Communication problems in children with autism and intellectual disability: Depicting the phenotype

Jarymke Maljaars

Printed by: Ipskamp Drukkers BV, the Netherlands (www.ipskampdrukkers.nl)
ISBN/EAN: 978-94-6191-294-7

Communication problems in children with autism and intellectual disability: Depicting the phenotype

PROEFSCHRIFT

ter verkrijging van
de graad van Doctor aan de Universiteit Leiden,
op gezag van Rector Magnificus prof.mr. P.F. van der Heijden,
volgens besluit van het College voor Promoties
te verdedigen op dinsdag 26 juni 2012
klokke 11.15 uur

door

Janna Pieternella Wilhelmina Maljaars

geboren te Middelburg
in 1982

Promotiecommissie

Promotores:
Prof. dr. I. A. van Berckelaer-Onnes (Universiteit Leiden)
Prof. dr. E. M. Scholte (Universiteit Leiden)

Co-promotor:
Prof. dr. I. L. J. Noens (Katholieke Universiteit Leuven, België)

Overige leden:
Prof. dr. L. J. M. van Balkom (Radboud Universiteit Nijmegen)
Prof. dr. M. J. Jongmans (Universiteit Utrecht)
Prof. dr. H. Roeyers (Universiteit Gent, België)
Prof. dr. J. T. Swaab (Universiteit Leiden)

Table of contents

1

General introduction

General introduction

Children with autism spectrum disorder (ASD) and co-occurring intellectual disability (ID) are distinctively different from children with ASD or ID alone. The co-occurrence of ASD and ID is known to cause severe problems for the child, the parents, and other caregivers and is a strong predictor of poor prognosis (Boucher et al., 2008). There is a need for a better understanding of the challenges and impairments which characterize the co-occurrence of these disorders (Matson & Shoemaker, 2009). Communication problems are one of the core symptoms of ASD. Children with ASD and co-occurring ID form a particularly vulnerable group regarding communication problems. Severe communication problems evidently influence the child's development and the child-rearing situation. Impairments in communication can lead to challenging behavior (Bott et al., 1997; Chamberlain et al., 1993; Sigafoos, 2000), and place a heavy burden on parents (Bebko et al., 1987). Valid and reliable diagnostic instruments and knowledge about strengths and weaknesses in communicative behavior are important to improve assessment and intervention for these children and their families.

Autistic disorder and intellectual disability

Autism spectrum disorders (ASDs) are lifelong, complex neurodevelopmental disorders, encompassing a wide spectrum of developmental disorders characterized by impairments in three behavioral domains: social interaction, communication, and range of interests and activities. In this thesis, the term 'autism' will be used to refer to autistic disorder (AD) specifically, whereas the abbreviation ASD will be used to refer to the broader spectrum of autistic disorder, Asperger syndrome, and Pervasive Developmental Disorder Not Otherwise Specified (PDD-NOS or atypical autism). The terms autism and autistic disorder are used interchangeably depending on journal policies. ASD corresponds to pervasive developmental disorder (PDD) as described in the Diagnostic and Statistical Manual of Mental Disorders, fourth edition – text revision (DSM-IV-TR; APA, 2000) and the International Classification of Diseases, tenth revision (ICD-10; WHO, 1992). Most of the literature used in this dissertation is based on the broader ASD, but given the classification difficulties in low-functioning children, the present study focused on AD, the core syndrome instead of the whole spectrum in order to create a more homogeneous group. According to the DSM-IV-TR (APA, 2000) AD is characterized by severe and pervasive impairments in social interaction and communication skills, and by the presence of restricted and stereotyped patterns of behavior, interests, and activities with onset prior to the age of three years. Criteria for these impairments are based on both quantitative (absence of normative, developmental milestones) and qualitative differences (presence of aberrant behaviors) from typical development (Klin et al., 2005). Each of these core features have to be present in order to diagnose AD. However, the clinical manifestations and severity of

symptoms of AD are often very heterogeneous (Beglinger & Smith, 2001; Hill & Frith, 2003).

Estimates of the prevalence of ASDs have gradually increased over the past decades. The estimates for ASDs from recent studies have been considered to be just over 1% (Harrington, 2010; Lord, 2011), and the average prevalence for AD is approximately 0.2% (Fombonne, 2009). Furthermore, ASDs are four times more common in males than in females, although some recent studies are questioning this ratio (Gould & Ashton-Smith, 2011; Kopp & Gillberg, 2011). Although ASDs are considered to be among the most heritable developmental disorders, the underlying genetic factors remain largely unknown (Glaser et al., 2010; Noor et al., 2010). Recent studies have led to the identification of several autism susceptibility genes and assessment of copy number variation has discovered numerous potentially important novel candidate loci (Abrahams & Geschwind, 2008). Many of these genes and genomic loci have been associated with ID, which suggests that both developmental disorders share a common genetic etiology (Betancur, 2011; Noor et al., 2010).

Epidemiological studies report varying percentages for the rate at which ID is present in individuals with ASD, ranging from about 15% to 70%, depending on diagnostic criteria and level of functioning (Bhaumik et al., 2010). The highest prevalence rates of ID are found in the more strictly defined group with AD (Charman et al., 2011). Furthermore, increased severity of ID is positively correlated with incidence of AD (Vig & Jedrysek, 1999). Conversely, about 3 to 50% of individuals with ID have ASD (De Bildt et al., 2004b). ASDs are one of the most frequently co-occurring disorders in individuals with ID (Matson & Shoemaker, 2009). ID is defined by significant limitations in intellectual functioning and adaptive functioning (two or more standard deviations below the population mean) (APA, 2000; Schalock et al., 2010). Particularly for research purposes, classification of the level of severity of ID is often based on IQ-scores only. Mild ID is defined by an IQ range of 50-55 to approximately 70, moderate ID by 35-40 to 50-55, and severe to profound ID by an IQ below 35-40 (APA, 2000).

Making an accurate diagnosis of ASD in ID is difficult, especially in those with severe or profound ID (Bhaumik et al., 2010; Vig & Jedrysek, 1999). The main reason for this is the overlap in behavioral characteristics between children with ASD and children with ID. Especially, the criteria formulated as quantitative impairments, i.e. absence of normative, developmental milestones, are difficult to apply in low-functioning children. Delays in social and communicative development and absence of specific behaviors may also be explained by low levels of cognitive functioning (Bhaumik et al., 2010). This makes some diagnostic criteria from the DSM-IV-TR difficult to apply. Individuals with severe or profound ID, with ASD as well as without ASD, may have no or limited expressive language abilities. In these cases, diagnostic criteria concerning atypical use of language and impairments in conversations are not applicable. Since symbolic abilities typically emerge just around 18 to 24 months (Thiemann-Bourque et al., 2011), symbolic play can also be absent due to cognitive level of functioning. Furthermore,

stereotyped and repetitive behaviors can also be present in individuals with ID without ASD (Bodfish et al., 2000; Bradley et al., 2004). In addition, common medical comorbidities in the ID population, such as deaf blindness, epilepsy, or cerebral palsy, may obscure the clinical presentation of ASD symptoms, and can result in a limited behavioral repertoire and restricted possibilities to express themselves (Bhaumik et al., 2010). Nevertheless, there are also clear distinct symptoms present in individuals with ID and co-occurring ASD compared to individuals with ID without ASD, mainly in severity of stereotyped and repetitive behaviors and relative delay in social and communicative development regarding their general level of functioning (Matson et al., 2008). In the case of ID without ASD, social and communicative skills are usually corresponding with the child's overall developmental level (Klin et al., 2005). The combination of ASD and ID results in a mutual aggravation of the impairments involved in both disorders and has consequences for the well-being of the individuals themselves and their families (Boucher et al., 2008). A correct diagnosis is important to choose the most indicated intervention strategies. Individuals with ID and ASD need to be treated in a different way than individuals with ID without ASD (Matson & Shoemaker, 2009). Although ASD and ID do co-occur, research in ASD is generally focused on individuals with intelligence in the normal range. An IQ below 70 or even 85 is frequently used as an exclusion criterion for autism research. Therefore, relatively little is known about children with ASD and co-occurring ID.

Communication in low-functioning children with autistic disorder

Impairments in communication comprise an important dimension of the ASD phenotype and involve both verbal and nonverbal communication problems (Landa, 2007; Weismer et al., 2010). Communication and language skills are extremely variable in individuals with ASD (Kjelgaard & Tager-Flusberg, 2001; Thurm et al., 2007). A significant part of mainly low-functioning individuals with ASD do not develop language or only a limited number of communicative words or gestures (Boucher et al., 2007). Particularly in case of severe or profound ID, often no verbal or symbolic communication is present. These individuals communicate through idiosyncratic gestures, vocalizations, or other behaviors (Snell, 2002). When children with AD do not gain language by the age of 6, they frequently remain nonverbal (Prior & Ozonoff, 1998). The levels of language development and communicative competence acquired by children with ASD have been found to be important predictors of later outcome in terms of adaptive functioning, intelligence, academic achievement, and level of independence in adulthood (Lord et al., 2004). Several studies in individuals with co-occurring ID reported that receptive communication is overestimated by caregivers of low-functioning individuals, resulting in a too high level of approach (Barlett & Bunning, 1997; Bradshaw, 2001; Cascella, 2004; McConkey et al., 1999). Overestimation of communication skills has consequences for quality of life, learning potential, and level of independence (Barlett & Bunning, 1997; Bradshaw, 2002). Communication problems

in children with ASD and ID are severe, resulting in increased caretaker demands, and may lead to stress in parents and professionals (Lecavalier et al., 2006). In addition, a relation between communication problems and the presence of challenging behavior is evident. Previous studies showed that problem behaviors increase in frequency, intensity, and duration, when communication problems are more severe (Bott et al., 1997; Chung et al., 1995; Van Berckelaer-Onnes et al., 2002). Despite these disconcerting results, relatively little is known about communicative abilities and impairments in children with ASD and ID, in particular in children who are either nonverbal or minimally verbal (Tager-Flusberg et al., 2005).

Exploring patterns in communicative strengths and weaknesses can contribute to a better understanding of the nature of these problems in low-functioning children with ASD. Moreover, it may lead to recommendations for assessment, development of more effective interventions, and suggestions about which children may benefit from specific intervention approaches. However, most standardized assessment measures focus primarily on formal aspects of language, which can only provide information on a limited number of aspects of communication for children with ASD and therefore are not sufficient, especially for low-functioning children with ASD (Lord & McGee, 2001; Twachtman, 1995). Communication problems in ASDs are often examined in a narrow way by focusing on the level of expressive language and abnormalities in language use. There is a need for both qualitative and quantitative information on a broad range of communicative skills. When assessing communication in children with ASD, it is useful to combine standardized and norm-referenced instruments with more descriptive and qualitative observational procedures to be able to identify developmental discontinuities and provide complementary information about more subtle differences between children (Klin et al., 2005). Assessment of communication can be divided in three important aspects, namely communicative *functions*, *forms*, and *content*. By integrating information gathered from the assessment of competences and impairments in the use and / or understanding of function, form, and content of communication, it should be possible to outline a picture of overall communicative competence (Twachtman, 1995).

A communicative *function* refers to the intention, goal, or purpose communication is used for. With respect to functions of communication, this study is confined to the most basic functions, because of our focus on low-functioning children. These functions concern communicative acts for behavior regulation, social interaction, and joint attention. Usually, intentionality in communication develops during the first year of life and is an important precursor for language development (Travis & Sigman, 2001; Watt et al., 2006). Children with ASD have problems in the development of intentionality or pragmatics, but severity can differ from no intentional communication of any kind to problems in complex pragmatic functions, such as using sarcasm or the intention to deceive another person (Twachtman, 1995).

Several *forms* or means can be used for communication, of which language is the most common and conventional form. Language involves a complex communication system that is based on a set of rules relating symbols to referents (Twachtman-Cullen & Twachtman-Reilly, 2007). The most basic intentions, such as behavior regulation and joint attention, can be communicated by either non-symbolic or symbolic communication forms. Language or other symbolic communication systems are required to express more complex communicative functions (Coupe O'Kane & Goldbart, 1998). Children with ASD have been found to use fewer vocalizations (Tager-Flusberg et al., 2005) and develop language at a slower pace compared with typical development (Eigsti et al., 2011; Weismer et al., 2010). Furthermore, the use of gestures is also limited. They often use only primitive presymbolic motoric gestures, such as pulling or manipulating another's hand, and lack the use of more conventional gestures, such as waving, pointing and nodding one's head (Lord & McGee, 2001; Shumway & Wetherby, 2009). The absence of conventional communicative means often leads to the development of idiosyncratic or undesirable behaviors, such as automutilation and aggression, to communicate (Wetherby et al., 2000).

Content of communication refers to the meaning of words or other communicative forms that is conveyed. Noens and Van Berckelaer-Onnes (2005) conceptualized communication problems in ASDs as a problem in sense-making or the perception of meaning. Sense-making can take place at different levels: sensation, presentation, representation, and metarepresentation (see Chapter 5 for a more detailed explanation). The levels of sensation and presentation refer to non-symbolic levels of understanding of communication. At these non-symbolic levels, children do not comprehend that a word or a picture can refer to something else. At the levels of representation and metarepresentation, the symbolic or referential meaning of communicative forms is understood (Noens & Van Berckelaer-Onnes, 2005). Children with ASD have difficulty acquiring conventional and symbolic aspects of communication. The use and comprehension of symbols require an understanding of the relationship between a symbol and its referent. A symbol stands for and is separate from its referent (Bates, 1979). Symbols enable people to exchange information efficiently by representing something else even beyond the here and now (Namy, 2005). During the first few years of life most children achieve important milestones in symbolic development (DeLoache, 1995).

In summary, assessment of communication in children with ASD should determine the communicative functions a child is currently expressing as well as the means, i.e. the repertoire of behaviors and level of language, by which the child attempts to do so. It is also important to assess the level of language comprehension in order to provide information which parents and professionals can use to adjust their communication level towards the child with ASD. Furthermore, assessment of the level of symbolic development or sense-making is of critical importance in planning intervention when assessing low-functioning children with minimal or no language.

Objectives and overview of the dissertation

The main objective of the current thesis is to provide insight into the communication problems of children with AD and co-occurring ID in order to improve assessment and intervention strategies in this vulnerable group. The five chapters that follow aim to shed light on the function, form, and content of communication in children with AD and ID. Each chapter concerns a published manuscript, which can be read separately. Due to this, some overlap between each of the chapters is inevitable. The study in Chapter 6 is based on a different research group than the other chapters. Figure 1.1 presents an overview of the different components that were examined in the various studies.

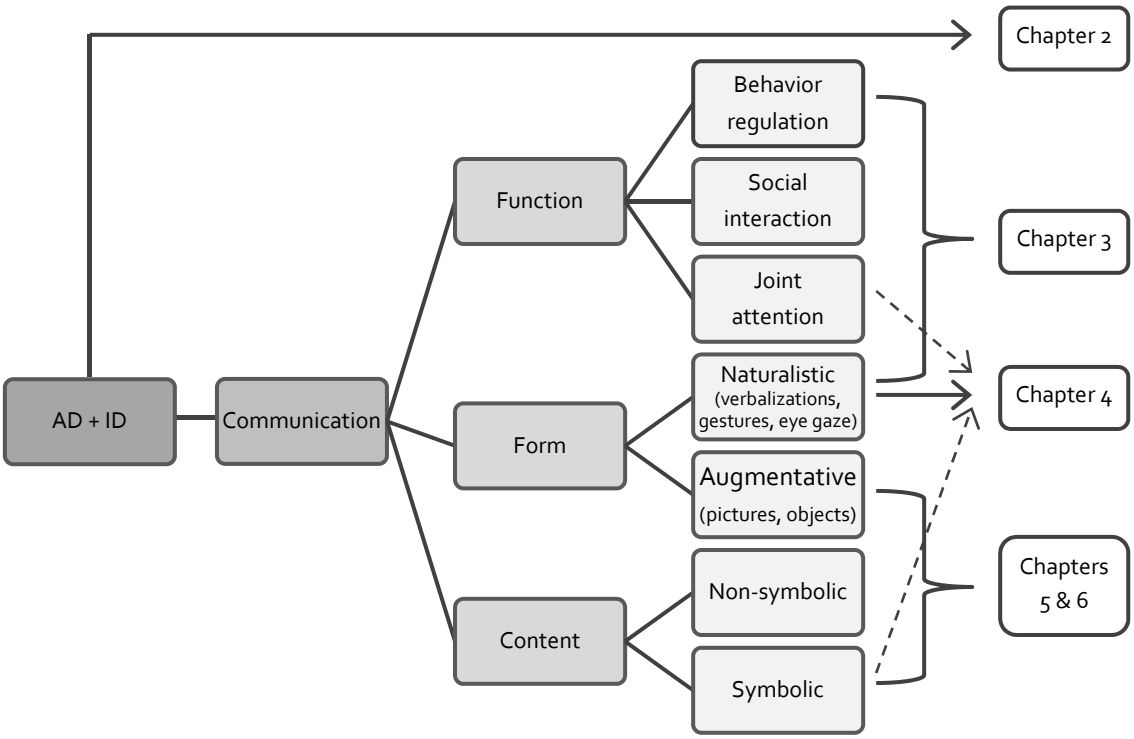


Figure 1.1 **Overview of different components of communication examined in the current study**

The figure can be elucidated as follows. Since a diagnosis of AD in ID is complicated, in Chapter 2 the validity of a semi-structured interview for the identification of ASD in children with ID was evaluated. The main focus of Chapter 3 is to examine functions and complexity of forms used to communicate in both verbal and nonverbal low-functioning children with AD. Chapter 4 extends the current knowledge on the most common form of communication, namely language in children with AD and ID. Particularly, attention is paid to the relation between expressive and receptive language and the role of joint attention and symbol formation in language development. In Chapters 5 and 6 sense-making of visual (and tactile) communication forms in low-functioning children or adults with AD is studied. The relation with adaptive behavior and autism symptomatology is examined in Chapter 5. Chapter 6

presents the findings of sense-making in specific subgroups of low-functioning individuals with AD. In this chapter, the influence of co-occurring deafness in individuals with AD and ID is explored. The discussion of Chapter 5 is mainly focused on clinical implications, while in Chapter 6 the theoretical implications are the focus of attention.

2

Validity of the DISCO in young and low-functioning children

Jarymke Maljaars
Ilse Noens
Evert Scholte
Ina van Berckelaer-Onnes

Autism (2011), doi: 10.1177/1362361311402857

Evaluation of the criterion and convergent validity of the Diagnostic Interview for Social and Communication Disorders in young and low-functioning children

Abstract

The Diagnostic Interview for Social and Communication Disorders (DISCO; Wing, 2006) is a standardized, semi-structured and interviewer-based schedule for diagnosis of autism spectrum disorder (ASD). The objective of this study was to evaluate the criterion and convergent validity of the DISCO-11 ICD-10 algorithm in young and low-functioning children. The DISCO-11, Autism Diagnostic Observation Schedule (ADOS), and Social Communication Questionnaire (SCQ) were administered to a Dutch sample of 115 children comprising 52 children with ASD (both with and without intellectual disability), 26 children with intellectual disability (non-ASD), and 37 typically developing children. Results indicated high sensitivity and specificity for DISCO-11 classifications in differentiating ASD from non-ASD according to the clinical classification in children with mild intellectual disability or average intelligence. Among children with a moderate or severe intellectual disability the sensitivity was equally high, but the specificity was significantly lower. The agreement between DISCO-11 and ADOS classifications was substantial, between DISCO-11 and SCQ moderate. The correlations between raw scores of the DISCO and ADOS algorithm or SCQ were both high. In conclusion, the DISCO-11 differentiates accurately between autistic disorder and non-ASD in young children with an average intelligence or mild intellectual disability, but is over inclusive in the lower levels of intellectual disability.

Introduction

A substantial proportion of individuals with autism spectrum disorder (ASD) also have an intellectual disability (ID). Reported percentages are variable (ranging from about 15% to 70%; Bhaumik et al., 2010), depending on diagnostic criteria and level of functioning. The diagnosis of ASD in ID can be complicated, especially in those with severe or profound ID (Bhaumik et al., 2010; Vig & Jedrysek, 1999). Nevertheless, diagnosis of ASD in ID is important in order to adjust treatment strategies and to provide access to specific services. A detailed interview with the parents about developmental history and current behavior is an essential part of the assessment procedure to diagnose ASD (Bishop et al., 2008; Filipek et al., 1999). Therefore, it is necessary to examine the applicability of standardized interviews in the ID population.

The Diagnostic Interview for Social and Communication Disorders – version 11 (DISCO-11) is a semi-structured interview to be administered with parents or other primary carers (Wing, 2006). The instrument can be used to assess individual needs and

to diagnose ASD and is suitable for individuals of all ages, levels of abilities, and the whole spectrum (for a more detailed description of the DISCO, see Wing et al., 2002). The DISCO is constructed in a dimensional rather than a categorical way, as a bottom-up approach was used to compose the items. Based on the broad-ranging nature of ASDs, the DISCO items cover a very wide range of skills and features. Originally, the DISCO was designed for clinical use to provide a comprehensive description of individual characteristics and needs, but the interview can also be used for classification in clinical practice or for research. For this purpose, computerized algorithms have been developed. These algorithms provide classifications based on official classification systems, such as the ICD-10, and other specific diagnostic categories (Wing et al., 2002). The algorithms for ICD-10 Childhood Autism and Atypical Autism are based on a selection of 93 DISCO items and were used in this study.

Thus far, the psychometric properties of the DISCO (versions 9 and 10) have only been tested in the United Kingdom and Sweden. With regard to the reliability, the inter-rater reliability proved to be moderate to good or excellent for the algorithm items (Leekam et al., 2002; Nygren et al., 2009), but also for the other items (Wing et al., 2002). As for validity, good agreement across clinical diagnoses and the DISCO algorithms for ICD-10 (κ = .57) and Wing and Gould ASD (κ = .79) has been demonstrated (Leekam et al., 2002). Nygren et al. (2009) also found a good agreement (89%) between the DISCO and ADI-R classification based on 57 interviews. Furthermore, age, non-verbal IQ, language production and comprehension were not significantly related to the DISCO classifications (Leekam et al., 2002).

As far as we know, studies into the psychometric properties of the DISCO are limited to the three aforementioned studies. No reports have focused specifically on young and low-functioning children. Since the DISCO was designed for any age and any level of ability (Wing et al., 2002), the DISCO may be useful for autism diagnoses in ID. Therefore, the aim of this study was to evaluate the criterion and convergent validity of the DISCO ICD-10 algorithms in young and low-functioning children. To this end, first the sensitivity and specificity of the DISCO ICD-10 classifications were explored, and second, the DISCO results were compared to the ADOS and SCQ outcomes.

Method

Participants

The sample for this study comprised three different subgroups: children with ASD, children with ID, and typically developing children (TD) (see Table 2.1). All children were less than twelve years old and had a developmental age below six years. The ASD group (n = 52) mainly consisted of boys and the majority had an associated ID. All children with ASD had received an independent clinical diagnosis by qualified professionals

before participation in this study (see Procedure). These were mainly children with a clinical classification of autistic disorder.

Table 2.1 ***Main characteristics of the participants***

	ASD (*n* = 52)			**ID (non-ASD)** (*n* = 26)			**TD** (*n* = 37)		
	Range	*M*	*SD*	Range	*M*	*SD*	Range	*M*	*SD*
Chronological age [a]	34 - 137	79.6	28.9	48 - 134	80.9	20.7	24 - 49	37.9	6.3
Nonverbal mental age [a]	<24 [b] - 70	43.0	14.2	<24 [b] - 63	38.2	12.5	29 - 65	42.5	9.2
Receptive language [a]	<14 [b] - 65	33.9	15.8	<14 [b] - 70	37.3	13.8	22 - 75	44.3	11.1
Expressive language [a]	<18 [b] - 75	37.6	16.4	<18 [b] - 53	34.7	11.9	22 - 72	43.6	9.5
	n	%		*n*	%		*n*	%	
Gender									
Male	43	83%		16	61%		15	41%	
Female	9	17%		10	39%		22	59%	
Level of ID									
No ID	11	15%		0	-		37	100%	
Borderline ID	6	23%		0	-		0	-	
Mild ID	15	29%		11	42%		0	-	
Moderate ID	12	12%		12	46%		0	-	
Severe ID	8	21%		3	12%		0	-	

Note. ASD = autism spectrum disorder; ID = intellectual disability; TD = typical development
[a] age / age equivalent in months; [b] minimum score

The ID group consisted of 26 children. These were children with either known causes of ID, such as Down syndrome, or unknown causes, but all without ASD. The level of ID ranged from mild to severe. About half of the children had associated conditions, such as ADHD or language disorder. The TD group contained 37 children between 2 and 4 years old.

Instruments

Diagnostic Interview for Social and Communication Disorders. The DISCO-11 can be administered to collect information about developmental history and description of a broad range of skills and behaviors (Wing, 2006; Wing et al., 2002). The Dutch translation of the 11th version of the interview was used in this study (Van Berckelaer-Onnes et al., 2008). The interview takes about two to three hours to administer and consists of more than 300 questions grouped under different sections: 'Family, medical background and identifying information', 'Infancy', 'Developmental skills', 'Repetitive, stereotyped activities', 'Emotions', 'Maladaptive behavior', and 'Interviewer's judgment of quality'. The sections concerning psychiatric illnesses, difficulties related to sexual behavior, and forensic problems were not used in this study, because they were not relevant for young and low-functioning children. The majority of the items are scored in three levels of severity: 'marked problem', 'minor problem', or 'no problem'. Furthermore, the DISCO distinguishes both 'ever' and 'current' ratings of the child's behavior. In this way, a distinction is made between current symptoms and behavior that has ever been present. For this study, the ICD-10 classifications Childhood Autism

and Atypical Autism were collapsed together to compose an ASD or non-ASD classification. Furthermore, only 'current' classifications were used, because no differences were found between current and ever results.

Autism Diagnostic Observation Schedule. The Autism Diagnostic Observation Schedule (ADOS) is a semi-structured, standardized assessment in which standardized toys and activities are used to provide opportunities to elicit social, communicative, play, and stereotyped behaviors relevant to the diagnosis of ASD (Lord et al., 1999). Each child was administered activities from module 1 or 2 based on the expressive language level by officially trained examiners. The revised algorithms and cut-offs were used to provide ASD and non-ASD classifications (Gotham et al., 2007). Inter-rater and test-retest reliability, as well as validity, have been demonstrated (e.g. Gotham et al., 2007, 2008; Lord et al., 2000). Validity of the ADOS was also examined in the ID population (De Bildt et al., 2004a).

Social Communication Questionnaire. The Social Communication Questionnaire (SCQ; Rutter et al., 2003; Dutch translation: Warreyn et al., 2004) is a parent-report questionnaire with 40 binary-scaled items to screen for ASD based on the Autism Diagnostic Interview – Revised (ADI-R; Lord et al., 1994). The recommended cut-off score for ASD is 15 (Rutter et al., 2003), and proved to be accurate in the ID population (Witwer & Lecavalier, 2007). The SCQ is available in two forms: lifetime and current. In this study the 'current' version which evaluates the child's behavior over the most recent 3 months was used. This version is also suitable for children under the age of 5 years. The SCQ has been found reliable (Rutter et al., 2003) and valid by several studies (e.g. Berument et al., 1999; Chandler et al., 2007). Reliability and validity of the Dutch translation have been tested in Belgium (Warreyn et al., 2004).

Other instruments. To measure nonverbal mental age a Dutch test for nonverbal intelligence, the SON-R 2½-7 (Tellegen et al., 1998) was used. Language abilities were individually assessed using the Reynell test for Dutch language comprehension (Van Eldik et al., 1995) and the Schlichting test for Dutch language production (Schlichting et al., 1995). Raw scores were converted into age equivalents. All three instruments have been found reliable and valid by the authors and the Committee for Psychometric Evaluation of Tests of the Dutch Institute of Psychologists (Evers et al., 2009).

Procedure

Families of children with ASD and/or ID were contacted through 23 special schools and day care centers. The professionals involved were asked to include children between 2 and 12 years old with an official ASD diagnosis or children without ASD, and to exclude children without official diagnosis. Severe sensory or severe motor impairments were exclusion criteria for all groups, because several items from the DISCO, ADOS, and SCQ

are difficult to code in these cases. TD children were recruited from four regular day care centers. All regular and special schools and day care centers were randomly selected from schools and day care centers situated in three different regions of the Netherlands (Zuid-Holland, Noord-Holland, and Utrecht). All ASD children were clinically diagnosed based on DSM-IV-TR criteria for pervasive developmental disorders prior to recruitment for the study by independent and qualified professionals. The DISCO was not used and none of the researchers of the current study were involved in the diagnostic procedure. The time gap between previous diagnosis and participation in this study varied between one and seven years. Parents were asked to send a copy of the diagnostic report from the child's clinical record. Informed consent was obtained from all parents. The original sample consisted of 129 cases, from which 14 children were excluded, because the clinical diagnosis of the child was unclear or the DISCO could not be administered due to refusal or insufficient Dutch language abilities of the parents. Eventually, 115 parents and children participated in this study. Comparisons to the ADOS were based on 109 cases and comparisons to the SCQ on 106 cases.

The DISCO interviews were conducted by 7 different interview couples at the parents' home. The interviewers were trained to use the DISCO specifically for the purpose of the study. In most of the cases they did not see the child before or during the interview. The interviewers and raters were blind to the previous diagnosis before the DISCO interview. The ADOS was administered at the university or at school in presence of one of the parents. The study was designed so that different researchers administered the interview and the ADOS. The parents filled out the Social Communication Questionnaire at home. The SON-R 2½-7, Reynell, and Schlichting were administered at the child's school or day care.

Data analyses

First, raw scores of the three subgroups on total and domain scores of the ICD-10 algorithm were compared to examine whether the DISCO differentiates between ASD and non-ASD cases. The domains Social Impairment, Communicative Impairment, and Repetitive Behavior and Activities are based on 38, 15, and 29 items, respectively. The raw scores were calculated by summing the number of recorded symptoms for each domain. The rating code that is demanded in the algorithm in order to count as positive for the relevant diagnostic criterion was used. Most of the DISCO items are coded as severe (0), minor (1), or no problem (2). To meet the criteria for a symptom in specific domain, a 'severe' code is needed in most of the cases. Differences in mean scores between the three groups were evaluated by conducting several analyses of variance followed by post hoc tests. The Games-Howell procedure was used, because Levene's tests reported unequal variances between the groups. Variances in the ASD group were higher than in the ID and TD groups. Second, sensitivity and specificity rates, positive and negative predictive values were computed for the DISCO, ADOS, and SCQ compared to the clinical diagnoses. Third, agreement between the different autism

instruments was examined by computing Kappas on the classification outcomes and correlations on the raw scores.

Results

DISCO ICD-10 algorithm scores

The mean raw scores on the different subcategories of the DISCO ICD-10 algorithm for the three different subgroups are presented in Table 2.2.

Table 2.2 ***Description and comparisons of raw DISCO ICD-10 algorithms scores for the subgroups***

	ASD (*n* = 52)		**ID** (*n* = 26)		**TD** (*n* = 37)		**ANOVA**			**Games-Howell**
DISCO [a]	*M*	*SD*	*M*	*SD*	*M*	*SD*	*F*	*p*	η^2	post-hoc differences
SOC	17.9	6.0	6.4	4.0	2.4	1.4	139.8	< .001	.71	ASD > ID > TD
COM	6.0	2.3	1.7	1.5	0.2	0.5	134.3	< .001	.71	ASD > ID > TD
RRB	6.0	3.8	1.6	2.2	0.2	0.8	52.3	< .001	.48	ASD > ID > TD
Total	29.9	9.5	9.7	6.0	2.8	1.9	172.9	< .001	.76	ASD > ID > TD

Note. SOC = social impairment; COM = communication impairment; RRB = restricted repetitive behaviors / activities
[a] scores were based on number of recorded symptoms for each domain

Significant differences were found between all three subgroups on the different domains and the total algorithm score (p < .001, large effect sizes). Post-hoc test revealed that for all domains the ASD group obtained significantly higher mean raw scores compared to both the ID and TD groups. Furthermore, the ID group scored significantly higher on all domains than the TD group (see Table 2.2).

Sensitivity and specificity

Sensitivity and specificity rates of the DISCO and the other autism instruments were computed (see Tables 2.3 and 2.4).

Table 2.3 ***Sensitivity, specificity, PPV and NPV for the DISCO, ADOS, and SCQ***

Instruments	Sample	*n*	Sens	95% CI	Spec	95% CI	PPV	NPV
DISCO	Total sample	115	.96	.86 - .99	.79	.67 - .88	.79	.96
(ICD-10 algorithm)	ASD+TD	89	.96	.86 - .99	1.00	.88 - 1.00	1.00	.95
	ASD+ID	78	.96	.86 - .99	.50	.29 - .68	.79	.87
ADOS	Total sample	109	.92	.81 - .98	.95	.84 - .99	.94	.93
(cut-off: ASD)	ASD+TD	85	.92	.81 - .98	.97	.82 - 1.00	.94	.93
	ASD+ID	76	.92	.81 - .98	.92	.72 - .99	.96	.85
SCQ	Total sample	106	.67	.51 - .79	.93	.82 - .98	.89	.78
(cut-off: 15)	ASD+TD	84	.67	.51 - .79	1.00	.87 - 1.00	1.00	.69
	ASD+ID	70	.67	.51 - .79	.82	.59 - .94	.89	.53

Note. Sens = sensitivity; Spec = specificity; PPV = positive predictive value; NPV = negative predictive value

For the DISCO, a sensitivity of .96 and a specificity of .79 were found. The specificity of the DISCO in relation to the ID group was much lower than in relation to

the TD group. The sensitivity of the ADOS was comparable to the DISCO, but the SCQ led to much more false negative classifications and therefore to a lower sensitivity. The specificity for both the ADOS and SCQ was higher than for the DISCO, although 95% confidence intervals of the three tests overlap to some extent.

When the DISCO classifications ASD or non-ASD were compared to the clinical diagnosis, the majority of the children were correctly classified as having ASD (n = 50) or no ASD (n = 50). Thirteen false positive cases and two false negative cases were identified. All of the 13 false positive cases had ID, thus no TD children were present in this category.

Table 2.4 ***Cross tab comparing DISCO-11 classifications to clinical diagnoses***

				Clinical dx ASD		
		n		Absent	Present	Totals
DISCO-11	Total sample	115	Test negative	50	2	52
(ICD-10 algorithm)			Test positive	13	50	63
			Totals	63	52	115
	ASD+TD	89	Test negative	37	2	39
			Test positive	0	50	50
			Totals	37	52	89
	ASD+ID	78	Test negative	13	2	15
			Test positive	13	50	63
			Totals	26	52	78

When the false positives (n = 13) and true negatives (n = 13) in the ID group were compared, no significant differences in gender ($\chi^2_{(1)}$ = 0.01, p = .99), age (U = 72.0, p = .55), language abilities (U = 38.0, p = .09), associated conditions ($\chi^2_{(1)}$ = 2.5, p = .12), or interviewers ($\chi^2_{(4)}$ = 3.7, p = .45) were observed. However, these analyses were based on very small sample sizes. The most obvious difference was seen in severity of ID ($\chi^2_{(1)}$ = 12.8, p < .001). The false positive group mainly consisted of children with a severe or moderate level of ID (92%), while most of the children in the true negative group had only a mild ID (77%).

Comparison to ADOS and SCQ

Kappas were computed as a measure of agreement between classifications on the three autism instruments. The agreement between DISCO and ADOS classifications was substantial (κ = .69, p < .001). However, in comparison to the SCQ a much lower Kappa and only a moderate agreement was found (κ = .49, p < .001). The SCQ had more false negatives and fewer false positives, in other words the opposite pattern of the DISCO classifications, which explains the lower Kappa. The Kappas for the ID group were much lower than for the TD group for both instruments.

The correlations between raw total scores of the DISCO algorithm with the ADOS ($r_{(107)}$ = .87, p < .001) and SCQ ($r_{(104)}$ = .86, p < .001) were both high. Regarding the domains, the correlation between DISCO and ADOS social / communication domain

scores ($r_{(107)}$ = .87, p < .001) was much higher than between the restricted / repetitive behavior domain scores ($r_{(107)}$ = .64, p < .001), but were both significant.

Discussion

The aim of the current study was to evaluate the criterion and convergent validity of the DISCO-11 ICD-10 algorithms in young and low-functioning children. Overall, the sensitivity rate was high, the specificity rate was acceptable, and the agreement with the ADOS and SCQ was sufficient. The results support the utility of the DISCO as an effective diagnostic tool for ASD in children with average intelligence or mild ID, since the results of the current study indicated a good criterion-related and convergent validity. However, among the individuals with moderate to severe levels of ID, the specificity was significantly lower than among individuals with a mild ID to an average level of functioning. In other words, the DISCO accurately identifies the low-functioning children with ASD, but is over inclusive. The DISCO classifications were more accurate at a mild level of ID. These findings were in line with previous studies (Leekam et al., 2002; Posserud et al., 2009).

