

Reconsidering core impairments in autism:

Diversity in empathy and social behavior

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**Reconsidering core impairments in autism:
Diversity in empathy and social behavior**

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‘If you have seen one person with autism,
you have seen one person with autism.’

Chapter 1

General Introduction

In the 1940's, Leo Kanner provided detailed descriptions of eleven children with an 'extreme autistic aloneness' (Kanner, 1943). These children were all characterized by profound problems to emotionally connect with other people and, instead, appeared to be more fascinated by objects. For instance, Kanner reported that young Donald T. 'was happiest when left alone, almost never cried to go with his mother, did not seem to notice his father's home-comings, and was indifferent to visiting relatives' (Kanner, 1943, p. 218). It was only until 1980 that 'infantile autism' was included as a distinct developmental disorder in the Diagnostic and Statistical Manual of Mental Disorders (DSM-III; APA, 1980). The present DSM-IV criteria for autism still show considerable overlap with Kanner's early clinical observations: (1) impairments in social interaction, (2) delayed language development and communication impairments, and (3) repetitive and restricted interests and behaviors (APA, 2000). In this thesis and in agreement with future DSM 5 amendments, the overarching term autism spectrum disorder (ASD) will be used, representing a range of autism-like disorders which vary in symptom severity and quality (www.dsm5.org).

Impairments in the social interaction domain are considered the *core aspect* of autism spectrum disorders (APA, 2000; Hobson, 2002; Kanner, 1943). Children with ASD have difficulties developing friendships with peers, spontaneously sharing experiences with others, using non-verbal behaviors such as eye contact or hand gestures to guide social interactions, and show a lack of social or emotional reciprocity (APA, 2000; Bauminger, Shulman, & Agam, 2003; Kasari, Locke, Gulsrud, & Rotheram-Fuller, 2011; Macintosh & Dissanayake, 2006; Mundy & Neal, 2001; Sigman & Ruskin, 1999). Although these social impairments form the crux of an ASD diagnosis, there is an ongoing debate on the potential mechanisms underlying these impairments (Chevallier, Kohls, Troiani, Brodtkin, & Schultz, 2012b). A widely known and influential theory on the social impairments in ASD, the social cognition hypothesis, states that individuals with ASD lack insight into own and others' mental states. Although many empirical findings support the social cognition hypothesis of ASD, a direct link between social cognition on the one hand and social behavior on the other hand has hardly been examined in children with ASD. In this thesis, we focus on both the socio-cognitive abilities and the social behavior of children and adolescents with ASD. Moreover, because previous research on social behavior primarily focused on the early childhood period or included children with ASD and an intellectual disability, we examine the social behavior of school-aged children and adolescents with ASD and a normal intellectual level (i.e., high-functioning ASD). Finally,

because individuals with ASD form a highly heterogeneous group, we explore the origins of their individual differences in social behavior.

Defining empathy

Social behavior hinges on the fundamental ability to empathize with others (de Waal, 2008). Due to the core impairments in social behavior, it is not surprising that ASD has also been described as an ‘empathy disorder’ (Decety & Meyer, 2008; Gillberg, 1992; Krahn & Fenton, 2009). In de Waal’s review on the evolution of empathy, he defines empathy as ‘the capacity to (a) be affected by and share the emotional state of another, (b) assess the reasons for the other’s state, and (c) identify with the other, adopting his or her perspective’ (de Waal, 2008, p. 281). This definition demonstrates the complex and multifaceted nature of empathy, including both affective and cognitive components (Blair, 2005; Davis, 1983). Affective empathy commonly refers to the experience of an emotion in response to and in harmony with a perceived emotion (Decety & Meyer, 2008; Jones, Happé, Gilbert, Burnett, & Viding, 2010). Cognitive empathy, on the other hand, refers to the ability to understand others’ emotions and thoughts by adopting their perspective (de Waal, 2008). Cognitive empathy has also been used interchangeably with the term ‘Theory of Mind’ (Blair, 2005; Jones et al., 2010).

Most research on empathy in ASD has been devoted to children’s conceptual understanding of others’ emotions and mental states, but comparatively few studies have been performed to examine the *behavioral* component of empathy, that is, whether children with ASD behave empathically in response to others’ emotions. This is particularly remarkable given the emphasis in diagnostic assessments of ASD on social behavioral rather than cognitive problems (APA, 2000; Lord et al., 2000; Lord, Rutter, & Le Couteur, 1994). Most studies on empathic responsiveness have focused on young children, highlighting their responses to an experimenter’s display of distress. For instance, an experimenter pretends to hurt his/her finger by hitting it with a toy hammer, expressing both verbal (“Ouch!”) and non-verbal (facial expression) cues of distress (Sigman, Kasari, Kwon, & Yirmiya, 1992). Typically developing one-year-olds generally respond to the experimenter’s distress by showing increased attention and emotional concern (Hutman et al., 2010; Zahn-Waxler, Radke-Yarrow, Wagner, & Chapman, 1992). However, even though young children with ASD and an intellectual disability are not oblivious to nor actively withdraw from the emotions of others (Corona, Dissanayake, Arbelle, Wellington, & Sigman, 1998; Dissanayake, Sigman, & Kasari, 1996), they tend to look less at the distressed adult, demonstrate less emotional concern on their face and provide fewer

sympathetic comments when compared to age or IQ-matched peers (Bacon, Fein, Morris, Waterhouse, & Allen, 1998; Hobson, Harris, García-Pérez, & Hobson, 2009; Loveland & Tunali, 1991; Scambler, Hepburn, Rutherford, Wehner, & Rogers, 2007; Sigman et al., 1992). These experimental studies all suggest a deficit in empathic responsiveness in preschoolers with ASD and an intellectual disability. Yet, it is currently unclear whether the observed lack of empathic responsiveness can be generalized to school-aged children and adolescents with HFASD. Both IQ and age have been indicated as promoting factors of empathic responsiveness in children and adolescents with ASD, giving rise to the possibility that they overcome these early observed limitations in empathic responsiveness (Bacon et al., 1998; Dissanayake et al., 1996; McGovern & Sigman, 2005). Moreover, it is unclear whether the observed lack of empathic responsiveness results from a lack of understanding others' emotions and mental states, or possibly from a lack of motivation to interact with others. Both of these hypotheses will be discussed more thoroughly below.

Social cognition hypothesis

The social cognition hypothesis of ASD asserts that the characteristic impairments of ASD are produced by a key deficit in Theory of Mind (Baron-Cohen, Leslie, & Frith, 1985). Theory of Mind (ToM) refers to the ability to interpret own and others' behaviors in terms of mental states such as intentions, beliefs, and desires (Baron-Cohen et al., 1985). In a now classic study, Baron-Cohen and colleagues (1985) used a first-order false belief task to examine the ToM abilities of children with autism and compared them to peers with Down Syndrome and typical development. A first-order false belief task assesses children's understanding that people act according to their own (sometimes false) beliefs about the objective world, rather than the objective world itself. Typically developing 5-year-olds are generally able to solve this task, but children with ASD are significantly delayed in their performance (Baron-Cohen et al., 1985; Baron-Cohen, 1989; Wellman, Cross, & Watson, 2001). The 1985-publication on an impaired ToM in autism boosted a new research area and numerous studies have replicated the finding (Boucher, 2012; Yirmiya, Erel, Shaked, & Solomonica-Levi, 1998). Failing to attend to or comprehend others' mental states may also entail problems in understanding others' emotions. Indeed, a wealth of studies has shown extensive impairments in recognizing and understanding the emotions of others in children with ASD compared to age-matched peers (for a review see Begeer, Koot, Rieffe, Terwogt, & Stegge, 2008b). Hence, there appears to be a convincing amount of empirical evidence for a lack of social cognition in children with ASD.

While the social cognition hypothesis of ASD is intuitively appealing, it has also been criticized (e.g., Bloom & German, 2000; Rajendran & Mitchell, 2007; Yirmiya et al., 1998). First, a ToM impairment is not uniquely related to ASD; similar impairments have been reported in schizophrenia, frontotemporal dementia, and anorexia nervosa (Brüne & Brüne-Cohrs, 2006; Russell, Schmidt, Doherty, Young, & Tchanturia, 2009; Yirmiya et al., 1998). Second, successful performance on a false belief task requires several other abilities (e.g., verbal ability, memory capacity, cognitive flexibility) besides an adequate ToM (Bloom & German, 2000). Thus, as is true for most psychological constructs, a successful ToM task performance may not be a pure reflection of adequate ToM. Similarly, an impaired or delayed performance on a false belief task may be due to linguistic rather than ToM impairments. Children's ToM task performance is indeed positively correlated with their verbal abilities (Fisher, Happé, & Dunn, 2005; Milligan, Astington, & Dack, 2007; Ronald, Viding, Happé, & Plomin, 2006). This association appears to be even stronger in children with ASD (Bauminger & Kasari, 1999; Happé, 1995), suggesting that children with ASD may rely disproportionately on their verbal abilities to solve mental reasoning tasks. Third, many individuals with high-functioning ASD (HFASD) are able to pass false belief tasks (e.g., Brent, Rios, Happé, & Charman, 2004; Happé, 1994) and show adequate understanding of basic emotions (e.g., Capps, Yirmiya, & Sigman, 1992; Downs & Smith, 2004), but they still experience problems understanding other minds and socially interacting with others (e.g., Kasari et al., 2011; Macintosh & Dissanayake, 2006; Peterson, Garnett, Kelly, & Attwood, 2009; Roeyers, Buysse, Ponnet, & Pichal, 2001). Most researchers, including Baron-Cohen, currently agree that a full understanding of the complexities of the human mind cannot be reduced to passing or failing a first-order false belief task (Bloom & German, 2000; Rajendran & Mitchell, 2007). Instead, children continue to improve their understanding of other minds, grasping complex mental concepts such as sarcasm later on in their development (Miller, 2009). For this reason, more advanced ToM tasks have been developed as sensitive and age adequate measures of ToM in older individuals with HFASD. Previous studies have highlighted advanced ToM impairments in children with HFASD (Beaumont & Sofronoff, 2008; Brent et al., 2004; Sobel, Capps, & Gopnik, 2005; White, Hill, Happé, & Frith, 2009), yet findings are equivocal with regard to advanced ToM impairments in adolescents and adults with HFASD (Ponnet, Buysse, Roeyers, & De Clercq, 2008; Roeyers et al., 2001; Senju, Southgate, White, & Frith, 2009; Spek, Scholte, & Van Berckelaer-Onnes, 2010).

Although adequate social cognition may be necessary to show appropriate social behavior, it may not be sufficient (Peterson et al., 2009). Thus, impairments in social

cognition may not entirely explain the social interaction problems in ASD. In developmental psychology, children's adequate social cognition is often understood as both cause and consequence of adequate social interactions (Bosacki & Astington, 1999; Caputi, Lecce, Pagnin, & Banerjee, 2012; Carpendale & Lewis, 2004; Findlay, Girardi, & Coplan, 2006). Yet, children may also use their social cognition for less honorable goals such as teasing and bullying (Harris, 1989; Hughes & Leekam, 2004). Also, studies in the field of social psychology have frequently demonstrated that the link between human social cognition and behavior is less than perfect. For instance, realizing that an unfamiliar person on the street is injured and may require medical help, does not automatically lead to helping behavior when other witnesses are present ('bystander effect'). Both of these examples illustrate that adequate social cognition does not guarantee adequate social behavior or an empathic response. A motivational aspect may help to explain the inconsistent relation between social cognition and social behavior.

Social motivation hypothesis

It has been hypothesized that children with ASD may find social interactions less inherently rewarding, because social interactions lack emotional significance for them (Dawson et al., 2004; Hobson, Chidambi, Lee, & Meyer, 2006; Kanner, 1943). Therefore, children with ASD may not be sufficiently motivated to share their experiences with others or respond adequately to the social bids of others. Multiple researchers in the autism field have already alluded to a negative cascading effect that an early lack of social motivation may have on the development of children with ASD (Chevallier et al., 2012b; Dawson et al., 2004; Klin, Jones, Schultz, & Volkmar, 2003; Mundy & Neal, 2001).

Indications for a reduced social motivation in ASD come from empirical studies showing a lack of social orienting (Jones & Klin, 2009; Mundy & Neal, 2001; Osterling, Dawson, & Munson, 2002), a lack of enjoyment induced by social interactions (Baron-Cohen & Wheelwright, 2003; Chevallier, Grèsez, Molesworth, Berthoz, & Happé, 2012a; Hauck, Fein, Waterhouse, & Feinstein, 1995; Macintosh & Dissanayake, 2006), and a lack of desire to maintain one's social status in children and adults with ASD (Begeer et al., 2008a; Chevallier et al., 2012b; Chevallier, Molesworth, & Happé, 2012c; Hobson et al., 2006; Izuma, Matsumoto, Camerer, & Adolphs, 2011). For instance, children with ASD show a reduced tendency to praise others' achievements for social reasons (Chevallier et al., 2012c) or to adjust their self-presentation to others' preferences (Begeer et al., 2008a). These findings seem to suggest that individuals with ASD are characterized by a reduced concern for what others might think or feel about them.

Summing up, a lack of social motivation may help to explain the socially deviant behaviors and the lack of empathic responsiveness seen in ASD. However, it should be noted that any single-deficit model of ASD, including the social motivation hypothesis, is probably limited in explaining all individual cases of ASD (Happé, Ronald, & Plomin, 2006; Pellicano, 2010) due to the large genetic heterogeneity of the disorder. A single genetic cause of ASD has not been identified, instead the genetic etiology of ASD is diverse and still unknown for 75-80% of all cases (Miles, 2011). Jones and Klin (2009) describe the genetic heterogeneity of ASD as ‘one of the greatest obstacles to identification of discrete causes of autism, and it presents a formidable hurdle to developing effective treatments targeting the causes of the syndrome and not only its behavioral symptoms’ (Jones & Klin, 2009, p. 471). Our current research and understanding of ASD is still predominantly based on group comparisons (comparing a group with ASD to a matched peer group without ASD), which falsely implies that individuals with an ASD diagnosis form a uniform group. Rather than trying to understand, diagnose, and treat all individuals with ASD alike, more research efforts should be made to disentangle and understand the individual differences within the autism spectrum. A better understanding of these individual differences may ultimately lead to an improvement in diagnostic assessments and an increase in interventions that are tailored to the specific needs of a subgroup or individual.

Social subtypes

Heterogeneity in the autism spectrum is not only recognized at a genetic level, but is also found at a neurological (McPartland, Coffman, & Pelphrey, 2011), cognitive (Pellicano, 2010; Towgood et al., 2009), and behavioral level (APA; 2000; Mundy, Henderson, Inge, & Coman, 2007; Wing & Gould, 1979). In order to address the large heterogeneity within the autism spectrum, several attempts have been made to identify and create clinically relevant ASD subgroups. For instance, the current DSM-IV differentiates autistic disorder, syndrome of Asperger, and PDD-NOS (APA, 2000). However, the validity and clinical usefulness of the DSM-IV diagnostic subgroups has been a topic of considerable debate, because differences between the subgroups are not consistently found (e.g., Macintosh & Dissanayake, 2004; Volkmar, State, & Klin, 2009; Witwer & Lecavalier, 2008). Therefore, in the upcoming DSM 5, all individuals with an ASD will be subsumed under the umbrella term of an autism spectrum disorder (www.dsm5.org). In this thesis, we intend to shed more light on the heterogeneity in ASD by considering individual differences in the core social interaction impairments.

Children with autism were originally described by Kanner (1943) as socially withdrawn children with a profound lack of interest in others. However, it is now recognized that while some individuals with ASD prefer to keep to themselves, others actively seek social interactions, albeit in an unusual manner (Mundy et al., 2007). For instance, a student with HFASD described her social behavior on an online blog as: ‘I persist in chattering inanely to everyone around me - classmates, professors, cashiers, people waiting in line, people in the same elevator. It's like I have no social boundaries. I'd probably address the President the exact same way I talk to the janitor in the hallway.’ (“Reports from a Resident Alien”, 2012). Thus, even though ASD is primarily defined by profound social interaction impairments (APA, 2000), individuals with ASD also demonstrate significant individual differences in the *quality* of their social impairments. Wing and Gould already suggested in 1979 that the social behavior of children with ASD could be categorized in three different subtypes: aloof, passive, and active-but-odd. Aloof children with ASD do not seek nor adequately respond to social interactions. Although they may ask for things such as food, they do not seem to care for social attention. Passive children on the other hand engage in social interactions when instigated by others, but fail to initiate social interactions themselves. Finally, the attempts of active-but-odd children to seek social contact are often inadequate or even inappropriate. For instance, they may stand too close to a conversation partner or talk endlessly about one particular interest. The validity of the ASD social subtypes has not only been confirmed in later studies (Castelloe & Dawson, 1993; Roeyers, 1997), but has also been substantiated by findings of distinct neuronal activity patterns (Burnette et al., 2011; Dawson, Klinger, Panagiotides, Lewy, & Castelloe, 1995; Sutton et al., 2005).

Note that the active approach behavior of active-but-odd children, although poorly adjusted to the social requirements, seems to imply some sort of social motivation (Chevallier et al., 2012b). Hence, socio-motivational deficits may not be universally present in individuals with ASD. Instead, the drive to socially interact with others and establish relationships may be preserved in some individuals with ASD, but impaired in others. This marks an important step away from the idea of one central impairment in ASD that explains all individual cases of ASD (Happé et al., 2006; Pellicano, 2010). Even though most researchers and clinicians agree that the autism spectrum is a heterogeneous collection of developmental disorders with an emphasis on social interaction impairments, there have been surprisingly few studies on within-group differences in social behavior.

Research aims

Previous empirical studies generally confirm that ASD is an empathy disorder, consistently showing impairments in the understanding of and empathically responding to others' emotions. However, these cognitive and behavioral components of empathy have not been systematically examined in a large sample of school-aged children and adolescents with HFASD. Furthermore, even though individual differences in social behavior are widely recognized in children with ASD (e.g., Mundy et al., 2007; Wing & Gould, 1979), it is unclear which variables underlie these individual differences. In this thesis, two research questions will therefore be addressed, both pertaining to group and individual differences in empathy and social behavior in HFASD.

Our first aim is to create a better understanding of the empathic abilities of school-aged children and adolescents with HFASD. Children's understanding of other minds (i.e., Theory of Mind) has already been measured extensively with first-order false belief tasks. However, adolescents with HFASD are generally able to solve these false belief tasks, even though they still show impairments during everyday social interactions. Hence, more complex and sensitive Theory of Mind (ToM) measures are needed to assess ToM in older children and adolescents with HFASD. While previous studies already demonstrated advanced ToM impairments in children with HFASD (6-12 years), results remain equivocal in older individuals with HFASD. In Chapter 2 of this thesis, we therefore compare more advanced ToM abilities of children and adolescents with HFASD to those of typically developing peers.

While a majority of the studies on empathy in ASD has concentrated on specific cognitive impairments, only few studies have directly looked at children's empathic behavior in response to others' emotions. Moreover, because previous studies on children's empathic responsiveness primarily included young children with ASD or children with an intellectual disability, it is uncertain whether the observed lack of empathic responsiveness in ASD can be generalized to school-aged children and adolescents with HFASD. In Chapter 3, we therefore examine the behavioral responses of children and adolescents with HFASD and typical development to others' emotional displays, both during a structured observation and by use of parental report.

In Chapter 4, we assess a very basic, yet vital element of everyday social behavior: introducing oneself to others. The ability to present oneself favorably to others while taking into account others' preferences, in other words a successful self-presentation, requires at least some understanding of what the audience wants to hear (social cognition), but may additionally, and perhaps crucially, depend on the wish to convey a positive image

of oneself (social motivation). Hence, any impairments found in the self-presentational skills of children and adolescents with HFASD are indirect evidence of socio-cognitive and/or socio-motivational deficits. In our study we compare the self-presentational skills of children and adolescents with HFASD to those of typically developing peers in both hypothetical (self-reported response) and real life (observed response) situations.

In the following two chapters, we shift from a group-based approach (HFASD vs. typical development) to an individual-based approach by looking at individual differences in the empathic (Chapter 5) and social behavior (Chapter 6) of children and adolescents with HFASD. The second aim of this thesis is to explore the underlying mechanisms of these individual social differences. In Chapter 5, we focus on the role of children's temperament and cognitive abilities in predicting their degree of empathic responsiveness. For instance, children with HFASD may fail to show an empathic response to someone's distress, because they have a low tendency to engage in social interactions (temperament). On the other hand, children with HFASD may also fail to respond empathically, because they lack insight into other's emotional states (Theory of Mind) or because they are impaired in the basic cognitive processes guiding their behavior (executive functions). Therefore, we study the unique contribution of children's temperament, Theory of Mind, and executive functioning to their empathic responsiveness.

Finally, in Chapter 6, we examine individual differences in social interaction style (aloof, passive, active-but-odd) of children and adolescents with HFASD and associated characteristics. We use multiple informants (child, parent, teacher) to establish behavioral, cognitive and socio-emotional profiles of the children. By doing so, we aim to increase the awareness and understanding of individual differences in social behavior of children and adolescents with HFASD. Social interaction style may be a useful dimension to differentiate children with HFASD.

Participants

A pilot-study was performed to test the practical feasibility of our newly developed empathy measures and to get a first impression of the responses of children and adolescents with and without HFASD. We included 15 children and 11 adolescents with a clinical diagnosis of HFASD via specialized schools or youth care institutions for children with ASD. A typically developing comparison group was included consisting of pupils from regular Dutch primary (n=16) and secondary schools (n=10). This sample is described in more detail in Chapter 4.

All other chapters in this thesis are based on the sample from the main study. Participants in the main study consisted of 214 individuals with a clinical diagnosis of HFASD (mean age: 13,7 years; range: 6,4 - 20,5 years) and 73 typically developing peers (mean age: 12,1 years; range: 6,0 - 17,1 years). The vast majority of the participants with HFASD (n=188; 90%) was admitted to specialized education. The remaining participants with HFASD either followed regular education (n=4) or lived in a youth care institution (n=22). We managed to obtain additional diagnostic information on the majority of the participants, based on parental reports of autism severity (Social Responsiveness Scale; n=168; 79%) and standardized observations of the participants' socio-communicative behavior (Autism Diagnostic Observation Schedule; n=197; 92%). The typical development group was derived from regular primary and secondary schools in the proximity of Amsterdam. The final size of the samples in each chapter may vary according to the specific variables of interest (HFASD: n = 121-194; Typical Development: n = 50-60).

Chapter 2

Rethinking Theory of Mind in high-functioning autism spectrum disorder

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Journal of Child Psychology and Psychiatry (in press)*

Abstract

Background: The socio-communicative problems in autism spectrum disorder (ASD) are traditionally linked to impairments in Theory of Mind (ToM), the ability to ascribe mental states to others. Although ToM impairments are consistently reported in young children with ASD, findings on more advanced ToM understanding in older individuals with high-functioning ASD (HFASD) are less straightforward. Therefore, we assessed the advanced ToM abilities of a large sample of school-aged children and adolescents with HFASD (n=194; 6-20 years) and compared them to a typically developing comparison group (n=60). *Methods:* Participants' advanced ToM was assessed with five social stories containing second-order false beliefs, display rules, double bluff, faux pas, and sarcasm. *Results:* Participants with HFASD performed equally well on each of the ToM stories as their typically developing peers. Consistent age effects were noticed with adolescents outperforming the children. Furthermore, advanced ToM was positively associated with participants' age, verbal abilities and general reasoning abilities. *Conclusions:* Counter to what the Theory of Mind theory of ASD would predict, school-aged children and adolescents with HFASD seem to be able to master the theoretical principles of advanced mental state reasoning. However, they may still fail to apply these theoretical principles during everyday social interactions.

Introduction

For nearly three decades, studies on Theory of Mind have dominated research on individuals with Autism Spectrum Disorders (ASD; Baron-Cohen, Leslie, & Frith, 1985). Theory of Mind (ToM) classically refers to the ability to ascribe mental states to people and to explain and predict their behavior in terms of underlying mental states (Baron-Cohen et al., 1985). A limited ToM ability may explain the characteristic impairments in socio-communicative behavior in individuals with ASD. Indeed, young children with ASD generally fail first-order ToM tasks (false belief tasks), but research findings are less straightforward with regard to more advanced ToM understanding in older individuals with ASD of normal intelligence (i.e., 'high-functioning' ASD; HFASD). In the current study, we examined advanced ToM understanding in a large sample of school-aged children and adolescents with HFASD, and a typically developing comparison group.

Children's ToM has been examined extensively with first-order false belief tasks (Wellman, Cross, & Watson, 2001; Wimmer & Perner, 1983). These tasks require children to predict a protagonist's actions or thoughts based on a false belief. Young children with ASD, especially those with an intellectual disability, generally fail these first-order ToM tasks (Baron-Cohen et al., 1985; Yirmiya, Erel, Shaked, & Solomonica-Levi, 1998) that are

mastered by typically developing children at four or five years of age (for a meta-analysis see Wellman et al., 2001). However, once the verbal abilities of children with ASD are equivalent to those of a typical 11- or 12-year-old, they too perform at ceiling on such tasks (Fisher, Happé, & Dunn, 2005; Happé, 1995). Hence, children's success on a ToM task seems in part dependent on their verbal ability. Yet, despite their ultimate success on first-order ToM tasks, children and adolescents with ASD still experience profound difficulties understanding others' thoughts and intentions in everyday life (Bauminger & Kasari, 1999; Peterson, Garnett, Kelly, & Attwood, 2009). There is a need for more sensitive measures which capture putative deviations in ToM understanding in these older groups.

In response to this need, various advanced ToM tests have been developed, such as the Strange Stories task (Happé, 1994). Advanced ToM tests consist of an eclectic mixture of social stories that all require a form of second-order reasoning: inferences about someone's thoughts and feelings, which are, in turn, about another person's mental states (Miller, 2009). In typical development, children are able to infer second-order false beliefs ('X falsely believes that Y thinks') when they are five or six years old (for a review see Miller, 2009). More complex forms of second-order reasoning such as the understanding of ironic remarks occur later in development, although exact ages have not yet been pinpointed (Filippova & Astington, 2010; Miller, 2009). Recently, advanced ToM tasks have been refined by the addition of physical state stories or questions that allow a specific impairment in mental state reasoning to be distinguished from more general impairments in reasoning or text comprehension (Kaland, Callesen, Moller-Nielsen, Mortensen, & Smith, 2008; White, Hill, Happé, & Frith, 2009).

The relatively small number of studies on advanced ToM in school-aged children (6 to 12 years) with HFASD indicate some level of advanced ToM impairment when compared to typically developing children (Beaumont & Sofronoff, 2008; Brent, Rios, Happé, & Charman, 2004; Kaland et al., 2008; Sobel, Capps, & Gopnik, 2005; White et al., 2009). Yet, White and colleagues (2009) also underscore the extent of individual differences in ToM task performance. In their study, a substantial proportion of children with HFASD performed similarly or even better than a typically developing comparison group on advanced mental state reasoning. At later ages, findings with regard to advanced ToM understanding are more equivocal in HFASD groups. While various studies have highlighted advanced ToM impairments (Baron-Cohen, Jolliffe, Mortimore, & Robertson, 1997; Kleinman, Marciano, & Ault, 2001), others have failed to document any limitations in ToM understanding (Ponnet, Buysse, Roeyers, & De Clerq, 2008; Roeyers, Buysse,

Ponnet, & Pichal, 2001; Senju, Southgate, White, & Frith, 2009; Spek, Scholte, & Van Berckelaer-Onnes, 2010). In sum, there is currently no consensus on whether adolescents and young adults with HFASD are impaired in their advanced ToM understanding.

In the current study we examined advanced ToM in children and adolescents with HFASD, and compared their performance to a typically developing group. Because we aimed to test a large sample with a wide age range, we devised a short, yet comprehensive collection of advanced ToM stories that could be administered to individuals between 6 and 20 years of age. First, we included the birthday puppy story, which is one of the most frequently used vignettes targeting *second-order false belief* reasoning (Sullivan, Zaitchik, & Tager-Flusberg, 1994). Second, we included an *emotional display rule* understanding task that highlights one of the most frequent ways of creating false beliefs in daily life: hiding one's true emotion by modifying one's facial expression (Begeer et al., 2011). The three remaining stories appeared in the Stories from Everyday Life (Kaland et al., 2008), which are very similar to those of the Strange Stories task. These stories comprised *double bluff*, social rule violation (*faux pas*), and *sarcasm*. Together, these five vignettes comprise a global index of advanced ToM reasoning that represent different but interconnected domains of mental state knowledge, some of which may be mastered earlier on (second order false belief, display rules), and some of which may be mastered relatively late in development (double bluff, faux pas, and sarcasm).

Based on previous findings, we expected that the majority of typically developing children and adolescents would pass the second order false belief and emotional display rule tasks (Begeer et al., 2011; Miller, 2009), but would show more difficulty understanding double bluff, faux pas, and sarcasm. Compared to typically developing children, we expected that children with HFASD (6 to 12 years) would show impaired performance on all five stories. Further, we expected that adolescents with HFASD (> 12 years) would only perform more poorly than their typically developing counterparts on the latter three stories.

The current study also allowed us to examine how maturation (i.e., chronological age), verbal ability and general reasoning abilities (i.e., physical state inferences) are related to advanced ToM in HFASD across a broad age-range. While verbal ability, in particular, has been shown to correlate strongly with ToM understanding in typically developing children and children with moderate learning difficulties (Fisher et al., 2005; Milligan, Astington, & Dack, 2007; Ronald, Viding, Happé, & Plomin, 2006), children with HFASD have been shown to need disproportionately advanced linguistic maturity before they can pass standard ToM tasks (Bauminger & Kasari, 1999; Happé, 1995). It is important to

establish, therefore, whether relations between verbal ability and advanced ToM are similarly manifest in both groups.

Method

Participants

Via a specialized school for normally intelligent children with ASD (Berg en Boschool) we recruited 214 children and adolescents with ASD. School admission criteria included a normal IQ ($IQ > 70$) and a clinical diagnosis of ASD. The clinical diagnoses were established prior to the recruitment according to DSM-IV-TR-criteria by psychiatrists/psychologists who worked independently from the school and the authors, and who were unaware of the goals and outcomes of the current study. The diagnostic process included anamneses, proxy reports, and psychiatric and neuropsychological examinations. Our strictly high-functioning ASD sample allowed for the inclusion of a comparison group with a normal IQ (Jarrold & Brock, 2004). The comparison group consisted of 73 typically developing children and adolescents and was recruited via public primary and secondary schools.

We decided to exclude 20 of the 214 participants with HFASD from the final analysis due to an incomplete IQ assessment (6), a verbal receptive IQ under 70 (4), or incomplete ToM data (10). Within the typically developing comparison group, 13 of the 73 participants were excluded from the final analysis due to technical problems (1), incomplete IQ assessment (3), incomplete ToM data (5), or a high level of parent-reported autistic characteristics on the Social Responsiveness Scale (4; Constantino & Gruber, 2007). All parents of the final sample of 60 children and adolescents in the comparison group confirmed that their child had no ASD diagnosis.

The resulting 194 participants with HFASD were significantly older than the 60 typically developing (TD) peers ($M_{HFASD} = 13.8$, $SD = 3.00$; $M_{TD} = 12.1$, $SD = 2.85$; $p < .001$), but mean receptive verbal IQ ($M_{HFASD} = 104.7$, $SD = 12.24$; $M_{TD} = 105.9$, $SD = 12.22$) and gender ratio (HFASD: 165 boys; TD: 52 boys) were comparable for both groups (see also Table 2.1). Following Jarrold and Brock (2004), we examined and, if necessary, statistically controlled for the possible influence of participants' age and verbal ability.

