Md* = 1.00), *U* = 203.000, *p* = .001, *r* = 0.45.

The total duration of stereotyped behaviour was divided by the frequency of stereotyped behaviour, finding the mean duration of each episode. Mann-Whitney U tests revealed no differences between people with and without ASD for episode length.

3.6 Clustering participants by stereotyped behaviour patterns

An hierarchical cluster analysis was performed to search for clusters of participants among the categories of stereotyped behaviour (Yim & Ramdeen, 2015). The duration of motor stereotyped behaviour, self-injurious stereotyped behaviour, vocal stereotyped behaviour and repetitive behaviour with objects were used as cluster variables. The agglomeration schedule and dendogram revealed three clusters of participants on these variables. See Table 4 for a comparison between clusters on these variables, as well as on total duration of stereotyped behaviour, age and OASID score.

As can be seen in Table 4, Cluster 1 consists of people with a relatively low total duration of stereotyped behaviour, reflected in all categories of stereotyped behaviour. Cluster 2 consists of people with a relatively high score on stereotyped behaviour, mostly in the category of motor stereotyped behaviours. Cluster three is a small cluster consisting of people with a high duration of stereotyped behaviour, especially seen in self-injurious stereotyped behaviour.

First we analysed differences on the variables used for clustering. Significant differences between clusters were found for motor stereotyped behaviour, where cluster 2 (*Md* = 333.67) scored higher than cluster 1 (Md = 6.37; *U* = 0, *p* < .001, *r* = .66) and cluster 3 (*Md* = 125.92; *U* = 0, *p* = .01, *r* = .70). On self-injurious stereotyped behaviour, cluster 3 (*Md* = 343.05) scored higher than cluster 1 (*Md* = 4.29; *U* = 0, *p* = .004, *r* = .41) and cluster 2 (*Md* = 2.19; *U* = 0, *p* = .01, *r* = .71). The other variables used for clustering did not differ significantly between groups.

Table 3 Prevalence, duration and frequency of stereotyped behaviours

	Number and percentage of individuals with stereotyped behaviour *n (%)*		Mean duration of stereotyped behaviours (in seconds) *M (SD)*		Frequency of stereotyped behaviours *M (SD)*	
	No ASD	ASD	No ASD	ASD	No ASD	ASD
Motor Stereotyped Behaviours	**19 (70.4%)**	**25 (78.1%)**	**71.97 (123.20)**	**108.75 (154.51)**	**6.52 (9.14)**	**14.00 (18.08)**
Cradle	8 (29.6%)	15 (46.9%)	26.84 (72.19)	41.95 (77.14)	2.67 (6.2)	4.72 (8.47)
Spinning	0	0	-	-	-	-
Pacing	2 (7.4%)	0	0.45 (20.09)	-	0.07 (0.27)	-
Head-rocking	8 (29.6%)	16 (50%)	37.42 (106.32)	49.35 (124.79)	2.44 (5.55)	5.28 (9.64)
Hands clapping	5 (18.5%)	7 (21.9%)	0.41 (1.15)	4.58 (17.0)	0.26 (0.66)	1.75 (4.79)
Hands flapping	5 (18.5%)	6 (18.7%)	1.53 (3.81)	3.0 (11.51)	0.70 (1.92)	1.25 (4.33)
Finger flicking	0	4 (12.5%)	-	9.58 (40.59)	-	0.94 (3.51)
Mouthing / Sucking	0	0	-	-	-	-
Hair pulling	2 (7.4%)	2 (6.3%)	5.31 (27.19)	0.29 (1.22)	0.37 (1.74)	0.06 (0.25)
Teeth grinding	0	0	-	-	-	-
Self-injurious Stereotyped Behaviours	**14 (51.9%)**	**27 (84.4%)**	**9.60 (20.03)**	**63.80 (100.50)**	**2.63 (5.30)**	**9.90 (11.84)**
Violent head banging	0	2 (6.3%)	-	1.58 (6.21)	-	0.19 (0.78)
Slapping	3 (11.1%)	11 (34.4%)	1.80 (6.47)	5.82 (17.64)	0.96 (3.17)	2.66 (8.09)
Eye pressing	4 (14.8%)	10 (31.3%)	2.14 (7.72)	9.29 (19.63)	0.22 (0.58)	1.06 (2.06)
Eye poking	1 (3.7%)	4 (12.5%)	0.04 (0.22)	9.41 (45.25)	0.04 (0.19)	0.72 (3.18)
Self biting	1 (3.7%)	3 (9.4%)	0.15 (0.80)	2.00 (7.84)	0.04 (0.19)	0.75 (2.83)
Self scratching	8 (29.6%)	11 (34.4%)	0.95 (2.0)	2.09 (5.42)	0.37 (0.63)	0.75 (1.85)
Self pinching	0	3 (9.4%)	-	2.85 (11.86)	-	0.53 (1.72)
Orifice poking	7 (25.9%)	12 (37.5%)	4.50 (12.82)	30.76 (89.13)	1.00 (2.30)	2.97 (6.52)
Nail picking	0	0	-	-	-	-

Vocal Stereotyped Behaviours	**3 (11.1%)**	**10 (31.3%)**	**0.44 (1.38)**	**5.00 (15.04)**	**0.85 (3.50)**	**1.50 (3.20)**
Yelling / Screaming	3 (11.1%)	10 (31.3%)	0.44 (1.38)	5.00 (15.04)	0.85 (3.50)	1.50 (3.20)
Repetitive Behaviours with Objects	**7 (25.9%)**	**11 (34.4%)**	**9.80 (23.75)**	**19.13 (47.32)**	**1.70 (4.16)**	**2.34 (4.70)**
Tick	5 (18.5%)	7 (21.9%)	4.20 (13.78)	5.21 (16.83)	0.85 (2.93)	1.09 (2.99)
Wiggle	4 (14.8%)	4 (12.5%)	2.28 (6.03)	8.10 (29.88)	0.41 (1.15)	0.88 (3.10)
Bounce	1 (3.7%)	0	0.25 (1.27)	-	0.11 (0.58)	-
Balance	1 (3.7%)	0	0.27 (1.43)	-	0.04 (0.19)	-
Classify / sort objects	0	0	-	-	-	-
Preoccupations / repetitive movements	1 (3.7%)	1 (3.1%)	2.81 (14.60)	5.82 (32.90)	0.30 (1.54)	0.38 (2.12)
TOTAL stereotyped behaviours	**23 (85.2%)**	**32 (100%)**	**91.81 (136.34)**	**196.67 (185.52)**	**11.70 (13.04)**	**27.44 (20.20)**

Then we analysed differences on the total duration of stereotyped behaviour. This was shorter in cluster 1 (*Md* = 36.74) than in cluster 2 (*Md* = 434.31; *U* = 0, *p* < .001, *r* = .66) and cluster 3 (*Md* = 477.64; *U* = 0, *p* = .004, *r* = .41). On OASID scores, persons in cluster 3 (*Md* = 47.00) scored higher than people in both cluster 1 (*Md* = 23.00; *U*= 12.0, *p*=.03, *r* = .34) and cluster 2 (*Md* = 24.50; *U*= 2.0, *p*=.02, *r* = .61), indicating more autistic symptoms in cluster 3. Based on OASID results, cluster 3 consisted of people with ASD only, when only about half of participants have ASD in cluster 1 and 2. No differences between clusters were found for duration of vocal stereotyped behaviour and stereotyped behaviour with objects, for age, visual impairment, auditory impairment or level of intellectual disability.

Table 4 Mean values on stereotyped behaviour, age and OASID scores for each cluster

	Cluster 1 (n = 46)	**Cluster 2 (n = 10)**	**Cluster 3 (n = 3)**
	M (*SD*)	*M* (*SD*)	*M* (*SD*)
Duration motor stereotyped behaviour (in seconds)	30.29 (51.85)	366.67 (93.02)	121.04 (108.54)
Duration self-injurious stereotyped behaviour (in seconds)	20.61 (33.34)	32.34 (46.70)	343.16 (40.82)
Duration vocal stereotyped behaviour (in seconds)	3.47 (12.67)	.30 (.94)	2.89 (5.01)
Duration stereotyped behaviour with objects (in seconds)	12.95 (37.02)	15.32 (33.86)	42.56 (73.72)
Total duration of stereotyped behaviour (in seconds)	67.33 (69.87)	414.62 (76.74)	509.66 (134.37)
Age (in years)	29.76 (15.40)	36.70 (10.88)	49 (*n*=1)
Score on OASID	26.24 (16.06)	25.30 (13.79)	54.33 (13.58)
Percentage of participants with ASD	50%	60%	100%

4. Discussion

The current study focused on stereotyped and repetitive behaviours in individuals with both an intellectual disability in addition to sensory impairments with or without ASD. Besides measuring the mere presence and type of stereotyped behaviour, this study zoomed in on the intensity of stereotyped behaviour, measured by its frequency and duration. People with sensory and intellectual disabilities show many topographical overlaps in behaviour with people with ASD (de Vaan, et al., 2013). Though for other

populations stereotyped and repetitive behaviours are differentiating factors for the presence of ASD, it might be not for people with sensory and intellectual disabilities, who seem to show stereotyped behaviour regardless of the presence of ASD (Poppes, et al., 2010; Tröster, et al., 1991). This study focused on the question whether stereotyped and repetitive behaviours are indeed characteristics of ASD and are differentiating factors for ASD and no ASD in people that have sensory and intellectual disabilities.

First, we made categories of stereotyped behaviour, based on the literature. Four categories were created, namely motor stereotyped behaviour, self-injurious stereotyped behaviours, vocal stereotyped behaviour and stereotyped behaviour with objects. A principle component analysis did not find the four a priori categories as the best factor solution but a solution with 10 factors. The analyses did not reveal clear latent structures in the stereotyped and repetitive behaviours. However, in our short session, we found an average of only 2 to 4 stereotyped behaviours per individual. This means that the remaining behaviours were not seen on an individual level. Possibly the frequency of stereotyped behaviours and sample size was too low to find any underlying structure.

Secondly, we analysed the prevalence of stereotyped behaviour in our groups of ASD and no ASD and the average number of different stereotyped behaviours each person showed. The groups were created based on a relatively new assessment instrument, OASID (de Vaan, Vervloed, Peters-Scheffer, et al., 2016; De Vaan, et al., 2018). Though this is a new instrument and has therefore not been studied extensively, other existing instruments have proven to be inappropriate for diagnosing ASD in this population (see de Vaan, Vervloed, Hoevenaars-van den Boom, et al., 2016 for a review). In the ASD group all participants showed stereotyped behaviours and in the no ASD group 85% of the participants showed stereotyped behaviours as well. Though still a very large proportion, it was significantly less than in the ASD group. This is in line with earlier findings from a study by Bodfish et al. (2000) who compared individuals with and without ASD with intellectual disabilities. Although stereotyped behaviour was present in both groups, it was more frequent in participants with ASD than in participants without ASD (Bodfish, et al., 2000). On a categorical level there was only a significant difference for self-injurious stereotyped behaviour, which is in line with Richards et al. (2012) who showed that self-injurious behaviour is more frequent among people with ASD and intellectual disability than in people with ASD alone. In addition, we found that people with ASD showed a larger number of different types of stereotyped behaviour than people without ASD. This is in line with earlier research that showed that in persons with intellectual disabilities, persons with ASD showed more stereotyped behaviours than without ASD (Bodfish, et al., 2000; Richards, et al., 2012; Rojahn, Wilkins, Matson, & Boisjoli, 2010). Specifically, we found that violent head banging and finger flicking occurred exclusively in people

with ASD, whereas pacing and bouncing and balancing of objects was only observed in people without ASD. Some other behaviours that we selected were not seen at all in our observations. This could be due to the limited observation time or because some higher order behaviours, such as classifying objects, are only seen in individuals with higher developmental levels (Militerni, et al., 2002). Other behaviours, such as nail picking and finger flicking, might not have been seen because they require fine motor skills that are often not well developed in people with multiple disabilities (Tröster, et al., 1991).