The agreement between DISCO-11 and ADOS classifications was substantial. Convergent validity was examined by calculating the relationship between the raw scores on the DISCO-11 and ADOS. In particular, the social and communication domain scores were strongly related. The correlation between restricted and repetitive behavior domain scores was weaker, but still satisfactory. The ADOS only contains three items in the restricted and repetitive behavior domain, whereas the DISCO score is based on 29 items and covers a broader range of repetitive behaviors and activities. The correlation between the raw scores on the DISCO and SCQ was comparable to the correlation with the ADOS, whereas the agreement between the DISCO and SCQ classifications was much lower. Probably the cut-off score of the SCQ is not accurate for this young and low-functioning group. It is important to notice that the SCQ is primarily designed for children of 4 years and older, and most of the children in this study were younger or lower functioning. The study of Witwer and Lecavalier (2007) indicated a cut-off score of 15 as accurate for individuals with ID. However, recent studies in young children (<5 years) showed that maximum sensitivity and specificity rates were achieved when the cut-off score was reduced (e.g., Corsello et al., 2007; Snow & Lecavalier, 2008; Wiggins et al., 2007). The data of our study support the latter findings. Again, the results indicated a lower convergent validity of the DISCO-11 in individuals with ID compared to individuals with an average intelligence.

Severity of ID often has a disturbing influence on the proper classification of diagnostic instruments. Several other diagnostic and screening instruments for ASD were also found to be over inclusive in the severe levels of ID (e.g., De Bildt et al., 2003, 2004a; Gray et al., 2008; Witwer & Lecavalier, 2007), because it is difficult to determine

whether specific behaviors are associated with ID or ASD. Some features, such as absence of language development and imagination, can be a consequence of a delay or low level of functioning, instead of an impaired development as seen in ASD (Bhaumik et al., 2010). When using the DISCO for very low-functioning children, some of the above mentioned features were recorded as ASD symptoms, without taking the general level of functioning into account. Therefore, a thorough examination of the intellectual and adaptive level of functioning with reliable and valid instruments is always necessary. These results underline the necessity of a comprehensive assessment by experienced clinicians to diagnose ASD.

Some limitations of the current study have to be mentioned. First, the sample size was relatively modest, in particular when specific subsamples were examined. Second, this study was not primarily designed as a validation study. In most of the cases the interviewers did not see the child before or during the interview, but in some cases the child was present at home during the interview. The interviewers and raters were blind to the previous diagnosis before the DISCO interview. However, some parents referred to the diagnosis during the conversation, which may have biased the outcomes. Third, all children with ASD were previously clinically diagnosed; as a result, parents' answers might have been influenced by familiarity with the types of questions and the knowledge of their child's diagnosis. And last, the ID group possibly contains undiagnosed ASD cases.

Given the influence of severity of ID on the classification, a more specific examination of the ICD-10 algorithm in different levels of intellectual functioning would be important in further research. The data of our study suggest that there are differences in raw scores between ASD and non-ASD, also at the lower levels of ID. Therefore, the possibilities for adjusted criteria can be explored. Since the majority of the participants received a clinical diagnosis of autistic disorder, the accurateness of the DISCO ICD-10 algorithm for broader ASDs in ID still has to be evaluated. This study and previous studies mainly focused on children, and therefore the psychometric properties for adults with ID also need attention in further research. Furthermore, only the algorithm results were used, whereas research into the possibilities of the use of the DISCO in a dimensional way to assess severity of symptoms can also be valuable, since ASDs are more and more considered to form a spectrum with different levels of severity (APA, 2010; Lord & Bishop, 2010). In that respect, the DISCO is a useful addition to other autism specific clinical and research instruments, as the DISCO covers many more areas of development and behavior, also aspects of ASD which are not included in the official diagnostic criteria (Wing et al., 2002).

3

Intentional communication in low-functioning children with autism

Jarymke Maljaars
Rianne Jansen
Ilse Noens
Evert Scholte
Ina van Berckelaer-Onnes

Journal of Communication Disorders (2011), *44*, 601-614

Intentional communication in nonverbal and verbal low-functioning children with autism

Abstract

In this study we characterized profiles of communicative functions and forms of children with autism and intellectual disability (*n* = 26), as compared to typically developing children (*n* = 26) with a comparable nonverbal mental age (2-5 years). Videotapes of the Communication and Symbolic Behavior Scales – Developmental Profile were analyzed using a standardized observation scheme in which three main functions were distinguished: behavior regulation, social interaction, and joint attention. Different forms of communication were also investigated: gestures, vocalizations / verbalizations, and eye gaze. Results indicated that in typically developing children the proportion of communication for the purpose of joint attention was much higher than for behavior regulation, whereas in children with autism the opposite pattern was seen. Low-functioning nonverbal children with autism mainly communicated for behavior regulation and not or only rarely for declarative purposes. Generally, this subgroup used the least complex forms to communicate. Low-functioning verbal children with autism differed from typically developing children only in the rate, not in the proportion of communication for specific functions. Combinations of three different communicative forms were used by verbal children with autism less frequently than by typically developing children.

Introduction

Communication in low-functioning children with ASD

Impairments in communication are among the most important features of autism spectrum disorders (ASD), and involve both verbal and nonverbal communication (APA, 2000). However, communication skills are extremely heterogeneous in individuals with ASD, particularly in children with an associated intellectual disability (ID). A large proportion of low-functioning individuals with ASD does not develop language or acquires at most a few functional words or signs (Boucher et al., 2007). Exact percentages of nonverbal individuals with ASD are difficult to determine, because of increased recognition of ASD in higher functioning and verbal individuals (Lord et al., 2004). Since research mainly focused on toddlers with ASD or higher functioning individuals with ASD, relatively little is known about language abilities and communication in children with ASD and ID (Boucher et al., 2007; Tager-Flusberg et al., 2005). Therefore, the purpose of this study was to identify the functions and forms of communication in verbal and nonverbal children with autism (AD) and associated ID, so as to evaluate their strengths and weaknesses in intentional communication.

Functions of communication in ASD

Limited intentionality is considered to be a core deficit in the communication of individuals with ASD (Noens & Van Berckelaer-Onnes, 2005; Travis & Sigman, 2001; Wetherby et al., 2000). Usually, intentionality is first seen when children start to use proto-imperative and proto-declarative expressions between 9 and 11 months of age (Bates, 1979). By the end of the first year of life, typically developing children communicate for a large number of functions, which can be grouped into three main functions: (a) behavior regulation, (b) social interaction, and (c) joint attention[1] (Shumway & Wetherby, 2009). Hence, communicative intentionality for different functions develops before the emergence of language by use of gestural and vocal communicative acts (Wetherby, 1986).

Several studies found a significantly lower rate of intentional communication in children with ASD compared to typically developing (TD) children or children with developmental delays (DD) (e.g., Chiang et al., 2008; Shumway & Wetherby, 2009). So far, a few studies into communication profiles of children with ASD have been conducted, but differences with respect to child characteristics (e.g., chronological age, level of functioning, verbal ability), and research methods (e.g., instruments, situations, and criteria used to assess intentional communication) complicate comparability of these studies. The most common finding in systematic observation of communication profiles in young children with ASD is a reduced rate of communication for all different communicative functions compared to children with TD or DD (Shumway & Wetherby, 2009), with the exception of initiation of acts for behavior regulation. Some studies reported reduced behavior regulation compared to TD children (Adrien et al., 2001; Chiang et al., 2008; Wetherby et al., 2007), but not compared to children with DD or ID (Stone et al., 1997; Viellard et al., 2007).

Several studies analyzed the proportions of communicative functions, thus determining patterns of communicative functions independent of the frequency of communication. Studies in young children with ASD reported a propensity to communicate for behavior regulation rather than for joint attention (Adrien et al., 2001; Shumway & Wetherby, 2009; Stone et al., 1997; Viellard et al., 2007). However, Chiang and colleagues (2008) only observed a lower proportion of joint attention in ASD, but equal proportions for behavior regulation and social interaction compared to TD children. With respect to social interaction, the least stable outcomes were found. Some studies described the initiation of social interaction as a relative strength (e.g., Shumway & Wetherby, 2009), while other studies (e.g., Adrien et al., 2001) reported social interaction together with joint attention as striking weaknesses.

Typically developing children learn to use different functions simultaneously (Bruinsma et al., 2004), whereas children with ASD seem to develop the different

[1] The term 'joint attention' refers in this paper to a specific communicative function and therefore is used in a broader definition than usual in autism literature.

functions consecutively. In ASD, development of communicative intentionality appears to follow a sequence from behavior regulation and social interaction to joint attention (Calloway et al., 1999; Crais et al., 2004). They continue to make progress in the use of different communicative functions as they get older (Calloway et al., 1999; Travis & Sigman, 2001). Yet, most prior studies of communication profiles in ASD focused on very young children only. Consequently, less is known about the relationship between level of functioning, language abilities, and communication profiles in ASD.

Intentionality has a central role in the acquisition of language (Bloom & Tinker, 2001). In typical development, joint attention abilities in young children are significantly related to expressive language (Carpenter et al., 1998; Delinicolas & Young, 2007; Mundy & Gomes, 1998; Watt et al., 2006). Mundy and colleagues (1995) also reported a link between behavior regulation and social interaction on the one hand and expressive and receptive language abilities on the other. With respect to children with ASD, several studies found a predictive relationship between joint attention and expressive language development (Dawson et al., 2004; Drew et al., 2007; McDuffie et al., 2005). Studies examining several communicative functions found that behavior regulation and social interaction were not significantly related to language development in ASD (Mundy et al., 1990; Watt et al., 2006). The study of Stone and Caro-Martinez (1990) demonstrated a wider range of communicative functions in verbal children with autism compared to nonverbal children with autism. According to the study by Wetherby and colleagues (2007), acts for behavior regulation and joint attention in the second year of life were significantly related to nonverbal intelligence at three years of age. Most of the other studies did not find a correlation between nonverbal intelligence and communicative functions in ASD (Charman et al., 2003; Shumway & Wetherby, 2009). All these studies focused on relatively young children with ASD and not specifically on children with associated ID.

Forms of communication in ASD

Communicative intentions can be expressed in several ways, differing in quality from very primitive forms (such as pulling one's hand) to more conventional forms (such as spoken language). Forms can be analyzed by focusing on quantity, diversity, or complexity. Previous studies revealed that in children with ASD eye gaze was seen less frequently and less integrated with other communicative forms (Stone et al., 1997). Children with ASD also used less pointing than TD children (Shumway & Wetherby, 2009; Stone et al., 1997), especially for joint attention purposes (Camaioni et al., 2003; Goodhart & Baron-Cohen, 1993). Children with ASD often compensate their reduced use of gestures and eye gaze with more primitive or unconventional forms, such as manipulations of hands or body (Freeman et al., 2002; Shumway & Wetherby, 2009; Stone et al., 1997). With respect to diversity, children with ASD showed a decreased variety in type of gestures (Colgan et al., 2006). In general, results on quantity and diversity of communicative forms are unambiguous, whereas the possible differences in

complexity seem to be less clear. Complexity refers to the degree by which communicative gestures are accompanied by eye gaze and/or vocalizing. Shumway and Wetherby (2009) reported no differences in combinations of forms used for behavior regulation, social interaction, and joint attention in young children with ASD when compared to TD children (aged between 18 and 24 months). However, a substantial variance within the ASD group was observed for the different levels of complexity. Therefore, they recommended further research into possible changes across the first years of life. Another study (Stone et al., 1997) reported significant differences in level of complexity between children with DD or language impairment and children with ASD. The latter of the two groups used a significantly lower proportion of acts involving a combination of gestures, vocalizations, and eye gaze, although no differentiation was made for the specific communicative functions.

Implications of communication problems in ASD

Together with the restricted use of (non)verbal symbolic communication forms, the limited communicative intentionality in ASD leads to reduced possibilities to control, understand, and participate in the social world. These communication impairments can result in challenging behavior, in particular in low-functioning nonverbal individuals (Chiang, 2008; McClintock et al., 2003; Sigafoos, 2000), and are a major stressor for parents (Bebko et al., 1987; Konstantareas & Homatidis, 1989; Lecavalier et al., 2006). In addition, the development of communication and language predicts better long-term outcomes (Mahwood et al., 2000), and improvement in communicative skills is closely related to reduction of challenging behavior (Goldstein, 2002; Van Berckelaer-Onnes et al., 2002). Therefore, research into the communication abilities of low-functioning children with ASD is essential. Research into low-functioning individuals with ASD is more complex, as severe communication problems make it harder to use standardized instruments and result in limitations in testability (Eagle, 2002). By using direct observation with a precise measure to describe and quantify communicative functions and forms, important information on the communicative weaknesses, but also on the communicative strengths can be provided. This will improve our understanding of the communication profiles of low-functioning children with ASD. For both verbal and nonverbal low-functioning children with ASD, it is important to characterize intentional communication skills in rate and quality, so as to provide important intervention targets to enhance intentional communication (Bruinsma et al., 2004; Iacono et al., 1998).

Purpose of the study

The main purpose of the current study was to explore the profile of intentional communication in low-functioning verbal and nonverbal children with autism (AD) compared to typically developing (TD) children matched on nonverbal mental age. Two important aspects of intentional communication were taken into account; first, the functions or purposes underlying the communicative efforts of the children, and

second, the complexity of forms of the communication they use. The aims of this study were: (a) to assess the differences in rate and proportion of communicative functions between low-functioning children with AD (both as a whole and divided into verbal and nonverbal subgroups) and TD children, (b) to investigate the relationship between nonverbal mental age, language abilities, and proportion of use of these communicative functions in the AD and TD groups, and (c) to examine differences in complexity of communicative forms used for the different communicative functions between verbal and nonverbal children with AD and ID compared to TD children. Based on the studies in toddlers with ASD, we expect a different communication profile in children with AD and ID compared to TD children with a tendency to communicate for behavior regulation rather than social interaction or joint attention. Furthermore, lower levels of complexity are expected in the AD and ID group, since children with ASD show difficulties in integrating different communicative forms. A relation between language and communication functions, especially joint attention, may be expected. Therefore, it is important to distinguish communication profiles regarding function and complexity of forms for verbal and nonverbal children with AD.

Method

Participants

Fifty-two children were included in this study. The main characteristics of the participants are presented in Table 3.1.

Table 3.1 ***Main characteristics of the participants***

	AD+ID group			**AD+ID subgroups**				**TD group**		
	Total (n = 26)			Nonverbal (n = 13)		Verbal (n = 13)		Total (n = 26)		
	Range	*M*	*SD*	*M*	*SD*	*M*	*SD*	Range	*M*	*SD*
Chronological age [a]	37 - 136	85.2	30.2	84.1	33.8	86.2	27.6	24 - 47	35.4	5.9
Nonverbal mental age [a]	26 - 68	39.7	11.1	34.7	5.5	44.8	13.1	29 - 65	40.8	10.0
Receptive language [a]	<14 [b] - 53	28.3	12.1	18.6	7.4	38.0	6.9	27 - 75	43.8	11.4
Expressive language [a]	<18 [b] - 55	31.8	12.8	- [c]	- [c]	41.7	7.8	22 - 67	43.5	9.9

Note. AD = autistic disorder; ID = intellectual disability; TD = typical development
[a] age / age equivalent in months; [b] minimum score; [c] nearly all cases achieved an age equivalent nearby or below the minimum score

The total autism group (AD group) comprised 26 children with autism and intellectual disability, whom all had received a formal classification of autistic disorder according to DSM-IV-TR criteria (299.00; APA, 2000) prior to the study. Diagnosis was confirmed by an algorithm score above the threshold for autism on the *Autism Diagnostic Observation Schedule* Modules 1 or 2 (ADOS; Gotham et al., 2007), and a classification of autistic disorder on the *Diagnostic Interview for Social and Communication Disorders – version 11* (DISCO-11; Wing, 2006). Levels of ID ranged from

mild to severe (APA, 2000) based on a standardized intelligence test. Given the classification difficulties in low-functioning individuals, this study focused on the core syndrome instead of the whole spectrum to create a more homogeneous group. Participants (23 boys; 3 girls) were aged between 3;1 and 11;4 years (*M* = 7;1, *SD* = 2;6). The nonverbal mental age ranged from 26 to 68 months (*M* = 39.7, *SD* = 11.1). Children with severe physical disabilities were excluded, since standard criteria for the identification of intentionality in communication are difficult to apply in this population (Iacono et al., 1998).

Each participant in the AD group was individually matched with a TD child by nonverbal mental age within 3 months, measured with *the Snijders-Oomen Non-Verbal Intelligence Test - Revised 2½-7 years* (SON-R; Tellegen et al., 1998). Therefore, the groups did not differ in SON-R scores ($t_{(50)}$ = -.36, p = .72). The children in the TD group did not meet diagnostic criteria for autism spectrum disorder on the ADOS nor the DISCO. Age of the participants in the TD group (11 boys; 15 girls) ranged from 2;0 to 3;11 years (*M* = 2;11, *SD* = 0;6). The TD group differed from the AD group on age ($t_{(50)}$ = 8.24, p < .001) and sex ($\chi^2_{(1)}$ = 12.24, p < .001), but there was no difference in rate of intentional communication between boys and girls in the control group ($t_{(24)}$ = 1.2, p = .25). Concerning the different communicative functions, boys in the TD group communicated more often for joint attention than girls, ($t_{(24)}$ = 2.5, p = .02). The impact of chronological age on the findings is negligible, since no significant and only weak correlations were found between chronological age, total rate of intentional communication, and the use of the three different functions in the AD group (see also Table 3.4).

For some analyses the AD group was subdivided into verbal and nonverbal subgroups. The children in the nonverbal group (n = 13) used no verbalizations or only one-word utterances during the Behavior Sample. Those with verbal abilities (n = 13) mainly used two- or multiword utterances based on the observation of communicative forms during the CSBS-DP Behavior Sample. All children in the TD group had phrase speech, except for one of the youngest children. The characteristics of the subgroups are presented in Table 3.1.

The verbal AD group did not differ from the typical control group on language production ($t_{(37)}$ = -.59, p = .56) and language comprehension ($t_{(37)}$ = -1.67, p = .10). The three groups were not comparable with respect to nonverbal mental age ($F_{(2, 49)}$ = 3.37, p = .04). The nonverbal AD group was significantly lower functioning than the verbal AD group and the TD group (p = .05), but the verbal AD group did not differ significantly from the TD group (p = .60).

Instruments

Intentional communication. The *Communication and Symbolic Behavior Scales – Developmental Profile Behavior Sample* (CSBS-DP; Wetherby & Prizant, 2002) is a structured assessment that encourages spontaneous communicative behavior. Six situations, such as play with a wind-up toy, a balloon, bubbles, or a book, offer the opportunity for the child to communicate. Intentional communication ratings were collected from CSBS-DP videotapes by two trained observers. Communication was coded during all Behavior Sample activities with exception of the comprehension probes. Every communicative act was coded (consistent with Shumway & Wetherby, 2009) to allow more in-depth analyses. The standard CSBS-DP scoring is restricted to a maximum of three communicative acts per activity and therefore has less opportunity to discriminate between the groups. Moreover, the norms for the CSBS-DP only go through 24 months of age. Therefore, only the materials and testing procedure and not the standard scoring procedure of the CSBS-DP were used.

To examine the communicative profiles, videotapes of CSBS-DP Behavior Samples were analyzed using a standardized observation scheme (based on Wetherby & Prizant, 1993), making it applicable to a broader (developmental) age range. A communicative act was determined using Wetherby and Prizant's (1993) criteria for intentional communication. An intentional communicative act requires a gesture, vocalization, or verbalization, that is directed toward the adult and that serves a communicative function. Gestures are directed when it concerns a movement toward, or touching the other person, or when combined with eye gaze or vocalization / verbalization. A vocal / verbal act must be accompanied by a gesture or eye gaze. Eye gaze alone and undirected vocalizations / verbalizations are not considered as intentional communication. Furthermore, only intentional communicative acts which were spontaneously initiated by the child were coded. The following rules were used to determine the spontaneity of communication**:** (a) only communicative acts following natural cues or manipulations of natural cues (e.g., presence or movement of objects, comments, or gestures) were coded, (b) communicative acts following a question or a direct prompt, or imitations were not included, and (c) non-interactive communicative acts such as utterances used to direct one's own actions, labeling objects, or emotional responses to a situation not directed to an adult were also excluded.

After identifying a communicative act, the function and form(s) used for this act were determined. Three mutually exclusive, main categories of communicative functions were coded: *Behavior Regulation* (BR), *Social Interaction* (SI), and *Joint Attention* (JA), according to the definitions described by Wetherby and Prizant (1993) (see Table 3.2 for definitions and examples). Due to the broader age range and verbal language abilities of this study compared to the target group of the CSBS-DP the specific functions within the categories SI and JA were extended with functions often seen in older, more verbal children, like expressing feelings or sharing information

(based on Buckley, 2003; Calloway et al., 1999). A fourth category was used for functions which were unclear or did not fit into one of the three categories, for example unintelligible, incomplete, and incoherent sentences which did not fit in the context in the communicative situation or touching another person without clear communicative intent.

Table 3.2 ***Description of the main categories of communicative functions and forms (based on Wetherby & Prizant, 1993)***

Category		Description	Examples
Functions	Behavior Regulation	Acts used to regulate another's behavior to obtain a specific result	*- request object / action* *- protest object / action*
	Social Interaction	Acts used to gain or maintain another's attention to oneself	*- request social routine / comfort / permission* *- call / greet / show off* *- express feelings / interest*
	Joint Attention	Acts used to direct another's attention to an object, event, or topic	*- comment on object / action* *- request / provide information* *- share pleasure / feelings*
Forms	Gestures	Gestures	*- reach / touch point / distal point* *- instrumental / descriptive gestures* *- nod / shake head*
		Manipulations of persons / objects and movements of objects	*- manipulate person's hand/body* *- move object toward / away from person* *- give / show object*
	Vocalizations / Verbalizations	Use of vocal or verbal communication	*- laugh / cry* *- meaningless / meaningful transcribable vocalizations* *- verbalizations (single- / multiword spoken utterances)*
	Eye gaze	Gaze toward a person's face immediately before, during, or after the act	

Forms used during the communicative acts were coded as gestures, vocalizations / verbalizations, and/or eye gaze. The forms coded within the main categories of gestures and vocalizations are mutually exclusive. The (combination of) forms used for communicative acts were divided into three different levels of complexity. Level 1 is used for communicative acts with only one form (gesture), and thus represents the least complex acts. Level 2 reflects an intermediate level of complexity, involving a combination of two different communicative forms, a combination of a gesture with either eye gaze or a vocalization, or a vocalization with eye gaze. Level 3 concerns the most complex level of communicative acts, in that a combination of three forms is used by the child: gesture, vocalization, and eye gaze (based on Stone et al., 1997).

Coding was conducted by two of the authors (JM and RJ). To assess inter-rater reliability, the two raters independently double-coded 27% of all videotaped Behavior Samples. Both authors were aware of the aims of the study, but the second researcher was blind to the children's clinical diagnosis and scores on the other measures. Inter-rater reliability was calculated using percentage agreement and intra-class correlations. Agreement between the two coders was computed as a percentage of scores for which

the observers completely agree compared to the jointly observed communicative acts, across 14 randomly selected participants (7 of these were drawn from the AD group and 7 from the TD group). Percentage agreement was uniformly above 89% for identification of communicative acts (*M* = 96.2; 95% confidence interval: 94.0 - 98.5). Agreement for functions ranged from 80 to 92%, and for forms from 82 to 94%. The intra-class correlation coefficient for identification of communicative acts (rates per minute) was .96 (95% confidence interval: .68 - .99). The generalizability coefficients for rates / percentages of function categories were .70 / .82 for function unclear, .90 / .98 for BR, .92 / .92 for SI, and .99 / .97 for JA. The coefficients for the rates of form categories were .95 (gestures), .96 (eye gaze), and .99 (vocalization / verbalization). All agreement percentages and intra-class correlations, except for the rate of the unclear function category, exceed .80, indicating a good to excellent reliability. The unclear function category is left out of consideration in most of the analyses.

Nonverbal mental age and language abilities. To determine nonverbal mental age a Dutch test for nonverbal intelligence, the SON-R 2½-7 (Tellegen et al., 1998) was used. Language abilities were individually assessed using the *Reynell* test for Dutch language comprehension (Van Eldik et al., 1995) and the *Schlichting* test for Dutch language production (Schlichting et al., 1995). The Reynell is an adaptation of the Reynell Developmental Language Scales (Reynell, 1985) and involves only tasks for verbal comprehension. The Schlichting measures expressive vocabulary requiring the naming of objects and pictures. Raw scores were converted into age equivalents. All three instruments have been found reliable and valid; internal consistency was above .80 and test-retest reliability ranged from .69 - .82 (Evers et al., 2009).

Procedure

Participants with AD were recruited from several special schools and special day care centers for children with ID in the Netherlands. TD children visited regular day care centers or nurseries. First, informed parental consent was obtained. Subsequently, all participants were assessed using the same instruments. In the first three sessions at school or day care, participants were given the SON-R, Reynell, and Schlichting. The DISCO-11 interview was conducted in the parents' home. Thereafter, the child and one of the parents were invited to visit the university, school, or day care of the child, where the ADOS and the CSBS-DP were administered. The ADOS and CSBS-DP were video-recorded for later rating.

Data analyses

SPSS 17.2 was used to conduct the analyses. Prior to analyses, the coded communicative acts were transformed into rates-per-minute to control for variations in the duration of the Behavior Sample for each child. The mean duration for the AD group was 17.2 minutes (range: 7.3 - 28.7, *SD* = 5.1) and for the TD group 17.3 minutes (range:

12.0 - 23.5 minutes; *SD* = 2.6). Furthermore, proportion of functions and forms were computed based on the total amount of communicative acts of the child. All frequency counts were acceptable to convert into rate and proportion metrics, since positive and linear relations were found between numerators and denominators, except for the function category unclear (Yoder & Symons, 2010).

All data were tested for normality and proportional data were arcsine transformed prior to statistical tests (Cohen et al., 2003). Several independent sample *t*-tests were computed to identify possible differences between the TD group and the AD group in total rate of communicative acts, and rates and proportions of communicative functions. Given the number of *t*-tests conducted, a Bonferroni correction was applied to the analyses, resulting in an adjusted significance level of $p < .006$. Cohen's *d* was computed as a measure of effect size. Repeated measures mixed analyses of variance were used to compare groups profiles on communicative functions, with one between-subjects factor of group (AD and TD), and one within-subject factor of function (BR, SI, and JA). Further, separate analyses for both groups were conducted to test differences in function within the two groups. Pearson correlations were calculated to examine the relationships between the proportions of communicative functions and other developmental characteristics. Bonferroni correction was applied for multiple comparisons, resulting in an adjusted significance level of $p < .0015$. A series of one-way analyses of variance were conducted to analyze group differences on communicative functions for two AD groups (verbal and nonverbal) and the TD group. The adopted significance level after Bonferroni correction was $p < .006$. Significant effects were followed up with post hoc tests to evaluate differences among the three groups. The Games-Howell procedure was used, because the variances between the groups were unequal. Analyses of covariance were also run to control for the influence of differences in nonverbal mental age results between the three subgroups. Differences in complexity of communicative forms were studied for the separate function categories using several one-way analyses of variance with an adjusted significance level of $p < .01$ after Bonferroni correction. Since the variances were equal for all levels of complexity, Bonferroni post hoc tests were conducted to provide pairwise comparisons. Eta squared was used as a measure of effect size.

Results

Differences in rate and proportion of communicative functions

Table 3.3 presents mean rates of communicative acts per minute for the two groups during the Behavior Sample. It is evident that children with AD communicated less frequently than TD children, $t_{(50)} = -8.2$, $p < .001$, Cohen's $d = 2.3$, large effect. The AD group communicated significantly less often for most of the communicative functions. However, acts with an unclear function occurred more frequently within the AD group.

All subsequent comparisons between the groups were based on both rate and percentages of the total amount of communicative acts. In this way, communicative patterns could also be studied independent of the frequency of communicative acts.

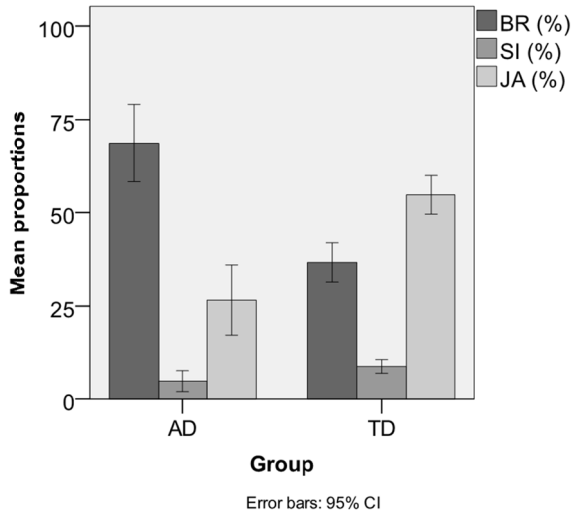
Table 3.3 ***Rate and proportion of communicative functions for AD and TD group***

	AD group (*n* = 26)				**TD group** (*n* = 26)				***t*-test**	**Effect size**
	Min	Max	*M*	*SD*	Min	Max	*M*	*SD*	*p*	Cohen's *d*
Total rate of acts p/m	0.17	2.51	1.22	0.63	0.89	4.18	2.87	0.81	<.001*	2.32 [a]
Functions - rate p/m:										
Behavior Regulation (rate)	0.17	1.66	0.74	0.36	0.37	1.78	1.02	0.38	.009	-
Social Interaction (rate)	0.00	0.33	0.06	0.09	0.05	0.50	0.24	0.14	<.001*	1.56 [a]
Joint Attention (rate)	0.00	1.27	0.42	0.43	0.32	2.50	1.61	0.63	<.001*	2.25 [a]
Function Unclear (rate) [b]	0.00	0.69	0.18	0.20	0.00	0.10	0.01	0.03	<.001*	1.21 [a]
Functions - proportion:										
Behavior Regulation (%)	14	100	68.7	25.8	18	68	36.7	13.0	<.001*	1.47 [a]
Social Interaction (%)	0	27	4.7	6.9	2	16	8.6	4.6	.021	-
Joint Attention (%)	0	78	26.6	23.2	26	73	54.7	12.8	<.001*	1.49 [a]

* $p < .006$ (adjusted significance level after Bonferroni correction)
[a] large effect size; [b] acts with unclear function were not included in the calculation of total rate of acts or proportions

When children in the AD group did communicate, it concerned acts to regulate the behavior of others most of the time, whereas communication for social purposes (SI and JA) was less represented. The high standard deviations for all functions are remarkable. In the TD group, the proportion of communicative acts for joint attention was much higher than for behavior regulation. In both groups, acts for social interaction were less common. *T*-tests showed that in terms of percentages AD children communicated significantly more often for behavior regulation than TD children ($t_{(50)}$ = 5.2, $p < .001$, Cohen's d = 1.5, large effect), and TD children communicated more often for joint attention than AD children ($t_{(50)}$ = -5.3, $p < .001$, Cohen's d = 1.5, large effect) (see Table 3.3).

Profile analysis revealed a significant difference in profiles between the two groups, Greenhouse-Geisser adjusted $F_{(1.10, 54.93)} = 27.40$, $p < .001$. The profiles of the two groups are presented in Figure 3.1. Within the AD group and the TD group, significant differences between the mean proportions of communicative functions were found (AD group: Greenhouse-Geisser adjusted $F_{(1.07, 26.80)} = 35.12$, $p < .001$; TD group: Greenhouse-Geisser adjusted $F_{(1.17, 29.26)} = 71.06$, $p < .001$). Pairwise comparisons revealed significant differences between the proportions of all three functions within the groups. In the AD group the mean proportion of social interaction was the lowest, behavior regulation was the highest, and joint attention was positioned in between, whereas in the TD group the results were the same for social interaction, but exactly the opposite for behavior regulation and joint attention (Bonferroni: $ps < .01$) (see Figure 3.1).



Note. BR = behavior regulation; SI = social interaction; JA = joint attention
Figure 3.1 **Communication profiles for AD and TD group**

Communicative functions in relation to verbal and nonverbal abilities

Correlations between chronological age, nonverbal intelligence, expressive and receptive language development, and use of communicative functions were computed separately for both groups (see Table 3.4).

Table 3.4 ***Correlations between age, nonverbal intelligence, language abilities, and communicative functions for AD and TD group***

	Age	Nonverbal mental age	Receptive language	Expressive language
AD group (n = 26)				
Total rate of communicative acts	-.07	.34	.60*	.52
Behavior Regulation (%)	-.15	-.34	-.70*	-.69*
Social Interaction (%)	.14	-.11	.04	.04
Joint Attention (%)	.16	.44	.70*	.71*
TD group (n = 26)				
Total rate of communicative acts	.24	.11	.17	.35
Behavior Regulation (%)	-.52	-.50	-.50	-.52
Social Interaction (%)	.09	.15	.19	.06
Joint Attention (%)	.52	.47	.46	.52

* $p < .0015$ (adjusted significance level after Bonferroni correction)

Receptive and expressive language were highly correlated with each other in the AD group ($r_{(24)} = .96$, $p < .001$), though the relation with nonverbal mental age was much weaker ($r_{(24)} = .60$ and $r = .58$ respectively; $ps < .01$). Strong significant negative correlations were found between both receptive and expressive language and the proportion of communicative acts for behavior regulation ($r_{(24)} = -.70$ and $r_{(24)} = -.69$, $ps < .001$), and also strong significant positive correlations with acts for joint attention ($r_{(24)} = .70$ and $r_{(24)} = .71$, $ps < .001$). The correlations between the functions and chronological age or nonverbal intelligence were weak or only modest in the AD group.

Nonverbal mental age and both language comprehension and production were strongly related to each other and to chronological age in the TD group ($r_{(24)}$ = .72 and higher, *ps* < .001). The relationships between these three developmental aspects and the proportion of communicative acts for behavior regulation and joint attention were only moderate in the TD group ($r_{(24)}$ ranges from (-).46 to .52, *ps* < .05), and no significant correlations with social interaction were found.

Communicative functions in nonverbal versus verbal children with AD

In Table 3.5 the results of the nonverbal and verbal AD group are presented separately and compared with each other and the TD group. The verbal AD group is positioned between the nonverbal AD group and the TD group in total rate of communicative acts, with significant differences between the three groups, $F_{(2, 49)}$ = 44.7, p < .001, η^2 = .14, large effect (see Table 3.5). The same pattern is more or less visible for the mean rates for the three different functions, but differences are not significant for behavior regulation. The verbal AD group demonstrated the highest rate of unclear communicative acts.

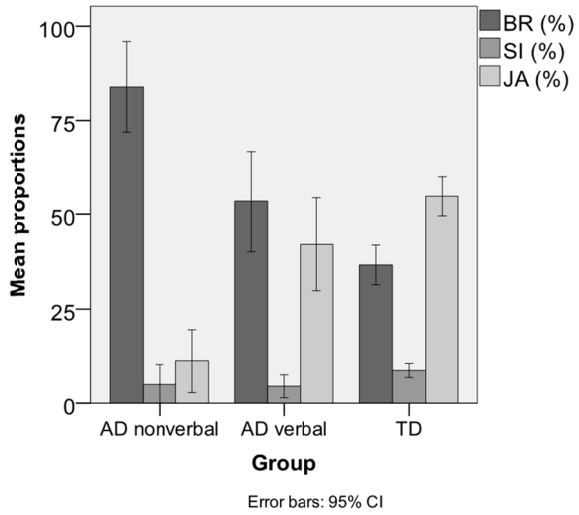
Table 3.5 ***Differences between nonverbal and verbal AD groups in use of communicative functions***

	AD1 nonverbal (*n* = 13)		**AD2 verbal** (*n* = 13)		**TD** (*n* = 26)		**ANOVA**		**Effect size**	**Post-hoc tests**
	M	*SD*	*M*	*SD*	*M*	*SD*	*F*	*p*	η^2	Games-Howell
Total rate of acts p/m	0.81	0.42	1.64	0.52	2.87	0.81	44.7	<.001*	.14 [a]	AD1 < AD2 < TD
Functions – rate p/m:										
Behavior Regulation	0.65	0.37	0.83	0.35	1.01	0.38	4.5	.016	-	-
Social Interaction	0.05	0.09	0.08	0.09	0.24	0.14	14.3	<.001*	.18 [a]	AD1 & AD2 < TD
Joint Attention	0.11	0.17	0.73	0.40	1.61	0.63	42.5	<.001*	.24 [a]	AD1 < AD2 < TD
Function Unclear	0.10	0.16	0.25	0.21	0.01	0.03	14.0	<.001*	.27 [a]	AD1 & TD < AD2
Functions – proportion:										
Behavior Regulation (%)	83.9	19.9	53.4	22.0	36.7	13.0	33.7	<.001*	.18 [a]	TD & AD2 < AD1
Social Interaction (%)	4.9	8.6	4.5	5.0	8.6	4.6	2.8	.069	-	-
Joint Attention (%)	11.1	13.9	42.1	20.3	54.7	12.8	32.6	<.001*	.15 [a]	AD1 < AD2 & TD

* p < .006 (adjusted significance level after Bonferroni correction)
[a] large effect size

The proportion of behavior regulation was much higher in the nonverbal AD group than in the two other groups, $F_{(2, 49)}$ = 33.7, p < .001, η^2 = .18, large effect (see also Figure 3.2). Nearly half of the nonverbal group (*n* = 6) did not show any acts for social interaction or joint attention at all, whereas in the verbal AD group and the TD group, all children communicated for all three functions. Therefore, the proportion of joint attention in the nonverbal AD group was significantly lower than in the verbal AD and TD groups, $F_{(2, 49)}$ = 32.6, p < .001, η^2 = .15, large effect. Post-hoc tests showed no difference between the verbal AD group and the TD group in the proportion of both behavior regulation and joint attention (p > .05) (Table 3.5). Though the profiles of the verbal AD group and the TD group were not significantly different, the verbal AD group

showed a relatively higher mean score for behavior regulation compared to joint attention. Since the nonverbal AD group differed from the verbal AD group and the TD group on nonverbal mental age, the analyses were rerun using nonverbal mental age as covariate. These results indicate that the covariate did not influence the results of the analyses of variance.