Table 2.1 Description of participants with high-functioning ASD (HFASD) and typically developing (TD) participants, split for children (6-11 years) and adolescents (12-20 years).

	Children				Adolescents			
	HFASD (n=59)		TD (n=27)		HFASD (n=135)		TD (n=33)	
	<i>M (SD)</i>	<i>Range</i>	<i>M (SD)</i>	<i>Range</i>	<i>M (SD)</i>	<i>Range</i>	<i>M (SD)</i>	<i>Range</i>
Age (in years)	10.2 (1.40)	6.4 – 11.9	9.5 (1.79)	6.0 – 11.9	15.3 (1.97)	12.0 – 20.5	14.3 (1.29)	12.5 – 17.1
Receptive verbal IQ	103.4 (12.67)	72 – 127	113.5 (9.72)	99 – 132	105.3 (12.06)	72 – 132	99.7 (10.48)	77 – 125
Receptive verbal raw score	120.8 (13.82)	93 – 149	123.7 (15.52)	93 – 151	155.1 (16.49)	106 – 191	143.6 (13.22)	115 – 183
Gender (boys; girls) (n)	53 ; 6		27 ; 0		112 ; 23		25 ; 8	
Clinical ASD diagnosis (n) (Autism; AS; PDD-NOS)	13 ; 5 ; 39		0 ; 0 ; 0		21 ; 22 ; 91		0 ; 0 ; 0	
ADOS score (SA + RRB)	6.2 (4.70)	0 – 18	-	-	5.1 (4.18)	0 – 18	-	-
ADOS severity score	3.7 (2.78)	1 – 10	-	-	3.0 (2.47)	1 – 10	-	-

Note. ASD = Autism Spectrum Disorder; AS = Asperger’s Syndrome; PDD-NOS = Pervasive Developmental Disorder – Not Otherwise Specified.

ADOS = Autism Diagnostic Observation Schedule; SA = Social Affect score; RRB = Repetitive and Restricted Behavior score.

In addition to clinical diagnoses of autism (34), Asperger's syndrome (27), or PDD-NOS (130), we gained diagnostic information on 178 participants with HFASD (92%) with the Autism Diagnostic Observation Schedule (ADOS; Lord et al., 2000). Despite their clinical diagnoses, admission to specialized education, and parental reports of high autistic traits on the Social Responsiveness Scale ($M = 80.0$, $SD = 22.41$), average ADOS score of the participants ($M = 5.5$, $SD = 4.37$) indicated that a large proportion in fact scored below the ADOS cutoff for an ASD (< 7). Indeed, 64% (114) of the participants received an ADOS score below the ASD cutoff using the revised ADOS algorithm (Gotham et al., 2008). These results suggest that a majority of our HFASD sample may show relatively mild autistic traits. To make sure that any possible group difference on advanced ToM task performance between our HFASD sample and typically developing (TD) sample would not be distorted by the relatively mild autistic symptoms in our HFASD sample, we checked in our statistical analyses whether autism severity had an impact on advanced ToM performance.

Measures

Advanced Theory of Mind (ToM)

The five advanced ToM tasks (second-order false belief, emotional display rule understanding, double bluff, faux pas, and sarcasm) appear in Appendix I (p. 162; Begeer et al., 2011; Kaland et al., 2008; Sullivan et al., 1994). These tasks were chosen because they were expected to elicit the strongest differences in scores between children with HFASD and typically developing comparison children (Kaland, personal communication).

All story narratives were read aloud by the experimenter and followed up with a physical state question (except second-order false belief) and a mental state question. The physical state question required reasoning about a non-mental event in the story, whereas the mental state question required reasoning about the mental state of the story protagonist. With the exception of the second-order false belief task, which included intermittent control questions, participants received a typed sheet for each story, which they could choose to read simultaneously. The typed sheet was taken away before questioning. Each physical state question was scored 1 (correct) or 0 (incorrect or 'don't know'). These scores were summed to yield a 0-4 physical state total index. Similarly, each of the mental state questions was scored 1 (correct) or 0 (incorrect or 'don't know'). The creation of a 0-5 advanced ToM index is discussed in the Results. Inter-rater reliability of the mental state questions was moderate to perfect (20% of the ToM data was coded double), with kappa's ranging from 0.57 to 1.00.

Peabody Picture Vocabulary Test-III-NL

The Peabody Picture Vocabulary Test (Dunn & Dunn, 2004) assesses receptive vocabulary and is highly correlated with more general measures of verbal IQ (Hodapp & Gerken, 1999). Participants had to select one of four pictures that corresponded with a given word. The test consists of 17 sets of 14 words, which increase in difficulty. Based on the PPVT, participants received an absolute measure of receptive verbal ability.

Autism Diagnostic Observation Schedule-Generic (ADOS-G)

The ADOS (Lord et al., 2000) is a diagnostic observation measure to assess the presence and severity of autistic symptoms in the domains of social reciprocity, communication, fantasy, and repetitive interests and behaviors. During a semi-structured observation, the ADOS-interviewer offers playful activities (e.g., reading a story book) and topics of discussion (e.g., peer problems) to assess the socio-communicative abilities of the participant. Each of the participant's behaviors is rated on a scale ranging from normal behavior (0) to clearly deviant and autistic behavior (2). An ADOS score of 7 or higher is indicative of an ASD. The ADOS has excellent internal consistency, inter-rater reliability, test-retest reliability, and discriminant validity (Lord et al., 2000).

Social Responsiveness Scale (SRS)

The SRS (Constantino & Gruber, 2007) is a parent or teacher questionnaire which assesses autistic traits. The SRS consists of 65 statements about the child's behavior that can be answered on a 4-point scale ranging from 0 (never true) to 3 (almost always true). A higher total score indicates more autistic traits. Good reliability and validity have been reported (Constantino & Gruber, 2007).

Procedure

Upon receiving informed consent from parents and participants over 11 years, each participant was individually tested at school. The advanced ToM tasks were part of a full battery of tests, described elsewhere (Scheeren, Koot, & Begeer, 2012). All interviews were videotaped and transcribed, and coded by graduate students who were blind to the diagnosis of the participants.

Table 2.2 Observed and predicted probability (between brackets) of passing the mental state question of each Theory of Mind story.

	Children		Adolescents		Contrast <i>p</i> -values	
	HFASD (n=59)	TD (n=27)	HFASD (n=135)	TD (n=33)	Group	Age
1. Second-order false belief ^a	.85 (.83)	.78 (.81)	.95 (.95)	.97 (.95)	n.s.	< .01
2. Emotional display rule ^b	.92 (.91)	.89 (.90)	.96 (.97)	.97 (.96)	n.s.	n.s.
3. Double bluff ^c	.46 (.48)	.44 (.41)	.65 (.64)	.55 (.58)	n.s.	< .05
4. Faux pas ^d	.49 (.50)	.44 (.43)	.68 (.68)	.61 (.62)	n.s.	< .01
5. Sarcasm ^e	.32 (.33)	.26 (.24)	.62 (.64)	.49 (.50)	n.s.	< .001

Note. ^aThe overall model for 1 was significant, $\chi^2_{(2)} = 10.54, p < .01$; ^b The overall model for 2 was not significant, $\chi^2_{(2)} = 3.04, p > .10$; ^cThe overall model for 3 was significant, $\chi^2_{(2)} = 8.15, p < .05$; ^dThe overall model for 4 was significant, $\chi^2_{(2)} = 9.27, p < .05$; ^eThe overall model for 5 was significant, $\chi^2_{(2)} = 22.18, p < .001$.

Results

Performance on the mental state questions for each ToM story is presented in Table 2.2 for Group (HFASD versus typically developing) and Age (child versus adolescent) separately. We conducted separate binary logistic regression analyses to establish the influence of Group and Age on each of the five stories. Table 2.2 summarizes each of these models and presents the observed and predicted (conditional) probabilities of passing each task (rather than odds ratios) by Group and Age. Overall, although some of the advanced ToM stories were clearly more difficult than others, there was no evidence that children with HFASD performed differently on the mental state questions than their TD counterparts. Furthermore, regardless of the type of story, adolescents performed consistently better than children.

Given such consistency, a composite advanced ToM score was created by summing the scores on the five mental state questions, resulting in a 0-5 score, $M = 3.5$, $SD = 1.17$. Despite the fact that the five stories tap distinct conceptual domains of advanced ToM, and emerge at different times developmentally, a reliability analysis was nonetheless conducted to examine whether any single task elicited qualitatively different response patterns. The advanced ToM score had a Cronbach's alpha of .46, which is modest, but all items were positively correlated (corrected item-total correlations ranged between .21 and .32) and the overall alpha was not improved by omitting any item. Moreover, these reliabilities confirm earlier findings on advanced ToM stories in ASD (Hughes et al., 2000; Peterson, Slaughter, & Paynter, 2007). Given the theoretical connection between these domains, the composite advanced ToM score was thus used in subsequent analyses. Below, we further examine the relation between advanced ToM and Group accounting for (1) chronological age and verbal ability, (2) physical state inference scores, and (3) autism severity (ADOS and SRS).

Table 2.3 shows significant positive correlations between participants' advanced ToM and their chronological age and verbal ability in both groups. However, partial correlations controlling for verbal ability revealed a non-significant correlation between chronological age and advanced ToM in both groups. Conversely, partial correlations controlling for chronological age revealed a significant correlation between verbal ability and advanced ToM in both groups. These analyses clearly illustrate that chronological age per se is not critical for advanced ToM understanding, rather it is the level of receptive verbal ability, in absolute terms, that is of importance for both participants with HFASD as well as TD children and adolescents.

Regarding physical state inferences, an initial 2 (Group) x 2 (Age) ANOVA was conducted with physical state score as a dependent variable. Results showed that there were no differences in physical state inferences between participants with HFASD and their typically developing counterparts, $F_{(1,253)} = 0.00$, $p = .96$, but there was a strong effect of Age, $F_{(1,253)} = 27.18$, $p < .001$. Table 2.3 shows that advanced ToM and physical state score were significantly positively correlated within both groups, but this association only remained significant for children and adolescents with HFASD once chronological age and verbal ability were controlled for using partial correlations.

Table 2.3 Pearson correlations between advanced ToM score, age, receptive verbal ability, physical state score, and autism severity (ADOS and SRS).

	Advanced ToM score	
	HFASD (n=194)	TD (n=60)
Age	.30 ***	.50 ***
Absolute receptive verbal ability	.38 ***	.56 ***
Physical state score	.40 ***	.39 **
ADOS severity score	-.04	-
SRS total score (HFASD: n=152; TD: n=47)	-.07	.05
Age (controlling for verbal ability)	-.01	.12
Age (controlling for physical state score)	.23 **	.38 **
Absolute receptive verbal ability (controlling for age)	.25 ***	.31 ***
Absolute receptive verbal ability (controlling for physical state score)	.22 **	.47 ***
Physical state score (controlling for age)	.37 ***	.17
Physical state score (controlling for verbal ability)	.29 ***	.19

Note. HFASD = high-functioning autism spectrum disorder; TD = typically developing; *** = $p < .001$; ** = $p < .01$.

Analysis of variance was used to examine possible effects of Group and Age on the composite advanced ToM score. Again, there was no main effect of Group on advanced ToM, $F_{(1,253)} = 2.34$, $p = .13$, but there was a highly significant effect of Age, $F_{(1,253)} = 22.80$, $p < .001$, with adolescents outperforming children. The interaction between Group and Age was not significant, $F_{(1,253)} = 0.05$, $p = .83$. We also conducted an analysis of

covariance (ANCOVA) to examine potential group effects on the total advanced ToM index, while controlling for the influences of chronological age, verbal ability, and physical state score. There was no main effect of Group, $F_{(1,251)} = 1.38, p = .24$.

Participants' scores of autism severity (both ADOS and SRS) were not significantly correlated with advanced ToM (see Table 2.3). Also, none of the subscales of the ADOS or SRS correlated significantly with advanced ToM (r 's ranging from -.13 to .11). In a final set of analyses, we examined whether autism severity nonetheless affected advanced ToM when other important variables were controlled for. We repeated an ANCOVA with advanced ToM score as dependent variable, Group as fixed factor with three levels (HFASD with a high (≥ 7) ADOS score; HFASD with a low (< 7) ADOS score; TD comparison), and chronological age, verbal ability, and physical state index as covariates. Again, no main effect of group was found, $F_{(2,234)} = 0.77, p = .46$, confirming that autism severity, as measured on the ADOS, had no significant effect on advanced ToM performance.

Discussion

Compared to the myriad of studies on false belief understanding in children with ASD and varying intellectual abilities, advanced Theory of Mind (ToM) understanding in children and adolescents with high-functioning ASD has received relatively little attention. Therefore, we used five advanced ToM stories to examine the performance of a large sample of children and adolescents with high-functioning ASD (HFASD), and compared them to typically developing peers. Performance on the second-order false belief story and the emotional display rule story approached ceiling, particularly for the adolescents, but the stories about double bluff, faux pas, and sarcasm were more difficult for all participants. However, counter to our expectations, no group differences were found on any of the stories. Furthermore, adolescents performed consistently better than children, irrespective of HFASD status. Yet, it was not their age, but rather absolute verbal abilities and general reasoning capacity that appeared to underlie better advanced ToM understanding.

Counter to some previous findings on advanced ToM in children with HFASD (Beaumont & Sofronoff, 2008; Brent et al., 2004; Sobel et al., 2005; White et al., 2009), we did not find an advanced ToM impairment in children with HFASD (6-12 years). Similarly, although previous results were equivocal concerning advanced ToM abilities in adolescents and adults with HFASD, we did not find support for an advanced ToM impairment in adolescents with HFASD (12-20 years). Both children and adolescents with

HFASD in our study performed equally well as their typically developing counterparts. Hence, when advanced ToM is operationalized by a set of complex social stories, high-functioning children and adolescents with ASD appear to be equally capable of inferring mental states of story protagonists. Although this finding is incompatible with the Theory of Mind theory of ASD, children and adolescents with HFASD may nonetheless show limited ability to infer mental states during social interactions occurring in everyday life. Indeed, parents report everyday mindreading problems in their child with ASD, even when the child succeeds on first-order ToM tasks (Peterson et al., 2009). Also, adults with HFASD who succeed on static advanced ToM tasks such as the Strange Stories, still show ToM problems when evaluating a videotaped social conversation (Ponnet et al., 2008; Roeyers et al., 2001). This apparent discrepancy may be due to the complexity of everyday social interactions compared to the social situations as described in ToM stories. Also, the demands of social interactions are not as explicitly defined as mental state questions. Therefore, other factors such as social attention and motivation may crucially determine whether or not the relatively intact conceptual understanding of mental states is activated and used in individuals with HFASD (Klin, Jones, Schultz, & Volkmar, 2003). Increased attention and motivation might also explain why a substantial proportion of our participants with HFASD only showed *subclinical* symptoms of autism on the ADOS, a semi-structured interaction with an adult experimenter, even though their clinical diagnoses imply that they experience profound social difficulties in their everyday lives.

Several alternative explanations for our non-significant results will be discussed below. First, because our participants with a clinical diagnosis of HFASD were characterized by relatively mild autism symptomatology with a substantial number not reaching the ASD cutoff on the ADOS (< 7), this mild autism severity might explain why our HFASD sample performed equally well on the advanced ToM task as typically developing peers. However, it must be noted that children's autism severity was not related to their ToM task performance, and a considerable number of our participants did meet ADOS criteria for ASD. Hence, mild autism severity in the HFASD group can be ruled out as an alternative explanation for the results. Second, it may be argued that the participants with HFASD, in particular the adolescent group, had a slight advantage on the advanced ToM task due to their significantly older age than the typically developing comparison group. However, even after controlling for chronological age, their advanced ToM task performance remained comparable to the typically developing group. Third, it could be that our measure of advanced ToM was not sensitive enough to detect group differences. The advanced ToM task consisted of a collection of five social stories, all

derived from previously validated ToM measures (Begeer et al., 2011; Kaland et al., 2008; Sullivan et al., 1994). Possibly, the first two stories were unable to differentiate participants with and without HFASD due to a ceiling effect. However, the same pattern of non-significant group differences was found for the latter three stories. Furthermore, each of the stories, except for the emotional display rule story, showed a significant age effect with adolescents performing better than children. This shows that the stories are in fact sensitive enough to detect potential group differences.

As expected, children's ToM task performance was positively associated with their chronological age, verbal ability, and physical state index (Bauminger & Kasari, 1999; Fisher et al., 2005; Happé, 1995). Importantly, these links were found in both groups. The positive association between age and ToM could be largely explained by a third factor: verbal abilities. Hence, it is not age per se, but an absolute growth in verbal abilities that increases a child's success on advanced ToM tasks. This may not be surprising given the highly verbal nature of the advanced ToM task. However, this finding also raises the interesting question whether differences in performance on an advanced ToM task are first and foremost determined by children's verbal abilities instead of their mental state reasoning. Future studies should try to include advanced ToM measures that are less intertwined with children's verbal abilities and more closely related to children's social competence in everyday life.

Positive correlations between physical state and mental state inferences were only found in the HFASD group, controlling for chronological age and verbal ability. This may indicate that children and adolescents with HFASD rely more heavily on their general reasoning abilities than typically developing peers do to solve advanced ToM tasks. Hence, children with HFASD may use non-social heuristics and general logic to understand others' intentions and desires (Peterson et al., 2009). In typical development, children's social understanding is embedded in their experience of social interactions (Carpendale & Lewis, 2004). Possibly, a different social interaction pattern from birth onwards results in a more analytical and theoretical understanding of what drives others' behavior (Klin et al., 2003). As the current study has shown, a different way of understanding other minds may not necessarily impair performance on a static ToM task. However, under less than perfect circumstances (implicit social demands, time constraints), such is the case with everyday social interactions, individuals with HFASD may still experience profound problems understanding the mental worlds of others. Hence, individuals with HFASD seem to master the concept of ToM without mastering the ability to use such insight in the service of their ongoing social interactions.

Chapter 3

Empathic responsiveness of children and adolescents with high-functioning autism spectrum disorder

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Abstract

Previous studies have shown reduced concern for and attention to others' emotions in preschoolers with autism spectrum disorder (ASD) and an intellectual disability. We examined the empathic responsiveness of normally intelligent school-aged children and adolescents with a clinical diagnosis of ASD (n=151) and typical development (n=50) using structured observations and parent reports. Participants' empathic responses to an interviewer were surprisingly similar. However, compared to parents of a typically developing child, parents of a child with ASD reported significantly fewer empathic responses, particularly when the child received a high score on the Autism Diagnostic Observation Schedule (ADOS). Hence, according to parents normally intelligent children and adolescents with ASD show impairments in their everyday empathic responsiveness compared to typically developing peers.

Introduction

Empathic responsiveness, the ability to respond to the emotional state of another person, is a core element of our social functioning (de Waal, 2008). A difficulty to empathize with others has been linked to autism (Blair, 2005) ever since the first clinical descriptions of autism by Leo Kanner who noted that 'these children have come into the world with an innate inability to form the usual, biologically provided emotional contact with people' (1943, page 250). However, surprisingly little work has been done on observed empathic responsiveness in children and adolescents with an autism spectrum disorder (ASD). The present study aims to fill this gap by examining the empathic responsiveness of a large group of normally intelligent children and adolescents with ASD with structured observations and parent reports, and comparing it to that of typically developing peers.

While there is much debate on the exact nature and definition of empathy (Preston & de Waal, 2002), most researchers agree that empathy includes both an affective and a cognitive component (Blair, 2005; Jones, Happé, Gilbert, Burnett, & Viding, 2010; Rogers, Dziobek, Hassenstab, Wolf & Convit, 2007; Yirmiya, Sigman, Kasari, & Mundy, 1992). The affective component of empathy generally refers to a vicarious emotional response to others' emotional states (Eisenberg & Miller, 1987; Knafo, Zahn-Waxler, Van Hulle, Robinson, & Rhee, 2008), while the cognitive component includes the ability to acknowledge and reason about own and others' mental states and emotions (Baron-Cohen, 1989). It is generally recognized that young children with ASD show limited cognitive empathic abilities compared to their typically developing peers (for a review see Yirmiya, Erel, Shaked, & Solomonica-Levi, 1998). Most research on empathy in ASD has

thus far mainly focused on these impairments in children's conceptual understanding of others' emotions and mental states and to a lesser extent to their emotionally sharing the emotions of others. Even fewer studies have looked at the *behavioral* component of empathy, that is, whether children with ASD behave empathically in response to others' emotions. This is remarkable given the emphasis in diagnostic assessments of ASD on social behavioral rather than cognitive problems (APA, 2000; Lord et al., 2000; Lord, Rutter, & Le Couteur, 1994).

Structured observations of empathic responsiveness in primarily pre-schoolers with ASD or children with ASD and an intellectual disability have been used in experimental designs where a parent or an experimenter displayed distress. For example, an adult experimenter would pretend to accidentally hurt him/herself by hitting a finger with a toy hammer (Sigman, Kasari, Kwon, & Yirmiya, 1992). Afterwards the child's empathic responsiveness was coded based on video footage (e.g., duration or frequency of looks to the experimenter). Typically developing one-year-olds respond to the experimenter's distress by showing increased attention and emotional concern (Hutman et al., 2010; Zahn-Waxler, Radke-Yarrow, Wagner, & Chapman, 1992). However, even though children with ASD and an intellectual disability are not oblivious to nor actively withdraw from the emotions of others (Corona, Dissanayake, Arbelle, Wellington, & Sigman, 1998; Dissanayake, Sigman, & Kasari, 1996), they look less at the distressed adult, show less emotional concern and make fewer sympathetic verbal responses when compared to matched controls (Bacon, Fein, Morris, Waterhouse, & Allen, 1998; Hobson, Ruth, García-Pérez, & Hobson, 2009; Loveland & Tunali, 1991; Scambler, Hepburn, Rutherford, Wehner, & Rogers, 2007; Sigman et al., 1992). Furthermore, prospective studies have also revealed poorer empathic responsiveness to other's distress in toddlers with varying intellectual abilities who were later diagnosed with ASD (Hutman et al., 2010; McDonald & Messinger, 2012). In short, previous experimental studies support a deficit in the empathic responsiveness of young children with ASD and children with ASD and an intellectual disability.

It is currently unclear whether the observed lack of empathic responsiveness can be generalized to school-aged children and adolescents with ASD and a normal intelligence (high-functioning ASD; HFASD). One of the few studies that examined empathic responsiveness to a distressed adult in a group of children with HFASD failed to show any differences with a typically developing comparison group (Bacon et al., 1998). This suggests that a normal IQ may benefit the empathic responsiveness of children with ASD, possibly to the extent that their presumed impairment in empathic responsiveness

disappears ('cognitive compensation'; cf. Yirmiya et al., 1992). In addition, as children grow older, their social experiences expand, and this may continue to improve their social competence. Indeed, parent reports suggest that children with ASD show an overall improvement in empathic responsiveness from middle school to late adolescence (McGovern & Sigman, 2005). In adolescence and adulthood, self-reports of individuals with ASD even suggest intact responses to others' emotional states (Dziobek et al., 2008; Jones et al., 2010; Rogers et al., 2006), though the validity of these self-reports has been debated (Johnson, Filliter, & Murphy, 2009). Still, normal IQ and older age likely promote children's empathic responsiveness.

Both structured observations and parent reports provide unique and diagnostically relevant perspectives on a child's behavior (Noterdaeme, Mildenerger, Sitter, & Amorosa, 2002; Stone, Hoffman, Lewis, & Ousley, 1994). Parents or caregivers provide necessary information about the child's (early) development and everyday behavior (Ozonoff et al., 2009). They have experience with their child's empathic responsiveness across a full range of everyday social situations, including their child's responses to their own emotional states. Reports from parents suggest that children with ASD respond less empathically to others' emotional states than typically developing children (Hudry & Slaughter, 2009; Johnson et al., 2009). Yet, although parent reports of children's behavior are generally reliable (e.g., Dirks & Boyle, 2010) and meaningful (Verhulst, Koot, & Van der Ende, 1994), agreement across informants tends to be low to moderate (Achenbach, McConaughy, & Howell, 1987; Posserud, Lundervold, & Gillberg, 2006). It seems plausible that parents interpret their child's behavior differently than objective viewers would (Capps, Kasari, Yirmiya, & Sigman, 1993). Indeed, only a fair amount of agreement has been found when comparing diagnostic outcomes based on the 'gold standard' parent interview for ASD (ADI-R; Lord, Rutter, & Le Couteur, 1994) and the 'gold standard' observation measure of ASD (ADOS-G; Lord et al., 2000) (Papanikolaou et al., 2009). Furthermore, parental reports of a child with ASD may be biased by parental psychopathology (Bennett et al., 2012) or incorrect recollections of the child's early development (Hus, Taylor, & Lord, 2011). Hence, in diagnostic assessments it is equally important that an independent party observes the child's behavior.

In the current study we examined children's empathic responsiveness in a large sample of children and adolescents with HFASD and typically developing peers by using two different methods. First, participants' behavioral responses to the emotional displays (happiness, sadness, pain) of an interviewer were videotaped and coded. To approximate real life social situations, the emotional cues of the interviewer were presented as naturally

occurring events during the interview. Second, parents were asked to describe the anticipated responses of their child in situations comparable to the ones used in the interview. We hypothesized that children and adolescents with HFASD would demonstrate fewer empathic responses to the emotional states of an adult interviewer when compared to the typically developing comparison group. Also, we expected parents of a child with HFASD to report limitations in their child's empathic responsiveness when compared to that of typically developing children.

Method

Participants

Children and adolescents with a clinical diagnosis of ASD were recruited via a specialized school for normally intelligent children with ASD (Berg en Boschschool). School admission criteria included a normal IQ (IQ > 70) and a clinical diagnosis of ASD. The diagnostic classification of ASD in the Netherlands is commonly given by a psychiatrist according to established DSM-IV-TR-criteria and based on an elaborate examination, both observations and parent interviews, by multiple experienced clinicians (psychologists, psychiatrists and educationalists). The comparison group consisted of typically developing children and adolescents and was recruited via public primary and secondary schools.

Thirty-nine of the 214 participants with HFASD were excluded from the final analysis, because parents did not return the parent questionnaire about their child's empathic responsiveness. Twenty-four more participants with HFASD were excluded from the analysis due to a poor video record (n=1), inadequate IQ assessment (n=4), a verbal receptive IQ under 70 (n=3) and inadequate or incomplete parent report (n=2) or observation data (n=14). Within the comparison group, 23 of the 73 typically developing participants were excluded from the final analysis due to a missing parent questionnaire (n=16), a poor video record (n=1), inadequate observation (n=1), inadequate IQ assessment (n=1), or a high level of autistic characteristics as indicated by a high score (> 70) on the parent version of the Social Responsiveness Scale (n=4; Constantino & Gruber, 2007). All parents of the final sample of 50 children (44 boys; 6 girls) in the comparison group confirmed that their child had no ASD diagnosis, see also Table 3.1.

Table 3.1 Descriptives for the three groups of participants.

	High ADOS Group (n = 56)		Low ADOS Group (n = 95)		Comparison Group (n= 50)		Group differences
Child variables	<i>M (SD)</i>	<i>Range</i>	<i>M (SD)</i>	<i>Range</i>	<i>M (SD)</i>	<i>Range</i>	
Age (in years)	13.0 (2.98)	6.4 – 18.8	13.7 (2.97)	6.4 – 18.8	11.6 (2.72)	6.0 – 16.8	High & Low > C
Receptive verbal IQ	103.4 (12.85)	72 - 126	106.5 (12.94)	76 - 132	107.2 (12.22)	85 - 132	n.s.
Gender (boys; girls) (n)	54; 2		76; 19		44; 6		Girls: High < Low
Clinical ASD diagnosis (n) (Autism; AS; PDD-NOS)	9; 5; 42		20; 15; 60		0; 0; 0		High & Low > C
Total ADOS (module 3 or 4)	10.2 (2.85)	7 - 19	3.1 (1.77)	0 - 6	-	-	High > Low
Total SRS	84.5 (21.18)	36 - 128	78.2 (23.59)	23 - 133	31.2 (11.89)	13 - 63	High & Low > C
Environment variables	<i>M (SD)</i>	<i>Range</i>	<i>M (SD)</i>	<i>Range</i>	<i>M (SD)</i>	<i>Range</i>	
Number living with both biological parents; other (n)	46; 10		73; 22		39; 8 (3 miss)		n.s.
Educational level mother ^a	4.7 (1.56)	1 - 7	4.7 (1.55)	1 - 7	5.2 (1.73)	2 - 7	n.s.
Educational level father ^a	4.2 (1.84)	1 - 7	4.9 (1.61)	1 - 7	5.0 (1.46)	2 - 7	High < Low
Level of profession mother ^b	3.0 (1.15)	0 - 5	3.1 (1.08)	0 - 5	3.4 (1.31)	0 - 5	n.s.
Level of profession father ^b	3.1 (0.94)	0 - 5	3.4 (0.85)	1 - 5	3.5 (1.01)	2 - 5	High < C

Note. ASD = Autism Spectrum Disorder; AS = Asperger's syndrome; PDD-NOS = Pervasive Developmental Disorder-Not Otherwise Specified; ADOS = Autism Diagnostic Observation Schedule; SRS = Social Responsiveness Scale; High = High ADOS group with HFASD; Low = Low ADOS group

with HFASD; C = Typically developing comparison group; n.s. = no significant group differences. a 1 = elementary school; 2 = lower professional; 3 = middle secondary; 4 = middle professional; 5 = higher secondary; 6 = higher professional; 7 = academic education; b 0 = no profession; 1 = elementary; 2 = lower; 3 = middle; 4 = higher; 5 = academic level.

The final group of participants with HFASD consisted of 151 children and adolescents (130 boys; 21 girls) with a clinical diagnosis of autism (n=29), Asperger's syndrome (n=20), or PDD-NOS (n=102). Participants with HFASD were also assessed with the Autism Diagnostic Observation Schedule (ADOS; Lord et al., 2000). Previous studies already indicated a relatively poor sensitivity of the ADOS to classify individuals with PDD-NOS and identify adults with high-functioning ASD (Bastiaansen et al., 2011; Gotham et al., 2008). Indeed, based on ADOS scores, participants were divided in two groups: (1) a group with both a clinical diagnosis of ASD and a research diagnosis of ASD, that is, a total ADOS score at/above the ASD cutoff of 7 (n=56; from now on referred to as 'high ADOS group'), and (2) a group with a clinical diagnosis of ASD, but an ADOS score below the ASD cutoff (n=95; from now on referred to as 'low ADOS group'). The high and low ADOS groups were both significantly older and received significantly higher scores on the Social Responsiveness Scale (SRS; Constantino & Gruber, 2007) than the comparison group (see Table 3.1), confirming that both groups showed more parent reported autistic traits than children from the comparison group. The high and low ADOS group however did not differ significantly in their SRS scores, nor in their distribution of clinical diagnoses (autistic disorder, Asperger's syndrome and PDD-NOS).

Measures

Structured observation of empathic responsiveness.

Empathy was evoked by a display of emotion by the interviewer on three different occasions during the test procedure. The displays of emotion were designed to look like naturally occurring events during the procedure. This method was derived from previous studies (e.g., Loveland & Tunali, 1991; Scambler et al., 2007) and adapted to be suitable for a high-functioning group with a wide age range. All situations were first tried out during a pilot study with children and adolescents with and without HFASD (n=52). During the pilot study none of the children noticed anything strange about the interviewer's behavior except for one typically developing adolescent girl. Therefore, the situations were considered as sufficiently realistic.