Earlier studies indicated that the intensity of stereotyped behaviour is more severe in people with ASD as opposed to people without ASD (Turner, 1999; Watt, et al., 2008). We tested if this was also the case in people with sensory and intellectual disabilities, by looking at duration, frequency and mean duration per episode. We found that the duration and frequency of stereotyped behaviours was indeed higher for people with ASD; this was found for total duration and self-injurious stereotyped behaviours. However, this difference could have been biased by the lower proportion of people that showed stereotyped behaviours in the no ASD group. It was therefore important first to see if the results could be replicated if only participants that showed stereotyped behaviours were analysed. Secondly we had to take a closer look at the duration of each episode. Our results revealed that when people without stereotyped behaviours were excluded from the analyses, people with ASD only scored higher than people without ASD on total duration and not on self-injurious stereotyped behaviours anymore. In fact, no differences between ASD and no ASD were found for the duration of each stereotyped behaviour episode. Based on our data no conclusions about the presence of ASD can be drawn from the length of an episode.

On average people with ASD showed more types of stereotyped behaviours, they had a higher frequency of stereotyped behaviours and on average they showed stereotyped behaviour of longer durations. However, even in participants without ASD stereotyped behaviours were observed frequently. So, though a high frequency and long duration of stereotyped behaviour can be alarming, especially when it is self-injurious, it does not necessarily indicate the presence of ASD.

Our findings raised the question which factors were involved in the occurrence of stereotyped and repetitive behaviours in people with sensory and intellectual disabilities. To answer this final question, a cluster analysis was performed to search for clusters among participants who showed similar patterns of stereotyped behaviour. Our analysis revealed three clusters, consisting of one cluster with people showing stereotyped behaviour of limited duration, and two clusters with people showing long lasting stereotyped behaviours. Participants in cluster 3 clearly had higher ASD scores and also more self-injurious stereotyped behaviours than persons in the other two clusters. The persons in cluster 2 showed many stereotyped behaviours but they scored the same amount of other ASD characteristics as the

persons in cluster 1 who showed limited numbers of stereotyped behaviour. Apparently in people with both sensory and intellectual disabilities there is a distinct group of people without ASD that show high amounts of stereotyped behaviours, especially in the category of motor stereotyped behaviours. Our further analyses revealed that these clusters did not differ for different levels of intellectual disability, visual impairment, auditory impairment or age. As a result these variables cannot explain the number of stereotyped behaviours. Other factors than ASD or sensory and intellectual disabilities must be related to the occurrence of stereotyped behaviours, such as amount of social reinforcement (Janssen, Schuengel, & Stolk, 2002), level of stimulation that proceeds the behaviour (Hall, Thorns, & Oliver, 2003), or stress and anxiety (Leekam, Prior, & Uljarevic, 2011; Rodgers, Glod, Connolly, & McConachie, 2012).

The study has several limitations that have to be taken into account. A first limitation of this study is that for the principal component analysis the sample size might be too small. A larger number of participants than in this study is normally recommended (Pallant, 2010). The small number of participants could have played a part in not finding clear latent structures among our variables. A second limitation is that OASID assessments were used in both the observations of stereotyped behaviour and in assessing the presence of ASD. This could have resulted in overlap between both measures, because stereotyped behaviour is also one of the criteria for ASD (American Psychiatric Association, 2013) that was also observed in the OASID assessment (de Vaan, Vervloed, Peters-Scheffer, et al., 2016). However, OASID only observed whether stereotyped behaviour was present, whereas in the current observations, we zoomed in on the types, duration, frequency and duration per episode.

The current study showed that, on average, in people with both sensory and intellectual disabilities, the persons with ASD show more stereotyped behaviours than persons without ASD, especially self-injurious stereotyped behaviours. However, as this behaviour also occurs in people without ASD and the duration of each episode does not differ between people with and without ASD, the mere occurrence of stereotyped behaviour cannot be used as a differentiating factor for ASD. In fact, there was a distinct group within our population that do show stereotyped behaviours, but have a low number of autistic symptoms. Hence, the assumed over-diagnosis of ASD in people with sensory and intellectual disabilities can be understood (Andrews & Wyver, 2005; Hoevenaars-van den Boom, et al., 2009). Future research should look at factors that cause stereotyped behaviour in people with sensory and intellectual disabilities, so that over-diagnosis of ASD is prevented and treatment can be aimed at reducing stereotyped behaviours, especially the types that harm the individual.

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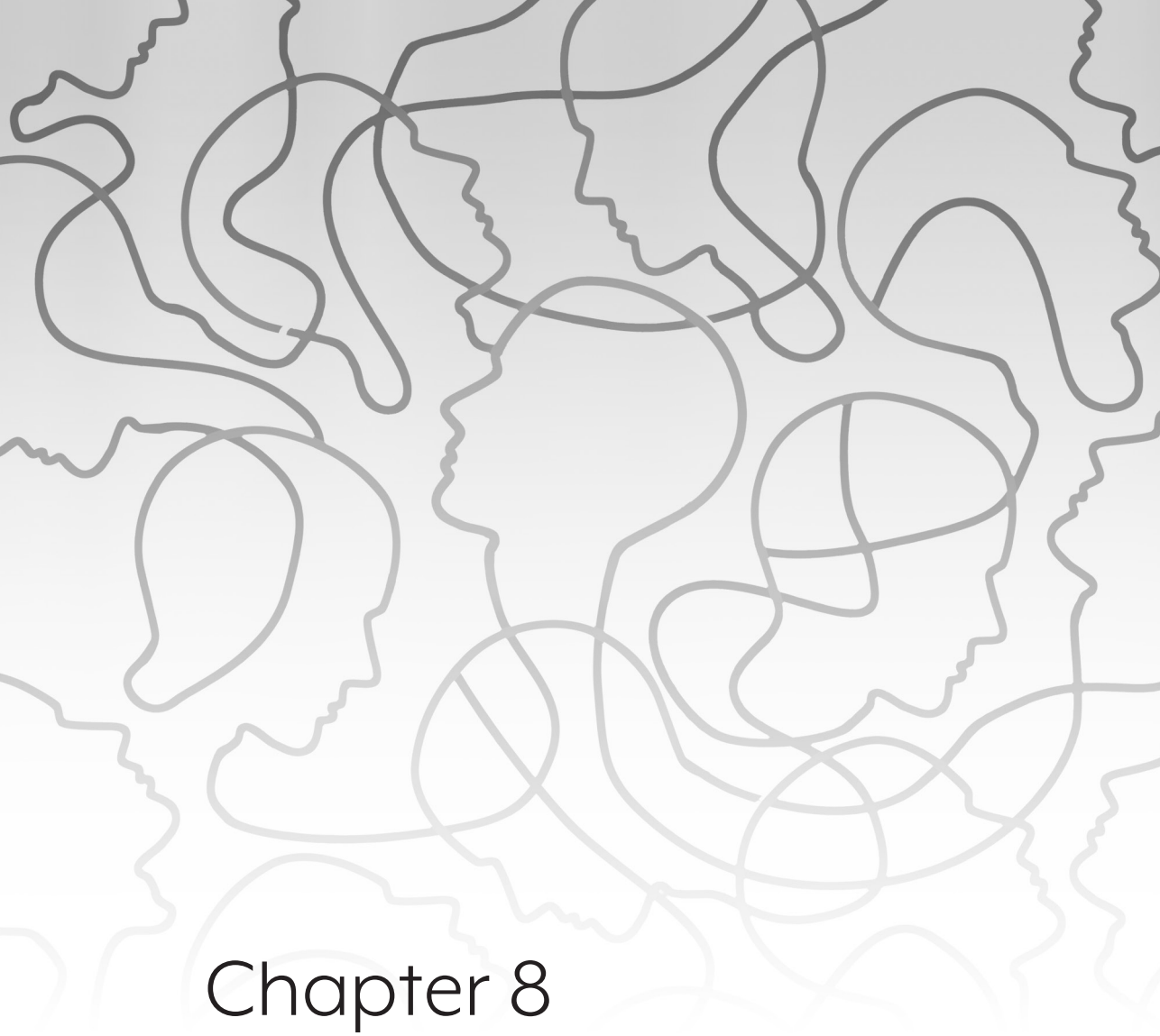

Chapter 8

The relationship between stress and autism spectrum disorder in people with sensory and intellectual disabilities: Evidence from cortisol levels

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Abstract

Individuals with combined sensory and intellectual disabilities are more sensitive to stress than people without disabilities, especially when they have an Autism Spectrum Disorder (ASD). Reversely, stress can also trigger ASD symptoms. The current study investigated the relationship between stress and ASD symptoms in this population.

Participants (n=46) were individuals with combined sensory and intellectual disabilities. The presence of ASD was assessed with Observation of Autism in people with Sensory and Intellectual Disabilities (OASID). This assessment also served as a stressor. Stress levels were measured with salivary cortisol during the OASID assessment and on a control day.

There were no differences in cortisol levels between participants with and without ASD, or between the OASID test day and control day. Half the study sample showed high cortisol levels compared to a reference group. Cortisol levels were positively related to the presence of stereotyped and repetitive behaviours.

No differences were found in stress levels after administration of OASID between people with or without ASD. OASID was found not to produce increases in cortisol. Cortisol levels were correlated with stereotyped and repetitive behaviours, which makes it likely that these behaviours are stress reactions.

1. Introduction

Individuals with a combination of an intellectual disability and sensory impairments with or without an Autism Spectrum Disorder (ASD) can be more susceptible to stress than people without impairments. Missing visual and auditory information from the environment can make situations more unpredictable, more difficult to interpret and to control, hence making these situations more stressful (see Bloeming-Wolbrink et al., 2012; Corbett, Mendoza, Abdullah, Wegelin, & Levine, 2006; Dickerson & Kemeny, 2004). The experience of stress can be defined as a reaction that occurs when a person perceives a threat to their well-being. This reaction can be based on an actual threat or something that is interpreted as a threat (Morilak et al., 2005). This reaction may include psychological reactions, such as feelings of helplessness, and physiological reactions, such as increased heartrate, muscle tension and transpiration (Lovallo, 1997; Schuengel & Janssen, 2006). Also, the stress hormone cortisol is released. This hormone is often measured in saliva, urine or blood (Hellhammer, Wüst, & Kudielka, 2009). The current study will compare individuals with sensory and intellectual disabilities with and without ASD on salivary cortisol concentrations and will assess the relationship between salivary cortisol and ASD characteristics. The relationship between ASD characteristics and stress in this population is expected to be complex and has never been studied before.

Not only are people with sensory and intellectual disabilities more susceptible to stress than people without disabilities, but they additionally lack the ability to adequately cope with stressful situations (Schuengel & Janssen, 2006), which means it may take them longer to recover from stress. For example, a typical way for children or individuals with developmental disabilities to cope with the feeling of stress is to seek comfort with an attachment figure such as a parent or caregiver. This may be difficult for people with intellectual or multiple disabilities, because they are often less securely attached or they lack the behaviours to seek comfort. In general, the experience of stress, coping with stress, attachment behaviours and disabilities seem to be closely related in this population (Giltaij, Sterkenburg, & Schuengel, 2016; Janssen, Schuengel, & Stolk, 2002; Schuengel, de Schipper, Sterkenburg, & Kef, 2013; Schuengel & Janssen, 2006).

Next to cortisol reactivity, cortisol circadian rhythms can look different in individuals with sensory and intellectual disabilities. The circadian rhythm of cortisol is often atypical in people with visual impairments, as this rhythm is influenced by light perception (Lockley, Arendt, & Skene, 2007). Sterkenburg (2008) showed that in people with intellectual disabilities and visual impairment the morning peak is lower and the evening cortisol values are higher than in people without disabilities. This may be related to the attachment problems that were described earlier, since Sterkenburg (2008) found that an attachment-based intervention, that is an intervention that first improved bonding between client and therapist before starting applied behaviour

analysis, led to more typical cortisol patterns in people with intellectual disabilities and visual impairments. Contrarily, the cortisol curves of people with intellectual disabilities and congenital deafblindness were found to be quite normal in a small study by Bloeming-Wolbrink et al. (Bloeming-Wolbrink, et al., 2012).

Not only people with intellectual and sensory disabilities, but also people with ASD may be more susceptible to stress. Though they show similar daily patterns of salivary cortisol levels to healthy controls, there is more variability in stress levels between individuals with ASD (Corbett, Mendoza, Wegelin, Carmean, & Levine, 2008). In people with ASD the initial reaction to novel stimuli is larger than in typically developing individuals (Corbett, et al., 2006) and they are also known to show a more prolonged cortisol response and slower recovery from (social) stressors than people without ASD (Corbett, Schupp, & Lanni, 2012; Spratt et al., 2012).

