

# A longitudinal study of parent-child and child-child interactions in very young siblings of children with autism spectrum disorder

**Chloè Bontinck**

Supervisor: Prof. dr. Herbert Roeyers

Co-supervisor: dr. Petra Warreyn

A dissertation submitted to Ghent University in partial  
fulfilment of the requirements for the degree of  
Doctor of Psychology

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## DANKWOORD

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# CHAPTER **1**

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## GENERAL INTRODUCTION

In this first chapter, we start with a general description of autism spectrum disorder (ASD). Furthermore, the increased familial risk for ASD as well as the developmental trajectories of younger siblings of children with ASD are reported. Next, the importance and characteristics of the early social environment are discussed, both for typically developing children and for children with (an increased risk of) ASD. In addition, the use of different coding methods of social interaction is elaborated upon. We conclude this chapter with the formulation of the research objectives and an overview of the chapters included in this dissertation.

## AUTISM SPECTRUM DISORDER

Autism spectrum disorder is a neurodevelopmental disorder characterised by persistent deficits in social communication and social interaction across multiple contexts (American Psychiatric Association [APA], 2013). According to the Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-5; APA, 2013, p. 50) this includes 1) deficits in social-emotional reciprocity, 2) deficits in nonverbal communicative behaviours used for social interaction, and 3) deficits in developing, maintaining and understanding relationships. These symptoms co-occur with restrictive and repetitive patterns of behaviour, interests, or activities, expressed by at least two of the following criteria: 1) stereotyped or repetitive motor movements, use of objects, or activities; 2) insistence on sameness, inflexible adherence to routines, or ritualised patterns of verbal or nonverbal behaviour; 3) highly restricted, fixated interests that are abnormal in intensity or focus; or 4) hyper- or hyporeactivity to sensory input or unusual interest in sensory aspects of the environment (APA, 2013, p. 50). The prevalence of ASD is estimated around 60-70 per 10,000 children (Elsabbagh et al., 2012). In addition, epidemiological studies report a higher prevalence of ASD in males with a 4:1 ratio of males to females (Elsabbagh et al., 2012; Fombonne, 2009). Although evidence suggests that a reliable ASD diagnosis is possible from the age of 2 onwards (Charman et al., 2005; Chawarska, Klin, Paul, Macari, & Volkmar, 2009), only a minority (17-23%) of the children with ASD is identified before the age of 3 years (Barbaro & Dissanayake, 2009; Sheldrick, Maye, & Carter, 2017).

Early ASD characteristics become visible during the first years of life, especially in the period between 12 and 24 months (Bryson et al., 2007; Landa & Garrett-Mayer, 2006). In this period, ASD is characterised by phenotypic heterogeneity, complicating diagnostic assessment (Szatmari et al., 2016). First, early atypicalities are visible in the social communication and interaction domain (e.g., Bryson et al., 2007). These include impairments in the use of gestures, response to joint attention, social engagement/interest, eye contact, sharing interest, orientation to name, imitation, and social smiling (Chawarska et al., 2014; Mitchell et al., 2006; Ozonoff et al., 2014; Sullivan et al., 2007; Wetherby, Watt, Morgan, & Shumway, 2007; Zwaigenbaum et al., 2005). Second, infants later diagnosed with ASD show higher levels of repetitive behaviours



(Chawarska et al., 2014; Wolff et al., 2014). Finally, studies demonstrate delays in both receptive and expressive language between 12 and 24 months (Barbaro & Dissanayake, 2012; Mitchell et al., 2006; Ozonoff et al., 2014; Zwaigenbaum et al., 2005).

The aetiology of ASD is complex and both environmental and genetic influences need to be considered in the emergence (onset) and development (course) of the ASD phenotype (Mandy & Lai, 2016). Tick, Bolton, Happé, Rutter, and Rijdsdijk (2016) point out that the heritability of ASD is estimated around 64-91%, depending on whether or not subclinical cases of ASD (i.e., the broader autism phenotype; BAP) are included. However, for at least 70% of individuals with ASD the genetic aetiology is unknown and there are as many as 400-1,000 candidate genes linked to ASD (Masi, DeMayo, Glozier, & Guastella, 2017). Because genetic factors cannot explain the full ASD phenotype, environmental factors and the gene-environment interaction should be considered as well, especially during critical periods of development when brain plasticity is high (Dennis et al., 2013; Elsabbagh & Johnson, 2010; Inguaggiato, Sgandurra, & Cioni, 2017). Different combinations of genetic and environmental factors can result in different ASD phenotypes (Elsabbagh & Johnson, 2010). Several potential environmental risk (e.g., hypoxia, maternal/paternal age) and protective (e.g., prenatal folate) factors have been identified, including the early social environment (Inguaggiato et al., 2017; Mandy & Lai, 2016).

This dissertation will focus on the younger siblings of children with ASD. More specifically, we focus on characteristics of the early social environment (i.e., parents, parent-child interaction, sibling interaction) and the association with the developmental trajectories of these younger siblings. In what follows, characteristics of both parents and younger siblings of children with ASD as well as the parent-child and sibling interaction are discussed to illustrate how the early social environment of younger siblings of children with ASD differs from typically developing children and how this might impact on their development. In addition, we elaborate upon the two coding methods most frequently used to describe these early social interactions (i.e., global rating vs. frequency coding).

## YOUNGER SIBLINGS OF CHILDREN WITH ASD

The prevalence of ASD is significantly higher in first-degree relatives of children with ASD (Ozonoff et al., 2011; Wheelwright, Auyeung, Allison, & Baron-Cohen, 2010). More specifically, the general recurrence rate of ASD in younger biological siblings of children with ASD (*hereafter* high-risk siblings; HR-sibs) is 18.7%, with an even higher recurrence rate in males (25.9%) and multiplex families (32.2%) (Ozonoff et al., 2011). Of the HR-sibs who do not go on to meet the criteria for ASD, there is a substantial proportion that shows mild but qualitatively similar characteristics of ASD, also referred to as the ‘broader autism phenotype (BAP)’ (Bailey, Palferman, Heavey, & Le Couteur, 1998; Georgiades et al., 2013; Sucksmith, Roth, & Hoekstra, 2011).

During the past decades, the BAP has been investigated by means of observational studies, performance measures (e.g., cognition), and neuroimaging techniques, revealing a broad range of traits possibly characteristic for the BAP (Sucksmith et al., 2011). The BAP in HR-sibs includes language-related difficulties, impairments in reciprocal social interaction (e.g., joint attention, eye contact, response to and initiation of social interaction, gestures), and to some extent repetitive or stereotyped behaviours (Sucksmith et al., 2011). Unfortunately, studies do not always distinguish between HR-sibs with or without ASD. To determine whether particular impairments are part of the BAP or the full ASD phenotype, it is necessary to consider HR-sibs with and without ASD as separate groups. Developmental trajectories of HR-sibs without ASD are more frequently characterised by early deficits in language development, cognitive functioning, quality of social approaches, sharing affect, and social-communicative functioning (Brian et al., 2014; Georgiades et al., 2013; Hudry et al., 2014; Landa & Holman, 2007; Toth, Dawson, Meltzoff, Greenson, & Fein, 2007), which provides evidence for the presence of BAP features in young HR-sibs irrespective of a later ASD diagnosis.

Prospective longitudinal studies of HR-sibs have led to important insights into the early behavioural markers of ASD. Given their increased risk of developing ASD, following HR-sibs from the first year of life through (at least) 36 months provides researchers the opportunity to explore the ASD phenotype as it emerges during the first years of life (Szatmari et al., 2016). In addition, the findings on HR-sibs without ASD

emphasise the need to monitor and possibly support HR-sibs even when they do not go on to meet all ASD criteria. As stated by Szatmari et al. (2016):

A closer investigation of the individual and family factors that are associated with each sibling's position on the broader autism phenotype–ASD boundary is needed. This may enhance our efforts to identify the potential mechanisms involved in determining the developmental pathways and outcomes of all infant siblings of probands with ASD (p. 183).

### **PARENTS OF CHILDREN WITH ASD AND THE EARLY SOCIAL ENVIRONMENT**

Parents of children with ASD, both mothers and fathers, are more likely to show BAP features compared to the general population, with prevalence rates ranging between 14-23% for parents of children with ASD and between 5-9% for control parents (Sasson et al., 2013). BAP characteristics in parents include difficulties in the pragmatic use of language, fewer friendships, a lower preference for social activities and behaviours, alexithymia, and rigid/perfectionistic traits or special interest patterns (Sucksmith et al., 2011). In addition to the possible expression of BAP or ASD in parents, raising a child with ASD can be challenging for parents. A recent review of the impact of ASD on parents and families reports lower feelings of parental self-efficacy, more parenting stress or psychological distress, more parental conflict, lower feelings of well-being and more mental health concerns (e.g., depression, anxiety) in parents of children with ASD (Karst & van Hecke, 2012). It should be noted that this partly depends on both parent (e.g., personality) and child (e.g., ASD symptoms, emotional, functional, and behavioural problems) characteristics and that the relation between the child and family environment is transactional and bidirectional (Karst & van Hecke, 2012).

In addition to a genetic susceptibility in HR-sibs, demonstrated by for example the increased recurrence rate of ASD, HR-sibs also face differences in their early social environment. The presence of ASD in one or more children impacts the broader family context (e.g., parents' well-being, family functioning; Karst & van Hecke, 2012). Moreover, given the possible presence of ASD/BAP in parents and the presence of ASD in their older sibling, HR-sibs' parent-child and sibling interactions are likely to differ

from those of typically developing children without a sibling with ASD. The combination of these genetic and environmental factors might impact on the (atypical) developmental trajectories of HR-sibs.

### **EARLY SOCIAL INTERACTIONS**

Early social interactions (e.g., with parents or siblings) provide an important learning context and have the potential to influence child development, both in typically developing children and in children (at risk for) ASD (e.g., Clifford & Dissanayake, 2009; Harrist et al., 2014; Russel, 2011). However, given that the social environment of HR-sibs is more frequently characterised by difficulties in parental well-being and family functioning, and that HR-sibs more often show early vulnerabilities in their social-communicative functioning (e.g., Karst & van Hecke, 2012; Szatmari et al., 2016), HR-sibs' social interactions are likely to differ from those of typically developing children. Although the early social environment is unlikely to *cause* atypicalities in the developmental trajectories of HR-sibs, they could influence the manifestation of the ASD phenotype (Mandy & Lai, 2016). Nevertheless, research including the early social experiences of HR-sibs is limited.

The possible influence of the social environment on child development is not unidirectional. Rather, characteristics of the child and social environment influence each other in a dynamic and bidirectional way (Dawson, 2008; Gottlieb, 2007) and the nature and quality of social interactions depend on both interaction partners. First, the possible presence of characteristics of ASD or BAP in the HR-sib could lead to a reduced quantity or quality of social experiences (Dawson, 2008; Mandy & Lai, 2016). Second, whether or not prompted by characteristics or behaviours of the HR-sib, differences in the behaviours of the parent, sibling or peer may influence the relationship with and/or development of HR-sibs. For example, parents' interaction style might be influenced by prior experiences with their child with ASD or by the fact that they experience social-communicative difficulties themselves (i.e., BAP/ASD). Finally, children differ in their susceptibility to influences of the environment, with research suggesting a differential susceptibility in at-risk samples (Baker, Messinger, Lyons, & Grantz, 2010; Russel, 2011). Combined, altered social interactions could influence the development of HR-sibs (Baker

et al., 2010; Mandy & Lai, 2016). Consequently, the study of early social interactions could shed light on the early behavioural manifestations of ASD or BAP in HR-sibs and possible risk or protective factors in the development of ASD/BAP characteristics.

### **Parent-child interactions**

The parent-child relationship is one of the most important influences in child development (Russel, 2011). Research highlights the role of parent-child interactions for children's adjustment and the development of for example social competence and executive functioning (Feldman, 2010; Feldman, Bamberger, & Kanat-Maymon, 2013; Houck & LeCuyer-Maus, 2002; Lucassen et al., 2015; Russel, 2011; van der Sluis, van Steensel, & Bögels, 2015). Different studies have also emphasised the beneficial influence of positive parenting behaviours (e.g., responsiveness) on the development of children with ASD (Clifford & Dissanayake, 2009; Haebig, McDuffie, & Weismer, 2013; Ruble, McDuffie, King, & Lorenz, 2008). Furthermore, there is support for the effectiveness of parent-mediated interventions (e.g., improving parental responsiveness or dyadic reciprocity) on the social-communicative and language development of children with ASD (Casenhiser, Shanker, & Stieben, 2013; Green et al., 2010; Oono, Honey, & McConachie, 2013; Siller, Hutman, & Sigman, 2013), emphasising the importance of early parent-child interactions. Given the specific characteristics of children with ASD or HR-sibs (e.g., BAP or ASD) and their parents (e.g., BAP/ASD, stress, internalising problems), parent-child interactions including children with ASD or HR-sibs are likely to differ from parent-child interactions including typically developing children. The parent-child relationship shapes and is being shaped by specific parent and/or child characteristics (Pettit & Arsiwalla, 2008).

First, results from both within-family and between-family studies suggest that parent-child interactions including children with ASD are characterised by more parent directiveness and lower responsiveness compared to parent-child interactions in typically developing children (Doussard-Roosevelt, Joe, Bazhenova, & Porges, 2003; Freeman & Kasari, 2013; Shapiro, Frosch, & Arnold, 1987). On the other hand, parents of children with ASD appear equally sensitive (van Ijzendoorn et al., 2007) and show signs of positive adaptability to their child's shortcomings (e.g., symbol highlighting, more social initiations, stimulating higher levels of play) (Adamson, Bakeman, Deckner, &

Nelson, 2012; Lemanek, Stone, & Fishel, 1993; Meirsschaut, Roeyers, & Warreyn, 2010). Differences in child behaviours include lower responsiveness to the mother, deficits in the use of eye contact, and lower child involvement in children with ASD (Dawson, Hill, Spencer, Galpert, & Watson, 1990; Dolev, Oppenheim, Koren-Karie, & Yirmiya, 2009; Doussard–Roosevelt et al., 2003).

Second, there is some evidence for altered parent-child interactions in HR-sibs. Studies report reduced infant liveliness, attentiveness and positive affect during the parent-child interaction as well as higher parent directiveness and lower parent sensitive responsiveness and synchrony (Wan et al., 2012, 2013; Yirmiya et al., 2006). In contrast, Rozga et al. (2011) did not find differences between parent-child interactions of HR-sibs and parent-child interactions of typically developing children. Further research on the early parent-child interactions of HR-sibs is needed.

### **Sibling interactions**

Sibling relationships are involuntary, lifelong social relations that have the potential to significantly influence children’s development, even beyond the influence of other family relationships (Feinberg, Solmeyer, & McHale, 2012; Howe & Recchia, 2014; Oh, Volling, & Gonzalez, 2015; Stocker, Burwell, & Briggs, 2002). Sibling relationships can be complementary/asymmetrical when one of the siblings dominates the other, or they can be reciprocal/symmetrical when both siblings have an equal position (Harrist et al., 2014). During early and middle childhood, both types of interaction are equally represented, with older siblings as the most dominant sibling during complementary exchanges (Harrist et al., 2014).

Siblings can influence one another directly through their day-to-day social exchanges and experiences growing up together (McHale, Updegraff, & Feinberg, 2016). Sibling relationship dynamics are most commonly explained by the *social learning theory* (Bandura, 1977; Whiteman, McHale, & Soli, 2011). By observing and imitating one another and reinforcing either positive or negative behaviours, siblings shape their mutual relationship. As such, sibling interactions provide important learning opportunities. Considering that children more frequently imitate individuals who are nurturing, warm, and high in status, older siblings can serve as powerful models (Bandura, 1977; Whiteman et al., 2011). Accordingly, studies found that nurturing,

positive sibling interactions can benefit the social-communicative, emotional, cognitive, and behavioural development of both children, whereas negative sibling interactions led to poorer child outcomes (Bank, Patterson, & Reid, 1996; Brody, 2004; Buist & Vermande, 2014; Harrist et al., 2014; Tucker, Updegraff, McHale, & Crouter, 1999). Studies show that conflict in itself is associated with internalising problems and lower academic and social competence, but a balance between conflict and warmth could also strengthen children's anger management and conflict resolution skills (Brody, Stoneman, & Mackinnon, 1982; Buist & Vermande, 2014).

The social learning theory further implies that modelling processes depend on the sibling dyad constellation. More specifically, older and same-gender siblings are more likely to serve as models (Whiteman et al., 2011). In addition, other studies suggest gender differences in sibling relationship quality, with sister pairs characterised as more intimate and harmonious than brother pairs (Buist & Vermande, 2014; Kim, McHale, Wayne Osgood, & Crouter, 2006).

Research on early sibling interactions between children with ASD and HR-sibs is limited. Nevertheless, the presence of ASD in an older sibling as well as possible social-communicative impairments of the HR-sib can have important consequences for the quality (e.g., more negative interactions, differences in role (a)symmetry) and quantity (e.g., fewer social interactions) of sibling interactions. This could in turn influence the HR-sib's learning opportunities. Moreover, in line with the social learning theory, HR-sibs could learn ASD-specific behaviours from their older sibling through modelling or imitation (Bandura, 1977). As expected, the few studies that have been conducted on sibling interactions including children with ASD report significant differences from other control groups (typical development, Down Syndrome). Children with ASD show fewer social approaches towards their sibling and are less responsive, whereas typically developing HR-sibs are less involved and more avoidant during the sibling interaction (Knott, Lewis, & Williams, 1995; Walton & Ingersoll, 2015). In addition, siblings of children with ASD report less conflict compared to siblings of typically developing children (Kaminsky & Dewey, 2001). Given the importance of early social interactions for child development, altered early sibling interactions could influence the development of HR-sibs. However, research is needed to confirm the association between possible perturbations in early sibling interactions and the developmental trajectories of HR-sibs.

## Peer interaction

Similar to parent-child and sibling relationships, peer relationships can benefit child development. Peer interactions are characterised by processes such as conflict, cooperation and friendship that contribute to the emotion-regulation skills, self-esteem, and social development of young children (Cheah, Nelson, & Rubin, 2001; Denham et al., 2011). ASD is characterised by deficits in social interaction, including social relationships with peers (APA, 2013). Accordingly, research shows that children with ASD more frequently struggle with peer relationships and display deficits in social relatedness with same-aged peers (Boyd, Conroy, Asmus, & McKenney, 2011; Kasari, Locke, Gulsrud, & Rotheram-Fuller, 2011). Although it is intuitive to assume that the presence of social-communicative difficulties in HR-sibs are likely to influence their peer relationships, to date there are no studies on the peer relationships of HR-sibs.

Different theories propose different relations between early parent-child or sibling relationships and later peer relationships. The *epigenetic hypothesis* suggests a strong interdependence between parent-child, sibling and peer relationships, with the attachment with the caregiver as basis for all future relationships, whereas the *social network model* assumes a relative independence between interactional systems (Lewis, 2005; Roskam, Meunier, & Stievenart, 2015). Investigating both theories, Roskam et al. (2015) concluded that especially sibling relationships have a consistent and enduring effect on later peer relationships. On the one hand, it is possible that the pattern of interaction between siblings is carried over to the interaction between these siblings and their peers. Siblings who experience more positive sibling interactions might show a more positive interaction style in interaction with a peer (Howe, Ross, & Recchia, 2011; Lockwood, Kitzmann, & Cohen, 2001). On the other hand, siblings who experience negativity or other problems during the sibling interaction might attempt to compensate for these deficits by turning to peers (Howe et al., 2011; Mendelson, Aboud, & Lanthier, 1994; Stocker, 1994). In this way, negative sibling interactions might be associated with positive peer interactions. Further research is needed to evaluate both the quality of HR-sibs' peer relationships as well as the association with later outcome.



### **Coding social interaction**

Even though social interactions are frequently studied in different populations, there is little consensus regarding the coding method best used for coding social interactions and there is usually no motivation for the coding scheme that is selected. Nevertheless, reliable coding methods are essential for empirical research as well as clinical practice (Ruble, 2008).

Both frequency and global coding schemes are frequently used and both methods have advantages and disadvantages. At a *microanalytic* level, a frequency coding method can provide a detailed and numeric-rich picture of social interactions, including data on frequencies and durations of clearly defined behaviours as well as temporal sequences. As such, frequency coding is suited to address questions of quantity, but the coding process can be very time-consuming. At a *macrolevel*, global rating scales involve the evaluation of both qualitative and quantitative attributes of domains of interaction. Moreover, given that social interactions are often influenced by contextual information and that the behaviours of different interaction partners are interdependent, global ratings may be more suited to address questions of relationships or interactions by abstracting and integrating information (Adamson, 2012; Bakeman, 2011; Grotevant, 1989; Wan, 2012). Therefore, the choice of coding scheme depends on the research objectives and researchers should elaborate upon the coding method being used.

## **RESEARCH OBJECTIVES OF THE DISSERTATION**

The main objective of this dissertation was to gain more insight into the early social experiences of younger siblings of children with ASD. Early developmental trajectories of HR-sibs have been relatively well documented (e.g., Hudry et al., 2014; Toth et al., 2007; Zwaigenbaum et al., 2005), but early social interactions are frequently neglected. First, research on the **parent-child interaction** including HR-sibs during the first years of life suggests meaningful differences from parent-child interactions in typical development (Wan et al., 2012, 2013; Yirmiya et al., 2006). However, these differences are not consistently found (Rozga et al., 2011), especially before the age of 12 months. Therefore, we aimed to investigate early parent-child interactions including HR-sibs at the ages of 5 and 10 months. In addition, studies have used both frequency and global

coding schemes to describe the parent-child interaction, but there is insufficient research evidence to make an informed choice between the two methods. For this reason, we evaluated a global and frequency coding scheme in a within-family study of parent-child interactions including children with ASD.

Second, studies on **sibling interactions** including children with ASD are scarce. Although highly valuable, the studies that are available entail some limitations. To start with, the participants mainly include school-aged children, with a very wide age range. Research shows that social-communicative skills already develop at a very young age and social-communicative impairments in children with ASD or BAP clearly emerge during the second year of life (Carpenter, Nagell, Tomasello, Butterworth, & Moore, 1998; Szatmari et al., 2016). Therefore, HR-sibs' sibling interactions should also be evaluated during the first years of life. Next, sibling interactions have predominantly been studied with questionnaires (e.g., Kaminsky & Dewey, 2001; Walton & Ingersoll, 2015), which have disadvantages such as rater bias (Stone, Hoffman, Lewis, & Ousley, 1994). A naturalistic, observational method could provide a broader and more reliable assessment of social interaction (Hastings & Petalas, 2014; Lobato, Miller, Barbour, Hall, & Pezzullo, 1991). Finally, previous studies mainly focus on *typically developing* siblings of children with ASD. However, sibling interactions might be especially compromised when the younger HR-sib experiences social-communicative impairments as well. Thus, we aimed to evaluate early sibling interactions of 18-, 24-, and 36-month-old HR-sibs, irrespective of outcome at 36 months, using a naturalistic, observational method.

Another aim of this dissertation was to test the hypothesis that early social experiences have the potential to influence the developmental trajectories of HR-sibs. Different authors have suggested that early social experiences may have the potential to mediate the association between early vulnerabilities of HR-sibs and later outcome (Dawson, 2008; Mandy & Lai, 2016), but there is little empirical support for the association between early social interactions and the subsequent development of HR-sibs. The only evidence available is provided by Wan et al. (2013), reporting an association between early parent-child interaction characteristics and ASD at 36 months. Consequently, we aimed to evaluate the association between early parent-child interactions (5-10 months) and sibling interactions (24 and 36 months) and the development of HR-sibs at 24 and 36 months.

Table 1 provides an overview of the different samples included in this dissertation.

**Table 1**

*Overview of the research samples*

	Age (months)	<i>n</i>	Objective
Chapter 2	Without ASD: 68.06 (11.56) With ASD: 47.75 (13.02)	16	Cross-sectional analysis of parent-child interactions in a within-family design and the evaluation of coding method
Chapter 3 <sup>a</sup>	5 months: LR: 5.48(.58) HR: 5.33(.66)	47LR 63 HR	Cross-sectional analysis of parent-child interactions in a between-family design at 5 and 10 months (European sample); Predictive value of parent-child interactions for child development at 24 months (Belgian subsample)
	10 months: LR: 10.18(.46) HR: 10.29(.53)	73LR 93HR	
Chapter 4 <sup>ab</sup>	LR: 18.37 (.54) HR: 18.52 (.85)	29 LR 22 HR	Detailed, cross-sectional analysis of sibling interactions
Chapter 5 <sup>ab</sup>	LR: 24.75 (.77) HR: 24.69 (.77)	32 LR 24 HR	Cross-sectional analysis of sibling interactions and the prediction of the social-communicative and language development
Chapter 6 <sup>ab</sup>	LR: 37.39 (.79) HR: 37.77 (1.28)	31 LR 15 HR	Cross-sectional analysis of sibling interactions, the association with peer interactions, and the association with ASD characteristics

*Note.* LR=low-risk, HR=high-risk; <sup>a</sup>These studies are (partly) based on the same sample (for Chapter 3, only the Belgian subsample); <sup>b</sup>age of the youngest sibling

## OVERVIEW OF THE CHAPTERS

In *Chapter 2*, parent-child interactions were observed between mothers and their child with and without ASD (within-family design), using both a frequency coding scheme and global rating scales. The first aim was to compare both coding methods to establish which aspects of the parent-child interaction are best captured by which

coding method. Second, group differences in the parent-child interaction between mothers and their child with and without ASD were evaluated with each coding method to determine the value of each method to detect group differences in parent-child interactions.

In *Chapter 3*, parent-child interactions were evaluated in the first year of life (5 and 10 months) in a European sample of HR-sibs and a typically developing control group (i.e., between-family design). A new global coding scheme was developed based on an extensive literature review. The first aim was to explore potential differences in the parent-child interaction between HR-sibs and a low-risk control group at the ages of 5 and 10 months. Second, the predictive value of parent-child interactions at 5-10 months for child development at 24 months was investigated in the Belgian subsample.

*Chapters 4-6* focus on the evaluation of sibling interactions in a sample of HR-sibs included in the prospective follow-up study of younger siblings of children with ASD at Ghent University. In *Chapter 4*, HR-sibs' sibling interactions are evaluated and compared with a low-risk control group at the age of 18 months. The aim was threefold: 1) to evaluate the characteristics of the sibling interaction in both groups; 2) to explore the presence of role (a)symmetries; 3) to investigate the influence of gender of the oldest sibling on the quality of the sibling relationship.

In *Chapter 5*, we again compared sibling interactions of HR-sibs with sibling interactions in a low-risk control group, but at the age of 24 months. In addition, to evaluate the concept of social learning, we evaluated to what degree HR-sibs imitated their older sibling with ASD. Finally, the association between sibling interactions and the child's social-communicative and language development at 24 months was investigated.

In *Chapter 6*, sibling and peer relationships of HR-sibs at 36 months were evaluated. To get a general idea of sibling interaction characteristics, we first compared sibling interactions from HR-sibs with a low-risk control group. Second, the association between HR-sibs sibling and peer interactions was explored. Finally, the predictive value of ASD characteristics of HR-sibs and children with ASD for the quality of the sibling or peer relationship was investigated.

Finally, *Chapter 7* provides an integrated overview and general discussion of the main findings of this dissertation. In addition, methodological considerations, theoretical and clinical implications, limitations and guidelines for future research are outlined.

It should be noted that this dissertation consists of several research papers, which have been published, are currently under editorial review or have been submitted. Since each of the manuscripts is a self-contained manuscript, which should be able to stand on its own, the text of some of the chapters may partially overlap.

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**PARENT-CHILD INTERACTION IN CHILDREN  
WITH AUTISM SPECTRUM DISORDER AND THEIR  
SIBLINGS: CHOOSING A CODING STRATEGY<sup>1</sup>**

ABSTRACT

The parent-child interaction strongly influences the emotional, behavioural, and cognitive development of young children. The nature of parent-child interactions differs in families with children with autism spectrum disorder (ASD), but research still entails a lot of inconsistencies and there is no consensus as to how these interactions should be coded. We evaluated differences in parent-child interaction between sixteen mothers and their child with ASD ( $M_{age} = 68$  months) and a younger sibling without ASD ( $M_{age} = 48$  months) in a within-family study using global rating scales. Global and frequency codes of the same sample were compared to explore the added value of each coding method and how they could complement each other. We found that mothers used an interaction style characterised by more support and structure, and clearer instructions in interaction with their children without ASD. In addition, global rating results suggested that within the ASD group, mothers may adapt their behaviour to the specific abilities of their child. Regarding the evaluation of coding method, results showed overlap between conceptually similar constructs included in both coding schemes. Although frequency coding clearly has its value, more qualitative aspects of the interaction were better captured by global rating scales and global rating was more time efficient. For this purpose, global ratings might be preferable over frequency coding.