To increase the participant's attention for the interviewer's emotions the interviewer showed each emotion in between tasks that were offered as part of a large battery of psychological tests. The entire test procedure lasted 1.5 hours. Each display of the interviewer's emotion was separated by approximately 20 minutes in the test procedure. The order of the three types of emotions shown by the interviewer depended on the interview version. In one version, after 30 minutes into the interview, the

interviewer showed happiness, followed by sadness later on in the interview. In the other version the order of happiness and sadness was reversed. Pain was always shown last. The two interview versions were counter-balanced across groups.

Response to happiness; At the beginning of the test procedure, the interviewer said that he/she would leave his/her mobile phone on, because he/she was expecting an important message or call. We reasoned this announcement would reduce the participant's surprise as the interviewer pretended to check a text message on the mobile phone. The interviewer displayed happiness to the participant by pretending to receive a positive text message. Depending on the version of the interview, the interviewer either said: 'Oh, that's nice [prompt 1]. My friend invites me to the cinema tonight [prompt 2]', or: 'Oh, that's good [prompt 1]. My friend is going to be fine. He/she can leave the hospital [prompt 2]'. Between the first and second prompt, the interviewer paused for two seconds. The interviewer showed a happy facial expression corresponding to the emotional content of the text message and looked at the participant.

Response to sadness; The interviewer displayed sadness by pretending to receive a negative text message on his/her phone. Depending on the version of the interview, the interviewer either said: 'Oh, that's a shame [prompt 1]. My friend tells me the tickets for the movie are sold out [prompt 2]', or: 'Oh, that's bad [prompt 1]. My friend had a bike accident. He/she broke an arm [prompt 2]'. Between the first and second prompt, the interviewer paused for two seconds. The interviewer showed a sad facial expression and looked at the participant.

Response to pain; The interviewer displayed distress by pretending to have neck pain. The interviewer would rub his/her neck, have a distressed facial expression and moan, while not looking at the participant. After ten seconds the interviewer would give a second prompt by saying: 'I had neck pain for a while.'

Parent reports of empathic responsiveness

Empathy vignettes were modeled after the empathy evoking situations during the interview and were used to assess parental views on the empathic responsiveness of their child. Each vignette described a social situation which resembled one of the empathy evoking situations during the interview. In the vignette the emotions were displayed by a teacher, because this resembled the situation with the adult interviewer as closely as possible (fully matched vignettes would require parents to imagine and describe their child's responses to simulated emotions of an adult interviewer during a psychological test procedure, which would be unrealistic). After each vignette parents were asked to describe

the child's anticipated response. They were instructed to write down the most probable response, but they were free to report as many responses as they wished. Instructions and the vignettes are shown in Appendix II (p. 167).

Peabody Picture Vocabulary Test-III-NL

The Peabody Picture Vocabulary Test (Dunn & Dunn, 2004) assesses receptive vocabulary and is highly correlated with more general measures of verbal IQ (Hodapp & Gerken, 1999). Based on the PPVT participants received a receptive verbal IQ-score standardized for age.

Autism Diagnostic Observation Schedule-Generic (ADOS-G)

The ADOS (Lord et al., 2000) is a semi-structured diagnostic observation measure to assess the presence and severity of ASD-specific impairments in social reciprocity, communication, fantasy, and repetitive interests and behaviors. The ADOS-interviewer uses playful activities (e.g., reading a story book) and topics of discussion (e.g., peer problems) to assess the socio-communicative abilities of the child. Each of the child's behaviors is rated on a 3-point-scale (0 = normal behavior; 1 = slightly abnormal behavior; 2 = clearly abnormal behavior). Items in the social reciprocity domain and the communication domain are added to make up a total ADOS score. An ADOS score of 7 or higher is indicative of an ASD. The ADOS has excellent internal consistency, interrater reliability, test-retest reliability, and discriminant validity (Lord et al., 2000).

Procedure

After receiving informed consent from parents and participants themselves (if 12 years or older), each participant was individually tested at school. The test procedure involved a full battery of tests, including the structured observation of empathic responsiveness. Interviewers were 14 trained graduate students in psychology, health science or medicine. It was not possible to keep the interviewers blind for the child's clinical status, because the location of testing gave it away. However, all interviews were videotaped and transcribed, and coded by three graduate students who were blind to the diagnosis of the participants. After children participated in the study, their parents received a booklet of questionnaires at home concerning their child's behavior. This booklet also contained the empathy vignettes discussed in the present study.

Coding

Structured observation of empathic responsiveness. Participants' responses to each prompt of the interviewer during the empathy evoking situations were coded from video recording into five different and mutually exclusive response categories containing both verbal and non-verbal behaviors (see Table 3.2), based on Loveland and Tunali (1991) and Bacon et al. (1998). Because each situation contained two prompts a participant could show a response 0 to 6 times (2 prompts x 3 situations) during the interview. Hence, for each of the five response categories a participant received a score ranging from 0 to 6.

Coders of the children's responses to the interviewer were three graduate students who were not informed about the children's diagnoses and ADOS scores. Coder 1 and 2, who were responsible for 88% of all coding of the structured observations, double coded 30 participants (coder 1 and 3: 10 participants). Exact agreement between coder 1 and coder 2 on children's observed responses ranged between 79% and 90% (coder 1 and 3: 60-90%), with kappa's ranging from .68 to .85 (adequate to good agreement).

Parent reports of empathic responsiveness. Parent reports of the child's empathic responses were assigned to the same five response categories that were used for the structured observation. The frequency of each type of response was tallied across the vignettes. Coder 1 and 2 double coded the parent reports of 30 participants. Exact agreement ranged between 97% and 100%, with the three computed kappa's showing a perfect agreement of 1.00.

Manipulation check

At the end of the test session each participant was asked whether he/she had noticed anything unusual during the interview. None of the participants stated to have noticed anything unusual about the interviewer's behavior.

Statistical analysis

Multivariate analyses of covariance (MANCOVA's) were performed to test for possible group differences (high ADOS group, low ADOS group, comparison group) in the proportion scores of the five response categories (i.e., proportion of each type of response compared to total number of responses) in both observed behavior and parent report. Age was added as a covariate in the analyses, because participants with HFASD were significantly older than the comparison group and age was correlated with some of the dependent variables. Finally, for each measure of empathic responsiveness a series of multiple regression analyses was conducted to examine the unique contribution of

Table 3.2 Categories of responses obtained from structured observations and parent reports.

Category	Definition	Examples of responses to other's emotional states		
		Happiness (e.g., going to the movies)	Sadness (e.g., friend broke an arm)	Pain (e.g., neck pain)
Empathic response	Child gives a relevant verbal response including an empathic reference to the other's emotional state, or offers solutions to alleviate the other's distress.	- 'That sounds like fun.' - 'That's nice.'	- 'I'm sorry to hear that.' - 'Would you like to call your friend?'	- 'Are you okay?' - offering or getting help to alleviate the pain
Relevant response	Child gives a relevant verbal response, but response does not include an empathic reference to the other's emotional state or solutions to alleviate the other's distress.	- 'Which movie?'	- 'I broke my arm once.'	- 'I have that after playing video games.'
Confirmatory response	Child briefly confirms that he/she has heard the other person.	- nodding, smiling - 'Ok', 'Yes'	- nodding - 'Ok', 'Yes'	- nodding - 'Ok', 'Yes'
Attention without response	Child attends to the other person, but does not give a response.	- looking, but no response	- looking, but no response	- looking, but no response
No response or irrelevant response	Child does not attend or respond to the other person, or gives an irrelevant or inappropriate response.	- no attention or response - 'When do we have a break?'	- no attention or response - 'What kind of phone is that?'	- no attention or response - laughing

receptive verbal IQ and age to the variance in empathic responsiveness above and beyond the variance explained by group status. In the multiple regression analysis, group was entered in the first step of the model, followed either by receptive verbal IQ or age in the second step. To check for possible group differences in the association of receptive verbal IQ or age with children's empathic responsiveness, this was followed up by separate regression analyses for each group.

Results

First, we checked whether there was a bias in the responses to the 14 different interviewers. A MANOVA was run with interviewer as fixed factor and proportion scores of the five response categories as dependent variables. No significant differences were found in responses to the interviewers (all p 's $> .10$) except for one interviewer (J.) receiving significantly more empathic responses than two other interviewers (B. and A.). This was likely due to the older age of J's participants with HFASD ($M = 17.6$ years, $n = 5$) compared to the participants with HFASD of B. ($M = 13.5$ years, $n = 10$) and A. ($M = 12.7$ years, $n = 10$). However, removing J's participants from the analyses yielded the same results, therefore it was decided to keep them.

Second, it was checked whether the three emotional situations during the interview differed in proportion of elicited empathic responses. Sadness generally evoked more empathic responses ($M = .20$) than happiness ($M = .13$; $t(198) = 3.59, p < .001$) or pain ($M = .14$; $t(193) = 2.66, p < .01$), which is in line with recent reports (Bandstra, Chambers, McGrath, & Moore, 2011). Separate analyses within each of the three participant groups showed that the interviewer's sadness generated more empathic responses than happiness ($p < .05$) and pain ($p < .05$) in the comparison group, but these differences in empathic responses were non-significant in the high ADOS group, albeit in the same direction. Within the low ADOS group, participants also responded with significantly more empathic responses to the interviewer's sadness than happiness ($p < .05$), but the difference between sadness and pain was non-significant.

Structured observation of empathic responsiveness

A MANCOVA with Group (high ADOS group, low ADOS group, comparison group) as fixed factor, proportion scores of the five response categories as dependent variables, and age as a covariate, showed no main effect of Group on the proportion of empathic responses. While 48% of the participants in the high ADOS group showed at least one or more empathic responses during the interview, this was also true for 59% of

the low ADOS group and 52% of the comparison group. In fact, Group effects were not found on any of the response categories except for the response category ‘no response or irrelevant response’ ($F(1, 199) = 3.70, p < .05, \text{partial } \eta^2 = .04$). This group effect was followed up by post-hoc Bonferroni corrected tests, which revealed that the high ADOS group more frequently displayed no or irrelevant responses than participants from the comparison group ($p < .05$) (see Table 3.3). No significant difference was noted in this response category between the high and the low ADOS group, and the low ADOS group and comparison group.

Parent reports of empathic responsiveness

MANCOVA's on parent reports of their children's empathic responsiveness demonstrated a main effect of Group on proportion of empathic responses ($F(1, 198) = 22.46, p < .001, \text{partial } \eta^2 = .19$), relevant responses ($F(1, 198) = 4.27, p < .05, \text{partial } \eta^2 = .04$), attention without response ($F(1, 198) = 7.03, p < .01, \text{partial } \eta^2 = .07$), and non-responses or irrelevant responses ($F(1, 198) = 8.34, p < .001, \text{partial } \eta^2 = .08$). Subsequent Bonferroni corrected post-hoc analyses indicated that parents expected their child with a high ADOS score to show fewer empathic responses than children from the low ADOS group ($p < .001$), who in turn were expected to show fewer empathic responses than the comparison group ($p < .01$), see Table 3.3. To illustrate these differences: 23% of the parents with a child from the high ADOS group did not report any empathic response, compared to 10% of parents in the low ADOS group and only 2% in the comparison group. Moreover, 16% of parents with a child from the high ADOS group reported four or more empathic responses, compared to 40% in the low ADOS group and 62% in the comparison group.

In contrast to the empathic responses, participants in the high ADOS group were expected to show significantly more relevant responses than the comparison group ($p < .05$). Also, compared to both other groups, the high ADOS group was expected to show more often ‘attention without response’ (low ADOS: $p < .05$; comparison group: $p < .01$) and ‘no response or irrelevant response’ (low ADOS: $p < .01$; comparison group: $p < .001$).

Table 3.3 Mean proportion scores and standard deviations of each response type for both the structured observation and parent report of empathic responsiveness.

	High ADOS group (n = 56)	Low ADOS Group (n = 95)	Comparison group (n = 50)	Group differences (Mancova)
Structured observation:	<i>M (SD)</i>	<i>M (SD)</i>	<i>M (SD)</i>	
Empathic response	.12 (.16)	.18 (.19)	.13 (.15)	n.s.
Relevant response	.22 (.24)	.23 (.20)	.19 (.17)	n.s.
Confirmatory response	.17 (.17)	.16 (.17)	.20 (.15)	n.s.
Attention without response	.25 (.25)	.27 (.21)	.34 (.18)	n.s.
No or irrelevant response	.23 (.20)	.16 (.19)	.14 (.17)	High > C
Parent report:	<i>M (SD)</i>	<i>M (SD)</i>	<i>M (SD)</i>	
Empathic response	.41 (.33)	.61 (.30)	.78 (.24)	High < Low < C
Relevant response	.27 (.23)	.22 (.22)	.17 (.21)	High > C
Confirmatory response	.02 (.06)	.03 (.07)	.00 (.03)	n.s.
Attention without response	.15 (.28)	.07 (.17)	.01 (.07)	High > Low & C
No or irrelevant response	.15 (.19)	.07 (.14)	.04 (.10)	High > Low & C

Note. High = High ADOS group with HFASD; Low = Low ADOS group with HFASD; C = Typically developing comparison group; n.s. = no significant group differences.

Verbal IQ and age

Receptive verbal IQ failed to add a significant amount of explained variance in empathic responses during the structured observation or in parent reports above and beyond the variance already explained by group status. Separate regression analyses within each group (high ADOS group, low ADOS group, comparison group) all confirmed a non-significant association between children's receptive verbal IQ and their empathic responses in the interview or parent reports. However, when age was added in the second step of the regression model, a significantly positive association was found with proportion of empathic responses during the structured observation ($\beta = .26$, $\Delta R^2 = .06$, $p < .001$) and proportion of empathic responses in parent reports ($\beta = .13$, $\Delta R^2 = .02$, $p < .05$). Hence, an increase in participant's age was associated with an increase in empathic responses. Separate regression analyses within each group (high ADOS group, low ADOS group, comparison group) further showed that this was particularly true for the

comparison group, where age was positively associated with empathic responses both during the structured observation ($\beta = .33, \Delta R^2 = .11, p < .05$) and in parent reports ($\beta = .27, \Delta R^2 = .07, p = .06$). This positive association between age and empathic responses was weaker within the low ADOS group (observation: $\beta = .29, \Delta R^2 = .08, p < .01$; parent report: $\beta = .14, \Delta R^2 = .02, p > .10$) and non-significant within the high ADOS group (observation: $\beta = .01, \Delta R^2 = .00, p > .10$; parent report: $\beta = .07, \Delta R^2 = .00, p > .10$).

Discussion

Empathic responsiveness of a large sample of normally intelligent children and adolescents with a diagnosis of autism spectrum disorder (HFASD) and typically developing peers was systematically examined using structured observations and parent reports. Counter to our expectation, observed responses to the emotional states of an interviewer were largely comparable for participants with and without HFASD. However, children's empathic responsiveness as described by parents was substantially reduced in the group of participants with HFASD and a high ADOS score, both compared to the low ADOS group and the typically developing comparison group.

Previous experimental studies have consistently demonstrated reduced attention and concern towards distressed adults in children with ASD and an intellectual disability compared to a matched comparison group (Bacon et al., 1998; Scambler et al., 2007; Sigman et al., 1992). Considering these results, it was surprising that responses during the structured observation were highly similar for children with and without HFASD. Children and adolescents with HFASD equally often provided an empathic response, a relevant response, or a brief response indicating that they had listened to the interviewer. They also equally often paid attention to the interviewer after his/her emotional display. A modest group difference was only noticed in the 'lower order' response category of non-responses, irrelevant and inappropriate responses to the interviewer's emotions. Compared to the typical comparison group, participants with more severe ASD symptoms (ADOS score ≥ 7) showed these types of responses more frequently after the interviewer's emotional display.

Overall, children and adolescents with HFASD may be equally able as typically developing peers to respond empathically to the emotions of an unfamiliar adult during a semi-structured interaction. Thus, when circumstances are kept relatively simple (a dyadic interaction, display of one basic emotion) children with and without HFASD are equally likely to give an overt empathic response. Our observation data correspond with self-reports of adequate affective empathy in adolescents and adults with ASD (Dziobek et al.,

2008; Jones et al., 2010; Rogers et al., 2006). Alternatively, it could be that a normal IQ serves as a protective factor against an impaired empathic responsiveness in children and adolescents with HFASD (Yirmiya et al., 1992). Children with HFASD may rely actively on their cognitive abilities to respond adequately to others' emotions. While cognitive compensation would explain the observational data, the lack of association between receptive verbal IQ and empathic responsiveness within this normal IQ sample suggests that intellectual abilities may no longer add meaningfully to variance in empathic responsiveness once above a particular threshold. Compared to the (pre)school age of participants in earlier studies, the older age of the participants in this study may be another factor to explain the apparent contrast with previous findings. School-aged children and adolescents with HFASD may continue to develop their empathic responsiveness, as has already been suggested by longitudinal studies using parent reports (McGovern & Sigman, 2005). Indeed, in our study older participants tended to show more empathic responses. However, a closer examination showed that this age effect was present within the typically developing group and the low ADOS group, but missing within the group of participants with HFASD and high ADOS scores. This suggests that the empathic responsiveness of children and adolescents with HFASD and more severe autistic traits may be less sensitive to developmental growth.

Parent reports clearly pointed to a reduced everyday empathic responsiveness in children and adolescents with HFASD, which agrees with previous findings (Hudry & Slaughter, 2009; Johnson et al., 2009). According to parents, their child with HFASD will respond less empathically and more passively to the emotional displays of an adult. This is particularly true for children and adolescents with an HFASD diagnosis and more severe autistic characteristics as indicated by a high ADOS score. However, it should also be noted that even among children and adolescents with HFASD, approximately half of all parent reported responses were coded as empathic responses (41% in high ADOS group, 61% in low ADOS group). Apparently, most parents with a child with HFASD do expect at least some adequate empathic responses from their child, although substantially less so when compared to the 78% of all responses reported by parents of a typically developing child. Importantly, parent reports of children's empathy also proved to be sensitive to variance in autistic traits as evidenced by a significantly higher frequency of anticipated empathic responses in the comparison group compared to the low ADOS group, that in turn was expected to show more empathic responses than the high ADOS group. Thus, parent reports of empathic responsiveness appear to be inversely associated with children's autism severity.

The present study warrants caution when using standardized observations of empathic responsiveness during a social interaction with an unfamiliar adult (e.g., a psychiatrist) in diagnostic assessments of ASD. These observations are commonly included in diagnostic assessments of children referred with concerns of ASD (Lord et al., 2000). The results of our study suggest that explicit empathic responses from a normally intelligent school-aged child or adolescent during a structured observation do not refute an ASD diagnosis. However, unresponsiveness or irrelevant responses to other's emotions may be seen in normally intelligent children with more severe forms of ASD.

This study has several limitations. First, although none of the participants stated to have noticed anything unusual about the interviewer's behavior, we cannot be certain that all participants found the emotions of the interviewer clear and credible. However, particular care was taken to make the emotional displays of the interviewer age appropriate and realistic for our participants. The emotions shown by the interviewer were purposely made more subtle than the distress displayed by interviewers in previous studies with toddlers (Sigman et al., 1992). Second, judgments of the empathic responsiveness of participants were primarily based on the verbal content of their responses, although helping behavior and looking behavior was also included. An advantage of coding these explicit behaviors is that these can be coded easily (no facial coding system required) and reliably (reduced subjectivity of the observer). Yet, more subtle impairments in the empathic responding of children with HFASD might still exist, but these were not targeted in the present study. This might also explain why parent reports do point to a reduced empathic responsiveness in children and adolescents with HFASD, while the structured observation failed to demonstrate this reduction in empathy. Future studies on empathic responsiveness should therefore include both explicit and more implicit (e.g., facial expression, response timing) behavioral measures. Also, it would be informative to examine children's empathic responses to different social partners. For instance, a peer may evoke more empathic responses in a child than an experimenter or a teacher due to increased familiarity and similarity (Preston & de Waal, 2002; Saarni, 2001).

An impaired ability to empathize with others has been continually emphasized in clinical observations and diagnostic guidelines of autism (APA, 2000; Kanner, 1943; Lord et al., 2000). Our observation data partly refine the idea of an empathy deficit in ASD by showing that school-aged children and adolescents with and without HFASD show equal levels of empathic responses to an interviewer's display of affect. However, parent reports did point to a limited empathic responsiveness in children and adolescents with HFASD compared to typically developing peers. Notably, parents still expected their child with

HFASD to respond empathically in approximately half of all situations. In sum, our main findings highlight that it is not the presence of empathic responses which indicates typical development; rather it is the marked *absence* of adequate responses to others' emotions which indicates an atypical, and possibly autistic, development.

Chapter 4

*Can you tell me something about yourself?
Self-presentation in children and adolescents with
high-functioning autism spectrum disorder
in hypothetical and real life situations*

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Abstract

The self-presentation skills of children and adolescents with high-functioning autistic spectrum disorder (HFASD) and typically developing (TD) controls were compared, in response to both hypothetical and real life situations. In both situations, 26 HFASD and 26 TD participants were prompted to describe themselves twice, first in a baseline condition, and later in a goal-directed condition where specific information was given about the preferences and demands of the audience. Confirming and extending previous research, both TD and HFASD participants exhibited a tendency to be more positive when describing themselves in a goal-directed condition. However, HFASD participants were less strategic than TD participants in responding to the information they were given about the audience preferences and demands. Possible explanations and implications of the results are discussed.

Introduction

Self-presentation refers to an individual's efforts to shape the self-image that is portrayed to others. Thus, in the process of self-presentation an actor regulates his or her behaviour in order to manipulate the impression being made on an audience (Levine & Feldman, 1997). In order to do so, the actor has to: a) understand the expectations of the audience; b) be aware of his or her own abilities and characteristics in this respect; and c) be able to match the two in such a way that he or she creates the desired impression. The first two abilities – awareness of the normative values and preferences of others and awareness of one's own assets – are central to the development of the self. Children as young as 8 years have been found to be sensitive to audience preferences by selecting particularly those self-descriptions that are relevant to the audience (Aloise-Young, 1993; Banerjee, 2002). Children continue to develop these skills throughout childhood. In the current study we focus on self-presentation skills in children and adolescents with autism spectrum disorder (ASD).

There are several reasons to hypothesise that self-presentation skills may be deviant or delayed in individuals with ASD. An abundant number of studies has shown that individuals with ASD find it difficult to understand the intentions and feelings of others (e.g., Klin, Jones, Schultz, & Volkmar, 2003). This may cause them to misinterpret the preferences and evaluations of their audience, in turn making it difficult to achieve successful self-presentation. Furthermore, individuals with ASD often insist on routines and strict rules (e.g., Corbett, Constantine, Hendren, Rocke, & Ozonoff, 2009; Russo et al., 2007). This lack of flexibility complicates the adaptation of their self-presentation to different audiences. While direct studies on self-presentation are sparse, research on self-

awareness and processing of self-related information are in line with the suggested poor self-presentation in ASD.

In a series of experiments, Hobson, Chidambi, Lee and Meyer (2006) demonstrate that children with ASD show awareness of other's engagement with themselves to a lesser extent than verbally matched children without autism. Social emotions that are often considered the consequence of such engagement, like embarrassment, coyness (self-conscious, embarrassed smiling) or guilt, were less frequently found and were also considered to be of a slightly different quality in children with ASD. However, when explaining social emotions, these children do refer to others' perspectives on themselves, suggesting at least some understanding of basic self-presentational processes (Bauminger, 2004; Heerey, Keltner, & Capps, 2003; Hillier & Allinson, 2002). In a similar way, emotional display rules seem to be a stumbling block for individuals with ASD, even when their intelligence is average or above average (High Functioning ASD, HFASD). Compared to typically developing control children, those with HFASD were less sophisticated in inhibiting the expression of their true emotions (i.e., applying an emotional display rule), despite their adequate explanations of display rules (Barbaro & Dissanayake, 2007; Dennis, Lockyer, & Lazenby, 2000; Peterson et al., 2005). In short, the awareness of others' perspectives on oneself is not wholly absent, but is likely not applied appropriately in children with HFASD.

Several studies have hinted at a deviant processing of self-related information in autism. The processing of personal experiences in individuals with ASD may be more rule-based than emotion-driven (Williams & Happé, 2009a). This could diminish superior knowledge of one's own mental states, which is typically accompanied by emotions, over the mental states of others. However, studies have produced mixed results about superior recognition or recall of *self*-related information over *other*-related information in individuals with ASD. Some studies did not find this so called self-reference effect in individuals with ASD when compared to typically developing controls (Henderson et al., 2009; Toichi et al., 2002), whereas others did (Lind & Bowler, 2009; Williams & Happé, 2009b). Typically developing individuals do assign more knowledge about their inner states to themselves than to a close other, but this effect of privileged inner insight was not observed within a group with HFASD (Mitchell & O'Keefe, 2008). This detached and objective perspective on the self has been suggested as part of the allocentric perspective of individuals with autism, described by Frith and De Vignemont (2005), and merits further research.

In a preliminary investigation of self-presentational processes in autistic children, Begeer et al. (2008a) asked children with HFASD and typically developing children how

they would present themselves in two different hypothetical situations. In a baseline condition children were asked to describe themselves without knowledge of audience preferences. In a self-promotion condition children were encouraged to convince the audience of their competence, as they were told they might get selected for a prize-winning game. In the baseline condition children with HFASD spoke less positively about themselves than typically developing children. Both groups of children, however, were receptive to the condition manipulation: they expressed more positive statements about themselves in the self-promotion condition compared to the baseline condition. Yet, children with HFASD were less strategic than typically developing children in the self-promotion condition: their self-presentation was less relevant to competencies that were particularly useful for the desired activity (e.g., game-related skills).

The present study was designed to extend Begeer et al.'s (2008a) research by including a wider range of self-presentational contexts. Specifically, the study of Begeer et al. (2008a) could not shed light on how children's self-presentation strategies are used in real life situations, and how these compare with their responses in hypothetical situations. Older children with HFASD may perform reasonably well on cognitive social tasks (e.g., Capps, Yirmiya, & Sigman, 1992), but they may still experience subtle difficulties delineating social situations in real life (e.g., Channon, Charman, Heap, Crawford, & Rios, 2001). For this reason, the current study not only focused on self-presentation in response to hypothetical situations, but also included real life interactions with an interviewer.

The present study also covered a wider age range, including both school aged children and adolescents. It is not yet clear how self presentation strategies develop in individuals with HFASD. On the one hand, self-presentation strategies of adolescents with HFASD could be expected to benefit from developmental growth compared to younger children with HFASD. There are clear indications of improvement in behaviour from childhood to adolescence and adulthood in individuals with ASD (McGovern & Sigman, 2005; Seltzer et al., 2003). On the other hand, it is not uncommon for individuals with ASD to develop psychiatric and psychosocial problems in adulthood (Hofvander et al., 2009; Howlin, Mahwood, & Rutter, 2000). Due to an accumulation of social experiences, adolescents with HFASD may become increasingly aware of their own social difficulties (Meyer, Mundy, Van Hecke, Durocher, 2006; White, Oswald, Ollendick, & Scahill, 2009). This may induce a less positive self-presentation. Indeed, research suggests older children with HFASD provide lower evaluations of their social competence than younger children with HFASD (Vickerstaff, Heriot, Wong, Lopes, & Dossetor, 2007). This could be

indicative of an increasingly realistic perspective of individuals with HFASD on their own social competence.

To sum up, the present study compared the self-presentation skills of children and adolescents with HFASD to typically developing controls in response to hypothetical and real life situations. Based on the findings of Begeer et al. (2008a), we expected children from both HFASD and typically developing groups to speak more positively about themselves in the goal-directed conditions compared to the baseline conditions, both in response to hypothetical situations as well as during a real life interaction with an interviewer. Second, we predicted that the HFASD group would express fewer positive statements about themselves than the typically developing group in the baseline conditions, and investigated the extent to which this presumed group difference would be more pronounced among adolescents. Crucially, based on the existing evidence regarding social cognition, self-understanding, and self-focus in ASD samples, we also expected that the HFASD group would exhibit a tendency to be less strategic (i.e., less responsive to specific audience demands and preferences) than typically developing controls in the goal-directed conditions. We also evaluated the extent to which this group difference would be attenuated among adolescents.

Method

Participants

The HFASD group consisted of 15 school aged children and 11 adolescents and young adults with HFASD, henceforward ‘adolescents with HFASD’ (see Table 4.1). The typically developing control group consisted of 16 children and 10 adolescents. The diagnostic classification of the HFASD participants was based on assessments by a psychiatrist and multiple informants (psychologists and educationalists). All participants fulfilled established diagnostic criteria according to the DSM-IV-TR (APA, 2000). Additional diagnostic information about the participants was obtained from the parents with the Social Responsiveness Scale (Constantino & Gruber, 2007). As would be expected, the HFASD group scored substantially higher on the SRS than typically developing controls ($t(51) = 10.31, p < .001, d = 3.04$). In the HFASD group there was a wide range of raw SRS scores: 44-152. Despite their clinical diagnosis six adolescents received SRS scores below 70: the recommended cutpoint to screen for ASD. Five of the six adolescents with relatively low SRS-scores were university students and two lived independently. The SRS might be a less suitable measure to assess ASD in intelligent adolescents and young adults with ASD. Importantly, post hoc t-tests concerning our

most important variables showed no difference between adolescents scoring below and above the ASD cutpoint. Also, none of the typically developing participants approached or surpassed this threshold (range of raw SRS scores: 11-46). The HFASD group and typically developing group did not differ on age ($t(51) = 1.29, p > .10, d = 0.36$), gender ratio ($\chi^2(1) = 1.65, p > .10, \phi = 0.06$), or receptive verbal IQ ($t(51) = 0.13, p > .10, d = 0.04$) as assessed by the Dutch version of the Peabody Picture Vocabulary Test-III (Dunn & Dunn, 2004).

Table 4.1 Sample description.

	Group with HFASD			Typically developing group		
	Child (n = 15)	Adolescent (n = 11)	Total (n = 26)	Child (n = 16)	Adolescent (n = 10)	Total (n = 26)
	<i>M (SD)</i>	<i>M (SD)</i>	<i>M (SD)</i>	<i>M (SD)</i>	<i>M (SD)</i>	<i>M (SD)</i>
Age (in years)	8.8 (1.36)	20.3 (2.08)	13.6 (6.05)	7.8 (0.78)	17.8 (1.09)	11.7 (5.07)
Gender (boys ; girls)	14;1	9;2	23;3	16;0	6;4	22;4
Receptive verbal IQ	109 (15.92)	110 (5.11)	109 (12.17)	109 (10.35)	110 (8.40)	109 (9.49)
Social responsiveness (SRS)	94 (15.17)	82 (35.07)	88 (26.73)	30 (7.44)	24 (10.41)	28 (8.78)

Measures

Hypothetical Task

Baseline condition. Hypothetical self-presentation was examined using two vignettes about peer interactions (adapted from the peer interaction scenarios of Banerjee, 2002). In the baseline condition participants were told the following: ‘Imagine you move to a different neighbourhood. The family living next to you has a son/daughter [*matched to participant’s gender*]. The boy/girl next door introduces him/herself to you and you want the boy/girl to like you. What would you tell him/her about yourself?’ Participants were asked to explain their choice of self-description.