Note. BR = behavior regulation; SI = social interaction; JA = joint attention

Figure 3.2 **Communication profiles for AD groups and TD group**

Differences in complexity of communicative forms

In order to examine the complexity of communicative forms used for the different communicative functions, proportions were calculated for three different levels of complexity from the total acts per function. Table 3.6 shows the proportion of complexity levels used for the functions behavior regulation and joint attention. Social interaction was excluded because of the low rates of communicative acts for this function in all three groups.

Table 3.6 ***Differences between AD and TD groups in levels of complexity of communicative forms***

	AD1 nonverbal (n = 13)		**AD2 verbal** (n = 13)		**TD** (n = 26)		**ANOVA**		**Effect size**	**Post-hoc tests**
	M	*SD*	*M*	*SD*	*M*	*SD*	*F*	*p*	η^2	Bonferroni
Behavior Regulation										
Complexity 1 (%)	49.9	20.9	14.0	12.4	10.0	9.9	37.6	<.001*	.32 [a]	AD2 & TD < AD1
Complexity 2 (%)	41.1	18.2	68.1	11.5	58.6	15.0	10.9	<.001*	.03 [c]	AD1 < AD2 & TD
Complexity 3 (%)	9.1	12.1	17.9	11.9	31.4	13.5	14.4	<.001*	.12 [b]	AD1 & AD2 < TD
Joint Attention	(n = 7) [d]		(n = 13)		(n = 26)					
Complexity 1 (%) [e]	-	-	-	-	-	-	-	-	-	-
Complexity 2 (%)	93.5	17.2	88.4	10.8	77.2	12.0	11.2	<.001*	.03 [c]	TD < AD1 & AD2
Complexity 3 (%)	6.5	17.2	11.2	10.4	22.4	11.7	6.3	.004*	.09 [b]	AD1 & AD2 < TD

* p < .01 (adjusted significance level after Bonferroni correction)

[a] large effect size; [b] medium effect size; [c] small effect size; [d] 6 children did not show any communicative acts for joint attention; [e] percentages were too small to analyze

Analyses of the acts for behavior regulation revealed that children in the nonverbal AD group used the least complex form significantly more often than the TD group and the verbal AD group. However, the combination of communicative forms used by verbal children with AD was only significantly distinct from the TD group at the most complex level, that is the combination of a gesture, verbalization, and eye gaze (p = .009). For behavior regulation, the verbal AD group showed a significant higher complexity of communicative forms than the nonverbal AD group. In comparison to the TD group, significant differences only were found at the highest level of complexity. Post hoc analyses on the joint attention data indicated that both AD groups used forms with an intermediate level of complexity more often than the TD group and combined three different communicative forms significantly less in comparison with the TD group. In other words, the level of complexity is significantly lower in both the nonverbal and the verbal AD groups than in the TD group.

Discussion

Communication problems in individuals with ASD and associated ID are often very severe and heterogeneous (Boucher et al., 2007). The goal of our study was to describe the communication abilities of low-functioning children with autism (AD) in a comprehensive way by examining both communicative functions and complexity of communicative forms. Communication profiles of low-functioning verbal and nonverbal children with AD were compared with those in a control group consisting of typically developing children. The children were individually matched on nonverbal developmental level.

Rate and functions of communication in low-functioning children with autism

The results of this study indicated that the amount of intentional communication is significantly lower in children with co-occurring AD and ID than in typically developing children with a comparable developmental level. Reduced frequency of communication is one of the core deficits of ASD, appearing already at a very young age, as demonstrated in several studies on toddlers (Landa et al., 2007). Our study focused on older children with AD and a co-occurring ID, thus the results indicate that the amount of intentional communication remains reduced while growing up. The lower frequency of communication can reflect a lower intrinsic motivation to communicate and interact with others, or it can be due to a reduced ability to initiate social bids and respond to the social environment, resulting in fewer opportunities to influence the social environment (Shumway & Wetherby, 2009).

With regard to the functions of communication, the results of our study indicated that low-functioning children with AD show a specific communication profile as compared to typically developing children. Children with AD and ID prefer to

communicate for the purpose of behavior regulation rather than social interaction and joint attention, while in TD children the opposite profile is present. A previous study found the same profile in very young children with ASD (Shumway & Wetherby, 2009). Communication for behavior regulation, such as requesting or protesting, can be characterized as less complex and less social than communication for joint attention purposes (Landa, 2007; Wetherby et al., 1997). To learn to interact with others, children need to pay attention to socially relevant stimuli, such as faces and voices, in their environment. Several studies have shown that from early on children with ASD are less focused on such stimuli (Dawson et al., 1998; Klin et al., 2003). As a consequence, children with ASD participate less frequently in social interactions, which impedes the development of social cognitive abilities, such as joint attention (Dawson et al., 2004; Klin et al., 2002).

Communication profiles of nonverbal and verbal children with autism

A considerable amount of heterogeneity in communication was observed within the AD group. For example, some children with AD communicated only a few times during the whole assessment, and all communicative acts served the purpose of behavior regulation, while other children with AD communicated as often as TD children, even for joint attention purposes. Language development (both expressive and receptive) showed the strongest relation with the proportional use of communication for behavior regulation and joint attention compared to other characteristics, such as chronological age and nonverbal mental age. The development of joint attention is usually intertwined with that of language acquisition: joint attention is essential to create shared situations, in which children can learn from the language of others, but the acquisition of language also increases the possibilities for joint attention, because the scope of situations extends from present objects and activities to objects out of sight, future and past events, and internal states (Adamson et al., 2009; Carpenter & Tomasello, 2000).

In order to further explore the role of language, communication profiles were examined in both a verbal and a nonverbal subgroup of children with AD. In our study, low-functioning nonverbal children with AD showed the most atypical and limited communication profile compared to verbal children with AD and typically developing children. These findings are in line with the results of the study by Stone and Caro-Martinez (1990) in which verbal and nonverbal children with AD (including children with and without ID) were compared in a naturalistic setting. Communicative acts for social purposes seemed to be strongly related to language abilities in low-functioning children with AD. Nearly half of the nonverbal group in our study did not show any communication aimed at social interaction or joint attention, and the other half only for a very small part. In contrast, typically developing children are able already before language development to communicate extensively for social interaction and joint attention (Coupe O'Kane & Goldbart, 1998; Wetherby et al., 1997). On the basis of the

results of this study, communication on the part of low-functioning nonverbal children with AD can mainly be characterized as communication intended to regulate somebody else's behavior, to obtain or protest against certain objects or activities. Furthermore, these children communicate by means of very primitive forms of the least complex level, such as manipulations of persons or objects and movements of objects or simple gestures. Nonverbal children with AD have very restricted communicative abilities to express their needs and desires to others, and consequently are less able to influence their social environment. It is harder to understand nonverbal children with AD, especially for unfamiliar individuals, since these children use very unconventional and sometimes very subtle ways to communicate (Wetherby et al., 1997). Particularly in the nonverbal group, challenging behavior is often seen and this may be used for communicative ends (Schuler et al., 1997).

In our study, significant differences in frequency of communicative acts were found between verbal children with AD and TD children, but there were no differences in the proportional use of the communicative functions between these two groups. This suggests that in the verbal group with AD differences in the use of communicative functions as compared to TD children are quantitative rather than qualitative in nature. However, only basic communicative functions were included in this study. The higher rate of communication for unclear functions in the verbal group as compared to both other groups may be explained by the amount of echolalia or other aberrant language features observed in this group. The verbal AD group made more efforts to communicate, as the rate of communication was significantly higher than in the nonverbal AD group, but these children were not always successful in communicating their intentions in an effective way. Low-functioning verbal children with AD also differ significantly from nonverbal children with AD in the proportion of communicative acts for the purpose of joint attention. In a previous study by Stone and Caro-Martinez (1990) the same differences between verbal and nonverbal children with AD were found during observations in a naturalistic setting. With respect to forms, verbal children with AD were more likely to use a combination of two communicative forms instead of three than TD children for both behavior regulation and joint attention.

Limitations and recommendations for future research

Using observational methods permits an examination of communication patterns in low-functioning individuals with AD in great detail. As such, this is a valuable technique in research into ID, since no standardized tests for communication are available which provide norms for this low-functioning population. However, there are some methodological issues that limit the strengths of the present findings. The first limitation concerns the small sample size, in particular when specific subsamples were examined. Nevertheless, the number of participants compares well with other studies on communication profiles. Second, the situations of the CSBS-DP are specifically developed to elicit communicative behavior and are not completely comparable to

communication in daily life. However, a standardized observational measurement procedure, such as the CSBS-DP provides a more stable estimate of a child's true performance, because error introduced through uncontrolled factors is minimized (Yoder & Symons, 2010). The CSBS situations mainly focused on behavior regulation and joint attention, because most items are designed for triadic interaction, i.e., person-object-person or object-person-object interaction, resulting in a possible underestimation of the level of social interaction and overestimation of the proportions for the other two functions. Therefore, future studies should examine communication during more varied activities, including social activities without play materials. Another weakness of this study concerns the sex difference between the AD and TD groups. A comparison group matched on sex is recommended for future research. In addition, it would be useful to have a control group of verbal and nonverbal children with an ID without ASD and a comparison group consisting of children with AD without ID. In this way it is possible to control for the effect of experience and to analyze which characteristics may be due to the AD diagnosis and which to the ID diagnosis. In this study the TD control group was matched on nonverbal mental age, not on chronological age, in order to create groups of comparable levels of functioning.

Implications for clinical practice

More insight into communication strengths and difficulties in low-functioning children with AD is valuable for both assessment and treatment goals. Knowledge of the child's actual communication level regarding functions and forms can change the caregivers' expectations and subsequently their communication strategies (Yoder & Warren, 2001). Because of the heterogeneity in the low-functioning AD group, an individual approach regarding assessment and intervention is necessary. A comprehensive evaluation of communication, with respect to both functions and forms, is recommended to choose the intervention best suited for improving communication abilities.

The use of basic communicative functions is impaired mainly in low-functioning nonverbal children with AD. Several treatment strategies have been developed to stimulate the communication abilities of children with ASD. In most cases the focus of these interventions is on improvement in the use of communicative forms, especially language, or the development of one specific function, namely joint attention. In line with the results of this study, improvement of communicative intentionality seems to be equally or even more important than an emphasis on communicative forms. After all, the ability to use (conventional) communicative forms is useless when children do not know how and when to communicate their needs and desires (Schuler et al., 1997; Wetherby et al., 1997). Purposefully communicative behavior enables a child to influence the environment and has a positive influence on other developmental aspects (Coupe O'Kane & Goldbart, 1998). Therefore, establishment of intentionality and expansion of the range of communicative functions are important goals for interventions in low-functioning nonverbal children with AD. To accomplish these aims

individual strengths, motivations, and preferred activities are useful starting points (Schuler et al., 1997). It is possible to stimulate intentionality, for example by interrupting well-known routines, placing a favorite play object out of reach, or encouraging the child to choose between favorite snacks or activities. Other developmental aspects, such as the presence or absence of object permanence, are important to determine how and in which situations communication may be encouraged.

Regarding low-functioning verbal children with AD the focus of communication intervention shifts to the effective use of conventional communication forms and skills in more complex situations. Since verbal children with AD try to initiate communication much more often than nonverbal children with AD, but also commit more communicative acts with an unclear function, the results suggest that some of these children are making an effort to communicate, but do not always succeed in communicating their intention in an effective way. This group may be particularly ripe for interventions focusing on the improvement of the quality and diversity of communicative functions and the use of more conventional ways to communicate. Intervention goals need to be based on individual needs and may also lie beyond the scope of this study, such as vocabulary expansion, supporting specific pragmatic skills, and stimulating communication about decontextualized topics (Tager-Flusberg et al., 2005).

The generic goals for interventions described above, all concentrated on expressive communication, since our study focused on initiation of intentional communication. Especially in the case of children with AD and ID, problems in receptive communication are usually even more severe than problems in expressive communication. Therefore, stimulation of communicative competence should be focused on the support of receptive communication as well (Noens & Van Berckelaer-Onnes, 2004).

Conclusions

Since our study focused on a particular subgroup – children with co-occurring AD and ID – which is often neglected in research, it constitutes an important contribution to the current knowledge on communication in ASD. Characteristics of children with co-occurring AD and ID are not comparable to characteristics of other subgroups of children with ASD right away and need to be studied separately. The results of this study suggest that communication profiles in low-functioning children with AD are extremely variable. A striking difference between the profiles of verbal and nonverbal children with AD and ID was found. Communication in nonverbal children with AD was very limited and mainly served imperative purposes to regulate behaviors of others. Verbal children with AD communicated more often for declarative purposes, such as

joint attention, than the nonverbal subgroup. The nonverbal AD group used less complex forms of communication in comparison to the verbal AD group and typically developing children with a comparable developmental level. The verbal AD group differed from typically developing children only in rate of communication, not in proportion of communication for specific functions. Only at the most complex level of communicative forms (a combination of three different forms) a significant difference in complexity was found between the verbal AD group and the TD group. The results of this study indicate the presence of severe communication impairments in low-functioning children with ASD, especially in nonverbal children, and emphasize the importance of suitable communication interventions for this vulnerable group.

4

Language in low-functioning children with autism

Jarymke Maljaars
Ilse Noens
Evert Scholte
Ina van Berckelaer-Onnes

Journal of Autism and Developmental Disorders (2012), doi:10.1007/s10803-012-1476-1

Language in low-functioning children with autistic disorder: Differences between receptive and expressive skills and concurrent predictors of language

Abstract

Language profiles of children with autistic disorder and intellectual disability (*n* = 36) were significantly different from the comparison groups of children with intellectual disability (*n* = 26) and typically developing children (*n* = 34). The group low-functioning children with autistic disorder obtained a higher mean score on expressive than on receptive language, whereas both comparison groups showed the reverse pattern. Nonverbal mental age, joint attention, and symbolic understanding of pictures were analyzed in relation to concurrent receptive and expressive language abilities. In the group with autistic disorder and intellectual disability, symbol understanding and joint attention were most strongly related to language abilities. Nonverbal mental age was the most important predictor of language abilities in the comparison groups.

Introduction

The development of language skills is one of the most important achievements of early childhood. Language acquisition leads to new opportunities in other developmental areas, such as cognitive, social, and emotional development and is a cornerstone for successful outcomes later in life (Schlichting et al., 1995). Although language ability usually develops spontaneously, it does not progress typically for a substantial number of children. Children with autism spectrum disorder (ASD) show impairments in language development (Tager-Flusberg et al., 2005; Eigsti et al., 2011). The development of functional language serves an important prognostic function (Mahwood et al., 2000). Thus, gaining a better understanding of language impairments in ASD is important for development and improvement of interventions.

In order to provide a context in which we can evaluate the language impairments in ASD, typical language development will be described briefly. Bates (1979) indicated two critical transitions in early childhood which precede language development: 1) development of communicative intentionality, and 2) symbol formation. The first transition is the onset of communicative intentionality, when children become aware that their signals influence behavior of others. In typical development, intentionality emerges when children start to use proto-imperative and proto-declarative behaviors, i.e., to obtain a desired object / event or to share attention / interest between persons on a common focus, by the end of their first year of life (9-13 months) (Baron-Cohen, 1989; Bates, 1979; Camaioni, 1997).

The second transition concerns the emergence of symbol understanding and use. According to Bates (1979) symbol formation requires an understanding of the relationship between a sign and its referent. The sign can replace that referent in a variety of situations, even if the referent is not currently present. During the first years of life children gradually learn to understand and use symbols, which is reflected in the development of conventional gestures, words, and symbolic play (Wetherby et al., 1998). Language is a complex symbolic communication system, for which symbol-referent relations cannot be inferred through observations, but have to be learned step by step. Children may use first words before they have any explicit symbolic understanding. Only when a word is generalized beyond the initial learning environment, it may be granted as symbol use and understanding (Namy & Waxman, 2005). Next to the verbal domain, the process of symbol development can also be examined in the nonverbal domain. Symbolic play requires the ability to symbolize, and awareness of the relation between the present play object or action and its absent referent in daily life (Jarrold et al., 1997). Symbolic play has been linked to language development in typical children and young children with ASD (e.g., Lewis et al., 2000; McCune, 1995; Toth et al., 2006). Other symbolic abilities, such as understanding of pictures, have not been studied in relation to language abilities yet. Stephenson and Linfoot (1996) argued that the use of pictures as symbols has clear parallels to the use of words as symbols.

In typical semantic development, language comprehension always precedes production (Fenson et al., 2000a). Word comprehension emerges around 9 months of age and by the first birthday toddlers usually say their first recognizable words. By this age, they can understand many more words and even some simple phrases (Fenson et al., 1994). Initially, meaning is linked to a specific context. Between 12 and 18 months of age, receptive and expressive vocabulary gradually increases and becomes less context-bound (Tager-Flusberg, 1997). The production of phrases starts between 18 and 24 months of age (Fenson et al., 1994). During the preschool period, the vocabulary and complexity of grammar expand rapidly (Tager-Flusberg et al., 2005).

Limited intentionality and symbol formation are considered to be core deficits in communication of individuals with ASD (Noens & Van Berckelaer-Onnes, 2005; Travis & Sigman, 2001; Wetherby et al., 2000). Young children with ASD exhibit intentional communication less frequently than typically developing children or children with developmental delays (e.g., Chiang et al., 2008; Shumway & Wetherby, 2009). A reduced amount of intentional communication is also seen in older children with autistic disorder and associated ID (Maljaars et al., 2011 (Chapter 3)). The most striking impairments in ASD are found in proto-declarative communication or joint attention (e.g., Mundy & Burnette, 2005). Several studies found a predictive relationship between joint attention and expressive language development for young children with ASD (e.g., Charman et al., 2003; McDuffie et al., 2005; Smith et al., 2007; Watt et al., 2006). Symbolic impairments have already long been associated with ASD (Hammes &

Langdel, 1981; Ricks & Wing, 1975), but it remains unclear whether a more general impairment in symbol formation is related to language impairments in ASD.

Children with ASD vary widely in their language abilities and features. Only a few studies analyzed differences in expressive versus receptive language abilities based on a within-group design (Tager-Flusberg et al., 2005). In toddlers with ASD, lower levels of both expressive and receptive language compared to typical control or norm groups were found. In contrast to typical development, language comprehension is often even more delayed relative to production (Charman et al., 2003; Hudry et al., 2010; Luyster et al., 2008; Weismer et al., 2010). Generally, in older, high-functioning children with ASD the discrepancy between receptive and expressive language decreases (Rapin & Dunn, 2003; Tager-Flusberg et al., 2005). Jarrold et al. (1997) described relatively uniform language profiles in children and adolescents with ASD with equal levels of expressive and receptive language. Another study from Kjelgaard and Tager-Flusberg (2001) found that receptive skills were comparable to expressive skills on vocabulary tests.

Research into ASD has focused on high-functioning, rather than low-functioning children with ASD. In case of ASD with co-occurring intellectual disability (ID), children are 'double impaired' with respect to language abilities. A large proportion of low-functioning individuals with ASD does not develop language or acquires at most a few functional words or signs (Boucher et al., 2007; Rapin & Dunn, 2003). Language comprehension is always impaired in low-functioning individuals with ASD, although severity varies (Boucher et al., 2007). A higher level of expressive language compared to receptive language can for example be due to echolalia or the ability to label or to reproduce memorized language in specific contexts (Tager-Flusberg et al., 2005). In this way, true level of language understanding is concealed. Not surprisingly, impairments in receptive language are strongly associated with severe behavior problems (Sigafoos, 2000). Language abilities and characteristics in children with ASD are closely related to nonverbal cognitive level of functioning (Luyster et al., 2008; Weismer et al., 2010).

Identifying factors that are associated with concurrent language abilities and possibly influence differential outcomes is necessary for improving treatments. General intellectual level of functioning is probably the most important factor associated with language development in children with ASD (Luyster et al., 2008; Weismer et al., 2010), but this factor does not explain all variance in language abilities. Other developmental factors, such as social and cognitive abilities, are also crucial (Prizant, 1996; Thurm et al., 2007). Low-functioning children with ASD often exhibit more severe delays in language development relative to their nonverbal cognitive level. The question is whether limitations in language abilities in older children with ASD and ID are associated with social aspects, such as the ability to initiate and respond to joint attention, or with cognitive aspects, such as the understanding of symbols. Both limited intentionality and symbol formation are considered to be core deficits in communication development of individuals with ASD (Travis & Sigman, 2001). Joint attention cannot be the only explaining factor for language impairment in children with

ASD, because older children with ASD sometimes have impaired joint attention skills but adequate levels of language (Bloom, 2000). Indirect evidence for an association between symbol formation and language abilities is that individuals with ASD are impaired in their development of different skills that require symbolic abilities, such as language and symbolic play. Furthermore, in contrast with for example specific language impairment, problems in language development seem to be amodal in ASD (Boucher et al., 2007). Nonverbal children with ASD often fail to compensate their lack of verbal communication with other modalities, such as gestures (Mundy et al., 1994). These findings support the hypothesis of a relationship between language development and symbolic capacities in the nonverbal domain.

Research into specific language profiles and the relation with other developmental aspects in low-functioning children with ASD is important from the perspective of clinical practice, but also for research. First, a lack of sufficient functional language can result in challenging behavior, in particular in low-functioning nonverbal individuals (Chiang, 2008; McClintock et al., 2003), and form a major stressor for parents (Lecavalier et al., 2006). Language abilities are closely related to later prognosis (Mawhood et al., 2000). Therefore, more insight into developmental factors which are associated with language development in low-functioning children with ASD is important. Better understanding of language impairments in low-functioning children is necessary in order to improve interventions and care. Second, the results can be useful for genetic and neurobiological studies. Individuals with ASD and associated ID are more often included in genetic studies than in behavioral or cognitive studies. Subsetting by language phenotypes has proven to be useful in genetic research, and also led to identification of genome-wide significant loci (Abrahams & Geschwind, 2008). Therefore, the main purposes of this study are: 1) to study differences and similarities between receptive and expressive language abilities in children with autistic disorder and ID compared to children with ID without ASD and typically developing children; and 2) to examine whether and how precursors of language (joint attention and symbol understanding) are related to concurrent receptive and expressive language abilities in these groups.

Method

Participants

The first group comprised 36 children with autistic disorder and intellectual disability (AD+ID). Participants were included in the AD+ID group if they had received a formal classification of autistic disorder and ID conform DSM-IV-TR criteria (APA, 2000) prior to the study. All children had an algorithm score above the threshold for autistic disorder on the Autism Diagnostic Observation Schedule Modules 1 or 2 (ADOS; Gotham et al., 2007) and a classification of autistic disorder on the Diagnostic Interview

for Social and Communication Disorders – version 11 (DISCO-11; Wing, 2006). Given the classification difficulties in low-functioning individuals, this study focused on the core syndrome instead of the whole spectrum to create a more homogeneous group. Levels of ID ranged from mild to severe (APA, 2000). A nonverbal intelligence test confirmed that IQ-scores of all participants were below 70. The AD+ID group consisted of 32 boys and 4 girls with a mean chronological age of 7.1 years (*SD* = 2.3; range: 3.3 - 11.3). The mean nonverbal mental age was 39.0 months (*SD* = 13.7). In some children additional conditions, such as ADHD (*n* = 3), Down syndrome (*n* = 1), and Fragile X syndrome (*n* = 1), were present. Two comparison groups were used: a group of children with intellectual disability (ID; *n* = 26) with 15 boys and 11 girls, and a group of young typically developing children (TD; *n* = 34) with 14 boys and 20 girls. Children in the ID and TD groups did not meet diagnostic criteria for autism spectrum disorder on the ADOS. Children in the ID group all had a formal classification of ID, ranging from mild to severe (APA, 2000), confirmed by a nonverbal intelligence test. The mean chronological age for the ID group was 6.5 years (*SD* = 1.5; range: 4.3 - 11.0) and for the TD group 3.0 years (*SD* = 0.5; range: 1.9 - 3.9). Mean nonverbal mental age for both groups was 38.5 months (*SD* = 12.7) and 4.3 months (*SD* = 9.5), respectively. Additional conditions identified in the ID group were: Down syndrome (*n* = 11), Williams syndrome (*n* = 1), Velo-Cardio-Facial syndrome (*n* = 1), ADHD (*n* = 1), and epilepsy (*n* = 1). The main characteristics of the groups are presented in Table 4.1.

Table 4.1 ***Main characteristics of the subsamples***

		Nonverbal mental age equivalent (in months)			**Chronological age** (in months)			**Gender** (in %)	
Groups	*n*	Range	*M*	*SD*	Range	*M*	*SD*	Male	Female
AD+ID	36	<24 [a] - 68	39.0	13.71	40 - 136	85.1	28.01	89	11
ID	26	<24 [a] - 63	38.5	12.65	51 - 132	78.1	17.59	58	42
TD	34	29 - 65	42.3	9.53	23 - 47	36.5	5.97	41	59

Note. AD = autistic disorder; ID = intellectual disability; TD = typical development
[a] minimum score

The AD+ID group and the two comparison groups were comparable with respect to nonverbal mental age ($F_{(2,93)} = .96$, $p = .39$) (see Table 4.1), since groups were matched on this variable. There were significant between group differences with respect to chronological age ($F_{(2,93)} = 59.46$, $p < .001$), and gender ($\chi^2_{(2)} = 17.71$, $p < .001$). Logically, post hoc tests revealed that participants in both groups with ID were significantly older than the TD group ($p < .05$), because comparable groups were created based on level of functioning instead of chronological age. In the AD+ID group significantly more males were present than in the two comparison groups. This has not threatened the validity of the study, as there was no difference in language outcomes between boys and girls within the three subsamples ($p > .05$).

Instruments

Language abilities

Receptive language. Receptive language scores were derived from two different instruments: (a) Reynell test for Dutch language comprehension (Reynell; Van Eldik et al., 1995), or (b) Dutch Communicative Development Inventories – short forms (D-CDI; Zink & Lejaegere, 2003). Most language scores were based on the Reynell, which evaluates receptive language between 14 and 75 months. Only for children who did not obtain the basal score (<14 months) on the Reynell (5% of the participants), D-CDI was used to determine a receptive language score. The Reynell is an adaptation of the Reynell Developmental Language Scales (Reynell, 1985); the Dutch adaptation only contains tasks for verbal comprehension. The Reynell has to be administered individually and consists of 87 items concerning comprehension from single words to complex sentences. Because several participants in this study were chronologically older than the normative sample of the receptive and expressive language instruments, age equivalents based on raw scores were used for analyses. The Reynell was found to exhibit good psychometric properties. A good internal consistency ($\alpha > .80$) and a sufficient test-retest reliability ($r = .69$) were reported. In addition, there were strong correlations between the Reynell scores and chronological age or other language instruments, which supported validity (Van Eldik et al., 1995; Evers et al., 2009). The Dutch adaptation of the MacArthur Communicative Development Inventories (Fenson et al., 2000b), D-CDI list 1, assesses vocabulary skills between 8 and 16 months of age, based on a parental report. The raw comprehension score was converted into an age equivalent based on percentile 50. The Dutch adaptation of the CDI list 1 has been demonstrated to have very high internal consistency ($\alpha > .97$), and a high convergent and sufficient predictive validity (Zink & Lejaegere, 2003).

Expressive language. Language production age equivalents were also derived from two different instruments: (a) Schlichting test for Dutch language production (Schlichting; Schlichting et al., 1995), and (b) Dutch Communicative Development Inventories – short forms (D-CDI; Zink & Lejaegere, 2003). The same procedure as for receptive language was used. Children with an expressive language level below 17.5 months (12% of the participants) were assigned a score based on the D-CDI. The Schlichting measures language production and two different subscales were used: (a) Schlichting vocabulary development (14-75 months), requiring the naming of objects and pictures (62 items), and (b) Schlichting syntax development (21-75 months), assessing sentence structure (40 items). An overall expressive language age equivalent was calculated by averaging vocabulary and syntax age equivalent scores. The psychometric properties of the Schlichting are sufficient to good. The test was found to have a good internal consistency ($\alpha > .80$) and adequate test-retest reliability ($r = .75$). Correlations between Schlichting scores and other language tests are adequate (Schlichting et al., 1995; Evers

et al., 2009). The Dutch CDI list 1, filled out by parents, also evaluates expressive vocabulary (see description above).

Predictors of language development

As explained in the introduction, possible concurrent predictors of language development included in this study were nonverbal mental age, joint attention, and symbolic abilities.

Non-verbal intelligence. To measure nonverbal mental age, a Dutch test for nonverbal intelligence, the SON-R 2½-7 (Tellegen et al., 1998) was used. The SON-R 2½-7 is an individually administered measure, which consists of six subtests: three reasoning tests (Categories, Analogies, and Situations) and three spatial, performance tests (Mosaics, Puzzles, and Patterns). Total raw scores were converted into nonverbal mental age equivalents (NVMA). The SON-R 2½-7 has been found reliable and valid (Evers et al., 2009).

Joint attention abilities. Children's joint attention (JA) abilities were based on ADOS modules 1 or 2 (Lord et al., 1999). The items included were 'Spontaneous Initiation of Joint Attention' (Module 1 item B10 or Module 2 item B6) and 'Response to Joint Attention' (Module 1 item B11 or Module 2 item B7). Initiation of JA codes the child's attempts to draw another person's attention to objects that are out of reach for the purpose of sharing. The item about response to JA addresses the child's response to the examiner's use of gaze and/or pointing to direct the child's attention to a specific object. The raw scores on both items were combined to a total joint attention score.

Symbol understanding. Symbol understanding was measured by using an adaptation of different symbolic representation tasks developed by Bebko, McCrimmon and McFee (McFee, 2006), based on the procedure of DeLoache and Burns (1994). The adapted version of the dollhouse experiment involves a reduced language protocol and comprises six tasks increasing in difficulty with each task containing six trials. The first three tasks are used to establish understanding of the procedure without the need of verbal instructions by modeling the first item of each task and to practice the search procedure. In task 1 participants had to match identical figurines and in task 2 identical photographs of the figurines were used, which had to be combined with the correct figurine. These two tasks were developed to examine memory and object identification skills, which are necessary for success on the subsequent object-retrieval tasks. The following four tasks consist of search tasks; participants were asked to find a figurine hidden under one of six pieces of household furniture in a 3-floor dollhouse. Photographs of the search location within the dollhouse were shown to the child. The first location searched by the child was recorded as the response. In the 3rd task, the figurine was placed in full view on one of the pieces of furniture; therefore this task can

be seen as a training search task. The other three tasks require the participant to use a picture as a symbol. During task 4, children were shown a photograph of the target item partially visible in its location within the dollhouse, while in the dollhouse the figurine was hidden out of direct view of the child. The 5th task was similar to task 4 with one modification: the photograph did not show the figurine, but only the hiding location. For task 6 the same photographs as for task 5 were used, but the procedure was changed based on the 'Hide-Picture' condition in the study of DeLoache (1991). In this condition, the children were shown a duplicate figurine and the experimenter positioned the photograph in front of the figurine to give a search instruction (McFee, 2006). Tasks 4, 5, and 6 were used to compose a symbol understanding score, ranging from 0-18 based on the sum of correct trials.

Procedure

Participants were recruited from special schools and day care centers for children with mild to severe ID, and from regular day care centers in the Netherlands. Informed consent from the parents was obtained. Parents were asked to complete the D-CDI questionnaire. Each child was individually tested using the SON-R, Reynell, and Schlichting over the course of three or more sessions at school or day care. The DISCO-11 was administered with one or both parents at home. Thereafter, the child and one of the parents were invited to visit the university, school, or day care of the child, where participants were assessed using the ADOS and the dollhouse experiment.

Data analyses

SPSS 18 was used to conduct the analyses. First, receptive and expressive language abilities were evaluated by conducting two analyses of covariance in combination with planned contrasts to compare three subsamples. A repeated measures analysis of covariance (RM-ANCOVA) was used to compare language profiles. Nonverbal mental age was included as covariate in the analyses. Six hierarchical regression analyses were conducted to determine predictors of receptive and expressive language abilities for the separate subsamples. In each analysis, nonverbal mental age was entered in the first step to control for the influence of level of functioning before entering the other predictors. Joint attention and symbol understanding were both entered in the second step, except for joint attention in the TD group, since the distribution of this variable was too skewed and correlations revealed no significant relationship with language outcomes in this sample. Independent variables were checked for multicollinearity. Correlations between predictors were below $r = .70$, except for the correlation between symbol understanding and NVMA in the AD+ID group ($r = .75$, $p < .001$) and in the ID group ($r = .83$, $p < .001$). All other basic assumptions for regression analyses were satisfied. Cases with standardized residuals greater than two standard deviations were excluded from the analyses, but the number of excluded cases did not exceed two per

analysis. The variable symbol understanding contained one missing value in the AD+ID group. An alpha level of .05 was established throughout all analyses.

Results

Receptive and expressive language abilities

Although groups were comparable with respect to mean and range of nonverbal mental age, there were significant differences between language abilities of the three groups (Table 4.2 and Figure 4.1).

Table 4.2 ***Language and nonverbal mental age equivalents for each subsample***

		Receptive language age equivalent [a]			Expressive language age equivalent [a]			Nonverbal mental age equivalent [a]		
Groups	*n*	Range	*M*	*SD*	Range	*M*	*SD*	Range	*M*	*SD*
AD+ID	36	9 - 65	27.8	14.44	8 - 75	29.4	16.85	<24 [b] - 68	39.0	13.71
ID	26	17 - 70	37.3	12.87	13 - 59	33.1	12.78	<24 [b] - 63	38.5	12.65
TD	34	27 - 75	45.6	11.47	23 - 74	43.3	9.92	29 - 65	42.3	9.53

[a] in months; [b] minimum score

Analyses of covariance with nonverbal mental age as covariate, revealed significant differences between the groups in receptive language ($F_{(2,92)} = 31.60$, $p < .001$, partial $\eta^2 = .41$) and expressive language ($F_{(2,92)} = 13.38$, $p < .001$, partial $\eta^2 = .23$). Based on contrasts, the mean receptive age equivalent of the AD+ID group was significantly lower than in the ID group ($p < .001$), whereas the ID group scored significantly lower than the TD group ($p = .02$). With respect to expressive language there was no difference between the ID groups with and without AD ($p = .08$), but both groups received a significantly lower score than the TD group ($ps < .01$).

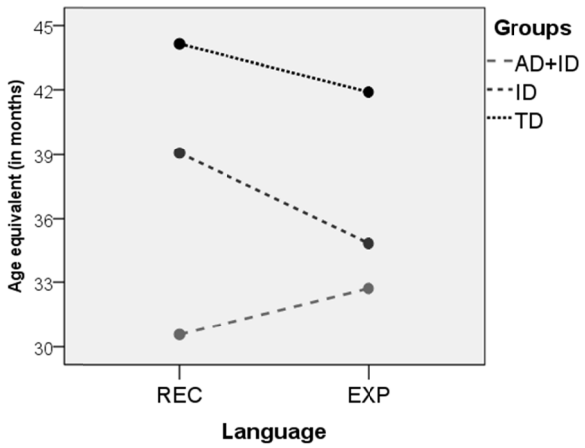


Figure 4.1 **Language profiles for the three groups**

In the AD+ID group 5 children obtained a receptive and expressive language age equivalent below 1;0 year, which means that they did not use or understand any words or at least some simple single words (Table 4.3). Therefore these children were excluded from the analyses concerning differences between receptive and expressive language levels.

Table 4.3 ***Cross tab including differences between receptive and expressive language levels***

	Difference	AD+ID		ID		TD		Total
REC > EXP	≥ 6 months	1	(3)	8	(14)	8	(15)	17
	≥ 3 - 5 months	2		6		7		15
REC = EXP		17		8		11		36
EXP > REC	≥ 3 - 5 months	5	(11)	2	(4)	6	(8)	13
	≥ 6 months	6		2		2		10
Language levels too low to compare [a]		5		0		0		5
Total		36		26		34		96

Note. REC = receptive language; EXP = expressive language
[a] REC & EXP ≤ 12 months

The most striking difference in Table 4.3 is seen between the AD+ID group versus the two comparison groups, $\chi^2_{(4)} = 14.19$, $p = .007$. In the AD+ID group 36% of the children obtained a higher score on the test for expressive language compared to receptive language, whereas in the ID and TD groups, 54% and 44% respectively showed better language comprehension than production skills. In contrast, in the AD+ID group a higher score on receptive language was very uncommon (10%).