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<sup>1</sup> Based on Bontinck, C., Warreyn, P., Meirsschaut, M., & Roeyers, H. (2017). Parent-child interaction in children with autism spectrum disorder and their siblings: Choosing a coding strategy. *Journal of Child and Family Studies*. <https://doi.org/10.1007/s10826-017-0877-3>

## INTRODUCTION

Persistent deficits in social communication and social interaction constitute one of the core impairments of autism spectrum disorders (ASD) (American Psychiatric Association, 2013). During the first years of life, children who later develop ASD show social-communicative difficulties such as problems in gaze following, joint attention, verbal and non-verbal communication (Bedford et al., 2012; Mitchell et al., 2006; Sullivan et al., 2007). Genetic factors play an important role in the development of ASD characteristics (Elsabbagh & Johnson, 2010), which is also reflected in the higher number of ASD diagnoses and milder/subclinical features of ASD (Broader Autism Phenotype; BAP) in siblings (*hereafter*, high-risk siblings) and parents of children diagnosed with ASD (Ozonoff et al., 2014; Sucksmith, Roth, & Hoekstra, 2011). However, genetic factors cannot fully account for the variability in outcome found in children with ASD or BAP.

In addition to the genetic component, the possibility of a gene-environment interaction should be taken into account. Different combinations of genetic and environmental factors can result in different ASD phenotypes (Elsabbagh & Johnson, 2010). Studies suggest a bidirectional influence between individuals and their social environment including the parent-child relationship (Dawson, 2008; Gottlieb, 2007). First, early social-communicative deficits of high-risk siblings or children with ASD may influence their ability to engage in social interactions with their parents, which could in turn influence the child's social experiences and developmental outcome (Dawson, 2008). Second, parents of children with ASD are more likely to experience social-communicative difficulties themselves. Third, it has been demonstrated that parenting children with ASD involves specific challenges (Estes et al., 2009). Parents of children with ASD report higher levels of stress and psychological distress (depression and anxiety) and lower feelings of self-efficacy and competency as a parent (Davis & Carter, 2008; Estes et al., 2009; Meirsschaut, Roeyers, & Warreyn, 2010).

Vulnerabilities of both children with ASD or high-risk siblings and their parents might result in altered parent-child relationships. There is some evidence from *between-family studies* that parent-child interactions in families with children with ASD differ from parent-child interactions in families without ASD. First, there are differences in the relationship between parents and their child with ASD. Whereas some studies report

more negative parental behaviours (e.g., more directive/controlling behaviour or commanding of play) (Freeman & Kasari, 2013; Shapiro, Frosch, & Arnold, 1987), other studies refer to the positive adaptability of mothers in light of their child's ASD diagnosis (e.g., more symbol highlighting, more social initiations, stimulating higher levels of play, high levels of sensitivity) (Adamson, Bakeman, Deckner, & Nelson, 2012; Lemanek, Stone, & Fishel, 1993; Meirsschaut et al., 2010; van Ijzendoorn et al., 2007). In addition, children with ASD are less contingent to their mothers' approaches or requests, integrate their smiles less frequently with eye contact or show lower responsiveness to mothers' smiles (Dawson, Hill, Spencer, Galpert, & Watson, 1990; Doussard-Roosevelt, Joe, Bazhenova, & Porges, 2003). Second, differences are observed in the interaction between mothers and their other children (i.e., high-risk siblings). Studies show that mothers are more directive and less synchronous and that high-risk siblings are less active than low-risk controls (Wan et al., 2012; Yirmiya et al., 2006). In contrast, Rozga et al. (2011) found no group differences in social-communicative behaviour of the child towards the mother.

To gain a full understanding of social interactions in families with children with ASD, it is important to evaluate the parent-child interaction *within* families. The interaction style of parents of children with ASD could be influenced by prior experiences with their child with ASD. In turn, it is possible that parents generalise this interaction style to their other (typically developing) children. As a result, parental behaviours in interaction with a typically developing child are likely to differ between parents with only typically developing children and parents with child(ren) with ASD. In addition, in between-family designs there is not only a significant difference in child characteristics (i.e., ASD vs. non-ASD), but also variability in parent characteristics and experiences. In a within-family design, the same parent is observed in interaction with both a child with and without ASD and differences in previous experiences as well as variability in parent characteristics are not an issue. Unfortunately, within-family studies in families with children with ASD are scarce. Using a within-family design, Meirsschaut, Warreyn, and Roeyers (2011) found that mothers were more responsive to their non-ASD child compared to their child with ASD, but, contrary to expectations, mothers used comparable amounts of initiatives (both declarative and imperative) towards both children. Similarly, Doussard-Roosevelt et al. (2003) found no differences in the amount

of maternal approaches with children with ASD in comparison with their sibling. However, there were qualitative differences between both groups. Mothers used fewer social verbal approaches and more physical contact in interaction with their children with ASD than with their non-ASD children.

Parent-child interactions need to be considered when evaluating the development of children with ASD and high-risk siblings. First, studies show that the relationship between parents and their child with ASD (e.g., sharing attention, following the child's focus, parental responsiveness) is positively associated with child outcome (Clifford & Dissanayake, 2009; Haebig, McDuffie, & Weismer, 2013; Ruble, McDuffie, King, & Lorenz, 2008). In addition, Wan et al. (2013) investigated the interactions between mothers and high-risk siblings and concluded that dyadic mutuality, infant positive affect and infant attentiveness to the mother at 12 months predicted 3-year ASD outcome. Research including other clinical populations also emphasises the association between the parent-child interaction and children's internalising and externalising problems (van der Sluis, van Steensel, & Bögels, 2015; van Doorn et al., 2016). For example, a higher level of psychological control exerted by the mother was associated with more externalising problems of the child (van Doorn et al., 2016).

Second, interventions often focus on these parent-child interactions to promote child development. A recent review including children with ASD provides evidence for the beneficial effects of parent-delivered interventions on child outcomes such as language development and ASD characteristics (Oono, Honey, & McConachie, 2013). Positive changes in parent-child interaction and parental communication resulted in positive long-term outcomes in children with ASD in terms of social-communicative and language skills, and ASD core symptoms (e.g., reciprocity) (Aldred, Green, & Adams, 2004; Casenhiser, Shanker, & Stieben, 2013; Green et al., 2010; Siller, Hutman, & Sigman, 2013; Siller & Sigman, 2002). Characteristics of the parent-child interaction that were related to positive child functioning include lower levels of controlling and intrusive responses, higher levels of joining the child, enjoyment of the child and support of reciprocity, higher levels of parental responsiveness, and higher parental synchrony.

Effective parent-mediated interventions should be based on a reliable and comprehensive assessment of parent-child interactions (Ruble et al., 2008). Differences in for example the content of the intervention impede the comparison of different



parent-mediated interventions (Oono et al., 2013). In addition, inconsistencies in the existing literature may at least be partly explained by differences in how parent-child interactions are measured. This calls for the development of reliable, valid and feasible measurement tools, both in empirical research and clinical practice (Ruble et al., 2008). To date, there is no consensus as to how parent-child interactions should be measured to achieve the most accurate and reliable assessment.

Two coding methods are frequently used to code social interactions: moment-by-moment frequency coding and global rating scales. Moment-by-moment frequency coding is relatively objective and yields detailed information about frequencies, durations and sequences, but the coding process is time consuming and often requires specific event logging software. Global ratings are more time-efficient, but are based on the subjective judgement of the coder and require extensive training in the interpretation of the coded concepts. Given the interdependence of interaction partners during social interaction, global ratings may be more suited to address questions of relationships or interactions by abstracting and integrating information. In addition, the quality of interactive behaviours (e.g., distinction directive behaviour and scaffolding, appropriateness/sensitivity of parental behaviour) may be better captured by rating scales in comparison to a frequency count. Hence, global ratings might provide a broader view on parent-child interactions in typically developing toddlers and young children with developmental disorders (Adamson et al., 2012). On the other hand, frequency coding allows for sequential analysis enabling the coder to assess specific processes and is more suited to address questions of quantity (e.g., total number of initiations/responses, rate per minute) (Adamson et al., 2012; Bakeman & Quera, 2011; Grotevant & Carlson, 1989; Ruble et al., 2008).

The within-family studies of Meirsschaut et al. (2011) and Doussard-Roosevelt et al. (2003) both used frequency counts to code initiations/approaches and responses during parent-child interaction instead of global rating scales. Consequently, characteristics of the coding method might explain why differences found in previously mentioned between-family studies were not replicated in these within-family studies. Several studies reporting differences in parent-child interaction between typically developing children and children with developmental disorders used global rating scales. Adamson et al. (2012), using 7-point rating scales to code joint engagement, found that parents

used more symbol highlighting in interaction with children with developmental disorders. In the study of Wan et al. (2012), showing higher levels of parental directiveness and lower responsiveness in interaction with high-risk siblings, 7-point rating scales were also used. In addition, studies using global rating scales found that characteristics of the parent-child interaction are associated with child outcome, providing evidence for the value of global rating scales. For example, higher levels of parental responsiveness were associated with better social skills in children with ASD (Mahoney & Perales, 2003; Ruble et al., 2008). In addition, higher maternal sensitivity at 18 months was associated with a growth in expressive language between age 2 and 3 years for children with emergent ASD, but not for children without an ASD diagnosis (Baker, Messinger, Lyons, & Grantz, 2010).

To date, there is insufficient research evidence to make an informed decision on which coding method is best used (frequency vs. global) or on how to measure parent-child interactions in an effective and accurate way. The current study's main aim was to evaluate the usefulness of both coding approaches regarding different aspects of the parent-child interaction. To this end, data from a prior within-family study of Meirsschaut et al. (2011), who used frequency coding, were reanalysed using a selection of the global observation scales of Erickson (Erickson, Sroufe, & Egeland, 1985). First, the association between the global rating scales and frequency codes was evaluated to assess which aspects of the parent-child interaction were captured by both coding schemes and for which aspects one specific method was preferable. Second, the value of each coding scheme to detect differences in parent-child interactions between mothers and her child with and without ASD was evaluated. More specifically, the group differences found by means of the global ratings were reviewed in light of the results previously found by Meirsschaut et al. (2011) based on the frequency codes. Sample characteristics such as age, nonverbal IQ and word comprehension were taken into consideration.

## METHOD

### Participants

The sample comprised 16 mothers with both a child with autism spectrum disorder (ASD) and a child without ASD. In all families the child with ASD was the oldest of the two children and the ASD diagnosis was given after an extensive diagnostic procedure by an experienced multidisciplinary team. Diagnostic status was confirmed using the Social Communication Questionnaire, lifetime version (SCQ; Rutter, Le Couteur, & Lord, 2003; Dutch translation by Warreyn, Raymaekers, & Roeyers, 2004) and the Autism Diagnostic Observation Schedule (ADOS; Lord, Rutter, Dilavore, & Risi, 1999). Participant characteristics are presented in Table 1. The sample was predominantly male: 6 'brother-brother' dyads (ASD – non-ASD), 7 'brother-sister' dyads, 2 'sister-brother' dyads and one 'sister-sister' dyad. A chi-square analysis revealed no significant difference in sex ratio between the ASD and the non-ASD group ( $\chi^2(1)=1.25, p=.458$ ). Nonverbal mental age was assessed using the Snijders-Oomen non-verbal intelligence test 2<sup>1/2</sup>-7 (SON-R; Tellegen, Winkel, Wijnberg-Williams, & Laros, 1998) and word comprehension was measured with the Peabody Picture Vocabulary Test-III-NL (PPVT-III-NL; Dunn & Dunn, 2005). For more information on the participants, we refer to Meirsschaut et al. (2011).

Mothers were on average 33.87 years old ( $SD=4.77$ , range: 27-47) with a social status of 42.88 ( $SD=8.11$ , range: 27-53). Social status was calculated by means of the Hollingshead four factor index and was based on the mother's occupation and education (Hollingshead, 1975). The mothers' social status in the current study reflects an average social status and corresponds with the middle three (stratum 2: machine operators, semiskilled workers; stratum 3: skilled craftsmen, clerical, sales workers; stratum 4: machine operators, semiskilled workers) of the five social strata defined by Hollingshead.

**Table 1***Sample characteristics*

	ASD ( <i>n</i> = 16)		Non-ASD ( <i>n</i> = 16)		
Chronological age					
<i>M</i> ( <i>sd</i> )	68.06	(11.56)	47.75	(13.02)	$F(1,30) = 21.78^{***}$
Range	46-84		29-67		
Sex ratio (M:F)	12:4		9:7		$\chi^2(1) = 1.25$
Social-communicative abilities					
<i>M</i> ( <i>sd</i> )	18.71	(6.23)	4.31	(3.25)	$F(1,25) = 55.37^{***}$
Range	10-29		1-10		
Word comprehension (percentiles)					
<i>M</i> ( <i>sd</i> )	36.69	(37.95)	54.81	(27.73)	$F(1,30) = 2.38$
Range	1-99		5-98		
Nonverbal mental age					
<i>M</i> ( <i>sd</i> )	61.40	(19.25)	50.02	(15.48)	$F(1,30) = 3.39$
Range	30-93		29-74		

*Note.* Chronological and mental age data are reported in months; *ASD* = children with autism spectrum disorder; *Non-ASD* = children without autism spectrum disorder; Social-communicative abilities are obtained with the Social Communication Questionnaire (SCQ), word comprehension with the Peabody Picture Vocabulary Test-III-NL (PPVT-III-NL), and nonverbal mental age with the Snijders-Oomen non-verbal intelligence test 2<sup>1/2</sup>-7 (SON-R).

\*\*\* $p < .001$ .

**Procedure**

The current study is a secondary analysis of a prior study by Meirsschaut et al. (2011), investigating parent-child interactions with a within-family design to evaluate whether mothers differentiate in their interactive behaviour between their child with

and without ASD. In the study of Meirsschaut et al. (2011), parent-child interactions were observed during both a play and task situation. Because the task situation was associated with more active/directive behaviours of the mother and given that the global rating scales also assess structuring behaviours (e.g., quality of instruction, structure and limit setting), the task situation seemed more suited for the purpose of this study. Furthermore, mothers were more responsive during the task situation and differences in child behaviours between contexts were limited.

Mother and child were observed during a short task interaction in which they were instructed to build as many block constructions as possible from a book of construction photos. Mothers were asked to interact with their child as they would at home. During the observation of the mother-child interaction with one child, the other child's word comprehension was tested with the Peabody Picture Vocabulary Test-III-NL (PPVT-III-NL; Dunn & Dunn, 2005). At the end of the session, both children's nonverbal IQ was simultaneously measured with the Snijders-Oomen Niet-verbale Intelligentietest (SON-R; Tellegen et al., 1998). At the beginning of each session, mothers signed an informed consent and completed a sociodemographic form. For more details on the procedure, we refer to Meirsschaut et al. (2011).

All task interactions were recorded digitally and the middle 5 of the 7 minutes of mother-child interaction were coded. Clips were rated blind to all participant information. For both the frequency and global coding scheme, coders were trained in the use of coding scheme using several practice tapes of children not included in the study. For the frequency coding scheme, the first author of Meirsschaut et al. (2011) provided the criterion against which the coder's performance was compared. Training on the practice tapes continued until the coder's degree of agreement with the criterion reached an acceptable standard. To evaluate interrater reliability, approximately 15% of the mother-child interactions were randomly selected for double coding. Kappa was .81 (range .70 - .90) for child's and mother's behaviours, i.e., the social initiatives and responses. Kappa was .74 (range .61 - .85) for agreement in coding of the content (e.g., declarative versus imperative initiative and confirming versus non-confirming response) of child's and mother's behaviour. Regarding the global rating scales, the first author of the present study coded all clips using the Erickson observation scales. A random selection of clips (15%) from the sample of Oosterling et al. (2010) was double coded by

the first author to determine interrater reliability. Cronbach's alpha ranged from .79 (Compliance) to 1.00 (Supportive presence).

### **Measures**

The nonverbal IQ was tested with the Snijders-Oomen Niet-verbale Intelligentietest (SON-R; Tellegen et al., 1998), a nonverbal intelligence test suited for children with ASD or other social-communicative, hearing or language difficulties. The test can be administered without the use of written or spoken language. To assess social-communicative functioning, the Social Communication Questionnaire was used (SCQ; Rutter, Bailey, & Lord, 2003; Warreyn et al., 2004). The SCQ is a screening questionnaire for ASD (parent-report), derived from the Autism Diagnostic Interview-Revised (ADI-R; Rutter, Le Couteur, et al., 2003). Finally, word comprehension was measured with the Peabody Picture Vocabulary Test-III-NL (PPVT-III-NL; Dunn & Dunn, 2005), a reliable measure for word comprehension/vocabulary. The SON-R and PPVT-III-NL were available for all children. Three families did not complete the SCQ. As a result, SCQ scores for 6 children (three ASD and three non-ASD children) were missing. Because missing data were missing completely at random (Little's MCAR test:  $\chi^2(5)=5.498$ ,  $p=.358$ ), participants were not excluded from the sample.

### **Mother-child interaction: Global rating.**

A selection of the widely used Erickson observation scales (Erickson et al., 1985) was used as a global measure of mother-child interaction. In accordance with Oosterling et al. (2010), we included only those scales reflecting interactive behaviour. Scales defined in terms of subjective experiences of the mother or child (confidence, enthusiasm, quality of experience, reliance on mother) were excluded due to their subjective nature and lack of relevance for the current research goals.

Although more comprehensive global coding schemes are available to code parent-child interactions, the Erickson global rating scales were selected for several reasons. First, even though other studies found associations between parental behaviours and child functioning (e.g., Ruble et al., 2008), this was not the main focus of the current study. Therefore, the inclusion of constructs that best predicted subsequent development was not our primary focus. Nevertheless, there is some overlap between

the constructs included in the Erickson global rating scales and constructs predicting child development. For example, the global rating scale supportive presence shows conceptual overlap with other constructs predicting development such as parent responsiveness or parent sensitivity. Second, comprehensive measures for rating parent-child interactions often require extensive training. For the current study, the focus was on a time-efficient coding scheme, with a straightforward and concise training to achieve interrater reliability. Third, for the purpose of comparing the global and frequency codes, the selected global coding scheme should contain both rating scales that show conceptual overlap with the frequency coding scheme of Meirsschaut et al. (2011) and rating scales that are conceptually different. For example, the global rating scale supportive presence relies more on a subjective evaluation of behaviour and is therefore unlikely to be captured by frequency codes. Other global rating scales such as structure and limit setting or compliance are somewhat more quantifiable and show overlap with the frequency codes. Structure and limit setting could be captured by the mother's imperative initiations and compliance could be captured by the child's confirming responses.

Five scales for the mother's social behaviour (supportive presence, respect for child's autonomy, structure and limit setting, quality of instruction and (non-)hostility) and 4 scales for the child's social behaviour ((non-)negativity, (non-)avoidance, compliance and affection) were included. *Supportive presence* refers to positive regard and emotional support the mother expresses to the child. This may occur by acknowledging the child's accomplishments, encouraging the child and other ways of letting the child know that he/she has her support and confidence. *Respect for the child's autonomy* reflects the degree to which the mother acted in a way that recognised and respected the validity of the child's individuality, motives, and perspectives. *Structure and limit setting* expresses how adequately the mother attempted to establish her expectations for the child's behaviour. *Quality of instruction* involves the rating of how well the mother structures the situation so that the child knows what the task objectives are and receives hints of corrections while solving the problems. *(Non-)hostility* reflects the mother's expression of anger, discounting, or rejecting of the child. *(Non-) negativity* refers to the degree to which the child shows anger, dislike or hostility towards the mother. *(Non-)avoidance* is a measure of the child's tendencies or clear

attempts to avoid interacting with the mother. *Compliance* assesses the degree to which the child shows willingness to listen to mother's suggestions and to comply to her requests in a reasonable manner. Finally, *affection* reflects whether there was a substantial period of positive regard and sharing of happy feelings of the child towards the mother. All scales were 7-point rating scales ranging from 1 (very low/maladaptive behaviour) to 7 (very high/adaptive behaviour).

### **Mother-child interaction: Frequency coding.**

The frequency coding scheme included the social initiatives (declarative (i.e., social, sharing interest), imperative (i.e., directive, requesting), or neutral) and social responses (confirming, non-confirming, neutral, or attempt to comply) of both mother and child (see Appendix). Social initiatives and responses could be either verbal or non-verbal. For more details on this coding scheme, we refer to Meirsschaut et al. (2011).

### **Data-analysis**

To answer the *first research question*, correlations between the global ratings and the frequency codes of the parent-child interaction were evaluated. Because assumptions for parametric testing were not met for the global rating scales (i.e., non-normal distribution), Spearman's rank correlation coefficients were calculated. For the *second research question*, it was evaluated to what extent global or frequency codes could detect differences in parent-child interaction between mothers and their child with and without ASD.

We first report a summary of the results based on the frequency codes of Meirsschaut et al. (2011). For all frequency coding variables, assumptions for parametric analyses were met and data were analysed using repeated measures ANOVA's. For the mother's social behaviour, diagnosis of the child (ASD vs. non-ASD) and context (play vs. task) were entered as within-subject factors. For the child's social behaviour, diagnosis of the child was entered as between-subject factor and context as within-subject factor. Because the comparison between coding schemes applies to the task situation, only the results for the task situation are reported in the results section.

Second, the parent-child interaction was compared between groups by means of the global rating scales. Due to a lack of variance, the global rating scales respect for the



child's autonomy, (non-)hostility and (non-)avoidance were excluded from further analyses. In the non-ASD group, all participants obtained the same score on respect for the child's autonomy (score 5), (non-)hostility (score 7) and (non-)avoidance (score 7). In the ASD group, 94% of the participants obtained the same score for (non-)hostility (score 7) and (non-)avoidance (score 7), whereas for respect for the child's autonomy 88% of the participants obtained the same score (score 5). Next, the interaction between mothers and their child with and without ASD was compared using the Wilcoxon signed-rank test for two related samples. Finally, correlations between the global rating scales and child characteristics were evaluated using Spearman's rank correlation coefficients.

## RESULTS

### **Association global and frequency coding**

To evaluate the hypothesised overlap and differences between both global and frequency coding, the frequency coding of Meirsschaut et al. (2011) was correlated with the global rating of the mother-child interaction of the same sample. The results are shown in Table 2. There were significant correlations between the global rating scales for the *mother's* behaviour and the frequency coding scheme. For structure and limit setting, results showed a significant negative correlation with the child's total amount of initiatives. In addition, there was a significant positive correlation between structure and limit setting and the mother's imperative initiatives and between structure and limit setting and the mother's total amount of initiatives. Concerning quality of instruction, there was a positive correlation with the mother's total amount of initiatives. There were no significant correlations for the global scale supportive presence. Correlations between the global rating scales for the *child's* behaviour and frequency coding were less apparent. There were no significant correlations between the child's (non-)negativity or compliance and the frequency codes. For the global rating scale affection, there was a positive correlation with the total amount of child responses.

**Table 2**

*Correlations between the frequency coding of Meirsschaut et al. (2011) and the Erickson's global rating scales.*

	SuppPres	StructLimit	QualInstr	Negativity	Compliance	Affection
C_DeclNit	.28	.18	.22	.08	.04	-.10
C_ImplNit	-.12	-.11	-.34	-.15	-.16	-.17
C_TotalNit	-.03	-.53**	-.33	.03	.18	-.18
C_ComplResp	.18	-.03	.03	.08	.31	.15
C_NoncomplResp	-.05	.16	-.06	-.11	-.21	-.04
C_TotalResp	-.09	-.08	-.17	.00	.15	.36*
M_DeclNit	.30	-.24	.09	-.06	.03	-.15
M_ImplNit	-.20	.35*	-.02	.05	-.09	.08
M_TotalNit	.11	.53**	.50**	.12	-.24	-.04
M_ComplResp	-.12	.16	.11	.10	.19	.07
M_NoncomplResp	.13	-.24	-.18	-.08	-.16	-.01
M_TotalResp	.10	.44*	.30	.19	.09	.10

*Note.* *SuppPres* = Supportive Presence, *StructLimit* = Structure and Limit Setting, *QualInstr* = Quality of Instructions. *C\_DeclNit* = proportion declarative child initiatives, *C\_ImplNit* = proportion imperative child initiatives *C\_TotalNit* = total amount of child initiatives, *C\_ComplResp* = proportion compliant/confirming child responses, *C\_NoncomplResp* = proportion non-compliant/non-confirming child responses, *C\_TotalResp* = total amount of child responses. *M\_DeclNit* = proportion declarative mother initiatives, *M\_ImplNit* = proportion imperative mother initiatives, *M\_TotalNit* = total amount of mother initiatives, *M\_ComplResp* = proportion compliant/confirming mother responses, *M\_NoncomplResp* = proportion non-compliant/non-confirming mother responses, *M\_TotalResp* = total amount of mother responses.

\* $p < 0.05$ , \*\* $p < 0.01$ .

Regarding the group differences explored by means of frequency coding, Meirsschaut et al. (2011) found no interaction effect between context (play vs. task) and diagnosis (ASD vs. non-ASD), meaning that group differences did not differ depending on the context. There was a significant effect of *diagnosis* for the parents' responsiveness (i.e., proportion of a child's social initiatives followed by a reaction of the mother) and the content of the children's initiations. Mothers showed comparable amounts of

initiatives (both declarative and imperative) towards their child with ASD and their non-ASD child, but were more responsive to their non-ASD child compared to their child with ASD. No differences were found with regard to the content of the responses (confirming vs. non-confirming). ASD and non-ASD children used comparable amounts of total initiatives, but ASD children used more imperative initiatives whereas their non-ASD siblings used more declarative initiatives. Total child responsiveness was comparable in both groups.

With respect to the global rating scales, there was a significant group effect for the mother's social behaviour. With their child with ASD, mothers were less supportive, less structuring and showed a lower quality of instruction. There were no significant group differences in the child's social behaviour. The results are presented in Table 3. Furthermore, correlational analyses revealed a few significant intercorrelations between the global rating scales. First, there was a significant positive correlation between the mother's quality of instruction and mother's supportive presence ( $\rho=.43$ ,  $p=.013$ ) and between the mother's quality of instruction and mother's structure and limit setting ( $\rho=.59$ ,  $p<.001$ ). Second, there was a significant negative correlation between mother's quality of instruction and child compliance ( $\rho=-.43$ ,  $p=.015$ ).

**Table 3**

*Means (standard deviations) of the global rating of mother-child interaction.*

	ASD		Non-ASD		Z
Supportive Presence	4.56	(1.63)	5.94	(1.24)	-2.34*
Structure and Limit Setting	4.06	(1.98)	6.00	(1.41)	-2.93**
Quality of Instruction	4.37	(1.41)	5.69	(0.70)	-2.62**
(Non-)negativity	6.50	(1.15)	6.87	(.50)	-1.13
Compliance	6.19	(1.60)	6.12	(1.09)	-.48
Affection	2.00	(1.15)	2.00	(1.32)	-.18

*Note.* Z = test statistic Wilcoxon signed-ranks test; ASD = children with autism spectrum disorder; Non-ASD = children without autism spectrum disorder

\* $p < 0.05$ , \*\* $p < 0.01$ .

Finally, the possible role of child characteristics in the parent-child interaction was evaluated. The global rating scale structure and limit setting correlated negatively with chronological age ( $\rho = -.48$ ,  $p = .005$ ) and social-communicative abilities ( $\rho = -.48$ ,  $p = .011$ ). Also quality of instruction correlated negatively with chronological age ( $\rho = -.62$ ,  $p < .001$ ) and social-communicative abilities ( $\rho = -.50$ ,  $p = .008$ ). Finally, there was a positive correlation between the child's compliance and chronological age ( $\rho = .47$ ,  $p = .007$ ). However, when looking at the ASD and non-ASD group separately, a different pattern emerged. Results are shown in Table 4.

**Table 4**

*Correlations between child characteristics and mother's and child's social behaviour in children with and without ASD.*

	Chronological age	SON-R	SCQ	PPVT-III-NL
ASD				
Supportive Presence	.11	-.14	.22	-.11
Structure and Limit Setting	-.57*	-.21	-.12	-.58*
Quality of Instructions	-.45	-.27	.08	-.52*
(Non-)negativity	.41	.05	.06	.35
Compliance	.57*	.58*	.26	.76**
Affection	.15	.51*	.12	.45
Non-ASD				
Supportive Presence	-.14	.26	.53	.44
Structure and Limit Setting	.16	-.25	-.44	-.18
Quality of Instructions	-.45	-.43	.01	-.19
(Non-)negativity	-.23	.14	-.43	-.37
Compliance	.48	.01	-.38	-.13
Affection	.01	.03	-.02	.21

*Note.* ASD = children with autism spectrum disorder; Non-ASD = children without autism spectrum disorder; SCQ = Social Communication Questionnaire; PPVT-III-NL = Peabody Picture Vocabulary Test-III-NL; SON-R = Snijders-Oomen non-verbal intelligence test 2<sup>1/2</sup>-7.

\* $p < 0.05$ , \*\* $p < 0.01$ .

In the non-ASD group, there were no significant correlations between child characteristics and the global rating of parent child interaction. In the ASD group, there was a significant positive correlation between chronological age and child compliance and a significant negative correlation between chronological age and structure and limit setting. There were also significant positive correlations between nonverbal mental age and compliance and between nonverbal mental age and affection. For the child's word comprehension, results showed significant negative correlations between word comprehension and structure and limit setting and between word comprehension and quality of instruction. In addition, there was a significant positive correlation between word comprehension and child compliance. There were no significant correlations between the global rating scales and the child's social-communicative abilities (SCQ).

## DISCUSSION

Given the importance of early parent-child interactions in stimulating the development of young children with developmental disorders (e.g., Siller & Sigman, 2002, 2008), parents have been playing an important role in early interventions in children with ASD. However, there is a lack of consensus as to how parent-child interactions should be measured (global vs. frequency). Therefore, the current study aimed to evaluate the value of both frequency codes and global rating scales for coding parent-child interactions.