Goal-directed condition. In the goal-directed condition participants were told the following: ‘Imagine you go to a new school where you know none of the students. The only thing you know about the person sitting next you is that he/she [*matched to participant’s gender*] likes animals very much and he /she also likes people who like animals. You want

the boy/girl to like you. What would you tell him/her about yourself?' Participants were asked to explain their choice of self-description.

Real Life Task

Baseline condition. The Real Life Task is based on the scenarios described in Begeer et al. (2008a). In the baseline condition the interviewer first introduced him/herself and then asked the participant: 'Can you tell me something about yourself?' After the child's first answer the interviewer would ask: 'Can you tell me something more about yourself?'

Goal-directed condition. In the goal-directed condition the interviewer said: 'A couple of the participating children will be picked to play a game where you can win lots of cool prizes. To determine who should be picked for this game with prizes, I ask everyone to tell me something about him/herself. So, can you tell me something about yourself?' To ensure children would not purposely leave out information they had already mentioned in the baseline condition, children were told information on this paper would later be used to choose children for the game. After the child's first answer the interviewer would ask: 'Can you tell me something more about yourself?' Because the type of game (e.g. physical/intellectual game, competitive/cooperative game) was not specified, game preferences were not expected to influence self-promoting. Still, three participants (one child with HFASD, one typically developing adolescent and one adolescent with HFASD) were not interested in the game. Exclusion of these participants did not affect the comparability of the two groups with regard to age, gender ratio and verbal IQ (all t 's ≤ 1.07 ; all p 's $\geq .29$).

Procedure

These tasks were part of a longer battery of assessments. The interview was either conducted by a psychology graduate student or a PhD student. Because it was natural for the interviewer and child to get acquainted with each other at the very beginning of the interview, every session started with the baseline condition of real life self-presentation. To reduce transfer risk (e.g. learning) from one task to the other, all tasks were separated by at least 20 minutes, with the exception of the two conditions of hypothetical self-presentation. Thus, an hour after the real life baseline condition, the real life goal-directed condition started. After another 20 minutes this was followed by hypothetical self-presentation. In line with the Real Life Task, the baseline condition was always offered first. Children's responses were taped and transcribed.

Coding

Positive self-statements. In both the Hypothetical Task as well as the Real Life Task, all self-statements were counted and evaluated. A self-statement was defined as a self-referring sentence with 'I' as grammatical subject or other self-referring statement. Positive self-statements included positive affect (like, enjoy), abilities (good at something) or socially desirable attributes (being nice). For examples, please see Table 4.2.

Strategic self-statements. Every positive self-statement in the goal-directed conditions was categorized as strategic or non-strategic. Strategic self-statements included those positive self-statements that were relevant to a particular preference of the audience. Hence, in the Hypothetical Task a self-statement was coded as strategic when it was a positive self-statement about animals. In the Real Life Task a self-statement was coded as strategic when it was a positive self-statement about games.

Social justifications. Justifications of answers in the Hypothetical Task were coded as either social or non-social (other). In a social justification the importance of a social relationship with another peer is acknowledged.

References to honesty or truth. In the Hypothetical Task, presence of one or more references to honesty or truth was coded.

Results

Positive self-statements in baseline vs. goal-directed conditions

The percentage of positive self-statements was analyzed using a 2 (Group: HFASD vs. TD) x 2 (Age: children vs. adolescents) x 2 (Task: Hypothetical vs. Real Life) x 2 (Condition: baseline vs. goal-directed) analysis of variance (for descriptive statistics see Table 4.3). All participants expressed more positive self-statements in the Hypothetical Task than in the Real Life Task ($F(1, 28) = 5.90, p < .05, d = 0.40$; 44 % vs. 30 % respectively). As predicted, participants were more positive about themselves in the goal-directed conditions compared to the baseline conditions ($F(1, 28) = 67.32, p < .001, d = 1.36$). Importantly, this effect did not interact with Group ($F(1, 28) = 0.26, p > .10, d = 0.08$). Furthermore, an Age x Condition interaction ($F(1, 28) = 19.74, p < .001, d = 0.74$) emanated from adolescents' tendency to give more positive self-statements than children in the baseline condition ($t(28) = 1.81, p = .08, d = 0.67$), whereas children gave far more such statements than adolescents in the goal-direction condition ($t(28) = 4.10, p < .001, d = 1.34$). No other two-, three- or four-way-interactions were found (all F 's ≤ 1.29 ; all p 's $\geq .27$; all d 's ≤ 0.19).

Table 4.2 Coding scheme of dependent variables.

Variable	Definition	Example	Interrater reliability (*)
Self-statement	Self-referring sentence with “I” as grammatical subject or other self-referring statement	<i>‘I am eight years old’</i> <i>‘My name is ...’</i>	$\alpha = .95 - .98$
Positive self-statement	Self-statement which includes a positive affect, abilities or socially desirable attributes	Positive affect: <i>‘I like dancing’</i> Abilities: <i>‘I am really smart’</i> Socially desirable attributes: <i>‘I am kind to other children’</i>	$\alpha = .68 - .89$
Strategic self-statement	Positive self-statement that is relevant to the preference of an audience	Hypothetical Task: <i>‘I love animals’</i> Real Life Task: <i>‘I like winning with monopoly’</i>	$\alpha = .78 - .88$
Non-strategic self-statement	Positive self-statement that is irrelevant to the preference of an audience	Hypothetical Task: <i>‘I can make a good drawing’</i> Real Life Task: <i>‘I love airplanes’</i>	$\alpha = .70 - .96$
Social justification	Participant’s justification of answers where the importance of peer relations is acknowledged	<i>‘Because I want to be friends’</i> <i>‘So he can get along with me’</i>	$\kappa = .50$
Reference to honesty or truth	Reference of the participant to being honest or truthful	<i>‘Because in real life that is really true’</i> <i>‘Because that’s just the way it is, who I am’</i>	$\kappa = .58$

* Agreement between two independent raters was obtained for 63% of the self-statements (in the baseline and goal-directed condition of the Hypothetical Task and Real Life Task) and 100 % of the social justifications and references to honesty or truth (in the Hypothetical Task). Social justifications and references to honesty and truth were coded as present (1) or absent (0), therefore kappa values were calculated.

Table 4.3 Proportions of positive self-statements in baseline and goal-directed conditions averaged over Hypothetical and Real Life Task (range = 0-1).

Condition	Group							
	Children				Adolescents			
	HFASD (n = 7)		TD (n = 10)		HFASD (n = 9)		TD (n = 9)	
	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>
Baseline	.15	.18	.07	.07	.23	.22	.33	.19
Goal-directed	.73	.24	.75	.26	.33	.19	.55	.20

Table 4.4 Proportions of strategic and non-strategic self-statements averaged over Hypothetical and Real Life Task (range = 0-1).

Condition	Group							
	Children				Adolescents			
	HFASD (n = 10)		TD (n = 9)		HFASD (n = 10)		TD (n = 9)	
	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>
Strategic	.62	.21	.71	.32	.20	.16	.46	.17
Non-strategic	.14	.19	.04	.08	.18	.15	.09	.09

Strategic self-statements in goal-directed conditions

The percentages of all self-statements that were categorized as positive and strategic or positive and non-strategic, in the goal-directed conditions only, were analyzed using a 2 (Group: HFASD vs. TD) x 2 (Age: children vs. adolescents) x 2 (Task: Hypothetical vs. Real Life) x 2 (Strategy: strategic vs. non-strategic) analysis of variance (for descriptive statistics see Table 4.4). All participants expressed more positive self-statements in the Hypothetical Task than in the Real Life Task ($F(1, 37) = 4.39, p < .05, d = 0.30$; 67 % vs. 51 % respectively). More importantly, participants used more strategic self-statements than non-strategic self-statements in the goal-directed conditions ($F(1, 37) = 67.79, p < .001, d = 1.19$). A Group x Strategy interaction ($F(1,37) = 8.41, p < .01, d = 0.42$) originated from the HFASD group expressing significantly more non-strategic self-statements ($t(37) = 2.15, p < .05$ (one-tailed), $d = 0.70$) and fewer strategic self-statements ($t(37) = 1.96, p < .05$ (one-tailed), $d = 0.64$) than the TD group, averaged over both tasks. Furthermore, an Age x Strategy interaction ($F(1, 37) = 15.96, p < .001, d = 0.58$) was observed: children in general expressed more strategic self-statements than adolescents ($t(37) = 4.36, p < .001, d = 1.41$), but there was no age effect on non-strategic self-

statements ($t(37) = 0.91, p > .10, d = 0.14$). A Task x Strategy interaction was produced ($F(1, 37) = 14.76, p < .01, d = 0.56$), because in the Hypothetical Task all participants expressed relatively more strategic self-statements ($t(37) = 3.41, p < .01, d = 0.55$) and fewer non-strategic self-statements ($t(37) = 2.75, p < .01, d = 0.45$) than in the Real Life Task. Finally, a Group x Task trend was found ($F(1, 37) = 3.56, p = .07, d = 0.28$), because controls used more positive self-statements (strategic and non-strategic self-statements combined) than the HFASD group in the goal-directed condition of the Hypothetical Task ($t(37) = 2.00, p = .05, d = 0.65$), while no such group difference was found in the goal-directed condition of the Real Life Task. However, because this finding was based on combined strategic and non-strategic statements, it was not informative about strategic self-presentation. No other two-, three- or four-way-interactions were found (all F 's ≤ 1.78 ; all p 's $\geq .19$; all d 's ≤ 0.19).

Social justifications

In the Hypothetical Task, all participants were asked to explain their choice of self-description, and we tallied the number of participants providing one or more social justifications. Eleven of 20 participants with HFASD (55 %) mentioned at least one social justification against 12 of 17 TD participants (71 %) ($\chi^2(1) = 0.95, p > .10, \phi = .16$).

References to honesty or truth

In the Hypothetical Task, we also tallied the number of participants providing one or more references to honesty or truth. Eleven of 20 participants with HFASD (55 %) mentioned at least once their tendency to be honest or telling the truth, whereas only 3 of the 17 TD participants did (18 %) ($\chi^2(1) = 5.45, p < .05, \phi = .38$).

Discussion

The present study adds to our understanding of a neglected aspect of self-related reasoning and behaviour in ASD samples. First of all, it must be noted that the HFASD group performed surprisingly well. Compared to typically developing controls, the children and adolescents with HFASD expressed a similar proportion of positive self-statements in the baseline conditions and they also showed an increase in positive self-statements in the goal-directed conditions. Yet, importantly, the present study also extends one of the key findings reported by Begeer et al. (2008a). Compared to typically developing peers, children and adolescents with HFASD gave fewer self-descriptions that were specifically

relevant for audience preferences. In other words, their self-presentation was less strategic than the self-presentation of typically developing peers.

Because the HFASD group did not differ from the control group in their increase of positive self-statements from baseline to goal-directed condition, the HFASD group does appear to be receptive to audience preferences when sufficiently motivated. This builds on mounting evidence that HFASD children are receptive to manipulations focused on personal gain (Begeer, Rieffe, Meerum Terwogt, & Stockman, 2003, 2006). There was no interaction of this effect with task, suggesting that the increase in positive self-descriptions was generated to a similar degree by a real life prize incentive and by a hypothetical social incentive (to be liked by a peer audience). Interestingly, the hypothetical context in general elicited relatively more positive self-statements than real interactions with an interviewer, which could be interpreted as an overestimation of real life behaviour. Typically developing controls were even more positive in the goal-directed condition of the Hypothetical Task compared to the children and adolescent with HFASD, while no such group difference was observed in the Real Life Task. Also, although in both types of tasks participants mentioned more strategic than non-strategic self-statements in the goal-directed conditions, this pattern was more pronounced in the Hypothetical Task than in the Real Life Task. An overestimation of real life behaviour may be influenced by a self-enhancement bias: the tendency for mentally healthy people to describe themselves more positively compared to a normative criterion (e.g., Taylor & Brown, 1988). Another possible reason for the less positive self-presentation in real life may be the distracting element of complex and ambiguous real life situations compared to clearly circumscribed hypothetical situations. However, the two tasks used in the present study were different in several important ways; hence further research is needed to explore task differences more systematically.

Our analysis also showed that adolescents, both typically developing and with HFASD, were more positive than children in the baseline conditions, but less positive than children in the goal-directed conditions. Nonetheless, it is important to note that both children and adolescents did significantly increase positivity in the goal-directed conditions. It seems possible that the specific content of the goals/preferences in the two tasks used in this study (prizes and liking animals) was effective in eliciting positive self-descriptions across the age range, but was relatively less effective in engaging the enthusiasm and motivation of adolescents compared to children. This could also explain why the children, who were expected to have less efficient self-presentation strategies than adolescents, actually expressed more strategic self-statements compared to the adolescents.

Hence, no support was found for a developmental increase in strategic self-presentation, in either group. Future research should include an activity or goal that is empirically determined to be equally motivating for younger and older youths.

Contrary to the Begeer et al. (2008a) study, there was no evidence for any group difference in positive self-statements. This may be due to the fact that in the present study, participants generally provided fewer positive self-statements in the baseline conditions, thus making it more difficult to find robust group differences. Furthermore, we did not find support for a specific developmental decline in positive self-perceptions within the HFASD group, because we did not find a Group x Age interaction. However, because of the small sample size for each age group in this study, we advocate further research on this question.

Importantly, we also demonstrated that the lower levels of strategic self-promotion reported by Begeer et al. (2008a) in HFASD children can be generalised to other self-presentation tasks and to a wider age range. Specifically, in the goal-directed conditions, individuals with HFASD expressed fewer strategic self-statements and more non-strategic self-statements than typically developing controls. The lower levels of strategic responding in the HFASD group, compared to typically developing controls, cannot be attributed to a different evaluation of the incentives for the two groups. As noted earlier, both children and adolescents with HFASD seemed to be broadly responsive to the goal-directed conditions, as shown by an increase in positive self-statements. Indeed, in the hypothetical situation, the basic motive of being liked by others was identified as often by the HFASD group as by the TD group. Other research also suggests that individuals with ASD have a comparable desire for friendships as evinced by feelings of loneliness (Bauminger & Kasari, 2000).

There may be an alternative explanation that helps to account for the low strategic score of the HFASD group, and the HFASD adolescents in particular. A closer look at the qualitative data of the Hypothetical Task showed that four of 11 adolescents with HFASD made it explicitly clear they would not make strategic self-statements (e.g., 'I like cats'), because they considered it to be dishonest or false. Therefore, it seems some adolescents with HFASD do in fact know when self-promotion strategies are expected, yet are reluctant to change their self-presentation to the varying requirements of an audience. Indeed, statements that referred to being honest or telling the truth were found more often in the HFASD than the control group.

This pattern is thrown into sharp relief by existing research showing that typically developing adults are more likely to lie about themselves when instructed to self-promote

in a dyadic conversation, than when they are only instructed to have a neutral conversation (Feldman, Forrest, & Happ, 2002): sixty percent of all participants indicated they had told at least one lie during a 10 minute conversation. Hence, individuals who find it hard to deceive, may also find it hard to self-promote. The difficulties of individuals with HFASD in deceiving others (e.g., Barbaro & Dissanayake, 2007; Yirmiya, Solomonica-Levi, & Shulman, 1996) may be the result of poor perspective-taking or Theory of Mind abilities (e.g., Baron-Cohen, 1992). Yet, some adolescents with HFASD clearly did comprehend how they could make a positive impression on an audience, but did not act accordingly. Hence, the perspective-taking skills of this group did not appear to be hampered. An alternative, socio-motivational explanation of less strategic self-presentation in individuals with HFASD – despite intact knowledge about self-presentation – lies in their tendency to rigidly stick to moral and social rules (e.g., Begeer et al., 2010), even at the expense of possible self-presentational gains. A possibly heightened awareness of the norms and conventions of the general public (e.g. ‘Lying is bad’) may stem from a lack of superior focus on self- rather than other-related information (Henderson et al., 2009; Toichi et al., 2002). A socio-motivational explanation of this kind can also help us understand why this pattern may become more pronounced with age, since social norms are likely to become more internalized over the course of development.

The present study has some other important limitations that can be addressed in future work. First, as noted earlier, the specific choice of tasks in the present study was varied, because we aimed to evaluate the generalisability of the effects reported by Begeer et al. (2008a). However, this diversity makes it somewhat difficult to draw conclusions about specific task features that influence self-presentational behaviour. There were some main effects of task in the present study, suggesting that future studies of situational variables that could influence self-presentation (e.g., adult versus peer audience, social versus non-social goal, audience preference) could be fruitful. All of these variables can also be examined in both real life and hypothetical contexts, but it must be acknowledged that precisely matched real life and hypothetical contexts in a repeated measures design will increase the risk of transfer effects from one task to the other.

Second, the sample size of the present study was rather small, which may have influenced the ability to detect subtle effects. A four-way ANOVA on data from such a small sample warrants some caution while interpreting the results. We did replicate a key finding of Begeer et al. (2008a), but further investigations of other variables such as age and task may require a larger sample. Indeed, it would be helpful to have a more complete picture of the development of self-presentation in participants with HFASD. The present

study has shed some light on the generalisability of self-presentational patterns across a wider age range than previously studied, but the gap between 10 and 16 years needs to be filled. Longitudinal research in particular would be valuable for determining how self-presentational patterns emerge in youths with autism. Finally, the present study did not directly assess the motivational and cognitive factors that could underpin self-presentation skills. What milestones does a child have to reach to make a successful self-presentation possible? Is this different for typically developing children and children with HFASD? In our discussion above, we have identified Theory of Mind, flexibility, deceptive skills, and motivation as potentially relevant, and these deserve systematic exploration in future research.

Chapter 5

The role of temperament, Theory of Mind, and executive functioning in the empathic responsiveness of children with autism spectrum disorder

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Abstract

Children with autism spectrum disorder (ASD) have shown reduced empathic responsiveness to others' emotions. Given the large individual differences frequently noted within the autism spectrum, we examined the role of individual differences in temperament, Theory of Mind, and executive functioning in the empathic responsiveness of children and adolescents with ASD and a normal intelligence (n=121; 6-19 years). Empathic responsiveness was based on structured observations and parental reports. A hierarchical multiple regression analysis revealed that temperament explained significant variance (15%) in children's empathic responsiveness, whereas Theory of Mind and executive functioning failed to add explained variance. Hence, the empathic responsiveness of children and adolescents with ASD is not merely determined by their ASD diagnosis, but also by individual temperamental characteristics.

Introduction

Autism is often described as a developmental disorder with a core deficit in empathy. Some theorists have proposed a lack of affective empathy in autism, in particular a lack of emotional relatedness to the emotions of others (Hobson, 2002; Kanner, 1943). Instead, others have emphasized a lack of cognitive empathy in autism, indicating an impaired understanding of others' mental states (Baron-Cohen, 1989; Blair, 2005). A lack of cognitive empathy is one of the major cognitive deficit models of autism (Rajendran & Mitchell, 2007). Not surprisingly, empathy research in children with autism spectrum disorder (ASD) has thus far mainly focused on impairments in cognitive empathy by looking at children's performances on Theory of Mind tasks (see Boucher, 2012, for a recent review), leaving their empathic responsiveness in everyday social interactions relatively under-examined. We define empathic responsiveness here as the ability to respond appropriately to others' emotions. The few studies that directly observed children's responses to an experimenter's display of emotion (mostly distress) have shown reduced attention and concern in children with ASD compared to matched peers (Bacon, Fein, Morris, Waterhouse, & Allen, 1998; Scambler, Hepburn, Rutherford, Wehner, & Rogers, 2007; Sigman, Kasari, Kwon, & Yirmiya, 1992). However, these studies mainly included (preschool) children with ASD and an intellectual disability. Therefore, in a previously submitted paper the authors addressed the question whether limitations in empathic responsiveness would also be evident in school-aged children and adolescents with ASD and a normal intelligence (high-functioning ASD; HFASD). Children's empathic responses to the emotional displays of an experimenter were coded by independent observers and parents were asked to describe their child's anticipated

empathic responses. Surprisingly, children with HFASD and typically developing IQ matched peers responded largely similar to the emotions of the experimenter. However, parents of a child with HFASD reported substantially fewer empathic responses than parents of a typically developing child. Still, parents of a child with HFASD expected their child to respond empathically in approximately half of all situations. Our research findings suggest that empathic responsiveness may not be universally impaired in children with ASD and may also vary according to the measure that is used (structured observation or parent report). In the present paper we will address factors that may account for individual differences in the empathic responsiveness of children and adolescents with HFASD.

Research on the empathic responsiveness of typically developing children has devoted attention to developmental changes (Young, Fox, & Zahn-Waxler, 1999) and individual temperamental differences (Eisenberg, Wentzel, & Harris, 1998) that underpin variation in children's empathic responsiveness. However, most experimental studies on the empathic responsiveness of children with ASD have restricted their analyses to group-level comparisons (ASD vs. typical development or intellectual disability) thereby ignoring individual differences within the autism spectrum. Even though a striking variation in (dis)abilities of children with ASD is consistently reported (Jones & Klin, 2009; Mundy, Henderson, Inge, & Coman, 2007; Pellicano, 2010; Scheeren, Koot, & Begeer, 2012), factors that contribute to this variation remain largely unexplored. Age and intelligence have thus far been identified as potentially promoting factors of children's empathic responsiveness in ASD (Bacon et al., 1998; Dissanayake, Sigman, & Kasari, 1996; McGovern & Sigman, 2005). Research in typical development has already demonstrated an important role for temperament in children's empathic responsiveness (Eisenberg et al., 1998), but the impact of temperament on empathy has not yet been examined in children with ASD. Instead, cognitive abilities such as Theory of Mind and executive functioning are more common factors to consider when explaining ASD symptomatology (Rajendran & Mitchell, 2007), and these cognitive factors are therefore considered candidate predictors of children's (lack of) empathic responsiveness in the present study. In sum, we examined whether individual differences in temperament, Theory of Mind and executive functioning could be meaningfully related to individual differences in empathic responsiveness in children and adolescents with HFASD. The potential influence of these factors is discussed below.

In typical development, children's temperamental traits are known to have a large impact on their social development including their empathic responsiveness (Eisenberg et al., 1998; Fox & Henderson, 1999; Sanson, Hemphill, & Smart, 2004). Temperament has

been defined as biologically based individual differences in behavioral style visible from early childhood (Sanson et al., 2004). There is growing consensus in the literature that broad aspects of temperament include ‘*negative emotionality*’ (display of negative emotions), ‘*self-regulation*’ (effortful regulation of attention and emotions) and ‘*sociability*’ (tendency to seek out new situations and social interactions; Sanson et al., 2004). Typically developing children high in negative emotionality and low in self-regulation tend to show less empathy (or ‘*sympathy*’ following the terminology in Eisenberg et al., 1998). Also, children low in sociability and high in shyness tend to have poorer social skills than peers who are high in sociability (Fox, Henderson, Rubin, Calkins, & Schmidt, 2001; Sanson, Letcher, Smart, & Prior, 2009). These temperamental traits, which account for variation in the expression of empathy in children with typical development, may well affect children with HFASD in a similar fashion.

While a specific temperamental make-up may make an empathic response more likely, a child’s empathic responsiveness may crucially rely on his/her ability to ascribe mental states (such as emotions) to others (Rogers, Dziobek, Hassenstab, Wolf, & Convit, 2007), which is a fundamental aspect of Theory of Mind (ToM). Indirect evidence for this conceivable link between ToM and empathic responsiveness comes from studies showing a positive association between ToM and social competence in typically developing children (e.g., Bosacki & Astington, 1999; Caputi, Lecce, Pagnin, & Banerjee, 2012). However, to date, it has been difficult to find a direct link between ToM task performance and everyday social competence in children with ASD (Joseph & Tager-Flusberg, 2004; Travis, Sigman, & Ruskin, 2001). In their study, Peterson, Garnett, Kelly, and Attwood (2009) found half of the children with HFASD who passed ToM tasks were still impaired in their everyday use of ToM during social interactions compared to typically developing children who failed ToM tasks. They tentatively interpreted this finding as an indication that ToM may be necessary, but not sufficient for adequate social interactions. Therefore, in the present study ToM task performances of children with HFASD were expected to be positively, but only modestly related to their empathic responsiveness.

Executive functions (EF), the cognitive abilities which guide all goal-directed behaviors (Hill, 2004), play a central role in a second major cognitive deficit model of ASD (Hill, 2004; Rajendran & Mitchell, 2007). Profound and general EF problems have been observed in ASD when compared to typical development, such as impairments in working memory, cognitive flexibility, vigilance and inhibitory control (Corbett, Constantine, Hendren, Rock, & Ozonoff, 2009; see Willcutt, Sonuga-Barke, Nigg, & Sergeant, 2008, for a review). Indirect support for a link between EF and empathic responsiveness comes

from studies showing positive associations between EF task performances and (later) social competence of children with ASD, ADHD and typical development (McEvoy, Rogers, & Pennington, 1993; Nigg, Quamma, Greenberg, & Kusché, 1998; Rinsky & Hinshaw, 2011). However, other studies have failed to show this association in children with ASD (Joseph & Tager-Flusberg, 2004; Landa & Goldberg, 2005). The inconsistent association between EF task performances and children's social competence may be related to the explicit and often artificial demands and constraints of EF laboratory tasks (e.g., order a set of cards based on color, shape, or number) compared to the dynamics of real life social interactions (Geurts, Corbett, & Solomon, 2009). In order to increase the ecological validity of the EF measure in the present study we used a parent report measure of the child's everyday executive functioning (example item 'My child has difficulty finishing things (chores, homework)').

In the present study we examined whether individual differences in temperament, Theory of Mind, and executive functioning could be meaningfully linked with differences in the empathic responsiveness of children and adolescents with HFASD. Similar to what has been found in typical development, we expected that children's emotionality and shyness would be negatively related to empathic responsiveness, whereas sociability was expected to be positively related with empathy. Also, based on the idea of specific cognitive deficits in ASD, we expected children's empathic responsiveness to benefit from better Theory of Mind understanding and better executive functions. Age and level of intelligence were statistically controlled for in order to estimate the unique contribution of temperament, Theory of Mind and executive functioning to variance in children's empathic responsiveness. Finally, we checked whether children and adolescents with HFASD and varying degrees of autism severity differed in their associations between their empathic responsiveness and the three main predictors.

Method

Participants

The initial sample of participants consisted of 214 (183 boys; 31 girls) Dutch children and adolescents with HFASD recruited via a specialized school for normally intelligent children with ASD (Berg en Boshschool). Only those participants were included in the present study if participant's receptive verbal IQ was 70 or higher, parents returned a completed parent questionnaire (including questions on the child's temperament and executive functioning), and we had complete data on the participant's empathic responsiveness. Hence, 121 participants with a clinical diagnosis of HFASD (104

boys, 17 girls) were included in the final analysis with a mean age of 13.3 years and a mean receptive verbal IQ of 106.1 (see Table 5.1 for complete description). The group of selected participants did not significantly differ from the group of participants with missing data with regard to their age, receptive verbal IQ, gender ratio, or autism severity scores on the Social Responsiveness Scale (SRS; Constantino & Gruber, 2007) and the Autism Diagnostic Observation Schedule (ADOS; Lord et al., 2000).

Twenty-three participants of the final sample (19%) had a clinical diagnosis of autism, 18 (15%) were diagnosed with Asperger's syndrome and 80 (66%) with PDD-NOS. The diagnostic classifications were given by psychiatrists in regular youth mental health care who worked independently from the school and the authors, and who were blind to the goals and outcomes of the present study. Diagnostic information was obtained through examination by multiple experienced clinicians (psychologists, psychiatrists and educationalists) according to established DSM-IV-TR-criteria (APA, 2000). The diagnostic process includes anamneses, proxy reports, and psychiatric, neuropsychological and logopedic examinations. In the present study extra information on children's autism severity was gathered by administering the SRS (Constantino & Gruber, 2007) and the ADOS (Lord et al., 2000; see Table 5.1).

Measures

Empathic responsiveness

Structured observation of empathic responsiveness; On three preset occasions during an individual psychological assessment of 90 minutes, the interviewer displayed an emotion (happiness, sadness, or pain) according to a standardized protocol. Participants' empathic responsiveness to the interviewer's emotions were videotaped and coded by independent observers. The empathy evoking situations were based on previous studies (e.g., Dissanayake et al., 1996; Loveland & Tunali, 1991; Scambler et al., 2007) and adapted to be suitable for a normally intelligent group of a wide age range.

At the beginning of each interview, the interviewer explained that he/she expected an important message or call and therefore would leave his/her mobile phone on. The interviewer pretended to receive two text messages during the interview, each text message was separated by approximately 20 minutes. After reading the fake text message the interviewer would either show happiness or sadness. The emotional display always consisted of two verbal prompts separated by a pause of two seconds. For instance, the interviewer's first happy prompt would be: 'O that's nice', followed by a second prompt: 'My friend invites me to the cinema tonight.'

Table 5.1 Descriptives for the participants (n=121).

Child background	<i>M (SD)</i>	Range
Age (in years)	13.3 (2.72)	6.9 - 18.8
Receptive verbal IQ	106.1 (12.38)	72 - 132
Gender (n)	104 boys;17 girls	
Clinical diagnosis (n): autism; Asperger's Syndrome; PDD-NOS	23; 18; 80	
SRS score	80.8 (22.29)	29 - 133
ADOS score (SA + RRB)	5.7 (4.43)	0 - 18
ADOS severity score	3.37 (2.67)	1 - 10
Dependent variables		
Joint measure ER	0.34 (0.20)	0.00 - 0.75
Observed ER	0.15 (0.17)	0.00 - 0.60
Parent reported ER	0.53 (0.32)	0.00 - 1.00
Independent variables		
EAS - Emotionality	14.7 (4.11)	5 - 24
EAS - Activity	14.3 (3.91)	6 - 23
EAS - Sociability	14.9 (3.57)	7 - 25
EAS - Shyness	15.0 (3.93)	5 - 24
Theory of Mind performance	3.5 (1.14)	0 - 5
BRIEF - Executive Functioning	155.2 (20.25)	103 - 196

Note. PDD-NOS = Pervasive Developmental Disorder - Not Otherwise Specified; SRS = Social Responsiveness Scale; ADOS = Autism Diagnostic Observation Schedule; SA = Social Affect score; RRB = Repetitive and Restricted Behavior score; ER = empathic responsiveness; EAS = Emotionality Activity Sociability Temperament Survey; BRIEF = Behavior Rating Inventory of Executive Function.

The verbal cues were accompanied by a corresponding facial expression and tone of voice. The third and last emotion that the interviewer displayed during the interview was neck pain. For ten seconds the interviewer would rub his/her neck, have a distressed facial expression and moan, while not looking at the participant. Afterwards, a verbal prompt followed: 'I had neck pain for a while.'

Parent reports of empathic responsiveness; Empathy vignettes were used to assess parental perceptions of the empathic responsiveness of their child. Each vignette depicted a social situation where a teacher would either show happiness, sadness or pain, similar to the emotions shown by the interviewer. After each vignette parents were instructed to write down their child's anticipated and most probable response, but they were free to report as many responses as they wished. Instructions and the vignettes are shown in Appendix II (p. 167).