Differences in receptive and expressive age equivalents were further examined using a RM-ANCOVA with nonverbal mental age as covariate (Figure 4.1). A significant interaction effect between groups and language abilities was found ($F_{(2,87)} = 7.64$, $p = .001$, partial $\eta^2 = .15$), thus language profiles differed among the three groups. Pairwise comparisons revealed that the AD+ID group obtained significantly higher scores on expressive language than on receptive language ($t_{(30)} = -2.65$, $p = .013$), whereas the ID group ($t_{(25)} = 2.50$, $p = .019$) and the TD group ($t_{(33)} = 2.274$, $p = .030$) showed the opposite profile with significantly higher age equivalents on receptive than on expressive language (Table 4.2).

Concurrent predictors of receptive and expressive language

Hierarchical regression analyses were performed to explore the relationship between joint attention, symbolic abilities, and level of language comprehension and production in each subsample. Regression analyses were used to control for nonverbal mental age and to assess differences in predictive value of joint attention and symbol understanding. Results of the regression analyses are shown in Table 4.4.

For the group of low-functioning children with AD, symbol understanding and joint attention were most strongly related to concurrent language abilities. Together, the predictors explained 81% of the variance in receptive language ($F = 41.49$; $p < .001$)

and 72% of the variance in expressive language abilities (F = 25.09; p < .001). Symbol understanding showed a positive relationship with language abilities and joint attention was inversely correlated with language abilities, since a higher score on joint attention implied more problems with joint attention. With respect to language comprehension, symbol understanding was a stronger predictor (β = .56) than joint attention (β = -.30) (see Table 4.4).

Table 4.4 ***Hierarchical regression analyses for receptive and expressive language including nonverbal mental age, joint attention, and symbolic abilities as predictors***

Group	*n*	Language	Step	Predictor	Beta	*t*	*p*	R^2	$p\ \Delta R^2$
AD+ID	33	Receptive	1	NVMA	.73	5.94	<.001	.53	<.001
			2	NVMA	.15	1.23	.230	.81	<.001
				JA	-.30	-2.68	.012		
				Symbol	.56	4.25	<.001		
	33	Expressive	1	NVMA	.72	5.69	<.001	.51	<.001
			2	NVMA	.21	1.40	.171		
				JA	-.33	-2.47	.019	.72	<.001
				Symbol	.42	2.69	.033		
ID	25	Receptive	1	NVMA	.89	9.25	<.001	.79	<.001
			2	NVMA	.61	4.22	<.001	.83	.02
				Symbol	.35	2.47	.022		
	26	Expressive	1	NVMA	.84	7.65	<.001	.71	<.001
TD	34	Receptive	1	NVMA	.79	7.28	<.001	.62	<.001
	33	Expressive	1	NVMA	.75	6.28	<.001	.56	<.001

Note. NVMA = nonverbal mental age; JA = joint attention; Symbol = symbol understanding

For the ID group, nonverbal mental age and symbolic abilities were significant predictors for receptive language. These variables accounted for 83% of the variance. Standardized coefficients showed that nonverbal mental age was the strongest predictor, followed by symbolic abilities. With respect to expressive language in the ID group, and both expressive and receptive language in the TD group, the final model only consisted of nonverbal mental age contributing significantly to the variance (see Table 4.4).

Discussion

There is growing interest in studying early language acquisition in toddlers with ASD. Although language is often extremely impaired in older children with ASD and associated intellectual disability (ID), less is known about language profiles and related factors in this low-functioning group with ASD. The first purpose of our study was to examine within- and between-group differences regarding receptive and expressive language in children with autistic disorder (AD) and ID, children with ID without ASD, and typically developing children. Nonverbal mental age was comparable across groups. The second purpose of this study was to examine the contribution of nonverbal

cognitive level of functioning and two important precursors of language, joint attention and symbol formation, to concurrent language abilities.

Several key findings emerged from this study of language profiles in low-functioning children with autistic disorder. First, lower scores for both receptive and expressive language were found in the AD group compared to the other groups, while groups scored the same on nonverbal mental age. These results confirm the presence of severe communication problems in low-functioning children with ASD (Noens & Van Berckelaer-Onnes, 2004). Second, a substantial discrepancy between expressive and receptive language skills on the one hand and nonverbal cognitive level on the other was found in the group of low-functioning children with autistic disorder. In contrast, in both comparison groups language levels approximated the nonverbal cognitive level, except for expressive language in the ID group. Hence, developmental profiles in low-functioning children with ASD were more uneven than in both comparison groups. Third, language profiles differed significantly across the three groups. Children with AD and ID achieved an expressive language level above their receptive language level, whereas children in the ID and the TD groups showed the opposite profile. However, not in all cases with ASD and ID receptive language was relatively more impaired than expressive language. Approximately half of the group achieved equal levels for receptive as for expressive language, but a substantial part showed a considerably lower level for comprehension than for production. All findings about language profiles are in line with the results for toddlers with ASD (e.g., Hudry et al., 2010; Weismer et al., 2010). However, studies in older high-functioning children with ASD found no differences between expressive and receptive language abilities (Jarrold et al., 1997; Kjelgaard & Tager-Flusberg, 2001). These previous results, combined with the results of this study, suggest a discrepancy in language profiles between high and low-functioning children with ASD.

The second purpose of this study was to examine the relation between nonverbal mental age, social and symbolic skills with concurrent language abilities. The results suggest that joint attention, symbol understanding, and nonverbal mental age share a substantial amount of variance in the TD and ID groups. However, in the low-functioning AD group other skills made a significant and unique contribution to language outcome. In the TD and ID groups nonverbal mental age was the most robust concurrent predictor of both language comprehension and production. Although nonverbal mental age was correlated with expressive and receptive language skills in the AD group, it did not remain significant in the final regression models, where joint attention and symbol understanding were the major predictors. In general, the role of joint attention in predicting language skills is consistent with previous findings in toddlers with ASD (e.g., Charman et al., 2003; McDuffie et al., 2005). However, the literature reported mixed findings regarding the association between joint attention and language abilities, with several studies indicating joint attention as a stronger

predictor for expressive than receptive language (e.g., Watt et al., 2006), but other studies reporting opposite results (e.g., Luyster et al., 2008).

The current data suggest that symbol understanding plays an important role in both expressive and receptive language development in low-functioning children with AD. Nonverbal children with ASD also have difficulties in understanding the symbolic nature of pictures. Therefore, impairments in symbol formation could be a fundamental deficit underlying problems in language development in children with ASD (Bates, 1979; Travis & Sigman, 2001). The nature of the relationship between joint attention and symbol formation and the relative contribution of both precursors to language development remain unclear. Joint attention is supportive, but not necessary for the acquisition of symbols (Travis & Sigman, 2001). On the basis of a longitudinal study in young children with ASD, Toth and colleagues (2006) suggest that joint attention seems to be an important skill for the start of language development, while symbolic play - which refers to more representational or symbolic skills - is associated with the continued development and the expansion of language abilities in later years.

A limitation of our study is the use of different instruments for assessing receptive and expressive language abilities, involving both direct assessment and parent questionnaires. The parental reports were used to correct the floor effects of the direct assessment measures in the lowest functioning children in our study. This strategy was chosen, because several previous studies have suggested that there is a very close agreement among these different language measures (e.g., Fenson et al., 1994; Luyster et al., 2008; Weismer et al., 2010; Zwaigenbaum et al., 2005). No instruments were available which evaluate language comprehension and production for the whole age range of the participants included in this study. The Reynell / Schlichting and the D-CDI have the same measurement goal, i.e. assessment of receptive and expressive language, but for a different age range, and all instruments result in an age equivalent score.

The results of our study have clinical relevance with respect to assessment and intervention regarding language abilities in low-functioning children with ASD. Regarding language assessment, our findings indicate that all children with ASD and ID demonstrate significant delays in receptive and expressive language abilities, even relative to their nonverbal cognitive level. Assessment should always include language measurements, in order to elicit strengths and weaknesses so that intervention targets may be adjusted to individual needs. Moreover, both expressive and receptive language abilities need attention separately, because substantial and varying discrepancies might be present. Many low-functioning children with ASD have receptive skills that are more impaired than their expressive language skills. Particularly in the case of more verbal children, communication partners will be misled by the level of expressive language abilities, assuming a comparable level of comprehensions skills (Hudry et al., 2010; Noens & Van Berckelaer-Onnes, 2004). In typical development, comprehension always precedes language production. From a developmental perspective, parents, teachers,

and caregivers are used to simplifying their language. They often use words and sentence structures just above the expressive language level of children in order to encourage development. Consequently, language comprehension in low-functioning children with ASD is often overestimated, when they are approached at the level of their expressive language (Hudry et al., 2010). Several previous studies demonstrated a significant relation between communication difficulties and the severity of challenging behavior in low-functioning individuals (Bott et al., 1997; Chamberlain et al., 1993; McClintock et al., 2003). The study of Sigafoos (2000) showed that more severe deficits in receptive language compared to expressive language are associated with challenging behavior. Problem behaviors may serve different functions (Day et al., 1994; Hanley et al., 2003; Reese et al., 2005). Some specific behavior problems might be related to receptive communication problems (e.g., to escape too difficult demands) and other to expressive communication problem (e.g., to get a desirable object which is not within reach). Unfortunately, little attention has so far been given to comprehension problems in interventions for children with ASD (Kevan, 2003; Sigafoos, 2000). Interventions and approaches should have improvement and support of language comprehension as a key target; interventions which only aim for expressive language improvement will widen the gap between comprehension and production. Further, the findings from our study have implications for designing interventions to improve language comprehension. Pictures or objects are often used in a symbolic and representational way to support communication in children with ASD (Wendt, 2009). This strategy is problematic when language impairments are accompanied by impairments in symbolic understanding of pictures. For these children, augmentative communication adapted to their level of sense making is a recommended treatment strategy (Noens & Van Berckelaer-Onnes 2004). Facilitating joint attention development is also an important target for intervention in low-functioning children with ASD. Several studies have described techniques that may be effective (e.g. Kasari et al., 2008; Siller & Sigman, 2002). Future research will need to examine the relation between symbolic understanding and language development more closely in younger children with ASD, as well as in ASD children with different levels of ID. It is important to differentiate between factors contributing to the emergence and expansion of language skills.

5

Sense-making in low-functioning children with autism

Jarymke Maljaars
Ilse Noens
Evert Scholte
Ina van Berckelaer-Onnes

Research in Autism Spectrum Disorders (2011), *6*, 806-814

Level of sense-making in children with autistic disorder and intellectual disability: Patterns of delay and deviance in development

Abstract

The present study examined levels of sense-making in relation to adaptive functioning and autism symptomatology in low-functioning children with autistic disorder. Thirty-six children with autistic disorder and intellectual disability were compared with 27 children with intellectual disability and 33 typically developing children with a comparable nonverbal mental age (2-5 years). Level of sense-making was measured with the ComFor. Delays and deviant behaviors were assessed by using the Vineland Screener 0-6-NL and the Diagnostic Interview for Social and Communication Disorders. Levels of sense-making were substantially lower in the group with autistic disorder. At non-symbolic levels of sense-making, children with autistic disorder and intellectual disability were much lower functioning in terms of social and communicative adaptive behavior than the children in the comparison groups with the same level of sense-making. Within the autism group, lower levels of sense-making were associated with more severe autism symptomatology in the domains of social interaction, communication, and imagination.

Introduction

During the first few years of life most children achieve important milestones in symbolic development (DeLoache, 2004). They gradually learn to understand and use symbols, such as words and pictures (Callaghan, 2000). Symbolic knowledge requires understanding of symbol-referent relations, even if the referent is not currently present. Symbolic development is important to increase communication opportunities, as symbols enable people to exchange information efficiently by representing something else even beyond the here and now (Namy & Waxman, 2005). Problems in symbol formation are considered to be a core deficit in children with autism spectrum disorder (ASD) (Noens & Van Berckelaer-Onnes, 2005; Travis & Sigman, 2001, Wetherby et al., 2000). Many studies focused on children with ASD with symbolic communication; children who have not achieved symbol understanding yet have seldom been evaluated (Granlund & Olsson, 1999). Especially in children with ASD and an associated intellectual disability (ID) understanding of symbol-referent relations does not develop in all cases (Preissler, 2008).

Noens and Van Berckelaer-Onnes (2005) conceptualized communication problems in ASD as a problem in 'sense-making', the perception of meaning. Sense-making refers to the cognitive process underlying receptive and expressive communication. The level of sense-making describes the way someone perceives and

interprets incoming stimuli gathered by different senses. Since children with ASD often find it difficult to integrate several pieces of information (Happé & Booth, 2008), it is very likely that they may fail to encode and relate all of the available cues of communication. Comprehension and production of symbolic communication appeals to complex information processing abilities, because various stimuli need to be integrated simultaneously and interpreted in a specific context.

Communication and sense-making can take place at the following four levels: sensation, presentation, representation, and metarepresentation (Verpoorten, 1996). At the level of *sensation*, communication consists of sensory experiences through different senses, such as vision and hearing, but also smell, taste, touch, balance, and kinesthesia. A regularly offered sensory stimulus can appear familiar, but has no functional or symbolic meaning, which is congruent to the developmental level of young infants (Noens & Van Berckelaer-Onnes, 2004). Individuals with ASD who communicate at the level of sensation only, often have a severe to profound ID. Lack of any use and understanding of language is one of the main characteristics of this level.

At the level of *presentation*, one perceives information within a concrete context. The individual understands the functional meaning of objects when they are literally present. At this level, children start to use conventional means of communication, such as gestures and words, but still in a non-symbolic way (Noens & Van Berckelaer-Onnes, 2004). A word or a gesture is used in a specific context where the relationship between the sign and the referent is limited to that context (Stephenson & Linfoot, 1996). For the transition from presentation to representation, generalization and decontextualization are needed to be able to use symbolic means without contextual constraints. This process requires the ability to take a symbol one has learned in one situation and apply it in a new and different context (DeLoache, 1989). It includes for example applying the word to a variety of referents, even when the referents are absent, or using the word with different listeners. Only when a word is generalized beyond the initial learning environment, it may be granted as symbol use and understanding (Namy & Waxman, 2005). This is considered to be the same for the use of pictures. The capacity to match a picture to a referent in one restricted context or to label a picture is also context-bound (Stephenson & Linfoot, 1996). Generally, in typical development, this transition from presentation to representation or presymbolic to symbolic functioning takes up a very short period of a couple of months (Wilkinson & McIlvane, 2001). From studies in children with ASD, it can be found that such limitations in symbol use can be observed for more extended time periods (Wetherby et al., 2000). In case of low-functioning children with ASD, true symbolic capacity and communication may not develop at all (McLean, 1993; Noens & Van Berckelaer-Onnes, 2004). When low-functioning children with ASD do not reach the level of representation, this is also reflected in other problems such as lack of symbolic play and delayed imitation (Blanc et al., 2005).

The level of *representation* is achieved when someone understands the full referential or symbolic function of communication. Symbol formation and object permanence are necessary to achieve the level of representation (Noens & Van Berckelaer-Onnes, 2004). Verbal children with ASD generally communicate at the level of representation, except for children with speech mainly consisting of echolalia (Wetherby et al., 2000). Communication at the level of representation can be characterized as symbolic communication. Language is a complex symbolic communication system, for which symbol-referent relations are arbitrary and conventional. Some non-verbal children with ASD are able to understand and use gestures, objects, or pictures as symbols. In that case, the level of representation has also been reached (Noens & Van Berckelaer-Onnes, 2004).

At the level of *metarepresentation*, information beyond the literal meaning or the primary information is perceived. Human language is interlarded with metarepresentations, such as proverbs and metaphors (Noens et al., 2006). Even for verbal children with ASD, sense-making is often fragmentary or literal. Generally, sense-making at the level of metarepresentation will be less flexible or will be missed, unless the meaning behind the words is directly taught (Twachtman-Cullen, 1998).

So far, no studies sought to investigate level of sense-making in relation to the core behavioral characteristics of ASD. Since ASD is considered as a developmental disorder, it is important to make a distinction between delayed and deviant patterns of development, in particular in relation to adaptive functioning and autism symptomatology (Klin et al., 2007). In ASD, both delays and deviances are present, but the severity of symptoms varies. The aim of the present study was to give more insight into the level of sense-making in low-functioning children with AD in order to formulate recommendations for assessment and intervention for this vulnerable group. To this end, levels of sense-making of a group of children with AD and ID were compared to groups of children with ID without ASD and typically developing children. Also, the relationships between level of sense-making and clinical presentation of adaptive functioning and autism symptomatology in low-functioning children with AD were examined.

Method

Participants

A group of 36 children with AD and ID participated in this study. Participants were included in the AD+ID group if they had received a formal AD and ID classification according to DSM-IV-TR criteria (APA, 2000) prior to the study. All children met diagnostic criteria for AD according to the Autism Diagnostic Observation Schedule Modules 1 or 2 (ADOS; Gotham et al., 2007). Given the classification difficulties in low-functioning individuals, this study focused on the core syndrome instead of the whole

spectrum to create a more homogeneous group. The mean nonverbal mental age of the children with AD and ID was 3;3 years (range 2;0 - 5;8 years). The comparison groups consisted of 27 children with ID without ASD and 33 typically developing children (TD). Children in the ID and TD groups did not meet diagnostic criteria for ASD on the ADOS. Children in the ID group all have a formal ID classification (APA, 2000). Levels of ID in the groups with and without autism ranged from mild to severe (APA, 2000). Results on a Dutch nonverbal intelligence test (SON-R 2½-7; Tellegen et al., 1998) confirmed that IQ-scores of participants in both groups were below 70. The main characteristics of the groups are presented in Table 5.1.

Table 5.1 ***Main characteristics of the subsamples***

		Nonverbal mental age equivalent (in months)			**Chronological age** (in months)			**Gender** (in %)	
Groups	*n*	Range	*M*	*SD*	Range	*M*	*SD*	Male	Female
AD+ID	36	<24 [a] - 68	39.0	13.71	41 - 136	85.6	27.86	89	11
ID	27	<24 [a] - 63	39.1	12.71	50 - 132	80.6	19.12	61	39
TD	33	29 - 65	42.7	9.42	24 - 50	37.9	6.36	41	59

Note. AD = autistic disorder; ID = intellectual disability; TD = typical development
[a] minimum score

The TD children were significantly younger than the children in the other two groups ($F_{(2,93)}$ = 55.55, p < .001), but all groups were comparable with respect to nonverbal mental age ($F_{(2,93)}$ = 1.02, p = .37), since groups were matched on the latter variable. In the AD+ID group significantly more boys were present than in the two comparison groups, $\chi^2_{(2)}$ = 18.50, p < .001. There were no differences in outcome variables (level of sense-making, adaptive functioning, and autism symptomatology) between boys and girls within the three subsamples (p > .05).

Instruments

SON-R 2½-7. To measure nonverbal mental age, a Dutch test for nonverbal intelligence, the SON-R 2½-7 (Tellegen et al., 1998) was used. Total raw scores were converted into nonverbal mental age equivalents. The SON-R 2½-7 has been found reliable and valid (Evers et al., 2009).

ComFor. The ComFor is a clinical instrument for the indication of augmentative communication in people with ASD and ID, which evaluates visual perception and sense-making (Verpoorten et al., 2004, 2008). The target group of the ComFor consists of individuals with ASD and ID, with limited or no verbal communication and a developmental level of approximately 12 to 60 months. The ComFor addresses two core questions: first, the most suitable form of augmentation, and second, the level of sense-making at which the forms chosen can be offered. With respect to the level of sense-making, three general indications are possible: sensation, presentation, and

representation. Sense-making is made operational in the following way. At the level of presentation, identical objects or pictures have to be sorted according to shape, color, matter, and size. Thus, the tasks can be resolved on the basis of concrete, literally perceptible features. At the level of representation, non-identical objects or pictures have to be sorted on the base of sense-making beyond the concrete, literally perceptible features. The level of presentation consists of three series with a total of 23 sorting tasks and the level of representation comprises two series with a total of 13 sorting tasks. In series 1 sorting is first trained by different assembling tasks. Based on the profile of individual scores on all ComFor items an indication of level of sense-making can be given. Both reliability and validity of the ComFor were found to be satisfactory. The coefficients for inter-rater reliability, test-retest reliability, and internal consistency are .80 or higher. Analyses of internal structure, convergent and divergent patterns have shown adequate construct validity; criterion-related validity is not yet determined (Noens et al., 2006).

Vineland Screener 0-6. The Vineland Screener 0-6yrs-NL is a questionnaire which can be used to determine the level of adaptive functioning in age equivalents. This screener is adapted from the American Vineland Screeners, versions 0-2;11 and 3-5;11, developed by Sparrow et al. (1993) and standardized on a Dutch sample. Based on 72 items, this questionnaire distinguishes four domains of adaptive functioning: communication, daily living skills, socialization, and motor skills. The psychometric properties of reliability and validity for the Dutch version are good (Scholte et al., 2008).

DISCO. The Diagnostic Interview for Social and Communication Disorders – version 11 (DISCO-11) is a semi-structured interview to be administered with parents or other primary carers (Wing, 2006). The Dutch translation of the 11th version of the interview was used in this study (Van Berckelaer-Onnes et al., 2008). The instrument can be used to provide a comprehensive description of individual characteristics and is suitable for individuals of all ages, levels of abilities and the whole autism spectrum. Through the DISCO-11 information about developmental history and a broad range of skills and behaviors can be gathered (Wing, 2006). The interview takes about two to three hours to administer and consists of more than 300 questions grouped under different sections. Only section 7 about the quality of social interaction, communication, imagination, and activities was used for the analyses. The DISCO distinguishes both 'ever' and 'current' ratings of the child's behavior for all items, but only 'current' items and classifications were analyzed in the present study. The quality of social interaction, communication, imagination, and pattern of activities is rated by choosing the type and description that is the most characteristic for a person. With regard to the reliability, the inter-rater reliability proved to be high (Wing et al., 2002).

Procedure

Children with ID with and without autism were recruited from several special day care centers and schools for children with ID. The sample of typically developing children was obtained from regular day care centers. After written informed consent was received, parents were asked to complete the Vineland Screener 0-6. Subsequently, participants were individually administered a non-verbal intelligence test and the ComFor at their day care or school in three or four visits. The child and one of the parents were invited to visit the university, school or day care of the child, where the ADOS was administered. The DISCO interviews were conducted by seven different interview couples at the parents' home. For three children in the AD+ID group, the DISCO could not be administered due to refusal or insufficient Dutch language abilities of the parents. The interviewers were blind to the levels of functioning and sense-making of the children. All data were analyzed using SPSS 18.0. Considering that many variables were not distributed normally and that the groups, particularly subgroups within the AD+ID group, were relatively small, analyses were conducted using non-parametric tests. Chi-square tests were used to compare the distribution of level of sense-making between the three groups. Further, adaptive levels of functioning and autism characteristics were compared using Kruskal-Wallis tests. Mann-Whitney *U*-tests were applied for post hoc comparisons with adjusted *p*-values according to the Bonferroni method (adjusted *p*-value is .017). Typology of autism characteristics was only examined in the AD+ID group.

Results

Levels of sense-making

Table 5.2 illustrates the percentages of participants in each group in relation to level of sense-making as measured with the ComFor. A Chi-square test indicated a significant association between level of sense-making and diagnosis ($\chi^2_{(4)} = 11.41$, $p = .02$).

Table 5.2 ***Cross tabulation of groups versus level of sense-making***

		Level of sense-making			
Groups		Sensation	Presentation	Representation	Total
AD+ID	*n*	9	13	14	36
	%	25%	36%	39%	100%
ID	*n*	2	11	14	27
	%	7%	41%	52%	100%
TD	*n*	0	14	19	33
	%	0%	42%	58%	100%

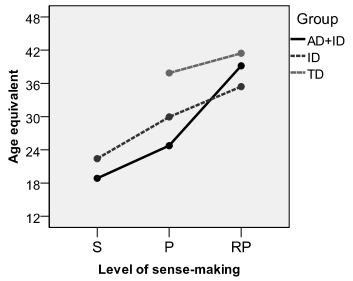
The distribution of level of sense-making is comparable for the ID and TD groups. About half of the participants have reached the level of representation, and the

other half were grouped at the level of presentation or some in the ID group at the level of sensation. A fourth part of the AD+ID group perceives the world at the level of sense-making, 36% at the level of presentation and 39% at the level of representation. It is important to notice that nonverbal mental age was comparable across all subgroups for each level of sense-making ($p > .05$).

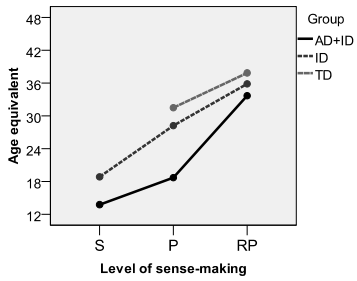
Levels of sense-making and adaptive functioning

Adaptive skills were compared within and between groups according to level of sense-making (see Figures 5.1a, 5.1b, and 5.1c). Kruskal-Wallis tests were conducted to compare mean levels of adaptive functioning between levels of sense-making within the AD+ID group. Significant differences were found for Communication ($H_{(2)} = 12.59$, $p = .002$), Socialization ($H_{(2)} = 11.29$, $p = .004$), and Daily Living Skills ($H_{(2)} = 10.51$, $p = .005$), between low-functioning children with AD at the level of sensation or presentation versus the level of representation. Within the ID group and TD group a comparison was made between the levels of presentation and representation. Mann-Whitney *U*-tests revealed significant differences for the domain of Daily Livings Skills for children at the level of presentation versus the level of representation ($U = 33.0$, $p = .015$ and $U = 74.0$, $p = .031$, respectively).

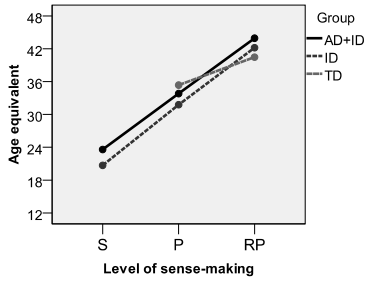
5.1a Communication



5.1b Socialization



5.1c Daily Living Skills



Note. S = sensation; P = presentation; RP = representation

Figures 5.1a, 5.1b, and 5.1c **Mean adaptive age equivalents based on level of sense-making for the three groups**

Kruskal-Wallis and Mann-Whitney *U*-tests showed that children with AD+ID at the level of presentation achieved significantly lower scores on the domains Communication ($H_{(2)} = 7.90$, $p = .019$) and Socialization ($H_{(2)} = 9.83$, $p = .007$), but not on Daily Living Skills ($H_{(2)} = 0.39$, $p = .822$) in comparison with children with ID and TD at the same level of sense-making. For the Communication domain, only the difference between the AD+ID group and the TD group turned out to be significant ($U = 40.0$, $p = .013$). At the level of representation, the AD+ID group showed comparable levels of adaptive functioning to the ID and TD groups on all three domains. Due to the lack of comparison groups, no analyses could be performed for the level of sensation.

Level of sense-making and autism symptomatology

Quality of social interaction, communication, imagination, and patterns of activity as measured with the DISCO-11 were compared across the different levels of sense-making within the AD+ID group (Table 5.3). Ratings for quality are on an ordinal scale and compared using Kruskal-Wallis tests. In all domains higher ratings indicated relatively higher quality of skills.

Table 5.3 ***Rates and percentages of quality of social interaction, communication, imagination, and pattern of activities for AD+ID-subgroups based on level of sense-making***

	Level of sense-making					
	Sensation (n = 8)		Presentation (n = 12)		Representation (n = 13)	
DISCO part 7 [a]	*n*	%	*n*	%	*n*	%
Quality of social interaction						
0. Does not interact; aloof and indifferent	1	12	0	0	0	0
1. Interacts to obtain needs, otherwise indifferent	6	76	6	50	4	31
2. Generally does not initiate, but responds to social contact; joins in passively	1	12	3	25	2	15
3. Makes social approaches actively, but usually inappropriate, 'one-sided', naive and peculiar	0	0	3	25	7	54
Quality of communication						
0. No social communication	1	13	2	17	0	0
1. Communicates only / mainly own needs	7	87	4	33	3	23
2. Adequate language; no wish to engage in communication; only brief answers or essential comments	0	0	2	17	1	8
3. Communicates regarding his / her own interests	0	0	4	33	9	69
Quality of imagination						
0. No pretend / role play or imaginative activities	7	88	8	66	3	24
1. Some pretend / role play, but specifically taught	0	0	2	17	2	15
2. Pretend / role play is copied from others	0	0	2	17	4	31
3. Self-invented pretend / role play, but repetitive in theme / content	1	12	0	0	2	15
4. Pretend / role play appears to be shared with others, but restricted to own themes / rules	0	0	0	0	2	15
Quality of pattern of activities						
0. Only repetitive activities	4	50	2	16	3	23
1. Some varied interests / activities	4	50	8	68	7	54
2. Varied and flexible activities	0	0	2	16	3	23

[a] Only ratings seen in ASD are listed

Significant differences between levels of sense-making were found for Quality of Social Interaction ($H_{(2)}$ = 10.42, p = .005), Communication ($\chi^2_{(2)}$ = 10.85, p = .004), and Imagination ($H_{(2)}$ = 9.56, p = .008). Quality of Social Interaction was significantly worse at the level of sensation compared to presentation (U = 21.0, p = .014) and representation (U = 14.0, p = .002). For Quality of Communication children at the level of sensation achieved significantly lower scores than children at the level of representation (U = 10.5, p = .001). Children at the levels of sensation and presentation showed significantly less Quality of Imagination than at the level of representation (U = 20.5, p = .015 and U = 34.0, p = .012, respectively). No significant differences were found for Quality of Patterns of Activities ($H_{(2)}$ = 3.59, p = .166).

Discussion

The way someone perceives and interprets incoming stimuli defines the level of sense-making or perception of meaning. The level of sense-making of visual stimuli can be determined by using the ComFor. Based on the ComFor results a clinical and individualized indication of an augmentative communication strategy can be provided (Noens et al., 2006). In addition to the ComFor results, it is also important to take other developmental and behavioral aspects in account in order to adapt the intervention strategy to individual needs. Furthermore, sense-making plays an important role in different areas of development such as communication, but also social interaction and imagination (Noens & Van Berckelaer-Onnes, 2004). It is important to study level of sense-making, because it determines the most suitable level of approach in daily life situations. In the present study, we characterized level of sense-making and its relation with adaptive functioning and autism symptomatology in low-functioning children with AD. It is important to make a distinction between delayed and deviant patterns of development. The first approach compares the child with the normative course of development and the second approach places the child in a dimensional continuum of severity of autism symptomatology (Klin et al., 2005). Relating adaptive functioning and autism symptomatology to the level of sense-making provides the opportunity to give more specific recommendations for interventions. In this way, interventions can be adapted to abilities and disabilities of low-functioning children with AD, because each level of sense-making requires particular communication strategies.

First, the results of our study indicate that difficulties in sense-making are more pervasive in children with AD and ID compared to typically developing children, but also compared to children with ID without ASD with a comparable nonverbal mental age. In the group with AD significantly fewer children achieved the level of representation. A large part of this group is only able to perceive the world at the level of sensation or presentation, which implies that they do not (completely) understand symbolic or representational means of communication. It must be noted that the ComFor only evaluates visual perception and sense-making. From the studies of DeLoache (2004) and DeLoache and Burns (2004) it is known that in typical development the understanding of language precedes the representational understanding of pictures. The findings from the present study confirm the hypothesis of Noens and colleagues (2006) that in children with AD the development of sense-making evolves at a slower pace or in a different way. In case of AD, the transitions from sensation to presentation and from presentation to representation possibly take place in a later stage of development compared to children without ASD. However, longitudinal research is necessary to validate this hypothesis. A complicating factor is that there is no consensus in literature when symbolic capacities are completely developed. Generalization and decontextualization are necessary to speak about symbolic use and understanding of language or pictures, but there are no guidelines as to how much generalization and

decontextualization is needed to identify true symbolic behavior (Wilkinson & McIlvane, 2001). As a result, the duration of the transitional phase before the level of representation is difficult to determine, also in typical development. Nevertheless, there are clear indications that children with ASD have more difficulties with generalization and decontextualization (e.g., MacDuff et al., 1993), as reflected in the distribution of level of sense-making within the groups in the present study.

Second, the results of the study showed that at the level of representation all three groups have comparable levels of adaptive behavior on all domains, also with respect to communication and socialization. This is consistent with other studies in individuals with ASD and co-occurring ID, which indicate that in this low-functioning group the overall level of adaptive functioning is fairly comparable to the level of intelligence (Bölte & Poustka, 2002), whereas in higher functioning individuals adaptive behavior is relatively more impaired (Liss et al., 2001; Perry et al., 2009). Other reports indicated that children with ASD and ID are relatively more impaired in the social domain than children with ID (Loveland & Kelley, 1991), but this is not supported by our results for children at the level of representation. However, the children with AD and ID at the levels of sensation and presentation are much lower functioning on social and communicative functioning compared to the control groups with the same level of sense-making. In other words, mainly children at the lower levels of sense-making are impaired in adaptive functioning in comparison with their nonverbal mental age matched controls. This implies that at these levels of sense-making developmental level does not entirely determine adaptive scores on the domains of communication and socialization. Thus, level of sense-making is an important underlying factor. This could possibly suggest that low-functioning children with AD have problems in translating their cognitive potential into real-life skills (Klin et al., 2007). An explanation for this finding is that particularly these aspects of adaptive functioning interfere with autism symptomatology or the lack of symbolic skills at the lower levels of sense-making. Another explanation is that it might be difficult for the environment to give instructions and provide interventions to improve adaptive behavior adjusted to these lower levels of sense-making.

Third, level of sense-making is related to autism symptomatology for the domains of communication, social interaction, and imagination. Lower levels of sense-making are associated with more severe problems in those domains and vice versa. Children making sense at the level of sensation are often indifferent in social contact, communicate only to obtain their needs, and have no symbolic play or imaginative activities. At the level of presentation, more variation in types of social interaction, communication, and imagination was observed, but the majority was still very impaired. According to the social typologies defined by Wing and Gould (1979), children making sense at the levels of sensation and presentation might be characterized as aloof or passive. At the level of representation, the subtypes aloof and passive are still present, but to a smaller extent. Half of the group can be characterized as active but

odd. They accept or search for social interaction, but interact in an odd or unconventional way. A greater frequency of initiation of social interaction and communication, as well as more symbolic play are seen in this group. The results are in line with previous studies which found that severity of autism symptomatology or social typologies were related to level of functioning (e.g., Joseph et al., 2002; Volkmar et al., 1989).

Some limitations of our study have to be acknowledged. First, the present study examined sense-making in young children with AD, thus additional research would be necessary to extend the findings to older samples with AD and ID and to samples with other ASDs. Another limitation is that the sample of children with AD and ID is rather small when subdivided by level of sense-making in the analyses regarding adaptive functioning and autism symptomatology. Further, a screener version was used to evaluate adaptive level of functioning, whereas the extended interview version would provide a more differentiated picture of adaptive functioning. This study extends previous studies by examining level of sense-making and differentiating between symbolic and non-symbolic communicators regarding the clinical manifestation of ASD. Moreover, only children with a co-occurring ID were included in the AD group, while other studies often use groups of mixed levels of IQ or only higher functioning children.

This research has a number of important implications for clinical practice with respect to assessment and intervention strategies. The findings from the current study suggest that clinicians would benefit from evaluating level of sense-making in children with AD and ID, not only because sense-making might be weaker than expected on the basis of their nonverbal mental age, but also because sense-making is an important underlying factor for social and communicative functioning. The most common communication strategies, naturalistic as well as augmentative, presume symbolic abilities. Augmentative communication is also possible at the levels of sensation or presentation, provided that pictures, objects, or other forms of communication are used in a different way (see Noens & Van Berckelaer-Onnes, 2004). Visualizations might be helpful, since visual perception is a strength of many children with ASD (Quill, 1997). Sometimes visualizations are not sufficient, in that case tactile or other sensory strategies need to be implemented. Communication tailored to the individual level of sense-making is crucial to improve quality of life, because overestimation and limited comprehension of communication may result in poorer outcome results and challenging behaviors (Bradshaw, 2002). As level of sense-making plays an important role in level and quality of social and communicative functioning, level of sense-making need to be improved within the limits of the individual's possibilities. Furthermore, adjustments of the environment as per level of sense-making can offer new opportunities of change in other areas of functioning as well.

The profiles of adaptive behavior and typologies of autism characteristics indicate that, particularly at the levels of sensation and presentation, improving social, communication, and play skills are important goals for intervention in low-functioning

children with AD. At the level of sensation it is impossible to announce activities or to explain different steps within an activity, by using verbal instructions or pictures. At this level, the environment and all activities need to be highly structured and caregivers should stick to fixed routines and activity scenarios. Communication at the level of presentation is limited to the 'here and now' (Noens & Van Berckelaer-Onnes, 2004). Therefore, more independence in daily life skills or more initiation in communication and social interaction can be achieved by using non-symbolic communication strategies in the situation itself. Routines to stimulate initiation of social interaction and communication can be set up around daily routines. The use of pictures as symbols is inadvisable at this level of sense-making. Previous studies showed a difference between concept identification and concept formation abilities in children with ASD (e.g., Minshew et al., 2002). Some children may be able to label a picture, but this does not mean that the child can use the picture in a representational communicative way (Twachtman-Cullen & Twachtman-Reilly, 2007). In case of a child at the level of representation with adaptive functioning below its mental age matched peers, intervention can focus on more independence in several daily life situations. These situations can be explained and practiced by using augmentative communication at the level of representation, for example visual schedules or protocols to indicate the different steps or task components. Due to their non-transient nature, such schedules make instructions clear, help the child to stay focused, and diminish the need for adult support and reinforcement (Wendt, 2009).