ASD may not only make people more prone to stress, but stress may also elicit behaviours that are topographically similar to behaviours that are characteristics of ASD. ASD consist of two components, social communication and interaction on one hand, and stereotyped and repetitive behaviour on the other. Both of these behavioural components may be affected by stress. For example, when feeling stressed or helpless a person might revert to stereotyped behaviours (Kraijer, 2004) or social withdrawal (Rubin, Coplan, & Bowker, 2013), both of which are also symptoms of ASD (American Psychiatric Association, 2013; Frith, 2003). So, on the one hand, the presence of ASD may cause people to be more stressed in specific situations (Corbett, et al., 2006). On the other hand, stress may lead to more ASD typical behaviours in people with sensory and intellectual disabilities, regardless of the actual presence of ASD.This complicated relationship between ASD symptoms and stress reactions is even more complex in individuals with sensory and intellectual disabilities, as they are known to show ASD typical behaviours regardless of the presence of stress or ASD (Dammeyer, 2014; De Vaan et al., 2016b; De Vaan, Vervloed, Knoors, & Verhoeven, 2013; Hoevenaars-van den Boom, Antonissen, Knoors, & Vervloed, 2009; Jure, Rapin, & Tuchman, 1991)

The goal of the present study is to begin to clarify the complex relationship between stress and ASD in people with both sensory and intellectual disabilities. Though the target group of people with sensory and intellectual disabilities has been studied with regard to salivary cortisol levels before, this was always done in small samples or single case studies (e.g. Bloeming-Wolbrink, et al., 2012; Nelson, Greenfield, Hyte, & Schaffer, 2013; Sterkenburg, 2008). The current study is the first to include a large group of individuals, enhancing the possibility to generalize the findings to the population of people with multiple disabilities. The sample size and the use of multilevel statistics should make the results also more robust to potential problems of missing data. Furthermore, our study is the first to relate the hormonal stress reaction to the behavioural characteristics of participants with sensory and

intellectual disabilities with and without ASD. The current study hopes to give more insight in the relationship between salivary cortisol levels and the expression of ASD characteristics in individuals with multiple disabilities.

In the current study, an assessment session with an unfamiliar researcher will serve as the stressor. This session is a novel situation performed by an unfamiliar psychologist and includes social evaluation, and thus is potentially stressful, especially for participants with ASD (Dickerson & Kemeny, 2004). The stress reactions will be assessed by studying cortisol levels during and after the assessment and will be compared with similar measures taken on a typical day to correct for individual variation in salivary cortisol.

This study has two research questions. The first question focuses on whether the stress reaction differs in people with combined sensory and intellectual disabilities with and without ASD. Based on the literature we expect that individuals with combined sensory and intellectual disabilities and ASD will show a stronger stress reaction and a slower recovery from stress than individuals with combined sensory and intellectual disabilities without ASD. In addition, we will compare the cortisol concentrations to earlier reported reference values (Miller et al., 2016). The second question focuses on whether the stress reaction is related to autism-typical behaviours in individuals with combined sensory and intellectual disabilities, regardless of an ASD diagnosis. We expect to find a positive relationship between stress reactivity and autistic behaviour, as experiencing stress could lead to behaviours that are also typical for ASD. These behaviours will be assessed on both of the two major components of ASD typical behaviours as described in the DSM-5 (American Psychiatric Association, 2013), 'stereotyped and repetitive behaviour' and 'social communication and interaction'.

2. Method

2.1 Participants

Participants were recruited in four residential facilities and three schools for people with combined sensory and intellectual disabilities. Participants were recruited by a contact person from each facility. The inclusion criteria were a moderate to profound intellectual disability combined with a visual impairment. The participants were allowed to have an additional auditory impairment. Information about visual impairments, auditory impairments and intellectual disabilities were retrieved directly from the participants' records kept at the facilities.

Sixty participants with combined sensory and intellectual disabilities were recruited for this study. In five cases legal representatives gave no consent to collect saliva, in four cases participants did not accept saliva sampling and in five cases not

enough saliva was collected to analyse cortisol levels. Overall, saliva samples of 46 participants were included (a response rate of 77%).

There were 31 male and 15 female participants with a mean age of 33.8 years (*SD* = 14.74, range 6-55). Participants had a moderate (*n*=9), severe (*n*=17) or profound (*n*=20) intellectual disability. All participants had a visual impairment, 19 of them were blind without light perception. Ten participants had an additional auditory impairment and three were completely deaf. As part of this study, two groups were made using the OASID (see materials section), ASD and no ASD. The no ASD group consisted of 22 individuals (15 males) with a mean age of 35.7 (*SD* = 13.1), the ASD group consisted of 24 individuals (16 males) with a mean age of 31.81 (*SD* = 16.2).

This study was approved by the local Committee on Research Involving Human Subjects and conformed to the Ethical Principles for Medical Research Involving Human Subjects of the World Medical Association declaration of Helsinki (World Medical Association, 2013). All participants were legally incapable, so parents or legal representatives were asked for informed consent before the study started.

2.2 Materials

2.2.1 Autism Spectrum Disorder

The presence of ASD was assessed using Observation of Autism in people with Sensory and Intellectual Disabilities (OASID) (De Vaan et al., 2016a; De Vaan, et al., 2016b). OASID is an assessment tool consisting of a semi-structured play session. During the play session the experimenter played five tasks with the participants using toys and games. The session is adapted to each individual by taking into account the participant's intellectual disabilities, sensory impairments and communication skills. The assessment lasted between 30 and 60 minutes.

The assessment was recorded on video and observed and scored afterwards, using a 40 item questionnaire. Each item was scored on a Likert scale from 0-2, where a higher score corresponded with more autistic behaviours. Item scores were added to calculate two scale scores, based on the two main criteria for ASD as described in DSM-5 (American Psychiatric Association, 2013): 'Social Interaction and Communication' and 'Repetitive and Stereotyped Behaviour'. Scores on both scales were used to assess the presence of ASD according to the guidelines of OASID (De Vaan, et al., 2016a).

OASID was found to be both a valid and reliable measurement tool to assess ASD symptoms in individuals with a combination of intellectual disabilities and sensory impairments (De Vaan, et al., 2016a, 2016b). In this study OASID was used to assess the presence of ASD. The OASID play session was also used as the stressor for the cortisol measure. Though OASID was designed as a non-stressful measurement, because it is still an assessment this may be stressful for participants nevertheless.

2.2.2 Cortisol

Levels of physiological stress were determined using cortisol measurements in saliva, following the protocol of Bloeming-Wolbrink et al. (2012). Saliva was collected using Salivettes, cotton rolls that were used to swab the participant's mouth. Saliva sampling was done by a familiar caregiver of the participant who was instructed on the procedures by letter and video. Caregivers were instructed to wear medical gloves in order to keep the cotton rolls sterile. The salivettes were stored in a fridge immediately after sampling, and frozen within a few days after sampling at -20 °C until further analysis. Salivary cortisol was measured by the University Medical Centre in Utrecht the Netherlands and was measured without extraction using an in house competitive radio-immunoassay employing a polyclonal anticortisol-antibody (K7348). [1,2-3H(N)]-Hydrocortisone (PerkinElmer NET396250UC) was used as a tracer. The lower limit of detection was 1.0 nmol/l and inter- and intraassay variations were below 10%.

Saliva was collected six times, three times on the OASID test day, and three times on a control day. Caregivers were instructed that the participants could not eat, drink (except water if necessary) or brush their teeth an hour before the saliva samples were taken (see also Bloeming-Wolbrink, et al., 2012). On the test day, saliva samples were taken before the beginning of the OASID assessment (prestressor, T1), 35 minutes after the beginning of OASID (stress reaction, T2) and 75 minutes after the beginning of OASID; which is 35 minutes after the end of OASID (the average duration of OASID is 40 minutes - recovery, T3). A cortisol reaction is visible in saliva around 25 minutes after the stressor, although inter-individual differences exist. To take this inter-individual variation into account, the samples were taken 35 minutes after the stressor. On the control day, saliva samples were taken at the same times as on the test day to assess the cortisol pattern during a standard day. To control for the cortisol awakening response (see Lovallo, 1997), all the OASID assessments were done after 11.00 AM. For 29 participants all six samples were analysed, for 5 participants there was one missing sample, for 4 participants there were two, for 6 participants there were three, for one participant there were four and for one participant there were five missing samples.

2.3 Procedure

After written consent was given, the OASID assessment was planned and caregivers were informed about the procedure. They were asked to be present during the assessment and to perform the saliva sampling. For the comfort of the participant, we chose to let familiar caregivers perform the saliva sampling instead of doing this ourselves as unfamiliar researchers. Before the assessment, the caregivers received information about the protocol in text and video, with instructions for saliva sampling. They also received all of the necessary materials, including salivettes, with some

additional salivettes for practice and medical gloves. They were given the opportunity to ask questions about this procedure. The OASID assessment was performed and the saliva samples were taken. Saliva samples on the control day were taken at the exact same times as on the test day. This control day was before or after the test day and the same procedure was followed for collecting and storing cortisol. Saliva samples of all participants were stored in a freezer until analysis.

2.4 Statistical analyses

First, all variables were checked for outliers (>3 SD difference from the mean). One outlier was detected for the cortisol measurements on the OASID test day, and four outliers for the cortisol measurements on the control day. Since an advantage of multilevel analyses is its robustness for missing data, these outliers were removed before analysis (Tabachnik & Fidell, 2007). The residuals were normally distributed.

To test whether the cortisol response to OASID differed between people with and without ASD, two longitudinal regression analyses were performed using mixed-model (multilevel) designs. One analysis aimed to test the difference between people with and without ASD, and the other analysis aimed to test whether the continuous scales of 'Social interaction and communication' and 'Repetitive and stereotyped behaviour' were able to predict the people's cortisol response to the administration of OASID.

In these analyses, the three repeated cortisol measures (T1-T3) were used at Level 1 and nested within the participants at Level 2. To examine whether the nested structure was required, the intraclass correlation (ICC) was calculated using a null model for the area under the curve (AUC). The ICC for children's cortisol AUC measure was 0.7772, indicating that 77.72% of the variability in cortisol responses to the OASID was associated with differences between participants, meaning that multilevel analyses were applicable.

Thereafter, a build-up strategy was followed in which variables were added one-by-one to the model with random intercept (allowing the intercept of the regression line to vary per participant). After adding each variable, the change in deviance on the -2 log likelihood ratio scale after generalized least square estimations was assessed. Variables that did not improve the model by significantly reducing the deviance were excluded. Time (considered as a random factor, allowing the slope of the regression line to vary per participant) and quadratic time (to indicate a cortisol response to OASID) were entered into the model first. Secondly, the confounders were entered into the time models. The following confounders were taken into account separately: all three cortisol measurements on the control day, sex, age, level of visual and auditory impairments, level of intellectual disability and time of the day that the OASID took place. Lastly, the predictors and the interactions between the predictors and time were entered into the model. To test whether cortisol response to OASID differed between people with and without ASD, the first multilevel model

contained the predictor ASD or no ASD. To test whether the continuous scales of OASID, regardless of ASD diagnosis, were able to predict the participant's cortisol response to OASID, the second multilevel model contained the predictors 'Social interaction and communication' and 'Repetitive and stereotyped behaviour' instead of ASD diagnosis.

3. Results

3.1 Differences in cortisol between participants with ASD and without ASD

Two groups were created based on OASID scores, no ASD symptoms (n=22) and ASD symptoms (n=24). The mean cortisol level in nmol/l for each group and the moment of measurement is provided in Table 1. Independent samples T-tests revealed no differences in cortisol level between ASD and no ASD for any of the cortisol measures on both the test- and the control day. Also within groups no differences were found on cortisol level between the different measurement times.

The absolute values of these cortisol levels have been compared to reference values described by Miller et al. (2016) to see if they fall into the normal range (i.e. between the 5th and 95th percentile). All individual values were compared to the reference values of the corresponding sex, age and number of hours after awakening. This was done separately for T1 and C1. The other cortisol measures were not taken into account because these values may be influenced by any stress caused by the administration of OASID or saliva sampling. On T1, 45.5% of the no ASD sample and 50% of the ASD sample were within the normal range, whereas the others had a cortisol value above the 95th percentile. On C1, 47.6% of the participants without ASD and 50% of the participants with ASD had a cortisol value within the normal range, the others had values above the 95th percentile that was described by Miller et al. (2016).