Each of the participants' responses to the emotions of an adult interviewer (structured observation) or a teacher in a hypothetical situation (parent report) were coded into five different and mutually exclusive response categories, based on Loveland and Tunali (1991). A response was coded as an empathic response when the participant gave a relevant verbal response including an empathic reference to the other's emotional state (e.g., 'Are you okay?') or solutions to alleviate the other's distress (e.g., 'Can I get something for you?'). The other four response categories included a relevant response (e.g., 'I have neck pain sometimes'), a confirmatory response (e.g., 'O.k. '), attention without a response (e.g., participant looks at interviewer without a response), and a final response category of no responses, irrelevant or inappropriate responses (e.g., 'When do we have a break?'). Two independent coders double coded the responses of 30 participants. Exact agreement between the coders on children's responses during the structured observation ranged between 79% and 90%, with kappa's ranging from .68 to .85 (adequate to good agreement). Exact agreement between the coders on parent reported responses ranged between 97% and 100%, with the three computed kappa's showing a perfect agreement of 1.00.

Joint measure of empathic responsiveness; We decided to aggregate the verbal empathic responses as observed by independent raters and the verbal empathic responses as reported by parents, because the sum of multiple empathy estimations is a more stable and unbiased estimator than any single empathy measurement (Rushton, Brainerd, & Pressley, 1983). Aggregating empathy estimations over multiple informants helps to partial out error variance due to unreliability of each particular measurement (Denham, Wyatt, Bassett, Echeverria, & Knox, 2009). A significantly positive, though modest correlation was found

between the proportion scores of verbal empathic responses in the structured observation and parent report ($r = .28, p = .002, n = 121$), suggesting that both measures indeed relate to the same latent variable, but also add unique variance. A joint measure of empathic responsiveness was created by averaging the proportion scores of empathic responses.

Temperament

Emotionality Activity Sociability Temperament Survey (EAS); The EAS (Buss & Plomin, 1984) is a 20-item parental temperament questionnaire consisting of four subscales: emotionality (e.g., ‘Child gets easily upset’), activity (e.g., ‘Child is always on the go’), sociability (e.g., ‘Child likes to be with people’) and shyness (e.g., ‘Child takes long time to warm up to people’). Each scale consists of five statements that can be answered on a 5-point scale ranging from 1 (not at all descriptive of my child) to 5 (very well descriptive of my child). A higher score on a scale indicates a stronger presence of this temperamental trait. Average Cronbach’s alpha of the EAS scales was .78 in a Dutch elementary school sample, which is acceptable given the low number of items in each scale (Boer & Westenberg, 1994). In the present HFASD sample, Cronbach’s alpha’s of the EAS scales ranged from .59 (sociability) to .81 (emotionality).

Cognitive abilities

Peabody Picture Vocabulary Test-III-NL; The Peabody Picture Vocabulary Test (Dunn & Dunn, 2004) assesses receptive vocabulary and is highly correlated with more general measures of verbal IQ (Hodapp & Gerken, 1999). The participant has to select one of four pictures that corresponds with a given word. The test consists of 17 sets of 14 words which increase in difficulty. Based on the PPVT participants received a verbal IQ-score standardized for age.

Advanced Theory of Mind task. The advanced Theory of Mind task consists of five social stories, each story assessing the understanding of a protagonist’s mental state (intention, belief, emotion). The first story is a second order false belief story (‘birthday puppy story’) derived from Sullivan, Zaitchik, and Tager-Flusberg (1994). The second story addresses understanding of emotional display rules (hiding one’s true emotion in certain social contexts) and comes from Begeer et al. (2011). The three final stories address social rule violation, double bluff, and irony and are derived from Kaland, Callesen, Moller-Nielsen, Mortensen, and Smith (2008). After a story is read out loud to the participant, a question follows about the protagonist’s mental state. Each of the five mental state questions is rewarded one point (correct) or zero points (incorrect or ‘don’t

know') and add up to a total score of 0-5. Interrater reliability of the mental state questions was moderate to very good (20 % of the data was coded double), with kappa's ranging from 0.57 to 1.00.

Behavior Rating Inventory of Executive Function (BRIEF). In the BRIEF (Gioia, Isquith, Kenworthy, & Barton, 2002) parents rate the frequency (1 = never; 2 = sometimes; 3 = often) of 86 behaviors describing their child's executive functioning in everyday life. The BRIEF assesses several domains of executive functioning, including inhibition, cognitive flexibility, emotion regulation, initiative, and behavioral evaluation. A higher score on the BRIEF signifies a higher frequency of the described behavior and a higher degree of executive function problems. Adequate psychometric properties have been reported (Gioia, Isquith, Kenworthy, & Barton, 2002). Cronbach's alpha of the BRIEF in the present sample was .95.

Autism severity

Autism Diagnostic Observation Schedule-Generic (ADOS-G); The ADOS (Lord et al., 2000) is a diagnostic observation measure to assess the presence and severity of ASD-specific impairments in social reciprocity, communication, fantasy, and repetitive interests and behaviors. In a semi-structured fashion the ADOS-interviewer offers playful activities (e.g., reading a story book) and topics of discussion (e.g., peer problems) to assess the socio-communicative abilities of the participant. Each of the participant's behaviors is rated on a 3-point-scale (0 = normal behavior; 1 = slightly deviant behavior; 2 = clearly deviant and autistic behavior). In the present study we used the revised ADOS algorithm to calculate a total ADOS score consisting of a Social Affect score (social reciprocity domain and language and communication domain) and a Repetitive and Restricted Behavior score (Gotham, Risi, Pickles, & Lord, 2007). An ADOS score of 7 or higher is indicative of an ASD. The ADOS has excellent internal consistency, interrater reliability, test-retest reliability, and discriminant validity (Lord et al., 2000; Gotham et al., 2007). In the analyses we employed the ADOS *severity* score as a continuous measure of participants' autism severity (Gotham, Pickles, & Lord, 2009).

Procedure

We received written informed consent from parents and participants (if 12 years or older at time of testing). Each participant was seen during two individual test sessions at school, separated by one week to one month. During one session the ADOS was administered. The other session consisted of a full battery of tests, including the PPVT,

the advanced Theory of Mind test and the structured observation of empathic responsiveness to the interviewer. Interviewers were trained graduate students in psychology, health science or medicine. All interviews were taped and verbatim transcribed. After children participated in the study, parents received a questionnaire about their child's behavior.

Results

First, a correlation table was created to examine the relations between the dependent and independent variables. As would be expected, the three measures of empathic responsiveness (observation, parent report, joint measure) were significantly positively correlated (see Table 5.2). Also, they were all significantly negatively correlated with ADOS severity score, indicating that participants with higher ADOS severity scores were characterized by fewer empathic verbal responses, both during the interaction with an interviewer and according to parent reports. Furthermore, the joint measure of empathic responsiveness was significantly positively associated with the temperamental trait sociability ($r = .30, p < .01$), but negatively with emotionality ($r = -.22, p < .01$) and shyness ($r = -.29, p < .01$). Hence, children with HFASD who were rated by their parents as more sociable, less emotional and less shy tended to show more empathic responses. Also, a near significant positive correlation was found between participants' empathic responsiveness and their Theory of Mind task performance ($r = .15, p = .05$), indicating that participants with a high performance on the ToM task tended to respond more empathically to others' emotions than participants with a low ToM performance. Finally, participants' age, receptive verbal IQ (PPVT), activity (EAS) and executive functioning (BRIEF) were not directly associated with their empathic responsiveness.

Table 5.2 Pearson correlations between the measures of empathic responsiveness (ER) and all independent variables.

	1	2	3	4	5	6	7	8	9	10	11	12
1. Joint measure ER	-											
2. Observed ER	.65***	-										
3. Parent reported ER	.91***	.28**	-									
4. Age	.14	.13	.11	-								
5. Verbal IQ	.05	.04	.04	.05	-							
6. ADOS severity score	-.33***	-.23*	-.30**	-.11	-.13	-						
7. EAS - Emotionality	-.22**	-.10	-.23*	-.33***	-.04	.04	-					
8. EAS - Activity	.13	.12	.10	-.17*	-.07	.01	.20*	-				
9. EAS - Sociability	.30***	.15	.30**	-.11	-.08	-.12	-.01	.32***	-			
10. EAS - Shyness	-.29**	-.20*	-.23*	.09	-.08	.13	.15*	-.08	-.63***	-		
11. Theory of Mind	.15†	.13	.12	.28**	.26**	-.04	-.00	.06	-.03	-.01	-	
12. BRIEF – EF problems	-.01	.11	-.08	-.20*	.12	.00	.47***	.12	-.12	.08	.05	-

Note. ER = Empathic responsiveness; ADOS = Autism Diagnostic Observation Schedule; EAS = Emotionality Activity Sociability Temperament Survey ; BRIEF = Behavior Rating Inventory of Executive Function; EF = Executive Functioning; *** = $p < .001$; ** = $p < .01$; * = $p < .05$; † = $p < .10$.

A hierarchical multiple regression analysis was performed to check for the unique contribution of temperament, Theory of Mind and executive functioning to the prediction of children's empathic responsiveness (see Table 5.3). In the regression model the joint measure of empathic responsiveness was used as a dependent variable. Age and receptive verbal IQ were entered as first predictors (step 1), followed by the four temperamental scales of the EAS (step 2), and the two cognitive factors Theory of Mind and executive functioning in the final step (step 3). Temperament explained a significant amount of variance (15%) in children's empathic responsiveness over and above age and receptive verbal IQ. Emotionality and shyness were negatively associated with children's empathic responsiveness (β emotionality = $-.18$, $p = .05$; β shyness = $-.10$, $p > .10$). Sociability and activity were positively associated with children's empathic responsiveness (β sociability = $.22$, $p < .10$; β activity = $.11$, $p > .10$). Adding Theory of Mind and executive functioning (BRIEF) into the regression model did not add explained variance in empathic responsiveness over and above the variance already explained by age, receptive verbal IQ and temperament.

Since results of the regression analysis may also vary according to the measure that is used for children's empathic responsiveness, we ran the model again for both measures separately (structured observation and parent report; see Table 5.3). None of the individual steps in the regression model added a significant amount of explained variance in verbal empathic responses during the structured observation, although the total model explained a near significant 12% of variance ($p < .10$). Yet, an unexpected positive association was noted between children's daily executive functioning problems as reported by parents (BRIEF) and children's empathic responsiveness during the structured observation. This unexpected finding was therefore followed up by an extra regression analysis to check which aspects of executive functioning (i.e., which scales of the BRIEF) were significantly associated with children's empathic responses to the interviewer. After controlling for age and receptive verbal IQ in the first step, the 'Inhibition' scale of the BRIEF was the only scale that was significantly and positively associated with empathic responses ($\beta = .28$, $p < .05$), indicating that participants with increased inhibition problems showed more empathic responses to the interviewer. Furthermore, there was a trend for a negative association between the BRIEF scale 'Emotion regulation' and empathic responses ($\beta = -.26$, $p < .10$).

Table 5.3 Results of three hierarchical multiple regression analyses with children's empathic responsiveness as dependent variable.

Predictors	Empathic responsiveness					
	Joint measure		Observation		Parent report	
	ΔR^2	β	ΔR^2	β	ΔR^2	β
Step 1	.02		.02		.01	
Age		.15		.12		.10
Verbal IQ		.03		.03		.04
Step 2	.15**		.06		.14**	
Emotionality		-.18 [†]		-.05		-.21*
Activity		.11		.14		.07
Sociability		.22 [†]		.00		.28*
Shyness		-.10		-.19		-.02
Step 3	.03		.04 [†]		.01	
Theory of Mind		.12		.08		.10
EF problems		.15		.21*		.07
Total R ²	.20**		.12 [†]		.17**	

Note. ** = $p < .01$; * = $p < .05$; † = $p < .10$.

When parent reported empathic responses were taken as the dependent variable, the results of the hierarchical multiple regression analysis resembled the results with the joint measure of empathic responsiveness as dependent variable. Children's temperament explained 14% of variance in empathic responses ($p < .01$), which was mainly driven by a negative association between emotionality and empathy (β emotionality = $-.21$, $p < .05$) and a positive association between sociability and empathy (β sociability = $.28$, $p < .05$). Adding Theory of Mind task performance and executive functioning problems in the third step of the model did not add a significant amount of explained variance in parent reported empathic responses.

Because ADOS severity scores were significantly negatively correlated with all three measures of empathic responsiveness (Table 5.2), we checked whether ADOS severity score would also affect the *associations* between children's empathic responsiveness and the three main predictors. In the regression model the joint measure of empathic responsiveness was used as a dependent variable. Children's age and receptive verbal IQ were entered in the first step of the model, followed by temperamental scales in the

second step, and ADOS severity score in the third step. In the final step the interaction term between the ADOS severity score and a score on a temperamental scale was entered into the model. Results showed an 8% increase in explained variance of empathic responsiveness after adding ADOS severity score into the regression model ($\beta = -.28$, $\Delta R^2 = .08$, $p = .001$), confirming that children with more severe autistic traits tended to show fewer empathic responses than children with relatively mild autistic traits as indicated by their ADOS scores. Interaction terms of ADOS severity score and each of the temperamental scales did not add a significant amount of explained variance in empathic responsiveness. However, a trend was observed when adding the interaction term between ADOS severity score and sociability ($\beta = .15$, $\Delta R^2 = .02$, $p < .10$), suggesting that the positive association between a child's sociability and the degree of empathic responses was slightly stronger among children with higher ADOS severity scores. Still, the associations between temperament and empathic responsiveness appeared to be largely independent of participants' autism severity.

When entering Theory of Mind and executive functioning problems in the second step of the model, followed by ADOS severity score (third step), and interaction terms between ADOS severity score and the cognitive factors (final step), ADOS severity score again predicted a higher empathic responsiveness ($\beta = -.32$, $\Delta R^2 = .10$, $p < .001$), but the interaction terms of ADOS severity score and the cognitive factors did not add explained variance. Thus, the lack of association of Theory of Mind and executive functioning with the joint measure of empathic responsiveness was independent of participants' autism severity.

Discussion

In the present study we examined the role of temperament, Theory of Mind and executive functioning in the empathic responsiveness to others' emotions of school-aged children and adolescents with high-functioning autism spectrum disorder (HFASD). Individual differences in temperament explained 15% of the variance in empathic responsiveness, based on behavioral observations and parent reports. However, counter to our expectations, neither Theory of Mind nor executive functioning were significantly associated with children's empathic responsiveness. A wide range of autism severity (ADOS severity score) was further noted among our participants with HFASD, but autism severity did not affect the (lack of) associations between participants' empathic responsiveness and their temperament, Theory of Mind, and executive functioning.

Even though temperament has a deep impact on children's social development and empathic responsiveness (Eisenberg et al., 1998; Sanson et al., 2004), individual differences in temperament had thus far hardly been examined among children with HFASD. Based on the previously found associations in typical development, we hypothesized that children's empathic responsiveness would be negatively related with emotionality, but positively with sociability. Indeed, children with HFASD who easily experienced strong negative emotions according to their parents (high emotionality) or showed a preference to be alone rather than to be with others (low sociability) tended to respond less empathically to the emotions of others. Associations between children's empathic responsiveness and their level of shyness and activity were less robust, but in the anticipated direction. Shyness was negatively related with children's empathy, whereas activity was positively related. Further analyses showed that temperamental traits were strongly associated with parent reported empathic responses, but did not significantly affect children's empathic responses to the interviewer. It is plausible that parents made a prediction about their child's empathic responses based on their full range of experiences with the child's everyday responsiveness to others' emotions, thereby creating a relatively stable and predictable measure of empathy. On the other hand, participants' empathic responses during the interview may have been more subject to mood and context effects, thus creating a large amount of unexplained variance.

Overall, our findings suggest that the role of temperament in children's empathic responsiveness extends to children with HFASD. The link between children's temperament and empathy may have evolved, because temperament affects the way children attend to and interpret the social environment (Caspi, 1998 in Fox & Henderson, 1999). For instance, children high in emotionality may experience personal distress while watching an adult in distress, thereby making an empathic response less likely (Decety & Meyer, 2008; Eisenberg et al., 1998). A specific temperament may also evoke specific responses from the environment or may generate a selective bias towards a particular social environment (Caspi, 1998 in Fox & Henderson, 1999). In turn, this social environment may stimulate or hinder a child's empathic responsiveness.

We reasoned that impairments in Theory of Mind understanding in ASD (e.g., Baron-Cohen, 1989; Blair, 2005; Jones, Happé, Gilbert, Burnett, & Viding, 2010) might explain limitations in empathic responsiveness observed in some children with ASD (Rogers et al., 2007). Indeed, a positive association was found between children's performance on an advanced Theory of Mind (ToM) task and their empathic responsiveness. However, after controlling for age and receptive verbal IQ, ToM did not

add unique variance to children's empathic responsiveness, whether measured with a structured observation, parent report, or both. Thus, the positive link between ToM and empathy could be partially mediated by children's age and receptive verbal IQ. With increased age and verbal IQ children with HFASD will do better on a Theory of Mind task (Happé, 1995) and may at the same time respond more empathically to others' emotions. However, in the regression model age and receptive verbal IQ both did not add explained variance in children's empathic responsiveness. The association between ToM and empathic responsiveness may be modest, because ToM task performances of children and adolescents with HFASD may depend predominantly on socio-cognitive aspects of ToM (Hughes & Leekam, 2004) or non-social heuristic strategies (Peterson et al., 2009), whereas their empathic responsiveness may rely more on socio-perceptual aspects of ToM. That is, the ability to understand emotions and intentions in a story character probably is a different ability than to recognize and adequately respond to emotions and intentions of others in real life.

Counter to our expectation, individual differences in everyday executive functioning (EF) were unrelated to children's empathic responsiveness. A separate analysis on empathic responses as described by parents confirmed that EF problems in everyday life did not predict children's empathic responsiveness in hypothetical situations. However, EF problems were unexpectedly *positively* associated with children's empathic responses during the structured observation. Hence, children scoring high on EF problems were more likely to give a verbal empathic response to the interviewer than children scoring relatively low on EF problems. Inhibition problems were primarily responsible for the association. Children with inhibition problems have the tendency to respond impulsively and may therefore also respond more readily to the emotions of an unfamiliar adult.

Consistent with the diagnostic criteria for ASD (APA, 2000) and current movements towards a more dimensional approach of ASD (APA, 2011), our study demonstrated a negative association between children's autism severity and their empathic responsiveness. Hence, children and adolescents with more severe autistic traits as indicated by their ADOS severity score (Gotham et al., 2009) were less inclined to give an empathic response, both during a structured observation and according to parents, than children and adolescents with relatively mild autistic traits. Importantly, autism severity did not have a significant effect on the (lack of) associations between children's empathic responsiveness and their temperament, Theory of Mind, and executive functioning.

The diagnostic process of ASD is challenging for clinicians due to the large individual differences found within the autism spectrum, including differences in empathic responsiveness. The identification of factors that contribute to individual differences in children's empathic responsiveness will provide a better understanding of what level of empathy can be expected from children with and without ASD considering their developmental age, intelligence, and temperamental features. Clinicians should be aware that findings on children's empathic responsiveness may also vary across clinical observations and parent reports. The currently used semi-structured interaction with an unknown adult resembles a diagnostic assessment situation with a psychiatrist. Under these circumstances, children with HFASD who lack inhibitory control may also be the ones who respond instantly and adequately to the adult's emotions. However, when relying on parent information, children's temperament appears to be a significant predictor of empathy. Hence, clinicians should be aware that a child's (lack of) empathic responsiveness is not a plain indicator of a child's clinical status, but should be seen as the outcome of multiple factors including unique characteristics of the child and the context in which the empathic responses were measured.

This study has several limitations. First, our measure of empathic responsiveness was primarily based on verbal expressions. Although a verbal empathic response to someone's distress evinces an adequate understanding of the social emotional situation and competence to act accordingly, it remains a topic of debate whether a verbal expression of empathy also reflects an *experience* of empathy. For instance, children's empathic comments may also follow from social rule learning rather than the experience of true empathy. On a related note, given the multitude of empathy definitions (Decety & Meyer, 2008), it may be difficult for researchers to decide on a measure that reflects 'true empathy'. Furthermore, it should be noted that in the structured observation the degree of verbal empathic responses was relatively low. Possibly the older age and authority of the interviewer might have inhibited some children to respond empathically to the interviewer's emotions. Future studies on real life empathy should therefore ideally include measures of children's empathic responsiveness to peers.

Previous studies on children's empathic responsiveness indicated reduced empathic responses in children with ASD compared to matched peers (Bacon et al., 1998; Scambler et al., 2007; Sigman et al., 1992). Yet, these group-level comparisons ignored the large heterogeneity within the autism spectrum (Jones & Klin, 2009; Lord & Jones, 2012). Findings from the present study show that the empathic responsiveness of children and adolescents with HFASD is not merely defined by their ASD diagnosis. Similar to

previous findings in typical development, variance in empathic responsiveness has been meaningfully linked with individual child characteristics. Individual differences in temperament determine for a significant part the empathic responsiveness of children with HFASD.

Chapter 6

Social interaction style of children and adolescents with high-functioning autism spectrum disorder

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Abstract

Qualitative differences in social interaction style exist *within* the autism spectrum. In this study we examined whether these differences are associated with (1) the severity of autistic symptoms and comorbid disruptive behavior problems, (2) the child's psycho-social health, and (3) executive functioning and perspective taking skills. The social interaction style of 156 children and adolescents (6-19 years) with high-functioning autism spectrum disorder (HFASD) was determined with the Wing Subgroups Questionnaire. An active-but-odd social interaction style was positively associated with symptoms of autism, attention deficit and hyperactivity. Furthermore, an active-but-odd social interaction style was negatively associated with children's psycho-social health and positively with executive functioning problems. Social interaction style explains part of the heterogeneity among children with HFASD.

Introduction

All individuals with a diagnosis of an autism spectrum disorder (ASD) show qualitative impairments in social interaction, as stated in the DSM-IV (APA, 2000). Yet, the social interaction impairments of children with ASD can take many different forms (e.g., Jones & Klin, 2009; Mundy, Henderson, Inge, & Coman, 2007). Diversity in social interaction style likely yields diversity in intervention needs and responsiveness (Beglinger & Smith, 2005). In the current study we therefore focus on individual differences in social interaction style in ASD and associated factors.

The child with autism was first described by child psychiatrist Leo Kanner as a withdrawn child who does not seek interaction with others (Kanner, 1943). Indeed, empirical studies on peer interaction of children with ASD have repeatedly shown that children with ASD show less social play, fewer social interactions, and lack reciprocal friendships compared to typically developing children (Bauminger, Shulman, Agam, 2003; Hauck, Fein, Waterhouse, & Feinstein, 1995; Kasari, Locke, Gulsrud, & Rotheram-Fuller, 2011; Macintosh & Dissanayake, 2006; Sigman & Ruskin, 1999). However, considerable individual differences have also been documented between children with ASD in the quality and quantity of interaction with peers (Kasari et al., 2011; Sigman & Ruskin, 1999).

Wing and Gould (1979) first differentiated individuals with ASD based on their social interaction style. They systematically described three different social subtypes of autism. First, the aloof child seeks no social interactions, nor does the child respond socially to the approaches of others. The passive child does not initiate social interaction, but responds appropriately to the social initiatives of others. Finally, the active-but-odd child actively seeks interactions with others, albeit in an unusual way (e.g., holding a

monologue about a particular interest, or standing too close to a conversation partner). The Wing's social subtype of a child with ASD can be reliably ascertained by observations (Roeyers, 1997) or a parental questionnaire (Wing Subgroups Questionnaire; Castelloe & Dawson, 1993).

The different social interaction styles may be associated with different degrees of autism severity. To date, research with primarily children with ASD and an intellectual disability has shown that active-but-odd children tend to have a higher intelligence, better adaptive behaviors, and lower autism severity scores compared to aloof children (as measured by the Childhood Autism Rating Scale or the Autism Behavior Checklist), and they are more often diagnosed with PDD-NOS or Asperger's Syndrome instead of autism (Althaus, Minderaa, & Dienske, 1994; Castelloe & Dawson, 1993; Ghaziuddin, 2008; O'Brien, 1996; Roeyers, 1997; Waterhouse et al., 1996). However, medical records also suggest that active-but-odd children have a higher rate of comorbidity, defined by deficits in attention, motor control, and perception, than passive and aloof children (Bonde, 2000). Overall, the passive subtype appears to hold an intermediate position between the aloof and active-but-odd group. For instance, passive children are generally reported to be more intelligent than aloof children, but less intelligent than the active-but-odd group (Borden & Ollendick, 1994). Yet, a limitation of the aforementioned studies is that none made a distinction between low-functioning (IQ < 70) and high-functioning (IQ > 70) children with ASD.

Intelligence could be a major confounding factor when examining the associated characteristics of the social interaction styles. Research has already shown that children with high-functioning ASD (HFASD) are generally more active in initiating of and responding to social interactions and show more developmental progress in social interaction skills than children with ASD and an intellectual disability (Bauminger et al., 2003; Eagle, Romanczyk, & Lenzenweger, 2010). Furthermore, aloofness could be confounded by an intellectual disability given the overlap in characteristics (e.g. inability to use speech effectively). Therefore, research within a sample of children with low-functioning ASD does not lead to conclusive results about the associated characteristics of social interaction styles. Research on the social interaction styles of children with HFASD would provide a better understanding of these issues.

In the present study, differences in social interaction styles are examined in a large sample of children and adolescents with HFASD. In a clinical setting, the differentiation of individuals is still strongly based on a categorical system (presence or absence of the disorder). However, we believe a dimensional approach may refine our perspective on the

heterogeneity within the autism spectrum (Pellicano, 2010; Volkmar, State, & Klin, 2009), which is in line with proposals for the upcoming DSM-V (APA, 2011). Therefore, rather than forming social subtype categories to distinguish and compare individuals with ASD, we use a continuous measure of each social interaction style. Furthermore, to be able to understand the unique contribution of each social interaction style, the influence of age, gender, verbal IQ, and all other social interaction styles are statistically controlled for.

Different social interaction styles may be linked with different needs for and responsiveness to interventions (Beglinger & Smith, 2005). Therefore, in the current study we first explore whether the degree to which a child with HFASD shows each social interaction style is associated with his/her needs for intervention, by examining (1) the severity of the child's psychopathology in terms of autistic symptoms and comorbid disruptive behavior problems and (2) the child's psycho-social health. Secondly, we want to shed light on possible cognitive underpinnings of the social interaction styles to encourage customized intervention methods and enhance intervention responsiveness. More specifically, associations are examined between social interaction styles on the one hand and executive functioning and perspective taking skills (Theory of Mind) on the other hand. Information about the child's competence and behavior was obtained in a multi-method (observation, test performance, questionnaires) and multi-informant design (children, parents and teachers).

Method

Participants

Participants were 214 (183 boys; 31 girls) Dutch children and adolescents with HFASD. Participants were recruited via a specialized school for normally intelligent children and adolescents with an ASD diagnosis. The diagnostic classification of ASD was given by a psychiatrist according to established DSM-IV-TR criteria and based on examination by multiple experienced clinicians (psychologists, psychiatrists and educationalists). The diagnostic process included anamneses, heteroanamneses, and psychiatric, neuropsychological and logopedic examinations.

The following inclusion criteria were used for the data analyses: (1) the child has a verbal IQ of 70 or higher, as shown by performance on the Dutch version of the Peabody Picture Vocabulary Test-III (Dunn & Dunn, 2004), and (2) parents completed the Wing Subgroups Questionnaire (WSQ). Consequently, 156 of the original 214 participants (73%) were included in the analyses. The final sample consisted of 134 boys and 22 girls with a clinical diagnosis of autism ($n = 29$), Asperger's Syndrome ($n = 22$), or PDD-NOS

($n = 105$). Mean age of the final sample was 13.4 years ($SD = 3.0$; range = 6.4-18.9) and mean receptive verbal IQ was 105 ($SD = 12.8$; range = 72-132). Children of the final sample were significantly younger ($p < .01$) than children whose parents did not complete the WSQ, but no differences were observed in verbal IQ, gender ratio or clinical diagnosis.

All participants were assessed with the Autism Diagnostic Observation Schedule (ADOS; Lord et al., 2000). Despite the extensive diagnostic procedures, only thirty seven percent of the participants ($n = 57$) received a total score on the ADOS at or above the cutoff point for ASD (≥ 7). Earlier studies have already shown a relatively poor sensitivity of the ADOS (ranging from .49 - .80) in classifying individuals with PDD-NOS (Bastiaansen et al., 2011; Gotham et al., 2008). Therefore, all statistical analyses were repeated to check whether results differed between individuals scoring below or at/above the ADOS cutoff point.

Measures

Below are the measures described for social interaction style, severity of psychopathology, psycho-social health, and cognitive factors respectively. Internal consistencies for the different measures in the study sample are indicated in the final column in Table 6.1.

Social interaction style

Wing Subgroups Questionnaire (WSQ). The WSQ (Castelloe & Dawson, 1993) is a parent or teacher questionnaire to determine the Wing social subtype of a child with ASD. For the present study the WSQ was translated into Dutch with a forward-backward-translation method. The WSQ contains 13 descriptions of each of the three Wing subtypes (active-but-odd, passive, aloof) and 13 descriptions of typical socio-communicative behaviors. Parents or teachers evaluate how well each statement describes the child's behavior in everyday activities on a 7-point Likert scale ranging from 0 (never) to 6 (always). All item-scores belonging to one subtype add up to a scale-score. Traditionally, a child is assigned to that particular social subtype with the highest scale-score. However, instead of assigning participants to a specific category, in the present study all continuous scale-scores are included in the analyses. Internal consistency of the four WSQ scales was moderate to good in previous samples with ASD and an intellectual disability (Castelloe & Dawson, 1993; O'Brien, 1996). Pearson correlations between the different WSQ scales were: .28 (active-but-odd - passive), .25 (active-but-odd - aloof), -.46 (active-but-odd - typical), .58 (passive - aloof), -.37 (passive - typical), and -.42 (aloof - typical).

Psychopathology

Autism Diagnostic Observation Schedule-Generic (ADOS-G). The ADOS (Lord et al., 2000) is a semi-structured diagnostic observation measure to assess the presence and severity of the main problem areas in autism: social reciprocity, communication, fantasy, and repetitive interests and behaviors. The ADOS-interviewer offers several playful activities (e.g. reading a story book) and topics of discussion (e.g. peer problems) to assess the socio-communicative abilities of the participant. The ADOS has excellent internal consistency, interrater reliability, test-retest reliability, and discriminant validity (Lord et al., 2000).

Social Responsiveness Scale (SRS). The SRS (Constantino & Gruber, 2007) is a parent- or teacher questionnaire which assesses autistic traits. The SRS consists of five scales: social awareness, social cognition, social communication, social motivation, and autistic mannerisms. Each of the 65 statements about the child's behavior can be answered on a 4-point scale ranging from 0 (never true) to 3 (almost always true). A higher total score indicates more autistic traits. Good reliability and validity have been reported (Constantino & Gruber, 2007).

Disruptive Behavior Disorders rating scale (DBD). The DBD (Pelham, Gnagy, Greenslade, & Milich, 1992) is a parent or teacher questionnaire developed to assess externalizing problem behaviors in children. It consists of symptom descriptions of four disorders: ADHD Inattentive subtype, ADHD Hyperactive/Impulsive subtype, Oppositional Defiant Disorder, and Conduct Disorder. Each statement has to be rated on how well it describes the child's behavior on a 4-point scale ranging from 0 (not at all) to 3 (very well). A higher score indicates more symptoms of externalizing problem behaviors. Adequate psychometric properties of the DBD have been reported (Pelham et al., 1992). Pearson correlations between parent and teacher scores on the DBD in this study were .49 for the inattention scale, .47 for the hyperactivity scale, .53 for the ODD scale, and .13 for the CD scale, which compare favorably to expectable correlations between parent and teacher ratings (cf. Achenbach, McConaughy, & Howell, 1987).