In conclusion, difficulties in sense-making are more pervasive in children with AD and ID, hence increasing their need for support in daily life and interventions adapted to their level of sense-making. Future research needs to determine the effectiveness of augmentative communication to improve communication, social interaction, and daily life skills in low-functioning children with AD. Although visualizations, such as visual schedules within and between activities, are used very often in home, institution, and school settings, research in this area is surprisingly scarce (Wendt, 2009).

6

Visual perception in low-functioning individuals with autism and deafness

Jarymke Maljaars
Ilse Noens
Roger Verpoorten
Evert Scholte
Ina van Berckelaer-Onnes

Journal of Intellectual Disability Research (2011), *55*, 95-105

Visual local and global processing in low-functioning deaf individuals with and without autism spectrum disorder

Abstract

The ComFor study has indicated that individuals with intellectual disability (ID) and autism spectrum disorder (ASD) show enhanced visual local processing compared with individuals with ID only. Items of the ComFor with meaningless materials provided the best discrimination between the two samples. These results can be explained by the weak central coherence account. The main focus of the present study is to examine whether enhanced visual perception is also present in low-functioning deaf individuals with and without ASD compared with individuals with ID, and to evaluate the underlying cognitive style in deaf and hearing individuals with ASD. Different sorting tasks (selected from the ComFor) were administered from four subsamples: (1) individuals with ID (n = 68), (2) individuals with ID and ASD (n = 72), (3) individuals with ID and deafness (n = 22), and (4) individuals with ID, ASD and deafness (n = 15). Differences in performance on sorting tasks with meaningful and meaningless materials between the four subgroups were analyzed. Age and level of functioning were taken into account. Analyses of covariance revealed that results of deaf individuals with ID and ASD are in line with the results of hearing individuals with ID and ASD. Both groups showed enhanced visual perception, especially on meaningless sorting tasks, when compared with hearing individuals with ID, but not compared with deaf individuals with ID. In ASD either with or without deafness, enhanced visual perception for meaningless information can be understood within the framework of the central coherence theory, whereas in deafness, enhancement in visual perception might be due to a more generally enhanced visual perception as a result of auditory deprivation.

Introduction

The debate on cognitive theories explaining behavior and finding indications for possible brain dysfunctions in autism spectrum disorders (ASD) is still on-going. Frith (1989) introduced the concept of 'weak central coherence' (WCC). In ASD the drive for central coherence is rather weak, whereas in the normal cognitive system there is a natural tendency to integrate incoming stimuli and to interpret them as a whole based on context. This weaker drive for central coherence is reflected in both local superiority and global weaknesses (Happé & Booth, 2008). Local processing refers to featural and detail-focused information processing, whereas global processing is the ability to extract global form and meaning from incoming information (Happé & Frith, 2006). Alternative explanations like 'enhanced perceptual functioning' (EPF) emphasize enhanced low-level perception leading up to superior local processing without invoking

a deficit in global processing (Mottron et al., 2006; Wang et al., 2007). Nowadays, local and global processing are more and more considered as two independent mechanisms (Happé & Booth, 2008; Lopez et al., 2008). Despite robust findings of strong local processing in ASD, evidence of impaired global processing is mixed (Happé & Frith, 2006). However, in many studies it is difficult to disentangle local versus global preferences (Happé & Booth, 2008).

Local and global processing can be examined in the visual / visuo-spatial and auditory / verbal domain. Preferences in visual perception are often studied in high-functioning individuals with ASD. Despite the relatively high prevalence of intellectual disability (ID) in ASD (Fombonne, 2005; Goin-Kochel et al., 2008), the group of low-functioning individuals with ASD is generally neglected in research. Enhanced local perception in individuals with ASD and ID has been previously determined (Noens & Van Berckelaer-Onnes, 2008), but the specificity of this enhancement warrants further investigation. Therefore, in the present study visual perception is examined by comparing deaf and hearing individuals with ID with and without ASD. Furthermore, the underlying cognitive style of visual processing in deaf and hearing individuals with ASD is evaluated.

The style of visual processing in individuals with both ASD and ID has been investigated only incidentally. The study of Van Lang et al. (2006) showed that adolescents with ASD and a moderate or mild ID have a weaker drive for central coherence based on two cognitive tasks: Block Design and Child Embedded Figures Test, than IQ-matched adolescents without ASD. However, a recent study in individuals with mild ID found no difference in performance on the Block Design test between participants with high versus low scores on the Social Communication Questionnaire (Harris et al., 2007). These mixed results can possibly be due to differences in level of ID or the criteria used to compose the ASD group.

For individuals with moderate to severe ID, these standard cognitive tasks are often too complicated. In comparison with construction or search tasks, sorting tasks are simpler, because they are unambiguous in nature and no verbal instruction is needed. Even individuals with severe ID are capable of making same versus different judgments or can learn to make these (Serna et al., 1997). The perception of similarities or dissimilarities, as in sorting tasks, involves local and global processing (Förster, 2009). Local processing is helpful in sorting identical materials on perceptual differences, like shape and color (matching). A distinction can be made between matching tasks with meaningless and meaningful materials. Meaningless tasks can be resolved on the basis of local information processing only (shape discrimination). Meaningful tasks can be resolved in the same way, but global information processing can be helpful even though it is not necessary. In contrast, global processing is needed in sorting similar, but non-identical materials on the basis of broader concepts or categories (Noens & Van Berckelaer-Onnes, 2008). The items of the ComFor (Forerunners in Communication) (Verpoorten et al., 2004, 2008) give the opportunity to study visual perception in

individuals with moderate to severe ID, because of the use of different above-mentioned sorting tasks. The results of the ComFor study (Noens & Van Berckelaer-Onnes, 2008) revealed enhanced visual perception in individuals with ASD and ID compared with individuals with ID. In this study, enhancement is a relative term, as enhancement implies superior performance compared with individuals with ID without ASD, thus 'less impaired' and not an enhanced visual perception in comparison with the general population. Item analyses in the ComFor study showed better visual shape discrimination in individuals with ASD and ID compared with individuals with ID, especially in tasks with meaningless materials in which the establishment of meaning is not supportive to sort these materials. These findings are consistent with the WCC account. The present study concerns an extension of the previous ComFor study (Noens & Van Berckelaer-Onnes, 2008) and examines differences in visual perception in ID and ASD and other groups with ID as well. Moreover, the specificity of the WCC account for individuals with ASD and ID compared with other clinical groups is evaluated.

Deafness provides a unique opportunity to examine the influence of auditory deprivation on visual perception (Rettenbach et al., 1999). The question whether intact senses can compensate for a sensory impairment has often been a reason for research (Marschark, 2005). Half of the studies indicate improved visual performance, whereas other studies showed no compensation or even impairment in the remaining senses as well. Age of participants, attention to and localization of the visual stimuli seem to influence the visual performances of the participants (e.g., Bavelier et al., 2006, Dye et al., 2009). Because ID and deafness may have mutual causes, an increased prevalence of deafness in ID can be presumed (Denmark, 1994). The exact prevalence of auditory impairments in individuals with ID is difficult to determine (Evenhuis et al., 2004), but a recent study showed a prevalence of approximately 30%, which is two times higher than in the general population (Meuwese-Jongejeugd et al., 2006). As a result of the increased risks for both auditory impairments and ASD in ID, comorbidity is also expected to occur frequently. However, the prevalence is unknown, possibly because this group is often neglected in research and also complicated to assess (Vernon & Rhodes, 2009). Scarce epidemiological studies have shown that auditory problems are more common in ASD and vice versa than in the general population (e.g., Gallaudet Research Institute, 2007; Kielinen et al., 2004; Rosenhall et al., 1999).

The specificity of enhanced visual perception with meaningless materials in individuals with ASD and moderate to severe ID will be evaluated by investigating whether deaf individuals with ID (with and without ASD) also show an enhanced visual perception compared with individuals with ID. The group of individuals with ID is very heterogeneous and therefore it is important to study specific subgroups with ID. Different disorders can bring along different assets and deficits in functioning. Strengths or weaknesses in visual perception in low-functioning deaf individuals with and without ASD will have consequences for treatment. In the present study, visual perception is examined by comparing the results on sorting tasks of four subgroups:

individuals with ID, with ID and ASD, with ID and deafness, and with ID, ASD and deafness. As far as we know, this is the first study to explore visual perception in low-functioning deaf individuals with and without ASD.

The following hypotheses will be tested. First, both groups with deafness are expected to show enhanced visual perception in comparison with the ID group, as a result of auditory deprivation. Second, the results on meaningful and meaningless tasks of individuals with ID, ASD and deafness are expected to resemble the results of hearing individuals with ASD and ID, because ASD entails the same underlying cognitive style in both groups. Consistent with results of the previous ComFor study (Noens & Van Berckelaer-Onnes, 2008) individuals with ASD and ID are expected to show increased scores in comparison with the ID group when the same materials have to be sorted according to perceptual differences, because of strong local perception in ASD. In accordance with the WCC theory these increased scores are mainly driven by enhanced scores on tasks with meaningless materials where global processing is not helpful, but not on tasks with meaningful materials. No differences are expected in sorting tasks with different meaningful materials, because these materials only concern very simple global meanings. Third, in case of deafness without ASD an enhanced score on sorting tasks compared with ID is also to be expected, but not for meaningless or meaningful tasks specifically. Individuals with ASD benefit from relatively superior local processing, whereas individuals with deafness without ASD are expected to have a general enhanced visual perception compared with hearing individuals with ID.

Method

Participants

The total sample consists of 177 children and adults with an adaptive age between 12 and 60 months for the domain of daily living skills measured with the Vineland Screener 0-6yrs-NL (Scholte et al., 2008) (see Table 6.1).

Table 6.1 ***Main characteristics of the four subsamples***

		Adaptive age equivalent (in months)			**Chronological age** (in months)			**Gender** (in %)	
Subsamples	*n*	*M*	Range	*SD*	*M*	Range	*SD*	Male	Female
ID	68	42	25 - 53	7.3	181	73 - 580	143.3	48%	52%
ID+ASD	72	42	27 - 53	6.6	171	73 - 605	111.2	75%	25%
ID+D	22	43	26 - 53	7.6	290	97 - 590	124.7	68%	32%
ID+ASD+D	15	40	19 - 54	10.8	248	64 - 381	102.1	60%	40%

Note. ID = intellectual disability; ASD = autism spectrum disorder; D = deafness

The total sample comprises four subsamples: (1) individuals with ID (*n* = 68), (2) individuals with ID and ASD (*n* = 72), (3) individuals with ID and deafness (D) (*n* = 22), and (4) individuals with ID, ASD and D (*n* = 15). The developmental level for adaptive

functioning ranges from 19 to 54 months in groups 3 and 4. Groups 1 and 2 were selected from the data of the ComFor standardization study (Noens et al., 2006). The selection criteria for this study were both a developmental level consistent with groups 3 and 4 and a chronological age above 6 years old. All participants are recruited through institutions where ID (IQ < 70) is a condition for admittance and all participants have a moderate to severe ID based on their adaptive level of functioning on the Vineland Screener. ASD was classified according to the criteria of the Diagnostic and Statistical Manual of Mental Disorders, 4th edition, test revision (DSM-IV-TR) [Autistic Disorder or Pervasive Developmental Disorder - Not Otherwise Specified (PDD-NOS)] by qualified professionals (APA, 1994, 2000). Deaf participants suffer from a severe to profound hearing loss (> 70db loss in the better ear) and have no cochlear implant. Individuals with severe motor impairments and (uncorrected) visual impairments were excluded from all subsamples. The four groups are comparable with respect to adaptive age equivalent ($F_{(3,173)} = .43$, $p = .73$), but the groups with and without D differ in mean chronological age; the latter are significantly younger ($F_{(3,173)} = 6.29$, $p < .01$). There is also a difference in distribution of gender across the subsamples ($\chi^2_{(3)} = 10.83$, $p < .05$), but no significant differences were found between mean scores of men and women on the main sorting outcomes ($p > .05$).

Instruments

Vineland Screener 0-6. The Vineland Screener 0-6yrs-NL is a questionnaire which can be used to determine the level of adaptive functioning. This screener is adapted from the American Vineland Screeners, versions 0-2;11 and 3-5;11, developed by Sparrow et al. (1993) and recently standardized on a Dutch sample. In this questionnaire with 72 items, a distinction is made between four domains of adaptive functioning: Communication, Daily Living Skills, Socialization and Motor Skills (Scholte et al., 2008). In low-functioning individuals with ASD, the level of adaptive functioning is fairly comparable to the level of intelligence (Bölte & Poustka, 2002; Liss et al., 2001). Research with individuals with ASD and ID indicated the domain of Daily Living Skills as the best indicator of general IQ (Bölte & Poustka, 2002). Therefore, the adaptive age equivalent for the domain of Daily Living Skills was used to compare the developmental level of the four subsamples. The psychometric properties of reliability and validity for the Dutch version of the Vineland Screener are good (Scholte et al., 2008).

Sorting tasks. Sorting was first trained by completing different assembling tasks. The scores on the assembling tasks were not used to measure visual perception. Only participants who were able to complete the six assembling tasks were assessed by using the sorting tasks. The participants who failed on these items did not have the basic prerequisites for sorting. From the original sample ($N = 200$), 23 participants made one

or more mistakes during the assembling tasks, as they were too low functioning or unable to manipulate the materials adequately because of motor problems.

A selection of 18 different sorting tasks derived from the ComFor (Verpoorten et al., 2004, 2008) was used to measure visual perception (see Table 6.2).

Table 6.2 ***Short description of the sorting tasks***

Tasks	Item	Description
Assembling: prerequisite for sorting		
Objects	*1*	*Assembling balls in a closed box*
	2	*Assembling blocks in a closed box*
	3	*Assembling rings in a closed box*
	4	*Assembling rings and balls in closed boxes*
	5	*Assembling blocks, balls and rings in closed boxes*
	6	*Assembling blocks and balls in closed boxes, spoons in open box*
1. Sorting meaningful identical materials		
→ local information processing is satisfactory, but global information processing can be helpful		
1a. Objects	1	Sorting identical checkers, screws, little sticks
	2	Sorting identical spoons, knives, forks
	3	Sorting identical dessert, tea, soup spoons
1b. Pictures	1	Sorting identical black / white photographs of combing hair, eating, brushing teeth
	2	Sorting identical black / white photographs of one person reading, writing, drinking
	3	Sorting identical line drawings of combing hair, eating, brushing teeth
2. Sorting meaningless identical materials		
→ local information processing only		
2a. Objects	1	Sorting identical blue, yellow, red checkers
	2	Sorting identical small circles, big circles, ovals
	3	Sorting identical cubes, thin blocks, wide blocks
2b. Pictures	1	Sorting identical letters E, F, L
	2	Sorting identical numbers 25, 26, 29
	3	Sorting identical words bed, bad, kam (English: bed, bath, comb)
3. Sorting meaningful different materials		
→ global information processing, local information processing is contra productive		
3a. Objects	1	Sorting different balls, dolls, cars
	2	Sorting different spoons, knives, forks
	3	Sorting functionally related objects: toothbrush-toothpaste, pencil-notebook, fork-plate
3b. Pictures	1	Sorting photographs of woman, ball, car in different situations
	2	Sorting photographs of comb, toothbrush, spoon in different situations
	3	Sorting photographs of writing, combing hair, drinking in complex context with pen, comb, cup

Because there are no common used tasks or instruments to evaluate visual perception and WCC in individuals with moderate to severe ID, the tasks used in the ComFor offer a good alternative. As mentioned in the introduction, sorting tasks are very suitable to measure visual perception in individuals with moderate to severe ID. To complete a sorting task, the participant has to sort nine objects or pictures correctly in three boxes with one example in each of the boxes.

The sorting tasks were divided in three categories with each six items: meaningful identical materials, meaningless identical materials and meaningful different materials. Within these categories, a distinction is made between the subcategories objects and pictures. For each subcategory of sorting tasks, only the three most difficult items were selected. In this way, non-discriminating items and items which were not clearly meaningful or meaningless were not used in this study. In the first two categories of sorting tasks, the identical objects or pictures have to be

matched according to shape, color, matter and size. Thus, these tasks can be resolved on the basis of concrete, literally perceptible features. A further distinction can be made between tasks with meaningless and meaningful materials. Meaningless materials include for example black abstract shapes, numbers or small words. For most individuals with moderate to severe ID items with numbers and words are meaningless, because they are not familiar with written language and are not able to read, whereas examples of meaningful materials are pictures of utensils or simple actions, like drinking or combing hair. Meaningless and meaningful items can be resolved on the basis of local information processing only (e.g., shape discrimination), global information processing can only be helpful in meaningful items. In the third category, non-identical objects or pictures have to be sorted on the basis of sense-making beyond the concrete, literally perceptible features. To solve these tasks global information processing is indispensable, but it only concerns primary and functional meanings. In sorting different meaningful materials, a focus on local information is usually contra productive (Noens et al., 2006). The reliability of the measurements of the various categories is .76 and above (Cronbach's alpha).

Procedure

Participants were recruited from several institutions for persons with ID and/or hearing impairments in the Netherlands and Flanders (Belgium). First, informed consent was obtained from the parents. The caregivers of the participants in the institutions were asked to complete the Vineland Screener 0-6yrs-NL. Subsequently, all participants were assessed using the sorting tasks. Assessments were carried out in the participant's institution by one of the authors (JM) or trained master's students of the Leiden University. The administrations were all videotaped to allow inspection of assessment procedure and consensus scoring.

Data analyses

SPSS 16.0 was used to conduct the analyses. First, performances of the four groups on the total score for the sorting tasks were examined to determine a possibly enhanced visual perception in the subsamples with ASD and/or D in comparison with the subsample with ID. The scores were calculated by summing the item scores. Second, differences between the subgroups on meaningful and meaningless items were analyzed to evaluate differences in visual perception between the four subgroups and the underlying cognitive style in the subgroups with ASD. Differences in mean scores between the four groups were evaluated by conducting several analyses of covariance (ANCOVA) in combination with planned comparisons with two contrasts. The first contrast concerned the comparisons between the subsamples ID vs. ID+ASD, ID vs. ID+D and ID vs. ID+ASD+D, the second contrast between the subsamples ID+ASD vs. ID+D and ID+ASD+D vs. ID+D. Given the differences in mean chronological age between the groups and the variance in adaptive age equivalents (Daily Living Skills)

within the groups, these variables were included as covariates in the General Linear Model. The validation study of the ComFor revealed a significant relation between ComFor scores and the domain of Daily Living Skills (Noens et al., 2006). Gender was not used in the model, because it was not significantly related to the outcome variables ($p > .05$ for all analyses). Furthermore, the differences between tasks with objects or pictures were studied in more detail. A more stringent significance level of .01 was established throughout all analyses, because of the relatively large number of analyses of variance conducted.

Results

For all sorting tasks, the subsample with individuals with three classifications (ID+ASD+D) obtained the highest mean scores compared with the other subsamples. Furthermore, the subsamples with ID+D and ID+ASD performed also better than the subsample with ID only (see Table 6.3).

Table 6.3 ***Description of adjusted means of the ComFor-scores***

		Sorting identical materials						Sorting different materials	
		Total (12 items)		Meaningful (6 items)		Meaningless (6 items)		Meaningful (6 items)	
Subsamples	*n*	*M*	*SD*	*M*	*SD*	*M*	*SD*	*M*	*SD*
ID	68	5.2	.37	3.0	.19	2.1	.20	1.7	.22
ID+ASD	72	7.2 [b]	.36	3.7 [a]	.19	3.5 [b]	.20	2.3 [a]	.22
ID+D	22	6.4 [a]	.67	3.5 [a]	.35	2.9 [a]	.37	1.9 [a]	.41
ID+ASD+D	15	8.0 [b]	.79	4.2 [a]	.41	3.7 [b]	.43	2.1 [a]	.48

[a] no difference with ID; [b] significantly higher than ID, no difference with ID+D

The analyses of covariance revealed that both covariates, adaptive and chronological age, are significantly related to the results on the different sorting tasks ($p < .01$), with the exception of chronological age in relation to the sorting tasks concerning different meaningful materials ($p > .01$). There was also a significant effect of diagnosis on the total score for sorting identical materials ($F_{(3,166)} = 6.87$, $p < .001$). Planned contrasts revealed that the scores were significantly higher for the subsamples with ID+ASD and ID+ASD+D compared with ID ($p < .01$). The mean score of the subsample ID+D was not significantly different from the higher scores in the ID+ASD and ID+ASD+D groups, or from the lower score in the ID group ($p > .01$). This group seems to take a position in the middle of these groups. The differences between the groups on tasks with identical materials were significant for meaningless tasks ($F_{(3,166)} = 9.44$, $p < .001$) and planned contrasts showed the same significant differences as mentioned above for sorting identical materials ($p < .001$). No significant differences were found for meaningful tasks with identical materials ($F_{(3,166)} = 3.51$, $p > .01$) or different materials ($F_{(3,166)} = 1.04$, $p > .01$).

Discussion

The ComFor study (Noens & Van Berckelaer-Onnes, 2008) has indicated that individuals with ID and ASD show enhanced visual local processing compared with individuals with ID only. Item analyses revealed that this enhancement can be explained by WCC in ASD. The main objective of this study was to examine the performances on sorting tasks of the ComFor; first, to determine whether an enhanced visual perception is also present in low-functioning deaf individuals with and without ASD, and second, to evaluate the underlying cognitive style in the two subgroups with ASD.

The results from this study showed that in addition to individuals with ID and ASD, also deaf individuals with ID and ASD obtained a higher total score on the sorting tasks compared with individuals with ID, but not when compared with individuals with ID and deafness. These findings indicate enhanced visual perception in low-functioning individuals with ASD with and without deafness in comparison with individuals with the same adaptive level of functioning without ASD. The scores of the deaf groups indicate that visual perception is also enhanced in these groups, but the enhancement is less clear in the deaf group without ASD. Other studies into deafness mainly found differences in visual processing when visual stimuli were presented in the peripheral visual field (e.g., Bavelier et al., 2006; Dye et al., 2009; Reynolds, 1993), whereas the sorting tasks concerned both stimuli in the central and the peripheral visual field. One of the main reasons why some studies found no differences in or even worse visual sensitivity in the central field in deaf individuals concerns their increased visual distractibility by irrelevant peripheral stimuli (Dye et al., 2008). Hence, the ComFor was administered by one person in a separate, quiet and visually predictable environment allowing participants to focus upon the tasks at hand. However, all mentioned studies into visual perception and deafness are difficult to compare with this study because in all other studies only participants with cognitive abilities in the mean or even higher functioning range took part.

The evaluation of the total performance on the sorting tasks does not address the question which underlying cognitive style is responsible for the enhanced performances in the different groups with ASD. Therefore, differences in comparisons between groups for meaningless and meaningful items were taken into account. The group with ID and ASD obtained higher scores on meaningless items only compared with the ID group, which indicates enhanced local information processing alongside reduced global processing (Noens & Van Berckelaer-Onnes, 2008). All sorting tasks concerning identical materials could be sorted according to shape. To solve the meaningless items, global information processing and sense-making beyond literally perceptible features cannot offer any support and the tasks have to be sorted on the base of local information processing. Results could not be explained by good shape discrimination alone, otherwise the differences would be found in both meaningful and meaningless sorting tasks. Therefore, the findings of the analyses for meaningful and

meaningless tasks support the hypothesis of the WCC theory and are not consistent with the EPF hypothesis. In contrary to the WCC theory, the EPF predicts higher scores on both meaningless and meaningful items, because of superior local processing which becomes more evident in the tasks concerning the most difficult shape discriminations regardless of the content of the materials.

Despite the smaller groups of deaf individuals, also a significantly higher score on meaningless tasks and not on meaningful tasks was found for deaf individuals with ASD, whereas no differences were found on meaningless or meaningful tasks for deaf individuals without ASD. As expected, only the results of the deaf individuals with ID and ASD are in line with the results of the hearing individuals with ID and ASD. These results indicate the same underlying cognitive style in both deaf and hearing individuals with ASD.

In summary, like in individuals with ID and ASD, enhanced performance on the sorting tasks in deaf individuals with ID and ASD compared with individuals with ID can be understood within the framework of the central coherence theory, whereas in deaf individuals with ID without ASD the enhancement might be due to a more generally enhanced visual perception. WCC in both groups with ASD is reflected in superior performance on meaningless sorting tasks where the ability to process local information is advantageous, because the groups without ASD cannot benefit from a more global processing style.

In the study of Van Lang et al. (2006) also indications for a weaker drive for central coherence in individuals with ID and ASD were found in the performances on the Block Design Test and the Child Embedded Figures Test, whereas Harris et al. (2007) failed to find the same results on the Block Design Test. The most important difference between these two studies is the level of functioning of the participants. Both studies focused on participants with ID, but the former study included mild and moderate ID while the latter only addressed mild ID or even borderline intellectual functioning. Based on the results of before-mentioned studies, it can be hypothesized that a WCC in the visual or visuo-spatial domain becomes clearer in performances on less complex tasks as the level of cognitive functioning decreases. To study WCC in the visual domain among high-functioning individuals with ASD even more complex visual tasks which require global perception of complex symbolic meanings are necessary to reveal this processing style (see for example Nakahachi et al., 2008). In addition, all studies into WCC in individuals with ASD and ID only examined the style of processing in the visual / visuo-spatial domain. Studies into the auditory / verbal domain are difficult to carry out, because low-functioning individuals often lack the verbal abilities to perform the tasks.

Results of the present study have to be interpreted in the context of the study's limitations. The first shortcoming is related to the instrument used in this study. Several sorting tasks derived from the ComFor are applied to measure visual perception, whereas the main purpose of this clinical instrument concerns the assessment for the indication of augmentative communication based on perception and level of sense-

making (Verpoorten et al., 2004, 2008). However, the sorting procedure used in this instrument is very suitable to measure visual perception in low-functioning individuals. Furthermore, there are differences in size and chronological age between the subsamples. The groups with deafness are smaller and significantly older than the hearing groups with ID and/or ASD. Therefore, results have to be interpreted with caution, because ANCOVA is less robust to violations of homogeneity of variance when group sizes are unequal. Recruiting (young) deaf individuals with ID and ASD turned out to be very difficult, also aggravated by the strict criteria we had to use for deafness and ASD in ID. Only individuals who passed a hearing test and had an official DSM-classification for autistic disorder or PDD-NOS were considered for participation, as we lacked a suitable (screening) instrument to classify ASD in deafness (Vernon & Rhodes, 2009). Although ASD can be diagnosed in deaf individuals, it is much later recognized than in the hearing population, often not before adolescence (Roper et al., 2003). A broader international collaboration is necessary to improve research into individuals with ID, ASD and deafness. Finally, a control group of individuals with ID is used. This group can be characterized as a very heterogeneous group, but the common characteristics are no hearing loss or ASD. To reveal strengths and weaknesses in specific conditions with ID, the use of a heterogeneous control group of ID is much more preferable than a typical or specific control group like Down syndrome (Hodapp & Dykens, 2001). To ensure comparability between the subsamples, all groups were definitely matched for level of adaptive functioning according to the criteria for matching (Mervis & Klein-Tasman, 2004).

Despite these limitations, the current findings do have implications for our understanding of deafness and ASD in ID, and the processes involved in visual perception and sense-making. A clearer understanding of the nature of mechanisms underlying problems in perception and sense-making has the potential to improve interventions. WCC is not only present in hearing, but also in deaf individuals with ID and ASD. This study shows that ASD and deafness do not only result in deficits, but can bring along assets, like a strong local visual perception, in functioning as well. Because ASD and deafness have communication problems in common, the presence of both conditions in one person can lead to more severe communication problems, therefore treatment becomes more complex (Vernon & Rhodes, 2009). The understanding and use of sign language is not always achievable in low-functioning deaf individuals with ASD. Therefore, other communication strategies have to be applied to maximize communication. In clinical practice, it is important to realize that individuals with associated disabilities may benefit from other augmentative communication strategies compared with individuals with ID only. Strengths in visual perception and abilities in sense-making can be used to offer the best suitable intervention to support communication.

7

General conclusions and discussion

General conclusions and discussion

Children with an autism spectrum disorder (ASD) and an intellectual disability (ID) form a particularly vulnerable group, as both disorders have a significant impact on the way and level of information processing and communication (Boucher et al., 2007; Noens & Van Berckelaer-Onnes, 2004). ASD and ID are usually accompanied by severe communication problems, which may lead to challenging behavior (Bott et al., 1997; Chung et al., 1995). However, children with ASD and co-occurring ID are often excluded from research. Knowledge with regard to the communication skills of this particular group is greatly lacking. Therefore, the main objective of this dissertation concerned the assessment of communication characteristics in children with autistic disorder (AD) and co-occurring ID in order to determine how children with AD and ID are similar and different in their communication compared with children with ID and typical development (TD). The study focused on problems in functions, forms, and content of communication. In this chapter, the general conclusions will be presented and discussed in relation to limitations and implications for clinical practice and research purposes.

Assessment of autism spectrum disorder in children with intellectual disability

The first step in the present study was to examine the psychometric properties and clinical value of instruments for identification of ASD in children with ID. The diagnosis of ASD in individuals with ID can be complicated, especially in those with severe or profound ID (Bhaumik et al., 2010). It is therefore important to evaluate the tools used to diagnose ASD in the ID population. The Diagnostic Interview for Social and Communication Disorders version 11 (DISCO; Wing, 2006) is a standardized interview that professionals can use to diagnose ASD. It was designed for any age and any level of ability, but the psychometric properties of the DISCO for ASD diagnosis in ID had never been determined. This study evaluated how well the DISCO algorithm, relative to other tools commonly used to diagnose ASD, can differentiate between children with and without ASD at different levels of intellectual ability. The instruments used for comparison were the Social Communication Questionnaire (SCQ; Rutter et al., 2003) and the Autism Diagnostic Observation Schedule (ADOS; Lord et al., 1999; Gotham et al., 2007). For this study, we focused on the whole ASD spectrum, because not all instruments differentiate between AD and ASD. The results of this study into the psychometric properties of ASD screening and diagnostic instruments were used in order to decide which instruments would be applied to compose the research and comparison groups for our subsequent studies.

With respect to the utility of the DISCO as a diagnostic instrument for ASD in individuals with ID, two main conclusions can be drawn (see Chapter 2). First, sensitivity and specificity rates for the three instruments were compared. The results indicated that the DISCO is a very sensitive instrument, which implies that a high proportion of

children with and without ID were correctly classified as having ASD. However, the DISCO proved to be over-inclusive in the group of children with moderate or severe ID. In this low-functioning group, children without a clinical ASD diagnosis were frequently incorrectly classified as having ASD on the DISCO. The SCQ proved to be a very specific screening instrument with a considerable likelihood of false negative outcomes. The best rate between sensitivity and specificity was found for the ADOS. Second, the agreement between the DISCO and other tools for screening or diagnosing ASD was moderate to substantial.

Possible explanations for different outcomes of the three instruments include the content of information gathered, the sources of information, and the proposed target group of the instruments. With respect to the content of information, the SCQ is the shortest and least time consuming instrument with the most limited scope of information of the three instruments. Furthermore, the questions about communication impairments mainly focus on language characteristics and are therefore less applicable for nonverbal low-functioning children. In the ADOS, the emphasis is also on the three main criteria for ASD, but the items are more specified and the standardization is explicitly customized for verbal and nonverbal children. The DISCO covers the widest range of developmental domains and atypical behaviors, but no difference is made between verbal and nonverbal individuals in the algorithms used for the classifications. It is also important to take into account that the classifications of the instruments are based on different sources of information. The SCQ is a parent questionnaire, whereas the DISCO is an interview administered from the parents, but scored by a professional. The assessment of the ADOS concerns an observation of the child by a professional. Parents and professionals may have different perceptions about the behavior of children (Barnhill et al., 2000; De Los Reyes & Kazdin, 2005). The SCQ is specifically developed for children of four years and older or with a mental age over two years, whereas the ADOS and DISCO cover a broader developmental and chronological age range. However, our participants were partly younger or lower functioning than the target group of the SCQ. Furthermore, the SCQ was designed for screening purposes and both the ADOS and the DISCO as diagnostic instruments. A screening instrument is designed to identify individuals at risk for a disorder; hence a greater emphasis should be placed on the sensitivity. In our study, the SCQ was not effective as a screening instrument in low-functioning individuals. The major problem of the relatively poor sensitivity of the SCQ seems to be the prescribed cut-off score, as higher sensitivity rates would be achieved when the cut-off score was reduced, in line with previous studies with young children (e.g., Corsello et al., 2007; Snow & Lecavalier, 2008). Based on the raw scores, the DISCO has the potential to make an adequate differentiation between children with ID with and without ASD, but the current algorithms and cut-off scores are not sufficiently adjusted to this low-functioning group. The present findings suggest that clinicians and researchers should use caution when using the current

standards for classification of the DISCO and SCQ with individuals with moderate and severe ID.

Assessment of ASD involves an in-depth investigation by experienced clinicians. An ASD diagnosis is based on clinical judgment, aided by the combined results of various instruments and the guidelines as formulated in the DSM-IV-TR and ICD-10 (Filipek et al., 1999). A classification on the basis of a single screening or diagnostic instrument is not sufficient in order to make a clinical diagnosis. The current study only evaluated the final classification on the instruments. The diagnostic instruments also provide an extensive amount of qualitative information, but the usefulness of this information for the diagnostic process was not evaluated in this study. The ADOS and DISCO together provide a broad description of the child's behavior. The assessment with the ADOS gives the opportunity to observe the child's behavior in structured and semi-structured situations (Lord et al., 2000). A detailed interview with the parents, such as the DISCO, gives information about the developmental history, current behavior, and level of functioning in several developmental domains at home or in other settings (Wing et al., 2002). Research has shown that a combination of both methods is more reliable and stable over time (Risi et al., 2006). The ADOS and DISCO can shed light on the child's strengths and weaknesses. This information can be used for an individualized, descriptive diagnosis and to devise an individualized treatment plan (Wing, 2006).

To compose our research group with AD and ID, the ADOS and DISCO together were used to confirm the previously given clinical diagnosis. For the comparison groups of children with ID without ASD and the typically developing group, the ADOS was administered to exclude possibly undiagnosed ASD cases.

Communication in low-functioning children with autistic disorder

Communication characteristics of children were examined with respect to communicative functions, forms, and content. The results of the present study will be discussed in this order. The main findings of the studies are summarized in Table 7.1.

Communicative functions

As described in Chapter 3, we observed communicative functions of low-functioning children with autistic disorder in comparison with typically developing children groupwise matched on nonverbal developmental level. Three basic functions were assessed: behavior regulation (acts to regulate another's behavior for purposes of requesting or protesting), social interaction (acts to draw another's attention to oneself for social purposes), and joint attention (acts to direct another's attention to interesting objects or events for the purpose of sharing). Overall the findings suggest that low-functioning children with AD communicated at a significantly lower rate than typically developing children with a comparable developmental level. For this reason, the absolute frequencies of functions were converted into relative frequencies or

proportional use in order to compare the communication profiles independent of the total amount of intentional communication. Nonverbal children with AD were severely impaired in both the absolute and relative use of the different communicative functions. In the nonverbal group some children communicated only a few times during the whole observation. These children did not seem to know how to influence the behaviors of others in an intentional way. Other children in the nonverbal group showed intentional communication, but communicative acts for behavior regulation covered the greater part of their communication initiations. They did not communicate for social interaction and joint attention purposes or only to a small extent. An important difference in the type of functions which are coded within each of these communication functions is that behavior regulation serves a less social function compared to social interaction and joint attention (Wetherby & Prizant, 2002). The differences between verbal children with AD and typically developing children were less pronounced. The relative frequency of function use is comparable between both groups, although the absolute frequency of communication for joint attention is lower for verbal children with AD.

Table 7.1 ***Main findings on communication in children with autistic disorder and intellectual disability***

		Children with autistic disorder and intellectual disability	
		Nonverbal	*Verbal*
Function	*Behavior regulation*	- Relative strength (Ch3)	- Relative strength (Ch3)
	Social interaction	- Not or only scarcely present (Ch3)	- Only differences in absolute not in relative frequencies compared with TD (Ch3)
	Joint attention	- Not or only scarcely present (Ch3)	- Only differences in absolute not in relative frequencies compared with TD (Ch3)
	Unclear	- Not or only scarcely present (Ch3)	- More frequently present than in nonverbal AD+ID and TD (Ch3)
Form	*Nonverbal*	- Overall substantially lower complexity than verbal AD+ID & TD (Ch3)	- Verbalizations less often combined with both gestures and eye gaze for joint attention compared with TD (Ch3)
	Verbal [a]	- Significant delays in receptive and expressive language (Ch4) - Receptive and expressive language significantly related to joint attention and symbol understanding (Ch4)	
		- Not applicable	- Opposite profile from TD and ID: expressive > receptive language (Ch4)
	Augmentative [a]	- Stronger local processing of pictures and objects compared to ID with and without deafness (Ch6)	
Content	*Level of sense-making* [a]	- Significantly lower levels of sense-making compared with ID and TD (Ch5) - At non-symbolic levels of sense-making: more severe problems in social interaction and communication (Ch5) - Level of sense-making is an important underlying factor for adaptive delays (Ch5) - Non-symbolic levels of sense-making are associated with more severe autism symptomatology (Ch5)	

[a] Analyzed for the AD+ID group as a whole, no subdivision in verbal and nonverbal groups was made.