Multilevel analyses were performed to assess the effect of ASD on the cortisol response to the OASID administration. Table 2 represents the best fitting multilevel model. The predictors time and time quadratic were not significant, indicating that the OASID did not provoke a significant cortisol response for the whole group. Furthermore, the dummy indicating whether people had ASD symptoms or not, based on OASID, was not significant, indicating that the cortisol response levels of people with and without ASD symptoms were similar to the assessment, so with no significant cortisol responses. The cortisol measurements on the control day significantly predicted the matched cortisol responses to the OASID. Higher cortisol concentrations on the control day predicted higher cortisol concentrations on the OASID test day.

Table 1 Mean cortisol levels in nmol/l for participants with and without ASD

	No ASD		ASD	
Cortisol measure[1]	*n*	*M(SD)*	*n*	*M(SD)*
T1	19	10.79 (5.07)	20	10.65 (4.35)
T2	18	10.47 (3.18)	21	10.50 (3.69)
T3	16	9.26 (2.42)	20	9.92 (4.08)
C1	21	10.40 (3.21)	19	13.03 (7.07)
C2	21	10.92 (4.81)	21	12.20 (11.03)
C3	19	9.82 (3.84)	21	11.52 (9.12)

Note. [1]T1-T3 assessments on test day, C1-C3 assessments on control day

Table 2 Best fitting multilevel model studying the effect of ASD on cortisol response

	Cortisol response to the OASID		
	Estimate	*SE*	*p*
Model 1 ASD and no-ASD*			
Intercept	7.8744	1.8005	.000
Time	-.0088	.0065	.186
Cortisol measurements control day	.2123	.0854	.015
Age	.0053	.0390	.894
ASD classification (0=no-ASD, 1=ASD)	.3705	1.0687	.731
Deviance	448.897		

Note. Repeating the analyses without the cortisol measurements on the control day led to similar results.

3.2 Cortisol responses and relations with autistic behaviour

Another multilevel analysis was performed to assess how cortisol responses related to autistic behaviour. Instead of dichotomizing the ASD diagnosis, the total scores on the two scales of OASID, 'Social interaction and communication' and 'Repetitive and stereotyped behaviour', were used. Table 3 represents the best fitting multilevel model predicting the cortisol response after administering OASID with the scores on the two OASID scales. More repetitive and stereotyped behaviour during OASID administration significantly predicted higher cortisol concentrations on the OASID test day, after controlling for cortisol concentrations on the control day. No such relationship was found for the other ASD behavioural aspect, 'Social interaction and communication'. Lastly, higher cortisol concentrations on the control day significantly predicted higher cortisol concentrations on the OASID test day.

Table 3 Best fitting multilevel model studying the association between ASD symptoms measured with OASID scales and cortisol response

	Cortisol response to the OASID		
	Estimate	*SE*	*p*
Model 2 Continuous OASID scales*			
Intercept	6.8163	1.7587	.000
Time	-.0087	.0065	.186
Cortisol measurements control day	.1943	.0849	.025
Age	-.0017	.0372	.963
Repetitive and stereotyped behaviour	.1834	.0885	.046
Deviance	444.943		

Note. Repeating the analyses without the cortisol measurements on the control day led to similar results.

4. Discussion

The current study investigated whether cortisol responses to a stressor are related to ASD and ASD symptoms in people who have sensory impairments in combination with intellectual disabilities. Both people with ASD and people with sensory and intellectual disabilities are more prone to experiencing stress in novel, unpredictable and uncontrollable situations (e.g. Bloeming-Wolbrink, et al., 2012; Corbett, et al., 2006; Schuengel & Janssen, 2006). However, it is unknown what the additional effect of ASD is on stress levels when a person has these multiple disabilities. In addition, this study explored the relationship between salivary cortisol levels and the behavioural characteristics of ASD in this target population. No differences in cortisol levels or cortisol responses were found between participants with and without ASD, but a higher cortisol response was found to be related to more stereotyped and repetitive behaviours, one of the core characteristics of ASD.

The results showed that there were no differences in cortisol levels between participants with and without ASD, and also no difference in cortisol levels between the OASID test day and cortisol levels on a typical day (i.e. control day) for either group. The absolute values of salivary cortisol concentrations are quite high as compared to reference values described by Miller et al. (2016). A little over half of our participants, with and without ASD, had cortisol values above the 95th percentile of normal cortisol values that were described by Miller et al. (2016). Possibly, for some participants a ceiling effect had occurred. If this is true, additional stress would not be visible in salivary cortisol levels. However, any conclusions regarding ceiling effects should be drawn with care, because of the large individual variations in baseline

cortisol levels. Moreover, the norm values that Miller et al. (2016) described were all based on saliva samples analysed in only two different laboratories (Trier and Dresden, Germany) which was different from the lab where our cortisol analyses were done (Utrecht, the Netherlands).

Our multilevel analyses further confirmed the preliminary findings that cortisol levels did not differ between groups or moment of testing, as neither the assessment with OASID nor the presence of ASD predicted cortisol levels. The only significant predictor of cortisol levels on the OASID test day was the cortisol level on the control day. Baseline cortisol levels can vary between individuals (Bartels, Van den Berg, Sluyter, Boomsma, & de Geus, 2003; Smyth et al., 1997). This individual variation in salivary cortisol levels were the only significant predictor for salivary cortisol levels on an OASID test day. Neither the presence of ASD, nor the assessment of OASID was related to higher cortisol levels.

Possibly, OASID was too mild of a stressor for any of the participants to show stress reactions in the first place. We chose the administration of the OASID assessment as a stressor because it is an assessment which may be stress producing for participants. However, OASID did not cause cortisol reactivity in either group, nor in the group as a whole. Neither of the groups showed a cortisol response to OASID, which could be the cause of not finding any differences between the groups on cortisol reactivity. Though the assessment was never designed to produce stress, in fact, precautions were taken to prevent stress (De Vaan, et al., 2016b), we still expected the session to be stressful to some extent. For example, because of the social evaluative aspects of the administration (Dickerson & Kemeny, 2004) and the fact that the session was with an unfamiliar researcher in an unfamiliar setting. The fact that OASID was not found to be stressful for participants could be seen as a limitation of this study because the intended stressor did not appear to be stressful. On a more positive note, this finding shows that OASID can measure autistic behaviours without producing physiological stress and that the precautions taken to prevent stress may have been successful.

In the second part of this study, we assessed if any of the behavioural aspects typical for ASD were related to stress. One behavioural domain of ASD, namely 'stereotyped and repetitive behaviour' was correlated with cortisol concentrations on the OASID test day, while controlling for cortisol concentrations of the control day. It is known that stress or anxiety can lead to stereotyped behaviour (Leekam, Prior, & Uljarevic, 2011; Rodgers, Glod, Connolly, & McConachie, 2012) but also that specific movements such as body rocking could be a possible way to cope with stress (Bloeming-Wolbrink, et al., 2012). At the moment it is, given the design used in the current study, impossible to determine any causal relation between repetitive and stereotyped behaviour and cortisol responses. We can only verify a correlation between both variables. The other aspect of ASD, 'social interaction and

communication', was unrelated to cortisol levels. Although stress can lead to social withdrawal (Rubin, et al., 2013) and negative feelings as a result of social evaluation may lead to stress (Dickerson & Kemeny, 2004), our data did not find an association between social behaviours and cortisol responses.

This study has some limitations. Since multilevel analyses are robust against missing data we could study a relatively large group of 46 individuals, but all six cortisol samples could only be analysed for 29 participants. For the remaining 17 participants the number of missing measurements ranged from 1 to 5. For nine participants there were no usable cortisol samples, so their data were not included in the analyses. This shows that it was rather difficult to collect saliva in this target population. Missing values were caused by too little saliva and by the participants refusing to provide any saliva. Some participants did not accept the cotton swab in their mouths for more than a few seconds or did not accept it at all. Because our sample consisted of people with a moderate to profound intellectual disability (IQ $<$ 50), combined with additional sensory impairments, there were only limited possibilities to communicate with them, to explain the intention of the saliva swab, and to persuade them to produce saliva. Though sampling of salivary cortisol is described as non-invasive and stress-free (Levine, Zagoory-Sharon, Feldman, Lewis, & Weller, 2007), this may not have been so for our study population. In fact, perhaps the persons that refused saliva sampling were stressed and only the non-stressed individuals were therefore included in our study. This could have led to our lack of differences between groups.

Another possible reason for the fact that we could not extract enough saliva might also be due to staff not using the salivettes correctly. For reasons of comfort the saliva sampling was done by familiar caregivers of the participant. However, these caregivers had no experience or expertise with saliva sampling. They received instructions on how to collect saliva by video and text, but they were not trained in person in sampling saliva. In order to increase the sampling success in future studies, it would be recommendable to give more training to the persons collecting saliva, or have the saliva collected by a more experienced researcher in the presence of a familiar caregiver. Despite these difficulties in collecting saliva in this multiple disabled population, we still believe it to be the best method to measure cortisol levels in this group. Cortisol can also be measured through blood sampling, but this is painful, expensive and requires medical staff (Levine, et al., 2007). Other stress measures such as heart rate or skin conductance (e.g. Meehan, Insko, Whitton, & Frederick P. Brooks, 2002) require equipment strange to the participants to be placed on their bodies, which participants with multiple disabilities may not understand and reject. Finally, the communication difficulties of this population make it challenging if not impossible to validly use self-report scales to assess stress levels. Hence, salivary cortisol is still the best way to measure cortisol responses in this target group.

The current study revealed no differences in cortisol levels between people with and without ASD when they have a combination of sensory and intellectual disabilities. This implies that an additional ASD does not lead to higher cortisol responses, and perhaps more stress, in multiple disabled individuals. We did however find stereotyped and repetitive behaviours were related to cortisol responses on OASID. Clinical practice should take this into account when treating individuals with multiple disabilities that show stereotyped behaviours, especially as this may be a self-regulating process of coping with experienced stress (Bloeming-Wolbrink, et al., 2012). Stereotyped behaviour in this sense is then a good warning signal for stress and treatment could then be focused on the reduction of stress instead of on reducing the stereotyped behaviours. This finding could also have strong implications for diagnosing ASD in this group. The observation of ASD typical behaviours such as stereotyped movements is not necessarily indicative of ASD but could be a symptom of stress. This is in line with earlier studies that have shown that in this target group the biggest differences between individuals with and without ASD is on the social and communicative domain (De Vaan, et al., 2016a, 2016b; Hoevenaars-van den Boom, et al., 2009). More research should be done on the relationship between stress and stereotyped behaviours in this target population, especially if this relationship turns out to be causal.

5. Conclusion

This study did not reveal differences in cortisol responses between participants with and without ASD, but it has revealed a relationship between cortisol and a behavioural characteristic that is related to ASD, stereotyped and repetitive behaviour. The OASID assessment did not provoke rises in cortisol concentrations in participants of this study. This could be due to a ceiling effect for some participants but it could also imply that OASID was not stressful and may be used without problems as an assessment instrument for ASD. At the same time, the results indicate that OASID cannot be used as an experimental stressor in future research. In order to compare cortisol responses in people with and without ASD, future studies should look at other ecologically relevant and ethically acceptable situations, occurring naturally, that are potentially more stressful events, such as medical examinations, visits to the dentist, or vaccinations. Finally, the collection of saliva in order to measure cortisol was challenging in this population. Nonetheless, we recommend this procedure over alternative stress measures provided that the staff is well trained in sampling saliva to ensure comfortable sampling while preventing high numbers of missing values.