Psycho-social health

Pediatric Quality of Life Inventory (PedsQL). The PedsQL (Varni, Seid, & Kurtin, 2001) is a 23-item questionnaire about the quality of life of children and can be filled in by parents and children. The PedsQL assesses the occurrence of problems in the past four weeks in several domains of functioning: physical, social, emotional, and school-

functioning. Each item can be answered on a 5-point scale ranging from 100 (never) to 0 (almost always). Good reliability and validity have been reported (Varni et al., 2001).

Cognitive underpinnings

Behavior Rating Inventory of Executive Function (BRIEF). The BRIEF (Gioia, Isquith, Kenworthy, & Barton, 2002) is an 86-item parent questionnaire about children's executive functioning in daily life. The BRIEF assesses several domains: inhibition, cognitive flexibility, emotion regulation, initiative, working memory, planning, orderliness, and behavioral evaluation. Each item is coded 1 (never), 2 (sometimes), or 3 (often). A higher score indicates more executive functioning problems in daily life. Adequate psychometric properties have been reported (Gioia et al., 2002).

Theory of Mind task. The Theory of Mind task used in the present study consists of five social stories, derived from Sullivan, Zaitchik, and Tager-Flusberg (1994), Begeer et al. (2011), and Kaland, Callesen, Moller-Nielsen, Mortensen, and Smith (2008). Each story is read out loud to the participant and is followed by a question about the mental state of one of the story characters. The stories assess understanding of second order false belief, emotional display rules, violation of social rules, double bluff, and irony. Each of five mental state questions is rewarded one point (correct) or zero points (incorrect or 'don't know') and add up to a total score of 0-5. One of the social stories is about a man, Johan, who makes a faux pas while talking to an old lady. An example of a mental state question in this story would be: 'How do you think Mrs. Smit is feeling when she hears what Johan tells her?' Interrater reliability of the mental state questions was moderate to very good (20 % of the data was coded double), with kappa's ranging from 0.57 (story 4) to 1.00 (story 1).

Interpersonal Reactivity Index (IRI). Two subscales of the IRI (Davis, 1983), Perspective Taking and Fantasy, assess the tendency of an individual to adopt the perspectives of others in real life, books or movies. The IRI is a self-report questionnaire with adequate psychometric properties (Davis, 1983). For this study an adapted child version of the IRI was used, consisting of 24 instead of 28 items. The child has to evaluate how well each statement describes him/her on a 5-point scale ranging from 0 (not at all) to 4 (very well). An example of a statement is: 'When I'm angry at someone, I also try to imagine how he/she is feeling.' A higher score indicates more perspective taking.

Procedure

We received parental informed consent for participation as well as children's consent when the child was 12 years or older at the time of testing. Each participant went to two individual test sessions at school, separated by one week to one month. During one session the ADOS was presented. The other session involved a complete battery of tests, including the Theory of Mind task and two self-report questionnaires (PedsQL and IRI). After the test sessions parents and teachers received questionnaires about the participant's behavior.

Statistical analysis

Because age, gender, and verbal IQ were found to correlate significantly with one or more WSQ scales and/or total scores on the main outcome measures, it was decided to statistically control for the confounding influence of age, verbal IQ and gender. For instance, age correlated negatively with the active-but-odd WSQ scale ($r = .23, p < .01$), but positively with the passive WSQ scale ($r = .24, p < .01$). To test the extent to which each of the WSQ scales was uniquely related to the child characteristics, a series of multiple regression analyses was conducted with each WSQ scale as independent variable, and measures of autism severity, disruptive behavior problems, psycho-social health, executive functioning, and perspective taking as dependent variables, controlling for age, gender, and verbal IQ, and for all other WSQ scales. Age, verbal IQ and gender were entered in the first step of the model, all three non-targeted scales of the WSQ in the second step, and the fourth scale of the WSQ (the scale of interest) in the final step (for descriptive statistics of the WSQ scales and outcome measures see Table 6.1). The analyses were repeated, with each WSQ scale as final predictor in the model, to examine the unique contribution of each social interaction style to the outcome measures above and beyond the predictive power of the other social interaction styles. The results of the multiple regression analyses are shown in Table 6.2. All analyses with significant outcomes were repeated while controlling for possible group differences between individuals scoring below and at/above the ADOS cutoff point for ASD (score ≥ 7).

Table 6.1 Descriptive statistics for the predictors (WSQ scales) and main outcome measures.

	<i>M (SD)</i>	<i>Range</i>	<i>Cr. a</i>
Social interaction styles			
WSQ – active-but-odd (P)	33.0 (10.1)	2 – 56	.84
WSQ – passive (P)	37.6 (12.6)	6 – 71	.73
WSQ – aloof (P)	21.0 (9.3)	2 – 53	.69
WSQ – typical (P)	35.3 (11.7)	4 – 63	.86
Autistic symptoms			
ADOS Module 3 (C)	6.3 (4.4)	0 – 19	.82
ADOS Module 4 (C)	5.6 (3.9)	0 – 16	.88
SRS Total (P)	80.6 (22.4)	23 – 133	.93
Comorbid disruptive behavioral problems			
DBD Attention deficit (P)	11.4 (5.1)	0 – 25	.82
DBD Hyperactivity (P)	9.2 (5.2)	0 – 24	.84
DBD Attention deficit (T)	8.9 (5.5)	0 – 24	.85
DBD Hyperactivity (T)	7.2 (5.6)	0 – 23	.87
Psycho-social health			
PedsQL Total (C)	75.6 (12.1)	34 – 99	.84
PedsQL Total (P)	64.7 (12.1)	22 – 97	.84
Executive functioning			
BRIEF Total (P)	155.0 (20.1)	103 – 196	.95
Perspective taking			
Theory of Mind task (C)	3.5 (1.2)	0 – 5	.46
IRI Fantasy (C)	12.9 (5.4)	0 – 23	.72
IRI Perspective taking (C)	11.8 (4.8)	0 – 22	.77

Note. (C) = Child informant; (P) = Parent informant; (T) = Teacher informant; ADOS = Autism Diagnostic Observation Schedule; SRS = Social Responsiveness Scale; DBD = Disruptive Behavior Disorders rating scale; PedsQL = Pediatric Quality of Life Inventory; BRIEF = Behavior Rating Inventory of Executive Function; IRI = Interpersonal Reactivity Index.

Results

Psychopathology

The active-but-odd scale of the WSQ accounted for a small, but significant amount of variance on the ADOS above and beyond the explained variance by age, verbal IQ, gender, and the three other WSQ scales ($\beta = -.18$, $\Delta R^2 = .02$, $p = .05$). The active-but-odd scale also explained a significant amount of variance on the SRS above and beyond all other variables ($\beta = .35$, $\Delta R^2 = .09$, $p < .001$; all SRS subscales with the exception of Social motivation: $\beta > .28$, $\Delta R^2 > .05$, $p < .001$). Analyses with the passive WSQ scale as final predictor in the regression model failed to show any meaningful associations with the psychopathology outcome measures, with the exception of a small positive association with the Social Motivation subscale of the SRS ($\beta = .18$, $\Delta R^2 = .02$, $p < .05$). The aloof scale of the WSQ also contributed modestly, yet significantly to variance in total score of the SRS ($\beta = .21$, $\Delta R^2 = .03$, $p < .001$). The typical scale of the WSQ, which indicates the degree of normal social interactions, was negatively associated with total scores on the ADOS ($\beta = -.28$, $\Delta R^2 = .05$, $p < .01$) and the SRS ($\beta = -.46$, $\Delta R^2 = .14$, $p < .001$).

With regard to disruptive behavior problems, the active-but-odd scale was most strongly and positively associated with symptoms of hyperactivity on the DBD (parent report: $\beta = .58$, $\Delta R^2 = .24$, $p < .001$; teacher report: $\beta = .32$, $\Delta R^2 = .07$, $p < .01$). The other WSQ scales did not contribute to variance in disruptive behavior problems.

Psycho-social health

Variance on the active-but-odd scale did not account for any significant variance on self-reported quality of life (PedsQL). Yet, when children's quality of life as reported by parents was taken as dependent variable in the regression analysis, the active-but-odd scale showed a significant negative association with quality of life ($\beta = -.34$, $\Delta R^2 = .08$, $p < .001$). All other WSQ scales did not contribute to variance in quality of life reports.

Cognitive underpinnings

Firstly, the active-but-odd scale explained a significant amount of all variance on the total score of the BRIEF ($\beta = .51$, $\Delta R^2 = .19$, $p < .001$), particularly the Inhibition scale ($\beta = .61$, $\Delta R^2 = .27$, $p < .001$). This indicates that a higher degree of an active-but-odd social interaction style is associated with a higher degree of executive dysfunctioning. The passive scale only had a negative association with the Orderliness subscale of the BRIEF ($\beta = -.26$, $\Delta R^2 = .04$, $p < .01$). Furthermore, the aloof scale also had a modest positive association with the BRIEF ($\beta = .20$, $\Delta R^2 = .03$, $p < .05$), particularly the BRIEF-subscale

cognitive flexibility ($\beta = .30$, $\Delta R^2 = .06$, $p < .01$). Finally, a negative association was noted between the typical scale and the BRIEF-subscale Initiative ($\beta = -.28$, $\Delta R^2 = .05$, $p < .01$). All other associations were found not significant. Variance on any of the WSQ scales did not account for significant variance on the Theory of Mind task nor self-reported perspective taking (IRI).

Control analyses

Additional analyses were performed to check for a possible interaction effect between the active-but-odd predictor and ADOS status (i.e. score below or at/above the cutoff point for ASD) on the outcome measures. No significant interactions were found between the active-but-odd scale and ADOS status on the outcome measures with the exception of Theory of Mind task performance ($\beta = .16$, $\Delta R^2 = .02$, $p = .05$). This signifies that only for the group at/above the ADOS cutoff point the active-but-odd scale is modestly and positively associated with Theory of Mind task performance.

Finally, to rule out the possibility that associations between WSQ scales and several outcome measures (SRS, DBD hyperactivity scale, BRIEF) were due to an overlap in item-content, the analyses were repeated exclusive of overlapping items. Positive associations between the active-but-odd scale and the outcome measures all remained significant. Associations between other WSQ scales and outcome measures remained stable, except for the association between the aloof scale and total score on the BRIEF, which became non-significant.

Table 6.2 Outcome of a series of multiple linear regression analyses with the unique contribution of each scale on the Wing Subgroups Questionnaire while controlling for age, verbal IQ, gender and the three other WSQ scales.

<i>Dependent variable</i>	<i>Predictor</i>								
	Active-but-odd scale		Passive scale		Aloof scale		Typical scale		Total R ²
	β	R ² change	β	R ² change	β	R ² change	β	R ² change	
Autistic symptoms									
ADOS Total (C)	-.18	.02 *	-.04	.00	.11	.01	-.28	.05 **	.18 ***
SRS Social awareness (P)	.29	.06 ***	-.14	.01	.10	.01	-.45	.14 ***	.48 ***
SRS Social cognition (P)	.29	.06 ***	.15	.01	.17	.02 *	-.15	.02	.41 ***
SRS Social communication (P)	.37	.09 ***	.04	.00	.15	.01 *	-.46	.15 ***	.63 ***
SRS Social motivation (P)	-.01	.00	.18	.02 *	.26	.04 **	-.46	.14 ***	.51 ***
SRS Autistic Mannerisms (P)	.46	.15 ***	-.07	.00	.22	.03 **	-.36	.09 ***	.58 ***
SRS Total (P)	.35	.09 ***	.05	.00	.21	.03 ***	-.46	.14 ***	.70 ***
Comorbid behavioral problems									
DBD Attention deficit (P)	.26	.05 **	-.17	.02	.03	.00	-.12	.01	.11 **
DBD Hyperactivity (P)	.58	.24 ***	-.16	.02	.13	.01	.03	.00	.38 ***
DBD ODD (P)	.21	.03 *	-.08	.00	.13	.01	-.01	.00	.10 *
DBD CD (P)	.08	.01	-.01	.00	.12	.01	-.02	.00	.04
DBD Attention deficit (T)	.13	.02	-.15	.01	.00	.00	.08	.01	.09
DBD Hyperactivity (T)	.32	.07 **	-.17	.02	-.05	.00	.15	.02	.25 ***
DBD ODD (T)	.11	.01	.09	.01	-.06	.00	.03	.00	.06
DBD CD (T)	-.17	.02	-.09	.00	-.07	.00	.03	.00	.07
Psycho-social health									
PedsQL Social scale (C)	-.13	.01	.04	.00	-.09	.01	.12	.01	.07
PedsQL Emotional scale (C)	-.09	.01	-.05	.00	.12	.01	-.03	.00	.08

<i>Dependent variable</i>	Active-but-odd scale		Passive scale		Aloof scale		Typical scale		
	β	R ² change	β	R ² change	β	R ² change	β	R ² change	R ² change
PedsQL Total (C)	-.07	.00	.06	.00	-.07	.00	.08	.00	.05
PedsQL Social (P)	-.45	.14 ***	.02	.00	.03	.00	-.01	.00	.19 ***
PedsQL Emotional (P)	-.25	.05 **	-.06	.00	-.08	.00	.00	.00	.19 ***
PedsQL Total (P)	-.34	.08 ***	.06	.00	-.16	.02	.11	.01	.28 ***
Executive functioning									
BRIEF inhibition (P)	.61	.27 ***	-.05	.00	.08	.00	.10	.01	.39 ***
BRIEF cognitive flexibility (P)	.19	.03 *	.14	.01	.30	.06 **	-.06	.00	.30 ***
BRIEF emotion regulation (P)	.19	.03 *	.02	.00	.17	.02	-.08	.00	.23 ***
BRIEF initiative (P)	.12	.01	.16	.02	-.03	.00	-.28	.05 **	.18 ***
BRIEF working memory (P)	.37	.10 ***	-.05	.00	.21	.03 *	.11	.01	.17 **
BRIEF planning (P)	.29	.06 **	-.12	.01	.12	.01	.09	.01	.13 **
BRIEF orderliness (P)	.47	.16 ***	-.26	.04 **	.20	.03 *	.10	.01	.26 ***
BRIEF behavior evaluation (P)	.50	.18 ***	-.10	.01	-.02	.00	.00	.00	.24 ***
BRIEF Total (P)	.51	.19 ***	-.07	.00	.20	.03 *	.01	.00	.32 ***
Perspective taking									
Theory of Mind task (C)	.13	.01	-.14	.01	-.01	.00	.06	.00	.19 ***
IRI Fantasy (C)	.11	.01	-.14	.01	.03	.00	.06	.00	.07
IRI Perspective taking (C)	.07	.00	-.16	.02	-.02	.00	.09	.01	.11 *

Note. *** = $p < .001$; ** = $p < .01$; * = $p < .05$; Beta's are standardized beta's for the full model, i.e. the value of the beta when all predictors were included. (C) = Child informant; (P) = Parent informant; (T) = Teacher informant; ADOS = Autism Diagnostic Observation Schedule; SRS = Social Responsiveness Scale; DBD = Disruptive Behavior Disorders rating scale; ODD = Oppositional Defiant Disorder; CD = Conduct Disorder; PedsQL = Pediatric Quality of Life Inventory; BRIEF = Behavior Rating Inventory of Executive Function; IRI = Interpersonal Reactivity Index.

Discussion

The present study examined to what extent the social interaction styles of children with HFASD are associated with their level of autistic symptoms, disruptive behavior problems and psychosocial health. A second focus of the study was to explore the relations of social interaction styles with executive functioning and perspective taking skills. Results showed that both an active-but-odd social interaction style as well as an aloof social interaction style were positively associated with ASD symptoms on the SRS. Yet, a modest negative association was found between active-but-odd social interaction style and ASD symptoms as measured by the ADOS. As would be expected, the level of typical social interaction style was negatively related to both measures of autism severity. Furthermore, an active-but-odd social interaction style was positively associated with characteristics of ADHD, ODD, and socio-emotional problems as reported by parents. Also, the active-but-odd style was strongly related to executive functioning problems, particularly inhibition problems. Additional analyses showed that children's performance on the Theory of Mind task was only related to an active-but-odd social interaction style in the group of individuals with ADOS scores above the cutoff point for an ASD. All associations with a passive social interaction style lacked significance after statistically controlling for age, verbal IQ, gender and other social interaction styles.

Previous research with low-functioning samples of ASD found active-but-odd children to be more intelligent and to have less severe forms of autism than passive and aloof children (Borden & Ollendick, 1994; Castelloe & Dawson, 1993; O'Brien, 1996; Roeyers, 1997). However, in the present sample active-but-odd behavior was both negatively (ADOS) and positively (SRS) associated with autistic symptoms. One must note that the basis for ADOS and SRS ratings is different in several important ways: informant (researcher/clinician vs. parent), relevant time frame (one hour vs. six months), and purpose of the measure (categorical vs. dimensional differentiation). The ADOS intends to differentiate between typical development and autistic development, and is less focused on a differentiation *within* the autism spectrum. Therefore, corresponding to DSM-IV criteria, most item descriptions in the ADOS are globally formulated and would fit a passive child as well as an active-but-odd child. However, the social approaches of a child with an active-but-odd interaction style may not be as readily recognized as socially deviant behavior during a one hour session, which might explain the modest negative association found in this study between ADOS and the active-but-odd style. The SRS is specifically designed to measure the severity of autistic symptoms, implying a sensitivity to mild variations within the autism spectrum. Even after excluding overlapping items between

SRS and WSQ, an active-but-odd social interaction style was positively associated with autistic symptoms on the SRS. This indicates that parents observe more autistic symptoms in children with a high degree of active-but-odd social behavior.

Consistent with earlier reports of more deficits in attention, motor control, and perception in active-but-odd children (Bonde, 2000), an active-but-odd social interaction style was associated with elevated levels of disruptive behaviors such as ADHD-symptoms. The question that is raised by this result is whether the social approaches of active-but-odd children are driven by an overall higher level of activity. Associations of an active-but-odd social interaction style with increased ASD and ADHD-symptoms as reported by parents underline the clinical relevance of social interaction style as a dimension to distinguish children and adolescents with HFASD.

Despite a general increase in autistic and disruptive behaviors, an active-but-odd social interaction style was not related to an increase in self-reported psycho-social problems. In fact, average quality of life scores of all HFASD participants in this study were comparable to previous reports of typically developing peers (Bastiaansen, Koot, Ferdinand, & Verhulst, 2004). Thus, children and adolescents did not experience the psycho-social problems their ASD-diagnosis seems to imply. A lack of self-reported psycho-social concerns in ASD has been supported by previous studies (Foley Nicpon, Doobay, & Assouline, 2010). Parents in this study generally did report more psycho-social problems of their children with HFASD. This discrepancy between children's and parents' reports could have been the result of children comparing themselves to other peers with HFASD (all children in this sample received specialized education) and their parents comparing them to typically developing children. Parents reported that particularly children with an active-but-odd interaction style showed more social and emotional problems. This agrees with the clinical observation by Wing and Gould (1979) that active-but-odd children were sometimes rejected by their peers because of their peculiar behavior.

The observed heterogeneity in social interaction style of children with HFASD may in part be produced by heterogeneity in cognitive underpinnings of autistic symptomatology. Indeed, the three proposed cognitive keystones of ASD - perspective taking difficulties, executive dysfunction, and weak central coherence - are not universally present in all children with ASD (e.g., Pellicano, 2010). Our study extends these findings by showing that the degree of active-but-odd behavior was strongly related to the degree of executive functioning problems in daily life. A difficulty to inhibit impulses and regulate behavior could explain the active-but-odd social behaviors seen in some children with

HFASD. For those individuals scoring at/above the ADOS cutoff point, an active-but-odd social interaction was also positively related to performance on the Theory of Mind task. Plausibly, the increased social interactions of active-but-odd children induces more feedback from the social environment, which in turn increases their opportunities to learn about social rules and stimulates social cognition as reflected in the Theory of Mind task.

The association between different social interaction styles and distinct patterns of strengths and weaknesses may be used as a starting point for interventions to improve social skills (see Schreiber, 2011, for a review). The present study shows that children with HFASD and an active-but-odd social interaction style seem in special need of support and interventions given their autism severity, ADHD-symptoms, poor executive functioning and psycho-social problems as reported by parents. Since these children already actively seek contact with others, interventions that are specifically focused on increasing social motivation seem less appropriate. Furthermore, because perspective taking abilities in this study were either independent of (in the less severely autistic group) or positively related to (in the more severely autistic group) an active-but-odd social interaction style, it seems unlikely that active-but-odd children will benefit more from social cognition interventions than passive or aloof children with HFASD. Interventions for children with an active-but-odd social interaction style may be particularly useful when they focus on executive functioning problems, for instance, self-regulation of behavior and control of impulses. These types of interventions may decrease the number of awkward social missteps of active-but-odd children.

The present study has several limitations. First, associations between social interaction style and outcome measures may in part be produced by overlap in informant (parent). Yet, this critique can be partly refuted, because teacher ratings of hyperactivity were similarly associated with the child's active-but-odd interaction style. Secondly, the results and implications of the present study only apply to children and adolescents with HFASD. Associations will need to be confirmed in ASD-samples with an intellectual disability, while controlling for the confounding influence of intelligence. Finally, it should be noted that more than half of the participants in this study did not meet the ADOS cutoff for having an ASD. Hence, our findings might not fully generalize to children and adolescents with more severe forms of ASD. Earlier studies have already shown a relatively poor sensitivity of the ADOS (ranging from .49 - .80) in classifying individuals with PDD-NOS (Bastiaansen et al., 2011; Gotham et al., 2008). However, it should also be noted that in the current study the distribution of clinical diagnoses (autism, syndrome of Asperger, PDD-NOS) was not significantly different for participants scoring above or

below the ADOS cutoff for an ASD. Possibly, ADOS scores are more influenced by the level of intelligence of a child with ASD rather than its particular clinical diagnosis.

It is striking that the aloof and passive social interaction style lacked significant associations with a majority of the outcome measures. Both an aloof and a passive social interaction style were modestly related to a lower social motivation as shown by higher scores on the social motivation scale of the SRS. The lack of social initiations shown by some children with HFASD may be produced by social anxiety rather than an inability to start social interactions. As yet, aloof and passive social behavior remains multi-interpretable. Different causes may underlie a lack of social initiative, for example a lack of social motivation or a lack of social competence. Thus, the aloof and passive group may still be a rather heterogeneous group, leading to few significant associations with other behavioral measures.

A topic of ongoing debate is whether the current DSM-IV categorical system is a meaningful way to differentiate children within the autism spectrum (APA, 2011; Volkmar et al., 2009). More than thirty years ago, Wing and Gould (1979) proposed social interaction style as a clinically relevant distinction among children with ASD. The results of the current study confirm the clinical relevance of the different social interaction styles of children with ASD. While controlling for the confounding influence of intelligence, this study has provided new insights into the associated characteristics of different social interaction styles in HFASD, and has offered possible suggestions for interventions. Future studies will need to identify the mechanisms behind these findings. For instance, it would be useful to examine whether differences in social interaction styles are driven by differences in social motivation. Another important area of interest is change and continuity in social interaction style. Age was found to correlate negatively with an active-but-odd social interaction style, yet positively with a passive interaction style. To find out whether there is a true developmental shift in social interaction style, it is necessary to study the social interaction styles in a longitudinal design. Besides changes over time, children may also adopt different social interaction styles depending on their social partner. Research already indicates that children with ASD show more social interaction problems with peers than adults (Hauck et al., 1995). Therefore it would be useful in future studies to make a distinction between social partners. A combination of multiple settings, multiple informants, and multiple methods will promote a better understanding of the heterogeneity in social interaction styles among those with autism spectrum disorders.

Chapter 7

General Discussion

Research on empathy deficits in autism spectrum disorder (ASD) has thus far mainly focused on specific impairments in children's understanding of others' emotions and mental states. In this thesis, we examined both cognitive and behavioral components of empathy in a large sample of school-aged children and adolescents at the higher-functioning (i.e., normally intelligent) end of the autism spectrum, and compared them to a typically developing peer group. Furthermore, even though individual differences in social behavior are consistently noted *within* the autism spectrum (e.g., Castelleo & Dawson, 1993; Mundy, Henderson, Inge, & Coman, 2007; Wing & Gould, 1979), they are poorly understood. Therefore, a second aim of this thesis was to explore possible underlying mechanisms and associated characteristics of individual differences in empathic and social behavior of children and adolescents with high-functioning ASD.

Empathy and social behavior

It has been theorized that the characteristic social interaction impairments in individuals with ASD are the result of a key deficit in cognitive empathy, better known as Theory of Mind (Baron-Cohen, Leslie, & Frith, 1985). Theory of Mind is the ability to interpret own and others' behaviors in terms of mental states such as intentions and beliefs. A myriad of studies has shown Theory of Mind impairments in children with ASD, as operationalized by their poor performances on first-order false belief tasks (Boucher, 2012; Yirmiya, Erel, Shaked, & Solomonica-Levi, 1998). However, relatively few studies have focused on more advanced mental state reasoning in older and high-functioning individuals with ASD (HFASD). In the second chapter of this thesis, we showed that a large sample of school-aged children and adolescents with HFASD and a typically developing peer group performed equally well on a collection of advanced Theory of Mind (ToM) tasks. Thus, counter to what the social cognition hypothesis would predict and counter to some earlier findings on advanced ToM in smaller samples of children with HFASD (Brent, Rios, Happé, & Charman, 2004; Sobel, Capps, & Gopnik, 2005; White, Hill, Happé, & Frith, 2009), we found that children and adolescents with HFASD were equally able to identify and predict the mental states of story protagonists. They demonstrated a comparable understanding of complex mental constructs such as sarcasm and double bluff. Yet, despite their unexpected adequate ToM task performance, these children and adolescents undeniably show impairments in their everyday social functioning as evidenced by their clinical diagnoses and parental reports (e.g., Social Responsiveness Scale). This discrepancy between social cognition and behavior suggests that social cognition may *not* be the key deficit underlying the social interaction impairments of

children and adolescents with HFASD. Alternatively, there may be something special about everyday inter-personal communication of emotions and mental states that makes it more difficult for individuals with ASD to understand than the mental states described in ToM tasks. For instance, while listening to a Theory of Mind story, one's attention is explicitly and verbally directed to the mental states of story characters. Yet, during a real life social interaction attention is less explicitly drawn to the other's emotions and mental states and these mental states are in part communicated through non-verbal channels such as eye gaze, facial expression and tone of voice. Therefore, the extent to which children and adolescents with HFASD are able to correctly identify and understand others' mental states may crucially depend on attention processes which in turn may depend on the verbal and explicit nature of emotional and mental cues. For this reason, 'real world' ToM may still be hampered in children and adolescents with HFASD, even though their ToM is relatively intact when assessed with a standard verbal ToM task.

To date, most studies on empathy in ASD have specifically focused on ToM impairments, that is, the cognitive component of empathy. However, the behavioral component of empathy, including the ability to adequately respond to others' emotional states, has received relatively little attention in autism research. In the study described in Chapter 3, we aimed to get more insight into the empathic responsiveness of children and adolescents with HFASD to others' emotions by using both structured observations and parental reports. Parents were asked to describe their child's responses in hypothetical empathy evoking situations such as a teacher spilling hot coffee. In agreement with the diagnostic criteria for ASD (APA, 2000) and previous studies (Hudry & Slaughter, 2009; Johnson, Filliter, & Murphy, 2009), parents of a child with HFASD expected that their child would show fewer empathic responses than parents of a typically developing child. Hence, despite the normal intelligence and wide age range of our participants with ASD, parents indicated that their child with HFASD had a reduced empathic responsiveness. Although parents provide highly relevant and unique information about their child's developmental history and everyday behavior (Ozonoff et al., 2009), parental information should also be interpreted with care (Bennett et al., 2012; Hus, Taylor, & Lord, 2011). The fact that parents have experience with their child's empathic responses across a broad range of everyday social situations, including the child's responses to the parents' own emotional states, may both be an advantage (high ecological validity) and a disadvantage (risk of subjectivity). Therefore, to get a more objective overview of children's empathic behavior, we also included a direct observation of children's responses to the emotional displays of an experimenter.

At three preset occasions during an individual interview, the interviewer expressed an emotional state according to a standardized protocol including both verbal and non-verbal cues (facial expression, tone of voice). Children's behavioral responses to the interviewer's emotional displays were independently coded based on video footage. Earlier observational studies had demonstrated a reduced empathic responsiveness to an experimenter's display of distress in young children with ASD and an intellectual disability (Bacon, Fein, Morris, Waterhouse, & Allen, 1998; Hobson, Harris, García-Pérez, & Hobson, 2009; Loveland & Tunali, 1991; Scambler, Hepburn, Rutherford, Wehner, & Rogers, 2007; Sigman, Kasari, Kwon, & Yirmiya, 1992). However, compared to a typically developing peer group, we found that children and adolescents with a diagnosis of HFASD equally often provided an empathic response, a relevant response, or paid attention to the interviewer after his/her emotional display. Thus, following these objective behavioral observations, the empathic behavior of children and adolescents with HFASD was not different from that of typically developing peers. However, more subtle impairments in children's empathic responsiveness may still exist, as also indicated by parent reports. Possibly, children and adolescents with HFASD show particular problems when it comes to adequately timing an empathic response or expressing empathy non-verbally. It should also be noted that the proportion of empathic responses to the interviewer's emotional states was relatively low in both groups compared to other types of responses (e.g., relevant verbal response), and this proportion was notably lower than in parent reports. Earlier studies on the empathic responsiveness of young children also documented low frequencies of explicit empathic or pro-social responses to others' distress (Bacon et al., 1998; Loveland & Tunali, 1991). Thus, we should consider the possibility that an explicit empathic response to an unfamiliar adult is not a standard response for a child or adolescent, regardless of clinical status. This also has important clinical implications, because diagnostic assessments of ASD commonly include an observation of a child's behavior while interacting with an unfamiliar adult (e.g., a psychiatrist). We will turn to this issue later in the discussion.

As we pointed out in the introduction of this thesis, social cognition may be necessary, but not sufficient to show adequate social behavior (Peterson, Garnett, Kelly, & Attwood, 2009). Previous research has shown that children and adolescents with HFASD may do relatively well on tasks assessing their basic understanding of emotions and social situations (e.g., false belief tasks), yet, they still experience problems during everyday social interactions (e.g., Kasari, Locke, Gulsrud, & Rotheram-Fuller, 2011; Macintosh & Dissanayake, 2006; Peterson et al., 2009). Therefore, in the study discussed in Chapter 4,

we assessed children's abilities to present themselves favorably to an audience, both during an actual interaction with an interviewer ('real life task') and in a hypothetical situation ('hypothetical task'). Overall, participants created a more positive image of themselves in the hypothetical scenario compared to the interaction with the interviewer. Similar to the typically developing comparison group, participants with HFASD talked more positively about themselves when motivated by the prospect of participation in a prize-winning game in the real life task, or to be liked by a classmate in the hypothetical task. However, their positive self-statements were less strategic, because they were less likely to highlight their game-related abilities or emphasize a shared interest in case of the classmate. When participants were asked to justify their way of self-presenting, some adolescents with HFASD explained that they made a trade-off for honesty and 'staying true to themselves' rather than a strategic self-promotion. This result agrees with anecdotal parental comments of the remarkable honesty of their child with HFASD and empirical findings of a reduced likelihood of children with HFASD to praise others' performances for the sake of being nice rather than honest (Chevallier, Molesworth, & Happé, 2012c). Thus, although children and adolescents with HFASD can be motivated to make a good impression on others, their social motivation may still be limited compared to typical development. In HFASD, social behavior may be more rule-based than emotion-driven, that is, they may be less emotionally affected by what others might think or feel about them.