Concerning the first research question, we evaluated the overlap and differences between frequency coding and global rating. In line with our expectations, there were no correlations between supportive presence, which is a more qualitative scale, and the frequency coding scheme. The global scales structure and limit setting and quality of instruction, somewhat more quantifiable, did show correlations with the frequency coding. Mothers providing more structure and better quality instructions had a higher frequency of initiatives. As providing structure and instructions requires parental initiatives, this relationship was evident. In addition, mothers who provided more structure also showed higher levels of imperative initiations. This means that they were more directive, which is in line with the definition of structure and limit setting.

Furthermore, when mothers showed higher levels of structure and limit setting, children were less likely to initiate the interaction. Either higher levels of structure prevented the children from initiating interaction themselves, or lower levels of social initiatives prompted parents to increase the level of structure. Both parent and child will influence each other in a bidirectional interaction. Regarding the second research question, global coding revealed differences in the parent-child interaction. Mothers were more supporting and provided more structure and better instructions in interaction with their children without ASD. Concerning the child scales, there were no differences between children with ASD and their non-ASD sibling.

Next, we evaluated whether child characteristics influenced the parent-child interaction. Because the sample consisted of children with ASD and their younger siblings, the age difference between both children may have influenced the results. More specifically, mothers used more adequate instructions and structure as their children were younger and children were more compliant as they were older. However, when looking at both groups separately, parent-child interaction was only correlated with child characteristics in the ASD group, reducing the likelihood that differences in parent-child interaction are only explained by chronological age. In interaction with their child with ASD, mothers adapted their structure and quality of interaction to the child's age and word comprehension. Furthermore, the negative correlation between social-communicative abilities (SCQ) and structure and limit setting suggests that mothers used more structure in the task situation as their children showed better social-communicative abilities.

Differences in supportive presence between groups were not related to or better explained by the child characteristics included in this study. As stated previously, parents of children with ASD more often show higher levels of parental stress with regard to their child with ASD (Davis & Carter, 2008; Estes et al., 2009; Hoffman, Sweeney, Hodge, Lopez-Wagner, & Looney, 2009), which might explain lower levels of support in interaction with their child with ASD. Accordingly, studies report a negative association between parents' stress levels and closeness to their child or self-perceived involvement in interaction with their child (Hoffman et al., 2009; Osborne & Reed, 2010). Parental stress in the current sample was evaluated in a previous study by Meirsschaut et al. (2010), confirming the relation between higher levels of stress related to parenting

incompetence and role restrictions concerning the child with ASD. However, in our data supportive presence was not related to parental stress, so other mechanisms may be involved. For example, mothers might experience negative cognitions or emotions related to the ASD diagnosis of their child, which could be related to a less supportive interaction style (e.g., Wachtel & Carter, 2008). Studies also show that children with ASD are less sensitive to social rewards, an important aspect of supportive presence (i.e., acknowledging the child's achievements) (Delmonte et al., 2012; Demurie, Roeyers, Baeyens, & Sonuga-Barke, 2011; Kohls et al., 2013). It is possible that mothers less frequently praise or encourage their child with ASD because they learned that their child does not always respond to these social rewards, explaining the lower score on supportive presence.

To conclude, results revealed some overlap between those constructs of frequency and global coding that showed conceptual similarities, but not consistently. In addition, the more qualitative global rating scales (supportive presence, quality of instruction, negativity, affection) were not optimally captured by these frequency codes and certain behaviours such as the mother's number of responses only seem to be reflected in the frequency codes. Furthermore, the combination of these frequency and global coding schemes may provide relevant insights into the dynamics of parent-child interaction such as the correlation between global parental behaviours (e.g., structure and limit setting) and child behaviours (e.g., child initiations). Regarding the group comparisons, the global rating of parent-child interaction revealed several differences between parental behaviour in interaction with a child with ASD and parental behaviour in interaction with a child without ASD. These differences were not evident when frequency codes were used (Meirsschaut et al., 2011). The global rating scales also suggested that, within the ASD group, mothers may adapt their behaviour to the specific abilities (nonverbal mental age and word comprehension) of their child, whereas this is not the case in interaction with their child without ASD. This could reflect real world differences. The benefit of specific parental behaviours might depend on the child's risk status (ASD vs. non-ASD) (Baker et al., 2010), which stresses the need for a coding scheme that is also sensitive for child characteristics. The frequency coding scheme was not able to detect these group differences.

The current study attempted to provide empirical support for the choice of either global rating scales or frequency codes. On the one hand, frequency codes are obviously necessary when there is a need to know absolute frequencies (e.g., number of communicative utterances per minute). If the frequency counts also include time stamps, it is possible to compute sequences of behaviours (e.g., how often is a gesture preceded or followed by eye contact), which is not possible when using global coding scales. On the other hand, global rating scales are more suited for behaviours that require a qualitative evaluation (e.g., affect, appropriateness/sensitivity of parents' responses, scaffolding vs. directive behaviour). These characteristics of parent-child interaction are very difficult to quantify with frequency coding (e.g., how do you count 'warmth' of a relation?). Accordingly, there was no association between the global scale supportive presence and the frequency coding scheme. There was also overlap between both coding schemes used in the present study, indicating that certain behaviours might be captured equally well by both global ratings and frequency codes. For example, structure and limit setting was associated with higher levels of (imperative) initiations.

Thus, depending on the specific research questions and behaviours of interest, a combination of frequency and global coding could be desirable to provide a detailed description of the parent-child interaction. However, this is not always possible due to limited resources. When time and resources are restricted, the choice of coding scheme should be guided by the research questions (absolute frequencies/sequences vs. qualitative evaluation). Given the overlap, certain behaviours (e.g., structuring behaviours) might be captured by both coding methods. Although researchers can opt for either one of the coding methods for these behaviours, the current results suggest that global ratings might be preferable to a frequency coding scheme. Global rating seems more efficient in capturing a variety of information and were able to capture a significant amount of information in a limited period of time. Whereas a narrow selection of relevant constructs has to be made to limit the time constraint in frequency coding, global rating is more time efficient, enabling the coder to include more relevant constructs.



### **Limitations and Future Research**

Certain limitations are worth mentioning. First, only a task situation was included. Although certain global rating scales such as quality of instruction were more applicable to a task situation (Erickson et al., 1985), certain aspects of the parent-child interaction such as parental sensitivity to child signals or child affect might be easier to evaluate in a free play context. A second limitation is that only younger non-ASD siblings were included. To exclude the possibility that the mother's social behaviour is adapted to the child's age rather than the child's diagnostic status, both younger and older non-ASD siblings of children with ASD should be included. However, because very little families consisted of three children with only the middle child having ASD, this was not evaluated in the current study. Third, the sample size was relatively small. For this reason, the power of the study was limited, possibly influencing the found results. In case of a larger sample size, the distribution of the global rating scales would most likely be normal, enabling more elaborate analyses. Also due to the small sample size, it was decided not to correct for multiple testing to prevent a further decrease of power. Finally, given that the study was cross-sectional and only correlational analyses were used, it was not possible to determine causality. Therefore, the possibility that parenting behaviours influence child characteristics in children with ASD cannot be excluded. With regard to word comprehension, this would mean that higher levels of structure and better quality instructions are associated with lower word comprehension. Nevertheless, because parents cannot influence the child's chronological age, there must be at least some adaptation of the parent to the child. For that reason, it is more likely that, in interaction with children with ASD, parents may adapt their parental behaviours to the specific child characteristics. This could in turn have an impact on the developmental trajectories of children with ASD.

Future research should continue to focus on the comparison between frequency coding and global rating scales. Recruiting a larger sample will allow for comparative statistical analyses, which can in turn provide more insights into the added value of each coding method, including the influence of sample characteristics. Second, the current study only focused on a task situation because of the association with more involvement and structuring behaviours of the mother. Given that Meirsschaut et al. (2011) did not find an interaction effect between context and diagnosis and that children with ASD on

average perform equally well or better on block or pattern construction tasks (Charman et al., 2011; Ehlers et al., 1997; Pellicano, Maybery, Durkin, & Maley, 2006), we did not expect the block construction task to have influenced the found group differences. However, the value of a coding strategy might depend on the specific context in which it is used. Therefore, future studies should include both structured and unstructured contexts when evaluating different coding methods. Third, coding schemes should be evaluated on their ability to detect group differences, but also on their value for predicting subsequent developmental outcomes. Hierarchical regression analysis including both coding methods as predictors could shed light on the predictive value of each coding method. Finally, not only the predictive value of each coding method but also the predictive value of behaviours during the parent-child interaction should be further explored. If certain parent or child behaviours during the parent-child interaction are associated with developmental outcomes and differ between groups, these behaviours might be important targets for future interventions. However, more research is needed to confirm possible associations between the parent-child interaction in children with ASD and subsequent child development.

## APPENDIX

*Frequency coding scheme used by Meirsschaut et al. (2011)*

Social initiative	Attempt to interact with someone; Social initiatives are addressed to a person with the intention to get a response from that person and they can be verbal or non-verbal (e.g. pointing, showing, or seeking physical proximity combined with eye contact)
	Declarative Social, to share interest in something with someone (e.g., "I'll feed the doll")
	Imperative Directive, to request something from someone (e.g., "Put that away!")
	Neutral no clear declarative or imperative intention (e.g., "Ok, what's next?")
Social response	Reaction to a social initiative or response and following the preceding attempt within 3 s. Social responses can be verbal and/or non-verbal and are always addressed to the other person.
	Confirming/ Compliant The response confirms the preceding initiative or response (e.g., "Yes, good idea!")
	Non-confirming/ Non-compliant The response denies the preceding initiative or response (e.g., "No, she is not hungry")
	Neutral The response is not clearly confirming or denying (e.g., "mmh")
	Attempt to comply (child scale only) e.g., "I don't know" as a response to mother's question "What colour is this?"
Mothers' responsiveness	The proportion of a child's social initiatives followed by a reaction of the mother (within 3 s)

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**THE QUALITY OF THE PARENT-CHILD  
INTERACTION IN INFANTS AT RISK FOR  
AUTISM SPECTRUM DISORDER AND THE  
ASSOCIATION WITH DEVELOPMENT  
AT 24 MONTHS****ABSTRACT**

Differences in the developmental trajectories of younger siblings of children with ASD (i.e., high-risk siblings; HR-sibs) are already visible during the first years of life. Even though research in typically developing children has provided evidence for the association between parent-child interactions and child development, this has barely been studied in HR-sibs. Through bidirectional processes, parent-child interactions could impact on the atypical developmental trajectories of HR-sibs. With a newly developed, comprehensive coding scheme, this study first aimed to evaluate early parent-child interactions of HR-sibs as compared to siblings of typically developing children (i.e., low-risk siblings; LR-sibs) at the ages of 5 and 10 months. Second, the association between these early interactions and child development at 24 months was explored. Results revealed no significant differences between the parent-child interaction of HR- and LR-sibs. We did, however, find an association with development at 24 months. At 5 months, negative affect of the HR-sib predicted better gross motor skills at 24 months. In addition, parent sensitive responsiveness predicted lower receptive language scores in HR-sibs. At this age, there were no significant results for the LR group. At 10 months, child negative affect and positive parenting behaviours predicted better language scores in LR-sibs. There were no significant associations in the HR group. These findings confirm the positive association between parenting behaviours and language development in typically developing children. The finding that the association with development was different in the LR group than in the HR group suggests that the influence of parent-child interactions on development might be different in high-risk populations.

## INTRODUCTION

Infants with autism spectrum disorder (ASD) show increased difficulties in verbal and nonverbal social communication (e.g., eye contact, gestures, social engagement, imitation) as well as higher levels of repetitive behaviours (Chawarska et al., 2014; Wolff et al., 2014; Zwaigenbaum et al., 2005). Studies have also provided important insights into the developmental trajectories of siblings of children with autism spectrum disorder (ASD) (*hereafter* high-risk siblings; HR-sibs). During the first years of life, HR-sibs more frequently show atypicalities in different developmental domains, with a developmental deceleration starting around 12 months (Rogers, 2009). This includes a higher recurrence rate of ASD and the broader autism phenotype (BAP) compared to the general population as well as other behavioural features such as language delays, cognitive deficits, and internalising or externalising problems (Brian et al., 2014; Gamliel, Yirmiya, & Sigman, 2007; Hudry et al., 2014; Rogers, 2009; Szatmari et al., 2016; Toth, Dawson, Meltzoff, Greenson, & Fein, 2007). One possible area of interest often neglected in studies including HR-sibs is the early social environment. Although early social interactions are unlikely to *cause* atypical developmental trajectories, they could influence their manifestation (Mandy & Lai, 2016).

First, child characteristics may influence the social environment. Difficulties in social interaction and social interest/engagement can result in fewer exchanges with social interaction partners and lower levels of social input (Dawson, 2008; Mandy & Lai, 2016). In addition, specific child behaviours could trigger different parenting styles. Studies including parent-child interactions in children with ASD or HR-sibs found evidence for lower child involvement and/or lower child responsiveness (Dolev, Oppenheim, Koren-Karie, & Yirmiya, 2009; Wan et al., 2013). Lower child involvement during social interaction gives parents fewer leads to respond to and signals lacking social quality are more easily missed (Hudry et al., 2013; van Ijzendoorn et al., 2007). This could in turn elicit different parental behaviours. Dolev et al. (2009), for example, found that child involvement and responsiveness decreased and maternal intrusiveness increased as ASD severity increased. Other studies provide further evidence for a more directive behavioural pattern in parents of children with ASD (e.g., Freeman & Kasari, 2013;

Lemanek, Stone, & Fishel, 1993; Wan et al., 2012), which could reflect an attempt of parents to compensate for the child's limitations.

Second, the social environment, including the interaction with parents or caregivers, dynamically shapes an individual's (social-communicative) development (Mandy & Lai, 2016; Pettit & Arsiwalla, 2008). In typical development, positive parenting characteristics such as warmth, responding appropriately and quickly to the child's signals, and mutuality can positively influence child socialisation (Russel, 2011), emphasising the protective potential of parent-child interactions. Consequently, early parent-child interactions could play an important role in maximising beneficial outcomes among HR-sibs (Baker, Messinger, Lyons, & Grantz, 2010; Dawson, 2008). However, impoverished parent-child interactions or parent-child interactions characterised by higher levels of directive behaviour/intrusiveness could interact with HR-sibs' early susceptibility, which, in turn, may contribute to HR-sibs' atypical developmental trajectories (Dawson, 2008; Mandy & Lai, 2016). Wan et al. (2012), for example, suggest that increased parent directiveness is associated with reduced developmental progress in infants. On the other hand, enriched early social interactions, characterised by sensitivity and warmth, could promote positive developmental outcomes (Baker et al., 2010).

The child and social environment operate in transaction with each other and child characteristics also determine to what extent the social environment influences child behaviour. Children differ in their responsiveness to socialisation and early behavioural atypicalities can influence the child's reactivity to the environment (Russel, 2011). Theories propose a differential susceptibility to the environment in at-risk samples, either in a positive (i.e., parenting fostering child competence) or negative (i.e., parenting exacerbating child risk) way (Baker et al., 2010). In addition, the child's engagement during the parent-child interaction determines to what extent children learn from social interaction (Siller & Sigman, 2002). Accordingly, Tomasello and Farrar (1986) report that *joint* engagement was predictive for language learning. Feldman et al. (2013) found that reciprocity during the parent-child interaction was associated with lower levels of aggression and higher social competence in preschool. Moreover, reciprocity during the father-child interaction was associated with a better capacity to handle conflict whereas mother-child reciprocity contributed to the child's dialogical skills. This suggests that the deficits in social relatedness specific to ASD might limit the

extent to which children with ASD or HR-sibs can benefit from positive parenting (Baker et al., 2010; van Ijzendoorn et al., 2007), and there is evidence suggesting lower levels of dyadic mutuality or synchrony during parent-child interactions with HR-sibs in general (Yirmiya et al., 2006) or HR-sibs with ASD (Wan et al., 2013).

The effectiveness of parent-mediated interventions in children with ASD provides further evidence for the role of the social environment. For example, the increase in responsive parental behaviours or parental synchrony during the parent-child interaction leads to gains in the child's language skills (Oono, Honey, & McConachie, 2013; Siller, Hutman, & Sigman, 2013). Unfortunately, less is known about the role of parenting and parent-child interactions in the (atypical) developmental trajectories of HR-sibs and studies investigating the parent-child interaction in HR-sibs are limited. Wan and colleagues evaluated the parent-child interaction at 6-10 and 12-15 months (Wan et al., 2012, 2013). At 6-10 months, HR infants showed lower levels of liveliness and their parents showed more directive behaviours. At 12-15 months, HR infants were less attentive to their parents and showed lower levels of positive affect whereas their parent showed more directive behaviours. In addition, dyadic mutuality was lower in the HR group. Finally, infant attentiveness, infant positive affect, and dyadic mutuality at 12 months predicted ASD at 3 years. Rozga et al. (2011) on the other hand did not find any significant differences in looks, smiles, and vocalisations of the child directed to the mother between HR infants with ASD, HR infants without ASD, and typically developing low-risk (LR) infants. Lastly, at the age of 4 months, Yirmiya et al. (2006) found lower maternal synchrony during infant-led interactions of high-risk siblings.

In sum, differences in the quality of the parent-child interaction including HR-sibs are limited to subtle differences in parent behaviours during the first year of life. From 12 months onwards, differences in parent and child behaviours as well as in dyadic mutuality seem more pronounced. Although Wan et al. (2013) investigated the predictive value of the parent-child interaction for later ASD, the association with other developmental outcomes has not yet been evaluated in HR-sibs. Given their risk status, HR-sibs might be especially vulnerable to an altered or impoverished social environment (Wan et al., 2012). Therefore, the association with general developmental outcomes should be considered in further research.

## Present study

In the current study, parent-infant interactions were evaluated across different European sites. For this purpose, a new coding scheme was developed based on recent literature on parent-child interaction and its role for subsequent development. This global coding scheme includes both child scales (i.e., initiations, attentiveness, sharing affect, positive affect, absence of negative affect), parent scales (i.e., sensitive responsiveness, absence of negative control, scaffolding, positive affect, absence of negative affect), and the scale 'dyadic reciprocity'.

First, the parent-child interaction was compared between HR-sibs and a low-risk control group at the ages of 5 and 10 months. Research on early ASD suggests that differences between HR and LR siblings only become clear around the age of 12 months (Szatmari et al., 2016). In addition, existing studies did not find clear differences in child behaviour during the first year of life (Rozga et al., 2011; Wan et al., 2012; Yirmiya et al., 2006). Regarding child affect, studies report lower levels of positive affect (smiling, laughing) and higher levels of negative affect or distress in HR infants, with some evidence of reduced positive affect in the first year of life (Clifford, Hudry, Elsabbagh, Charman, & Johnson, 2013; Garon et al., 2009; Zwaigenbaum et al., 2005). Based on these findings, we did not expect to find clear differences in child behaviour. Nevertheless, given the use of a new and more comprehensive measure, the goal was to explore whether early differences in social approaches/responses or affect between HR- and LR-sibs would become visible during the parent-child interaction. Next, we evaluated early differences in parental behaviours. More specifically, the objective was to investigate whether parents of HR-sibs would be more directive or structuring (i.e., more negative control and scaffolding), compared to parents of LR-sibs (e.g., Wan et al., 2012). Regarding the parent's sensitive behaviours with children with ASD or HR-sibs, previous studies did not report significant differences (e.g., Baker et al., 2010; van Ijzendoorn et al., 2007; Wan et al., 2012). Therefore we did not expect significant group differences in parents' sensitive responsiveness. Finally, we evaluated differences between the HR and LR group in terms of dyadic reciprocity. Given that findings of previous studies on synchrony or mutuality report both similarities and differences (in favour of the LR group) during the first year of life (Wan et al., 2012; Yirmiya et al., 2006), we did not have any specific hypotheses.

Second, we aimed to evaluate to what extent parent-child interaction characteristics predicted development at 24 months. Child development was measured using the Mullen Scales of Early Learning (Mullen, 1995), which includes a broad range of developmental domains relevant for infants with or at high risk for ASD (Landa & Garrett-Mayer, 2006). More specifically, the goal was to evaluate the association between the parent-child interaction and the five developmental domains (gross/fine motor skills, visual perception, receptive/expressive language). In addition, because at-risk populations might show a different susceptibility to effects of the social environment, associations were evaluated for each group (HR vs. LR) separately. Given the lack of previous research, no specific predictions were made. However, Baker et al. (2010) found that parental sensitivity predicted better expressive language development for children later diagnosed with ASD, but not for children without ASD.

## METHOD

### Participants

Participants were recruited through four European research labs, all part of the European Babysibs Autism Research Network (Eurosibs) and belonging to the following universities: Ghent University (Belgium), Birkbeck University of London (U.K.), Radboud University Nijmegen Medical Centre/University Medical Centre Utrecht (The Netherlands), and Karolinska Institutet (Sweden).

At 5 months the sample included 110 infants and their parent (108 mothers, 2 fathers) from Belgium, The Netherlands and the U.K., of which 63 infant siblings of children with a formal diagnosis of ASD (HR group) and 47 infant siblings of typically developing children (LR group). At 10 months the sample consisted of 166 infants and their parent (73 LR-sibs, 93 HR-sibs; 156 mothers, 10 fathers) from all four sites. The HR and LR group were gender- and age-matched as much as possible. Sample characteristics are shown in Table 1. Informed consents were obtained from the parents of all participants.

Inclusion criteria for the HR group included the presence of at least one full sibling with ASD (community clinical diagnosis). Participants were excluded if the manifestation

of ASD symptoms was related to the presence of a genetic syndrome. Infants in the LR group had at least one older, typically developing sibling (based on parent-report). Infants were excluded in case of first-degree relatives with an ASD diagnosis or if parents had ASD-specific concerns about their child. Information regarding genetic or medical conditions was obtained via parent-report.

**Table 1**

*Sample characteristics*

	5 months			10 months		
	LR ( <i>n</i> =47)	HR ( <i>n</i> =63)		LR ( <i>n</i> =73)	HR ( <i>n</i> =93)	
<b>Age</b>	5.48(.58)	5.33(.66)	$F(1,108)=1.687$	10.18(.46)	10.29(.53)	$F(1,164)=1.812$
<b>Sex ratio (M:F)</b>	27:20	32:31	$\chi^2(1)=.479$	37:36	47:46	$\chi^2(1)=.000$

*Note.* Age: Chronological age (mean (standard deviation)), reported in months; LR=low-risk, HR=high-risk

**Procedure**

The current study is part of a prospective follow-up study of HR-sibs and a LR control group. Children were followed from 5 to 36 months, including assessments at 5, 10, 14, 24, and 36 months. Results from the parent-child interaction at 5 and 10 months are reported for the entire European sample. For practical reasons, the association with the Mullen Scales of Early Learning (MSEL; Mullen, 1995) at 24 months is reported for the Belgian sample only. Data from 36 months, including the diagnostic status of HR-sibs, were not yet available.

**Mullen Scales of Early Learning.** The MSEL (Mullen, 1995) is a comprehensive measure of five developmental domains for infants and preschool children (0-68 months): gross motor, fine motor, visual reception, receptive language, and expressive language. Overall cognitive ability is represented by the Early Learning Composite (ELC; standard score), derived from the *T* scores of the four cognitive scales (fine motor, visual reception, receptive/expressive language). Each scale or domain is distinct and has sufficient specificity to be interpretable. The MSEL has demonstrated good internal consistency and test-retest stability (Mullen 1995).

**Parent-child interaction.** Parent and child were observed during a ten-minute period of unstructured play on a floor mat when the children were 5 and 10 months of

age. Parent-child interactions were videotaped and the first five minutes of each clip were selected for coding. A standardised set of age-appropriate toys was made available to the parents and parents were asked to play with their child as they typically would at home.

The coding scheme used in this study (i.e., Parent-Infant/Toddler Coding of Interaction [PInTCI]; Bontinck, Pijl, Warreyn, & Oosterling, 2015) was developed after an extensive literature review, focusing on characteristics of parent-child interaction that predicted subsequent child development in autism research. The coding scheme consists of 11 7-point global rating scales: 5 child scales (initiations, attentiveness, shared affect, positive affect, absence of negative affect), 5 parent scales (sensitive responsiveness, absence of negative control, scaffolding, positive affect, absence of negative affect) and 1 dyadic scale (dyadic reciprocity). Scale definitions are presented in Table 2. A score of 1 consistently reflects maladjusted/negative behaviours, while a score of 7 reflects more adaptive behaviours. In order to be applicable in different age groups across early development (5-36 months), scales from existing measures were adapted. The coding scheme was based on a combination and adaptation of the following measures of interaction: Coding Interactive Behavior (CIB; Feldman, 1998), Manchester Assessment of Caregiver-Infant Interaction (MACI; Wan et al., 2012, 2013), coding scheme for the Communication Play Protocol (Adamson & Bakeman, 1999; Adamson et al., 2012), Erickson coding scales (Erickson et al., 1985), Maternal Behavior Rating Scale (Mahoney & Perales, 2003; Mahoney et al., 1986), Siller's and Sigman's coding scheme (Siller & Sigman, 2002), Dyadic Communication Measure for Autism (DCMA; Aldred et al., 2004), Social Interaction Rating Scale (SIRS; Ruble et al., 2008), infant coding scales (Clifford & Dissanayake, 2009), scaffolding scales (Baker et al., 2007; Dieterich et al., 2006; Hoffman et al., 2006), and coding maternal response behaviors (Flynn & Masur, 2007; Landry et al., 2006; Lloyd & Masur, 2014).

**Coding procedure.** At each research site, clips were independently rated by trained coders blind to the infant's risk status (2 coders from Belgium, 2 coders from the Netherlands, 1 coder from the U.K., 2 coders from Sweden). Each coder followed an intensive group training followed by individual feedback to ensure sufficient interrater reliability. To determine the interrater reliability (IRR), approximately 15% of the clips were randomly selected and coded by all seven coders.



**Table 2***Definition global rating scales PInTCI*

Child initiations	Amount and quality of social initiations directed to the parent, either verbal (e.g., vocalising, babbling, talking) or non-verbal (e.g., showing, giving, pointing).
Child attentiveness	Amount and quality of: 1) The child's spontaneous orientation to the parent, not triggered by parental behaviours; 2) The child's responsiveness to parental behaviours, either positive or negative.
Child shared affect	The degree to which children share and direct their affective state with/to the parent. Affect can be either positive or negative, but must be shared with the parent.
Child positive affect	Examples: Relaxed body language, smiles, laughs, giggles, happiness, enthusiasm, elevation, excitement, positive vocalisations, positive facial expressions.
Child negative affect	Examples: Body language (e.g., tension, discomfort, restlessness), showing anger, dislike or hostility, negative facial expressions, negative vocalisations, negative bodily gestures (e.g., distress, rejection).
Parent sensitive responsiveness	1) The accuracy of identification and interpretation of the child's cues/needs, and 2) the timing and appropriateness of the parent's responses to these cues.
Parent absence of negative control	The degree to which the interaction is determined by the child's preferences and the child's focus of attention. A low score on this scale means that parents try to control the physical behaviour or attentional focus of their child instead of following the child's lead.
Parent scaffolding	The level of adequately facilitating the child's development and guiding the child's actions so that the child can do and say things that he/she would likely not achieve without guidance and encouragement.
Parent positive affect	Examples: positive tone of voice, enthusiasm, smiles/laughter, happy facial expressions, relaxed body posture, and physical affection towards the child.
Parent negative affect	Examples: negative tone of voice, tightened or angry facial expressions, tense body posture and angry or hostile acts.
Dyadic reciprocity	The amount and quality of engagement, mutuality, cooperation, reciprocity and sharedness between parent and child.

Given the language barrier, all IRR clips included English speaking parents from the U.K. sample. Next, average measure intraclass correlation coefficients (ICC) were calculated. ICC's between .60-.74 reflect good interrater agreement and ICC's between .75-1.00 reflect excellent interrater agreement (Cicchetti, 1994). At 5 months, all but two scales were coded reliably with ICC's ranging from .68 (shared affect) to .95 (child positive affect). Child initiations and parents' absence of negative affect were not coded reliably, most likely caused by too little variation in the scales, and therefore excluded from further analyses. At 10 months, all scales were coded reliably. ICC's ranged between .79 (scaffolding) and .96 (attentiveness).

### **Data-analysis**

The internal consistency of the global coding scheme was determined separately at 5 and 10 months. At 5 months, Cronbach's alpha for the nine rating scales (child initiations and parents' absence of negative affect excluded) was .81 across both groups. At 10 months, Cronbach's alpha was .83 for all eleven global rating scales. To assess the underlying structure of the global rating scales and reduce the number of variables for the regression analyses, a principal component factor analysis was performed. Both at 5 and 10 months, there was a three-factor solution (eigenvalues >1, see Table 3). At 5 months, three factors were produced, explaining 70% of the variance. The first factor consists of two parent scales (scaffolding, sensitive responsiveness), one child scale (attentiveness) and dyadic reciprocity. The second factor included two child scales (positive affect, shared affect) and one parent scale (positive affect). Finally, the third factor included the absence of parent negative control and the absence of child negative affect. At 10 months, the three-factor solution explained 63% of the variance. The first factor included four child scales (initiations, attentiveness, shared affect, positive affect) and dyadic reciprocity. The second factor consisted of three parent scales (sensitive responsiveness, scaffolding, positive affect). Finally, the third factor consisted of the absence of parent and child negative affect, and the absence of parent negative control.