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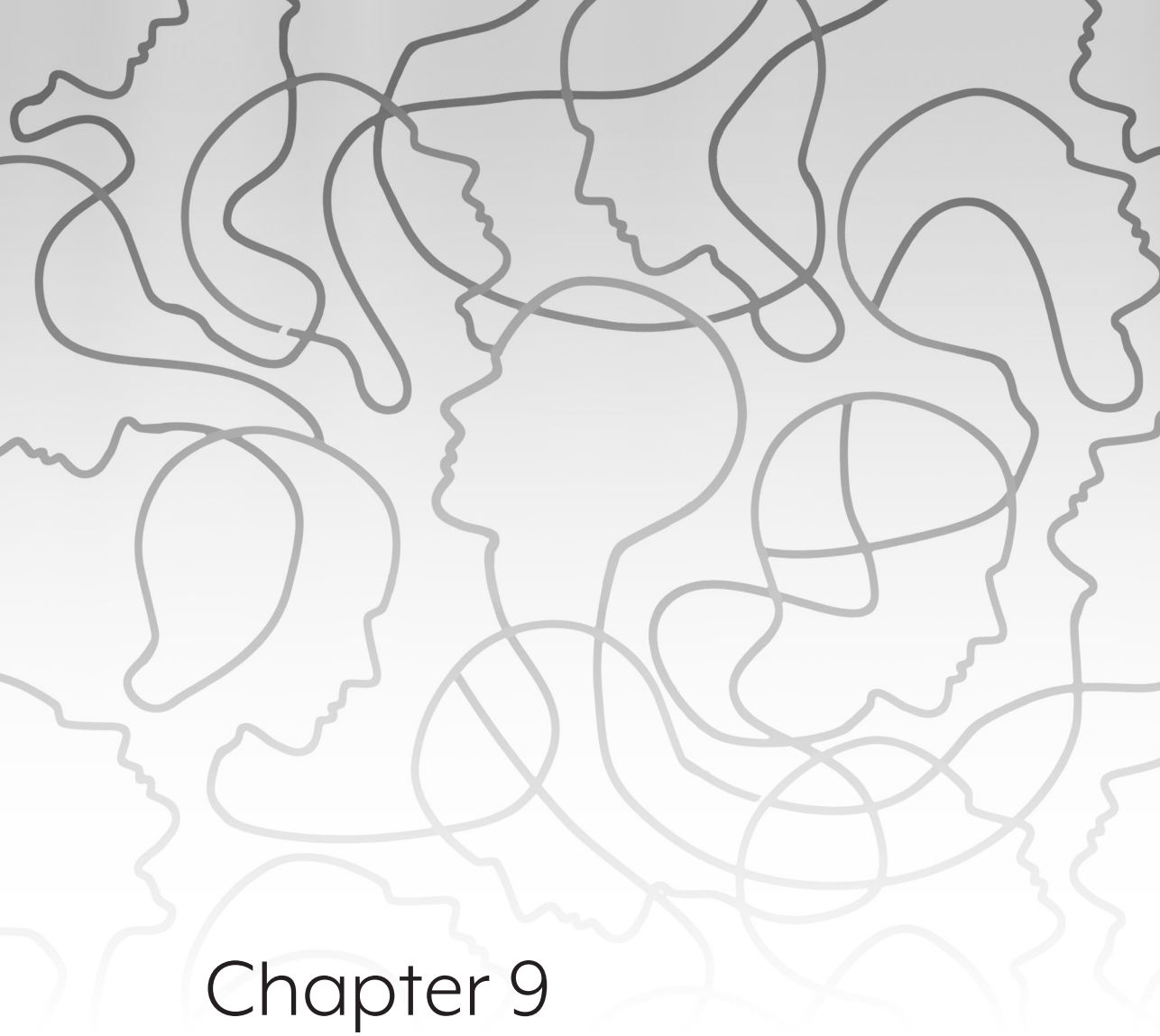

Chapter 9

General Discussion

People who have combined sensory and intellectual disabilities show behaviours that could be indicative of Autism Spectrum Disorder (ASD), such as social withdrawal or stereotyped behaviour. Although these behaviours can indeed be caused by ASD, they may also be caused by sensory or intellectual disabilities (Evenhuis, Sjoukes, Koot, & Kooijman, 2009; Hoevenaars-van den Boom, Antonissen, Knoors, & Vervloed, 2009) or by other mental health problems such as an insecure attachment style, stress, anxiety or mood disorders (Janssen, Schuengel, & Stolk, 2002; Rodgers, Glod, Connolly, & McConachie, 2012; Rubin, Coplan, & Bowker, 2013). There is a lot of overlap in behaviour between multiple disabilities, ASD and mental health problems. The purpose of this thesis was to unravel this overlap in two ways: firstly, by developing an instrument that could help in the diagnosis of ASD within people with sensory and intellectual disabilities, and secondly by using this instrument to assess the presence of ASD in persons with multiple disabilities to gain more insight into their behavioural characteristics that underlie possible mental health problems.

9.1 Screening for ASD in individuals with multiple disabilities

It is considered difficult to screen for ASD symptoms in individuals with combined sensory and intellectual disabilities. This is so challenging because of the behavioural overlap in characteristics of individuals with sensory and intellectual disabilities with symptoms of ASD. Chapter 2 showed that all characteristics of ASD as described by the DSM-IV-TR (American Psychiatric Association, 2000) are, at least to some extent, also present in individuals with multiple disabilities, regardless of the presence of ASD. Though the criteria for ASD have been slightly changed in the DSM-5 (American Psychiatric Association, 2013), the overlap in characteristics is still evident. For example, children who are blind may show a lack of reciprocity in social interaction (Celeste, 2006; Fraiberg, 1977), persons with intellectual disabilities may show fewer signs of joint attention (Osterling, Dawson, & Munson, 2002) and generally more language impairments (Tager-Flusberg, Paul, & Lord, 2005). Additionally, repetitive and stereotyped behaviour is often seen in individuals with intellectual disabilities and visual impairments (Gense & Gense, 2005; Kraijer, 2004; Warren, 1994).

In addition to the overlap in behavioural characteristics, another difficulty in recognizing symptoms of ASD is the lack of valid instruments for screening and diagnosis of ASD within this population. Instruments that are commonly used to assess the presence of ASD show adequate psychometric properties for the intended population. Unfortunately, most instruments were not designed to be used in persons with multiple disabilities. Of all of the instruments that were reviewed in Chapter 3, at least 25% of items were not suitable to be used in persons with combined intellectual and sensory disabilities. This is due to what we called a sensory bias; these items could not be interpreted in the same way as in persons without sensory impairments.

The inability to correctly attribute behaviours to either ASD or other present disabilities, can lead to both an over- and under-diagnosis of ASD. In clinical practice, this can have problematic consequences. The clinical classification of an individual often serves as the basis for treatment (Howlin, 2000; Rutter, 2006). If anything goes wrong in the diagnostic process, possibly due to overlapping behaviours or to a lack of valid diagnostic instruments, a person may end up with a wrong diagnosis of ASD. This may lead to the wrong treatment which can result in counter-effective results. The need for a new instrument that can validly recognize symptoms of ASD within this target population is evident. This new instrument should focus on the behavioural characteristics that differ between persons with and without ASD within this population. For example, persons with multiple disabilities without ASD show more openness for social contact and more pleasure when engaging in social contact than persons with multiple disabilities with ASD (Hoevenaars-van den Boom, et al., 2009). Persons without ASD may also show less severe and persistent stereotyped behaviours that can be interrupted more easily than in individuals with ASD (Bodfish, Symons, Parker, & Lewis, 2000; Gense & Gense, 2005).

9.2 Validating the observation of ASD symptoms

An instrument was developed that could differentiate ASD symptoms from behavioural characteristics of sensory and intellectual disabilities. A starting point was the O-ADD that was designed for Individuals with profound intellectual disabilities and deaf-blindness (Hoevenaars-van den Boom, et al., 2009). The new instrument that was developed had to be suitable for a broader range of individuals with intellectual disabilities (moderate to profound), and for visual impairments with or without deaf-blindness. Next to being reliable and valid, administration had to be shorter in duration and not stressful for participants. This resulted in a newly developed instrument, OASID: Observation of Autism in individuals with Sensory and Intellectual Disabilities. Items of OASID were based on the O-ADB, the differentiating characteristics of ASD and multiple disabilities that were retrieved from Chapter 1 and the diagnostic criteria for ASD that are described in the DSM-5 (American Psychiatric Association, 2013). The OAD-B was adjusted in procedure and items, making the assessment less stressful and time consuming, and making the scoring process easier.

Both the pilot study on 18 participants and the larger study on 60 participants showed promising results regarding psychometric properties. The pilot study was described in Chapter 4 and showed substantial interrater reliability, good discriminant validity with the list of disturbed attachment behaviour (Boris & Zeanah, 2005) and moderate convergent validity with the Pervasive Developmental Disorder in Mental Retardation Scale (PDD-MRS; Kraijer, 1999). Additionally, this study showed that there was no overlap in OASID scores between individuals with and without ASD when the diagnosis was based on expert consensus. Chapter 5 showed the results

of our larger study with 60 participants. We found an almost perfect intra-rater reliability, good interrater reliability and good internal consistency for both scales of OASID. The criterion validity of OASID was found good in comparison with the PDD-MRS (Kraijer, 1999), the Childhood Autism Rating Scale 2 (CARS2; Schopler, Van Bourgondien, Wellman, & Love, 2010) and moderate with the judgments about the presence of ASD done by two experts. Finally, in Chapter 5, reference points for the interpretation of OASID scores were introduced. OASID scores can be translated into descriptions on the severity of symptoms: No ASD symptoms, Mild ASD symptoms, Severe ASD symptoms or Profound ASD symptoms. This is in line with the idea that ASD occurs on a severity spectrum.

From both studies it appeared that OASID was valid and reliable in its purpose to assess ASD symptoms in individuals with combined intellectual and sensory disabilities with more certainty. No tools existed that were specifically designed for this population and purpose. Additionally, OASID can not only assist in assessing the presence of symptoms but also the severity of symptoms. Since ASD is said to occur on a spectrum, an instrument that can assess the severity of symptoms is of added value in clinical practice (Risi et al., 2006). However, for a complete diagnostic evaluation OASID only is not enough. A multidisciplinary and multimethod approach is required and differential diagnosis is an important part of this process. Furthermore, for treatment planning a broader assessment needs to take place, including an assessment of the person's cognitive abilities, adaptive skills and communication skills (Volkmar et al., 2014). Finally, follow-up procedures regarding treatment need to be planned so that the diagnostic process is not without purpose (Oosterling et al., 2010)

9.3 ASD related behaviour and mental health problems

The behaviours that are known as symptoms of ASD can indeed be an indication for ASD, but they can also be caused by a person's disabilities or even by other mental health problems. For example, mental health problems can lead to stereotyped behaviours, one of the core characteristics of ASD (Kraijer, 2004; Rubin, et al., 2013; Stewart, Barnard, Pearson, Hasan, & O'Brien, 2006). Simultaneously, both the presence of ASD and multiple disabilities can make a person more sensitive to developing mental health problems, such as stress, attachment problems or mood disorders (Bloeming-Wolbrink et al., 2012; Corbett, Mendoza, Abdullah, Wegelin, & Levine, 2006; Hurley, 2006; Janssen, et al., 2002; Rutgers, Bakermans-Kranenburg, Van IJzendoorn, & Van Berckelaer-Onnes, 2004). The presence of mental health problems can further complicate the matter of attributing behaviour patterns to a disability or disorder within persons with multiple disabilities.

Chapter 6 looked at the presence and severity of mental health problems in individuals with multiple disabilities and the relationship between ASD and mental health problems. The presence of ASD was assessed using OASID. It appeared that

mood disorders or stressors were not remarkably prevalent in our population of persons with combined sensory and intellectual disabilities. On the contrary, almost everyone from our sample showed signs of a disturbed attachment relationship, especially so in individuals with ASD. Possibly, this means that it is more difficult for our target group to develop secure attachment relations, perhaps because of the nature of their disabilities or due to living in an institution (Zeanah, Smyke, Koga, Carlson, & The Bucharest Early Intervention Project Core, 2005). However, it is also possible that the symptoms of an insecure attachment overlap with symptoms of ASD or multiple disabilities, making it difficult to recognize an insecure attachment style in this population. For example, an insecure attachment style may result in being socially withdrawn and a lack of showing affection (Boris & Zeanah, 2005), which can also be interpreted as signs of having an ASD.

Additionally, stereotyped and repetitive behaviours are part of the diagnostic criteria for ASD. However, in persons with multiple disabilities they can be related to a variety of factors, including stress and anxiety (Leekam, Prior, & Uljarevic, 2011; Rodgers, et al., 2012). In fact, Chapter 7 showed that stereotyped behaviours occur in individuals with multiple disabilities, both with and without ASD, making it impossible to use stereotyped behaviours as a differentiating factor for ASD in individual cases. However, as a group, persons with ASD do show on average more types, a higher frequency and a longer duration of stereotyped behaviours. The remaining question is still what the underlying cause is for these behaviours. ASD cannot be the only cause as stereotyped behaviours are also seen by individuals without ASD. Part of the answer to this question can be found in Chapter 8 that revealed that the presence of stereotyped and repetitive behaviours was related to the level of the stress hormone cortisol. This is in line with earlier findings that stress or anxiety can cause stereotyped and repetitive behaviours (Leekam, et al., 2011; Rodgers, et al., 2012).