Summing up, in the first three chapters of this thesis it was shown that children and adolescents with high-functioning ASD were generally surprisingly similar to a typically developing comparison group with regard to their understanding of others' mental states (Chapter 2), their observed responses to others' emotions (Chapter 3), and their ability to present themselves favorably to others (Chapter 4). However, although the empathic and social impairments are not as compelling in our HFASD sample, we cannot conclude that their empathic abilities 'work' in the same way as in typical development. We speculate that children and adolescents with HFASD may rely more strongly on verbal rather than non-verbal cues to infer others' emotions and mental states compared to 'neurotypicals'. This could explain why children and adolescents with HFASD performed equal to typically developing peers on the highly verbal ToM task. Lindner and Rosén (2006) also hinted at an 'over-reliance on verbal content as a compensatory strategy in social interactions' in children and adolescents with HFASD (Lindner & Rosén, 2006, p. 769). Furthermore, when the demand characteristics of a given social situation are relatively structured and explicit, this raises children's social attention and therefore may

increase their social performance. Yet, even when social attention and understanding is sufficiently triggered, some children and adolescents with HFASD may feel intrinsically less motivated to show the corresponding social behavior.

Individual differences in empathic and social behavior

Even though most researchers and clinicians now agree that the autism spectrum consists of a collection of developmental disorders which strongly vary in clinical presentation (e.g., Jones & Klin, 2009; Mundy et al., 2007), individual differences in social interaction impairments, the core impairment in ASD, are still largely neglected. Therefore, in Chapter 5 and 6, we aimed to create a better understanding of individual differences in the empathic and social behavior of children and adolescents with HFASD. Consistent with earlier reports of a higher within-group variability in cognitive performances in a group with ASD compared to a control group (Towgood, Meuwese, Gilbert, Turner, & Burgess, 2009), our group of participants with HFASD showed a marginally larger variance in empathic responding than the typically developing comparison group. For instance, when parents were asked to describe the anticipated responses of their child with HFASD in four hypothetical empathy evoking situations, 14% of the parents predicted that their child would not respond empathically in any of the situations (2% in comparison group), while 31% anticipated that their child would show four or even more empathic responses (62% in comparison group). Hence, even though a lack of empathic responsiveness may characterize a group with HFASD, as our parent reports indeed suggest, this does not apply to each individual child or adolescent with HFASD. As is true for typically developing children, individual child characteristics may importantly affect the degree of empathic responsiveness.

In Chapter 5 of this thesis, we examined the potential contribution of children's temperament, Theory of Mind (ToM) and executive functioning to variance in their empathic responsiveness. Similar to previous findings in typical development (Eisenberg, Wentzel, & Harris, 1998; Sanson, Hemphill, & Smart 2004), individual differences in temperament of children and adolescents with HFASD were significantly related to their empathic responsiveness. More specifically, after controlling for age and receptive verbal ability, participants who were high in emotionality (tendency to become easily emotional) and low in sociability (preference to be alone rather than with others) according to their parents, were less likely to respond empathically to others' emotions. On the other hand, participants' ToM was not associated with their empathic responsiveness after controlling for age and verbal ability. As discussed above, it may be difficult to link ToM tasks to

actual social behavior due to the different characteristics (verbal vs. non-verbal) and demands (explicit vs. implicit) of a ToM task compared to a real life social interaction. Finally, counter to our expectations, those participants who showed a poor inhibitory control of their behavior tended to show *more* empathic responses to the emotions of the interviewer. Possibly, a child's impulsive nature lowers the threshold to respond to the emotional states of an unfamiliar adult. This also means that some children who did *not* respond empathically to the interviewer's emotions, may well have understood and shared the emotions of the interviewer, yet, still failed to show an empathic response due to their inhibited nature (Findlay, Girardi, & Coplan, 2006). In short, the findings of this study suggest that individual child characteristics such as temperament and inhibitory control may account for diagnostically meaningful differences in empathic behavior among children and adolescents with HFASD.

In Chapter 6, we further explored individual differences in social interaction style in HFASD. We used the three social subtypes described by Wing and Gould (1979) to characterize the social interaction style of our participants. Aloof children with ASD, also eloquently portrayed by Leo Kanner (1943), fail to respond adequately to social initiatives of others nor initiate social interactions themselves. Passive children can be motivated to engage in social play when others initiate it, while active-but-odd children actively seek social interactions, but do this in a peculiar or clumsy way. Our high-functioning ASD sample was characterized by a relatively high degree of active-but-odd social behavior. We found that those children who showed more active-but-odd social behavior, also tended to show more ASD symptoms, hyperactivity symptoms, executive functioning problems (particularly inhibition problems), and socio-emotional problems according to parents and teachers. This contrasts previous research among primarily low-functioning (i.e., intellectually disabled) children with ASD which showed that active-but-odd children generally have a lower autism severity compared to passive and aloof children (for a review see Beglinger & Smith, 2001; Borden & Ollendick, 1994; Castelleo & Dawson, 1993; Waterhouse et al., 1996). Within the lower-functioning range of intellectual abilities, the active-but-odd social subtype has also been linked to a higher intelligence compared to the passive and aloof subtype (Beglinger & Smith, 2005; Borden & Ollendick, 1994; Castelleo & Dawson, 1993; Roeyers, 1997). This may in part explain the overall lower autism severity in active-but-odd children in previous studies. However, within a normal range of intellectual functioning, children's intellectual abilities may no longer have a significant impact on their autism severity or social interaction style. Instead, other factors such as

children's ability to inhibit behavioral impulses (i.e., inhibitory control) could affect the social interaction style and autism severity of children and adolescents with HFASD.

An alternative explanation for the increased autism severity in children and adolescents with HFASD and an active-but-odd social interaction style may stem from increased parental awareness or parental stress due to the explicit nature of their child's social impairments. A child with an active-but-odd social interaction style occasionally transgresses social boundaries (e.g., standing too close to a conversation partner) and may thus evoke more negative feedback from the social environment compared to passive children. Therefore, their parents may be more aware of the social impairments of the child or may experience their child's social impairments as more stressful compared to parents with a passive child. Increased parental awareness or parental stress may in turn lead to increased ratings of children's autism severity. As yet, it remains inconclusive whether the social interaction style of children and adolescents with HFASD truly reflects meaningful differences in autism severity.

Findings of a reduced inhibitory control and increased ADHD symptoms in children and adolescents with HFASD and an active-but-odd social interaction style raise the question to what extent an active-but-odd child with HFASD is different from a child with HFASD and ADHD. Previous research has shown a certain degree of overlap between ASD and ADHD in social interaction impairments and deficits in executive functioning (for a review see Nijmeijer et al., 2008). Also, children with ASD and ADHD (or high ADHD symptomatology) show elevated levels of internalizing and externalizing problems and autistic symptoms compared to children with ASD without ADHD or low ADHD symptomatology (Holtmann, Bölte, & Poustka, 2007; Yerys et al., 2009). While the DSM-IV does not allow for a dual diagnosis of ASD and ADHD, this will be possible in the upcoming DSM 5. However, in our HFASD sample, only a small subgroup of children met clinical criteria for an attention deficit disorder (13%) or hyperactivity disorder (5%) as indicated by parent and teacher ratings on the Disruptive Behavior Disorders rating scale (DBD; Pelham, Gnagy, Greenslade, & Milich, 1992). Furthermore, within the group of participants with an active-but-odd social interaction style, their 'active-but-oddness' was unrelated to levels of hyperactivity or inattention. Therefore, the ADHD characteristics of children with HFASD and a predominant active-but-odd social interaction style may not be the only factor adding to the elevated level of social, but odd initiative in these children. Being active-but-odd seems to be something more than simply being (hyper)active.

Summing up, our findings point out that both a lack of empathic responsiveness (Chapter 5) and a lack of social initiative (Chapter 6) should not be considered universal characteristics of children and adolescents with HFASD. Instead, large differences are noted in their empathic and social behavior. Individual child characteristics (e.g., age, temperament, inhibitory control) have a significant impact on the expression of empathy and social behavior in HFASD.

Development of empathy and social behavior

Given the fact that ASD is a developmental disorder that entails lifelong social impairments (Howlin, Mawhood, & Rutter, 2000), it is surprising that comparatively few studies have looked at developmental changes in social behavior of children and adolescents with ASD. While we did not (yet) collect longitudinal data, the wide age range of our sample (6-20 years) does allow for a cross-sectional study of age effects on children's empathic and social abilities. As expected, adolescents demonstrated a better advanced Theory of Mind task performance than children, both in our HFASD and typically developing sample (Chapter 2). Yet, this age effect could be largely explained by an increase in absolute verbal ability. Hence, a growth in children's verbal abilities as they mature likely adds to their understanding of other minds, as also indicated by previous research (e.g., Fisher, Happé, & Dunn, 2005; Ronald, Viding, Happé, & Plomin, 2006). Language may importantly help children to represent others' mental states (Pellicano, 2010). On the other hand, the verbal nature of standard ToM tasks also encourages, and perhaps overemphasizes, the role of verbal ability.

In line with an increased understanding of other minds, typically developing adolescents tended to respond more empathically to the interviewer's emotions than their younger counterparts (Chapter 3). Thus, empathy is more frequently expressed at later stages in children's typical development. This is likely caused by an increased understanding of the social situation and how to show empathy adequately. Alternatively, an increased similarity (in age) between participant and interviewer possibly made it easier for older participants to identify themselves with the interviewer's emotional states (Preston & de Waal, 2002). The same age-related increase in empathic responses to the interviewer was noticed within the current HFASD sample, which corresponds with previous findings from a longitudinal study on empathic responsiveness in ASD (McGovern & Sigman, 2005). However, a closer examination of the data revealed that this age effect was not significant within the subgroup of participants with HFASD and high autism severity (as indicated by their score on the Autism Diagnostic Observation

Schedule). This suggests that developmental effects on children's empathic responsiveness are overruled by autism severity. In groups with and without HFASD, participants' verbal ability was unrelated to their empathic responsiveness to the interviewer's emotional states. Once above a particular verbal IQ threshold (>70), children's verbal IQ may no longer add meaningful variance in their empathic responsiveness. Although a basic level of verbal ability is obviously required to understand someone's verbal emotional cues and verbally respond to these, verbal ability does not crucially determine whether a school-aged child with a normal intelligence expresses empathy or not.

A child's social motivation is another characteristic, besides age and verbal ability, that could importantly contribute to individual differences in empathic and social behavior. Indeed, in Chapter 4 we already indicated that some adolescents with HFASD seemed to be less motivated than typically developing adolescents to make a positive impression on others when this interfered with their sense of honesty or staying true to themselves. Social motivation may also explain why some children with HFASD actively engage in social interactions, while others tend to be more passive or aloof. In our sample we noted a relatively high degree of active-but-odd social behavior, indicating that a certain degree of social initiative may be quite common among these children and adolescents. However, increased social initiative may not necessarily represent increased social motivation. As a blogger with HFASD, who was also mentioned in the Introduction, explained: 'I don't feel a personal connection with all these people, beyond the basic "this is a human being, therefore interesting and valuable." But every day I go out and I chatter away, and at the end of the day I'm ready to collapse, because no matter how much I wish I could just ignore the rest of the world, it doesn't ignore me. ... I wish I could just ignore people, be as aloof and oblivious as you're supposed to be if you're autistic.' ("Reports from a Resident Alien", 2012). This quote suggests that some individuals with ASD, even when they show an active-but-odd social interaction style, may not experience social interactions as inherently emotionally rewarding when compared to typical development. This is in line with an increasing amount of empirical evidence for a reduced social motivation in ASD (Chevallier, Kohls, Troiani, Brodtkin, & Schultz, 2012b).

In the discussion of Chapter 6, we briefly hint at a potential developmental shift in social interaction style – from active-but-odd to passive – of our participants with HFASD. Counter to some previous findings suggesting stability of social interaction style (Beadle-Brown et al., 2002) or an age-related change from aloof to active-but-odd (Beglinger & Smith, 2001) in low-functioning individuals with ASD, we demonstrated that active-but-odd social behavior decreased as a function of age, while passive social behavior

increased. If an active-but-odd social interaction style originates from a reduced or delayed development of inhibitory control, improvement in children's executive functioning across development (Pellicano, 2010) will result in a better regulation of social behavior and a decrease in active-but-odd behavior. Although not addressed in the present study, it has also been suggested that certain individuals with HFASD develop an overly formal or stilted social interaction style into adulthood (Wing, 1996). These individuals have learned to apply social rules such as giving a hand shake as a sign of formal farewell, but use these social rules in an inflexible manner (e.g., shaking hands with someone regardless of the social relation or informal setting). A longitudinal design will be necessary to determine whether the social interaction style of children and adolescents with HFASD truly changes over time, and which factors underlie this potential change.

Clinical implications

Our findings on empathy and social behavior in children with HFASD entail several clinical implications. First of all, our study showed that children and adolescents with HFASD can perform surprisingly adequate on advanced Theory of Mind tasks. Therefore, we may seriously question the usefulness of including an advanced ToM task in the diagnostic assessments of ASD in a school-aged child with a normal IQ. Although standard ToM tasks have consistently and elegantly shown ToM impairments in young children with ASD (e.g., Yirmiya et al., 1998), we propose that advanced ToM tasks may not be a valid indication of the everyday understanding of other minds in older and high-functioning individuals with ASD. An advanced ToM task may fail to reveal ToM impairments in an adolescent with HFASD due to its explicit demands and structure and the adolescent's adequate verbal abilities. Therefore, it may be time to reconsider our operationalization of ToM and use measures that are more closely related to our everyday judgments of others' intentions, beliefs and emotions. A possibly fruitful approach to gain insight into everyday ToM in individuals with HFASD may be to examine their social attention processes and their dependence on structure and verbal cues during social interactions.

Secondly, we found that children and adolescents with HFASD were just as likely as their typically developing age mates to give an empathic response to the emotional states of an unfamiliar interviewer. This is important to consider, because diagnostic observations of autistic behavior (e.g., ADOS) typically include an evaluation of the child's empathy for others' feelings. Our findings thus warrant caution when relying solely on children's observed empathic responses to an unfamiliar adult in a semi-structured

situation such as a psychiatric examination. In this situation it may be quite uncommon for any child to show an overt empathic response to an unfamiliar adult. However, if the child does express empathy, the clinician should be aware that this does not refute an HFASD diagnosis. Our findings further indicate that parents offer vital information on their child's empathic responsiveness in everyday life. Even though most parents with a child with HFASD indicate that their child does respond empathically in some situations, they report these empathic responses substantially less so compared to parents of a typically developing child. Yet, similar to typically developing children, the empathic responsiveness of children and adolescents with HFASD also varies according to individual child characteristics such as temperament.

Third, the large heterogeneity within the autism spectrum makes it particularly challenging to correctly diagnose ASD and to provide adequate interventions for individuals with ASD that meet their specific needs. Children's social interaction style could be a relevant and promising way of subtyping individuals with ASD, because it is right at the core of the disorder, and it could be linked to different needs for and responsiveness to behavioral interventions (Beglinger & Smith, 2005). Our study highlighted the heterogeneity in social interaction style among children and adolescents with HFASD and showed that an active-but-odd social interaction style was associated with increased levels of ASD symptoms, ADHD symptoms, socio-emotional problems, and executive functioning problems in daily life as indicated by parents. Therefore, a conservative interpretation of the results and our main message for clinicians would be that active-but-odd children with HFASD do *not* have fewer problems than children with HFASD who are more passive in social interactions. In fact, they may have more and/or qualitatively different types of problems which in turn ask for different types of interventions. We suggest that an executive functioning training may be a potentially useful intervention for children with HFASD and an active-but-odd social interaction style.

Limitations and recommendations for future research

Despite our large sample and wide range of measures, our study also has several limitations that future research should take into consideration. In several chapters of this thesis we have highlighted the fact that a small majority of our participants with a clinical diagnosis of autism, Asperger's syndrome or PDD-NOS, did not meet the cutoff criteria for an ASD on the Autism Diagnostic Observation Schedule (ADOS; Lord et al., 2000). This was all the more surprising given the fact that most parent reports of autism severity

(as rated on the Social Responsiveness Scale) did confirm the children's clinical diagnoses. We should therefore consider two different options: 1) the clinical diagnosis of the majority of our participants with HFASD is incorrect, or 2) the ADOS (modules 3 and 4) is not sensitive enough to detect ASD in high-functioning children and adolescents. Due to the Dutch mental health care system that requires people to be diagnosed in order to receive services, some of our participants may have been misdiagnosed with HFASD. Yet, it seems highly unlikely that parents would 'seek' an ASD diagnosis for their child when their child develops within the normal range. Assuming then that all of the participants in our study are correctly diagnosed with HFASD, the ADOS (modules 3 and 4) appears to miss a significant number of them. Earlier studies have also shown a relatively poor sensitivity of the ADOS (.49 - .80) in classifying individuals with PDD-NOS or identifying high-functioning adults with ASD (Bastiaansen et al., 2011; Gotham et al., 2008). The ADOS does have a high specificity (Bastiaansen et al., 2011; Lord et al., 2000), indicating that those individuals with an ADOS score above the ASD cutoff most likely have an ASD. Thus, in autism research the ADOS cutoff may be used as a conservative way to differentiate individuals with absolute full-blown ASD from individuals with milder and possibly sub-clinical ASD symptoms. On the other hand, if researchers want to use a valid measure to confirm the clinical ASD diagnoses of normally intelligent individuals, it may be better to use a parent interview or questionnaire (e.g., Autism Diagnostic Interview or Social Responsiveness Scale).

Secondly, contextual influences on children's empathic responsiveness were not addressed in the present study, even though they may be significant (Preston & de Waal, 2002). For instance, the authority of an adult interviewer may have inhibited the empathic responsiveness of some children. Indeed, parents predict that their child with HFASD will respond more empathically to them than to an unfamiliar adult (Hudry & Slaughter, 2009). On the other hand, Sigman et al. (1992) found that pre-school children, both with and without ASD, paid more attention to the distress of an unfamiliar adult compared to a distressed parent. Either way, it is expected that typically developing children will respond more empathically to a peer than to an adult due to increased social motivation or increased similarity (Preston & de Waal, 2002; Saarni, 2001), and this peer effect may be less pronounced among children and adolescents with HFASD (Hauck, Fein, Waterhouse, & Feinstein, 1995; Jackson et al., 2003). We therefore suggest that an observation of a peer interaction could add valuable information on the empathic responsiveness of children and adolescents with HFASD.

Finally, the diversity in social interaction style among children and adolescents with HFASD deserves further clarification. Because the social subtypes are based on observations of children's social behavior, we still know little about the underlying processes and motives. For instance, are children with an active-but-odd social interaction style more intrinsically motivated to seek social contact compared to their passive and aloof counterparts, or are they simply less inhibited? We would suggest to include self-reports, subtle measures of social attention (e.g., using eye tracking devices), as well as experimental manipulations of children's social motivation to answer this particular question. Also, because parents reported a lack of inhibitory control in their active-but-odd child's behavior, it would be worthwhile to test this inhibition hypothesis more directly with experimental tasks. Yet, we recognize that it may be difficult to relate a measure of everyday executive functioning to laboratory measures of executive functioning (Geurts, Corbett, & Solomon, 2009). Finally, due to the positive association found between an active-but-odd social interaction style and symptoms of ADHD, it would be interesting to test the level of 'active-but-oddness' in a sample of children with HFASD who also meet the full diagnostic criteria for ADHD. This would provide more definite answers on the (dis)similarity of HFASD combined with an active-but-odd social behavior and HFASD combined with ADHD.

Conclusion

A first main conclusion of this thesis is that the empathic and social disabilities of school-aged children and adolescents with high-functioning ASD are more refined and less clear-cut than expected based on previous research in primarily young or low-functioning children with ASD. When compared to typically developing peers, they show a comparable understanding of other minds and a comparable empathic responsiveness to others' emotions in a structured situation. In part, the structure of the test (situation) and their cognitive strengths may help these children to overcome or conceal some of their social and empathic disabilities. Nevertheless, our findings do point to a reduced empathic responsiveness in their everyday lives, as reported by parents, as well as a likely decreased social motivation to manage the impression they make on others.

A second important conclusion is that children and adolescents with HFASD show large individual differences in their empathic responsiveness to others' emotions and their social interaction style. These individual differences in empathic and social behavior can be meaningfully linked to differences in children's temperament, inhibitory control, and ADHD symptoms. Thus, we want to emphasize that the autism spectrum does not

represent a uniform group of individuals. Instead, individuals with ASD are indeed individuals who share qualitative impairments in their socio-communicative behavior. If we want to categorize people into boxes - which may be efficient, if not necessary, within a clinical setting - we may perceive this large within-group variability as a nuisance and a source of confusion. However, if we view these individual differences as meaningful instead, they may be used as an important step towards a better understanding of autism spectrum disorder.

Summary

Theoretical background

In the 1940's, psychiatrist Leo Kanner described the behavior of a boy, Donald, who seemed happiest when he could play with his blocks and others would leave him alone (Kanner, 1943). Donald showed remarkably little interest in his social environment. He seemed to be living in a glass bulb, isolated from the others. Today, Donald would be recognized as a child with an autism spectrum disorder (ASD). The current diagnostic criteria for autism still show considerable overlap with the first behavioral observations by Kanner, that is, impairments in the social interaction with others, communication difficulties, and repetitive and restricted interests and behaviors (APA, 2000). The social impairments of children with autism spectrum disorder (ASD) are considered the core impairment of the disorder (APA, 2000; Hobson, 2002; Kanner, 1943). For example, children with ASD find it difficult to form friendships or to share their personal experiences with others.

Our social behavior is largely driven by our ability to empathize with others (de Waal, 2008). Due to the central role of empathy in social behavior, much research in autism has focused on the empathic abilities of children with ASD. These empathic abilities are usually assessed by examining children's understanding of others' thoughts, intentions and emotions. In developmental psychology this understanding is also called *Theory of Mind*. Theory of Mind refers to our everyday ideas (theories) about the mental worlds of the people around us. This helps us to better understand and predict others' behavior. Children with ASD could have difficulties in understanding the thoughts and feelings of others, causing their socially deviant behavior. Indeed young children with ASD usually perform worse on so-called Theory of Mind tests compared to typically developing children (Baron-Cohen, Leslie, & Frith, 1985; Boucher, 2012; Yirmiya, Erel, Shaked, & Solo Monica-Levi, 1998). A classic Theory of Mind test assesses a child's understanding that people act on their own, sometimes erroneous, ideas about reality and not so much on the basis of objective reality itself. Adolescents with ASD and a normal intelligence level ('high-functioning' ASD) have no problems with these classic Theory of Mind tests. More complex tests are therefore needed in order to shed light on the Theory of Mind abilities of older children and adolescents with high-functioning ASD.

Compared to the large number of Theory of Mind studies surprisingly little empirical research has focused on the empathic behavior of children with ASD. For instance, how do children with ASD respond to someone's sorrow or pain? And in what respect do their responses differ from the behavior of typically developing children? Although children with ASD are not insensitive to the emotions of others, several studies

have shown that young children with ASD, often with an intellectual disability, show less attention and less concern when an unknown adult pretends he/she is hurt (Bacon, Fein, Morris, Waterhouse, & Allen, 1998; Scambler, Hepburn, Rutherford, Wehner, & Rogers, 2007; Sigman, Kasari, Kwon, & Yirmiya, 1992). It is still unclear whether this reduced empathic responsiveness also occurs among older children and adolescents with ASD, but without intellectual disabilities.

Although social impairments of children with ASD are considered the core problem of the disorder, there are also large individual differences in both the severity and the nature of these social impairments (APA, 2000, Mundy, Henderson, Inge, & Coman, 2007; Wing & Gould, 1979). In his first descriptions of children with autism, Leo Kanner (1943) emphasized that these children hardly seemed interested in social contact and they did not feel emotionally connected with others. Following Wing and Gould (1979), these children probably belong to the aloof social subtype. Wing and Gould (1979) distinguished three social subtypes within the autism spectrum. The aloof children hardly respond to the social initiative of others. These children often also have an intellectual disability (Beglinger & Smith, 2005; Borden & Ollendick, 1994; Eagle, Romanczyk, & Lenzenweger, 2010). The passive children initiate little social interaction, but do respond to the social initiatives of others. The active-but-odd children actively seek contact with others, but do this in an unusual way. For instance, they may talk endlessly about the same subject without checking if their conversation partner is still interested. Although most clinicians and researchers firmly agree that the autism spectrum encompasses a heterogeneous group of children, there is remarkably little research on individual differences in social behavior of children and adolescents with ASD.

The first objective of this thesis is to improve our understanding of the empathic abilities of school-aged children and adolescents with high-functioning ASD (HFASD). With 'empathic abilities' we refer to the understanding of others' thoughts, desires, and feelings (Theory of Mind) as well as the empathic responding to the emotions of others (empathic responsiveness). The second objective of this thesis is to identify and explain individual differences in social behavior within the autism spectrum.

Method

The results presented in this thesis are derived from two studies: (1) a pilot-study in children (7-10 years) and adolescents (16-22 years) with HFASD (6-20 years) ($n = 26$, 88% boys) and a typically developing comparison group ($n = 26$, 85% boys) (see Chapter 4), and (2) a main study involving 214 (86% boys) children and adolescents with HFASD

from 6 to 20 years and 73 (85% boys) typically developing children and adolescents (see Chapters 2, 3, 5, and 6). Participants with HFASD from the main study are largely (90%) recruited through a school for special education. The other participants with HFASD came from regular education or a youth care institution. The participants from the comparison group were recruited through regular primary and secondary schools in the proximity of Amsterdam.

Participants took part in individual psychological assessments at school, including measures of social competence. One of the measures consisted of five stories to assess the Theory of Mind skills of the participant. In addition, during the assessment the experimenter expressed particular emotions (joy, sadness, pain) to evoke empathic behavior in the participant. After the study, parents and teachers of the participants received a questionnaire about the participant's behavior.

Theory of Mind

A central question in Chapter 2 is whether school-aged children and adolescents with HFASD struggle to understand the mental world of others. Or put differently: Do they show limitations in their Theory of Mind? We used five complex Theory of Mind stories to ascertain participants' understanding of the mental states (such as intentions or emotions) of story characters. Children and adolescents with HFASD performed similarly on the Theory of Mind stories as the comparison group. This is particularly surprising, because previous research - mainly among young children with ASD - consistently showed that children with ASD perform worse on classic Theory of Mind tests. Our results therefore suggest that Theory of Mind, as operationalized in this study, is not a valid explanation for the social impairments seen in children and adolescents with HFASD during everyday social interactions. On the other hand, because of the verbal and explicit nature of the Theory of Mind stories, it may be easier for individuals with HFASD to understand the intentions and emotions of a story character than the often unspoken intentions and emotions of a conversation partner during an actual interaction. The extent to which children and adolescents with HFASD are able to correctly identify the intentions and emotions of others might be largely determined by their attention and the verbal and explicit nature of the emotional signals of others.

Empathic responsiveness

In the study described in Chapter 3, we used two different methods to map empathic responsiveness of children and adolescents with and without HFASD: (1) an

observation of the empathic responsiveness of the child towards the experimenter, and (2) empathic responsiveness of the child as reported by the parent. On three separate occasions during the assessment the experimenter showed an emotion (joy, sadness, pain) according to a standardized protocol. The responses of each participant were recorded on video and coded. An empathic response was a response where the participant appropriately addressed the experimenter's emotional state ('Are you okay?') or offered solutions to improve the experimenter's emotional state. Participants with HFASD responded largely similar to the emotions of the experimenter as participants with a typical development. Thus, we observed no difference in empathic responsiveness between the two groups. It must be noted that both groups showed relatively few explicit empathic responses. The non-significant group difference in empathic responsiveness in this study counters results from previous studies which primarily included young children with ASD and an intellectual disability. Therefore, it seems that children and adolescents with ASD and a normal intelligence are able to respond equally well (or poorly) as their peers without ASD to the emotions of an unfamiliar adult in a semi-structured situation. Their normal intelligence and higher age may play a positive part in their empathic responsiveness.

However, parents of a child with HFASD did expect their child to show significantly fewer empathic responses compared to parents of a typically developing child. This finding suggests that children and adolescents with HFASD respond less empathically to the emotions of others in daily life compared to their peers without HFASD. It is important to note that most parents with a child with HFASD indicated that their child would respond empathically in some cases (53% of all parent reported responses was an empathic response), but less frequently so when compared to children in the comparison group (78% of all reported responses was an empathic response).

Self-presentation

In the study reported in Chapter 4, we examined a basic but vital part of everyday social behavior: introducing oneself to others. More specifically, we examined whether children and adolescents with HFASD would be able to present themselves positively and whether their self-presentation would be strategically adjusted to the specific preferences of an audience. The ability to present yourself positively to another while taking the other's preferences into account, in other words, a successful self-presentation, requires an understanding of what the other person wants to hear (social understanding), but also depends on the motivation to portray a positive image of yourself (social motivation). We found that children and adolescents with HFASD, like the participants in the comparison

group, spoke more positively about themselves when their self-presentation served a positive objective (e.g., the prospect to take part in a prize winning game). Participants with HFASD were less strategic in their self-presentation: they are less likely to highlight those specific skills or interests that are most relevant to the listener. For instance, if the other person loves football, it may be wise to include positive things about football in your self-presentation. However, some participants with HFASD explained that they felt it was important to stay true to themselves, sometimes at the expense of a strategic self-presentation. Children and adolescents with HFASD possibly do not wish to create a positive image of themselves as much as their peers without HFASD. This outcome is in line with accumulating evidence for a reduced social motivation in children and adolescents with ASD (Chevallier, Kohls, Troiani, Brodtkin, & Schultz, 2012b).

Diversity in empathy

Even though children and adolescents with HFASD *as a group* respond less empathically in everyday life (as reported by parents) compared to peers without HFASD, this does not mean that every *individual* with HFASD shows less empathy. The study described in Chapter 5 builds on the premise that individual child characteristics play an important role in the expression of empathy. More specifically, in Chapter 5 we examined whether individual differences in temperament, Theory of Mind and executive functioning (i.e., cognitive functions that guide goal-oriented behavior) contribute to differences in empathic responsiveness within the group of children and adolescents with HFASD. We found that a child's temperament is predictive of his/her empathic responsiveness as reported by parents. The more emotional a child's temperament, the less he/she will be inclined to respond empathically to others' emotions. Also, children with a high level of sociability, thus children who prefer to be with others rather than being alone, respond more empathically to the emotions of others. In short, children and adolescents with HFASD vary in their temperamental make-up - just like their peers in the general population - and these temperamental differences can be meaningfully linked to differences in their empathic responsiveness. In addition, children who show a poor inhibitory control of their behavior according to their parents, were more likely to show empathic responses to the emotions of the experimenter. Children with HFASD who find it hard to control their behavior, possibly respond rather impulsively to the emotions of an unfamiliar adult. Theory of Mind, the ability to understand others' thoughts and feelings, was not related to the degree of empathic responsiveness. The lack of association between Theory of Mind task performance and empathic responsiveness may be because the ability

to identify the mental states of story characters (as in a Theory of Mind test) is not exactly the same as the ability to detect an emotion during a social interaction.