To evaluate the first research question, a multivariate analysis of variance (MANOVA) with diagnostic group (HR vs. LR) and research site (U.K., Sweden, Belgium, The Netherlands) as independent variables was conducted to compare child, parent and dyadic behaviours during the parent-child interaction. To investigate the stability in

parent-child interactions during the first year of life (i.e., from 5 to 10 months), correlations between the parent-child interaction variables at 5 and 10 months were calculated. In addition, two-way repeated measures ANOVA was used with group (HR vs. LR) as between-subjects factor and time (5 and 10 months) as within-subjects factor.

**Table 3**

*Principal Component Analysis structure matrix of the parent-child interaction scales*

	5M			10M		
	Factor 1	Factor 2	Factor 3	Factor 1	Factor 2	Factor 3
C_Initiations	/	/	/	<b>.732</b>	.101	.301
C_Attentiveness	<b>.736</b>	.668	.069	<b>.861</b>	.423	.183
C_Sharing	.267	<b>.881</b>	.061	<b>.830</b>	.222	-.045
C_PosAff	.310	<b>.891</b>	.285	<b>.797</b>	.238	.132
C_NegAff	.267	.097	<b>.748</b>	.069	-.162	<b>.593</b>
P_Sensitivity	<b>.830</b>	.226	.208	.245	<b>.869</b>	.282
P_NegCon	-.028	.155	<b>.766</b>	.271	.309	<b>.658</b>
P_Scaffolding	<b>.880</b>	.258	.045	.295	<b>.817</b>	.057
P_PosAff	.559	<b>.623</b>	.078	.517	<b>.698</b>	.103
P_NegAff	/	/	/	.065	.268	<b>.668</b>
D_DyadicRec	<b>.788</b>	.604	.132	<b>.823</b>	.531	.131

*Note.* PosAff = Positive affect; NegAff = Negative affect; Sensitivity = Sensitive responsiveness; NegCon = Negative control; DyadicRec = Dyadic reciprocity

For the second research question, regression models were tested to evaluate the association between the parent-child interaction at 5 and 10 months, and the child's development at 24 months as measured with the *Mullen Scales of Early Learning* (MSEL). The association with development was only tested in the Belgian sample, which included 39 participants at 5 months (13 LR, 24 HR) and 43 participants at 10 months (18 LR, 25 HR). Information on the children's development was not available for the other sites and therefore not included. Significant intercorrelations between the child, parent, and dyadic scales caused multicollinearity in the regression model. To address the problem of multicollinearity and reduce the number of variables in the regression model, the underlying factors were used to predict the MSEL scores at 24 months. Regression

models were tested for the MSEL domains that significantly correlated with one or more of the PCI factors (5 months: MSEL gross motor and receptive language; 10months: MSEL gross/fine motor, visual perception and receptive/expressive language).

## RESULTS

### Parent-child interaction characteristics

Both at 5 and 10 months, the multivariate result for group was not significant (5m: Wilks'  $\lambda$ =.978,  $F(9,96)$ =.239,  $p$ =.988; 10m: Wilks'  $\lambda$ =.923,  $F(11, 148)$ =1.118,  $p$ =.351), indicating that there were no differences in parent-child interaction between siblings of children with ASD and siblings of typically developing children. Descriptives for both groups are presented in Table 4.

**Table 4**

*Descriptives (mean(standard deviation)) of the global rating scales (HR vs. LR)*

	5 months		10 months	
	LR	HR	LR	HR
C_Initiations	/	/	2.89(1.21)	2.44(0.80)
C_Attentiveness	4.00(1.06)	3.92(0.94)	4.08(1.31)	3.96(1.08)
C_Sharing	2.34(1.43)	2.22(1.40)	2.48(1.41)	2.27(1.15)
C_PosAff	3.23(1.39)	3.13(1.31)	3.81(1.37)	3.60(1.28)
C_NegAff	5.85(1.22)	5.83(1.24)	6.41(1.05)	6.35(0.88)
P_Sensitivity	4.34(1.11)	4.40(1.23)	4.78(1.19)	4.70(1.01)
P_NegCon	4.74(1.15)	4.67(1.06)	4.77(1.07)	4.56(1.13)
P_Scaffolding	4.13(1.03)	4.14(1.09)	4.62(1.10)	4.59(1.03)
P_PosAff	5.11(1.46)	4.87(1.34)	4.97(1.41)	4.75(1.29)
P_NegAff	/	/	6.82(0.48)	6.81(0.56)
D_DyadicRec	3.53(1.06)	3.48(1.12)	3.66(1.12)	3.66(1.04)

*Note.* LR=low-risk, HR=high-risk; PosAff = Positive affect; NegAff = Absence of negative affect; Sensitivity = Sensitive responsiveness; NegCon = Absence of negative control; DyadicRec = Dyadic reciprocity

There was however a significant difference in parent-child interaction between the research sites (5m: Wilks'  $\lambda$ =.585,  $F(18, 192)$ =3.284,  $p$ <.001,  $\eta_p^2$ =.24; 10m: Wilks'  $\lambda$ =.483;

$F(33, 436.74)=3.705, p<.001, \eta_p^2=.22$ ). Descriptives and univariate test results for the comparison between research sites are presented in Table 5. The group\*site interaction was not significant at both time points (5m: Wilks'  $\lambda=.895, F(18,192)=.610, p=.889$ ; 10m: Wilks'  $\lambda=.780, F(33,436.74)=1.167, p=.246$ ).

**Table 5**

*Descriptives (mean(standard deviation)) of the global rating scales (comparison research sites)*

	5 months				
	NL	BE	SW	U.K.	F(2, 104)
C_Attentiveness	3.64(0.99)	4.38(0.92)	/	3.83(0.93)	5.205*
C_Sharing	2.30(1.24)	2.57(1.42)	/	1.98(1.49)	1.881
C_PosAff	3.09(1.33)	3.11(1.43)	/	3.30(1.29)	.188
C_NegAff	5.82(1.10)	5.81(1.47)	/	5.88(1.09)	.045
P_Sensitivity	4.18(0.98)	5.16(1.24)	/	3.80(0.82)	15.463***
P_NegCon	5.00(0.97)	4.54(1.02)	/	4.60(1.24)	1.847
P_Scaffolding	3.94(1.03)	4.49(1.12)	/	3.98(0.97)	2.649
P_PosAff	4.76(1.09)	5.38(1.34)	/	4.78(1.59)	3.442*
D_DyadicRec	3.12(1.14)	3.97(1.01)	/	3.38(0.98)	5.444**
	10 months				
	NL	BE	SW	U.K.	F(3,158)
C_Initiations	2.43(0.85)	3.14(1.26)	2.53(0.81)	2.42(0.96)	5.979**
C_Attentiveness	3.79(1.16)	4.47(1.12)	4.09(1.22)	3.61(1.09)	4.404**
C_Sharing	2.23(1.20)	3.00(1.51)	2.11(0.96)	2.03(1.14)	5.977**
C_PosAff	3.89(1.15)	4.40(1.35)	3.22(1.17)	3.10(1.25)	11.061***
C_NegAff	6.32(1.18)	6.26(0.98)	6.44(0.84)	6.55(0.68)	.858
P_Sensitivity	4.74(1.03)	5.14(1.06)	4.40(1.12)	4.65(1.05)	3.819*
P_NegCon	4.85(1.16)	4.81(1.03)	4.29(1.16)	4.65(0.95)	2.275
P_Scaffolding	4.34(1.01)	4.86(1.23)	4.62(0.98)	4.61(0.95)	2.091
P_PosAff	4.57(1.23)	5.70(1.15)	4.78(1.28)	4.19(1.35)	10.902***
P_NegAff	6.94(0.32)	6.91(0.29)	6.56(0.72)	6.87(0.56)	5.359**
D_DyadicRec	3.38(0.95)	4.28(1.12)	3.58(0.99)	3.32(0.98)	8.810***

Note. \* $p<.05$ , \*\* $p<.01$ , \*\*\* $p<.001$ ; NL = The Netherlands, BE = Belgium, U.K. = United Kingdom, SW = Sweden; PosAff = Positive affect; NegAff = Negative affect; Sensitivity = Sensitive responsiveness; NegCon = Negative control; DyadicRec = Dyadic reciprocity

At 5 months, research sites significantly differed in terms of child attentiveness, parent sensitive responsiveness, parent positive affect, and dyadic reciprocity. Tukey post hoc tests revealed that for child attentiveness, parent sensitive responsiveness and dyadic reciprocity, the Belgian sample received higher scores compared to the Netherlands and the U.K. For parent positive affect, pairwise comparisons were not significant. At 10 months, follow-up univariate tests revealed significant differences between sites in child initiations, child attentiveness, child shared affect, child positive affect, parent sensitive responsiveness, parent positive affect, absence of parent negative affect, and dyadic reciprocity. Tukey post hoc tests indicated that the scores of the Belgian sample were higher than the scores of the Netherlands, the U.K. and Sweden for child initiations, child sharing of affect, parent positive affect, and dyadic reciprocity. For parents' absence of negative affect, the Swedish scores were significantly lower than the Netherlands, Belgium and the U.K. Child positive affect differed between Belgium and Sweden and between Sweden and the Netherlands (i.e., lower scores in the Swedish sample), whereas child attentiveness differed between Belgium and the Netherlands and between Belgium and the U.K (i.e., higher scores in the Belgian sample). Finally, parent sensitive responsiveness only differed between Belgium and Sweden, with higher scores in the Belgian sample.

Next, the stability in parent-child interaction from 5 to 10 months was evaluated. Correlation analysis between 5 and 10 months for each parent-child interaction scale only revealed two significant correlations: 1) a significant positive correlation between absence of parent negative control at 5 and 10 months ( $r=.41$ ,  $p=.016$ ), and 2) a significant positive correlation between parent positive affect at 5 and 10 months ( $r=.53$ ,  $p=.001$ ). The repeated measures ANOVA further showed a significant effect for *time* (Wilks'  $\lambda=.629$ ,  $F(9,81)=5.308$ ,  $p<.001$ ,  $\eta^2=.37$ ), indicating a significant difference in parent-child interaction between the observations at 5 and 10 months in both groups. There was no significant effect for *group* (Wilks'  $\lambda=.965$ ,  $F(9,81)=.328$ ,  $p=.963$ ), nor a significant interaction effect (Wilks'  $\lambda=.985$ ,  $F(9,81)=.136$ ,  $p=.999$ ). Univariate tests revealed that four parent-child interaction variables changed over time. First, positive affect was significantly higher ( $F(1,89)=14.947$ ,  $p<.001$ ) at 10 months ( $mean=3.89$ ) than at 5 months ( $mean=3.20$ ). Second, the absence of negative affect also increased from 5 to 10 months ( $mean_{5months}=5.76$ ,  $mean_{10months}=6.33$ ;  $F(1,89)=12.316$ ,  $p=.001$ ). Finally,

both parent sensitive responsiveness ( $mean_{5months}=4.48$ ,  $mean_{10months}=4.96$ ;  $F(1,89)=9.552$ ,  $p=.003$ ) and parent scaffolding ( $mean_{5months}=4.22$ ,  $mean_{10months}=4.56$ ;  $F(1,89)=5.629$ ,  $p=.020$ ) increased from 5 months to 10 months.

### **Association with development**

**Parent-child interaction at 5 months.** In the LR group, there were no significant correlations between the three parent-child interaction (PCI) factors and the child's scores on the MSEL. Consequently, no regression models were tested. In the HR group, there was a significant positive correlation between PCI factor 3 (absence of parent negative control, absence of child negative affect) and MSEL gross motor ( $r=.49$ ,  $p=.024$ ), and a significant negative correlation between PCI factor 1 (parent scaffolding, parent sensitive responsiveness, child attentiveness, dyadic reciprocity) and MSEL receptive language ( $r=-.53$ ,  $p=.013$ ). Thus, two hierarchical regression models were tested for MSEL gross motor and MSEL receptive language. Results show that both models were not significant (see Table 6). However, results show that PCI factor 3 at 5 months significantly predicted gross motor development at 24 months ( $\beta=.533$ ,  $t=2.562$ ,  $p=.020$ ). The combination of lower parental negative control and lower child negative affect was associated with better gross motor skills. In addition, PCI factor 1 negatively predicted receptive language at 24 months ( $\beta=-.506$ ,  $t=-2.461$ ,  $p=.025$ ). Higher levels of parent scaffolding, parent sensitive responsiveness, child attentiveness, and dyadic reciprocity were associated with lower levels of receptive language at 24 months.

**Parent-child interaction at 10 months.** In the LR group, there were significant correlations between the PCI factors and the child's scores on all five of the MSEL domains. In the HR group, the parent-child interaction at 10 months was not associated with the scores on the MSEL at 24 months. Consequently, hierarchical regression models were only tested in the LR group. As shown in Table 6, PCI factors significantly predicted child development at 24 months. It is, however, possible that the association between PCI at 10 months and the MSEL scores at 24 months was better explained by child functioning at 10 months. To this end, correlations between MSEL scores at 10 and 24 months were evaluated. First, fine motor development at 24 months was significantly correlated with gross motor development at 10 months ( $r=.60$ ,  $p=.011$ ).

**Table 6**

*Association between PCI at 5 (HR group) or 10 (LR group) months and MSEL scores at 24 months*

		High-risk group		
		<i>B</i>	<i>SE B</i>	$\beta$
<b>MSEL_GM</b>	<b><math>R^2=.32, F(3,17)=2.722, p=.077</math></b>			
	Factor 1 5m	-.583	.435	-.274
	Factor 2 5m	.201	.362	.113
	Factor 3 5m	1.005	.392	.533*
<b>MSEL_RL</b>	<b><math>R^2=.32, F(3,17)=2.654, p=.082</math></b>			
	Factor 1 5m	-2.812	1.143	-.506*
	Factor 2 5m	-.743	.949	-.159
	Factor 3 5m	-.404	1.029	-.082
		Low-risk group		
		<i>B</i>	<i>SE B</i>	$\beta$
<b>MSEL_GM</b>	<b><math>R^2=.39, F(3,13)=2.710, p=.088</math></b>			
	Factor 1 10m	1.000	.578	.461
	Factor 2 10m	.213	.754	.075
	Factor 3 10m	.969	.706	.301
<b>MSEL_VP</b>	<b><math>R^2=.24, F(3,13)=1.366, p=.297</math></b>			
	Factor 1 10m	.744	.559	.394
	Factor 2 10m	.334	.729	.136
	Factor 3 10m	-.426	.683	-.152
<b>MSEL_FM</b>	<b><math>R^2=.38, F(3,13)=2.678, p=.090</math></b>			
	Factor 1 10m	.690	.443	.415
	Factor 2 10m	-.444	.578	-.206
	Factor 3 10m	1.225	.542	.498*
<b>MSEL_RL</b>	<b><math>R^2=.67, F(3,13)=8.804, p=.002</math></b>			
	Factor 1 10m	.322	.452	.139
	Factor 2 10m	.838	.589	.278
	Factor 3 10m	2.327	.552	.678**
<b>MSEL_EL</b>	<b><math>R^2=.59, F(3,13)=6.225, p=.007</math></b>			
	Factor 1 10m	.233	.442	.114
	Factor 2 10m	1.669	.577	.630*
	Factor 3 10m	.692	.541	.229

*Note.* \* $p<.05$ ; GM=gross motor, VP=visual perception, FM=fine motor, RL=receptive language, EL=expressive language



Second, receptive language at 24 months correlated significantly with receptive language at 10 months ( $r=.485$ ,  $p=.049$ ). Finally, there were marginally significant correlations between expressive language at 24 months and receptive ( $r=.476$ ,  $p=.053$ ) and expressive ( $r=.436$ ,  $p=.081$ ) language at 10 months. These MSEL variables at 10 months were entered in the regression models to determine whether PCI at 10 months still significantly predicted MSEL scores at 24 months when controlling for MSEL scores at 10 months. More specifically, gross motor development at 10 months was entered in the model for fine motor development at 24 months, receptive language at 10 months was entered in the model for receptive language at 24 months, and both receptive and expressive language at 10 months were added in the model for expressive language at 24 months.

The regression model for fine motor development, previously non-significant, became significant after adding gross motor scores at 10 months ( $R^2=.66$ ,  $F(4,12)=5.858$ ,  $p=.007$ ). Both gross motor development ( $\beta=.553$ ,  $t=3.146$ ,  $p=.008$ ) and PCI factor 3 (absence of parental negative control, absence of parent/child negative affect;  $\beta=.510$ ,  $t=3.007$ ,  $p=.011$ ) were significant, positive predictors. The regression model for receptive language remained significant after adding receptive language scores at 10 months ( $R^2=.72$ ,  $F(4,12)=7.697$ ,  $p=.003$ ) with only PCI factor 3 as a significant positive predictor ( $\beta=.665$ ,  $t=4.302$ ,  $p=.001$ ). Finally, the regression model for expressive language remained significant after adding receptive and expressive language scores at 10 months ( $R^2=.66$ ,  $F(5,11)=4.319$ ,  $p=.020$ ) with only PCI factor 2 (parent sensitive responsiveness, parent scaffolding, parent positive affect) as a significant predictor ( $\beta=.689$ ,  $t=2.662$ ,  $p=.022$ ).

## DISCUSSION

This study aimed to evaluate early parent-infant interactions in a European sample of high-risk siblings of children with ASD and a low-risk control group. To this end, a comprehensive coding scheme was developed including parent, child and dyadic scales, based on their association with child development. First, characteristics of the parent-child interaction at 5 and 10 months were evaluated and compared between both

groups. Second, the association with development was explored in a subsample for each group separately.

### **Parent-child interaction characteristics**

In line with our expectations and previous research (Rozga et al., 2011; Wan et al., 2012; Yirmiya et al., 2006), the HR and LR group did not significantly differ on the child scales at 5 or 10 months. Wan et al. (2012) did find a significant difference in infant liveliness, which mainly includes the child's physical activity and motor movements. As stated in their article, infant liveliness was not associated with other parent-child interaction scales and most likely taps a different underlying construct. Although the evaluation of motor movements has its value in the study of emerging ASD, this is not a core characteristic of the parent-child interaction. Therefore, these behaviours were not captured by the coding scheme used in the current study. The lack of differences in child behaviour is also in agreement with previous studies suggesting that differences between high- and low-risk infants only clearly appear by the age of 12 months (e.g., Rogers, 2009). Differences in child behaviour during the parent-child interaction become more pronounced during the second year of life. For example, Wan et al. (2013) found that at the age of 12-15 months, LR-sibs were more attentive to the parent and showed higher levels of positive affect than HR-sibs. In addition, Rozga et al., (2011) did not find significant differences between 6-month-old HR-sibs with ASD (HR-ASD), HR-sibs without ASD (HR-no ASD) and LR-sibs, but by the age of 12 months the HR-ASD group differed from the other two groups exhibiting lower levels of joint attention and requesting behaviours. Thus, the current results further support the finding that social-communicative differences between LR- and HR-sibs only begin to emerge around the child's first birthday.

Concerning the parent scales, Wan et al. (2012) found a significant difference in parent directiveness, with more directive behaviours in HR dyads compared to LR dyads. Despite the conceptual overlap between the scale *caregiver nondirectiveness* of Wan et al. (2012) and *absence of negative control*, these higher levels of parent directiveness were not found in the current study. It is important to consider the distinction between HR-sibs who are later diagnosed with ASD (HR-ASD) and those who are not (HR-no ASD). The sample of Wan et al. (2012) included 14 (31.1%) HR-sibs who met criteria for ASD

(Wan et al., 2013). This proportion is somewhat higher than the recurrence rate reported in other studies (e.g., Ozonoff et al., 2011). Furthermore, results from the follow-up of their sample at 12-15 months showed that the HR-ASD group scored higher in terms of parent directiveness compared to the HR-no ASD group (Wan et al., 2013). Consequently, the differences in parent directiveness at 6 months could be partially due to a higher prevalence of HR-ASD siblings in their sample. The study of Doussard-Roosevelt and colleagues (2003) also provides evidence for early differences in parent-child interactions between HR-ASD siblings and low-risk controls, rather than between all HR-sibs (ASD and no ASD) and low-risk controls. They found higher levels of parent directiveness during parent-child interactions between parents and their child with ASD compared to parents and a child without ASD, whereas they did not find differences between parents and a typically developing low-risk child (i.e., no older sibling with ASD) compared to parents and a typically developing high-risk child (i.e., older sibling with ASD). This is further supported by studies reporting higher levels of parent directiveness during parent-child interactions with a child with ASD (e.g., Freeman & Kasari, 2013). Although specific ASD characteristics may not be clearly visible during the first year of life, other challenging behaviours not central to an ASD diagnosis such as regulatory problems (e.g., problems in eating, sleeping and emotion regulation) could influence parents' behaviours during the parent-child interaction (Davis & Carter, 2008). This could explain differences in parents' behaviours with HR-ASD siblings compared to HR-no ASD siblings.

Unfortunately information regarding the HR-sibs' diagnostic status was not available for all the European research sites. For the Belgian sample, only thirteen HR-sibs were seen at 36 months. Of these thirteen, only 2 (15%) of the HR-sibs received an official ASD diagnosis based on research-reliable ADI-R and ADOS administrations. For the other HR-sibs, only a tentative conclusion can be formulated regarding their diagnostic status. Based on the ADOS administration at 14 and/or 24 months, another two (sample 5 months) or three (sample 10 months) HR-sibs could qualify for an ASD diagnosis at 36 months. Combined, we can carefully conclude that around 18% of the HR-sibs of the Belgian sample qualifies for an ASD diagnosis at 36 months. This is more in line with the recurrence rate reported in previous studies (18.7%; Ozonoff et al., 2011). As in the total sample, the parent-child interaction did not differ between the LR and HR group at 5

months in this Belgian subsample. At 10 months, there was a significant difference in positive affect of the child ( $U=138.50$ ,  $p=.027$ ) as well as positive affect of the parent ( $U=146.00$ ,  $p=.041$ ), but not in parents' absence of negative control ( $U=205.50$ ,  $p=.624$ ).

Finally, we did not find significant group differences in dyadic reciprocity. This is in line with the findings of Wan et al. (2012), reporting similar levels of dyadic mutuality between LR and HR dyads at the age of 6-10 months. Yirmiya et al. (2006) on the other hand found significant group differences in maternal synchrony at the age of 4 months. However, the definition of synchrony used by Yirmiya and colleagues was more technical, based on the occasions when there was a cross-correlation between the mother's and child's behaviour (e.g., child leads, mother follows). Dyadic reciprocity or mutuality as defined by the current coding scheme or Wan and colleagues (2012, 2013) also incorporates a more qualitative rating (e.g., the sense of togetherness during play). Thus, results from Yirmiya et al. (2006) are not directly comparable with the current study or the studies of Wan and colleagues (2012, 2013).

Two other findings require further attention. First, the results suggest that there is little stability in parent-child interaction from 5 to 10 months. The correlations between the scores on each scale measured at 5 and 10 months are limited and there are significant changes in several parental behaviours over time. It is therefore important to consider the infant's age at the time of the observation when studying parent-child interactions. This is further confirmed by the change in results in the study of Wan et al. (2012) after controlling for the infant's age. However, given that there was only one observation at both 5 and 10 months, the lack of stability between the two time points could also be due to random variations in the data (e.g., parent or child being tired or sick at one time point). Second, even though there were no differences between the LR and HR group, there was a significant effect of research site. The reason for this effect is unclear. Based on the test for interrater reliability prior to the coding process as well as on reliability clips coded by each coder during the coding process, all coders were considered reliable. Thus, the significant difference between sites does not seem attributable to unreliable coding. Furthermore, excluding the scores from the coder that most frequently differed from the others did not change the findings.

### Association with development

Next, it was evaluated whether parent-child interaction characteristics predicted development at 24 months. Due to the small sample size and multicollinearity in the data, development was predicted using the underlying factors rather than the individual parent-child interaction variables. Also, results should be interpreted with care and replication of these results in a larger sample is needed. Nevertheless, the results suggest that parent-child interactions at 5 and 10 months are associated with development at 24 months and that these associations differ depending on risk status. In what follows, the predictive value of the factors is discussed. In addition, to facilitate the interpretation of the results and to gain insight into which scale of the factor was most strongly correlated with MSEL scores, correlations between the individual scales within each factor and the outcome variables are discussed as well.

At 5 months, parent-child interaction variables predicted gross motor and receptive language development at 24 months in the HR group but not the LR group. PCI factor 3 (less parental control and less negative affect of the child) was associated with better gross motor skills. Looking at the individual scales, it was only the absence of child negative affect that was correlated with MSEL scores ( $r=.55$ ,  $p=.010$ ). The association with the absence of parent negative control was not significant ( $r=.09$ ,  $p=.690$ ). It is possible that HR-sibs showing higher levels of negative affect may rely more on the presence of their parent and are less inclined to explore the environment during the play observation or during play in general. This might lead to lower levels of activity or movement during the interaction, providing fewer opportunities to practice gross motor skills. In turn, weaker gross motor skills may lead to more frustration in infants, which is expressed in higher levels of negative affect.

Next, against our expectations, PCI factor 1 (more parent scaffolding and sensitive responsiveness, more child attentiveness and more dyadic reciprocity) was associated with lower receptive language abilities. More specifically, there was a strong negative association between parent sensitive responsiveness and receptive language ( $r=-.46$ ,  $p=.036$ ), which is somewhat counterintuitive. It is possible that HR-sibs who later show lower receptive language skills also show subtle difficulties with receptive language at 5 months. Based on the early items of receptive language on the *Mullen Scales of Early Learning* (Mullen, 1995), this could include difficulties in for example responding to

sounds or their own name, or social smiling. Because parents of HR-sibs already have a child with ASD, these subtle atypicalities may unsettle them, resulting in a more sensitive parenting style compared to parents of HR-sibs who do not show behavioural atypicalities. To support this hypothesis, we re-evaluated this association with two subgroups: HR-sibs who qualified for an ASD diagnosis at 36 months ( $n = 4$ ) and HR-sibs who did not ( $n = 17$ ). Given that the ASD diagnosis was not available for all HR-sibs and that the HR-ASD group was very small, the following needs to be interpreted with care. First, the HR-ASD group scored significantly lower on receptive language at both 10 ( $U=8.50, p=.018$ ) and 24 ( $U=2.00, p=.004$ ) months compared to the HR-no ASD group. Second, the correlation between parents' sensitive responsiveness at 5 months and receptive language at 24 months was remarkably larger in the HR-ASD group ( $r=-.704$ ) than in the HR-no ASD group ( $r=-.142$ ). Thus, HR-sibs who go on to develop ASD and show lower receptive language abilities as babies might alarm parents, triggering a more sensitive parenting style at 5 months. However, this association was no longer significant when looking at the parent-child interaction at 10 months. Whereas parents' concerns might be translated into more sensitivity at 5 months, it might trigger other parental behaviours at 10 months. Nevertheless, given the small sample size these results could be coincidental and future research with larger samples is needed to provide more clarity into the association between maternal sensitivity and the child's subsequent language development.

At 10 months, the parent-child interaction only predicted development at 24 months in the LR group. First, PCI factor 3 (less negative affect of the parent and the child and less parental control) was positively associated with receptive language as well as fine motor abilities. Regarding receptive language, it was mainly the child's absence of negative affect that was strongly correlated with language ( $r=.59, p=.012$ ). Parents might adopt a more verbal interaction style (e.g., motherese talk) with children who are less frustrated or distressed. More verbal communication, especially when well-timed and adapted to the child's needs, can stimulate children's language abilities (Kuhl, 2004). In addition, children who are in a neutral or positive mood may be more receptive for language input. The fact that PCI factor 2 (more parent sensitive responsiveness, scaffolding and positive affect) was also associated with better expressive language, further supports this explanation. Positive parenting behaviours such as warmth,

positive affect, acknowledging the child's needs and stimulating the child's behaviour are positively associated with language development (e.g., Russel, 2011). Even though the current results did not show significant associations for the high-risk sample at 10 months, further research is needed to explore the association between the parent-child interaction and HR-sibs' developmental trajectories.

### **Limitations and directions of future research**

Some study limitations require further discussion. At this time, information regarding the HR-sibs' ASD outcome was not yet available for all children. When all children are seen at the 36-month visit, further analyses could be done based on the diagnostic status of the HR-sibs. The recurrence rate in the current sample should be determined to estimate the proportion of HR-ASD siblings and both group differences in parent-child interactions and the association with outcome should be elaborated upon. Next, the prediction of outcome was only based on the Belgian sample, which was limited in terms of sample size. This impeded more elaborate analyses such as regression analyses with the separate parent-child interaction scales. Also important to note is that, in light of the differences found between different European research sites, results from the Belgian sample cannot be generalised to other samples. Because Belgian ratings tended to be higher than ratings from the other sites, predictions might be different in other samples or in the entire sample. In future studies, the predictive value of the parent-child interaction should be further assessed in all research sites, including the exploration of between-site differences.

### **Conclusion**

The results from the current study provide further evidence for the lack of behavioural atypicalities in HR-sibs before the age of 12 months. However, we did not replicate the findings of Wan et al. (2012) stating that parents of HR-sibs were more directive during the parent-child interaction. This is possibly due to a lower prevalence of HR-sibs with ASD in our sample. Based on our results, there were no differences between the parent-child interactions of HR- and LR-sibs. The parent-child interaction was associated with the child's gross motor and language abilities at 24 months, which emphasises the potential of parent-child interactions to influence child development.