In summary, ASD symptoms are found on two behavioural domains: Social behaviour and communication, and stereotyped and repetitive behaviours (American Psychiatric Association, 2013). When a person with multiple disabilities shows impairments on these domains, they may be caused by ASD, by their intellectual or sensory disabilities, but also by mental health problems. Our study showed that in persons with multiple disabilities attachment problems are very prevalent, which can be an underlying cause for behavioural impairments on the social domain. Also, we found stress to be related to the domain of stereotyped and repetitive behaviours. This means that impairments on both domains of ASD can be caused by other factors within our target population. It is therefore of high importance to screen not only for ASD but also for other potential causes of behaviour problems such as an attachment disorder or stress disorder. When an attachment or stress disorder is likely to be present, treatment should first be aimed at these disorders before concluding the person has ASD and focusing treatment in this direction.

9.4 Behavioural overlap

The large amount of behavioural overlap makes the diagnostic process difficult. Morton and Frith (2002) argued that we should not rely on behaviour alone when interpreting developmental disorders. The same behaviours may be caused by a variety of underlying reasons. This is also seen in our study, where sensory and intellectual disabilities, but also stress and attachment disorders can result in de same behaviours that are scored to diagnose ASD. Morton and Frith (2002) explained that in order to understand behaviour, we should look at the cognitive and biological cause of these behaviours. However, as the same behaviour can have multiple causes, multiple causal models need to be developed and tested to see which applies to which individual (Krol, Morton, & De Bruyn, 2004).

Contrary to this causal modelling approach is network analysis. Within the network approach discrete symptoms are described in their interaction with other symptoms; they activate and influence each other. This interplay can then create a network of symptoms that clinicians label with a certain diagnostic classification. The diagnosis is then the result of the symptoms, rather than the reverse in which the symptoms are seen the result of one underlying cause (Borsboom & Cramer, 2013; Boschloo et al., 2015; Ruzzano, Borsboom, & Geurts, 2015). When following the causal modelling approach, the cause is seen as the root of the symptoms and the cause would be treated if possible. However, according to the network approach, symptoms are more important to treat than the cause (Borsboom & Cramer, 2013).

The large overlap in symptoms that we see in our population can also be explained by the network analysis approach. As is explained in this approach, symptoms can activate other related symptoms. In our population, for example, this could mean that a person shows some type of motor stereotyped behaviour. The occurrence of this behaviour in turn activates related behaviours, such as another type of stereotyped behaviour, for instance the repetitive use of objects. Together, these symptoms can make a clinician believe ASD is present, when in fact, the symptoms could be the expression of something else. However, our number of participants and our dataset about their symptoms is too small to perform a network analysis and figure out how symptoms interplay and unravel the overlap.

9.5 Diagnosis of ASD and multiple disabilities revisited

This thesis showed that diagnosing ASD in persons with combined sensory and intellectual disabilities is very complex. No instruments were specifically developed for this population, and already available instruments developed for other populations consisted of many unsuitable items and are thus invalid. Additionally, our studies showed that there is a lot of behavioural overlap between ASD, multiple disabilities, attachment problems and stress, making it difficult to decide what the underlying cause is to each of those behaviours.

Perhaps the categorical diagnoses that are described in the DSM (American Psychiatric Association, 2013) did not deal with the complexity and individual variation that is present in clinical practice (Fried et al., 2017). In the population of individuals with sensory and intellectual disabilities, finding the right diagnosis and then finding appropriate treatment are extremely complex processes. The diagnostic process can go wrong in many aspects due to lack of appropriate instruments and due to overlap in symptoms. This thesis helped to partly unravel the complex overlap between ASD and sensory and intellectual disabilities on the one hand, and mental health problems, on the other hand.

The results of this thesis taught us a number of things. The first part of this thesis showed that despite a behavioural overlap between individuals with sensory and intellectual disabilities and ASD, a distinction can be made between persons with and without ASD. To do so, one needs to focus on the specific behaviours that differ between the two groups. For example, we know that in persons with multiple disabilities, both with and without ASD, impairments in social communication are present. However, we now know that individuals with multiple disabilities without ASD can show social interest, but they may show it in a different way: without making eye contact but by using physical contact, for instance. OASID can be used to disentangle this overlap. It can help clinicians in making the distinction between characteristics of persons with multiple disabilities and characteristics of ASD.

In the second part of this thesis, we focused on mental health problems that could potentially explain the symptoms found in people with multiple disabilities. We found a lot of overlap between in behaviour stemming from stress, attachment, and mood disorders. Our research showed evidence that persons with multiple disabilities often showed signs of an attachment disorder. Also, it showed that stereotyped or repetitive behaviours are strongly related to stress, irrespective of the presence of ASD. As such, stereotyped behaviours alone are not a distinguishing characteristic of ASD in this population. It is important for clinicians to be aware of the high prevalence of stereotyped behaviours, and to always screen for attachment and stress related problems as an alternative for the diagnosis of ASD. This is especially so when a person shows more signs that one of these other disorders is present. For mood disorders, we found no evidence of a high prevalence within this population. In our study population, ASD and mood disorders are clearly two distinct disorders that can be diagnosed independent of each other. ASD related behaviours are not necessarily related to present mood disorders, making it easier to distinguish between the two. When a person shows ASD typical behaviours, clinicians should always consider the presence of stress and should include screening for attachment problems in their diagnostic assessment. Since mood disorders are more clearly distinct from ASD, they should only be further tested for when additional symptoms are present.

A summary of our findings can be found **in** Figure 1. This figure illustrates that both sensory and intellectual disabilities lead to specific behaviours. When these behaviours occur together, one might assume an ASD is present. However, some of these behaviours can be indications of an insecure attachment style or stress. This is less likely the case for mood disorders. When it comes down to interpreting the behaviour patterns, OASID can help in this process. With the use of OASID, clinicians can more easily decide if ASD is present or not.

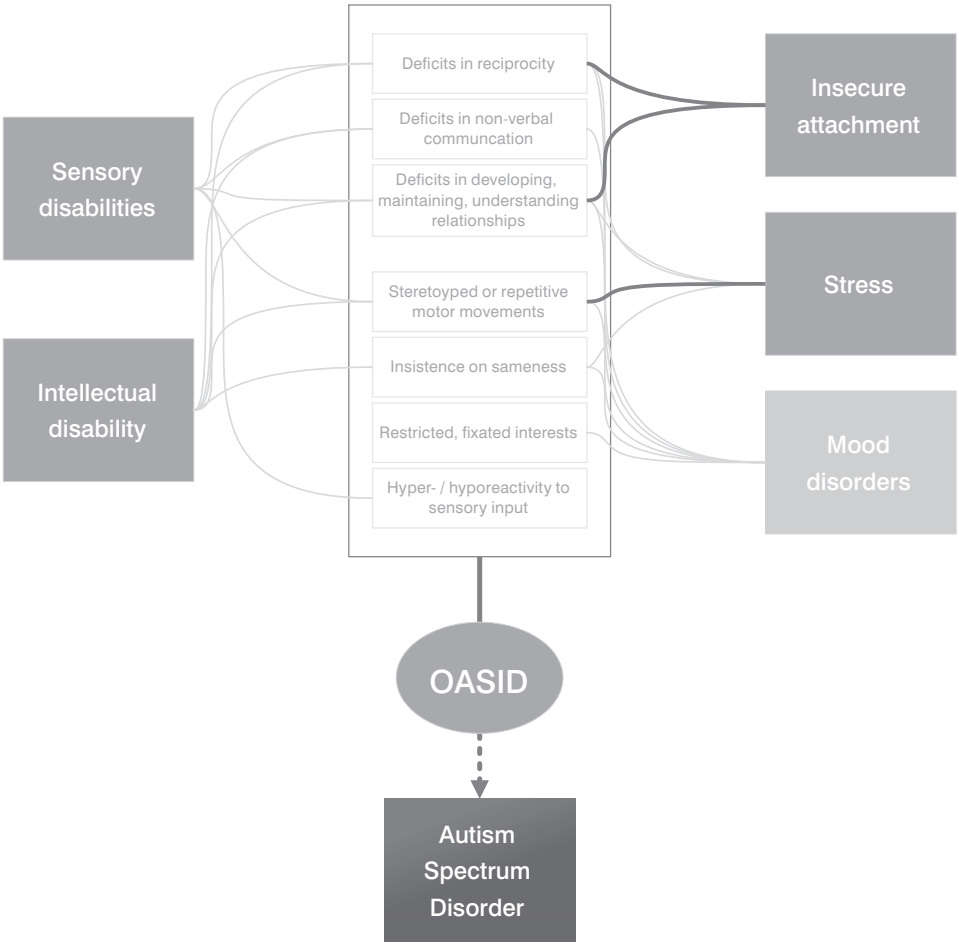

Figure 1 Behavioural overlap between intellectual and sensory disabilities and ASD, insecure attachment, stress and mood disorders.

9.6 Limitations and future perspectives

Of course the present study has some limitations. First, in general, our sample of 80 participants was rather small for the purpose of developing an instrument and determining reference points and norms. However, given the low incidence of combined intellectual and sensory disabilities, the size of this can be seen as reasonable. Nevertheless, the sample was too small to create separate norms for distinct groups based on, for example, level of intellectual disability, severity and presence of sensory disabilities, or age category. This would be an interesting step to take in future research and in perspective of the further development of OASID.

A second limitation of this study was that the OASID video material not only served as the basis for the OASID measurement, but also for the expert judgments used in Chapters 4 and 5 and the CARS2 scores from Chapter 5. Since these scores were all based on the same material, they may not be entirely independent from each other. For the expert judgments it would have been better if they had had the chance to see each participant face to face and assess the presence of ASD to their own insight. Similarly, the CARS2 should have been done on another play session, independent of OASID. The decision to base multiple measurements on the same video material was made after consultation of clinicians and caregivers of our target population. The main reason was not to burden the participants and their caregivers with too many assessments and tests. We believe the amount of possible contamination between the three assessments is minimalized because the scoring was done by independent raters who were unaware of the items and outcomes of questionnaires that the other raters filled in.

Finally, our scores of ASD behaviours were either based on questionnaires filled in by caregivers, or based on OASID video material. Because of limited communicative abilities of our participants, they were unable to answer questions or fill in questionnaires themselves so caregiver questionnaires were the most suitable alternative. The downside of these kind of indirect measurements is that the answers are based on the subjective interpretations of caregivers that filled in the questionnaires and not on the response of participants themselves. The other scores were based on OASID video material which lasted for about 20 to 60 minutes for each participant. The observations of stereotyped behaviour from Chapter 6 lasted only 10 minutes each. Our methods were a good starting point and successful in the sense that participants were not burdened with excessive testing. However, for a full behavioural assessment and to ensure a complete representation of a person's behavioural repertoire, we would recommend including elaborate behavioural observations in addition to questionnaires and assessments.

9.7 Clinical implications

Our study showed that care is needed when using assessment instruments in people with sensory and intellectual disabilities. Not only is there a lot of overlap in behaviour between different disabilities and disorders, we also found that current instruments regarding ASD related symptoms are not designed for this population. They contain biases, especially on the sensory domain, making many items inappropriate and invalid. Because there are still not many reliable and valid instruments available, it may not always be possible to completely refrain from using these instruments. Instead, we recommend to be aware of this bias and not to compare persons with multiple disabilities to norms for typically developing individuals.

OASID was developed to help in differentiating ASD symptoms from behavioural characteristics of individuals with sensory and intellectual disabilities. Our studies showed promising results regarding its psychometric properties and ability to assess the severity of ASD symptoms within this population. Although more research is required to further specify norms and to confirm its psychometric properties, a first step was made with the results from this thesis. In clinical practice, OASID can be used to assess the presence and severity of ASD related symptoms. However, for a full diagnostic evaluation a multimethod approach is still recommended, which would include making a differential diagnosis and assessing cognition, communication and adaptive skills of an individual.