Diversity in social behavior

In Chapter 6 we discuss the different social interaction styles of children and adolescents with ASD and a normal intelligence. To date, research into social interaction styles was mainly performed among individuals with ASD and intellectual disabilities. Because children with an aloof or passive interaction style often were less intelligent than children with an active-but-odd interaction style (Beglinger & Smith, 2005; Borden & Ollendick, 1994; Castelloe & Dawson, 1993; Roeyers, 1997), it remained unclear whether children's social interaction style was not just a feature of their intellectual level. In our study, 40% of children and adolescents with HFASD showed a high degree of active-but-odd social behavior, that is, they relatively often initiate social contact, but do this in an unusual or awkward way. Children with HFASD who showed more active-but-odd social behavior according to their parents, also showed more autistic traits, symptoms of hyperactivity, inhibition problems and socio-emotional problems according to parents and teachers. It is therefore important for clinicians to realize that socially active behavior of a child with HFASD does not necessarily indicate that this child experiences fewer problems than a child with HFASD and a predominantly passive behavioral style. In fact, children and adolescents who tend to show active-but-odd social behavior, may have more or different problems than those who remain more aloof or passive during social interactions. These differences can be used as potential leads for more personalized interventions for children and adolescents with HFASD.

Conclusion

Since the first descriptions of children with autism by Leo Kanner (1943), much has changed in our ideas of autism. Yet, most scientists and clinicians still agree that social impairments form the core of the disorder. A first conclusion of this thesis is that the empathic and social disabilities in ASD, as previously and consistently shown in young children with ASD (sometimes with an intellectual disability), are less straightforward in school-aged children and adolescents with ASD and a normal intelligence (HFASD). When compared to typically developing peers, they show a comparable understanding of other minds and a comparable empathic responsiveness to others' emotions in a structured situation. The structure of the test (situation) and intellectual abilities may help children and adolescents with HFASD to overcome or conceal some of their social and

empathic disabilities. However, parent reports indicate that children and adolescents with HFASD do in fact respond less empathically to others' emotions in everyday situations compared to peers. Also, it appears that some children and adolescents with HFASD are characterized by a reduced social motivation. More specifically, they seem to be less motivated than typically developing peers to make a positive impression on others when this impression jeopardizes their own sense of self.

A second important conclusion of this research is that children and adolescents with HFASD show large individual differences in their empathic and social behavior. A lack of empathic responsiveness or a lack of social initiative do not seem to be universal characteristics of HFASD per se, but they are shaped by individual differences in age, temperament, inhibitory control, and hyperactivity. It is important for both scientists and clinicians to appreciate that the autism spectrum represents a heterogeneous group of individuals. However, as long as we strive to categorize people (disorder versus no disorder) - which may be efficient, if not necessary, in a clinical setting -, the large diversity within the autism spectrum will remain a source of frustration and confusion. If we view these individual differences as meaningful instead, they may be used as an important step towards a better understanding of autism spectrum disorder.

Dutch Summary (Samenvatting)

Heroverweging van kernproblemen in autisme:

Diversiteit in empathie en sociaal gedrag

Theoretische achtergrond

In de jaren '40 beschreef de psychiater Leo Kanner het gedrag van een jongetje, Donald, dat het gelukkigst leek wanneer hij met zijn blokjes kon spelen en door anderen met rust gelaten werd (Kanner, 1943). Donald toonde opvallend weinig interesse in zijn sociale omgeving. Als leefde hij in een glazen bol, afgesloten van de anderen. Tegenwoordig zou Donald herkend worden als een kind met een autisme spectrum stoornis (ASS). De huidige diagnostische criteria voor autisme vertonen nog steeds veel overlap met de eerste gedragsobservaties door Kanner, namelijk beperkingen in de sociale omgang met anderen, communicatieve problemen, en herhalende en beperkte interesses en gedragingen (APA, 2000). De sociale beperkingen van kinderen met een autisme spectrum stoornis (ASS) worden beschouwd als het kernprobleem van de stoornis (APA, 2000; Hobson, 2002; Kanner, 1943). Kinderen met ASS vinden het bijvoorbeeld lastig om vriendschappen te vormen of om hun persoonlijke ervaringen te delen met anderen.

Ons sociale gedrag wordt in belangrijke mate aangedreven door ons vermogen om met anderen begaan te zijn, oftewel ons vermogen tot empathie (de Waal, 2008). Vanwege het belang van empathie voor sociaal gedrag is er veel onderzoek verricht naar de empathische vermogens van kinderen met ASS. Deze empathische vermogens zijn veelvuldig onderzocht door te kijken naar het inzicht dat kinderen met ASS hebben in de gedachten, intenties, en emoties van anderen. Dit inzicht wordt in de ontwikkelingspsychologie ook wel *Theory of Mind* genoemd. Theory of Mind verwijst naar onze alledaagse ideeën (theorieën) over de belevingswerelden van de mensen om ons heen. Op die manier kunnen we het gedrag van anderen beter begrijpen en voorspellen. Kinderen met ASS hebben mogelijk problemen om zich in te leven in de gedachten en gevoelens van anderen, waardoor zij afwijkend sociaal gedrag laten zien. Inderdaad presteren jonge kinderen met ASS doorgaans slechter op zogeheten Theory of Mind tests in vergelijking met zich normaal ontwikkelende kinderen (Baron-Cohen, Leslie, & Frith, 1985; Boucher, 2012; Yirmiya, Erel, Shaked, & Solomonica-Levi, 1998). Met een klassieke Theory of Mind test wordt onderzocht of een kind begrijpt dat mensen handelen op basis van hun, soms foutieve, *ideeën* over de werkelijkheid en niet zo zeer op basis van de objectieve werkelijkheid zelf. Adolescenten met ASS maar met een normaal intelligentieniveau ('hoog-functionerend' ASS) hebben geen moeite (meer) met deze klassieke Theory of Mind tests. Complexere tests zijn daarom nodig om licht te kunnen werpen op de Theory of Mind vermogens van oudere kinderen en adolescenten met hoog-functionerend ASS.

In vergelijking met de grote hoeveelheid Theory of Mind onderzoek is er nog verrassend weinig empirisch onderzoek verricht naar het empathische *gedrag* van kinderen met ASS. Hoe reageren kinderen met ASS bijvoorbeeld op het verdriet of de pijn van een ander? En in welk opzicht wijken die reacties af van het gedrag van zich normaal ontwikkelende kinderen? Hoewel kinderen met ASS niet ongevoelig zijn voor de emoties van anderen, hebben verschillende studies laten zien dat jonge kinderen met ASS, veelal met een verstandelijke beperking, minder aandacht en minder bezorgdheid tonen wanneer een onbekende volwassene doet alsof hij/zij zich heeft bezeerd (Bacon, Fein, Morris, Waterhouse, & Allen, 1998; Scambler, Hepburn, Rutherford, Wehner, & Rogers, 2007; Sigman, Kasari, Kwon, & Yirmiya, 1992). Het is echter nog onvoldoende uitgezocht of deze verminderde empathische responsiviteit ook voorkomt onder oudere kinderen en adolescenten met ASS maar zonder een verstandelijke beperking.

Hoewel sociale gedragsbeperkingen van kinderen met ASS worden gezien als het kernprobleem van de stoornis, bestaan er ook grote individuele verschillen in zowel de ernst als de aard van deze sociale beperkingen (APA; 2000; Mundy, Henderson, Inge, & Coman, 2007; Wing & Gould, 1979). Leo Kanner (1943) benadrukte in zijn eerste beschrijvingen van kinderen met autisme dat ze nauwelijks geïnteresseerd leken in sociaal contact en zich niet emotioneel verbonden voelden met de mensen om hen heen. De onderzoekers Wing en Gould (1979) zouden deze kinderen waarschijnlijk tot het afzijdige sociale subtype rekenen. Zij onderscheidden drie sociale subtypen binnen het autisme spectrum. De *afzijdige* kinderen reageren nauwelijks op het sociale initiatief van anderen. Vaak hebben deze kinderen ook nog eens een verstandelijke beperking (Beglinger & Smith, 2005; Borden & Ollendick, 1994; Eagle, Romanczyk, & Lenzenweger, 2010). De *passieve* kinderen initiëren zelf weinig sociale interactie, maar reageren wel op het sociale initiatief van de ander. De *actief-maar-vreemde* kinderen maken actief contact met anderen, maar doen dit op een ongebruikelijke manier. Ze kunnen bijvoorbeeld oeverloos doorpraten over hetzelfde onderwerp zonder te controleren of hun gesprekspartner het nog interessant vindt. Hoewel de meeste klinici en onderzoekers direct zullen erkennen dat het autisme spectrum een heterogene groep kinderen omvat, is er tot nog toe opvallend weinig onderzoek verricht naar individuele verschillen in het sociale gedrag van kinderen en adolescenten met ASS.

Dit proefschrift heeft als eerste doelstelling om meer inzicht te creëren in de empathische vermogens van schoolgaande kinderen en adolescenten met hoog-functionerend ASS (HFASS). Met empathische vermogens bedoelen we hier zowel inzicht in de gedachten, wensen, en gevoelens van anderen (Theory of Mind) als empathisch

reageren op de emoties van een ander (empathische responsiviteit). De tweede doelstelling van dit proefschrift is het in kaart brengen en verklaren van individuele verschillen in sociaal gedrag binnen het autisme spectrum.

Methode

De resultaten die in dit proefschrift besproken worden, zijn afkomstig van twee studies: (1) een voorstudie onder kinderen (7-10 jaar) en adolescenten (16-22 jaar) met HFASS (n=26; 88% jongens) en een zich normaal ontwikkelende vergelijkingsgroep (n=26; 85% jongens) (zie hoofdstuk 4), en (2) een hoofdstudie waaraan 214 (86% jongens) kinderen en adolescenten met HFASS deelnamen van 6 tot en met 20 jaar en 73 (85% jongens) zich normaal ontwikkelende kinderen en adolescenten die samen de vergelijkingsgroep vormen (zie hoofdstukken 2, 3, 5, en 6). Deelnemers met HFASS uit de hoofdstudie zijn grotendeels (90%) geworven via een school voor speciaal onderwijs. De overige deelnemers met HFASS zijn afkomstig van het reguliere onderwijs of een jeugdzorginstelling. De deelnemers uit de vergelijkingsgroep zijn geworven via reguliere basis- en middelbare scholen in de buurt van Amsterdam.

Deelname aan het onderzoek bestond uit een individueel psychologisch onderzoek op school waarin de sociale kennis en vaardigheden van het kind getoetst werden. Een van de tests bestond uit een vijftal verhalen om het Theory of Mind vermogen van de deelnemer te onderzoeken. Daarnaast liet de onderzoeker tijdens vastgelegde momenten in het onderzoek een bepaalde emotie (blijdschap, verdriet, pijn) zien om daarmee empathisch gedrag bij de deelnemer uit te lokken. Na het onderzoek ontvingen de ouders en de mentor van elke deelnemer een vragenlijst over het gedrag van de deelnemer.

Theory of Mind

In hoofdstuk 2 van dit proefschrift staat de vraag centraal of schoolgaande kinderen en adolescenten met HFASS moeite hebben om zich te verplaatsen in de belevingswereld van anderen, oftewel of zij beperkingen laten zien in hun *Theory of Mind*. Aan de hand van vijf complexe Theory of Mind verhalen is het inzicht van de deelnemers getoetst in de mentale toestanden (zoals een intentie of emotie) van personages. De prestaties van kinderen en adolescenten met HFASS op de Theory of Mind verhalen bleken vergelijkbaar met de prestaties van de vergelijkingsgroep. Dit is op z'n minst verrassend te noemen, omdat voorgaand onderzoek onder hoofdzakelijk jonge kinderen met ASS vaak heeft uitgewezen dat kinderen met ASS slechter presteren op klassieke Theory of Mind tests. Ons resultaat suggereert daarom dat Theory of Mind, zoals

geoperationaliseerd in deze studie, geen verklaring vormt voor de sociale beperkingen die kinderen en adolescenten met HFASS laten zien in hun alledaagse sociale interacties. Anderzijds is het vanwege de verbale en expliciete aard van Theory of Mind verhalen mogelijk makkelijker voor individuen met HFASS om de intenties en emoties van een personage uit een verhaal te begrijpen dan de vaak onuitgesproken intenties en emoties van een gesprekspartner tijdens een daadwerkelijke interactie. De mate waarin kinderen en adolescenten met HFASS in staat zijn om de intenties en emoties van anderen correct te identificeren zou weleens in belangrijke mate bepaald kunnen worden door hun aandacht en de mate van expliciete van de emotionele signalen van anderen.

Empathische responsiviteit

In het onderzoek beschreven in hoofdstuk 3 maakten we gebruik van twee verschillende methoden om de empathische responsiviteit van kinderen en adolescenten met en zonder HFASS in kaart te brengen: (1) een observatie van de empathische responsiviteit van het kind richting de onderzoeker, en (2) empathische responsiviteit van het kind zoals gerapporteerd door de ouder. Tijdens het individuele onderzoek liet de onderzoeker op drie verschillende momenten een emotie (blijdschap, verdriet, pijn) zien volgens een gestandaardiseerd protocol. De reacties van elke deelnemer werden op video vastgelegd en achteraf gecodeerd. Een empathische reactie werd gekenmerkt door de passende wijze waarop de deelnemer inging op de emotionele toestand van de onderzoeker ('Gaat het?') of oplossingen aanreikte om de toestand van de onderzoeker te verbeteren. Het bleek dat deelnemers met HFASS doorgaans hetzelfde reageerden op de getoonde emoties van de onderzoeker als de deelnemers met een normale ontwikkeling. Er werd dus geen verschil geobserveerd in empathische responsiviteit tussen de twee groepen. Daarbij moet worden opgemerkt dat beide groepen relatief weinig expliciete empathische reacties lieten zien. Het niet-significante groepsverschil in empathische responsiviteit in deze studie contrasteert met de resultaten van voorgaande onderzoeken onder hoofdzakelijk jonge kinderen met ASS en een verstandelijke beperking. Het lijkt er daarom op dat kinderen en adolescenten met ASS en een normale intelligentie net zo goed (of slecht) als hun leeftijdgenoten zonder ASS reageren op de emoties van een onbekende volwassene in een semigestructureerde situatie. Mogelijk speelt hun normale intelligentie en hun hogere leeftijd een positieve rol in hun empathische responsiviteit.

Ouders van een kind met HFASS bleken echter beduidend minder vaak een empathische reactie te verwachten van hun kind dan ouders van een zich normaal ontwikkelend kind. Deze uitkomst suggereert dat kinderen en adolescenten met HFASS in

het dagelijks leven minder empathisch op de emoties van anderen reageren in vergelijking met hun leeftijdgenoten zonder HFASS. Het is hierbij belangrijk om op te merken dat de meeste ouders met een kind met HFASS aangaven dat hun kind wel degelijk empathisch zou reageren in sommige gevallen (53% van alle door ouders gerapporteerde reacties was een empathische reactie), doch minder frequent in vergelijking met kinderen uit de vergelijkingsgroep (78% van alle gerapporteerde reacties was een empathische reactie).

Zelfpresentatie

In de studie gerapporteerd in hoofdstuk 4 werd een basaal, maar belangrijk onderdeel van het alledaagse sociale verkeer onderzocht: jezelf voorstellen aan een ander. Meer specifiek hebben we gekeken of kinderen en adolescenten met HFASS in staat zijn om zichzelf positief te presenteren en of zij hun zelfpresentatie strategisch kunnen aanpassen aan de specifieke voorkeuren van een toehoorder. Het vermogen om jezelf positief te presenteren aan een ander terwijl je rekening houdt met de voorkeuren van die ander, met andere woorden een succesvolle zelfpresentatie, vereist begrip van wat de ander wil horen (sociaal inzicht), maar hangt ook af van de motivatie om een positief beeld van jezelf neer te zetten (sociale motivatie). Wij vonden dat kinderen en adolescenten met HFASS, net als de deelnemers van de vergelijkingsgroep, positiever over zichzelf vertelden wanneer hun zelfpresentatie een positief doel diende (bijvoorbeeld het vooruitzicht om deel te nemen aan een prijzenspel). Deelnemers met HFASS waren echter minder strategisch in hun zelfpresentatie: zij belichtten minder vaak die specifieke vaardigheden of interesses van henzelf die vooral relevant zijn voor de toehoorder. Als de ander van voetbal houdt, is het bijvoorbeeld handig om je eigen affiniteit met voetbal ter sprake te brengen. Sommige deelnemers met HFASS vertelden echter dat ze het belangrijk vonden om zich niet anders voor te doen dan ze zijn, wat soms ten koste ging van een strategische zelfpresentatie. Mogelijk hechten kinderen en adolescenten met HFASS er minder belang aan een goede indruk te maken op anderen dan hun leeftijdgenoten zonder HFASS. Deze uitkomst ligt in de lijn van steeds meer bewijs voor een verminderde sociale motivatie bij kinderen en adolescenten met ASS (Chevallier, Kohls, Troiani, Brodtkin, & Schultz, 2012b).

Diversiteit in empathie

Wanneer kinderen en adolescenten met HFASS in het dagelijks leven *als groep* minder empathisch reageren in vergelijking met leeftijdgenoten zonder HFASS (zoals gerapporteerd door ouders), betekent dit niet dat elk *individu* met HFASS minder empathie

toont. De aanname van hoofdstuk 5 is dan ook dat individuele kindkenmerken een belangrijke rol spelen in het wel of niet tonen van empathisch gedrag. Meer specifiek onderzochten we in hoofdstuk 5 of individuele verschillen in temperament, Theory of Mind, en executief functioneren (d.w.z., cognitieve functies die doelgericht gedrag sturen) bijdragen aan verschillen in empathische responsiviteit binnen de groep kinderen en adolescenten met HFASS. Het bleek dat het temperament van een kind met HFASS voorspellend is voor zijn/haar empathische responsiviteit zoals gerapporteerd door ouders. Hoe emotioneler een kind van nature is, hoe minder hij/zij geneigd zal zijn om empathisch te reageren op de emotie van een ander. Daarnaast reageren kinderen met een hoge sociabiliteit, dat wil zeggen kinderen die liever met anderen zijn dan alleen, eerder empathisch op de emotie van een ander. Kortom, kinderen en adolescenten met HFASS verschillen - net als hun leeftijdgenoten in de normale populatie - in hun temperament en deze temperamentsverschillen kunnen betekenisvol worden gekoppeld aan verschillen in hun empathische responsiviteit. Daarnaast viel op dat kinderen die hun gedrag volgens ouders slecht konden remmen (inhibitieproblemen hadden), juist *meer* empathische reacties toonden naar aanleiding van de emoties van de onderzoeker. Kinderen met HFASS die moeite hebben om hun gedrag te controleren, reageren mogelijk impulsiever op de emoties van een onbekende volwassene. Theory of Mind, het vermogen om je in te leven in de gedachten en gevoelens van anderen, bleek niet gerelateerd te zijn aan de mate van empathische responsiviteit. Het gebrek aan samenhang tussen Theory of Mind prestaties en empathische responsiviteit wordt mogelijk veroorzaakt doordat de vaardigheid om de mentale toestand van een personage in een verhaal correct te identificeren (zoals in een Theory of Mind test) niet exact dezelfde is als de vaardigheid om de emotie van een ander op te pikken tijdens een sociale interactie.

Diversiteit in sociaal gedrag

In hoofdstuk 6 bespreken we de verschillende sociale gedragsstijlen van kinderen en adolescenten met ASS en een normale intelligentie. Voorgaand onderzoek naar sociale gedragsstijlen vond tot nog toe voornamelijk plaats onder individuen met ASS en een verstandelijke beperking. Omdat kinderen met een afzijdige of passieve gedragsstijl vaak minder intelligent bleken dan kinderen met een actief-maar-vreemde gedragsstijl (Beglinger & Smith, 2005; Borden & Ollendick, 1994; Castelloe & Dawson, 1993; Roeyers, 1997), bleef het de vraag of hun sociale gedragsstijl niet gewoon paste bij een bepaald intelligentieniveau. In onze studie vertoonde 40% van de kinderen en adolescenten met HFASS een hoge mate van actief-maar-vreemd sociaal gedrag, dat wil zeggen dat zij

relatief vaak sociaal contact initiëren, maar dit op een ongebruikelijke of onhandige manier doen. Kinderen met HFASS die meer actief-maar-vreemd sociaal gedrag lieten zien volgens hun ouders, vertoonden ook meer autistische kenmerken, kenmerken van hyperactiviteit, inhibitieproblemen en socio-emotionele problemen volgens ouders en leerkrachten. Het is daarom belangrijk voor klinici om zich te realiseren dat sociaal actief gedrag van een kind met HFASS niet hoeft te betekenen dat dit kind ook *minder* problemen ervaart dan een kind met HFASS en een overwegend passieve gedragsstijl. Kinderen en adolescenten die vooral actief-maar-vreemd sociaal gedrag laten zien, hebben mogelijk zelfs meer of andersoortige problemen dan zij die zich vooral afzijdig of passief opstellen in de sociale interactie. Deze verschillen kunnen gebruikt worden als mogelijke aanknopingspunten voor meer gepersonaliseerde interventies voor kinderen en adolescenten met HFASS.

Conclusie

Sinds de eerste gedragsbeschrijvingen van kinderen met autisme door Leo Kanner (1943) is er veel veranderd aan de kennis en het beeld dat we hebben van autisme. Toch zijn de meeste wetenschappers en klinici het er nog steeds over eens dat de sociale beperkingen de kern vormen van de stoornis. Een eerste conclusie van dit proefschrift is dat het empathische en sociale onvermogen, zoals dat consistent is vastgesteld in onderzoek bij met name jonge kinderen met ASS (soms met een verstandelijke beperking), minder duidelijk naar voren treedt bij schoolgaande kinderen en adolescenten met ASS en een normale intelligentie (HFASS). Dit onderzoek liet zien dat zij net zo goed als hun leeftijdgenoten zonder ASS in staat zijn om de mentale toestanden van personages te duiden of om empathisch te reageren op de emoties van een ander in een gestructureerde situatie. Het kan zijn dat de structuur van de test (situatie) en hun intellectuele vermogens er gedeeltelijk voor zorgen dat deze kinderen hun empathische en sociale beperkingen kunnen compenseren of verbergen. Ouders geven echter aan dat kinderen en adolescenten met HFASS in het dagelijks leven wel degelijk minder empathie tonen in vergelijking met leeftijdgenoten. Daarnaast lijken sommige kinderen en adolescenten met HFASS gekenmerkt te worden door een verminderde sociale motivatie. Zelfs wanneer ze zich bewust zijn van specifieke sociale eisen die aan hen gesteld worden, weigeren zij soms zich te conformeren aan dit sociaal wenselijke beeld.

Een tweede belangrijke conclusie van dit onderzoek is dat kinderen en adolescenten met HFASS grote onderlinge verschillen laten zien in hun empathische en sociale gedrag. Een gebrek aan empathische responsiviteit of een gebrek aan sociaal

initiatief lijken niet zo zeer universele kenmerken te zijn van HFASS, maar lijken mede te worden gevormd door individuele verschillen in leeftijd, temperament, inhibitievermogen, en hyperactiviteit. Het is belangrijk voor zowel wetenschappers als klinici om stil te staan bij het gegeven dat het autisme spectrum een heterogene groep van individuen representeert. Zolang we er echter naar streven om mensen op te delen in categorieën (stoornis versus geen stoornis) - wat wellicht in een klinische setting efficiënt en welhaast onvermijdelijk is -, zal de grote diversiteit binnen het autisme spectrum een bron van frustratie en verwarring blijven. Wanneer we deze individuele verschillen echter gaan beschouwen als betekenisvol, kunnen zij gebruikt worden als een belangrijke stap naar een beter begrip van het autisme spectrum.

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Appendices

Appendix I (Chapter 2)

1. Surprise (*derived from (Sullivan et al., 1994)*)

Tonight is Alex's Birthday, and Mom is surprising him with a puppy. She has hidden the puppy in the shed. Alex says, "Mum, I really hope you get me a puppy for my birthday" Mom wants to surprise Alex with the puppy so she says, "Sorry, Alex, I didn't get you a puppy for your birthday. I got you a really great toy instead."

1. Did Mom really get Alex a toy for his birthday? (probe question 1)
2. Did Mom tell Alex she got him a toy for his birthday? (probe question 2)

Alex goes outside to play. He decides to get his ball from the shed. In the shed, Alex finds the Birthday puppy! Alex says to himself, "Wow, Mom didn't get me a toy, she really got me a puppy for my Birthday." Mom doesn't see Alex go to the shed and find the Birthday puppy.

3. Does Alex know that his Mom got him a puppy for his birthday? (control question)
4. Does Mom know that Alex saw the Birthday puppy in the shed? (control question)

While Alex is outside, his Grandma comes to visit. Grandma asks Mom, "Does Alex know what you really got him for his birthday?"

3. What does Mom say to Grandma? (second-order ignorance question)

Then, Grandma says to Mom, "What does Alex think you got him for his birthday?"

4. What does Mom say to Grandma? (second-order false-belief question)
5. Why does Mom say that? (justification question)

2. The rollercoaster (*derived from (Begeer et al., 2011)*)

Karel is 10 years and is in Grade 4 at elementary school. He is really looking forward to going to school today because they're going to do something fun.

Today, Karel's teacher is having a Birthday. Karel is going to the Fun Park with all the other children in his class. After an hour on the bus, the children arrive at the Fun Park.

Everyone wants to go on the rollercoaster straight away. Karel doesn't like the rollercoaster, he is scared of the rollercoaster. When it's Karel's turn, he doesn't want to go in the rollercoaster at all. A boy that is standing in front of him says, 'Cool! A ride on the rollercoaster! Are you coming?' Karel says, 'You go ahead, I think the rollercoaster is for little kids, I'd rather eat an ice cream.'

1. Why do the children go to the amusement park? (physical state question)
2. What does Karel say when his classmate asks him to go in the rollercoaster? (control question)
3. Is it true what he tells his classmate?
4. Why does he say this? (mental state question)

3. The robbery (*derived from (Kaland et al., 2002)*)

Paul and his mother are walking in a dangerous part of the city on a dark evening. They're both a little scared because they've heard that people sometimes get robbed here.

Earlier that day, Paul's mother went to the bank to get \$500. Her washing machine is broken and she needs to buy a new one quickly. She put the money in the inner-pocket of her jacket instead of putting it in her purse.

Suddenly two armed men jump out of a dark alley. They shout, "Hands up, this is a robbery! Where's the money little lady?" Paul's mother takes a big risk and says that the money is in her inner pocket. The robbers grab her purse out of her hands and disappear into the darkness.

1. Why has Paul's mother gone to the bank to get money? (physical state question)
2. Where did Paul's mother hide her money? (control question)
3. Why does Paul's mother say that the money is in her inner pocket, where she really hid it, and not in her purse? (mental state question)

4. Little Mrs. Smit (*derived from (Kaland et al., 2002)*)

Johan is a piano tuner. He makes sure that pianos keep sounding beautiful.

One day Johan is called by Mrs. Emma Smit, who wants to have her piano tuned. A few minutes earlier, a small job that Johan was going to do that morning at the concert hall was cancelled.

A little later, Johan rings the bell at Mrs. Emma Smit's house. Johan sees that the old lady is rather short, she has short legs and a crooked back. Mrs. Emma Smit has a disease that causes her pain when she walks.

After an hour, Johan has tuned the piano. Johan, who is known to be a good piano tuner, says, "Little Emma, I noticed your piano seat was a little high. I adjusted the seat so you can reach the ground with your short little legs."

1. Why does Mrs. Emma Smit not have to wait to get her piano tuned? (physical state question)
2. What do people think about Johan as a piano tuner? (control question)
3. What do you think that Mrs. Emma Smit is feeling when she hears what Johan says to her? (mental state question)

5. Cleaning up the room (*derived from (Kaland et al., 2002)*)

Tom and Simon are brothers. Tom is 8 years and Simon is 14 years. Their mother is very strict and always wants them to clean up their rooms. Tom always makes a big mess of his room and is very untidy. His mother often complains about the mess. Simon is always very tidy.

Their mother says that they both need to clean up their room. After a while, mother asks them if they have finished. Simon says that he is done.

Mother asks Simon to check if Tom has also cleaned up his room. But Tom hasn't even started cleaning up! Simon opens the door of Tom's room and sees that it is still an enormous mess. He shouts to his mother, 'Mom, as usual, Tom has cleaned up very well!'

1. What does Simon's room look like? (physical state question)
2. What does mother ask Simon to do? (control question)
3. Simon says to his mother that Tom, as usual, has cleaned up very well. Is it true what he says?
4. Why does Simon say that? (mental state question)

Appendix II (Chapters 3 and 5)

Vignettes used for parent reports of children's empathic responsiveness

Situation response questionnaire

Four different **school situations** are described below. Please write down how you think your child would respond in each given situation. Write down the response that seems **most probable** to you. If the response of your child contains a verbal reply, please put this verbal reply between quotation marks: *Example.*'

1. Your child is standing alone with a teacher in the school hall way. The teacher tells your child enthusiastically that he/she will go to the theater tonight. Your child knows the teacher likes going to the theater. What would your child do or say?
2. The teacher is holding a pile of papers and has asked your child to keep the door open for him/her. The teacher stumbles over the doorstep and falls to the ground. The teacher gets up and rubs with a hand over the knee that hit the ground. Only your child has seen what has happened. What would your child do or say?
3. It is almost summer. Your child is standing alone with the teacher in the school hall way. The teacher tells your child disappointed that he/she will not be going on holiday this year. Your child knows that the teacher likes going on holiday. What would your child do or say?
4. The teacher knocks down a cup of coffee and gets the glowing hot coffee over his/her legs. The teacher jumps out of the chair. Only your child has seen what has happened. What would your child do or say?

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Acknowledgements (Dankwoord)

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Overview of publications and presentations

Anke Scheeren was born in Roermond, the Netherlands, in 1982. From 2000 to 2005 she studied (biological) psychology at the University of Maastricht. Her master thesis on the Broader Autism Phenotype was published in an international peer-reviewed journal. In December 2007 she continued her research on autism as a PhD student at the department of developmental psychology of the VU University in Amsterdam. After finishing her PhD thesis, and still not bored with autism, she now works as a post-doctoral researcher at the VU University.

Publications in peer-reviewed journals

Scheeren, A. M., de Rosnay, M., Koot, H. M., & Begeer, S. (in press). Rethinking Theory of Mind in high-functioning autism spectrum disorder. *Journal of Child Psychology and Psychiatry*.

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Scheeren, A. M., Begeer, S., & Koot, H. M. (2011). Factors associated with empathic behavior in children and adolescents with high-functioning ASD. Poster presented

11th Annual International Meeting for Autism Research (IMFAR): Toronto (2012, May 19).

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Backer van Ommeren, T., Begeer, S., **Scheeren, A. M.**, & Koot, H. M. (2011) Measuring reciprocity in high functioning children and adolescents with autism spectrum disorders. Poster presented 10th Annual International Meeting for Autism Research (IMFAR): San Diego (2011, May 12-14).

Scheeren, A. M., Begeer, S. & Koot, H. M. (2010). Advanced Theory of Mind from childhood to adolescence. Poster presented 9th Annual International Meeting for Autism Research (IMFAR): Philadelphia (2010, May 20).

Scheeren, A. M., Begeer, S.M., Wijhe, J. van & Koot, H. M. (2009). Empathic responses to hypothetical and real life situations in children and adolescents with autism spectrum disorder. Poster presented 8th Annual International Meeting for Autism Research (IMFAR): Chicago (2009, May 7-9).

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