It is important to note that the results reflect communication profiles during the Communication and Symbolic Behavior Scales – Developmental Profile (CSBS-DP) administration only, and can only be generalized to a certain extent. Factors that may influence the communication profiles are the semi-structured nature of situation,

unfamiliarity with the situation and the materials, and explicit prompting of communication. However, a standardized observational measurement procedure provides a more stable estimate of a child's performance and enables a reliable comparison between groups (Yoder & Simons, 2010).

In conclusion, communication for joint attention purposes is a major obstacle for low-functioning children with AD. Other studies pointed out that this characteristic also differentiates ASD from other developmental disorders in young children (Dawson et al., 2004; Wetherby et al., 1998). Joint attention involves triadic information processing, while behavior regulation and social interaction are mainly dyadic. When children are engaged in joint attention, they have to turn their attention to and process information about: an object or event, another person's attention and behavior related to the object, and information about their own attention to, and experience of the object and the situation (Mundy & Jarrold, 2010). Recently, Mundy and Jarrold (2010) proposed a new explanatory model of joint attention, called the Parallel and Distributed-Processing Model (PDPM). Although prerequisites and emergence of joint attention remain unclear, this model illustrates the complex and interactive nature of joint attention development. According to this model, three important aspects contribute to the development of joint attention. First, complex information processing of parallel self-referenced, other-referenced, and object- / event-referenced information is involved. Second, development originates from continual practice and children generate knowledge through their own initiated actions. Third, it is likely that this process is affected by and has an effect on the flow of information between brain areas (Mundy et al., 2009; Mundy & Jarrold, 2010; Mundy et al., 2010). It can be expected that problems will arise when any of these three components of joint attention development are disrupted or compromised. Children with ASD demonstrate significant difficulties in their ability to attend to and process information about another person's attention and behavior and incorporate the perspective of others into their communicative behavior (Nilsen & Fecica, 2011). Simultaneously, impairments in ASD could arise from disturbances in initiation, resulting in diminished experience and practice (Dawson et al., 2004; Klin et al., 2002). A growing body of evidence suggests that impairments in distributed brain systems are involved in joint attention problems seen in ASD (Müller, 2007; Rudie et al., 2011).

Communicative forms

Communicative intentions can be expressed in several ways, differing in quality from very primitive forms to more conventional forms, such as spoken language. In our study, several forms or means of communication were assessed. First, spontaneous naturalistic forms used for expressive communication were observed. A distinction was made between verbalizations, gestures, and use of eye gaze. Second, we zoomed in on the use and comprehension of verbal language. And third, comprehension of augmentative communication forms was examined.

Nonverbal and verbal communication. Previous studies examined the quantitative use of several expressive communication forms in children with ASD. In our study, not the quantity of specific communicative forms, but the complexity of forms was evaluated by examining the degree to which communicative gestures, vocalizations / verbalizations, and eye gaze were combined in one communicative act. The results of our study (see Chapter 3) indicated that mainly nonverbal children with AD used very primitive forms of communication and combined their communicative gestures or vocalizations less often with eye gaze than verbal children with AD and typically developing children. With respect to verbal children with AD, results revealed group differences at the most complex level of communicative forms only. Verbal children with AD used a significantly lower proportion of acts involving gestures, verbalizations, and eye gaze for the purpose of joint attention relative to the typically developing control group.

Receptive and expressive language. Language is the most conventional form of communication. As described in Chapter 4, we found that in the group of children with AD and ID, expressive and receptive language skills were substantially lower than their nonverbal cognitive level. In contrast, in both comparison groups language levels approximated the nonverbal cognitive level, except for expressive language in the ID group. Further, children with AD and ID had considerably better language production than comprehension skills, which is a pattern that is reversed in children with ID and typically developing children. These findings suggest that children with AD and ID who have some spoken language may well be overestimated on the basis of relatively good language production skills. The findings in low-functioning children with ID are in line with the results for toddlers with ASD (e.g., Hudry et al., 2010; Miniscalco et al., 2011; Weismer et al., 2010), but differ from results in older high-functioning children with ASD (Jarrold et al., 1997; Kjelgaard & Tager-Flusberg, 2001). The findings of the current and previous studies together indicate a discrepancy in language profiles between high and low-functioning children with ASD. In high-functioning children the difference between receptive and expressive skills seems to diminish as they grow older, while in low-functioning children with AD an atypical language profile remains present after early childhood.

Joint attention and symbolic thinking are defined as important precursors for language development in young children and are considered to be core deficits in communication development of children with ASD (Travis & Sigman, 2001). In our study both aspects were assessed in relation to concurrent receptive and expressive language abilities in children with AD and ID (see Chapter 4). In the comparison groups with ID and TD nonverbal mental age was strongly related to these precursors and the most robust concurrent predictor of both language comprehension and production. However, in the low-functioning AD group symbol understanding and joint attention made a significant and unique contribution to concurrent expressive and receptive language

abilities. This finding suggests that in contrast to the homogeneous development in typical development, developmental profiles in ASD are more disharmonious and language abilities are more independent from nonverbal cognitive level of functioning. Joint attention has often been indicated as an important predictor of language outcome in toddlers with ASD (e.g., Charman et al. 2003; McDuffie et al. 2005). The role of symbol understanding, however has seldom been scrutinized in relation to language skills. The nature of the relationship between joint attention and symbol formation and the relative contribution of both precursors to language development remain unclear. A study by Warreyn and colleagues (2005) suggested that social communicative skills, such as joint attention, develop independently from symbolic abilities in children with ASD. According to the PDPM of joint attention, the practice with joint attention contributes to the neurocognitive foundation of symbolic thinking (Mundy et al., 2009), as learning the meaning of a symbol usually implies a social activity consisting of both a producer who intends to convey information via a representational form and a recipient to whom this information is directed (DeLoache, 2004; Namy & Waxman, 2005). In this way joint attention can be helpful in learning symbol-referent relations (DeLoache, 2004). Accordingly, Toth and colleagues (2006) pointed out that joint attention is an important skill for the start of language development, while symbolic play - which refers to more representational or symbolic skills - is associated with the continued development and the expansion of language abilities in later years. In our study, both joint attention abilities and symbol understanding had a unique contribution to the level of concurrent language skills, but the contribution of symbol understanding was more pronounced. Also in children with AD, joint attention abilities increase with age (Naber et al., 2008), and probably become less important in language development in older children with AD, which explains the major role of symbol understanding. Furthermore, based on the results of Chapter 5 which highlights that problems in sense-making and symbol formation are specific for children with AD, it can be hypothesized that children with AD develop symbol understanding at a later stage or in an atypical way. Therefore, the role of symbol understanding might be more prominent in this group.

Augmentative communication forms. Many low-functioning children with ASD remain nonverbal or have only a limited number of communicative words or gestures. These individuals are dependent on other communication forms. Several communicative forms can be used to augment communication. Both three-dimensional forms (visuo-spatial objects) and visual, two-dimensional forms (pictures) were assessed with the ComFor. These assessments were used for the studies described in the Chapters 5 and 6. In Chapter 6, perception and processing of visual and tactile information (objects and pictures) were examined. Problems in symbolic communication in ASD, especially with respect to the content of communication or the establishment of meaning, can possibly be explained by a weaker central coherence. Central coherence refers to the natural tendency to process incoming stimuli in context, drawing pieces of information

together to derive meaning (Happé & Frith, 2006). In children with ASD, central coherence is rather weak, primarily resulting in a detail-focused processing style often accompanied by a failure to process information in context (Happé & Booth, 2008). Enhanced local processing skills were found in individuals with ASD and ID in a previous study by Noens and Van Berckelaer-Onnes (2008). We examined the effects of co-occurring deafness on visual perception in low-functioning individuals with ASD to investigate the specificity of this enhancement and explored whether a weak central coherence may be an underlying cognitive mechanism contributing to the visual processing style in individuals with ASD with and without deafness. The results showed that hearing as well as deaf individuals with ASD and ID have stronger local processing skills of pictures and objects compared to individuals with ID. It is important to stress that the terms enhanced visual perception and superior local processing in this context are used as relative statements, considering the ID of all participants. Although this processing style can hinder in tasks or situations where global information processing is useful or even necessary, our findings also provide evidence that individuals with ASD can 'benefit' from a weak central coherence. These relative strengths in visual perception can be used to augment communication.

Communicative content

Concerning communicative content, we focused on the question whether content is understood in a symbolic or non-symbolic way, and on the consequences of both ways of sense-making or perception of meaning. As described in Chapter 5, the present study revealed that difficulties in sense-making are more pervasive in children with AD and ID compared to typically developing children, but also compared to children with ID without ASD. In the group with AD, significantly fewer children achieved the level of representation. A large part of this group is only able to perceive the world at the levels of sensation or presentation, which implies that they do not (completely) understand the symbolic or representational content of communication. In particular low-functioning children with ASD are not always able to grasp the symbolic meaning of communication (Preissler, 2008). Communicative forms and contents must be directly linked to function and context in order to establish accurate meaning and sense-making (Quill, 1995). Children with ASD are less able to adapt and respond to the rapidly moving, fluid context of communication that demands flexibility (Landa, 2007). They can deal more effectively with concrete, non-transient stimuli. Therefore, children with ASD have a preference for visual-spatial information instead of temporal, auditory information (Twachtman, 1995). In nonverbal children, level of symbolic understanding of other communication forms, such as pictures or objects, also needs to be assessed (Paul, 2005). Based on our results, level of sense-making seems to be an important underlying factor for adaptive delays and more severe autism symptomatology. Mainly children with AD and ID at the non-symbolic levels (sensation and presentation) are impaired in adaptive functioning on the domains of communication and socialization

compared with their nonverbal mental age matched controls with either ID or typical development. Non-symbolic levels of sense-making are also associated with more severe autism symptomatology in terms of quality of communication, social interaction, and imagination.

Limitations and directions for future research

Research into communication in children with ASD and co-occurring ID is done less often compared to studies in higher functioning children with ASD. The results of this study in terms of impairments in intentionality, use of communicative functions, and language abilities, show many similarities with communication and language results in toddlers with ASD. Not completely surprisingly, as the developmental level of the children in our research group is situated in the toddler age range. However, the problems in the group with children with AD and ID seem to be more aggravated and pronounced. Unfortunately, we were not able to compare our results with a comparison group of young children with AD without ID with a comparable developmental level. The use of additional comparison groups, one with children with AD without ID (for all Chapters), and another with children with ID without AD (for Chapter 3), is an important recommendation for future research. Due to the very comprehensive way of data collection, especially concerning the observations of communicative functions and forms, this was currently not attainable in our study. It will be an important next step, but also a complicated one, as it is difficult to compose a comparison group of toddlers with AD without ID. When these groups have to be comparable with respect to nonverbal mental age, this has to be a relatively young group with autism. Although early diagnoses of ASD are nowadays more common (Charman & Baird, 2002; Zwaigenbaum et al., 2009), confirmation of the absence of an ID at a young age is less obvious, as many young children with ASD have developmental delays. Given that the focus of our research is a clearly defined group, for which communication profiles were not studied yet, the results of this study can be considered exploratory in nature. Based on the current data, it is not possible to draw conclusions about which characteristics may be solely due to the AD and which to the co-occurring ID. We have tried to describe the characteristics of the group with co-occurring AD and ID as a whole to provide suggestions for assessment and treatment of communication problems in this vulnerable group.

Another concern with respect to our comparison groups are the gender differences between the groups. As ASD is more common in boys than girls, the majority of this group is male, while the distribution male versus female is more equal in the comparison groups. In our study, groupwise matching on nonverbal mental age was given priority over the gender variable. Ideally, groups would be matched on both variables. For that reason, we controlled for the influence of gender on the outcome variables in all chapters.

Several related and important research topics were not addressed in this study. First, it is recommendable to further explore the development of symbol understanding in young and low-functioning children with ASD in order to gain more insight in the course of development and possible deviations from typical development. Second, based on the literature a relation between communication problems and challenging behavior can be expected (Bott et al., 1997; Chamberlain et al., 1993, McClintock et al., 2003). As communication problems are rather divers in low-functioning children with ASD, it is important to examine the association between specific communication problems (e.g., in terms of problems in functions, forms, or sense-making) and challenging behavior in order to improve prevention and interventions.

Clinical implications

To diagnose ASD in children with ID, cognitive abilities always have to be specified and are an important component of the diagnostic process. Information about the child's cognitive level of functioning provides the clinician with a context for evaluating behavioral symptomatology. This context can help to decide whether symptoms can possibly be explained by a global delay, and whether there is actually a developmental readiness for feature expression (Totsika et al., 2010; Vig & Jedrysek, 1999).

The results of this study showed that low-functioning children with AD have different impairments, at least in the social communication domain, compared to high-functioning children with AD. Concomitantly, a large heterogeneity of symptoms was observed within this low-functioning group. These results endorse the shift to a dimensional approach of impairments in ASD. The current classification systems are based on a predominantly categorical model with specific diagnostic criteria for different categories. In recent years, the conceptualization of ASD as a spectrum with different continua gained increased attention, partly due to the debates concerning the development of the DSM-5 (http://www.dsm5.org). The question is whether some major aspects of ASD might better be conceptualized in terms of quantitative dimensions (in analogy with for example IQ), instead of qualitatively distinct categories (Taylor & Rutter, 2002). In a categorical system behavioral characteristics are classified as either meeting criteria for a disorder or not based on qualitative criteria, in other words an 'all or nothing' approach (Kamp-Becker et al., 2010; Lilienfield & Landfield, 2008). The current categorical approach has been developed to improve reliability and validity of the classifications and research findings (Volkmar, 1998; Wing, 2005). However, reality is often more complex, resulting in the exclusion of a large number of children and overlap between several classifications. In line with the dimensional approach, difficulties in ASD are considered broad dimensions of individual variations that are widely distributed in the general population (Constantino & Todd, 2003). A disorder represents the severe end of continuous distributions of core symptoms (Constantino, 2011). Impairments in the three areas that characterize ASD are modified by many factors, for example chronological age, developmental level, language

functioning, and co-occurrence of other disorders (Hill & Frith, 2003; Volkmar & Lord, 1998). The ability to measure the severity of ASD dimensions would be useful for both research and clinical practice (Volkmar & Lord, 1998). Scientific evidence is necessary to validate the dimensions or spectra to diagnose and indicate severity.

Every child with ASD will have needs in the area of communication, since this is one of the core deficits in ASD. But the types and intensity of needs vary depending on age, level of functioning, and other characteristics (Paul, 2007). As we have seen in this study, even within the group of children with autistic disorder and co-occurring ID communication impairments are very heterogeneous. For this reason, assessment of communication always needs to be part of the diagnostic assessment of low-functioning children with ASD. In the case of nonverbal children, it will be particularly important to evaluate how they communicate in other ways, such as with gestures, gaze, and vocalizations, as well as their comprehension of the communication of others. Furthermore, the observation of intentionality and functions of communication needs to be included. Observation instruments, such as the Communication and Symbolic Behavior Scales – Developmental Profile (Wetherby & Prizant, 2002), or comparable situations which elicit the child to communicate, can be used in this target group. However, standardized scoring of the CSBS-DP is not sufficient. Many low-functioning children with ASD who lack the use of conventional means of communicating, may develop idiosyncratic, unconventional, or undesirable behaviors to communicate, such as self-injurious behavior or aggression (Paul, 2007; Wetherby et al., 2000). The use and functions of challenging behavior also need to be considered in the diagnostic process. For verbal children with ASD and ID, the usual and standardized tests for language development can be used. These will often need to be supplemented with observations of communication behavior in daily life situations to determine whether there is a disconnection between the level of language skills measured with standard tests and the use of language to achieve reciprocity and adequate exchange of information (Paul, 2007). Stimulation of communicative competence is an important goal in interventions in ASD. A common practice in communication intervention in young and low-functioning children with ASD is that of stimulation of verbal language (Twachtman, 1995). However, often there are not only severe delays in or a lack of speech, but also delays in the development of intentionality and symbol formation (see Chapters 3 and 4). This requires a broader communication intervention approach adjusted to their developmental level.

A significant part of mainly low-functioning children with ASD do not develop language or only a limited number of communicative words or gestures. Particularly in case of severe or profound ID, often no verbal or symbolic communication is present. These individuals are dependent on other communication forms, such as augmentative communication means. The need for individualized augmentative strategies to enhance the communication skills of low-functioning children with ASD is evident, given the severity of their language and communication impairments (Howlin, 2006). For low-

functioning children with ASD the common alternative and augmentative communication (AAC) systems are not directly applicable. Several principles of AAC, as formulated by the Committee on Augmentative Communication of American Speech Language Hearing Association (ASHA, 2005), do not correspond completely with the results of our study.

First, AAC is mainly focused on enhancing the expressive communication skills of individuals who have relatively good understanding of communication (Fossett & Mirenda, 2007; Wilkinson & Hennig, 2007). However, positive changes regarding this topic are included in the most recent definition of AAC, where also persons with comprehension problems are addressed. This definition draws attention to the unique communication needs of individuals who struggle to understand spoken language. In low-functioning and young children with ASD receptive skills are often relatively more impaired than expressive language abilities (see Chapter 4). In these target groups, initially strategies which aim to augment receptive communication need to be implemented. Augmentative communication can be used to announce activities or to help making transitions between activities. Further, it can be used to explain different steps within an activity or to teach sequences in several activities. When augmentative communication strategies are successfully implemented to support receptive communication, it can be used to encourage expressive communication as well. The child can be taught to use the same individualized strategies adjusted to the level of sense-making, for example to regulate the behavior of others or to make choices. The success of such an expressive intervention strategy is clearly related to the child's ability to communicate intentionally (Rowland & Schweigert, 2000). Nonverbal low-functioning children with ASD are often severely impaired in the use of intentional communication (see Chapter 3). When some intentionality is present, it is important to build upon the existing communicative functions and previously learned communication skills to extend the expressive communication skills. It is essential that the child's individual interests or needs are taken into account. Nevertheless, in many interventions which aim to stimulate the expressive communication, the focus of the training tends to be on teaching predetermined labeling skills. However, communication interventions are more likely to succeed if the child's existing and spontaneous attempts to communicate function as starting points. In this way, the child is provided with greater control over the environment (Howlin, 2006). Communication profiles in terms of function use are variable among low-functioning children with ASD, thus need to be characterized before the start of an intervention (see Chapter 3). Furthermore, because of generalizations problems, communication skills have to be taught in regularly occurring, daily life situations, instead of teaching them in isolation. Thus, the context for skill development needs to be the same as the context for skill use (Howlin, 2006; Twachtman-Cullen & Twachtman-Reilly, 2007).

Second, in the definition of AAC communicative means, such as photographs, printed words, objects, are being used in a symbolic way. However, the results

concerning level of sense-making pointed out that not all children with ASD and co-occurring ID are able to understand the symbolic meaning of communicative forms. When symbolic augmentative communication strategies are used when someone is unable to understand the meaning of symbols, the result is likely to be a frustration for both the child and the clinician (Rowland & Schweigert, 2000). A frequently used symbolic AAC system is the Picture Exchange Communication System by which children learn to exchange a picture for a desired item (PECS; Bondy & Frost, 2001). A recent study of McFee (2011) showed that symbolic understanding of pictures was one of the factors that was related to effectiveness of the PECS intervention. Our study showed that a part of the low-functioning children with ASD did not (yet) reach the required level of representation or symbolic understanding of communicative means. Because of the limited symbol understanding in ASD, the level of sense-making has to be taken into account before starting an intervention (Noens & Van Berckelaer-Onnes, 2004). Level of sense-making for the indication of an individualized strategy for augmentative communication can be measured by using the ComFor (Verpoorten et al., 2004, 2008). Several non-symbolic strategies for augmentative communication were developed at the levels of sensation and presentation (see Noens & Van Berckelaer-Onnes, 2004). As reviewed by Rowland and Schweigert (2003), low-functioning children with ASD are often excluded from AAC services because they fail to demonstrate so-called prerequisite skills, such as object permanence, and as a result are thought not to be ready for symbolic communication. Wilkinson and Hennig (2007) make a case for a broader view on AAC by proposing AAC as a continuum that can range from simple social or turn taking routines through more sophisticated use of symbols. In our opinion, functions, forms and levels of sense-making, are mixed up in this explanation of possible applications of AAC. Augmentative communication is possible for each level of sense-making, as also non-symbolic strategies can be utilized. Furthermore, forms are independent of levels of sense-making, thus for instance objects can be used both in a non-symbolic or symbolic way to support communication. And lastly, different communicative functions can be encouraged at each level of sense-making and have to be considered independently from form and level.

Concluding remarks

In conclusion, the results of our study emphasize that a successful strategy for augmentative communication must aim not only to provide the child with more effective expressive means of communication, but also improve understanding - both verbally and nonverbally - and increase motivation to communicate (Howlin, 2006). Moreover, augmentative communication strategies are accessible for everyone, also for non-symbolic communicators, if adjusted to individual needs. Augmentative communication strategies are often applied in clinical practice. Thus far, effectiveness of individualized augmentative communication strategies at the levels of sensation and presentation has not been demonstrated. This study focused on the description of

communicative strengths and weaknesses in low-functioning children with ASD. Based on the current findings several suggestions for assessment and treatment of communication in this group were formulated. However, another clinical question concerns the validity of communication measures or effect of interventions that are provided for children with ID and ASD. Extending the knowledge on these issues is therefore highly recommended as the focus of future research.

References

References

Abrahams, B. S., & Geschwind, D. H. (2008). Advances in autism genetics: On the threshold of a new neurobiology. *Nature Reviews Genetics, 9,* 341-355.

Adamson, L. B., Bakeman, R., Deckner, D. F., & Romski, M. A. (2009). Joint engagement and the emergence of language in children with autism and Down syndrome. *Journal of Autism and Developmental Disorders, 39,* 84-96.

Adrien, J. L., Rossignol-Deletang, N., Martineau, J., Couturier, G., & Barthelemy, C. (2001). Regulation of cognitive activity and early communication development in young autistic, mentally retarded and young normal children. *Developmental Psychobiology, 39,* 124-136.

American Psychiatric Association [APA] (1994). *Diagnostic and Statistical Manual of Mental Disorders* (4th ed.). Washington, DC: APA.

American Psychiatric Association [APA] (2000). *Diagnostic and Statistical Manual of Mental Disorders* (4th ed., text revision). Washington, DC: APA.

American Speech-Language-Hearing Association [ASHA] (2005). *Augmentative and alternative communication (AAC)*. Retrieved from http://www.asha.org/

Baron-Cohen, S. (1989). Perceptual role-taking and protodeclarative pointing in autism. *British Journal of Developmental Psychology, 7,* 113-127.

Barnhill, G. P., Hagiwara, T., Smith Myles, B., Simpson, R. L., Brick, M. L., & Griswold, D. E. (2000). Parent, teacher, and self-report of problem and adaptive behaviors in children and adolescents with Asperger syndrome. *Diagnostique, 25,* 147-167.

Bartlett, C., & Bunning, K. (1997). The importance of communication partnerships: A study to investigate the communicative exchanges between staff and adults with learning disabilities. *British Journal of Learning Disabilities, 25,* 148-153.

Bates, E. (1979). Intentions, conventions, and symbols. In E. Bates, L. Benigni, I. Bretherton, L. Camaioni, & V. Volterra (Eds.), *The emergence of symbols: Cognition and communication in infancy* (pp. 33-68). New York, NY: Academic.

Bavelier, D., Dye, M. W. G., & Hauser, P. C. (2006). Do deaf individuals see better? *Trends in Cognitive Sciences, 10,* 512-518.

Bebko, J. M., Konstantareas, M. M., & Springer, J. (1987). Parent and professional evaluations of family stress associated with characteristics of autism. *Journal of Autism and Developmental Disorders, 1,* 565-576.

Beglinger, L. J., & Smith, T. H. (2001). A review of subtyping in autism and proposed dimensional classification model. *Journal of Autism and Developmental Disorders, 31,* 411-422.

Berument, S. K., Rutter, M., Lord, C., Pickles, A., & Bailey, A. (1999). Autism Screening Questionnaire: Diagnostic validity. *The British Journal of Psychiatry, 175,* 444-451.

Betancur, C. (2011). Etiological heterogeneity in autism spectrum disorders: More than 100 genetic and genomic disorders and still counting. *Brain Research, 1380,* 42-77.

Bhaumik, S., Tyrer, F., Barrett, M., Tin, N., McGrother, C. W., & Kiani, R. (2010). The relationship between carers' report of autistic traits in clinical diagnoses of autism spectrum disorders in adults with intellectual disability. *Research in Developmental Disabilities, 31,* 705-712.

Bishop, S. L., Luyster, R., Richler, J., & Lord, C. (2008). Diagnostic assessment. In: K. Charwarska, A. Klin, & F. R. Volkmar (Eds.), *Autism spectrum disorders in infants and toddlers: Diagnosis, assessment, and treatment* (pp. 23-49). New York, NY: Guilford Press.

Blanc, R., Adrien, J.-L., Roux, S., & Barthélémy, C. (2005). Dysregulation of pretend play and communication development in children with autism. *Autism, 9,* 229-245.

Bloom, P. (2000). *How children learn the meaning of words.* Cambridge, MA: The MIT Press.

Bloom, L., & Tinker, E. (2001). The intentionality model and language acquisition: Engagement, effort, and the essential tension in development. *Monographs of the Society for Research in Child Development, 66,* 1-89.

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

Bölte, S. & Poustka, F. (2002). The relation between general cognitive level and adaptive behavior domains in individuals with autism with and without co-morbid mental retardation. *Child Psychiatry and Human Development, 33,* 165-172.

Bondy, A. & Frost, L. (2001). The Picture Exchange Communication System. *Behavior Modification, 25,* 725-744.

Bott, C., Farmer, R., & Rohde, J. (1997). Behavior problems associated with lack of speech in people with learning disabilities. *Journal of Intellectual Disability Research, 41,* 3-7.

Boucher, J., Bigham, S., Mayes, A., & Muskett, T. (2008). Recognition and language in low functioning autism. *Journal of Autism and Developmental Disorders, 38,* 1259-1269.

Boucher, J., Mayes, A., & Bigham, S. (2007). Memory, language and intellectual ability in low-functioning autism. In: J. Boucher & D. Bowler (Eds.), *Memory in autism: Theory and evidence* (pp. 268-290). Cambridge, UK: Cambridge University Press.

Bradley, E. A., Summers, J. A., Wood, H. L., & Bryson, S. E. (2004). Comparing rates of psychiatric and behavior disorders in adolescents and young adults with severe intellectual disability with and without autism. *Journal of Autism and Developmental Disorders, 34,* 151-161.

Bradshaw, J. (2001). Complexity of staff communication and reported level of understanding skills in adults with intellectual disability. *Journal of Intellectual Disability Research, 45,* 233-243.

Bradshaw, J. (2002). The management of challenging behavior within a communication framework. In S. Abudarham & A. Hurd (Eds.), *Management of communication needs of people with learning disability* (pp. 246-275). London: Whurr Publishers.

Bruinsma, Y., Koegel, R. L., & Koegel, L. K. (2004). Joint attention and children with autism: A review of the literature. *Mental Retardation and Developmental Disabilities Research Reviews, 10,* 169-174.

Buckley, B. (2003). *Children's communicative skills. From birth to five years.* London, UK: Routledge.

Callaghan, T.C. (2000). Factors affecting children's graphic symbol use in the third year: Language, similarity, and iconicity. *Cognitive Development, 15,* 195-214.

Calloway, C. J., Myles, B. S., & Earles, T. L. (1999). The development of communicative functions and means in students with autism. *Focus on Autism and Other Developmental Disabilities, 14*, 140-149.

Camaioni, L. (1997). The emergence of intentional communication in ontogeny, phylogeny, and pathology. *European Psychologist, 2,* 216-225.

Camaioni, L., Perucchini, P., Muratori, F., Parrini, B., & Cesari, A. (2003). The communicative use of pointing in autism: Developmental profile and factors related to change. *European Psychiatry, 18,* 6-12.

Carpenter, M., Nagell, K., Tomasello, M., Butterworth, G., & Moore, C. (1998). Social cognition, joint attention, and communicative competence from 9 to 15. *Monographs of the Society for Research in Child Development, 63*, 1-174.

Carpenter, M., & Tomasello, M. (2000). Joint attention, cultural learning, and language acquisition: Implications for children with autism. In A. M. Wetherby, & B. M. Prizant (Eds.), *Autism spectrum disorders: A transactional developmental perspective* (pp. 31-54). Baltimore, MD: Brookes.

Cascella, P. W. (2004). Receptive communication abilities among adults with significant disability. *Journal of Intellectual and Developmental Disability, 29,* 70-78.

Chamberlain, L., Chung, M. C., & Jenner, L. (1993). Preliminary findings on communication and challenging behavior in learning difficulty. *The British Journal of Developmental Disabilities, 35,* 118-125.

Chandler, S., Charman, T., Baird, G., Simonoff, E., Loucas, T., Meldrum, D., Scott, M., & Pickles, A. (2007). Validation of the Social Communication Questionnaire in a population cohort of children with autism spectrum disorders. *Journal of American Academic Child and Adolescent Psychiatry, 46,* 1324-1332.

Charman, T., & Baird, G. (2002). Practitioner review: Diagnosis of autism spectrum disorder in 2- and 3-year-old children. *Journal of Child Psychology and Psychiatry, 43,* 289-305.

Charman, T., Baron-Cohen, S., Swettenham, J., Baird, G., Drew, A., & Cox, A. (2003). Predicting language outcome in infants with autism and pervasive developmental disorder. *International Journal of Language and Communication Disorders, 38*, 265-285.

Charman, T., Pickles, A., Simonoff, E., Chandler, S., Loucas, T., & Baird, G. (2011). IQ in children with autism spectrum disorders: Data from the Special Needs and Autism Project (SNAP). *Psychological Medicine, 41,* 619-627.

Chiang, C.-H., Song, W.-T., Lin, T.-L., & Rogers, S. J. (2008). Nonverbal communication skills in young children with autism. *Journal of Autism and Developmental Disorders, 38*, 1898-1906.

Chiang, H.-M. (2008). Expressive communication of children with autism: The use of challenging behavior. *Journal of Intellectual Disability Research, 52*, 966-972.

Chung, M. C., Jenner, L., Chamberlain, L., & Corbett, J. (1995). One year follow up pilot study on communication skill and challenging behavior. *The European Journal of Psychiatry, 9,* 83-95.

Cohen, J., Cohen, P., West, S. G., & Aiken, L. S. (2003). *Applied multiple regression/correlation analysis for the behavioral sciences.* Mahwah, NJ: Lawrence Erlbaum.

Colgan, S. E., Lanter, E., McComish, C., Watson, L. R., Crais, E. R., & Baranek, G. T. (2006). Analysis of social interaction gestures in infants with autism. *Child Neuropsychology, 12,* 307-319.

Constantino, J. N. (2011). The quantitative nature of autistic social impairment. *Pediatric Research, 69,* 55R-62R.

Constantino, J. N., & Todd, R. D. (2003). Autistic traits in the general population: A twin study. *Archives of General Psychiatry, 60,* 524-530.

Corsello, C., Hus, V., Pickles, A., Risi, S., Cook, E. H. Jr., Leventhal, B. L., & Lord, C. (2007). Between a ROC and a hard place: Decision making and making decisions about using the SCQ. *Journal of Child Psychology and Psychiatry, 48,* 932-940.

Coupe O'Kane, J., & Goldbart, J. (1998). *Communication before speech: Development and assessment.* London, UK: David Fulton Publishers.

Crais, E., Douglas, D. D., & Campbell, C. C. (2004). The intersection of the development of gestures and intentionality. *Journal of Speech, Language, and Hearing Research, 47,* 678-694.

Dawson, G., Meltzoff, A. N., Osterling, J., Rinaldi, J., & Brown, E. (1998). Children with autism fail to orient to naturally occurring social stimuli. *Journal of Autism and Developmental Disorders, 28,* 479-485.

Dawson, G., Toth, K., Abbott, R., Osterling, J., Munson, J., Estes, A., & Liaw, J. (2004). Early social attention impairments in autism: Social orienting, joint attention, and attention to distress. *Developmental Psychology, 40*, 271-283.

Day, H. M., Horner, R. H., & O'Neill, R. E. (1994). Multiple functions of problem behaviors: Assessment and intervention. *Journal of Applied Behavior Analysis, 27,* 279-289.

De Bildt, A., De Jong, M., Lord, C., Rutter, M., DiLavore, P., & Risi, S. (2008). *ADOS Autisme Diagnostisch Observatie Schema*. Amsterdam, NL: Hogrefe.

De Bildt, A., Sytema, S., Ketelaars, C., Kraijer, D., Mulder, E., Volkmar, F., & Minderaa, R. (2004a). Interrelationship between Autism Diagnostic Observation Schedule-Generic (ADOS-G), Autism Diagnostic Interview-Revised (ADI-R), and the Diagnostic and Statistical Manual of Mental Disorders (DSM-IV-TR) classification in children and adolescents with mental retardation. *Journal of Autism and Developmental Disorders, 34,* 129-137.

De Bildt, A., Sytema, S., Ketelaars, C., Kraijer, D., Volkmar, F., & Minderaa, R. (2003). Measuring pervasive developmental disorders in children and adolescents with mental retardation: A comparison of two screening instruments used in a study of the total mentally retarded population from a designated area. *Journal of Autism and Developmental Disorders, 33,* 595-603.

De Bildt, A., Sytema, S., Kraijer, D., & Minderaa, R. (2004b). Prevalence of pervasive developmental disorders in children and adolescents with mental retardation. *Journal of Child Psychology and Psychiatry, 46,* 275-286.

Delinicolas, E. K., & Young, R. L. (2007). Joint attention, language, social relating, and stereotypical behaviors in children with autistic disorder. *Autism, 11*, 425-436.

DeLoache, J. (1989). The development of representation in young children. In H. Reese (Ed.), *Advances in child development and behavior* (pp. 2-39). New York: Academic Press.

DeLoache, J. S. (1991). Symbolic functioning in very young children: Understanding of pictures and models. *Child Development, 62,* 736-752.

DeLoache, J. S. (1995). Early symbol understanding and use. In D. Medin (Ed.), *The psychology of learning and motivation* (pp. 65-114). New York, NY: Academic Press.

DeLoache, J. S. (2004). Becoming symbol-minded. *Trends in Cognitive Sciences, 8,* 66-70.

DeLoache, J. S., & Burns, N. M. (1994). Early understanding of the representational function of pictures. *Cognition, 52,* 83-110.

De Los Reyes, A., & Kazdin, A. E. (2005). Informant discrepancies in the assessment of childhood psychopathology: A critical review, theoretical framework, and recommendations for further study. *Psychological Bulletin, 131,* 483-509.

Denmark, J. C. (1994). *Deafness and mental health.* London, UK: Jessica Kingsley.

Drew, A., Baird, G., Taylor, E., Milne, E., & Charman, T. (2007). The Social Communication Assessment for Toddlers with Autism (SCATA): An instrument to measure the frequency, form and function of communication in toddlers with autism spectrum disorder. *Journal of Autism and Developmental Disorders, 37,* 648-666.

Dye, M. W. G., Hauser, P. C., & Bavelier, D. (2008). Visual attention in deaf children and adults: implications for learning environments. In M. Marschark & P. C. Hauser (Eds.), *Deaf cognition: Foundations and outcomes* (pp. 250-263). New York, NY: Oxford University Press.

Dye, M. W. G., Hauser, P. C. & Bavelier, D. (2009). Is visual selective attention in deaf individuals enhanced or deficient? The case of the useful field of view. *PLoSONE 4(5): e5640. doi:10.1371/journal.pone.0005640*.

Eagle, R. S. (2002). Accessing and assessing intelligence in individuals with lower functioning autism. *Journal of Developmental Disabilities, 9*, 45-53.

Eigsti, I. M., De Marchena, A. B., Schuh, J. M., & Kelley, E. (2011). Language acquisition in autism spectrum disorders: A developmental review. *Research in Autism Spectrum Disorders, 5,* 681-691.

Evenhuis, H., Van Splunder, J., Vink, M., Weerdenburg, C., Van Zanten, B., & Stilma, J. (2004). Obstacles in large-scale epidemiological assessment of sensory impairments in a Dutch population with intellectual disabilities. *Journal of Intellectual Disability Research, 48*, 708-718.

Evers, A., Braak, M. S. L., Frima, R. M., & Van Vliet-Mulder, J. C. (2009). *Commissie Testaangelegenheden Nederland – Documentatie* [Committee for Psychometric Evaluation of Psychological Tests of the Dutch Institute of Psychologists – Documentation]. Amsterdam, NL: Boom Test Uitgevers.

Fenson, L., Bates, E., Dale, P., Goodman, J., Steven Reznick, J., & Thal, D. (2000a). Measuring variability in early child language: Don't shoot the messenger. *Child Development, 71*, 323-328.