Hopefully, the results of our study make clinicians aware that the behavioural repertoire of individuals with sensory and intellectual disabilities is often very complex. They should not only test for the most obvious or common disorders, but always check for alternative explanations for pathological behaviours. Different behaviours can be caused by a variety of underlying causes, disabilities or disorders which complicates the diagnostic process. There is an overlap in symptoms, not only between sensory disabilities, intellectual disabilities and ASD, but with other mental health problems as well. Both under- or over-diagnosis of ASD, attachment disorder, stress or anxiety are possible because these disorders share several symptoms. The current thesis is a starting point in disentangling this complex overlap. Similar behaviours can be caused by a variety of causes and in the current population it appears that an insecure attachment style and stress are important factors. It is important for clinicians to be aware of behavioural overlap and use elaborate assessments to study behaviour so that they can ensure the correct treatment for each individual.

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Nederlandse Samenvatting

Nederlandse samenvatting

Autisme spectrum stoornis (ASS) is een ontwikkelingsstoornis die wordt gekenmerkt door beperkingen op twee gebieden, namelijk: (1) Beperkingingen in sociale communicatie en sociale interactie en (2) beperkte en repetitieve gedragspatronen. De beperkingen in sociale communicatie en interactie zijn te herkennen aan beperkingen in sociaal-emotionele wederkerigheid, problemen in het gebruik van non-verbale communicatie en moeite in het ontwikkelen en behouden van relaties. De beperkte en repetitieve gedragspatronen worden onder andere gekenmerkt door motorisch stereotype of repetitief gedrag, vasthouden aan routines en niet goed kunnen omgaan met verandering, beperkte interesses en hyper- of hyporeactiviteit op zintuiglijke stimuli (American Psychiatric Association, 2013). De prevalentie van ASS is hoger bij mensen met een verstandelijke beperking, een visuele beperking of mensen met een combinatie van deze beperkingen dan bij mensen zonder een beperking (Brown, Hobson, Lee, & Stevenson, 1997; Carvill, 2001; Matson & Shoemaker, 2009). Echter, ongeacht de aanwezigheid van ASS, worden er kenmerken van ASS gezien bij mensen met verstandelijke beperkingen, visuele beperkingen, auditieve beperkingen of een combinatie hiervan (Brown, et al., 1997; Hoevenaars-van den Boom, Antonissen, Knoors, & Vervloed, 2009; Knoors & Vervloed, 2011; Matson & Shoemaker, 2009).

Een aantal gedragskenmerken komt dus zowel voor bij mensen met ASS als bij mensen met een combinatie van verstandelijke en zintuiglijke beperkingen (meervoudige beperkingen). Deze overlap in gedrag kan het moeilijk maken om ASS vast te stellen bij deze doelgroep. Sommige mensen met meervoudige beperkingen zullen onterecht gediagnosticeerd worden met ASS, terwijl hun gedrag eigenlijk veroorzaakt wordt door hun zintuigelijke of verstandelijke beperkingen (Andrews & Wyver, 2005; Cass, 1998). Andersom is het door de overlap in gedrag ook mogelijk dat de ASS wordt gemist omdat gedragskenmerken onterecht worden toegeschreven aan de aanwezige meervoudige beperkingen (Carvill, 2001).

Naast de overlappende gedragskenmerken speelt er een tweede probleem in de diagnostiek van ASS bij mensen met meervoudige beperkingen: het ontbreken van goede instrumenten. Wanneer er gedrag gezien wordt dat doet denken aan ASS, begint de diagnostische cyclus met het screenen van de persoon op ASS kenmerken. Daarna zal een meer uitgebreid diagnostische beoordeling plaatsvinden. Een groot deel hiervan gebeurt met instrumenten voor screening en diagnostiek van ASS (Nederlandse Vereniging voor Psychiatrie, 2009). Helaas zijn er geen instrumenten beschikbaar die specifiek gericht zijn op het vaststellen van ASS bij mensen met meevoudige beperkingen. Ook dit maakt de diagnostiek van ASS bij deze doelgroep lastig. Clinici nemen vaak de diagnose als uitgangspunt voor het bepalen van de behandeling (Howlin, 2000; Kraijer & Plas, 2006). In dat geval is het dus erg belangrijk om de juiste diagnose te stellen om zo ook de juiste behandeling te garanderen.

Het onderzoek beschreven in dit proefschrift richtte zich op het verbeteren van de dianogstiek van ASS en het beoordelen van gedrag van mensen met meervoudige beperkingen. Dit is op twee manieren gedaan en in deel 1 en deel 2 van dit proefschrift beschreven. Allereerst is er een instrument ontwikkeld waarmee ASS vastgesteld kan worden bij de de beschreven doelgroep. Daarnaast is er gekeken naar de gedragskenmerken van mensen met ASS en veelvoorkomende problemen in psychische gesteldheid.

De groep waar dit onderzoek zich op richtte bestond uit mensen met een combinatie van een verstandelijke beperking en een visuele beperking. De inclusiecriteria van dit onderzoek waren:

- Een leeftijd tussen de 6 en 60 jaar oud;
- Een matig, ernstig of diepe verstandelijke beperking (IQ $<$ 50), volgens de criteria van de AAIDD (2013) en WHO (2016);
- Een visuele beperking uit categorie 1-5, volgens de richtlijnen van de WHO (2016), of doofblindheid: een visuele beperking in combinatie met minimaal 35 debibel gehoorverlies, volgens de definitie van doofblind.nl (2017);
- De persoon heeft geen ernstige motorische beperkingen in de armen of volledige verlamming.

Overlappende kenmerken

Voordat er een instrument ontwikkeld kon worden om ASS vast te stellen moesten de overlappende en onderscheidende kenmerken van ASS en meervoudige beperkingen in kaart gebracht worden. Dit is gebeurd in deel 1 van dit proefschrift. **Hoofdstuk 2** liet zien dat veel gedrag van mensen met meervoudige beperkingen ook geïnterpreteerd zou kunnen worden als een kenmerk van ASS. Dit gedrag wordt dan niet veroorzaakt door ASS maar is puur een gevolg van de aanwezige beperkingen. Op het eerste domein van ASS, beperkingen in sociaal gedrag en sociale communicatie, is overlap te zien op het onderdeel wederkerigheid. Een voor de hand liggend voorbeeld is het maken van oogcontact en het reiken naar een ouder wanneer deze in beeld komt. Blinde kinderen zijn hier uiteraard niet toe in staat vanwege hun visuele beperkingen (Fraiberg, 1977), maar dit kan de indruk wekken dat het kind beperkingen heeft in het tonen van wederkerigheid in sociale interacties. Dit kan dan gezien worden als een kenmerk van ASS, terwijl het duidelijk wordt veroorzaakt door de visuele beperking. Dezelfde overlap is meer of minder duidelijk ook zichtbaar: in het begrijpen van non-verbaal gedrag (Gense & Gense, 2005), het laten zien van gedeelde aandacht (Osterling, Dawson & Munson, 2002) en de wijze van het voeren van gesprekken (Tager-Flusberg, Paul & Lord, 2005).

Ook op het gebied van stereotype en repetitief gedrag wordt overlap gezien tussen gedrag van mensen met ASS en met meervoudige beperkingen. Zo laten

zowel personen met ASS als personen met een meervoudige beperking stereotype gedrag zien met objecten of hun eigen lichaam (Gense & Gense, 2005; Tröster & Brambring, 1994; Warren, 1994). Er zijn echter wel verschillen tussen de gedragingen van personen met een meervoudige beperking met en zonder ASS. Hoevenaars-van den Boom et al. (2009) lieten zien dat personen met een meervoudige beperking zonder ASS meer open staan voor sociaal contact en meer plezier ervaren in dit contact dan mensen met een meervoudige beperking in combinatie met ASS. Deze eerste groep laat daarnaast meer initiatieven zien in het leggen van contact. Opgeteld is de grootste overlap in gedrag te vinden op het gebied van stereotype en reptitief gedrag, en minder op het gebied van sociale interacties.

Instrumenten voor screening en diagnostiek

Er is een breed aanbod van instrumenten die geschikt zijn voor het screenen van ASS-typisch gedrag en het diagnosticeren van ASS. De meeste van deze beschikbare instrumenten zijn echter specifiek gericht op de diagnostiek van mensen met een normale intelligentie en mensen zonder zintuiglijke beperkingen. Vanwege de grote overlap in gedragskenmerken bij mensen met een meervoudige beperking met en zonder ASS kunnen niet alle bestaande instrumenten gebruikt worden. Deel 1 van het proefschrift gaat verder in **Hoofdstuk 3** met het beschrijven van verschillende instrumenten om ASS te onderzoeken en hun geschiktheid voor mensen met meervoudige beperkingen. In totaal werden 13 screenings- en diagnostische instrumenten onderzocht en 7 instrumenten die onder andere kenmerken van ASS meten.

Alle instrumenten zijn geschikt om ASS vast te stellen bij de specifieke doelgroepen. Dit bleek onder andere uit adequate scores voor betrouwbaarheid en validiteit. Enkel de ABC (Krug, 1987) heeft normen voor mensen die doofdblind zijn. Geen enkel instrument was volledig afgestemd op de doelgroep van mensen met een gecombineerde verstandelijke en visuele beperking. De individuele items van alle instrumenten zijn ook onderzocht op geschiktheid voor de doelgroep. Hieruit bleek dat van alle instrumenten minimaal een kwart van de items ongeschikt was voor mensen met meervoudige beperkingen. Dit kwam omdat het item niet was af te nemen, onzinnig was om af te nemen of heel goed om andere redenen dan ASS vertoond zou kunnen worden. Items waren voornamelijk ongeschikt vanwege problemen op het zintuiglijke domein. De items waren niet aangepast aan de zintuiglijke beperken. Een veelvoorkomend voorbeeld is: “maakt de persoon oogcontact”, om te meten of iemand sociaal contact maakt. Dit overzicht in hoofdstuk 3 laat zien dat de bestaande instrumenten in hun huidige vorm niet bruikbaar zijn voor de doelgroep van dit onderzoek. Aanpassingen aan de testitems zijn noodzakelijk om het instrument bruikbaar te maken. Omdat er geen enkel volledig geschikt instrument beschikbaar was om ASS te diagnosticeren bij mensen met een meervoudige beperking is een nieuw instrument nodig wat dit wel mogelijk maakt.

De ontwikkeling van OASID

De diagnostiek van ASS bij mensen met een gecombineerde verstandelijke en zintuiglijke beperking is moeilijk. Dit wordt veroorzaakt door twee eerder genoemde problemen, namelijk een grote overlap in gedragskenmerken en een gebrek aan valide diagnostische instrumenten. Om die redenen was het logisch een nieuw instrument te ontwikkelen. Hoevenaars- van den Boom en collega's (2009) zijn hiermee gestart door de O-ADB te ontwikkelen (Observation of Autism in Deafblindness). De items van de O-ADB waren specifiek ontwikkeld voor mensen met een diepe verstandelijke beperking en doofblindheid. De O-ADB bleek inderdaad in staat te zijn om ASS vast te stellen bij deze doelgroep. De beperkingen van dit instrument waren echter de lange tijdsduur nodig voor de afname, en dat sommige mensen enige stress ondervonden bij de afname. Bovendien waren de items alleen geschikt voor mensen met een diepe verstandelijke beperking met doofblindheid. Een nieuw instrument zou dus makkelijker moeten zijn om af te nemen, geschikt moeten zijn voor een bredere groep mensen dan alleen mensen met doofblindheid en een diepe verstandelijke beperking, en bij voorkeur geen stressreacties opwekken tijdens de afname. Met dit doel is het instrument 'Observation of Autism in people with Sensory and Intellectual Disabilities' (OASID) ontwikkeld. De ontwikkeling van OASID en de pilot studie ervan staan beschreven in **Hoofdstuk 4**. Items van OASID zijn gebaseerd op de literatuur, bestaande instrumenten, de DSM-5, observaties en advies van experts op het gebied van meervoudige beperkingen. Uiteindelijk heeft dit geleid tot een semi-gestructureerd observatie-instrument. Met behulp van OASID neemt een onderzoeker spelenderwijs vijf taken af bij de deelnemers. De taken en de manier van afname worden ter plekke afgestemd op de mogelijkheden van de deelnemer, bijvoorbeeld in communicatie of de complexiteit van het spel. Deze testafname duurt tussen de 20 en 60 minuten en wordt opgenomen op video. Achteraf gebruikt de onderzoeker het videomateriaal om het gedrag van de deelnemer te scoren. Dit doet hij met behulp van 40 vragen verdeeld over de vijf taken. Gedrag wordt gescoord op een 3-punts schaal. In totaal waren er 21 items om sociaal gedrag en communicatie te onderzoeken en 19 items om repetetief en stereotype gedrag te onderzoeken. Een totaal score op beide schalen kon worden berekend door de score op individuele items bij elkaar op te tellen. Hoe hoger de score, des te meer het gedrag duidde op de aanwezigheid van ASS.