Finally, associations were different in the HR and LR group, emphasising the importance to consider the association between the social environment and later development within each group separately.



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# CHAPTER 4

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## THE EARLY DEVELOPMENT OF INFANT SIBLINGS OF CHILDREN WITH AUTISM SPECTRUM DISORDER: CHARACTERISTICS OF SIBLING INTERACTIONS<sup>1</sup>

### ABSTRACT

Although sibling interactions play an important role in children's early development, they are rarely studied in very young children with an older brother or sister with autism spectrum disorder (ASD). This study used a naturalistic, observational method to compare interactions between 18-month-old infants and their older sibling with ASD ( $n=22$ ) with a control group of 18-month-old infants and their typically developing (TD) older sibling ( $n=29$ ). In addition, role (a)symmetry and the influence of gender were evaluated. Sibling interactions in ASD-dyads were characterised by higher levels of negativity. Although somewhat less pronounced in ASD-dyads, role asymmetry was present in both groups, with the older child taking the dominant position. Finally, siblings pairs with an older sister were characterised by more positive behaviours. Since differences in sibling interactions may alter the developmental trajectories of both siblings, these early relationships should be taken into account in future ASD research and interventions.

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## INTRODUCTION

In early development, the social world mainly consists of interactions with caregivers and siblings (Lamb, 1978). Sibling interactions are long-term, intensive relationships that influence the development of both interaction partners (Feinberg, Solmeyer, & McHale, 2012; Oh, Volling, & Gonzalez, 2015). Furthermore, they have a unique socialisation function during infancy and childhood (Carpendale & Lewis, 2004; McHale, Updegraff, & Feinberg, 2016). Positive, nurturing sibling interactions can facilitate social behaviour and relationships. They promote the development of children's understanding of others' emotions and thoughts as well as their social competence (Harrist et al., 2014; Pike, Coldwell, & Dunn, 2005; Smith & Hart, 2002). Children who experience their sibling relationships as harmonious rather than conflictual report higher levels of academic and social competence and lower levels of internalising and externalising problems (Buist & Vermande, 2014). Along with positive interactions, sibling interactions usually entail a certain level of conflict. Since conflict helps children to learn to manage their anger and stimulates them in finding ways to resolve their quarrels, the presence of conflict combined with positive interactions/nurturance can promote their development (Brody, 2004).

The nature of sibling relationships partially depends on the gender of both interaction partners and gender differences in relationship quality have been reported. Sister pairs are more intimate and harmonious than brother pairs (Buist & Vermande, 2014; Kim, McHale, Wayne Osgood, & Crouter, 2006). In addition, research shows that boys experience higher levels of sibling rivalry in interaction with a brother or sister (either older or younger) and that dyads in which the older sibling is a boy are more conflictual (Buist & Vermande, 2014; Karavasilis Karos, Howe, & Aquan-Assee, 2007).

Sibling relationships are characterised by both complementary and reciprocal interactions (El-Ghoroury & Romanczyk, 1999; Harrist et al., 2014). Complementary interactions are hierarchical and imply that one interaction partner dominates the other due to greater power or more advanced cognitive skills (because of a difference in age or experience). During reciprocal interactions, both interaction partners have an equal position (Harrist et al., 2014; Hinde, 1979). Both types of interaction stimulate development. While complementary interactions promote the development of more

refined social skills, reciprocal interactions enhance the feeling of emotional support and help siblings develop shared meanings (Dunn, 1983; Lamb, 1982). Harrist and colleagues (Harrist et al., 2014) found that, in sibling interactions including very young children, both types of interaction are equally represented. With regard to complementary interactions, in typically developing sibling pairs older siblings often take the more dominant position, which is expressed by higher levels of both prosocial and agonistic initiations. In this case, younger siblings maintain the interaction and reinforce the older child's leadership position by responding positively to positive behaviour and by submitting to negative behaviour (Abramovitch, Corter, & Lando, 1979; Abramovitch, Corter, Pepler, & Stanhope, 1986; Harrist et al., 2014; Lamb, 1978). This asymmetric pattern is stable throughout early and middle childhood and is already visible during interactions between preschool-aged children and their younger infant sibling (Lamb, 1978). Reciprocal interactions are mainly characterised by playing together and sharing mutual goals/interests (with verbal/nonverbal communication) (Harrist et al., 2014).

The question arises how the impairments in social reciprocity of children with ASD may influence these complementary and reciprocal sibling interactions. Children with ASD show lower levels of social engagement (e.g., with peers) or social interest/motivation (Broekhof et al., 2015; Kasari, Locke, Gulsrud, & Rotheram-Fuller, 2011), and they report fewer reciprocal friendships (Kasari et al., 2011). Furthermore, difficulties are noticeable in both the initiation of social engagement as in the response to play bids or emotions of others (Sigman & Ruskin, 1999). Consequently, one can expect lower levels of reciprocal interactions in sibling pairs with a child with ASD. Concerning the complementary interactions, social-communicative deficits such as the great difficulty to produce effective social overtures (Roeyers, 1996), might prevent children with ASD from taking a dominant position, resulting in sibling interactions without a clear leader. However, further research on this topic is needed.

Deficits in social communication and social interaction are among the core features of autism spectrum disorder (ASD) (American Psychiatric Association, 2013). Research also shows that, in comparison with typically developing controls, siblings of individuals with ASD have an increased risk of developing ASD themselves (Ozonoff et al., 2011). In addition, they are at higher risk of showing subclinical features of ASD, referred to as the Broader Autism Phenotype (BAP) (Bailey, Palferman, Heavey, & Le Couteur, 1998; Piven,

Palmer, Jacobi, Childress, & Arndt, 1997; Sucksmith, Roth, & Hoekstra, 2011). During the first years of life, siblings of children with ASD may show atypicalities in their cognitive, motor, language, and/or social development (Bedford et al., 2012; Gamliel, Yirmiya, & Sigman, 2007; Ozonoff et al., 2014). Deficits are most prominent in the social-communicative domain (Ozonoff et al., 2014), including the use of gestures, orientation to name, language, and interpersonal relationships (Toth, Dawson, Meltzoff, Greenon, & Fein, 2007).

The contribution of genetic factors to the development of ASD and the BAP is substantial, with at least a moderate genetic heritability (e.g., Hallmayer et al., 2011). However, genetics cannot account for all the variability found in (siblings of) children with ASD. Environmental factors such as prenatal, perinatal and/or postnatal factors also contribute to the development of ASD (Chaste & Leboyer, 2012; Elsabbagh & Johnson, 2010; Tordjman et al., 2014). Gene-environment interactions are at the root of both typical and atypical development. Identical genotypes can result in different behavioural outcomes depending on differences in children's physical or social environment (e.g., life experiences, child rearing practices) (Elsabbagh & Johnson, 2010; Gottlieb, 2007).

As sibling interactions are an important aspect of the early social environment, they need to be considered when looking at the development of younger siblings of children with ASD and their increased risk of ASD/BAP (Dawson, 2008). Until now, researchers often focussed on the unidirectional impact of children with ASD on the development of their younger siblings while characteristics of the younger sibling need to be taken into account as well (Petalas et al., 2012). Siblings influence each other through bidirectional processes. In accordance with the diathesis-stress model, pre-existing vulnerabilities such as characteristics of the BAP may influence how children with ASD affect the adjustment of their younger sibling (Bauminger & Yirmiya, 2001; Petalas et al., 2012). Social approaches of children with ASD might lack social quality (e.g., more negative, unclear requests, fewer teacher or caregiver roles). On the one hand, typically developing HR-sibs may compensate for these difficulties by taking over the leader role, hereby stimulating social communication in their sibling with ASD (Knott, Lewis, & Williams, 2007). In addition, they may show more resilience or flexibility in light of conflict. On the other hand, HR-sibs who show characteristics of BAP/ASD might experience difficulties in initiating and responding to social approaches themselves.



More generally, a lack of positive social approaches of the child with ASD limits the opportunities for HR-sibs to practice adequate social responses while a lack of positive responses in children with ASD might discourage HR-sibs to initiate social interaction with their older sibling. This may lead to fewer learning opportunities for HR-sibs and a decrease in social input. Furthermore, in line with the theory of observational learning, siblings shape the relationship with each other by observing and imitating one another (Bandura, 1977; Whiteman, McHale, & Soli, 2011). Younger siblings of children with ASD may imitate certain ASD-specific behaviours of their older brother/sister. These processes can have a significant impact on the development of younger siblings of children with ASD, possibly resulting in behaviours that resemble ASD or the broader phenotype.

In the current study, the interactional processes involved in dyads including a child with ASD and a younger infant sibling will be explored as a first step to identify the importance of sibling interactions in families with a child with ASD. Existing studies on sibling interactions in children with ASD are rare. In an observational study, Knott and colleagues (1995) compared sibling relationships in pairs including a child with ASD (mean age: 6 years) and their younger/older sibling (mean age: 6.6 years) with pairs including a child with Down Syndrome (DS) (mean age: 5.2 years) and their younger/older sibling (mean age: 5.6 years). They found that children with ASD spent less time with their sibling, initiated the interaction less frequently and showed less variation in their prosocial behaviour than children with DS. Furthermore, children with DS were more responsive to their siblings' positive initiations, than children with ASD. Using parent-report, Walton and Ingersoll (2015) found that, in interaction with their brother/sister with ASD (mean age: 9.35 years), typically developing siblings (mean age: 10.43 years) showed lower levels of involvement and higher levels of avoidance compared to siblings of typically developing children. Based on child report, Kaminsky and Dewey (2001) concluded that siblings (mean age: 11.67 years) of children with ASD (mean age: 9.79) report less conflict with their sibling than siblings of typically developing children. Regarding role (a)symmetry, the studies of Knott and colleagues (1995, 2007) showed that in dyads with a child with ASD, the younger siblings (without ASD) initiate the interaction more often, taking over the dominant position of the oldest child with ASD and at the same time stimulating communication in children with ASD.

The previous studies report meaningful findings in school-aged children. However, early social-communicative deficits in children who later develop BAP/ASD are already visible in the second year of life (Bedford et al., 2012; Szatmari et al., 2016). Consequently, sibling interactions between children with ASD and their infant siblings need to be considered as well. Around the age of 15 months, typically developing infants show high levels of joint engagement, imitative learning or instrumental imitation, attentional following (e.g., following gaze) and communicative gestures (e.g., showing, pointing, giving) (Carpenter, Nagell, Tomasello, Butterworth, & Moore, 1998). This enables them to understand and interact with people and the environment. During sibling interactions, they can for example imitate actions of an older sibling, engage in joint play, share their interests by showing or giving something to their sibling, etc. Furthermore, abilities such as joint engagement and imitation are associated with later language development, social competence and theory of mind (Carpenter et al., 1998; Charman et al., 2000; Sheinkopf, Mundy, Claussen, & Willoughby, 2004). HR-sibs are at increased risk of showing deficits in these early social-communicative competencies. This could translate into differences in social engagement, imitation and sharing during sibling interactions, which may in turn impact on the outcome of HR-sibs. Although several studies evaluated the early development of HR-sibs (e.g., Brian et al., 2014; Toth et al., 2007), we are the first to consider the potential role that early sibling interactions can play in the expression of BAP or ASD in HR-sibs. The assessment of sibling interactions can shed light on the pathways between early vulnerabilities and later outcome (Dawson, 2008).

In addition to the focus on older children, there is a shortage of naturalistic observations. Sibling interactions have predominantly been studied by means of questionnaires (e.g., Kaminsky & Dewey, 2001; Rivers & Stoneman, 2008; Walton & Ingersoll, 2015), which have some disadvantages such as rater bias in parents (Stone, Hoffman, Lewis, & Ousley, 1994; Zwaigenbaum et al., 2005). Child-report also requires that children are old enough to report about their experiences. In addition, there is an important difference between reporting about sibling relations in terms of cognitions and/or perceptions and observing the specific dynamics of these interactions. These disadvantages can be avoided by using a naturalistic, observational method, which can

also provide a broader picture (Hastings & Petalas, 2014; Lobato, Miller, Barbour, Hall, & Pezzullo, 1991; Senapati & Hayes, 1988).

The present study aimed to evaluate three important aspects of sibling interactions: 1) The interactive behaviour of both siblings; 2) Role (a)symmetries; 3) The effect of gender. To this end, we observed sibling interactions in dyads including 18-month-old infants and their older sibling with ASD as well as in typically developing control dyads. To ensure the ecological validity of the observation, sibling interactions were observed in a naturalistic, familiar setting (i.e., the children's home). To our knowledge, our study is the first to observe sibling interactions among children with ASD and their younger infant sibling in comparison with a typically developing control group. First, sibling interactions were compared between groups to identify possible differences between dyads including a child with ASD and typically developing dyads. Given the social-communicative difficulties associated with ASD and based on previous research revealing fewer initiations and responses in children with ASD (Knott et al., 1995) as well as lower involvement of siblings of children with ASD (Walton & Ingersoll, 2015), lower levels of interactive behaviour were expected in dyads with children with ASD. Next, role (a)symmetry was evaluated for each group separately. To this end, the behaviour of the younger and older sibling within each group was compared in terms of positive and negative social initiations and responses. In line with previous research, we expected clear role asymmetry in the control group with the older child assuming a more dominant position (Abramovitch et al., 1979, 1986; Lamb, 1978). With regard to dyads with a child with ASD, Knott and colleagues (1995, 2007) suggested that not the children with ASD but their younger sibling would act as a leader. However, the younger siblings in the current sample were 18-month-old infants, whose social-communicative abilities are not as developed as in older children. Regarding the child with ASD, their social-communicative difficulties might inhibit them from assuming a dominant position as well. Consequently, given that both children might lack the abilities to act as a leader, we did not expect a clear role asymmetry in dyads with a child with ASD.

As a second research goal, the influence of gender of the oldest child on the quality of the sibling relationship was explored in the control group. In line with previous research, we expected dyads with an older sister to be more positive than dyads with an older brother (Buist & Vermande, 2014; Karavasilis Karos et al., 2007; Kim et al., 2006).

## METHOD

### Participants

Fifty-one sibling pairs participated in the study. All participants were recruited from an ongoing prospective follow-up study of younger siblings of children with ASD considered to be at increased risk for developing ASD (high-risk siblings; HR-sibs), and a typically developing control group at Ghent University. Twenty-two sibling pairs were high-risk (HR) dyads with an older child with ASD (hereafter, ASD-sib). Nine HR-sibs had one older sibling and another nine had two older siblings. Four HR-sibs had three or more older siblings and two HR-sibs had two siblings with ASD. The HR group consisted of 10 male-male, 9 female-male, 1 male-female and 2 female-female dyads (younger-older). Twenty-nine sibling pairs were low-risk (LR) dyads with a typically developing older child (typically developing sibling; TD-sib) and their younger sibling (low-risk sibling; LR-sib). The LR group had a family history without first-degree relatives with ASD. All LR-sibs had at least one older sibling and four had two older siblings. The LR group consisted of 7 male-male, 7 female-male, 11 male-female and 4 female-female dyads (younger-older). More HR- than LR-sibs had more than one older sibling (59% vs. 14%;  $\chi^2=11.55, p=.001$ ).

ASD-sibs received their ASD diagnosis after evaluation by a multidisciplinary team, including assessment of cognitive and social-communicative functioning. Diagnostic status of the oldest child in each group was confirmed using the Social Responsiveness Scale, Second Edition (SRS-2; Constantino & Gruber, 2012), and the Social Communication Questionnaire (SCQ; Rutter, Bailey, & Lord, 2003). In the HR group, the SCQ and SRS were available for all 22 children/ASD-sibs. 12 children scored above the threshold for ASD on both the SCQ and the SRS, the other ten scored above the threshold on the SRS. In the LR group, SRS and SCQ were available for 25 and 26 children/TD-sibs, respectively. All scored below the ASD threshold on the SCQ and all but one (total score of 69) scored below the ASD threshold on the SRS. Since further evaluation of this child (SCQ score, parent information/concerns) revealed no reasons to suspect ASD, we decided to include this dyad in further analyses. Cognitive functioning of children with ASD was assessed using either the Wechsler Intelligence Scale for Children (WISC-III-NL; Kort et al., 2005), the Wechsler Preschool and Primary Scale of

Intelligence (WPPSI-III-NL; Hendriksen & Hurks, 2009), the Snijders-Oomen Non-Verbal Intelligence Test (SON-R; Tellegen, Winkel, Wijnberg-Williams, & Laros, 1998), or the Bayley Scales of Infant Development (BSID-II-NL: Meulen, van der Ruitter, Spelberg, & Smrkovský, 2004); Bayley-III-NL: Van Baar, Steenis, Verhoeven, & Hessen, 2014). One child with ASD had a total intelligence quotient or developmental index that was very low ( $IQ < 55$ ), eight children scored below average ( $IQ < 85$ ), and eleven children scored in the normal range (IQ between 85-115). For two children, information on cognitive functioning was not available.

Sample characteristics are presented in Table 1. With regard to the youngest sibling, no differences were found in chronological age or sex ratio. As all younger siblings participated in the prospective follow-up study, information on their developmental level at the age of 14 months was available, as tested with the Mullen Scales of Early Learning (MSEL; Mullen, 1995). The average developmental quotient (DQ) was higher in the LR group ( $F(1,43)=4.78, p=.034$ ). Concerning the older siblings, ASD-sibs were older than TD-sibs ( $U=107.50, p<.001$ ) and there was a significant difference in sex ratio, with an overrepresentation of boys in ASD-sibs ( $\chi^2(1)=7.95, p=.007$ ). The families' socioeconomic status (SES) was calculated using Hollingshead's four factor index (Hollingshead, 1975). LR families had an average social status of 51.79 (range: 34.50-66.00) and HR families had an average social status of 40.77 (range: 22.00-66.00). The difference between groups was significant ( $U=151.50, p=.001$ ).

Additional information was collected with regard to the amount of time children spent playing together (seldom/sometimes/often) and to what extent both children attended school or day care centres. There were no significant differences between groups in time both children spent together while at home ( $\chi^2(1) = 5.71, p=.063$ ). However, LR-sibs more frequently went to day care centres than HR-sibs (93% vs. 64%;  $\chi^2=6.89, p=.013$ ). All older siblings attended school.

**Table 1***Sample characteristics*

	Low-risk (n=29)	High-risk (n=22)	
Youngest sibling			
Chronological age			
<i>M(sd)</i>	18.37 (.54)	18.52 (.85)	$F(1,49) = .59$
Range	17.17-19.33	17.37-20.43	
Sex ratio (M:F)	18:11	11:11	$\chi^2(1) = .74$
Developmental level (14 months)			
<i>M(sd)</i>	104.85 (9.29)	97.94 (11.86)	$F(1,43) = 4.78^*$
Range	92.00-126.00	79.00-120.00	
Oldest sibling			
Chronological age			
<i>M(sd)</i>	52.61 (14.85)	89.43 (39.10)	$U = 107.50^{***}$
Range	32.97-90.30	46.00-186.07	
Sex ratio (M:F)	14:15	19:3	$\chi^2(1) = 7.95^{**}$
Sibling pair			
Family SES ( <i>M(sd)</i> )	51.79 (6.96)	40.77 (12.28)	$U=151.50^{**}$
Time spent together (%)			$\chi^2(1) = 5.71$
<i>Never/seldom</i>	3,4%	22,7%	
<i>Sometimes</i>	34,5%	40,9%	
<i>Often/always</i>	62,1%	36,3%	
Day care attendance (%)	93%	64%	$\chi^2(1) = 6.89^*$

Note. Chronological age is reported in months; \* $p < .05$ , \*\* $p < .01$ , \*\*\* $p < .001$

**Procedure**

Sibling interactions were observed during a short play observation in a familiar context. In all but one cases, this was in the families' home. One play observation took place in the house of the grandparents. The children were offered a fixed set of toys, namely zoo-themed building blocks, a marble run and an animal sound keyboard, with which they could play consecutively for 10, 10 and 5 minutes, respectively. Before

offering a new toy, the previous toy was removed by the researcher. To facilitate the transition between each toy set, children were asked to help clear away the toys or the parent joined briefly to interact with the children. Children were encouraged to play together during the introduction of each toy set and they were given a short verbal instruction (“you can play together with these toys”). Once the children started playing, the researcher observed the play session from the background in order to observe spontaneous behaviour as much as possible. Different sets of toys were chosen to elicit different kinds of play. During play with blocks, the opportunities for parallel play (children play next to each other without interacting) were highest and opportunities for associative (children share or exchange toys, but there is no common goal and interaction is limited) or cooperative (children play together and they have a common goal, work together, make rules, etc.) play were limited. While playing with the marble run children had to share some objects (e.g., the marbles), but they were still able to play parallel as well. Therefore, opportunities for associative and cooperative play were higher during play with the marble run. Finally, during play with the keyboard opportunities for parallel play were limited since there was only one keyboard. By only providing one toy (i.e., limited resources), associative or cooperative play was encouraged. The more children had to share, the higher the risk of conflict.

To avoid unnecessary distractions, parents were asked to switch off all electronic devices (television, tablet) and to remove other toys as much as possible. At the beginning of each play session, parents received general information about the study and were asked to sign an informed consent. During the observation, one parent was always present in the room, continuing normal routines (e.g., household tasks or work).

*Coding procedure.* All play sessions were videotaped and coded afterwards using The Observer XT, version 11.5 (Noldus, 2013). The coding scheme developed for the current study was based on previous work by Abramovitch et al. (1987), Knott et al. (1995), and Roeyers (1996), but further adapted and elaborated. Interactive behaviour was coded in terms of social initiations and responses. *Social initiations* are communicative attempts to initiate a new interaction, directed towards another individual. *Responses* are related to and follow a previous initiation within five seconds. Initiations and responses can be either positive/prosocial (e.g., sharing a toy, allowing the other sibling to do something) or negative (e.g., refusing a request). The absence of a

response was also coded (*no response*). In addition to the initiations and responses, the time children spent in interaction with each other (*mutuality*), with the *parent* and with the *experimenter* was also coded. To account for the time not spent in interaction with another person, non-interactive behaviours were coded as well. The following behaviours were included in the present study: *orientation towards the sibling* (without interacting), time spent in a *purposeful activity* (e.g., play), *distressed behaviour* (e.g., anger tantrum, crying), *stereotypical/sensory behaviour*, and *doing nothing/looking at something random*.

Clips were rated by trained master students blind to participant information. 15% of the clips were randomly selected to determine interrater reliability and were coded by all coders. Intraclass correlation coefficients (ICC) were calculated for both the youngest and the oldest child and for each play context. ICC's ranged between .80 and .99 for the marble run and blocks, and between .78 and .97 for the keyboard. The behaviours 'no response', 'looking at random' 'distress' and 'stereotypical/sensory behaviour' were rarely coded, making reliability analysis impossible. Therefore, these behaviours were excluded from further analysis.

### **Data-analysis**

Potential outliers were detected using box plots and visual inspection of the data. Values higher/lower than the mean +/- 3 times the standard deviation (sd) were considered outliers. Assuming that extreme data were not random deviations but characteristic of the sample (e.g., high levels of initiations in high-functioning girls), outliers were replaced by the highest/lowest value allowed (mean +/- 3sd) rather than deleted.

Parametric group comparisons were not possible due to a lack of normal distribution in our data. Consequently, hierarchical regression analyses were used to analyse the degree to which the group status (high-risk vs. low-risk) predicted sibling interaction characteristics. Accordingly, at the first step of the regression model, group was added as a predictor of the sibling interaction variables (positive/negative initiations/responses, mutuality, orientation to sibling). Since the LR and HR group differed in terms of sex ratio and age of the oldest child, family SES, and developmental level of the youngest child, these sample characteristics were added at step 2 of the



regression model to evaluate whether sample characteristics changed the possible association between group status and sibling interactions. In addition, non-parametric analyses were used to analyse between-group differences (low-risk vs. high-risk; Mann-Whitney U) and within-group differences (youngest vs. oldest; Wilcoxon signed-rank).

To control for the inflation of the Type I error rate due to multiple comparisons, the (Holm-)Bonferroni correction was considered. However, both procedures lead to a substantial reduction of statistical power (Nakagawa, 2004; Perneger, 1998). Due to the combination of a lower statistical power because of the small sample size and the fact that we aim to detect small differences, applying a Bonferroni correction would greatly reduce the possibility of finding relevant group differences while there are in fact real world differences. Consequently, results without the Bonferroni correction are also discussed.

## RESULTS

The characteristics of the sibling interaction were similar for both the play with blocks and the marble run. Consequently, the data of these two play sets were combined. During play with the keyboard, results differed from the other two play sets and are therefore discussed separately.

### **Interactive vs. non-interactive behaviour**

First, the proportion of time children spent in social interaction (mutuality, interaction with experimenter, interaction with parent) compared to non-social activities (orientation towards sibling, involvement in a purposeful activity) was evaluated. During play with marble run/blocks, LR- and TD-sibs spent on average 15% and 16% of their time in social interaction, respectively. In the HR group, the average proportion of time spent in social interaction was 14% for the HR-sibs and 16% for the ASD-sibs. Differences between the LR and HR group were not significant (younger:  $U=287.00$ ,  $p=.552$ ; older:  $U=306.00$ ,  $p=.814$ ). While playing with the keyboard, the average proportion of time spent in social interaction was 15% for LR-sibs, 18% for TD-sibs, 15% for HR-sibs and 21% for ASD-sibs. Again, there were no significant differences between the LR and HR group

(younger:  $U=315.00$ ,  $p=.943$ ; older:  $U=294.00$ ,  $p=.644$ ). When only looking at social interaction between both siblings (mutuality), the average proportion of time spent in social interaction decreased to 3% for the LR group and 5% for the HR group during play with marble run/blocks, and 3% for the LR group and 2% for the HR group during play with keyboard. Differences between the LR and HR group were all non-significant (marble run/blocks:  $U=225.00$ ,  $p=.074$ ; keyboard:  $U=318.50$ ,  $p=.996$ ).

### Characteristics of sibling interactions

Descriptives of the sibling interaction are presented in Table 2. Results of the hierarchical regression models are shown in Table 3 (marble run/blocks) and Table 4 (keyboard).  $P$ -values that remained significant after the Bonferroni correction are marked in the tables.

**Table 2**

*Means (standard deviations) of sibling interaction characteristics*

	Marble run and Blocks		Keyboard	
	LR	HR	LR	HR
InNeg youngest <sup>a</sup>	0.63(0.69)	1.09(1.09)	1.48(2.18)	1.27(1.64)
InPos youngest <sup>a</sup>	2.31(2.64)	1.75(1.75)	0.91(1.79)	0.86(1.39)
ResNeg youngest <sup>a</sup>	1.41(1.65)	3.64(2.76)	2.24(2.54)	3.10(4.52)
ResPos youngest <sup>a</sup>	4.89(4.05)	6.32(4.98)	3.87(3.81)	2.08(2.16)
Mut <sup>b</sup>	15.50(16.92)	26.28(27.57)	15.38(21.03)	12.79(15.94)
Sib youngest <sup>b</sup>	47.08(18.12)	40.36(34.51)	26.41(21.82)	12.19(13.44)
	Marble run and Blocks		Keyboard	
	LR	HR	LR	HR
InNeg oldest <sup>a</sup>	3.57(2.42)	5.05(3.85)	3.86(3.23)	2.68(3.76)
InPos oldest <sup>a</sup>	4.15(4.53)	6.00(8.04)	2.79(4.48)	2.00(2.65)
ResNeg oldest <sup>a</sup>	0.95(1.41)	2.02(2.04)	1.59(2.10)	1.95(2.52)
ResPos oldest <sup>a</sup>	2.09(2.42)	2.66(3.19)	1.82(2.51)	1.36(1.94)
Mut <sup>b</sup>	15.50(16.92)	26.28(27.57)	15.38(21.03)	12.79(15.94)
Sib oldest <sup>b</sup>	21.55(27.78)	70.85(122.37)	19.56(22.65)	41.32(60.69)

*Note.* LR=low-risk, HR=high-risk; InNeg=negative initiations, InPos=positive initiations, ResNeg=negative responses, ResPos=positive responses, Mut=mutuality, Sib=orientation towards sibling; <sup>a</sup>results reflect absolute frequencies; <sup>b</sup>results reflect total duration (in seconds)

*Youngest child.* During play with marble run/blocks, group was a significant predictor for two sibling interaction characteristics: negative initiations and negative responses. For *negative initiations*, group initially accounted for 9.7% of the variance ( $F(1,43)=4.599, p=.038$ ). After adding the sample characteristics, the model accounted for 20% of the variance, but was no longer significant ( $F(5,39)=1.944, p=.109$ ). Nevertheless, group remained a significant predictor with higher levels of negative initiations in the high-risk group. Regarding *negative responses*, the model containing both group and the sample characteristics was significant, accounting for 28.2% of the variance ( $F(5,39)=3.071, p=.020$ ). Group was the only significant predictor with more negative responses in the high-risk group. During play with keyboard, group did not significantly predict sibling interaction characteristics after adding the sample characteristics in the model.

*Oldest child.* Play with marble run/blocks revealed two models in which group was a significant predictor. First, the model significantly predicted the *negative initiations*, accounting for 48.3% of the variance ( $F(5,39)=7.293, p<.001$ ). Group was a significant positive predictor with higher levels of negative initiations in the high-risk group. Second, group significantly predicted *negative responses*, meaning that negative responses were more frequent in the high-risk group. However, the overall model was not significant ( $R^2=.191, F(5,39)=1.846, p=.126$ ). In line with the youngest sibling, group did not significantly predict sibling interaction characteristics during play with keyboard.