Fenson, L., Dale, P., Reznick, J., Bates, E., Thal, D., & Pethick, S. (1994). Variability in early communicative development. *Monographs of the Society for Research in Child Development, 59*, 1-73.

Fenson, L., Pethick, S., Renda, C., Cox, J. L., Dale, P. S., & Reznick, J. S. (2000b). Short-form versions of the MacArthur Communicative Development Inventories. *Applied Psycholinguistics, 21,* 95-116.

Filipek, P. A., Accardo, P. J., Baranek, G. T., Cook, E. H. Jr., Dawson, G., Gordon, B., ... & Volkmar, F. R. (1999). The screening and diagnosis of autistic spectrum disorders. *Journal of Autism and Developmental Disorders, 29*, 439-484.

Fombonne, E. (2005). Epidemiological studies of pervasive developmental disorders. In F. Volkmar, R. Paul, A. Klin, & D. J. Cohen (Eds.). *Handbook of autism and pervasive developmental disorders* (3rd edn) (pp. 42-69). New York, NY: Wiley.

Fombonne, E. (2009). Epidemiology of pervasive developmental disorders. *Pediatric Research, 65,* 591-598.

Förster, J. (2009). Relations between perceptual and conceptual scope: how global versus local processing fits a focus on similarity versus dissimilarity. *Journal of Experimental Psychology: General, 138,* 88-111.

Fossett, B., & Mirenda, P. (2007). Augmentative and alternative communication. In S. L. Odom, R. H. Horner, M. E. Snell, & J. Blacher (Eds.), *Handbook of developmental disabilities* (pp. 330-348). New York, NY: Guilford Press.

Freeman, N. L., Perry, A., & Bebko, J. M. (2002). Behavior is communication: Nonverbal communicative behavior in students with autism and instructors' responsivity. *Journal on Developmental Disabilities, 9*, 145-155.

Frith, U. (1989). *Autism. Explaining the enigma.* Oxford, UK: Blackwell.

Gallaudet Research Institute [GRI] (2007). *Regional and national data from 2006-2007: annual survey of deaf and hard of hearing children and youth.* Washington, DC: Gallaudet University.

Glaser, S., Ouimet, T., & Shaw, S. R. (2010). Genetics of autism: Theories, findings, and implications. In P. McCabe & S.R. Shaw (Eds.), *Pediatric disorders: Current topics and interventions for educators.* Thousand Oaks, CA: Sage/Corwin Press.

Goin-Kochel, R. P., Peters S. U., & Treadwell-Deering, D. (2008). Parental reports on the prevalence of co-occurring intellectual disability among children with autism spectrum disorders. *Research in Autism Spectrum Disorders, 2,* 546-556.

Goldstein, H. (2002). Communication intervention for children with autism: A review of treatment efficacy. *Journal of Autism and Developmental Disorders, 32,* 373-396.

Goodhart, F., & Baron-Cohen, S. (1993). How many ways can the point be made? Evidence from children with and without autism. *First Language, 13*, 225-233.

Gotham, K., Risi, S., Pickles, A., & Lord, C. (2007). The Autism Diagnostic Observation Schedule: Revised algorithms for improved diagnostic validity. *Journal of Autism and Developmental Disabilities, 37,* 613-627.

Gotham, K., Risi, S., Dawson, G., Tager-Flusberg, H., Joseph, R., Carter, A., ... & Lord, C. (2008). A replication of the Autism Diagnostic Observation Schedule (ADOS) revised algorithms. *Journal of the American Academy of Child & Adolescent Psychiatry, 47,* 642-651.

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

Granlund, M., & Olsson, C. (1999). Efficacy of communication intervention for presymbolic communicators. *Augmentative and Alternative Communication, 15,* 25-37.

Gray, K. M., Tonge, B. J., & Sweeney, D. J. (2008). Using the Autism Diagnostic Interview-Revised and the Autism Diagnostic Observation Schedule with young children with developmental delay: Evaluating diagnostic validity. *Journal of Autism and Developmental Disorders, 38,* 657-667.

Hammes, J. G. W., & Langdel, T. (1981). Precursors of symbol formation and childhood autism. *Journal of Autism and Developmental Disorders, 11,* 331-346.

Hanley, G. P., Iwata, B. A., & McCord, B. E. (2003). Functional analysis of problem behavior: A review. *Journal of Applied Behavior Analysis, 36,* 147-185.

Happé, F. G. E., & Booth, R. D. L. (2008). The power of the positive: revisiting weak coherence in autism spectrum disorders. *The Quarterly Journal of Experimental Psychology, 61,* 50-63.

Happé, F., & Frith, U. (2006). The weak coherence account: detail-focused cognitive style in autism spectrum disorders. *Journal of Autism and Developmental Disorders, 36,* 5-25.

Harrington, J. W. (2010). The actual prevalence of autism: Are we there yet? *Pediatrics, 126,* e1257-e1258.

Harris, J. M., Best, C. S., Moffat, V. J., Spencer, M. D., Philip, R. C. M., Power, M. J., & Johnstone, E. C. (2007). Autistic traits and cognitive performance in young people with mild intellectual impairment. *Journal of Autism and Developmental Disorders, 38,* 1241-1249.

Hill, E. L., & Frith, U. (2003). Understanding autism: Insights from mind and brain. *Philosophical Transactions of the Royal Society of London, Series B, 358,* 281-289.

Hodapp, R. M., & Dykens, E. M. (2001). Strengthening behavioral research on genetic mental retardation syndromes. *American Journal on Mental Retardation, 106,* 4-15.

Howlin, P. (2006). Augmentative and alternative communication systems for children with autism. In T. Charman, & W. Stone (Ed.), *Social & communication development in autism spectrum disorders: Early identification, diagnosis, & intervention* (pp. 236-266). New York, NY: Guilford Press.

Hudry, K., Leadbitter, K., Temple, K., Slonims, V., McConachie, H., Aldred, C., Howlin, P., Charman, T., & PACT Consortium (2010). Preschoolers with autism show greater impairment in receptive compared with expressive language abilities. *International Journal of Language and Communication Disorders, 45,* 681-690.

Iacono, T., Carter, M., & Hook, J. (1998). Identification of intentional communication in students with severe and multiple disabilities. *Augmentative and Alternative Communication, 14,* 102-114.

Jarrold, C., Boucher, J., & Russell, J. (1997). Language profiles in children with autism: Theoretical and methodological implications. *Autism, 1,* 57-76.

Joseph, R. M., Tager-Flusberg, H., & Lord, C. (2002). Cognitive profiles and social-communicative functioning in children with autism spectrum disorder. *Journal of Child Psychology and Psychiatry, 43,* 807-821.

Kamp-Becker, I., Smidt, J., Ghahreman, M., Heinzel-Gutenbrunner, M., Becker, K., & Remschmidt, H. (2010). Categorical and dimensional structure of autism spectrum disorders: The nosologic validity of Asperger syndrome. *Journal of Autism and Developmental Disorders, 40,* 921-929.

Kasari, C., Paparella, T., Freeman, S., & Jahromi, L. B. (2008). Language outcome in autism: Randomized comparison of joint attention and play interventions. *Journal of Consulting and Clinical Psychology, 76,* 125-137.

Kevan, F. (2003). Link between communication and challenging behavior. *British Journal of Learning Disabilities, 31,* 75-80.

Kielinen, M., Rantala, H., Timonen, E., Linna, S. L., & Moilanen, I. (2004). Associated medical disorders and disabilities in children with autistic disorder. *Autism, 8,* 49-60.

Kjelgaard, M. M., & Tager-Flusberg, H. (2001). An investigation of language impairment in autism: Implications for genetic subgroups. *Language and Cognitive Processes, 2001,* 287-308.

Klin, A., Jones, W., Schultz, R., & Volkmar, F. (2003). The enactive mind, or from actions to cognition: Lessons from autism. *Philosophical Transactions of the Royal Society of London, 358,* 345-360.

Klin, A., Jones, W., Schultz, R., Volkmar, F., & Cohen, D. (2002). Visual fixation patterns during viewing of naturalistic social situations as predictors of social competence in individuals with autism. *Archives of General Psychiatry, 59,* 809-816.

Klin, A., Saulnier, C. A., Sparrow, S. S., Cicchetti, D. V., Volkmar, F. R., & Lord, C. (2007). Social and communication abilities and disabilities in higher functioning individuals with autism spectrum disorders: The Vineland and the ADOS. *Journal of Autism and Developmental Disorders, 37,* 748-759.

Klin, A., Saulnier, C., Tsatsanis, K., & Volkmar, F. R. (2005). Clinical evaluation in autism spectrum disorders: Psychological assessment within a transdisciplinary framework. In F. R. Volkmar, R. Paul, A. Klin, & D. Cohen (Eds.), *Handbook of autism and pervasive developmental disorders* (3rd ed.) (pp. 772-798). Hoboken, NJ: Wiley.

Konstantareas, M. M., & Homatidis, S. (1989). Assessing child symptom severity and stress in parents of autistic children. *Journal of Child Psychology and Psychiatry, 30,* 459-470.

Kopp, S., & Gillberg, C. (2011). The Autism Spectrum Screening Questionnaire (ASSQ)-Revised Extended Version (ASSQ-REV): An instrument for better capturing the autism phenotype in girls? A preliminary study involving 191 clinical cases and community controls. *Research in Developmental Disabilities, 32,* 2875-2888.

Landa, R. (2007). Early communication development and intervention for children with autism. *Mental Retardation and Developmental Disabilities Research Reviews, 13,* 16-25.

Landa, R. J., Holman, K. C., & Garrett-Mayer, E. (2007). Social and communication development in toddlers with early and later diagnosis of autism spectrum disorders. *Archives of General Psychiatry, 64,* 853-864.

Lecavalier, L., Leone, S., & Wiltz, J. (2006). The impact of behavior problems on caregiver stress in young people with autism spectrum disorders. *Journal of Intellectual Disability Research, 50,* 172-183.

Leekam, S. R., Libby, S. J., Wing, L., Gould, J., & Taylor, C. (2002). The Diagnostic Interview for Social and Communication Disorders: Algorithms for ICD-10 Childhood Autism and Wing & Gould autistic spectrum disorder. *Journal of Child Psychology and Psychiatry, 43,* 327-347.

Lewis, V., Boucher, J., Lupton, L., & Watson, S. (2000). Relationships between symbolic play, functional play, verbal and non-verbal ability in young children. *International Journal of Language and Communication Disorders, 35,* 117-127.

Lilienfield, S. O., & Landfield, K. (2008). Issues in diagnosis: categorical vs. dimensional. In W. E. Craighead, D. J. Miklowitz, & L. W. Craighead (Eds.), *Psychopathology: History, diagnosis, and empirical foundations* (pp. 1-33). Hoboken, NJ: Wiley.

Liss, M., Harel, B., Fein, D., Allen, D., Dunn, M., Feinstein, C., ... & Rapin, I. (2001). Predictors and correlates of adaptive functioning in children with developmental disorders. *Journal of Autism and Developmental Disorders, 31,* 219-230.

Lopez, B., Leekam, S. R., & Arts, G. R. J. (2008). How central is central coherence? Preliminary evidence on the link between conceptual and perceptual processing in children with autism. *Autism, 12,* 159-171.

Lord, C. (2011). How common is autism? *Nature, 474,* 166-168.

Lord, C., & Bishop, S. L. (2010). Autism spectrum disorders: Diagnosis, prevalence, and services for children and families. *Social Policy Report, 24,* 1-27.

Lord, C., & McGee, J. P. (2001). *Educating children with autism.* Washington, DC: National Academy Press.

Lord, C., Risi, S., Lambrecht, L., Cook, E. H. Jr., Leventhal, B. L., DiLavore, P. C., ... & Rutter, M. (2000). The Autism Diagnostic Observation Schedule – Generic: A standard measure of social and communication deficits associated with the spectrum of autism. *Journal of Autism and Developmental Disorders, 30,* 205-223.

Lord, C., Risi, S., & Pickles, A. (2004). Trajectory of language development in autistic spectrum disorders. In M. L. Rice & S. F. Warren (Eds.), *Developmental language disorders: From phenotypes to etiologies* (pp. 7-29). Mahwah, NJ: Lawrence Erlbaum Associates.

Lord, C., Rutter, M., DiLavore, P. C., & Risi, S. (1999). *Autism Diagnostic Observation Schedule (ADOS).* Los Angeles, CA: Western Psychological Services.

Lord, C., Rutter, M., & Le Couteur, A. (1994). Autism Diagnostic Interview-Revised: A revised version of a diagnostic interview for caregivers of individuals with possible pervasive developmental disorders. *Journal of Autism Developmental Disorders, 24,* 659-685.

Loveland, K. A., & Kelley, M. L. (1991). Development of adaptive behavior in preschoolers with autism or Down syndrome. *American Journal on Mental Retardation, 96,* 13-20.

Luyster, R. J., Kadlec, M. B., Carter, A., & Tager-Flusberg, H. (2008). Language assessment and development in toddlers with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 38,* 1426-1438.

MacDuff, G. S., Krantz, P. J., & McClannahan, L. E. (1993). Teaching children with autism to use photographic activity schedules: Maintenance and generalization of complex response chains. *Journal of Applied Behavior Analysis, 26,* 89-97.

Maljaars, J., & Noens, I. (2008). Communicatie zonder representatie: ondersteunende communicatie op maat. *Wetenschappelijk Tijdschrift Autisme, 2,* 58-66.

Marschark, M. (2005). *Psychological development of deaf children.* New York, NY: Oxford University Press.

Matson, J. L., & Shoemaker, M. (2009). Intellectual disability and its relationship to autism spectrum disorders. *Research in Developmental Disabilities, 30,* 1170-1114.

Matson, J. L., Wilkins, J., & Ancona, M. (2008). Autism in adults with severe intellectual disability: An empirical study of symptom presentation. *Journal of Intellectual and Developmental Disability, 33,* 36-42.

Mawhood, L., Howlin, P., & Rutter, M. (2000). Autism and developmental receptive language disorder: A comparative follow-up in early adult life. I: Cognitive and language outcomes. *Journal of Child Psychology and Psychiatry, 41,* 547-559.

McClintock, K., Hall, S., & Oliver, C. (2003). Risk markers associated with challenging behaviors in people with intellectual disabilities: A meta-analytic study. *Journal of Intellectual Disability Research, 47,* 405-416.

McConkey, R., Morris, I., & Purcell, M. (1999). Communications between staff and adults with intellectual disabilities in naturally occurring settings. *Journal of Intellectual Disability Research, 43,* 194-205.

McCune, L. (1995). A normative study of representational play at the transition to language. *Developmental Psychology, 31,* 198-206.

McDuffie, A., Yoder, P., & Stone, W. (2005). Prelinguistic predictors of vocabulary in young children with autism spectrum disorders. *Journal of Speech, Language and Hearing Research, 48,* 1080-1097.

McFee, K. H. L. (2006). *Understanding the symbolic nature of pictures in children with autism* [Master's thesis]. York University.

McFee, K. H. L. (2011). *Evaluation of child variables related to outcomes of the Picture Exchange Communication System in children with autism spectrum disorders*. Dissertation, York University.

McLean, L. K. (1993). Communication intervention for adults with severe mental retardation. *Topics in Language Disorders, 13,* 47-60.

Mervis, C. B., & Klein-Tasman, B. P. (2004). Methodological issues in group-matching designs: α levels for control variable comparisons and measurement characteristics of control and target variables. *Journal of Autism and Developmental Disorders, 34,* 7-17.

Meuwese-Jongejeugd, A., Vink, M., Van Zanten, B., Verschuure, H., Bernsen, R., & Evenhuis, H. (2006). Prevalence of hearing impairment in 1,598 adults with an intellectual disability: cross-sectional population-based study. *International Journal of Audiology, 45,* 660-669.

Miniscalco, C., Fränberg, J., Schachinger-Lorentzon, U., & Gillberg, C. (2011). Meaning what you say? Comprehension and word production skills in young children with autism. *Research in Autism Spectrum Disorders,* doi: 10.1016/j.rasd.2011.05.001.

Minshew, N. J., Meyer, J., & Goldstein, G. (2002). Abstract reasoning in autism: A disassociation between concept formation and concept identification. *Neuropsychology, 16,* 327-334.

Mottron, L., Dawson, M., Soulières, I., Hubert, B., & Burack, J. (2006). Enhanced perceptual functioning in autism: an update and eight principles of autistic perception. *Journal of Autism and Developmental Disorders, 36,* 27-43.

Müller, R. (2007). The study of autism as a distributed disorder. *Mental Retardation and Developmental Disabilities Research Reviews, 13,* 85-95.

Mundy, P. & Burnette, C. (2005). Joint attention and neurodevelopment. In F.R. Volkmar, R. Paul, A. Klin, & D. Cohen (Eds.), *Handbook of autism and pervasive developmental disorders. Volume I: Diagnosis, development, neurobiology, and behavior* (3rd ed.) (pp. 650-681). Hoboken, NJ: Wiley.

Mundy, P., & Gomes, A. (1998). Individual differences in joint attention skill development in the second year. *Infant Behavior and Development, 21*, 469-482.

Mundy, P., Gwaltney, M., & Henderson, H. (2010). Self-referenced processing, neurodevelopment and joint attention in autism. *Autism, 14,* 408-429.

Mundy, P., & Jarrold, W. (2010). Infant joint attention, neural networks and social cognition. *Neural Networks, 23,* 985-997.

Mundy, P., Kasari, C., Sigman, M., & Ruskin, E. (1995). Nonverbal communication and early language acquisition in children with Down syndrome and in normally developing children. *Journal of Speech and Hearing Research, 38,* 157-167.

Mundy, P., Sigman, M., & Kasari, C. (1990). A longitudinal study of joint attention and language development in autistic children. *Journal of Autism and Developmental Disorders, 20*, 115-128.

Mundy, P., Sigman, M., & Kasari, C. (1994). Joint attention, developmental level, and symptom presentation in autism. *Development and Psychopathology, 6,* 389-401.

Mundy, P., Sullivan, L., & Mastergeorge, A. M. (2009). A parallel and distributed-processing model of joint attention, social cognition and autism. *Autism Research, 2,* 2-21.

Naber, F. B. A., Bakermans-Kranenburg, M. J., Van IJzendoorn, M. H., Dietz, C., Van Daalen, E., Swinkels, S. H. N., ... & Van Engeland, H. (2008). Joint attention development in toddlers with autism. *European Child and Adolescent Psychiatry, 17,* 143-152.

Nakahachi, T., Yamashita, K., Iwase, M., Ishigami, W., Tanaka, C., Toyonaga, K., ... & Takeda, M. (2008). Disturbed holistic processing in autism spectrum disorders verified by two cognitive tasks requiring perception of complex visual stimuli. *Psychiatry Research, 156,* 330-338.

Namy, L. L. (2005). *Symbol use and symbolic representation: Developmental and comparative perspectives.* Mahwah, NJ: Erlbaum.

Namy, L. L., & Waxman, S. R. (2005). Symbols redefined. In L. L. Namy (Ed.), *Symbol use and symbolic representation* (pp. 269-277). Mahwah, NJ: Lawrence Erlbaum Associates.

Nilsen, E. S., & Fecica, A. M. (2011). A model of communicative perspective-taking for typical and atypical populations of children. *Developmental Review, 31,* 55-78.

Noens, I., & Van Berckelaer-Onnes, I. A. (2004). Making sense in a fragmentary world: Communication in people with autism and learning disability. *Autism, 8,* 197-218.

Noens, I., & Van Berckelaer-Onnes, I. A. (2005). Captured by details: Sense-making, language and communication in autism. *Journal of Communication Disorders, 38,* 123-141.

Noens, I. L. J., & Van Berckelaer-Onnes, I. A. (2008). The central coherence account of autism revisited: evidence from the ComFor study. *Research in Autism Spectrum Disorders, 2,* 209-222.

Noens, I., Van Berckelaer-Onnes, I., Verpoorten, R., & Van Duijn, G. (2006). The ComFor: an instrument for the indication of augmentative communication in people with autism and intellectual disability. *Journal of Intellectual Disability Research, 50,* 621-632.

Noor, A., Whibley, A., Marshall, C. R., Gianakopoulos, P. J., Piton, A., Carson, A. R. ... & Vincent, J. B. (2010). Disruption at the PTCHD1 locus on Xp22.11 in autism spectrum disorder and intellectual disability. *Science Translational Medicine, 2,* doi:10.1126/scitranslmed.3001267.

Nygren, G., Hagberg, B., Billstedt, E., Skoglund, A., Gillberg, C., & Johansson, M. (2009). The Swedish version of the Diagnostic Interview for Social and Communication Disorders (DISCO-10). Psychometric properties. *Journal of Autism and Developmental Disorders, 39,* 730-741.

Paul, R. (2005). Assessing communication in autism spectrum disorder. In F. R. Volkmar, R. Paul, A. Klin, & D. J. Cohen (Eds.), *Handbook of autism and pervasive developmental disorders* (3rd ed.) (pp. 799-816). Hoboken, NJ: Wiley.

Paul, R. (2007). Communication and its development in autism spectrum disorders. In F. R. Volkmar (Ed.), *Autism and pervasive developmental disorders* (2nd ed.) (pp. 129-155). Cambridge, UK: University Press.

Perry, A., Flanagan, H. E., Dunn Geier, J., & Freeman, N. L. (2009). Brief report: The Vineland Adaptive Behavior Scales in young children with autism spectrum disorders at different cognitive levels. *Journal of Autism and Developmental Disorder, 39,* 1066-1078.

Posserud, M., Lundervold, A. J., Lie, S. A., & Gillberg, C. (2009). The prevalence of autism spectrum disorders: impact of diagnostic instrument and non-response bias. *Social Psychiatry and Psychiatric Epidemiology, 45,* 319-327.

Preissler, M. A. (2008). Associative learning of pictures and words by low-functioning children with autism. *Autism, 12,* 231-248.

Prior, M., & Ozonoff, S. (1998). Psychological factors in autism. In F.R. Volkmar (Ed.), *Autism and pervasive developmental disorders* (2nd ed.) (pp. 64-108). Cambridge, UK: Cambridge University Press.

Prizant, B. M. (1996). Brief report: Communication, language, social, and emotional development. *Journal of Autism and Developmental Disorders, 26,* 173-178.

Quill, K. A. (1997). Instructional considerations for young children with autism: The rationale for visually cued instruction. *Journal of Autism and Developmental Disorders, 27,* 697-714.

Rapin, I., & Dunn, M. (2003). Update on the language disorders of individuals on the autistic spectrum. *Brain and Development, 25,* 166-172.

Reese, R. M., Richman, D. M., Belmont, J. M., & Morse, P. (2005). Functional characteristics of disruptive behavior in developmentally disabled children with and without autism. *Journal of Autism and Developmental Disorders, 35,* 419-428.

Rettenbach, R., Diller, G., & Sireteanu, R. (1999). Do deaf people see better? Texture segmentation and visual search compensate in adult but not in juvenile subjects. *Journal of Cognitive Neuroscience, 11,* 560-583.

Reynell, J. K. (1985). *Reynell Developmental Language Scales.* Windsor, UK: NFER-Nelson.

Reynolds, H. N. (1993). Effects of foveal stimulation on peripheral visual processing and laterality in deaf and hearing subjects. *The American Journal of Psychology, 106,* 523-540.

Ricks, D. M., & Wing, L. (1975). Language, communication, and the use of symbols in normal and autistic children. *Journal of Autism and Childhood Schizophrenia, 5,* 191-221.

Risi, S., Lord, C., Gotham, K., Corsello, C., Chrysler, C., Szatmari, P., ... Pickles, A. (2006). Combining information from multiple sources in the diagnosis of autism spectrum disorders. *Journal of the American Academy of Child & Adolescent Psychiatry, 45*, 1094-1103.

Rogers, S.J., & Dawson, G. (2010). Early Start Denver Model for young children with autism: Promoting language, learning, and engagement. New York, NY: The Guilford Press.

Roper, L., Arnold, P., & Monteiro, B. (2003). Co-occurrence of autism and deafness. *Autism, 7,* 245-253.

Rosenhall, U., Nordin, V., Sandström, M., Ahlsén, G., & Gilberg, G. (1999). Autism and hearing loss. *Journal of Autism and Developmental Disorders, 29,* 349-357.

Rowland, C., & Schweigert, P. (2000). Tangible symbols, tangible outcomes. *Augmentative and Alternative Communication, 16,* 61-78.

Rudie, J. D., Shehzad, Z., Hernandez, L. M., Colich, N. L., Bookheimer, S. Y., Iacoboni, M., & Dapretto, M. (2011). Reduced functional integration and segregation of distributed neural systems underlying social and emotional information processing in autism spectrum disorders. *Cerebral Cortex,* doi: 10.1093/cercor/bhr171.

Rutter, M., Bailey, A., & Lord, C. (2003). *SCQ: Social Communication Questionnaire*. Los Angeles, CA: Western Psychological Services.

Schalock, R. L., Borthwick-Duffy, S. A., Bradley, V. J., Buntinx, W. H. E., Coulter, D. L., Craig, E. M., ... & Yeager, M. H. (2010). *Intellectual disability: Definition, classification, and systems of support* (11th ed.). Washington, DC: American Association on Intellectual and Developmental Disabilities.

Schlichting, J. E. P. T., Van Eldik, M. C. M., Spelberg, H. C., Van der Meulen, S., & Van der Meulen, B. F. (1995). *Schlichting Test voor Taalproductie.* [Schlichting Test for Language production]. Nijmegen, NL: Berkhout.

Scholte, E., Van Duijn, G., Dijkxhoorn, Y., Noens, I., & Van Berckelaer-Onnes, I. (2008). *Handleiding Vineland Screener 0-6* [Manual Vineland Screener 0-6yrs-NL]. Leiden, NL: PITS.

Schuler, A. L., Prizant, B. M., & Wetherby, A. M. (1997). Enhancing language and communication development: Prelinguistic approaches. In D.J. Cohen & F.R. Volkmar (Eds.), *Handbook of autism and pervasive developmental disorders* (2nd ed., pp. 513-538). New York, NY: John Wiley & Sons.

Serna, R. W., Dube, W. V., & McIlvane, W. J. (1997). Assessing same/different judgments in individuals with severe intellectual disabilities: A status report. *Research in Developmental Disabilities, 18,* 343-368.

Shumway, S., & Wetherby, A. M. (2009). Communicative acts of children with autism spectrum disorders in the second year of life. *Journal of Speech, Language, and Hearing Research, 52,* 1139-1156.

Sigafoos, J. (2000). Communication development and aberrant behavior in children with developmental disabilities. *Education and Training in Mental Retardation and Developmental Disabilities, 35,* 168-176.

Siller, M., & Sigman, M. (2002). The behaviors of parents of children with autism predict the subsequent development of their children's communication. *Journal of Autism and Developmental Disorders, 32,* 77-90.

Smith, V., Mirenda, P., & Zaidman-Zait, A. (2007). Predictors of expressive vocabulary growth in children with autism. *Journal of Speech, Language, and Hearing Research, 50,* 149-160.

Snell, M. E. (2002). Using dynamic assessment with learners who communicate nonsymbolically. *Augmentative and Alternative Communication, 18*, 163-176.

Snow, A. V., & Lecavalier, L. (2008). Sensitivity and specificity of the Modified Checklist for Autism in Toddlers and the Social Communication Questionnaire in preschoolers suspected of having pervasive developmental disorders. *Autism, 12,* 627-644.

Sparrow, S. S., Carter, A. S., & Cicchetti, D. (1993). *Vineland Screener. overview, reliability, validity, administration and scoring.* New Haven, CT: Yale University.

Stephenson, J., & Linfoot, K. (1996). Pictures as communication symbols for students with severe intellectual disability. *Augmentative and Alternative Communication, 12,* 244-255.

Stone, W. L., & Caro-Martinez, L. M. (1990). Naturalistic observations of spontaneous communication in autistic children. *Journal of Autism and Developmental Disorders, 20*, 437-453.

Stone, W. L., Ousley, O. Y., Yoder, P. J., Hogan, K. L., & Hepburn, S. L. (1997). Nonverbal communication in two- and three-year-old children with autism. *Journal of Autism and Developmental Disorders, 27*, 677-696.

Tager-Flusberg, H. (1997). Language acquisition and theory of mind: Contributions from the study of autism. In L. B. Adamson & M. A. Romski (Eds.), *Research on communication and language disorders: Contributions to theories of language development.* Baltimore, MD: Paul Brookes Publishing.

Tager-Flusberg, H., Paul, R., & Lord, C. (2005). Language and communication in autism. In F. R. Volkmar, R. Paul, A. Klin, & D. Cohen (Eds.), *Handbook of autism and pervasive developmental disorders. Volume I: Diagnosis, development, neurobiology, and behavior* (3rd ed.) (pp. 335-364). Hoboken, NJ: Wiley.

Tager-Flusberg, H., Rogers, S., Cooper, J., Landa, R., Lord, C., Paul, R., ... & Yoder, P. (2009). Defining spoken language benchmarks and selecting measures of expressive language development for young children with autism spectrum disorder. *Journal of Speech, Language, and Hearing Research, 52,* 643-652.

Taylor, E., & Rutter, M. (2002). Classification: Conceptual issues and substantive findings. In M. Rutter & E. Taylor (Eds.), *Child and adolescent psychiatry* (4th ed., pp. 3-17). Malden, MA: Blackwell.

Tellegen, P. J., Winkel, M., Wijnberg-Williams, B. J., & Laros, J. A. (1998). *Snijders-Oomen Niet-verbale intelligentietest, SON-R 2½-7: handleiding en verantwoording.* [Snijders-Oomen Non-verbal intelligence test, SON-R 2½-7: Manual]. Lisse, NL: Swets & Zeitlinger.

Thiemann-Bourque, K. S., Brady, N. C., & Fleming, K. K. (2011). Symbolic play of preschoolers with severe communication impairments with autism and other developmental delays: More similarities than differences. *Journal of Autism and Developmental Disorders,* doi:10.1007/s10803-011-1317-7

Thurm, A., Lord, C., Lee, L.-C., & Newschaffer, C. (2007). Predictors of language acquisition in preschool children with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 37,* 1721-1734.

Toth, K., Munson, J., Meltzoff, A. N., & Dawson, G. (2006). Early predictors of communication development in young children with autism spectrum disorder: Joint attention, imitation, and toy play. *Journal of Autism and Developmental Disorders, 36,* 993-1005.

Totsika, V., Felce, D., Kerr, M., & Hastings, R. P. (2010). Behavior problems, psychiatric symptoms, and quality of life for older adults with intellectual disability with and without autism. *Journal of Autism and Developmental Disorders, 40,* 1171-1178.

Travis, L. L., & Sigman, M. (2001). Communicative intentions and symbols in autism: Examining a case of altered development. In J. A. Burack, T. Charman, N. Yirmiya & P. R. Zelazo (Eds.), *The development of autism. Perspectives from theory and research* (pp. 279-308). Mahwah, NJ: Erlbaum.

Twachtman, D. D. (1995). Enhancing communication in verbal children with ASD. In K.A. Quill (Ed.), *Teaching children with autism: Strategies to enhance communication and socialization* (pp. 133-162). Albany, NY: Delmar.

Twachtman-Cullen, D. (1998). Language and communication characteristics seen in high-functioning autism and Asperger syndrome. In E. Schopler, G.B. Mesibov & L.J. Kunce (Eds.), *Asperger syndrome or high-functioning autism?* (pp. 213-226). New York: Plenum Press.

Twachtman-Cullen, D., & Twachtman-Reilly, J. (2007). Communication and language issues in less able children. In: R. L. Gabriels & D. E. Hill (Eds.), *Growing up with autism: Working with school-age children and adolescents* (pp. 73-93). New York, NY: The Guilford Press.

Van Berckelaer-Onnes, I. A., Noens, I., & Dijkxhoorn, Y. (2008). *Diagnostic Interview for Social and Communication Disorders: Nederlandse Vertaling* [Diagnostic Interview for Social and Communication Disorders: Dutch Translation]. Leiden, NL: Universiteit Leiden.

Van Berckelaer-Onnes, I. A., Van Loon, J., & Peelen, A. (2002). Challenging behaviour: A challenge to change. *Autism, 8,* 123-124.

Van Eldik, M. C. M., Schlichting, J. E. P. T., Spelberg, H. C., Van der Meulen, S., & Van der Meulen, B. F. (1995) *Reynell Test voor Taalbegrip* [Reynell Test for Language comprehension]. Nijmegen, NL: Berkhout.

Van Lang, N. D. J., Bouma, A., Sytema, S., Kraijer, D. W., & Minderaa, R. B. (2006). A comparison of central coherence skills between adolescents with an intellectual disability with and without comorbid autism spectrum disorder. *Research in Developmental Disabilities, 27,* 217-226.

Vernon, M., & Rhodes, A. (2009). Deafness and autistic spectrum disorders. *American Annals of the Deaf, 154,* 5-14.

Verpoorten, R. A. W. (1996). Communicatie met verstandelijk gehandicapte autisten: een multidimensioneel communicatiemodel [Communication with mentally retarded people with autism: A multidimensional communication model]. *Nederlands Tijdschrift voor Zorg aan Verstandelijk Gehandicapten, 22,* 106-120.

Verpoorten, R., Noens, I., & Van Berckelaer-Onnes, I. (2004). *De ComVoor – Voorlopers in communicatie. Handleiding.* [The ComFor – Forerunners in communication. Manual]. Leiden, NL: PITS.

Verpoorten, R., Noens, I. & Van Berckelaer-Onnes, I. (2008). *The ComFor – Forerunners in communication. Manual.* Leiden, NL: PITS.

Viellard, M., Da Fonseca, D., De Martino, S., Girardot, A. M., Bastard-Rosset, D., Duverger, H., … & Poinso, F. (2007). Autisme et retard mental: étude de la communication sociale précoce. *Archives de Pédiatrie, 14,* 234-238.

Vig, S. & Jedrysek, E. (1999). Autistic features in young children with significant cognitive impairment: Autism or mental retardation? *Journal of Autism and Developmental, 29,* 235-248.

Volkmar, F. R. (1998). Categorical approaches to the diagnosis of autism: An overview of DSM-IV and ICD-10. *Autism, 2,* 45-59.

Volkmar, F. R., Cohen, D. J., Bregman, J. D., Hooks, M. Y., & Stevenson, J. M. (1989). An examination of social typologies in autism. *Journal of the American Academy of Child and Adolescent Psychiatry, 28,* 82-86.

Volkmar, F. R., & Lord, C. (1998). Diagnosis and definition of autism and other pervasive developmental disorders. In F. R. Volkmar (Ed.), *Autism and pervasive developmental disorders* (2nd ed.) (pp. 1-32). Cambridge, UK: Cambridge University Press.

Wang, L., Mottron, L., Peng, D., Berthiaume, C., & Dawson, M. (2007). Local bias and local-to-global interference without global deficit: a robust finding in autism under various conditions of attention, exposure time, and visual angle. *Cognitive Neuropsychology, 24,* 550-574.

Warreyn, P., Raymaekers, R., & Roeyers, H. (2004). *SCQ – Vragenlijst Sociale Communicatie* [Social Communication Questionnaire]. Destelbergen, BE: SIG.

Warreyn, P., Roeyers, H., & De Groote, I. (2005). Early social communicative behaviours of preschoolers with autism spectrum disorder during interaction with their mothers. *Autism, 9,* 342-361.

Watt, N., Wetherby, A., & Shumway, S. (2006). Prelinguistic predictors of language outcome at 3 years of age. *Journal of Speech, Language, and Hearing Research, 49,* 1224-1237.

Weismer, S. E., Lord, C., & Esler, A. (2010). Early language patterns of toddlers on the autism spectrum compared to toddlers with developmental delay. *Journal of Autism and Developmental Disorders, 40,* 1259-1273.

Wendt, O. (2009). Research on the use of manual signs and graphic symbols in autism spectrum disorders: A systematic review. In P. Mirenda, & T. Iacono (Eds.), *Autism spectrum disorders and AAC* (pp. 83-117). Baltimore, MD: Brookes.

Wetherby, A. (1986). Ontogeny of communicative functions in autism. *Journal of Autism and Developmental Disorders, 16,* 295-316.

Wetherby, A., & Prizant, B. (1993). *Communication and Symbolic Behavior Scales – Normed edition.* Baltimore, MD: Paul H. Brookes.

Wetherby, A., & Prizant, B. (2002). *Communication and Symbolic Behavior Scales: Developmental Profile – First normed edition*. Baltimore, MD: Paul H. Brookes.

Wetherby, A. M., Prizant, B. M., & Schuler, A. L. (2000). Understanding the nature of communication and language impairments. In S. F. Warren, M. E. Frey (Series Eds.), A. M. Wetherby, & B. M. Prizant (Vol. Eds.), *Communication and language intervention series: Volume 9. Autism spectrum disorders: A transactional perspective* (pp. 109-141). Baltimore, MD: Paul H. Brookes.

Wetherby, A. M., Reichle, J., & Pierce, P. L. (1998). The transition to symbolic communication. In A. M. Wetherby, S. F. Warren, & J. Reichle (Eds.), *Transitions in prelinguistic communication. Communication and language intervention series, Volume 7* (pp. 197-230). Baltimore, MD: Brookes.