De pilot studie uit **Hoofdstuk 4** bestond uit een testafname van OASID bij 18 deelnemers. Als gouden standaard voor de aanwezigheid van ASS werden de oordelen van twee experts gebruikt. Dit onderzoek liet zien dat OASID een goede interbeoordelaarsbetrouwbaarheid had, een goede interne consistentie van schalen en een goede constructvaliditeit. Ook liet dit onderzoek zien dat de scores op OASID sterk samenhingen met de expertoordelen. Er was een duidelijk onderscheid te maken tussen deelnemers met en zonder ASS.

Deel 1 van het proefschrift wordt afgesloten met **Hoofdstuk 5** waarin een uitbereiding staat van de pilot studie. In dit onderzoek werd OASID afgenomen op 60 deelnemers. Dit onderzoek bevestigde de eerdere bevindingen over de goede interbeoordelaarsbetrouwbaarheid. Daarnaast had OASID een goede intrabeoordelaarsbetrouwbaarheid en een goede interne consistentie. De criteriumvaliditeit werd bepaald door te onderzoeken in welke mate OASID vergelijkbare resultaten laat zien als instrumenten die hetzelfde psychologische construct beogen te meten. Dit bleek ook goed in vergelijking met twee andere ASS instrumenten en de expertoordelen. In **Hoofdstuk 5** zijn ook cut-off scores bepaald voor OASID. Omdat de DSM-5 (American Psychiatric Association, 2013) aangeeft dat beide domeinen van beperkingen aanwezig moeten zijn om van ASS te kunnen spreken, is er gekozen voor een aparte cut-off op beide domeinen. Daarnaast leiden de scores niet enkel tot de aanwezigheid of afwezigheid van ASS, maar wordt de ernst ook meegenomen om zo recht te doen aan het spectrum van autistische gedragingen. Het eindresultaat op OASID kan leiden tot een interpratie van 'geen ASS symptomen', 'milde ASS symptomen', 'ernstige ASS symptoimen' en 'zeer ernstige ASS symptomen'.

Gedrag en geestelijke gezondheid

Het tweede deel van dit proefschrift is gericht op het in kaart brengen van de psychische gesteldheid, het gedrag en eventueel daarbij optredende pathologie bij mensen met meervoudige beperkingen die al dan niet gedrag vertonen dat bij ASS hoort. In dit tweede deel is OASID gebruikt om het onderscheid te maken tussen personen met en zonder ASS. **Hoofdstuk 6** beschrijft in brede zin de psychische gesteldheid en gedragsproblemen. Mensen met meervoudige beperkingen vertonen een grotere gevoeligheid voor stress (Bloeming-Wolbrink et al., 2012), laten vaker een onveilige hechting zien (Janssen, Schuengel & Stolk, 2002) en zijn gevoeliger voor depressie en stemmingsstoornissen (Hurley, 2006). Binnen deze studie is onderzocht hoe dit gedrag naar voren komt bij de doelgroep van mensen met meervoudige beperkingen en in hoeverre deze gedragingen pathologisch waren. Ook is onderzocht wat de verschillen in gedrag zijn tussen mensen met en zonder ASS binnen deze doelgroep. Het onderzoek liet zien dat een groot aantal personen waarschijnlijk onveilig gehecht was. We konden geen stemmingsstoornissen en pathologische reacties op de meeste stresssituaties vinden. Wel liet bijna de helft van de deelnemers een verhoogde score zien op zintuiglijke en persoonlijke stress, zoals aangeraakt worden. Dit kan mogelijk gerelateerd zijn aan de zintuiglijke beperkingen. Er zijn ook verschillen gevonden tussen personen met en zonder ASS. Personen met ASS lieten meer sociale ontwijking en meer manisch en hyperactief gedrag zien dan personen zonder ASS. Andersom lieten personen zonder ASS meer stress zien in onprettige situaties, situaties die voor personen met ASS blijkbaar als minder stressvol werden ervaren.

In **Hoofdstuk 7** is stereotype en repetief gedrag onderzocht, van stereotype gedrag met het eigen lichaam tot dezelfde handeling herhalen met een object. Het voorkomen van stereotype gedrag is één van de criteria voor ASS, maar tegelijkertijd komt stereotype en repetitief gedrag ook vaak voor bij mensen met zowel verstandelijke als zintuiglijke beperkingen (Murdoch, 2007). Binnen deze studie werd het voorkomen van stereotype en repetitief gedrag geobseerveerd bij mensen met een verstandelijke en zintuiglijke beperking. Het type, het aantal gedragingen en de duur van het gedrag werden meegenomen. De resultaten lieten zien dat alle deelnemers met ASS inderdaad stereotype en repetitief gedrag lieten zien, echter 85% van de deelnemers zonder ASS lieten ook stereotype gedrag zien. Hiermee kan het voorkomen van stereotype op zich geen onderscheidend criterium vormen voor de diagnose van ASS. Verder onderzoek liet zien dat mensen met ASS meer verschillende typen stereotype gedrag lieten zien in het geobserveerde fragment dan mensen zonder ASS. Daarnaast was de duur van het stereotype gedrag in het algemeen langer dan bij mensen zonder ASS. Tot slot liet een clusteranalyse zien dat er drie groepen te onderscheiden waren binnen de participanten. Er was een groep deelnemers die volledig bestond uit mensen met ASS, zij lieten een lange duur en veel zelfverwondend stereotype gedrag zien. Beide overige groepen participanten bestonden ongeveer voor de helft uit mensen met ASS, waarbij één groep een een lange duur en veel motorisch stereotype gedrag liet zien en de andere groep maar weinig stereotype gedrag. Dit bevestigt het eerdere vermoeden dat stereotype gedrag op zich geen onderscheid kan maken tussen mensen met en zonder ASS en dat er waarschijnlijk nog een andere factor dan ASS aanwezig moet zijn die het stereotype gedrag zou kunnen verklaren.

Een mogelijke verklarende factor voor het voorkomen van stereotype gedrag bij mensen met meervoudige beperkingen zou een pathologische reactie op stress kunnen zijn. Eerder onderzoek liet al zien dat stress of angst factoren kunnen zijn die stereotype gedrag veroorzaken (Leekam, Prior & Uljarevic, 2011; Rodgers, Glod, Connolly & McConachie, 2012). Deze relatie is verder onderzocht in **Hoofdstuk 8**. Door middel van de aanwezigheid van het stresshormoon cortisol in mondspeeksel is de mate van stress onderzocht bij personen met en zonder ASS, op een dag dat OASID is afgenomen en op een normale dag. Allereerst bleek dat de cortisolwaardes niet verschilden tussen de afnamedag en de normale dag. Dit impliceert dat OASID niet stressvol is in afname. Daarnaast is gevonden dat de cortisolwaardes niet verschilden tussen personen met en zonder ASS. De aanwezigheid van ASS leidt dus niet tot meer stressreacties. Tot slot werd gevonden dat de cortisolwaardes wel gerelateerd waren aan het tonen van reptitief en stereotype gedrag tijdens de afname van OASID. Dit bevestigt het eerdere vermoeden dat stress mogelijk een factor is die gerelateerd is aan het tonen van stereotype en repetitief gedrag.

Conclusies en klinische implicaties

De onderzoeken beschreven in dit proefschrift laten zien dat veel instrumenten ongeschikt zijn voor gebruik in personen met gecombineerde zintuiglijke en verstandelijke beperkingen. Het zal niet altijd mogelijk zijn om volledig van het gebruik van deze instrumenten af te zien, maar het is wel belangrijk dat clinici op de hoogte zijn van deze beperkingen. Om de diagnostiek van ASS bij deze doelgroep te vergemakkelijken is OASID ontwikkeld. OASID laat veelbelovende resultaten zien en de betrouwbaarheid en validiteit lijken goed te zijn. Meer onderzoek naar OASID is wel noodzakelijk om met name de cut-off scores specifieker af te stemmen op verschillende doelgroepen binnen de populatie. Ondanks de veelbelovende resultaten raden we aan om voor een volledig diagnostisch onderzoek altijd nog gebruik te maken van meerdere diagnostische hulpmiddelen, waaronder ook gesprekken, observaties, differentieële diagnostiek en onderzoek naar cognitieve en communicatieve vaardigheden.

Het gedragsrepertoire van deze doelgroep is complex en ASS symptomen duiden niet altijd enkel op ASS. Om stereotype gedrag te verklaren is het daarom belangrijk eerst te onderzoeken of dit gedrag niet veroorzaakt wordt door een andere factor, zoals stress of een onveilige hechting. Gegeven de uitkomsten van dit onderzoek is het belangrijk bij deze doelgroep altijd onderzoek te doen naar hechtingsproblemen, voordat er conclusies getrokken worden over de diagnose ASS. Om elk persoon van de goede behandeling te voorzien is het belangrijk dat er een uitgebreid en nauwkeurig diagnostisch onderzoek wordt gedaan waarin wordt gekeken naar het gehele gedragsrepertoire en rekening wordt gehouden dat het gedrag niet alleen past bij ASS maar ook het gevolg kan zijn van hechtingsproblemen, de verstandelijke of zintuiglijke beperking of een reactie is op een stresssituatie.

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jouw nuchtere en kritische blik is vaak een eyeopener (ook wanneer ik dit niet wil toegeven). Je hebt zelfs één van de figuren in dit proefschrift gemaakt! Pap en mam, ook jullie wil ik bedanken voor alle hulp die ik van jullie heb gekregen. En nee, daarmee bedoel ik niet alleen het strijken, poetsen en klussen in huis. Pap, je hebt altijd een onwijs grote interesse getoond in alles waar ik mee bezig was. Je hebt me erg geholpen door je luisterend oor als ik het moeilijk had en door allerlei klusjes uit handen te nemen wanneer ik het te druk had. Mama, jij staat letterlijk altijd voor me klaar en begrijpt me beter dan wie dan ook, waarschijnlijk omdat ik zo op je lijk. Ik heb heel veel van je geleerd en ben ongelofelijk blij met hoe goed we met elkaar kunnen praten.

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Curriculum Vitae

Curriculum Vitae

Gitta de Vaan is geboren op 21 mei 1989 in 's-Hertogenbosch en is opgegroeid in Vlijmen. In 2007 behaalde zij haar vwo diploma aan het d'Oultremontcollege te Drunen. Na haar middelbare school is Gitta begonnen aan haar bachelor psychologie aan de Universiteit Maastricht. De bachelor werd gevolgd door de master 'Health and Social Psychology' aan dezelfde universiteit. Ze is in 2011 afgestudeerd op een onderzoek naar het eetgedrag van klanten van een bakkerij. Tijdens haar studie heeft Gitta als studentassistent werkgroepen begeleid van eerste en tweedejaars studenten psychologie op het gebied van klinische psychologie, cognitieve psychologie en communicatievaardigheden.

Omdat Gitta zowel geïnteresseerd was in het doen van onderzoek als in het geven van onderwijs is ze snel na haar master gestart aan haar promotieonderzoek aan de Radboud Universiteit. Hier heeft ze zich gericht op het onderzoeken van kenmerken van autisme spectrum stoornis bij mensen met een meervoudige beperking. Gitta heeft de resultaten van haar onderzoek gepubliceerd in internationale wetenschappelijke tijdschriften en gepresenteerd op nationale en internationale congressen. In 2015 won ze met haar presentatie op het jaarlijkse congres van de American Association of Intellectual and Developmental Disabilties een 'certificate of acedemic excellence' met bijbehorende beurs. Naast haar onderzoek heeft Gitta onderwijs verzorgd aan studenten van de opleiding 'PedagogischeWetenschappen en Onderwijskunde', over beroepsvaardigheden, leren en ontwikkeling, en heeft ze scripties begeleid van bachelor- en masterstudenten.

Haar passie voor het geven van onderwijs heeft Gitta voortgezet. Sinds 2016 is ze werkzaam bij de opleiding Informatica binnen de Academie voor Engineering en ICT van Avans Hogeschool. Hier geeft ze met veel plezier les in onderzoeksvaardigheden, professionele vaardigheden en communicatieve vaardigheden.

Publicatielijst

Publicatielijst

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