### **Role (a)symmetry**

Role (a)symmetry was based upon the number of initiations and responses of both siblings. Higher levels of initiations reflect a more dominant position, while higher levels of responses indicate a more following role. The younger and older child within each group were compared. As in the previous analyses, results for play with the marble run and play with the blocks were combined. Wilcoxon signed-rank tests were used to explore within-group differences. Results that remained significant after the Bonferroni correction are marked with an asterisk (\*).

**Table 3***Regression coefficients for significant predictors - Marble run/Blocks*

		Youngest sibling			Oldest sibling		
		<i>B</i> ( <i>SD</i> )	$\beta$	<i>R</i> <sup>2</sup>	<i>B</i> ( <i>SD</i> )	$\beta$	<i>R</i> <sup>2</sup>
<b>InPos</b>	1. (constant)	2.39(.46)		.01	1. (constant)	4.31(1.23)	.05
	Group	-.47(.73)	-.10		Group	2.95(1.95)	.23
	2. (constant)	3.08(3.79)		.28*	2. (constant)	10.19(9.12)	.44**** <sup>a</sup>
	Gender	1.85(.71)	.38*		Gender	4.67(1.72)	.35**
	Age	.03(.01)	.36 <sup>+</sup>		Age	.11(.03)	.56**** <sup>a</sup>
<b>InNeg</b>	1. (constant)	.66(.17)		.10*	1. (constant)	3.61(.61)	.08
	Group	.59(.27)	.31*		Group	1.83(.97)	.28
	2. (constant)	1.79(1.57)		.20	2. (constant)	1.27(4.43)	.48**** <sup>a</sup>
	Group	.98(.40)	.52*		Group	4.96(1.11)	.75**** <sup>a</sup>
	Age	-.01(.01)	-.40*		Age	-.04(.01)	-.44**
				SES	.18(.04)	.57**** <sup>a</sup>	
<b>ResPos</b>	1. (constant)	5.11(.85)		.07	1. (constant)	2.20(.54)	.03
	Group	2.34(1.34)	.26		Group	1.00(.86)	.17
	2. (constant)	11.45(6.93)		.33*	2. (constant)	3.01(4.30)	.35**
	Gender	2.74(1.30)	.30*		Gender	2.63(.81)	.46**** <sup>a</sup>
	Age	.05(.02)	.38*		Age	.03(.01)	.37*
	DQ	-.12(.06)	-.29*				
<b>ResNeg</b>	1. (constant)	1.42(.40)		.19**	1. (constant)	.96(.32)	.09
	Group	2.00(.63)	.43**		Group	1.01(.50)	.29
	2. (constant)	.20(3.64)		.28*	2. (constant)	.68(2.89)	.19
	Group	3.11(.91)	.68**** <sup>a</sup>		Group	2.06(.73)	.60**** <sup>a</sup>
	Age				Age	-.02(.01)	-.40*
<b>Sib</b>	1. (constant)	46.71(5.40)		.01	1. (constant)	22.20(16.51)	.12*
	Group	-6.04(8.55)	-.11		Group	62.49(26.11)	.34*
	2. (constant)	64.78(47.87)		.17	2. (constant)	64.59(110.59)	.57**** <sup>a</sup>
	Age	-.37(.16)	-.46*		Age	2.05(.36)	.78**** <sup>a</sup>

*Note.* <sup>+</sup>*p* = .053, \**p* < .05, \*\**p* < .01, \*\*\**p* < .001; InPos = Positive initiations; InNeg = Negative initiations; ResPos = Positive responses; ResNeg = Negative responses; Sib = Orientation to sibling; Gender = gender of the oldest sibling; Age = age of the oldest sibling; DQ = developmental quotient youngest sibling; SES = family SES; Group = high-risk vs. low-risk; <sup>a</sup>remained significant after Bonferroni correction

**Table 4**

*Regression coefficients for significant predictors - Keyboard*

	Youngest sibling			Oldest sibling				
		<i>B (SD)</i>	$\beta$	<i>R</i> <sup>2</sup>		<i>B (SD)</i>	$\beta$	<i>R</i> <sup>2</sup>
<b>InPos</b>	1. (constant)	.98(.33)		.01	1. (constant)	2.93(.77)		.01
	Group	.02(.52)	.01		Group	-.59(1.22)	-.07	
	2. (constant)	-2.77(3.02)		.10	2. (constant)	10.02(6.55)		.22
					Gender	3.22(1.23)	.40*	
<b>InNeg</b>	1. (constant)	1.59(.39)		.00	1. (constant)	3.96(.69)		.03
	Group	-.20(.62)	-.05		Group	-1.13(1.09)	-.16	
	2. (constant)	-6.75(3.23)		.26*	2. (constant)	.76(6.54)		.05
	DQ	.09(.03)	.50*** <sup>a</sup>					
<b>ResPos</b>	1. (constant)	4.01(.64)		.07	1. (constant)	1.96(.45)		.00
	Group	-1.81(1.02)	-.26		Group	-.29(.72)	-.06	
	2. (constant)	9.79(5.87)		.17	2. (constant)	-3.52(3.71)		.29*
					Gender	2.27(.70)	.48*** <sup>a</sup>	
<b>ResNeg</b>	1. (constant)	2.37(.57)		.01	1. (constant)	1.70(.42)		.00
	Group	.41(.91)	.07		Group	.19(.67)	.04	
	2. (constant)	1.66(5.40)		.05	2. (constant)	.91(3.96)		.05
<b>Sib</b>	1. (constant)	36.48(3.71)		.12*	1. (constant)	20.46(8.54)		.10*
	Group	-14.45(5.87)	-.35*		Group	28.97(13.51)	.31*	
	2. (constant)	76.61(31.12)		.34**	2. (constant)	27.71(55.19)		.60*** <sup>a</sup>
	Gender	13.06(5.85)	.31*		Age	1.04(.18)	.77*** <sup>a</sup>	
	DQ	-.60(.26)	-.32*		SES	-1.23(.52)	-.28*	
Sibling pair								
		<i>B (SD)</i>	$\beta$	<i>R</i> <sup>2</sup>				
<b>Mut</b>	1. (constant)	16.52(3.82)		.00				
	Group	-2.31(6.04)	-.06					
	2. (constant)	37.20(33.71)		.16				
	Gender	14.42(6.34)	.36*					

*Note.* \* $p < .05$ , \*\* $p < .01$ , \*\*\* $p < .001$ ; InPos = Positive initiations; InNeg = Negative initiations; ResPos = Positive responses; ResNeg = Negative responses; Sib = Orientation to sibling; Mut=Mutuality; Gender = gender of the oldest sibling; Age = age of the oldest sibling; DQ = developmental quotient youngest sibling; SES = family SES; Group = high-risk vs. low-risk; <sup>a</sup>remained significant after Bonferroni correction

*Low-risk dyads.* During play with the marble run/blocks and play with the keyboard, the older sibling assumed a more dominant position, reflected in higher levels of positive (marble run/blocks:  $z=-2.75$ ,  $p=.005$ ; keyboard:  $z=-2.77$ ,  $p=.004$ ), and negative (marble run/blocks:  $z=-4.49$ ,  $p<.001^*$ ; keyboard:  $z=-2.97$ ,  $p=.002^*$ ) initiations. The younger siblings followed more frequently, with higher levels of positive (marble run/blocks:  $z=-4.26$ ,  $p<.001^*$ ; keyboard:  $z=-2.38$ ,  $p=.015$ ), and negative (marble run/blocks:  $z=-2.44$ ,  $p=.014$ ) responses. During play with the keyboard, the difference in negative responses was not significant ( $z=-1.94$ ,  $p=.051$ ).

*High-risk dyads.* During play with the marble run and blocks, older children with ASD showed higher levels of positive ( $z=-2.78$ ,  $p=.004$ ) and negative initiations ( $z=-3.73$ ,  $p<.001^*$ ), and their younger HR-sibs showed higher levels of positive ( $z=-3.15$ ,  $p=.001^*$ ) and negative responses ( $z=-2.38$ ,  $p=.015$ ). While playing with the keyboard, children with ASD did not show a higher number of initiations. The younger HR-sibs showed more negative responses ( $z=-2.34$ ,  $p=.021$ ).

*Comparison high-risk and low-risk.* Finally, it was evaluated whether there was a difference in role (a)symmetry between groups. To determine the degree to which the oldest sibling was more dominant, the difference score between initiations of the oldest sibling and initiations of the youngest sibling was calculated. To evaluate to which extent the youngest sibling was more following, the difference score between responses of the youngest sibling and responses of the oldest sibling was calculated. These difference scores were compared between groups (Mann-Whitney U test). Results only revealed a marginally significant difference in the dominance of the oldest child during marble run/blocks ( $U=219.50$ ,  $p=.058$ ), indicating that the leader position of the oldest child was more pronounced in the LR group than in the HR group. There was no significant difference in the dominance of the oldest child during keyboard ( $U=243.50$ ,  $p=.152$ ), or the degree to which the youngest child followed (marble run/blocks:  $U=252.00$ ,  $p=.205$ ; keyboard:  $U=278.50$ ,  $p=.445$ ).

### **Sample characteristics**

The degree to which sample characteristics influenced the association between group status (high-risk vs. low-risk) and the sibling interactions was evaluated in the previously mentioned hierarchical regression models (see Table 3 and 4). Results are

discussed in more detail below. Results that remained significant after the Bonferroni correction are again marked in the table.

*Gender.* The gender of the oldest sibling was a significant predictor for positive initiations and responses of both the youngest and oldest child during marble run/blocks, and for orientation to sibling of the youngest child and positive initiations and responses of the oldest child during play with the keyboard. When the older sibling was a girl, positive initiations (youngest:  $\beta=.382$ ,  $p=.013$ ; oldest:  $\beta=.352$ ,  $p=.010$ ) and responses (youngest:  $\beta=.299$ ,  $p=.042$ ; oldest:  $\beta=.455$ ,  $p=.002^*$ ) were more frequent during play with marble run/blocks than when the older sibling was a boy. During play with keyboard, the youngest children more frequently oriented towards their sibling in sibling pairs with an older sister compared to an older brother ( $\beta=.314$ ,  $p=.032$ ), while the oldest children showed higher levels of positive initiations ( $\beta=.399$ ,  $p=.013$ ) and responses ( $\beta=.477$ ,  $p=.002^*$ ). In addition, although the regression model was not significant ( $F(5,39)=1.531$ ,  $p=.203$ ), mutuality was higher in dyads with an older sister ( $\beta=.360$ ,  $p=.029$ ).

For the positive responses of both children during play with marble run/blocks, the association between group and the sibling interaction was no longer significant when taking the gender of the oldest sibling into account. During play with the keyboard, the association between group and orientation to sibling of the youngest child was no longer significant after adding both gender of the oldest child and developmental level of the youngest child.

*Age.* During play with marble run/blocks, the age of the oldest sibling significantly predicted the negative initiations, positive responses and orientation to sibling of the youngest child, and positive initiations, positive responses, negative initiations, negative responses and orientation to sibling of the oldest child. Positive behaviours were more frequent when the older sibling was older (positive responses youngest:  $\beta=.384$ ,  $p=.031$ ; positive initiations oldest:  $\beta=.557$ ,  $p=.001^*$ ; positive responses oldest:  $\beta=.372$ ,  $p=.033$ ). On the other hand, negative behaviours were less frequent when the oldest child was older (negative initiations youngest:  $\beta=-.400$ ,  $p=.039$ ; negative initiations oldest:  $\beta=-.435$ ,  $p=.006$ ; negative responses oldest:  $\beta=-.399$ ,  $p=.040$ ). Finally, younger siblings were less frequently oriented to their older sibling when the oldest sibling was older ( $\beta=-.456$ ,  $p=.022$ ), while the older siblings were more frequently oriented to their younger sibling

( $\beta=.776, p<.001^*$ ). During play with keyboard, age of the oldest sibling only predicted orientation to sibling of the oldest child, with higher levels of orientation to sibling when the oldest child was older ( $\beta=.771, p<.001^*$ ).

The negative initiations of the youngest child and negative initiations and responses of the oldest child during marble run/blocks were still significantly predicted by group status (high-risk vs. low-risk) after adding the sample characteristics in the regression model. However, orientation to sibling of the oldest child was no longer predicted by group after adding the age of the oldest child, both during play with the marble run/blocks and play with keyboard.

*Developmental level youngest.* The developmental level of the youngest child was only associated with the positive responses of the youngest child during marble run/blocks. Younger siblings with a lower developmental level showed more positive responses ( $\beta=-.292, p=.044$ ). During play with the keyboard, developmental level of the youngest child significantly predicted the negative initiations and orientation to sibling of the youngest child. Younger siblings with a lower developmental level initiated the interaction less in a negative way ( $\beta=.500, p=.002^*$ ) and showed higher levels of orientation to their older sibling ( $\beta=-.316, p=.029$ ).

After adding developmental level of the youngest sibling and age of the oldest sibling in the hierarchical regression model, the association between group and orientation to sibling of the youngest child (keyboard) was no longer significant.

*Family SES.* During play with marble run/blocks, a higher socioeconomic status was associated with more negative initiations of the oldest child ( $\beta=.567, p<.001^*$ ). Nevertheless, group remained significant predictor for negative initiations of the oldest child. Second, during play with keyboard the oldest child was more frequently oriented towards their younger sibling when the family SES was lower ( $\beta=-.281, p=.022$ ). The significant association between group and orientation to sibling of the oldest sibling disappeared after adding age of the oldest sibling and family SES in the regression model.



## DISCUSSION

The main focus of this study was to evaluate the characteristics of sibling interactions in dyads involving a child with ASD and an infant sibling in comparison with typically developing dyads. Additionally, we investigated whether the role asymmetry that characterises early and middle childhood in typically developing dyads was also evident in dyads including a child with ASD. Finally, we explored the potential influence of child characteristics such as gender and age.

The present study revealed similarities as well as differences between dyads including a child with ASD and dyads with only typically developing children. Concerning positive behaviours, there were no clear differences between the HR and LR groups. Both showed a comparable amount of positive initiations and responses in all three play contexts. With regard to negative behaviours, differences were more pronounced. During play with marble run/blocks, HR-sibs and children with ASD showed higher levels of negative behaviours than LR-sibs and typically developing older children, both in terms of negative initiations and negative responses. Negative initiations included giving a command to or taking a toy from the other sibling, while negative responses included refusing to comply with a request (e.g., giving toy, following command) or counterattacks (e.g., resisting when the other sibling attempted to take a toy away). Play with keyboard did not reveal any group differences. The results are not in agreement with the expectation that children in HR dyads would be less interactive than children in LR dyads. Instead, there was a difference regarding the distinction between positive and negative behaviour. The HR group clearly showed more negative behaviours during play with marble run/blocks.

After exploring the differences in sibling interactions between both groups, we focussed on the role patterns between infants and their older sibling within each group. This has not yet been investigated in a very young age group. In all three play contexts and in both groups there was a pattern that older children initiated more frequently (dominant position) while younger children followed and responded more, this in terms of positive as well as negative initiations/responses. Especially during play with marble run/blocks, this asymmetry was clear in both the LR and HR group. The comparison of

role (a)symmetry between groups revealed no significant differences. Thus, against our expectations, role asymmetry was also found in HR dyads.

Our findings in the HR group are not in line with Knott and colleagues (1995, 2007), who showed that children with ASD initiate interaction less frequently and are less responsive to initiations of their sibling, nor with Walton and Ingersoll (2015), who found less involvement and more avoidance in HR-sibs. In addition, with regard to role (a)symmetry, Knott and colleagues (1995, 2007) found that the younger typically developing child took over the dominant position of the older child with ASD, while this was not the case in our study. It must be noted, however, that the children in the studies of Knott et al. (1995, 2007) and Walton and Ingersoll (2015) were on average older than the children in this study. Consequently, differences in results could be related to the younger sample. First, the level of social interaction in children and their *interest* in other interaction partners partially depends on the age of the child. In the present study the overall level of mutual interaction between the 18-month-old infants and their older sibling was low in the LR as well as in the HR group (5% or less of the total interaction). Previous research by Lamb (1978) also emphasised the limited amount of direct interaction between young siblings. In their study, younger infants and preschool-aged children showed more interest in interacting with their parent than with their sibling. Moreover, siblings stayed close to each other and often played with the same toys, but they were rather engaged in parallel than in mutual play. Second, the child's age determines the level of social-communicative development and the extent to which children are *capable* of participating in and/or leading social interactions. Infants will show fewer social-communicative abilities than toddlers/pre-schoolers or school-aged children. In addition, even though 18-month-old infants already display levels of simple social play during peer interactions, more complex forms of social play are not yet developed (Howes & Matheson, 1992). In sum, younger children could be less motivated and/or could lack the social-communicative abilities needed to participate in social interaction as well as to take on a dominant/leading position. Therefore, the young age of the current sample could explain the lower overall level of interaction, the limited group differences and the discrepancy between the current study and the studies of Knott et al. (1995, 2007) and Walton and Ingersoll (2015).

The limited time younger sibling pairs spend in mutual interaction and infants' lower levels of social-communicative abilities reduce the possibility of finding differences between ASD dyads and typically developing dyads. Nevertheless, the current study found significant group results, confirming that this younger age group should not be neglected. Especially the presence of negative behaviour in the HR group is noteworthy. During play with the marble run/blocks, sibling interactions in the HR group were more conflictual than in the LR group, possibly disturbing the balance between positive and negative interactions. This may impact on the development of HR-sibs. On the one hand, previous research has shown that sibling relationships high in conflict are associated with lower levels of social competence and self-worth, and higher levels of internalising and externalising problems (Buist & Vermande, 2014). On the other hand, when combined with warmth and nurturing interactions, conflict can lead to a more positive outcome (Brody, 2004). Participating in conflict rather than submitting to negative behaviour of the other sibling can benefit children's development, for example by promoting their problem-solving abilities.

As a result of the social-communicative difficulties associated with ASD, it can be expected that children with ASD experience difficulties in adequately initiating social interaction with a younger sibling or responding appropriately to their siblings' social approaches, resulting in higher levels of negative initiations and responses. Children with ASD also show higher rates of aggressive behaviour, depending on their cognitive functioning and language development (Dominick, Davis, Lainhart, Tager-Flusberg, & Folstein, 2007; Fitzpatrick, Srivorakiat, Wink, Pedapati, & Erickson, 2016; Hartley, Sikora, & McCoy, 2008). While negative behaviour in children with ASD can be associated with characteristics of the disorder, the origin of higher levels of negative behaviour in HR-sibs is less clear. First, through bidirectional processes, HR-sibs' responses may be influenced by repeated confrontation with the social-communicative difficulties of the child with ASD. For example, higher amounts of negative initiations or responses in children with ASD could trigger more negative initiations or responses in the HR-sib. After a while, this might become a learned interaction style. A second possibility is observational learning. Siblings learn and develop by observing and imitating one another, both in terms of positive (learning social competencies) and negative (conflict, hostility, aggression) behaviours (Bandura, 1977; Whiteman et al., 2011). Thus, HR-sibs

might imitate the more negative behaviours from their older brother/sister with ASD. Third, genetic factors can lead to (subclinical) characteristics of ASD in HR-sibs as well, leading to more negative behaviours during social interaction.

Child characteristics (i.e., negative initiations and responses) significantly differed between groups, which could have influenced the group differences found in the present study. Regarding the increased levels of negative behaviours in the HR group, regression analyses confirmed that sample characteristics could not better explain these group differences. Although the age of the oldest child influenced the association between group status (high-risk vs. low-risk) and negative behaviours, group status remained a significant predictor. Thus, the group differences in terms of negative behaviour are most likely genuine group differences between the LR and HR group. On the other hand, positive behaviours were associated with gender and not with group status. In general, positive behaviours were more frequent in sibling pairs with an older sister. Given the lower proportion of older sisters in the HR group, a lack of group differences in positive behaviour could be caused by group differences in terms of gender. Unfortunately, due to the limited number of girls with ASD, we could not compare HR sibling pairs with an older sister with HR sibling pairs with an older brother and evaluate gender differences within the HR group. We also need to take into consideration that LR-sibs attended day care more frequently than HR-sibs, meaning that LR-sibs had more social experiences than HR-sibs. These experiences could improve the LR-sibs' social-communicative abilities, which could in turn influence their behaviour during sibling interaction.

It is noteworthy that the group differences changed as a function of the play materials. One possible explanation lies in the type of interactions that the play materials trigger. Since there was only one keyboard (i.e., limited resources), the risk of conflict during play with keyboard was higher than during play with marble run/blocks. Accordingly, siblings in the LR group indeed showed higher levels of negative initiations during keyboard than during marble run/blocks. However, this was not the case in the HR group. It is possible that, while the LR group only showed an increase in negative behaviour as a response to the limited resources, levels of negative behaviour in the HR group were higher regardless of the play set. As a result, the difference in negative

behaviour between both groups was significant during play with marble run/blocks, but not during keyboard (since both groups display higher levels of negative behaviour).

This study has some limitations that need to be acknowledged. Although significant findings were observed, the small sample size reduced the power of the study and the likelihood of detecting significant results. After applying the Bonferroni correction, most significant results were no longer significant. This is possibly due to a decrease in power and does not necessarily mean that there are no real world differences. For negative initiations of the oldest sibling and negative responses of the youngest sibling (marble run/blocks), group remained a significant predictor even after correcting for multiple testing. Nevertheless, results need to be interpreted with caution. Second, parametric analyses, based on a larger sample size, would have allowed for more elaborate analyses such as controlling for the effect of sample characteristics. However, developmental level and family SES showed limited associations with characteristics of the sibling interaction. The age of the oldest sibling did predict the sibling interaction, but did not eliminate group as a significant predictor. Thus, it is unlikely that these sample characteristics influenced the group differences found in the current study. In addition, the average mental age of the children with ASD was somewhat lower than their chronological age, reducing the gap with typically developing older children. Since the cognitive functioning of several children with ASD was below average, this could have influenced the results as well. However, the association between cognitive functioning and the sibling interaction characteristics in the HR group was limited. Regarding the play with marble run/blocks, cognitive functioning was not correlated with the sibling interaction characteristics that differed between groups (i.e., negative initiations/responses). Regarding play with keyboard, only the association with negative initiations of the youngest child was significant. Gender differences in sibling relationships should be further evaluated in a larger sample, including a higher number of girls with ASD. We also need to take into account that these differences in sample characteristics could be inherent to the ASD population

Future research should focus on replicating the current results in a larger sample, controlling for relevant sample characteristics. Second, not only younger siblings' development is affected by the presence of a child with ASD in the family. The development of older siblings of children with ASD should be considered as well. Third,

the relationship between sibling interactions and child outcome could be influenced by social interactions with other family members. Due to practical reasons, the current study was unable to include sibling interactions between HR-sibs and an older sibling without ASD. Finally, the children were too young for diagnostic assessment. Therefore we were unable to evaluate differences between typically developing HR-sibs and HR-sibs with characteristics of BAP or ASD.

The results of the current study raise theoretical implications. While the development of HR-sibs is increasingly being studied, the current study is the first to consider sibling interactions between very young HR-sibs and their older sibling with ASD as a possible link between early vulnerabilities and later outcome. Even with the overall level of interaction being rather low, the sibling interactions in the HR group were clearly more negative than in the LR group. Altered early sibling interactions in terms of higher levels of negative behaviour could influence the learning environment of young HR-sibs, possibly changing their early developmental trajectory (Dawson, 2008; Seibert, Hogan, & Mundy, 1982). If the higher level of negative initiations and responses disturbs the balance between positive and negative, this could lead to higher levels of internalising or externalising problems and lower social competence in both children (Buist & Vermande, 2014). Although conflict and conflict resolution can benefit development as well (e.g., Bedford et al., 2012), negative behaviours in the current study are not always related to conflict (e.g., taking something from the sibling). Longitudinal studies could provide further evidence on the pathway between early sibling interactions and child outcome. For example, in their longitudinal study Wan et al. (2013) provided support for the association between early parent-child interactions at 12 months and ASD outcome at 36 months.

Second, if indeed differences in early sibling interactions lead to differences in developmental trajectories of HR-sibs, sibling interactions should be targeted in early interventions in ASD, taking into account the development of both siblings. In this way, interventions can promote positive sibling relationships and individual adjustment of both siblings (McHale et al., 2016). Interventions that target those aspects of sibling interactions that negatively influence child development, could improve the later outcome of both HR-sibs and children with ASD. Nevertheless, sibling interactions should be considered within the entire family system, including the interaction with

other family members (e.g., parents). Targeting sibling interactions could be part of a broader intervention or could be included in specific programs such as home guidance.

Despite the limitations, this study provides new insights in the early learning environment of HR-sibs. To date, the existing research on sibling relationships in families with a child with ASD is limited, especially including siblings as young as 18 months. The present study demonstrates that, by using naturalistic observations (i.e., observing behaviour in a natural setting), differences in early sibling interactions between ASD dyads and typically developing dyads can be detected. Further research is required to determine the impact of these sibling interactions on child development and family functioning, and to assess whether higher levels of negativity support or compromise the development of HR-sibs. This could in turn provide insights on the potential value of sibling interactions in early intervention.

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**SOCIAL INTERACTIONS BETWEEN  
24-MONTH-OLD CHILDREN AND THEIR OLDER  
SIBLING WITH AUTISM SPECTRUM DISORDER:  
CHARACTERISTICS AND ASSOCIATION WITH  
SOCIAL-COMMUNICATIVE DEVELOPMENT<sup>1</sup>**

ABSTRACT

This study compared sibling interactions between 24-month-old children and their older sibling with ASD (high-risk;  $n=24$ ) with 24-month-old children and their typically developing older sibling (low-risk;  $n=32$ ). First, high-risk sibling pairs showed lower levels of positive behaviour and younger siblings of children with ASD imitated their older sibling less. Second, in the high-risk group positive interactions were positively associated with the youngest child's language abilities. However, this association was no longer significant after controlling for language abilities at 14 months. Third, more total interactions in the high-risk group, both negative and positive, were associated with more ASD characteristics. Thus, early sibling interactions might reveal interesting information in light of the (atypical) developmental trajectories of younger siblings of children with ASD.

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## INTRODUCTION

Autism spectrum disorder (ASD) is characterised by persistent deficits in social communication and social interaction (American Psychiatric Association, 2013). These deficits, including atypicalities in eye contact, joint attention, responsiveness (to social cues), imitation, and social orienting or interest, are often evident in the first two years of life (Bryson et al., 2007; Osterling, Dawson, & Munson, 2002; Wetherby, Watt, Morgan, & Shumway, 2007; Zwaigenbaum et al., 2005). In addition, receptive as well as expressive language development is frequently delayed and/or deviant in children with ASD (Barbaro & Dissanayake, 2012) and clinically significant structural language impairments are common (Boucher, 2012).

Siblings of children with ASD (hereafter, *high-risk siblings*; HR-sibs) are up to 20 times more likely to be diagnosed with ASD compared to the general population (Ozonoff et al., 2011). In addition, HR-sibs more frequently show subclinical features of ASD, also referred to as the Broader Autism Phenotype (BAP) (Ozonoff et al., 2014; Sucksmith, Roth, & Hoekstra, 2011). This includes delays in social communication such as the use of eye contact, gestures, and orientation to name (Gamliel, Yirmiya, & Sigman, 2007; Gammer et al., 2015; Mitchell et al., 2006; Toth, Dawson, Meltzoff, Greenson, & Fein, 2007). Aside from BAP, HR-sibs without ASD also show more language difficulties, such as delays in receptive language (Hudry et al., 2014; Toth et al., 2007), or are delayed in their cognitive development during the first three years of life (Brian et al., 2014). Thus, the developmental trajectories of HR-sibs are often characterised by early deficits, irrespective of a later ASD diagnosis. Consequently, studies evaluating possible risk or protective factors for HR-sibs with atypical developmental trajectories would be valuable.

The heritability of the susceptibility to ASD is estimated between 64 and 91%, dependent on the prevalence rate used (from 1% up to 5% for BAP) (Tick, Bolton, Happé, Rutter, & Rijdsdijk, 2016). In addition, environmental factors and the gene-environment interaction need to be considered as well (Mandy & Lai, 2016), particularly at a young age when brain plasticity is high and social communication and language develop rapidly (Barbaro & Dissanayake, 2012; Elsabbagh & Johnson, 2010). Although it is clear that the social environment does not *cause* ASD, it can influence the manifestation of the ASD



phenotype and its functional impact (Mandy & Lai, 2016). Early child characteristics such as social-communicative and language impairments can impact upon the social interactions with family members. This can for example result in a diminished active engagement in social interaction, which may lead to a limited exposure to adequate social input. Since social input is needed to promote the development of social communication and language during early sensitive periods, altered social interactions can mediate the link between early susceptibilities and later outcome (Boucher, 2012; Dawson, 2008; Mandy & Lai, 2016). Nevertheless, in comparison to genetic and neurobiological research, research on the social environment in ASD is limited.