Wetherby, A., Schuler, A., & Prizant, B. (1997). Enhancing language and communication development: Theoretical foundations. In D.J. Cohen & F.R. Volkmar (Eds.), *Handbook of autism and pervasive developmental disorders* (2nd ed., pp. 513-538). New York, NY: John Wiley & Sons.

Wetherby, A., Watt, N., Morgan, L., & Shumway, S. (2007). Social communication profiles of children with autism spectrum disorders late in the second year of life. *Journal of Autism and Developmental Disorders, 37,* 960-975.

Wiggins, L. D., Bakeman, R., Adamson, L. B., & Robins, D. L. (2007). The utility of the Social Communication Questionnaire in screening for autism in children referred for early intervention. *Focus on Autism and Other Developmental Disabilities, 22,* 33-38.

Wilkinson, K. M., & Hennig, S. (2007). The state of research and practice in augmentative and alternative communication for children with developmental/intellectual disabilities. *Mental Retardation and Development Disabilities, 13,* 58-69.

Wilkinson, K. M. & McIlvane, W. J. (2001). Methods for studying symbolic behavior and category formation: Contributions of stimulus equivalence research. *Developmental Review, 21,* 355-374.

Wing, L. (2005). Problems of categorical classification systems. In F.R. Volkmar et al. (Eds.), *Handbook of autism and pervasive developmental disorders: Development, neurobiology, and behavior* (3rd ed., pp. 583-605). New Jersey: Wiley.

Wing, L. (2006). *Diagnostic Interview for Social and Communication Disorders* (11th ed.). Bromley, UK: Centre for Social and Communication Disorders.

Wing, L. & Gould, J. (1979). Severe impairments of social interaction and associated abnormalities in children: Epidemiology and classification. *Journal of Autism and Developmental Disorders, 9,* 11-30.

Wing, L., Leekam, S. R., Libby, S. J., Gould, J., & Larcombe, M. (2002). The Diagnostic Interview for Social and Communication Disorders: Background, inter-rater reliability and clinical use. *Journal of Child Psychology and Psychiatry, 43,* 307-325.

Witwer, A. N., & Lecavalier, L. (2007). Autism screening tools: An evaluation of the Social Communication Questionnaire and the Developmental Behaviour Checklist-Autism Screening Algorithm. *Journal of Intellectual & Developmental Disability Research, 32,* 179-187.

World Health Organization [WHO] (1992). *The ICD 10 classification of mental and behavioral disorders: Clinical descriptions and diagnostic guidelines.* Geneva, Switzerland: Author.

Yoder, P., & Symons, F. (2010). *Observational measurement of behavior.* New York, NY: Springer Publishing.

Yoder, P. J., & Warren, S. F. (2001). Intentional communication elicits language-facilitating maternal responses in dyads with children who have developmental disabilities. *American Journal on Mental Retardation, 106,* 327-335.

Zink, I., & Lejaegere, M. (2003). *N-CDI's: korte vormen.* [D-CDI's: short forms]. Antwerpen: ACCO.

Zwaigenbaum, L., Bryson, S., Rogers, T., Roberts, W., Brian, J., & Szatmari, P. (2005). Behavioral manifestations of autism in the first year of life. International Journal of Developmental Neuroscience, 23, 143-152.

Zwaigenbaum, L., Bryson, S., Lord, C., Rogers, S., Carter, A., Carver, L., … & Yirmiya, N. (2009). Clinical assessment and management of toddlers with suspected autism spectrum disorder: Insights from studies of high-risk infants. *Pediatrics, 123,* 1383-1391.

Nederlandse samenvatting

Nederlandse samenvatting

Introductie

Autismespectrumstoornissen (ASS) vormen een breed spectrum van ontwikkelingsstoornissen die worden gekenmerkt door beperkingen in drie gedragsdomeinen: sociale interactie, communicatie en het repertoire aan interesses en activiteiten. ASS gaat relatief gezien vaak samen met een verstandelijke beperking (VB) (Bhaumik et al., 2010). Het stellen van een accurate ASS-diagnose bij mensen met een VB is gecompliceerd, met name wanneer het om een ernstige of diepe VB gaat (Vig & Jedrysek, 1999). De belangrijkste reden die hieraan ten grondslag ligt, is de overlap in gedragskenmerken tussen personen met een ASS en personen met een VB. Achterstanden in de sociale en communicatieve ontwikkeling kunnen, evenals de aanwezigheid van specifieke gedragingen, namelijk ook verklaard worden door een laag niveau van functioneren (Bhaumik et al., 2010). Er zijn echter ook duidelijke verschillen tussen personen met een VB met en zonder een ASS; dat betreft met name de ernst van de stereotiepe en repetitieve gedragingen en de relatieve beperkingen op sociaal en communicatief vlak ten opzichte van het algemene niveau van functioneren (Klin et al., 2005; Matson et al., 2008). Een correcte diagnose is ook bij deze doelgroep van groot belang om de meest geschikte interventiestrategie te kunnen bepalen. De combinatie van ASS en VB zorgt voor wederzijdse versterking van de problemen behorend bij beide stoornissen en heeft grote consequenties voor het welzijn van de persoon zelf en diens omgeving (Boucher et al., 2008).

De communicatieproblemen van kinderen met een ASS en een bijkomende VB zijn veelal zeer ernstig en leiden vaak tot stress en handelingsverlegenheid bij zowel ouders als professionals (Lecavalier et al., 2006). Bovendien blijkt uit eerder onderzoek dat er een duidelijke relatie is tussen communicatieproblemen enerzijds en de aanwezigheid van probleemgedrag anderzijds (Bott et al., 1997; Chung et al., 1995; Van Berckelaer-Onnes et al., 2002). Ondanks dat is er slechts weinig onderzoek gedaan naar de communicatiemogelijkheden en -beperkingen van kinderen met een ASS en een VB, in het bijzonder wat betreft kinderen die niet of nauwelijks spreken (Tager-Flusberg et al., 2005). Het meeste onderzoek op het gebied van communicatie en taal bij kinderen met een ASS richt zich op peuters en/of kleuters met ASS of oudere kinderen met ASS zonder VB.

Om deze reden richt dit onderzoek zich specifiek op kinderen met een ASS met een bijkomende VB. Vanwege de classificatieproblemen bij laagfunctionerende kinderen concentreert de huidige studie zich op het kernsyndroom, de autistische stoornis (AS), in plaats van het volledige spectrum. Veel onderzoek op het gebied van communicatieproblemen bij ASS beperkt zich alleen tot het niveau van expressieve taal en de bijzonderheden in het taalgebruik (Lord & McGee, 2001; Twachtman, 1995). Het

huidige onderzoek bestudeert een bredere range van communicatievaardigheden opgedeeld in drie belangrijke aspecten van communicatie, namelijk functie, vorm en inhoud. Het in kaart brengen van sterke en zwakke kanten in het communicatieve profiel van deze doelgroep vergroot de kennis van de aard van de problemen bij laagfunctionerende kinderen met een ASS. Bovendien kunnen op basis hiervan aanbevelingen worden geformuleerd voor de diagnostiek van communicatieproblemen en levert dit onderzoek bouwstenen voor het vormgeven van meer effectieve interventies. Daarnaast worden suggesties gegeven over welke kinderen baat kunnen hebben bij specifieke interventiestrategieën.

Het hoofddoel van deze studie betreft het in kaart brengen van communicatie bij kinderen met AS en een bijkomende VB teneinde de diagnostiek en interventies op het gebied van communicatie voor deze kwetsbare doelgroep te verbeteren. In dit onderzoek wordt inzichtelijk gemaakt in hoeverre deze kinderen in communicatief opzicht overeenkomen en verschillen van kinderen met een VB zonder ASS en zich normaal ontwikkelende kinderen (Hoofdstukken 3 t/m 6). Voorafgaand hieraan worden eerst de psychometrische eigenschappen van diagnostische instrumenten die gebruikt kunnen worden om ASS vast te stellen bij kinderen met een VB geëvalueerd (Hoofdstuk 2).

Belangrijkste bevindingen

Gebruik van diagnostische instrumenten bij kinderen met een verstandelijke beperking

De Nederlandse vertaling van het *Diagnostic Interview for Social and Communication Disorders* versie 11 (DISCO; Wing, 2006; Nederlandse vertaling: Van Berckelaer-Onnes et al., 2008) is een diagnostisch interview dat professionals kunnen gebruiken om ASS te diagnosticeren. De betrouwbaarheid en validiteit van dit instrument zijn reeds eerder onderzocht (Leekam et al., 2002; Nygren et al., 2009; Wing et al., 2002), maar niet specifiek voor de doelgroep met een VB. Het huidige onderzoek richt zich op de vraag in hoeverre de DISCO in staat is om een onderscheid te maken tussen kinderen met en zonder ASS met verschillende niveaus van VB in vergelijking met andere instrumenten. De instrumenten die gebruikt zijn ter vergelijking zijn het Autisme Diagnostisch Observatie Schema (ADOS; Lord et al., 1999; Gotham et al., 2007; Nederlandse vertaling: De Bildt et al., 2008) en de Vragenlijst Sociale Communicatie (SCQ; Rutter et al., 2003; Nederlandse vertaling: Warreyn et al., 2004). Voor dit onderdeel van het onderzoek hebben we ons gericht op het gehele autismespectrum, aangezien niet alle instrumenten onderscheid maken tussen AS en ASS diagnoses.

In Hoofdstuk 2 zijn de sensitiviteit en specificiteit van de drie verschillende instrumenten met elkaar vergeleken. Met betrekking tot de bruikbaarheid van de DISCO als diagnostisch instrument kunnen er op basis van onderhavig onderzoek twee

conclusies worden getrokken. Ten eerste blijkt dat de DISCO een zeer sensitief instrument is, wat impliceert dat een groot gedeelte van de kinderen met en zonder een VB met een ASS correct geclassificeerd werd. De specificiteit van de DISCO is voldoende; het instrument is echter wel overinclusief in de groep kinderen met een matige tot ernstige VB. In deze laagfunctionerende groep werden kinderen zonder ASS geregeld ten onrechte geclassificeerd met ASS. De SCQ bleek een specifiek screeningsinstrument te zijn met een grote kans op vals-negatieven. De beste verhouding tussen sensitiviteit en specificiteit werd gevonden voor de ADOS. Ten tweede loopt de overeenstemming tussen de DISCO en de beide andere instrumenten uiteen van matig tot ruim voldoende. Op basis van deze resultaten is beslist dat de ADOS en DISCO voor de hieropvolgende studies worden gebruikt als inclusiecriteria. De SCQ is verder buiten beschouwing gelaten.

Communicatie bij kinderen met de autistische stoornis en een verstandelijke beperking

Communicatieve functies. Een communicatieve functie verwijst naar de intentie of het doel waarvoor de communicatie wordt ingezet. Dit onderzoek (Hoofdstuk 3) richt zich op drie basale functies van communicatie die al op zeer jonge leeftijd kunnen worden waargenomen (Travis & Sigman, 2001; Watt et al., 2006). Dit betreffen communicatieve gedragingen voor (1) gedragsregulatie: met als doel het gedrag van anderen te beïnvloeden om een iets te verkrijgen of te protesteren, (2) sociale interactie: met als doel de aandacht van een ander op zichzelf te richten om sociale redenen, en (3) joint attention ofwel gedeelde aandacht: met als doel de aandacht van de ander op een object of gebeurtenis te richten om interesse of plezier te delen (Wetherby & Prizant, 2002).

Over het geheel genomen communiceren laagfunctionerende kinderen met AS significant minder vaak dan zich normaal ontwikkelende kinderen met een vergelijkbaar ontwikkelingsniveau. Om deze reden zijn de absolute frequenties omgezet in relatieve frequenties waardoor een profielvergelijking tussen de groepen mogelijk wordt, onafhankelijk van de mate waarin een kind communiceert. Aangezien de frequentie van intentionele communicatie en het gebruik van de diverse functies sterk samenhangen met de expressieve taalvaardigheid van de kinderen met AS is deze groep voor de hierop volgende analyses onderverdeeld in een non-verbale en een verbale groep. De non-verbale laagfunctionerende kinderen met AS zijn het meest beperkt in hun intentionele communicatie en het gebruik van de verschillende communicatieve functies. Sommige kinderen in deze groep communiceerden slechts een aantal keer gedurende de hele afname. Deze kinderen bleken niet of nauwelijks in staat het gedrag van anderen doelbewust te beïnvloeden. Andere kinderen in de non-verbale groep waren wel in staat tot intentionele communicatie, maar het grootste deel van hun communicatie bestond uit gedragsregulatie. Zij communiceerden niet of nauwelijks voor sociale interactie en joint attention. De verschillen tussen verbale kinderen met AS

en zich normaal ontwikkelende kinderen waren minder opvallend. De relatieve frequentie van functiegebruik is vergelijkbaar tussen beide groepen, hoewel de absolute frequentie van communicatie voor joint attention wel significant lager ligt in de verbale groep met AS.

Vormen van communicatie. Verschillende vormen of middelen kunnen gebruikt worden om te communiceren, waarvan taal de meest conventionele vorm betreft. Het kwantitatieve gebruik van verschillende vormen van expressieve communicatie is reeds eerder onderzocht bij kinderen met AS. Hieruit bleek dat kinderen met AS een beperkte range aan non-verbale communicatievormen gebruiken (Stone et al., 1997). Het onderhavige onderzoek (Hoofdstuk 3) is gericht op de complexiteit van de communicatieve vormen. Hierbij werd onderzocht in hoeverre communicatieve gebaren, geluiden / woorden en blikrichting gecombineerd werden in één communicatieve handeling. De resultaten van ons onderzoek wezen uit dat voornamelijk non-verbale kinderen met AS gebruik maakten van zeer primitieve vormen van communicatie; ze hun communicatieve gebaren of geluiden significant minder vaak combineerden met oogcontact dan verbale kinderen met AS en zich normaal ontwikkelende kinderen. Bij verbale kinderen met AS waren alleen op het hoogste niveau van complexiteit (een combinatie van drie verschillende vormen) verschillen zichtbaar waren met de controlegroep. Verbale kinderen met AS maakt voor joint attention significant minder vaak gebruik van een combinatie van drie verschillende vormen, namelijk gebaren, woorden en oogcontact, in vergelijking met zich normaal ontwikkelende kinderen.

Daarnaast is het receptieve en expressieve taalniveau van kinderen met AS en een VB nader onderzocht (Hoofdstuk 4). Zowel expressieve als receptieve taalvaardigheden bleken bij kinderen met AS en VB significant lager te liggen dan hun non-verbale cognitieve ontwikkelingsniveau, dit in tegenstelling tot beide controlegroepen. Verder hadden de kinderen met AS en VB aanzienlijk betere vaardigheden op het gebied van taalproductie dan taalbegrip. Dit profiel is echter tegenovergesteld aan het profiel van kinderen met een VB en zich normaal ontwikkelende kinderen. Deze resultaten impliceren dat de receptieve taalvaardigheden van kinderen met AS en VB die over (enige) gesproken taal beschikken gemakkelijk overschat kunnen worden op basis van hun betere expressieve taalvaardigheden.

Joint attention en symboolbegrip worden gezien als belangrijke voorlopers van taalontwikkeling in jonge kinderen (Travis & Sigman, 2001). Deze beide aspecten zijn onderzocht in relatie tot de actuele receptieve en expressieve taalvaardigheden van kinderen met AS en VB (zie Hoofdstuk 4). In de controlegroepen van kinderen met VB en zich normaal ontwikkelende kinderen was er een sterke samenhang tussen het non-verbale cognitieve ontwikkelingsniveau en de beide voorlopers. Verder bleek het non-verbale ontwikkelingsniveau eveneens de meest robuuste voorspeller voor zowel

taalbegrip als taalproductie in beide groepen te zijn. In de groep laagfunctionerende kinderen met AS leverden zowel symboolbegrip als joint attention een significante en unieke bijdrage aan de verklaring van huidige expressieve en receptieve taalvaardigheden. In tegenstelling tot de homogene normale ontwikkeling, laten kinderen in de autismegroep een meer disharmonisch ontwikkelingsprofiel zien. Dit kan erop duiden dat de taalontwikkeling bij hen mogelijk minder afhankelijk is van het non-verbale cognitieve ontwikkelingsniveau dan in de vergelijkingsgroepen. Over de aard van de relatie tussen joint attention en symboolvorming en de relatieve bijdrage van beide voorlopers aan de taalontwikkeling kunnen echter geen uitspraken worden gedaan, aangezien dit geen longitudinaal onderzoek betreft.

Een deel van de kinderen met ASS en een VB ontwikkelt geen gesproken taal en is daardoor aangewezen op andere communicatiestrategieën, zoals ondersteunende communicatie. Zowel driedimensionele (voorwerpen) als tweedimensionele vormen (afbeeldingen) van ondersteunende communicatie kunnen worden onderzocht met behulp van de ComVoor, die gebruikt is voor de studies in de Hoofdstukken 5 en 6. In Hoofdstuk 6 werd de perceptie van visuele en tactiele informatie onderzocht. Problemen in de symbolische communicatie bij mensen met ASS en dan vooral met betrekking tot de inhoud van of de betekenisverlening aan communicatie kunnen mogelijk verklaard worden door een zwakke centrale coherentie. Centrale coherentie verwijst naar een natuurlijke geneigdheid om binnenkomende stimuli in de context te verwerken; verschillende stukjes informatie worden samengevoegd om er een zinvolle betekenis aan te verlenen (Happé & Frith, 2006). Het vermogen tot centrale coherentie is veelal zwak bij kinderen met ASS, wat leidt tot een detailgerichte verwerkingsstijl, vaak samengaand met een onvermogen om informatie globaal en in een context te verwerken (Happé & Booth, 2008). Uit een eerdere studie van Noens en Van Berckelaer-Onnes (2008) bleek dat personen met ASS en VB een sterkere lokale verwerking hebben dan personen met VB zonder ASS. In onze studie onderzochten we het effect van doofheid op visuele perceptie bij laagfunctionerende personen met ASS om de specificiteit van deze verwerkingsstijl te onderzoeken. Zowel horende als dove mensen met ASS en een VB bleken relatief sterke lokale verwerkingsvaardigheden bij voorwerpen en afbeeldingen te hebben in vergelijking met mensen met een VB zonder doofheid. De resultaten geven aan dat zowel dove als horende mensen met ASS profiteren van hun zwakke centrale coherentie wanneer het gaat om het matchen van betekenisloze voorwerpen en afbeeldingen. Deze relatieve sterktes in visuele perceptie kunnen worden gebruikt om de communicatie te ondersteunen, maar deze verwerkingsstijl kan ook problemen opleveren in situaties waarin globale informatie-verwerking nuttig of noodzakelijk is.

Inhoud van communicatie. Inhoud van communicatie verwijst naar de betekenis van woorden of andere communicatievormen. Noens en Van Berckelaer-Onnes (2005) omschreven communicatieproblemen bij ASS als problemen in betekenisverlening.

Waarneming en betekenisverlening kunnen op vier verschillende niveaus plaatsvinden, namelijk sensatie, presentatie, representatie en metarepresentatie (zie Hoofdstuk 5 voor een meer gedetailleerde uitleg). De niveaus van sensatie en presentatie verwijzen naar non-symbolische niveaus van communicatie. Op deze non-symbolische niveaus begrijpen kinderen nog niet dat een woord of een afbeelding kan verwijzen naar iets anders. Op het niveau van (meta)representatie wordt de symbolische of verwijzende betekenis van communicatievormen begrepen (Noens & Van Berckelaer-Onnes, 2005).

Wat betreft de inhoud van de communicatie, was het onderzoek met name gericht op de vraag of de inhoud al dan niet op een symbolische wijze wordt begrepen en wat de consequenties zijn van deze manieren van betekenisverlening. Uit Hoofdstuk 5 blijkt dat kinderen met AS en VB relatief meer problemen in betekenisverlening hebben dan zich normaal ontwikkelende kinderen, maar ook in vergelijking met kinderen met een VB zonder ASS. Kinderen met AS en VB behalen opvallend minder vaak het representatieniveau. Een groot deel van deze groep verleent betekenis op het sensatie- of presentatieniveau, wat inhoudt dat de symbolische of verwijzende betekenis van communicatie niet (volledig) begrepen wordt. Uit Hoofdstuk 5 blijkt tevens dat een laag niveau van betekenisverlening een belangrijke onderliggende factor is voor achterstanden in adaptief functioneren en ernst van de ASS-symptomatologie. In het bijzonder kinderen met AS en VB die nog niet in staat zijn tot symbolische betekenisverlening (sensatie- en presentatieniveau) laten opvallende achterstanden zien op het gebied van socialisatie en communicatie in vergelijking met de controlegroepen met VB en zich normaal ontwikkelende kinderen met een vergelijkbaar niveau van betekenisverlening. Bovendien laat deze groep significant ernstigere problemen zien in termen van kwaliteit van communicatie, sociale interactie en verbeelding.

Tekortkomingen en aanbevelingen voor vervolgonderzoek

De resultaten van dit onderzoek op het gebied van communicatie bij laagfunctionerende kinderen met AS vertonen veel overeenkomsten met eerdere onderzoeken bij peuters met ASS. De problemen in de onderhavige onderzoeksgroep lijken ernstiger en meer persistent te zijn. Helaas was het in dit onderzoek niet mogelijk om een directe vergelijking met maken met een groep jonge kinderen met ASS zonder VB. De toevoeging van een dergelijke extra vergelijkingsgroep (voor alle hoofdstukken) en een groep kinderen met VB zonder ASS (voor Hoofdstuk 3) is een belangrijke aanbeveling voor vervolgonderzoek. Hoewel de diagnose ASS op steeds jongere leeftijd wordt gesteld (Charman & Baird, 2002; Zwaigenbaum et al., 2009), zal het toch lastig blijven om deze vergelijkingsgroep samen te stellen. Aangezien veel kinderen met ASS al op jonge leeftijd een ontwikkelingsachterstand hebben, is het in de peuterleeftijd nog lastig om te differentiëren tussen kinderen met en zonder bijkomende VB. Een andere

beperking op het gebied van de onderzoeksgroepen betreft de scheve sekse-verhoudingen tussen de groepen in dit onderzoek. Omdat ASS vaker voorkomt bij jongens dan meisjes, bestaat de meerderheid van de AS+VB groep uit jongens, terwijl de verhouding jongen-meisje in de andere groepen meer evenwichtig is. Het groepsgewijs matchen op non-verbale ontwikkelingsleeftijd heeft in ons onderzoek voorrang gekregen op het matchen op sekse. Idealiter zouden de groepen op beide achtergrondvariabelen gematcht dienen te worden. Er is echter wel bij alle studies gecontroleerd op de invloed van sekse op de afhankelijke variabelen.

Naar aanleiding van deze studie zijn een aantal aanbevelingen voor vervolgonderzoek geformuleerd. Ten eerste is het aanbevelenswaardig om meer onderzoek te doen naar de symboolvorming bij jonge en laagfunctionerende kinderen met ASS om zo meer zicht te krijgen op het verloop van en afwijkingen in de ontwikkeling hiervan. Ten tweede blijkt uit de literatuur dat er een duidelijk verband is tussen communicatieproblemen en het voorkomen van gedragsproblemen bij laagfunctionerende personen met en zonder ASS. Aangezien de communicatie-problemen bij kinderen met AS en VB ernstig maar ook divers blijken te zijn, is het van belang om de relaties tussen specifieke communicatieproblemen en bepaalde gedragsproblemen (o.a. zelfverwondend gedrag, agressief / destructief gedrag) in kaart te brengen om met betrekking tot preventie en interventie van gedragsproblemen meer concrete adviezen te kunnen geven. Ten derde wordt er in Nederland in de klinische praktijk al vaak gebruik gemaakt van geïndividualiseerde ondersteunende communicatie (zie Klinische implicaties). Tot nu toe is echter de effectiviteit van een dergelijke interventie nog niet systematisch onderzocht. Het uitbreiden van kennis en inzichten op deze gebieden vereist dus nog de nodige aandacht in vervolgonderzoek.

Klinische implicaties

Diagnostiek

Om een ASS-diagnose te kunnen stellen bij kinderen met een VB is het altijd van belang om het cognitieve ontwikkelingsniveau vast te stellen. Deze informatie biedt de clinicus een context waarin hij / zij de symptomatologie op gedragsniveau kan interpreteren. Op deze manier kan de clinicus beslissen of de symptomen mogelijkerwijs te verklaren zijn uit een algemene ontwikkelingsachterstand of dat er een andere oorzaak aan ten grondslag ligt (Totsika, 2010). De ADOS lijkt het meest geschikte instrument voor de groep kinderen met ASS en een VB, aangezien in de algoritmes van dit instrument expliciet onderscheid wordt gemaakt tussen non-verbale en verbale kinderen. Op basis van de ruwe scores lijkt ook de DISCO in staat om een adequaat onderscheid te maken tussen kinderen met een VB met en zonder ASS, maar de huidige criteria en cut-off scores zijn nog niet voldoende aangepast aan deze laagfunctionerende groep. De huidige bevindingen geven aan dat clinici en onderzoekers zeer voorzichtig moeten zijn

met het gebruik van de SCQ bij kinderen met een VB. Bovendien is een classificatie op een screeningsvragenlijst of zelfs een diagnostisch instrument niet voldoende om een diagnose te kunnen stellen. Onderhavig onderzoek heeft alleen de bruikbaarheid van de uiteindelijke classificaties van drie verschillende instrumenten onderzocht. De diagnostische instrumenten bieden echter ook nog een grote hoeveelheid aan kwalitatieve informatie over een kind. De bruikbaarheid van deze informatie is echter niet geëvalueerd. Voor een diagnostisch onderzoek geven de ADOS en de DISCO samen een breed beeld van het gedrag van een kind. De ADOS biedt de mogelijkheid om het gedrag van een kind in zowel gestructureerde als ongestructureerde situaties te observeren (Lord et al., 2000). Een uitgebreid interview, zoals de DISCO, geeft de benodigde informatie over de ontwikkelingsgeschiedenis en het huidige gedrag en niveau van functioneren op verschillende domeinen in de thuissituatie of ander omgevingen (Wing et al., 2002). Onderzoek heeft uitgewezen dat een combinatie van beide methoden (observatie en interview) een meer betrouwbare en stabiele diagnose verschaft (Risi et al., 2006). De verkregen informatie uit de ADOS en DISCO kan tevens gebruikt worden om een passend interventieplan samen te stellen (Wing, 2006).

Ieder kind met ASS heeft problemen op het gebied van de communicatie, maar het type en de intensiteit van de behoefte aan stimulering en ondersteuning op dit gebied is afhankelijk van verschillende factoren, zoals de leeftijd en het niveau van functioneren (Paul, 2007). Uit de onderhavige studie blijkt dat zelfs binnen de groep kinderen met AS en een VB de heterogeniteit in communicatieproblemen groot is. Om deze reden is het van belang om communicatie altijd te betrekken in het diagnostisch of handelingsgericht onderzoek van laagfunctionerende kinderen met AS. Wanneer een kind niet spreekt, is het met name belangrijk om na te gaan in hoeverre hij / zij op andere manieren communiceert door middel van gebaren, blikrichting en geluiden. Daarnaast is het relevant om het begrip van de communicatie van anderen in kaart te brengen. Verder dient het onderzoek te worden aangevuld met het observeren van intentionaliteit en het gebruik van verschillende functies van communicatie. Hiervoor kan gebruik gemaakt worden van een observatie-instrument, zoals de *Communication and Symbolic Behavior Scales – Developmental Profile* (Wetherby & Prizant, 2002), of vergelijkbare situaties die communicatie uitlokken. De gestandaardiseerde scoringsprocedure is voor de doelgroep van dit onderzoek echter niet voldoende gedifferentieerd om een goed beeld te krijgen van de communicatieve functies. Wanneer een aangepaste, meer kwalitatieve analyse van deze observatie plaatsvindt, is het instrument wel goed bruikbaar (zie Hoofdstuk 3). Veel laagfunctionerende, niet-sprekende kinderen met AS ontwikkelen idiosyncratische, onconventionele en niet gewenste gedragingen om te communiceren, zoals zelfverwondend gedrag en agressie (Wetherby et al., 2000; Paul, 2007). In het geval van probleemgedrag, dient ook de functie van het probleemgedrag onder de loep te worden genomen tijdens het diagnostisch proces. Verder is het van belang om het niveau van betekenisverlening in kaart te brengen, niet alleen omdat dit veel lager kan liggen dan op basis van het

algemene ontwikkelingsniveau verwacht kan worden, maar ook omdat dit een belangrijke onderliggende factor voor het communicatieve en sociale functioneren blijkt te zijn (zie Hoofdstuk 5). Wanneer een kind wel spreekt, kunnen gestandaardiseerde tests voor taalproductie en taalbegrip gebruikt worden. Het zal echter vaak relevant zijn om deze tests aan te vullen met een observatie van communicatief gedrag in dagelijkse situaties om te bepalen in hoeverre er een discrepantie bestaat tussen het formele niveau van taalvaardigheid en het adequaat uitwisselen van informatie en de mate van wederkerigheid in dagelijkse communicatiesituaties (Paul, 2007).

Interventie

Het bevorderen van de communicatieve competenties is een belangrijk doel in interventies bij kinderen met ASS. Veelal richt de communicatieve interventie bij jonge en laagfunctionerende kinderen met ASS zich op het stimuleren van gesproken taal (Twachtman, 1995). Deze kinderen hebben echter vaak niet alleen een ernstige achterstand op het gebied van expressieve taal, maar er zijn in veel gevallen ook problemen in de receptieve taal en de ontwikkeling van intentionaliteit en symboolvorming (zie Hoofdstukken 3, 4 en 5). Hierdoor is een bredere benadering van communicatie-interventie vereist, aangepast aan het ontwikkelingsniveau.

Veel laagfunctionerende kinderen met ASS ontwikkelen geen gesproken taal of alleen een beperkt aantal communicatieve woorden of gebaren. Deze kinderen zijn aangewezen op andere communicatievormen, zoals ondersteunende communicatie. De resultaten van ons onderzoek leiden tot twee belangrijke aanbevelingen met betrekking tot ondersteunende communicatie. Ten eerste vereisen gebruikelijke strategieën voor ondersteunende communicatie vaak representatievaardigheden, omdat visualisaties worden ingezet als symbolen of verwijzers. De resultaten met betrekking tot het niveau van betekenisverlening (Hoofdstuk 5) laten echter zien dat niet alle kinderen met AS en een bijkomende VB in staat zijn om de symbolische betekenis van communicatie te begrijpen. Wanneer de ComVoor (Verpoorten et al., 2004, 2008) wordt ingezet om het niveau van betekenisverlening te bepalen, kan de ondersteunende communicatie worden afgestemd op het individuele niveau. Als een kind (nog) niet op representatieniveau functioneert, kunnen niet-symbolische strategieën worden ingezet (zie Maljaars & Noens, 2008). Ten tweede wordt ondersteunende communicatie meestal in eerste instantie ingezet om de expressieve communicatie te ondersteunen. De resultaten van Hoofdstuk 3 geven aan dat het in veel gevallen belangrijker zal zijn om eerst de receptieve communicatie te verbeteren om te voorkomen dat het verschil tussen receptieve en expressieve vaardigheden nog groter wordt. Ondersteunende communicatie kan bij deze doelgroep gebruikt worden om activiteiten aan te kondigen of te helpen bij het maken van overgangen tussen activiteiten. Daarnaast kan het gebruikt worden om verschillende stappen binnen activiteiten uit te leggen en een volgorde in activiteiten duidelijk te maken. Wanneer

ondersteunende communicatie succesvol is geïmplementeerd voor de receptieve communicatie, kan er gestart worden met het gebruik van vergelijkbare ondersteunende communicatievormen om de expressieve communicatie te bevorderen. Het welslagen van een dergelijke expressieve interventiestrategie is gerelateerd aan de mogelijkheden van een kind tot intentionele communicatie (Rowland & Schweigert, 2000).

Wat betreft de expressieve communicatie is met name bij laagfunctionerende non-verbale kinderen met AS het gebruik van basale communicatieve functies zeer beperkt (zie Hoofdstuk 3). In overeenstemming met de resultaten van dit onderzoek lijkt het insteken op het verbeteren van intentionaliteit minstens even belangrijk als nadruk op het gebruik van bepaalde conventionele communicatievormen. Het is namelijk zinloos om een bepaald woord te oefenen en te kunnen uiten, wanneer een kind niet weet hoe en wanneer het dit woord kan gebruiken (Schuler et al., 1997; Wetherby et al., 1997). Doelgericht communicatief gedrag stelt een kind in staat om invloed uit te kunnen oefenen op zijn omgeving en heeft bovendien een positieve invloed op andere ontwikkelingsaspecten (Coupe O'Kane & Goldbart, 1998). Om intentionaliteit bij laagfunctionerende non-verbale kinderen met AS te ontwikkelen is het belangrijk om de individuele sterke kanten, motivaties, voorkeuren en favoriete activiteiten als uitgangspunt te nemen (Rogers & Dawson, 2010; Schuler et al., 1997). Het is bijvoorbeeld mogelijk om intentionaliteit te stimuleren door bekende routines te onderbreken, een favoriet speelgoedstuk buiten bereik te plaatsen of een keuze tussen speelgoed of snacks aan te moedigen. Bij verbale kinderen met AS en VB verschuift de focus van de communicatieve interventie naar het effectief gebruik van conventionele communicatievormen en vaardigheden in meer complexe situaties. Aangezien uit het onderzoek blijkt dat verbale kinderen vaker het initiatief nemen tot communiceren dan non-verbale kinderen met AS, maar tevens meer communicatieve gedragingen laten zien waarvan de functie onduidelijk is, lijkt het erop dat deze kinderen wel pogingen doen om te communiceren, maar er niet altijd in slagen om hun intenties op een effectieve manier duidelijk te maken. In het bijzonder deze groep lijkt klaar voor interventies gericht op stimulering en uitbreiding van communicatieve functies en het gebruik van meer conventionele manieren om te communiceren.

Dankwoord

Dankwoord

Tot slot: dit proefschrift had niet tot stand kunnen komen zonder de bijdrage van vele anderen. Allereerst wil ik graag alle kinderen en ouders bedanken die bereid waren aan dit onderzoek mee te werken. Het was voor de kinderen uit onze doelgroep niet altijd vanzelfsprekend om naar een vreemde omgeving met vreemde personen te komen. Ik waardeer dan ook de grote inzet van al deze kinderen en hun ouders enorm. Ook de vele begeleiders, leerkrachten, orthopedagogen en psychologen op de verschillende kinderdagverblijven, peuterspeelzalen, medisch kinderdagverblijven, kinderdagcentra en cluster-3 scholen wil ik bedanken voor hun medewerking aan alle afnames. Daarnaast wil ik drie studentassistenten, Judith van Eekelen, Joren Roos en Rianne Jansen, hier noemen, vanwege hun flexibele inzet bij de afnames en het nauwkeurig invoeren en controleren van de data. Zonder de hulp van alle 47 masterscriptie-studenten was het niet mogelijk geweest om de intensieve dataverzameling te volbrengen, dank voor jullie inzet en enthousiasme. Mijn collega's bij de Universiteit Leiden, in het bijzonder mijn kamergenoten, de mede-AIO's en de onderzoeksgroep Ontwikkelingsstoornissen hebben mij de nodige ondersteuning en gezelligheid geboden. Dank ook aan mijn nieuwe collega's in Leuven voor de support tijdens de laatste loodjes van mijn doctoraat. Ineke Smit wil ik graag bedanken voor het nalezen en corrigeren van verscheidene introducties en discussies met betrekking tot het Engels. Verder waren ook nog heel wat andere diensten onmisbaar voor het slagen van dit onderzoek: het secretariaat van Orthopedagogiek, de Audio-Visuele Dienst, de FSW Servicedesk en de FSW receptie. Allemaal hartelijk dank. En als laatste, lieve vrienden en familie, zonder jullie belangstelling, steun en afleiding zou het traject er heel anders hebben uitgezien!

Curriculum vitae

Curriculum vitae

Jarymke Maljaars was born on March 12th 1982 in Middelburg, the Netherlands. In 2000, she completed her secondary education (VWO) at the Calvin College in Goes. She obtained her propedeuse in Education and Child Studies at the Leiden University in 2001 and continued her studies at the Department of Clinical Child and Adolescent Studies. During her studies, she was a student-assistant at the department of Clinical Child and Adolescent Studies and participated in a research project on assessment and treatment of young children with severe communication problems at the Koninklijke Effatha Guyot Groep in Zoetermeer. After graduation in 2005, she joined the staff of the Department of Clinical Child and Adolescent Studies at the Leiden University as a research and education assistant. In 2006, she started her PhD project on communication in children with autistic disorder and co-occurring intellectual disability. Besides her PhD, she was half-time appointed as a lecturer at the Department of Clinical Child and Adolescent Studies. She was also engaged in clinical work, in particular assessment and treatment of children with developmental disorders at the Ambulatorium of the Leiden University. In September 2011, she started working at the Parenting and Special Education Research Unit at the Katholieke Universiteit Leuven, Belgium, and became member of the Leuven Autism Research (LAuRes) consortium. The aim of her current research project is to evaluate the effectiveness of individualized augmentative communication for adults with severe intellectual disability, autism spectrum disorder, and challenging behavior.