During infancy and early childhood, the most important social interaction partners are caregivers and siblings (Lamb, 1978). Sibling interactions have an impact on the social-communicative, emotional, cognitive and behavioural development of young children (Buist & Vermande, 2014; Harrist et al., 2014). During these interactions there is a bidirectional influence of the characteristics and behaviours of both interaction partners (Gottlieb, 2007; Pettit & Arsiwalla, 2008), changing the nature of the interaction over time. Warm sibling interactions characterised by natural teaching and caregiving experiences benefit the development of both siblings (Brody, 2004; Buist & Vermande, 2014; Feinberg, Solmeyer, & McHale, 2012). In addition, more positivity in the sibling relationship and more positive behaviours of the older sibling are linked to better empathy development of the younger sibling (Tucker, Updegraff, McHale, & Crouter, 1999). Conversely, negative sibling interactions can lead to poorer developmental outcomes (Bank, Patterson, & Reid, 1996). Sibling interactions that mainly consist of conflict lead to higher levels of anxiety and depression, and lower levels of academic or social competence and global self-worth (Buist & Vermande, 2014). However, some level of conflict, in balance with warmth, can promote the development of anger management and conflict resolution skills (Brody, Stoneman, & Mackinnon, 1982).

Siblings influence each other through *social learning*, including observing each other, immediate or deferred imitation and modelling (Bandura, 1977; Feinberg et al., 2012; Whiteman, McHale, & Soli, 2011). Younger siblings are more likely to imitate their older brother or sister than vice versa (Whiteman, Bernard, & McHale, 2010). Through observing, remembering and imitating actions from their older sibling, HR-sibs might learn ASD-specific behaviours resulting in a behavioural phenotype that resembles the

BAP or the early ASD phenotype. In addition, due to the presence of social-communicative and language impairments in children with ASD and possibly in HR-sibs as well, their sibling interactions may lack social quality or occur less frequently (i.e., a reduction of adequate social input), which also influences the HR-sibs' development. There is some evidence suggesting that lower levels of social input or less positivity during sibling interactions are associated with deficits in the development of language and empathy (Kuhl, 2004; Tucker et al., 1999).

Although research on characteristics of sibling interactions including a child with ASD is scarce, it provides some support for the reduced social interactions within HR sibling pairs. The studies of Knott, Lewis, and Williams (1995, 2007) found that, in comparison to children with Down syndrome, children with ASD (age range: 3;10-9;0 years) initiated fewer interactions, were less responsive and spent less time with their younger/older sibling (age range: 1;11-12;5 years). Walton and Ingersoll (2015) reported that HR-sibs (mean age: 10.43 years) were less involved and more avoidant in interaction with their brother/sister with ASD (mean age: 9.35 years), compared to typically developing sibling pairs. In the study of Kaminsky and Dewey (2001), based on self-report, HR-sibs (mean age: 11.67 years) reported less conflict than siblings of typically developing children. However, since early signs of ASD are already visible in the first two years of life (e.g., Barbaro & Dissanayake, 2012; Zwaigenbaum et al., 2005), and given that the transactional processes between infants and the social environment start from birth onwards, studying sibling interactions in a younger age group is necessary to increase our understanding of the characteristics of sibling interactions including a child with ASD. In addition, since sibling interactions are associated with children's social-communicative functioning in typical development, these associations should also be evaluated in sibling pairs including a child with ASD.

The present study aimed to observe sibling interactions in 24-month-old HR-sibs and their older siblings with ASD. These HR sibling pairs were compared with low-risk (LR) sibling pairs of 24-month-old LR-sibs and a typically developing older sibling to evaluate whether sibling interactions differed between both groups. In line with the studies of Knott et al. (1995, 2007) and Walton and Ingersoll (2015), suggesting fewer interactions and less involvement in HR sibling pairs, and considering the social-communicative and language impairments in children with ASD as well as a considerable proportion of HR-

sibs, we expected lower levels of social interaction in HR sibling pairs compared to LR sibling pairs.

Second, to determine to what extent HR-sibs could learn from their sibling through *immediate* imitation (social learning), we evaluated to what degree HR-sibs imitated their older sibling with ASD in comparison with low-risk controls. In line with research reporting impaired immediate imitation in HR-sibs (Stone, McMahon, Yoder, & Walden, 2007; Zwaigenbaum et al., 2005), we expected that HR-sibs would imitate their sibling less than LR-sibs.

Finally, the association between the frequency of sibling interactions and the youngest siblings' social-communicative and language abilities at 24 months was evaluated. If the relationship between early susceptibilities of HR-sibs and later outcome is mediated through sibling interactions, we would expect an association between sibling interactions and the HR-sib's current development. Based on research in typically developing sibling pairs, we expected positive associations between warm/positive sibling interactions and social-communicative and language skills. Regarding negative sibling interactions, existing literature is inconsistent reporting both positive and negative associations with child development (Bedford, Volling, & Avioli, 2000; Buist & Vermande, 2014). Hence, we were not able to formulate specific hypotheses or expectations with regard to negative sibling interactions.

## METHOD

### Participants

Participants were 24-month-old children and their older sibling who were drawn from an ongoing prospective follow-up study of both younger siblings of children with ASD (high-risk siblings; HR-sibs) and a control group of younger sibling of typically developing children (low-risk siblings; LR-sibs). The sample comprised 56 sibling pairs, including 24 high-risk sibling pairs (9 male-male, 8 female-male, 2 male-female and 5 female-female; younger-older) and 32 low-risk sibling pairs (9 male-male, 9 female-male, 10 male-female and 4 female-female). LR sibling pairs consisted of LR-sibs and their older typically developing sibling (TD-sibs) without first- or second-degree relatives with ASD. HR sibling pairs included HR-sibs and their older sibling with a formal ASD diagnosis

(ASD-sibs). The ASD diagnosis was made by a multidisciplinary team and confirmed with the Social Responsiveness Scale, Second Edition (SRS-2; Constantino & Gruber, 2012), and the Social Communication Questionnaire (SCQ; Rutter, Bailey, & Lord, 2003). SCQ and SRS were available for all 24 children with ASD. Fifteen children scored above the threshold for ASD on both the SCQ and the SRS, the other nine scored above the threshold on the SRS. As part of the multidisciplinary assessment, cognitive functioning of children with ASD was evaluated using either the Wechsler Intelligence Scale for Children (WISC-III-NL; Kort et al., 2005), the Wechsler Preschool and Primary Scale of Intelligence (WPPSI-III-NL; Hendriksen and Hurks, 2009), the Snijders-Oomen Non-Verbal Intelligence Test (SON-R; Tellegen et al., 1998), or the Bayley Scales of Infant Development (BSID-II-NL; Meulen et al., 2004; Bayley-III-NL; Baar et al., 2014). Eleven of the children with ASD scored within the normal range (IQ between 85-115). Of the other 13 children, 3 scored very low (IQ<55), 9 children scored below average (IQ between 55-85), and 1 child scored above average (IQ>115).

Sample characteristics are presented in Table 1. To calculate the family's socioeconomic status (SES), Hollingshead's four factor index was used based on both parents' education level and occupation (Hollingshead, 1975). There were no significant group differences in the sex ratio of both younger and older siblings, chronological age of the youngest sibling or family SES. ASD-sibs were on average older than TD-sibs ( $F(1,54)=23.498, p<.001$ ). To assess the social experiences of the younger siblings, parents were asked whether or not their youngest child attended day care and how often both siblings were together at home (seldom/sometimes/often). As shown in Table 1, LR-sibs more frequently attended day care than HR-sibs (93% vs. 70%;  $\chi^2(1)=5.22, p=.031$ ). In addition, siblings in the LR group spent more time together than siblings in the HR group ( $\chi^2(1)=8.65, p=.013$ ).

## **Procedure**

As part of the prospective follow-up study, both HR- and LR-sibs were assessed at 24 months. This included the Mullen Scales of Early Learning (MSEL; Mullen, 1995), the Autism Diagnostic Observation Schedule-Second Edition (ADOS-2; Lord et al., 2012), the Dutch version of the MacArthur-Bates Communicative Development Inventory (N-CDI; Fenson et al., 1993; Zink & Lejaegere, 2002), and the Quantitative Checklist for Autism in Toddlers (Q-CHAT; Allison et al., 2008).

**Table 1***Sample characteristics and general description of the play observation*

	Low-risk (n=32)	High-risk (n=24)	
	Sibling pair		
<b>Family SES (M(sd))</b>	51.81 (7.00)	45.67 (11.39)	$U = 269.50$
<b>Time spent together (%)</b>			$\chi^2(1) = 8.65^*$
Never/seldom	7%	35%	
Sometimes	23%	30%	
Often/always	70%	35%	
<b>Day care attendance (%)</b>	93%	70%	$\chi^2(1) = 5.22^*$
<b>Representative? (% yes)</b>	<b>83.3%</b>	<b>78.3%</b>	$\chi^2(1) = .219$
	Youngest sibling		
<b>Chronological age</b>			
M(sd)	24.75 (.77)	24.69 (.77)	$F(1,54) = .072$
Range	23.23-27.03	23.23-26.40	
<b>Sex ratio (M:F)</b>	19:13	11:13	$\chi^2(1) = 1.01$
<b>Interaction (%)</b>	<b>19.4%</b>	<b>19.6%</b>	$U = 383.00$
Mutuality	4.2%	4.6%	$U = 332.00$
Interaction with parent	6.6%	7.8%	$U = 382.00$
Interaction with experimenter	8.6%	7.2%	$U = 342.00$
<b>Non-interaction (%)</b>	<b>80.6%</b>	<b>80.4%</b>	$U = 383.00$
Orientation to sibling	7.1%	9.3%	$U = 316.00$
Solitary play	73.5%	71.1%	$U = 330.00$
	Oldest sibling		
<b>Chronological age</b>			
M(sd)	55.69 (13.91)	87.85 (34.00)	$F(1,54) = 23.498^{***}$
Range	36.50-97.03	47.43-154.37	
<b>Sex ratio (M:F)</b>	18:14	17:07	$\chi^2(1) = 1.24$
<b>Interaction (%)</b>	<b>16.5%</b>	<b>18.4%</b>	$U = 339.00$
Mutuality	4.1%	4.7%	$U = 333.00$
Interaction with parent	4.1%	7.1%	$U = 372.00$
Interaction with experimenter	8.3%	6.6%	$U = 318.50$
<b>Non-interaction (%)</b>	<b>83.5%</b>	<b>81.6%</b>	$U = 339.00$
Orientation to sibling	4.2%	6.5%	$U = 309.50$
Solitary play	79.3%	75.1%	$U = 324.50$

Note. Chronological age is reported in months; \* $p < .05$ , \*\*\* $p < .001$ ; M=mean, sd=standard deviation

An additional appointment was scheduled at the participants' home to observe sibling interactions. Children were encouraged to play together at the beginning of each session. They were given zoo-themed building blocks, a marble run and an animal sound keyboard, with which they could play consecutively for 10, 10 and 5 minutes. Different sets of toys were chosen to elicit different kinds of play (parallel, associative and cooperative play). Since there were no clear systematic differences in sibling interaction characteristics between the three play contexts, the scores were summed and considered as one play interaction. During the observation, one parent was always present in the room, continuing normal routines (e.g., household tasks or work). Parents were asked not to interfere during the play observation. If children initiated social interaction with the parent, they could respond briefly as they normally would. At the beginning of each appointment, parents received general information about the study and were asked to sign an informed consent.

### **Measures**

The *ADOS-2* (Lord et al., 2012) is a semi-structured, standardised assessment of communication, social interaction, play/imaginative use of materials, and restricted and repetitive behaviours. Based on the child's language level, either the toddler module (82%) or module 2 (18%) was administered. In line with Shephard et al. (2016) Calibrated Severity Scores were used for Social Affect, Repetitive and Restricted Behaviours, and Total Score (Gotham, Pickles, & Lord, 2009; Hus, Gotham, & Lord, 2014) to account for differences in module administration and language level.

The *MSEL* (Mullen, 1995) is a comprehensive measure of five developmental domains for infants and preschool children (0-68 months): Gross Motor, Fine Motor, Visual Reception, Receptive Language, and Expressive Language. Overall cognitive ability is represented by the Early Learning Composite (ELC). The *MSEL* has demonstrated good internal consistency and test-retest stability (Mullen, 1995).

The *N-CDI* (Fenson et al., 1993; Zink & Lejaegere, 2002), is a parent-report measure of receptive and expressive vocabulary. It yields meaningful raw counts of word comprehension as well as word production.

The *Q-CHAT* (Allison et al., 2008) contains 25 items, scored on a 5-point scale, and is a screening tool to identify ASD-symptoms in toddlers. It is especially useful in the identification of threshold and sub-threshold autistic features and has potential as a quantitative phenotypic measure (Allison et al., 2008).

### **Sibling interaction**

All play sessions were videotaped. For play with marble run and blocks, both lasting 10 minutes, the middle 8 minutes were selected and coded using The Observer XT, version 11.5 (Noldus, 2013). For play with keyboard, lasting 5 minutes, the middle 4 minutes were selected for coding. First, a frequency coding scheme was used. Frequencies of social initiations and responses, both negative and positive, were coded. *Social initiations* are communicative attempts to initiate a new interaction, directed towards another individual. *Responses* are related to and follow a previous initiation within five seconds. Initiations and responses can be either positive/prosocial (e.g., sharing a toy, allowing the other sibling to do something) or negative (e.g., refusing a request). Next, the time children spent in interaction with each other (mutuality), with the parent and with the experimenter was also coded. To account for the time not spent in interaction with another person, the following non-interactive behaviours were coded: distress, doing nothing or looking at a random object, orientation towards the sibling or sibling's activity, repetitive/stereotyped behaviour, and time spent in a purposeful activity (e.g., play).

Second, to obtain a broader evaluation of the course of the play observation, five global rating scales were included. Each scale ranged from 1 (low frequency/quality) to 5 (high frequency/quality). *Interference of the parent* refers to the extent to which the parent interfered or interrupted during the play observation. *Proximity* indicates the distance between both children during play. In this scale, interpersonal distance is taken into account as well. Two children who are further away in distance but are in close interaction (e.g., dancing together from a distance), are considered to be in close proximity. *Imitation* of the younger as well as the older sibling was coded when the child shows behaviour that is a direct and exact repetition of the other child. Finally, *togetherness* reflects the degree to which both children are enjoying the interaction

*together*. Examples of togetherness are: warmth, positive affect, joint pleasure, engagement in a joint activity, mutuality, sharing, etc.

Clips were independently rated by trained master students blind to the participants' diagnostic status. Approximately 15% of the clips were randomly selected to determine interrater agreement and were coded by all coders. Next, single measures intraclass correlation coefficients (ICC) were calculated. ICC's between .60-.74 reflect good interrater agreement and ICC's between .75-1.00 reflect excellent interrater agreement (Cicchetti, 1994). Due to their low frequency, the following behaviours could not be coded reliably (ICC<.60) and are therefore excluded from further analyses: distress, doing nothing or looking at a random object, repetitive/stereotyped behaviour, and imitation by the oldest sibling. For the frequency coding scheme, ICC's of the included behaviours ranged between .74 and .95 for the youngest child and between .76 and .96 for the oldest child. For the global rating scales, ICC's ranged between .76 and .84.

### **Data-analysis**

Preliminary analyses revealed several outliers in the data (i.e., values higher/lower than the mean +/- 3 times the standard deviation (sd)). Since outliers were not considered to be random but characteristic of our sample, outliers were replaced by the highest/lowest value allowed (mean +/- 3sd) rather than deleted.

Concerning the *first research question*, we first provided a general description of the play observation. To this end, proportions were calculated of how long children were engaged in different types of behaviour (i.e., proportion of time spent in interaction, play, etc.) and several global scales (interference of parent, proximity, togetherness) were evaluated. Proportions and global ratings were compared between groups using non-parametric tests (Mann-Whitney U). Second, it was evaluated whether group status predicted social initiations and responses (positive and negative), accounting for sample characteristics that differed between groups (the age of the oldest sibling, day care attendance, and time spent together). Accordingly, regression models including 'group' (high-risk vs. low-risk) and these sample characteristics as predictors and sibling interaction characteristics as dependent variables were tested.



Regarding the *second research question*, it was evaluated whether group status predicted imitation of the youngest child. To this end, a regression model with group (high-risk vs. low-risk) and sample characteristics (age oldest child, day care attendance, time spent together) as predictors was tested with imitation of the youngest sibling as dependent variable.

To answer the *third research question* and evaluate the association between sibling interactions and child development, regression models including the sibling interaction characteristics as predictors and language and social-communicative abilities at 24 months as dependents were evaluated. However, it is possible that pre-existing language abilities of HR-sibs influenced the association between the sibling interaction characteristics and language (MSEL, N-CDI) at 24 months. Therefore, scores on the MSEL and N-CDI at 14 months were added as predictors in the regression model to determine whether the sibling interaction characteristics would still significantly predict development at 24 months when taking development at 14 months into account.

Correlation analyses revealed high intercorrelations between several child interaction variables, leading to multicollinearity in the regression model. Especially positive initiations and positive responses of both children were significantly ( $p < .05$ ) intercorrelated as well as negative initiations and negative responses. Correlations between positive behaviours ranged from  $r = .33$  to  $r = .85$  while correlations between negative behaviours ranged from  $r = .43$  to  $r = .82$ . To address the problem of multicollinearity, a total interaction composite was created by summing all behaviours, both positive and negative. This allowed us to evaluate whether more interaction, regardless of its nature, would predict development. In addition, positive initiations/responses of both children on the one hand and negative initiations/responses of both children on the other hand were summed to form two composite scores: *positive behaviour* and *negative behaviour*. Reliability analyses revealed a good internal consistency for both composite scores with Cronbach's alpha's of .81 for positive behaviour and .88 for negative behaviour.

## RESULTS

### General description of the play observation

To get a general idea of the course of the play observations, it was evaluated how much time children spent in *direct* mutuality with their sibling (i.e., a bout of interaction characterised by initiations and responses, either positive or negative, and lasting at least a few seconds), in interaction with the parent/researcher, or engaged in non-interactive activities. These proportions are presented in Table 1 and did not significantly differ between groups.

**Interaction.** In both groups, children spent 16-20% of the play observation in interaction with another interaction partner (sibling/parent/researcher). Of the total play session, siblings only spent less than 5% in *mutual interaction* with each other. The overall feel of *togetherness* (i.e., global rating of the degree to which both children are enjoying the interaction together) was 1.85 in the LR group and 1.68 in the HR group, meaning that there were short instances of togetherness between both children, but not frequently. The difference between groups was not significant ( $U=270.50$ ,  $p=.091$ ). The average *proximity* between both children was 3.92 (frequent proximity) in the LR group and 3.46 (occasional to frequent proximity) in the HR group, but did not significantly differ between groups ( $U=272.50$ ,  $p=.102$ ). In addition to the interaction with each other, children also interacted with their parent(s) (4-8%) or with the researcher (7-9%).

**Non-interaction.** Although often in close proximity, the majority of the play observation consisted of solitary play (71-79%). Of the remaining time, children spent 4 to 9% of their time observing their sibling.

Parents were asked to remain in the room while the children were playing and to only intervene when absolutely necessary. In both groups, the average score on *interference of the parent* was around 2, meaning that parents only sporadically intervened during the play observation. *Interference of the parent* did not significantly differ between groups ( $U=306.50$ ,  $p=.292$ ). In addition, the majority of parents indicated that the observed play observation was representative for a typical play observation at home (LR: 83%; HR: 78%).

### Group differences in social interaction and imitation

It was evaluated whether group status (high-risk vs. low-risk) predicted social initiations and responses as well as imitation of the youngest child while accounting for sample characteristics. Descriptives of the sibling interaction characteristics are shown in Table 2. Regression models and significant regression coefficients are presented in Table 3.

**Table 2**

*Descriptives (mean(standard deviation)) for sibling interaction characteristics*

	LR	HR
	Frequency - Youngest sibling	
Negative initiations <sup>a</sup>	4.04(5.03)	2.44(2.66)
Positive initiations <sup>a</sup>	5.48(4.51)	3.38(3.52)
Negative responses <sup>a</sup>	6.73(5.93)	7.83(6.69)
Positive responses <sup>a</sup>	16.49(8.93)	12.14(10.83)
Mutuality <sup>b</sup>	48.08(45.74)	54.68(79.76)
Interaction with experimenter <sup>b</sup>	99.41(78.74)	83.18(66.51)
Interaction with parent <sup>b</sup>	75.82(75.49)	90.62(113.86)
Orientation to sibling <sup>b</sup>	81.37(50.93)	106.54(75.76)
Play <sup>b</sup>	849.71(128.73)	823.45(118.83)
	Frequency - Oldest sibling	
Negative initiations <sup>a</sup>	10.13(5.93)	9.48(6.16)
Positive initiations <sup>a</sup>	10.95(8.72)	9.75(14.06)
Negative responses <sup>a</sup>	5.48(5.99)	5.11(4.98)
Positive responses <sup>a</sup>	7.93(6.36)	5.70(6.86)
Mutuality <sup>b</sup>	46.42(42.18)	54.28(79.63)
Interaction with experimenter <sup>b</sup>	97.55(91.02)	77.22(86.84)
Interaction with parent <sup>b</sup>	47.20(48.52)	83.50(114.87)
Orientation to sibling <sup>b</sup>	48.18(65.12)	76.06(108.00)
Play <sup>b</sup>	927.82(129.52)	880.50(165.92)
	Global rating scales	
Togetherness <sup>c</sup>	1.85(.55)	1.68(.84)
Proximity <sup>c</sup>	3.92(.84)	3.46(1.04)
Imitation youngest <sup>c</sup>	1.54 (.62)	1.20 (.26)
Interference <sup>c</sup>	1.98(.62)	2.25(.87)

Note. LR=low-risk, HR=high-risk; <sup>a</sup>results reflect absolute frequencies; <sup>b</sup>results reflect total duration (in seconds); <sup>c</sup>results reflect global rating (1-5)

**Table 3**

*Prediction of sibling interaction characteristics: Regression coefficients for significant predictors*

		<i>B</i>	<i>SE B</i>	$\beta$
	Youngest sibling			
<b>Positive initiations</b>	<b><math>R^2=.186, F(4,48)=2.734, p=.040</math></b>			
Group		-4.027	1.401	-.476**
Age oldest sibling		.051	.025	.362*
<b>Negative initiations</b>	<b><math>R^2=.076, F(4,48)=.981, p=.427</math></b>			
<b>Positive responses</b>	<b><math>R^2=.372, F(4,48)=7.119, p&lt;.001</math></b>			
Group		-11.192	2.934	-.555***
Age oldest sibling		.255	.052	.755***
<b>Negative responses</b>	<b><math>R^2=.092, F(4,48)=1.221, p=.314</math></b>			
<b>Imitation</b>	<b><math>R^2=.227, F(4,48)=3.458, p=.015</math></b>			
Group		-.410	.172	-.384*
Time spent together		.228	.100	.331*
	Oldest sibling			
<b>Positive initiations</b>	<b><math>R^2=.476, F(4,48)=10.890, p&lt;.001</math></b>			
Group		-10.673	3.028	-.469**
Age oldest sibling		.344	.053	.902***
<b>Negative initiations</b>	<b><math>R^2=.032, F(4,48)=.401, p=.807</math></b>			
<b>Positive responses</b>	<b><math>R^2=.389, F(4,48)=7.633, p&lt;.001</math></b>			
Group		-7.711	1.920	-.577***
Age oldest sibling		.165	.034	.735***
<b>Negative responses</b>	<b><math>R^2=.045, F(4,48)=.561, p=.692</math></b>			

Note. \* $p<.05$ , \*\* $p<.01$ , \*\*\* $p<.001$ ;

First, the regression models for positive behaviours of the youngest and oldest sibling were significant. Group status significantly predicted positive initiations of the youngest child ( $\beta=-.476, t=-2.874, p=.006$ ), positive responses of the youngest child ( $\beta=-.555, t=-3.814, p<.001$ ), positive initiations of the oldest child ( $\beta=-.469, t=-3.525, p=.001$ ),

and positive responses of the oldest child ( $\beta=-.577$ ,  $t=-4.016$ ,  $p<.001$ ). All four behaviours occurred more frequently in the LR group compared to the HR group. The regression models for negative behaviours were not significant.

Second, the regression model for imitation of the youngest child was also significant. Group significantly predicted imitation ( $\beta=-.384$ ,  $t=-2.384$ ,  $p=.021$ ), with higher levels of imitation in LR-sibs than in HR-sibs.

Third, sample characteristics significantly predicted characteristics of the sibling interaction. *Age of the oldest sibling* significantly predicted positive initiations of both children (youngest:  $\beta=.362$ ,  $t=2.077$ ,  $p=.043$ ; oldest:  $\beta=.902$ ,  $t=6.452$ ,  $p<.001$ ) and positive responses of both children (youngest:  $\beta=.755$ ,  $t=4.935$ ,  $p<.001$ ; oldest:  $\beta=.735$ ,  $t=4.871$ ,  $p<.001$ ). All four behaviours were more frequent in older children. In addition, *time spent together* positively predicted imitation of the youngest sibling ( $\beta=.331$ ,  $t=2.284$ ,  $p=.027$ ).

### **Association with social-communicative and language abilities**

Next, regression models were tested including the three sibling interaction composites (positive, negative, and total interaction) and imitation (at 24 months) as predictors. For each dependent variable, two regression models were tested. In a first model, the predictive value of the total interaction was tested. In a second model, positive and negative behaviour were added as two separate variables to evaluate whether the valence of the interaction would predict development.

Bivariate correlations between the sibling interaction characteristics (composite scores, imitation) and scores on the N-CDI, MSEL language scales, ADOS (Social Affect, Repetitive and Restricted Behaviours, and Total Score), and Q-Chat at 24 months were examined to guide the selection of dependent variables entered in the regression analysis. Because correlational patterns differed between groups, both groups were analysed separately. In the LR group, *total interaction* correlated negatively with N-CDI word comprehension ( $r=-.430$ ). In addition, the youngest child's imitation of their older sibling was positively correlated with N-CDI word production ( $r=.467$ ). In the HR group, *negative behaviour* correlated positively with N-CDI word production ( $r=.480$ ) and the Q-Chat total score ( $r=.478$ ). *Positive behaviour* correlated positively with MSEL receptive

language ( $r=.458$ ), MSEL expressive language ( $r=.453$ ), and the Q-Chat total score ( $r=.465$ ). *Total interaction* positively correlated with N-CDI word production ( $r=.468$ ), MSEL receptive language ( $r=.492$ ), MSEL expressive language ( $r=.505$ ) and Q-Chat total score ( $r=.566$ ). No significant correlations were found between the sibling interaction variables and the ADOS scores.

Next, regression models were tested. In the LR group, *total interaction* negatively predicted N-CDI word comprehension, accounting for 18.5% of the variance. Imitation of LR-sibs positively predicted N-CDI word production, accounting for 22% of the variance. In the HR group, *total interaction* positively predicted both MSEL receptive language and MSEL expressive language, explaining 24% and 27% of the variance, respectively. In addition, *total interaction* positively predicted the Q-Chat total score, accounting for 32% of the variance. Results are presented in Table 4.

**Pre-existing language abilities: language at 14 months.** To determine whether sibling interaction characteristics would still predict language at 24 months when controlling for language at 14 months, pre-existing language abilities were taken into consideration. In the *high-risk group*, correlational analyses revealed significant correlations between MSEL receptive language at 14 months and MSEL receptive language at 24 months ( $r=.546$ ), and between MSEL receptive language at 14 months and MSEL expressive language at 24 months ( $r=.750$ ). In addition, both N-CDI word production and N-CDI word comprehension at 14 months correlated significantly with N-CDI word comprehension (word comprehension:  $r=.795$ ; word production:  $r=.597$ ) and N-CDI word production (word comprehension:  $r=.814$ ; word production:  $r=.687$ ) at 24 months. In the *low-risk group*, there was a significant positive correlation between N-CDI word comprehension and N-CDI word production at 14 months and N-CDI word comprehension at 24 months (word comprehension:  $r=.702$ ; word production:  $r=.530$ ). In addition, N-CDI word comprehension at 14 months was associated with N-CDI word production at 24 months ( $r=.436$ ). Correlations between MSEL scores at 14 and 24 months were not significant.

Hierarchical regression models were tested for MSEL receptive language and MSEL expressive language in the high-risk group, and N-CDI word comprehension in the low-risk group. At step 1, the sibling interaction composite *total interaction* was added. At step 2, the MSEL or N-CDI scores at 14 months were added.

**Table 4**

*Prediction of language and social-communicative development: Regression models and predictor coefficients*

		Low-risk group		
		<i>B</i>	<i>SE B</i>	$\beta$
<b>N-CDI_WC</b>	<b>1. <math>R^2=.185, F(1,24)=5.448, p=.028</math></b>			
	Total interaction	-.221	.095	-.430*
	<b>2. <math>R^2=.189, F(2,23)=2.683, p=.090</math></b>			
	Positive behaviour	-.191	.130	-.276
	Negative behaviour	-.289	.170	-.320
<b>N-CDI_WP</b>	<b>1. <math>R^2=.22, F(1,24)=6.683, p=.016</math></b>			
	Imitation	17.204	6.655	.467*
		High-risk group		
<b>N-CDI_WP</b>	<b>1. <math>R^2=.22, F(1,43)=3.916, p=.068</math></b>			
	Total interaction	.340	.172	.468 <sup>+</sup>
	<b>2. <math>R^2=.27, F(2,13)=2.411, p=.129</math></b>			
	Positive behaviour	.205	.243	.210
	Negative behaviour	.677	.405	.416
<b>MSEL_RL</b>	<b>1. <math>R^2=.24, F(1,22)=7.031, p=.015</math></b>			
	Total interaction	.063	.024	.492*
	<b>2. <math>R^2=.25, F(2,21)=3.470, p=.050</math></b>			
	Positive behaviour	.068	.035	.389 <sup>+</sup>
	Negative behaviour	.057	.055	.208
<b>MSEL_EL</b>	<b>1. <math>R^2=.27, F(1,22)=8.221, p=.009</math></b>			
	Total interaction	.054	.020	.505*
	<b>2. <math>R^2=.28, F(2,21)=4.076, p=.032</math></b>			
	Positive behaviour	.055	.029	.369 <sup>+</sup>
	Negative behaviour	.059	.046	.252
<b>Q-Chat</b>	<b>1. <math>R^2=.32, F(1,11)=5.179, p=.044</math></b>			
	Total interaction	.088	.039	.566*
	<b>2. <math>R^2=.34, F(2,10)=2.525, p=.129</math></b>			
	Positive behaviour	.074	.058	.346
	Negative behaviour	.127	.095	.365

Note. <sup>+</sup> $p < .10$ , \* $p < .05$

N-CDI\_WC=N-CDI word comprehension; N-CDI\_WP=N-CDI word production; MSEL\_RL=MSEL receptive language; MSEL\_EL=MSEL expressive language; Q-Chat=Q-Chat total score

First, the models for MSEL receptive language and MSEL expressive language (HR group) were both significant (receptive:  $R^2=.591$ ,  $F(3,12)=5.792$ ,  $p=.011$ ; expressive:  $R^2=.666$ ,  $F(3,12)=10.972$ ,  $p=.001$ ) with MSEL receptive language at 14 months as a significant predictor in both models (receptive:  $\beta=.652$ ,  $t=3.171$ ,  $p=.008$ ; expressive:  $\beta=.811$ ,  $t=4.878$ ,  $p<.001$ ). The *total interaction* composite was no longer a significant predictor (receptive:  $\beta=.273$ ,  $t=1.267$ ,  $p=.229$ ; expressive:  $\beta=.117$ ,  $t=.672$ ,  $p=.514$ ). Second, the model for N-CDI word comprehension (LR group) was also significant ( $R^2=.546$ ,  $F(3,19)=7.629$ ,  $p=.002$ ) with N-CDI word comprehension at 14 months as a significant predictor ( $\beta=.613$ ,  $t=2.865$ ,  $p=.010$ ). Again, the *total interaction* composite was no longer a significant predictor ( $\beta=-.233$ ,  $t=-1.443$ ,  $p=.165$ ).

## DISCUSSION

### **Sibling interaction: high-risk vs. low-risk group**

The current study used a naturalistic, observational method to evaluate sibling interactions between 24-month-old children and their older sibling. With regard to the *first research question*, sibling interaction characteristics in the HR group (HR-sibs and their older sibling with ASD) were compared with those in the LR group (LR-sibs and their older typically developing sibling). Consistent with previous studies (Knott et al., 1995, 2007; Walton & Ingersoll, 2015), siblings interacted less frequently with each other in the HR group. More specifically, both siblings in HR-dyads showed lower levels of *positive* behaviour compared to the LR-dyads. HR-sibs and children with ASD were less likely to positively initiate social interaction (e.g., sharing, helping, smiling) and showed fewer positive responses (e.g., following an instruction, giving a toy upon request, returning a smile). Levels of conflict or negative behaviour did not differ between groups. Next, to answer the *second research question*, imitation of the youngest child was evaluated. Even though the frequency of imitation was relatively low in both groups, HR-sibs imitated their older siblings less frequently than LR-sibs during sibling interactions. This is in line with previous studies suggesting low levels of imitation in younger siblings of children with ASD (Stone et al., 2007; Zwaigenbaum et al., 2005). After controlling for age of the oldest sibling, day care attendance of the youngest sibling



and the amount of time both children spent together at home, group status (high-risk vs. low-risk) remained a significant predictor of both positive behaviour and imitation of the youngest child during the sibling interaction.

Previous studies have demonstrated the importance of (positive) sibling interactions for the development of both siblings (Brody, 2004; Feinberg et al., 2012; Kuhl, 2004; Tucker et al., 1999). However, when positive social approaches and responses of an older sibling with ASD are limited, possibly resulting in fewer bouts of positive interaction, younger HR-sibs might miss out on opportunities to practice adequate social behaviours. A decrease in social input may in turn contribute to the atypical developmental trajectories of HR-sibs (Dawson, 2008). The degree to which atypical social behaviour of the older sibling affects the HR-sib's development might also depend on characteristics of the HR-sib. For example, Knott and colleagues (2007) found that typically developing HR-sibs compensated for the impairments of their sibling with ASD by taking over the leadership position. This was not found in the current study, but the children in our sample were on average younger compared to the sample of Knott and colleagues (2007). It is possible that toddlers are less inclined or less able to take over the dominant position compared to school-aged children. In addition, HR-sibs who show signs of the BAP or early ASD might experience social-communicative difficulties themselves. Therefore, lower levels of social input during sibling interactions might influence vulnerable HR-sibs differently than typically developing HR-sibs.

### **Association with language and social communication**

Concerning the *third research question*, associations between sibling interaction characteristics and the youngest child's language and social-communicative abilities were evaluated. First, we found positive associations between the sibling interaction and language development at 24 months. In general, in the HR group but not in the LR group, a higher frequency of initiatives and responses was associated with better receptive and expressive language. In addition, it seemed that positive interactions more than negative interactions were associated with better language on the Mullen Scales of Early Learning (Mullen, 1995). In contrast, surprisingly, in the LR group there was a *negative* association between the sibling interaction and language comprehension. In addition, in the LR group there was a positive association between imitation of the

youngest sibling and language production, which is in line with existing research linking imitation to later expressive language (e.g., Charman et al., 2000). Due to the cross-sectional nature of these associations we cannot distinguish whether sibling interactions stimulate language development, or whether better language abilities lead to more (positive) sibling interactions. Nor can we exclude the possibility that other factors mediate the relationship between sibling interactions and language.

To conclude that sibling interaction characteristics promote development, we would not only expect a positive association between the sibling interaction and language, but we would also expect that this positive association remains significant after controlling for pre-existing language abilities at 14 months. To this end, the MSEL and N-CDI scores at 14 months were included. We could conclude that, for all significant regression models, language abilities at 14 months rather than sibling interaction characteristics at 24 months explained language development at 24 months. Therefore, based on these results, there is insufficient evidence to conclude that sibling interactions promote language.

In addition to pre-existing abilities, future research should also take the broader social context into account when evaluating the association between sibling interactions and HR-sibs' developmental trajectories. Parent-child interactions can also influence the development of their children. For example, parental behaviours such as sharing attention or responsive verbal language are important for later social responsiveness and language development in children with ASD (Clifford & Dissanayake, 2009; Haebig, McDuffie, & Weismer, 2013). It is therefore possible that parental behaviours compensate for lower levels of social input from the sibling interaction. Thus, the family context and parent-child interactions could also influence the association between sibling interactions and outcome.

Second, higher levels of *total* interaction (positive and negative) at 24 months were positively associated with more parent-reported ASD characteristics as measured with the Q-Chat (Allison et al., 2008), but not with the ADOS scores (Lord et al., 2012). Although the level of immediate imitation was lower in the HR group, this does not exclude the possibility that HR-sibs learn behaviours from their older sibling with ASD. New behaviours are often acquired through deferred imitation, modelling or social learning and older siblings can be powerful models (Bandura, 1977; Whiteman et al.,

2011). Consequently, HR-sibs might learn ASD-specific behaviours from their older siblings that are also measured by the Q-Chat (e.g., lining up toys, tip-toe walking, repetitive behaviours, echolalia). The correlations between the Q-Chat and ADOS scores were moderate (ADOS Social Affect:  $r=.440$ ; ADOS Restricted and Repetitive Behaviours:  $r=.450$ ; ADOS Total score:  $r=.411$ ), demonstrating a positive association between parent-report and a more comprehensive observation measure for ASD. Nevertheless, sibling interaction characteristics only predicted *parent-reported* ASD characteristics. It is possible that parents observe different behaviours at home or that they interpret the behaviour of their child differently (e.g., exaggerating subtle behaviours) than researchers, resulting in differences between parent-report and observational methods.

### **Implications and strengths**

The current study entails theoretical implications. Several studies have noted important differences between HR-sibs and siblings of typically developing children (e.g., Brian et al., 2014; Gamliel et al., 2007; Yirmiya et al., 2006), but sibling interactions have rarely been included in studies of HR-sibs. The current study was the first to assess both sibling interaction characteristics in sibling pairs with a child with ASD and the association with the language and social-communicative development of the youngest sibling. Not only were there significant differences between both groups in terms of positive initiations and responses, the association with the younger sibling's development was more pronounced in the HR group. The combination of early vulnerabilities and altered social interactions or social learning could contribute to the increased risk of ASD or the broader autism phenotype in HR-sibs. Although future research is needed to better understand the interplay between environmental and genetic/biological factors, the current study shows that the social environment, including sibling interactions, should be taken into account.

An (important) strength of this study is the use of a naturalistic, observational method. Compared to self-report or parent-report, observations in a naturalistic setting may provide more representative insights in sibling interactions (Hastings & Petalas, 2014; Lobato, Miller, Barbour, Hall, & Pezzullo, 1991; Senapati & Hayes, 1988). In addition, the sample included a very young age group. Given that interactions early in

life possibly impact on later development (Dawson, 2008; Seibert, Hogan, & Mundy, 1982), it is important to evaluate sibling interactions in younger populations.

### **Limitations and future research**

There are some limitations that need further consideration. The small sample size imposes several restrictions on the current study. First, it limits the generalisability of the study and the likelihood of detecting significant results. Second, only a limited number of predictors could be included in the regression model. As a result, we were restricted in the amount of regression models we could test. Third, the combination of the small sample size and the distribution of our data did not allow for more elaborate, parametric analyses.

To control for the inflation of the Type I error rate due to multiple comparisons, the (Holm-)Bonferroni correction was considered. However, this leads to a substantial reduction of statistical power (Nakagawa, 2004; Perneger, 1998). Due to the combination of a lower statistical power because of the small sample size and the fact that we expected to detect *small* differences, applying a Bonferroni correction would greatly reduce the possibility of finding relevant group differences while there are in fact real world differences. Therefore, the correction was not applied. Future research should focus on replicating the current results in a larger sample, matched on sample characteristics.

The cross-sectional nature of the analyses at 24 months limits our conclusions in terms of causality. In addition, as we only included measures for the development of the youngest child, we were unable to evaluate the association between sibling interactions and the development of children with ASD. More research, including longitudinal studies, is needed to assess to what extent sibling interactions might contribute to the development of both children.

At this point, since the prospective study is still ongoing, we were unable to evaluate the diagnostic status of the HR-sibs (ASD/BAP vs. no ASD) and distinguish HR-sibs with and without later ASD/BAP. This impeded us to draw conclusions regarding the value of sibling interactions for later ASD outcome. When all HR- and LR-sibs reach the age of 36 months, evaluations in terms of diagnostic status will be possible.

**Conclusion**

This study provides new insights into the association between the social environment of HR-sibs and their social-communicative and language development. Sibling interactions in sibling pairs with a child with ASD differ from sibling interactions between typically developing children. In addition, sibling interaction characteristics are associated with the HR-sib's ASD characteristics. Given that siblings are important interaction partners during early childhood, further evaluating the role of sibling interactions in the developmental trajectories of HR-sibs will be valuable to include in future research.

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**SIBLING AND PEER RELATIONSHIPS OF  
THREE-YEAR-OLD CHILDREN AT RISK FOR  
AUTISM SPECTRUM DISORDER<sup>1</sup>**

## ABSTRACT

The early social environment has the potential to influence later child development. Therefore, early social interactions need to be considered when studying the expression of the ASD phenotype in younger siblings of children with autism spectrum disorder (ASD). The current study observed early sibling and peer interactions in three-year-old younger siblings of children with ASD (high-risk group;  $n=15$ ). First, sibling interaction characteristics were evaluated and compared with sibling interactions in a typically developing control group (low-risk group;  $n=31$ ). Overall, sibling interactions in the high-risk group were characterised by fewer positive behaviours compared to the low-risk group. Second, differences and associations between sibling and peer interactions were evaluated in the high-risk group. The high-risk siblings showed more negative responses during sibling interactions than during peer interactions. Negative behaviours during sibling interactions also positively predicted negative behaviours during peer interactions. Third, the association between ASD characteristics and the sibling and peer interaction was explored in the high-risk group. In general, ASD characteristics were associated with fewer social behaviours (initiations and responses), either positive or negative. Thus, given that altered social experiences may influence the development of children at risk for ASD, these early social interactions should be further investigated in future research.

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## INTRODUCTION

During childhood, siblings typically spend more time together than with any other interaction partner (Berger & Nuzzo, 2008). Sibling relationships differ from peer interactions or friendships in the sense that they are involuntary and lifelong (Howe & Recchia, 2014). Moreover, given their shared family context, siblings co-construct a common history and a special understanding of one another (Dirks, Persram, Recchia, & Howe, 2015; McHale, Updegraff, & Feinberg, 2016). Recent literature increasingly focuses on sibling relationships as they can influence development even when accounting for other family relationships (Defoe et al., 2013; Dirks et al., 2015; Stocker, Burwell, & Briggs, 2002). Children can benefit from positive sibling relationships (Yucel & Downey, 2015) and positive sibling interactions and sibling warmth promote the development of empathy (Lam, Solmeyer, & McHale, 2012; Tucker, Updegraff, McHale, & Crouter, 1999). Moreover, older siblings are powerful models from whom younger siblings can learn a variety of behaviours (McHale et al., 2016). They initiate the interaction more frequently and take on a leadership role while younger siblings are more likely to follow and imitate their older brother or sister (Abramovitch, Corter, & Lando, 1979; Dirks et al., 2015; Lamb, 1978).

Autism spectrum disorder (ASD) is a neurodevelopmental disorder characterised by persistent deficits in social communication and social interaction (American Psychiatric Association [APA], 2013). Siblings of children with ASD (hereafter high-risk siblings; HR-sibs) are also at increased risk of developing ASD or showing ASD-related characteristics (Ozonoff et al., 2011). Although the high recurrence rate in HR-sibs emphasises the genetic susceptibility of family members of individuals with ASD (Risch et al., 2014), genetic factors are insufficient to explain the full ASD phenotype. Environmental factors need to be considered as well, including the social environment (Mandy & Lai, 2016). The manifestation of the ASD phenotype in HR-sibs and children with ASD could be influenced by early social interactions (Dawson, 2008; Mandy & Lai, 2016). Within the context of social interaction, the ASD phenotype mainly includes difficulties in initiating and responding to social communication and in maintaining social relationships. Studies report lower levels of social engagement and social interest/motivation and fewer reciprocal friendships in children with ASD (Broekhof et al., 2015; Kasari, Locke, Gulsrud,

& Rotheram-Fuller, 2011; Sigman & Ruskin, 1999). Consequently, sibling relationships between children with ASD and their younger siblings are likely to differ from sibling relationships between two typically developing siblings. Accordingly, the few studies that have been conducted on sibling relationships in sibling pairs with a child with ASD showed clear differences with sibling pairs without a child with ASD. Compared to siblings of typically developing children, HR-sibs (mean age: 9.35 years) were less involved and more avoidant (Walton & Ingersoll, 2015). Compared to children with Down Syndrome, children with ASD (mean age: 6.0 years) showed fewer social approaches and were less responsive to their sibling (mean age: 6.6 years). In addition, in sibling pairs including a child with ASD the typically developing sibling more frequently initiated the interaction than the child with ASD, taking over the leadership role (Knott, Lewis, & Williams, 1995, 2007). These altered social interactions could further influence the development of both interaction partners (Dawson, 2008; Mandy & Lai, 2016). Thus, the characterisation of early social interactions could help to shed light on the behavioural manifestation of ASD in HR-sibs.

As children enter preschool or kindergarten, interactions with peers increase. Similar to siblings, peers can significantly influence the social and emotional development of children (Denham et al., 2011). During social interaction with a peer (i.e., conflict, cooperation, friendship...) children can practice emotion regulation skills as well as develop their self-esteem (Cheah, Nelson, & Rubin, 2001). In contrast to older siblings or parents, peers are not necessarily more knowledgeable or skilful at social interaction than each other. In addition, they often share common interests and are like-minded (Howes, Rubin, Ross, & French, 1988). To our knowledge, peer interactions of young siblings of children with ASD have not yet been investigated. However, given that HR-sibs are at increased risk of showing early social-communicative and language impairments (e.g., Toth, Dawson, Meltzoff, Greenson, & Fein, 2007), HR-sibs' peer interactions might be characterised by more negative behaviours or by a lack of social interaction, at least for those HR-sibs showing increased ASD characteristics. Studies show that children with ASD more frequently experience difficulties in social interactions with peers (Kasari et al., 2011; Ozonoff & South, 2001; Zhang & Wheeler, 2011).

Sibling and peer relationships are closely related (Roskam, Meunier, & Stievenart, 2015). Two possible links between sibling and peer relationships have been suggested

(Howe, Ross, & Recchia, 2011). First, it is possible that children transfer the interaction style learned in one relationship to another. For example, studies report that positive sibling interactions are related to positive interactions with peers (e.g., Lockwood, Kitzmann, & Cohen, 2001). Second, children may compensate for deficits in one relationship (e.g., low-quality sibling relationships) by turning to other relationships (e.g., friendships). As such, negative behaviours in the sibling relationship could be associated with positive behaviours in the peer relationship (Mendelson, Aboud, & Lanthier, 1994; Stocker, 1994). To date, there are no studies exploring the association between sibling and peer interactions of younger siblings of children with ASD.

### **The present study**

This study investigated the sibling and peer relationships of three-year-old siblings of children with ASD. At the age of 3, children have just started school. Therefore, at this age, sibling interactions might be more influential than peer interactions.

First, sibling interactions between three-year-old children and their older sibling with ASD (HR group) were compared with sibling interactions between three-year-old children and their older typically developing sibling (low-risk (LR) group). In line with previous research (Knott et al., 1995, 2007; Walton & Ingersoll, 2015), we expected lower levels of social interaction in HR sibling pairs than in LR sibling pairs. This is consistent with the lower levels of social engagement and social interest/motivation found in children with ASD (Broekhof et al., 2015; Kasari et al., 2011; Sigman & Ruskin, 1999). In addition to the comparison between the HR and LR group, the role of the younger and older sibling within each group was evaluated. In the LR group, we expected that older children would be more dominant (i.e., more initiations) while younger children would be more following (i.e., more responses) (Abramovitch et al., 1979; Lamb, 1978). Concerning the HR group, Knott et al. (1995) suggested that typically developing HR-sibs would act as a leader. However, their sample included both younger and older siblings and the age range was wide. In our sample, HR-sibs were very young and both HR-sibs and children with ASD might experience social-communicative difficulties. Therefore, we did not have specific hypotheses for the HR group.

Second, the association between sibling and peer relationships was explored in the HR group. To this end, HR-sibs were also observed in interaction with a peer. To our

knowledge, this study is the first to compare sibling and peer relationships of HR-sibs. In addition, studies in typically developing children suggest that children either generalise their interaction style from one social context to another (i.e., correspondence between social relationships) or compensate for deficits in one relationship through other relationships (i.e., discrepancy between social relationships) (Howe et al., 2011). Hence, we were not able to formulate specific expectations with respect to the association between sibling and peer relationships.

Third, given the presence of ASD characteristics in children with ASD and possibly in HR-sibs as well, we evaluated the association between ASD characteristics of the ASD- and HR-sibs and the sibling and peer relationships of HR-sibs. Since ASD is characterised by lower levels of social interest, difficulties in initiating and responding to social communication and impairments in maintaining social relationships (APA, 2013; Zwaigenbaum et al., 2005), we expected a negative association between ASD characteristics and the frequency of social interactions.

## METHOD

### Participants

Sibling and peer relationships were evaluated in three-year-old younger siblings of children with or without ASD. The HR group ( $n=15$ ) included children with ASD (ASD-sib) and their younger sibling (HR-sib). The LR group ( $n=31$ ), without first- or second-degree relatives with ASD, consisted of typically developing children (TD-sib) and their younger sibling (LR-sib).

The ASD diagnosis of the older child with ASD was made by a multidisciplinary team and confirmed with the Social Responsiveness Scale, Second Edition (SRS-2; Constantino & Gruber, 2012; Dutch translation by Roeyers, Thys, Druart, De Schryver, & Schittekatte, 2011), and the Social Communication Questionnaire (SCQ; Rutter, Bailey, & Lord, 2003; Dutch translation by Warreyn, Raymaekers, & Roeyers, 2004). Fifteen children with ASD scored above the threshold for ASD on the SRS-2 and nine children scored above the threshold on both the SRS-2 and the SCQ. The multidisciplinary assessment also included an evaluation of cognitive functioning using the Wechsler Intelligence Scale for Children

(WISC-III-NL; Kort et al., 2005), the Wechsler Preschool and Primary Scale of Intelligence (WPPSI-III-NL; Hendriksen & Hurks, 2009), the Snijders-Oomen Non-Verbal Intelligence Test (SON-R; Tellegen, Winkel, Wijnberg-Williams, & Laros, 1998), or the Bayley Scales of Infant Development (BSID-II-NL: Meulen, van der Ruiters, Spelberg, & Smrkovský, 2004; Bayley-III-NL: Van Baar, Steenis, Verhoeven, & Hessen, 2014). Three children with ASD scored very low (IQ<55), four scored below average (IQ between 55-85), seven scored within the normal range (IQ between 85-115), and one child scored above average (IQ>115).

Regarding the sample characteristics (see Table 1), there were no significant differences between the LR and HR group in the sex ratio of the youngest or oldest child and the chronological age of the youngest child. However, ASD-sibs were on average older than TD-sibs ( $F(1,44)=14.156$ ,  $p<.001$ ). Family SES was calculated using Hollingshead's four factor index (Hollingshead, 1975). LR families had an average social status of 51.88 and HR families had an average social status of 46.37. There was no significant difference between both groups.

**Table 1***Sample characteristics*

	Low-risk ( $n=31$ )	High-risk ( $n=15$ )	
<b>Family SES (<math>M(sd)</math>)</b>	51.88 (7.22)	46.37 (11.88)	$F(1,43)=3.755$
	Youngest sibling		
<b>Chronological age</b>			$F(1,44)=1.521$
$M(sd)$	37.39 (.79)	37.77 (1.28)	
Range	35.33-39.73	35.60-41.70	
<b>Sex ratio (M:F)</b>	18:13	8:7	$\chi^2(1)=.092$
<b>ASD characteristics</b>			
SRS-2 ( $M(sd)$ )	24.73 (8.46)	36.64 (24.97)	$F(1,38)=5.824^*$
SCQ ( $M(sd)$ )	4.35 (2.50)	5.64 (4.88)	$U=170.50$
	Oldest sibling		
<b>Chronological age</b>			$F(1,44)=14.156^{***}$
$M(sd)$	68.44 (13.88)	92.93 (30.55)	
Range	52.37-109.00	60.30-165.03	
<b>Sex ratio (M:F)</b>	16:15	9:6	$\chi^2(1)=.287$

Note. Chronological age is reported in months; SRS-2=Social Responsiveness Scale, parent-report; SCQ=Social communication Questionnaire, parent-report; \* $p<.05$ , \*\*\* $p<.001$



To explore the presence of ASD characteristics in HR- and LR-sibs, the SCQ and SRS-2 scores were obtained from the youngest sibling. The total score on the SRS significantly differed between groups ( $F(1,38)=5.824, p=.021$ ), with an average score of 24.73 for LR-sibs and 36.64 for HR-sibs. There was no difference between LR- and HR-sibs concerning the SCQ total score (LR-sib: 4.35, HR-sib: 5.64;  $U=170.50, p=.653$ ). Two HR-sibs scored above the cut-off for ASD on both the SRS and SCQ while one additional HR-sib only scored above the cut-off for ASD on the SRS. None of the LR-sibs scored above the ASD cut-off on the SCQ or SRS.

### **Procedure**

Participants were recruited from an ongoing prospective follow-up study of the early social-communicative development of LR- and HR-sibs. At 36 months, two additional appointments were scheduled to observe sibling and peer interactions. The sibling interaction always preceded the peer interaction. The parents of all children were informed about the purpose of the study and were asked to sign an informed consent. At the time of the sibling interaction observation parents filled out questionnaires about the oldest child, including the SCQ and SRS-2. At the time of the school observation, parents of HR-sibs and their peers filled out the SCQ and the SRS-2 about their own child and the teacher filled out the SRS-2 about the HR-sibs.

The SCQ (Rutter et al., 2003) is a parent-reported, 40-item screening questionnaire for ASD. Although the SCQ was originally intended for children above age 4, the 'current' version can be used in children between 2 and 4 years (Allen, Silove, Williams, & Hutchins, 2007). For children over 4 years, it is recommended to use a cut-off score of 15 as a possible indication for ASD. For younger children, it is recommended to lower the cut-off score to 11 (Corsello et al., 2007).

The SRS-2 (Constantino & Gruber, 2012) is a 65-item dimensional measure that assesses ASD characteristics in children and adolescents between the ages of 3 and 18. It has five subscales (social awareness, social cognition, social communication, social motivation, and autistic mannerisms) and can be completed by either the parent or a teacher. The cut-off for ASD for three-year-olds is set at 48 (total raw score), the cut-off for ages 4-18 years is set at 51.

**Sibling interaction.** To observe sibling interactions, LR and HR sibling pairs were observed in the participants' home while playing with a standardised set of toys. They were encouraged to play 10 minutes with a Playmobil™ 1-2-3 farm, 10 minutes with a marble run, and 5 minutes with an animal sound keyboard, consecutively. Different sets of toys were chosen to elicit different kinds of play (parallel, associative and cooperative play). Since there were no clear systematic differences between the three play contexts, the scores were summed and considered as one play interaction. During the observation, one parent was always present in the room, continuing normal routines (e.g., household tasks or work). Parents were asked not to interfere during the play observation. If children initiated social interaction with the parent, they could respond briefly as they normally would.

**Peer interaction.** Fourteen high-risk siblings were observed while interacting with a peer. To maximise the comparability between sibling and peer observations, the same procedure was followed as during the sibling interaction observation, including the same set of toys. Teachers were also asked not to interfere during the play observation.

Teachers selected one classmate with whom the HR-sib was close and frequently engaged in play. Peer interactions were observed in the preschool classroom, a naturalistic setting to observe play interactions between peers (Cillessen & Bellmore, 2011). Although preschool classrooms can be noisy and busy, the involvement of both children was high in 12 out of 14 observations. Child involvement was rated by the observer on a three-point scale (0: low involvement; 1: moderate involvement; 2: high involvement). In one case, the HR-sib only showed moderate involvement and in another case both children only showed moderate involvement (e.g., distracted by other activities in the classroom). In the majority, other classmates did not disturb the play. In two cases, there was a slight degree of interference by other children (e.g., trying to join the play observation). In all cases, the teacher rated the behaviour of HR-sibs during the play observation as being representative for a typical play session.

**Coding procedure – Observer agreement.** All play sessions were videotaped. For play with the marble run and Playmobil™, the middle 8 minutes were selected and coded using The Observer XT, version 11.5 (Noldus, 2013). For play with keyboard, the middle 4 minutes were selected for coding. The same scheme was used to code both sibling and peer interactions. First, a frequency coding scheme was used. Frequencies of

social initiations and responses, both negative and positive, were coded. *Social initiations* are communicative attempts to initiate a new interaction, directed towards another individual. *Responses* are related to and follow a previous initiation within five seconds. Initiations and responses can be either positive/prosocial (e.g., sharing a toy) or negative (e.g., refusing a request). Next, the time children spent in interaction with each other (mutuality), with the parent/teacher and with the experimenter was also coded. To account for the time not spent in interaction with another person, the following non-interactive behaviours were coded: distress, doing nothing/looking at a random object, orientation towards the sibling/peer, repetitive/stereotyped behaviour, and time spent in a purposeful activity (e.g., play).

To obtain a broader evaluation of the course of the play observation, five global rating scales were included. Each scale ranged from 1 (low frequency/quality) to 5 (high frequency/quality). *Interference of the parent/teacher* refers to the extent to which the parent/teacher interfered or interrupted during the play observation. *Proximity* indicates the distance between both children during play. In this scale, interpersonal distance is taken into account as well. Two children who are further away in distance but are in close interaction (e.g., dancing together from a distance), are considered to be in close proximity. *Imitation* of both children is coded when the child shows behaviour that is a direct and exact repetition of the other child. Finally, *togetherness* reflects the degree to which both children are enjoying the interaction *together*.

Clips were independently rated by trained master students blind to participant information. 15% of the clips were randomly selected to determine interrater agreement and were coded by all coders. Next, average measures intraclass correlation coefficients (ICC) were calculated. ICC's between .60-.74 reflect good interrater agreement and ICC's between .75-1.00 reflect excellent interrater agreement (Cicchetti, 1994). Due to their low frequency, the following behaviours were not coded reliably (ICC<.60) and therefore excluded from further analyses: distress, doing nothing/looking at a random object, repetitive/stereotyped behaviour, imitation by the oldest sibling/peer. For the frequency coding scheme, ICC's of the included behaviours ranged between .84 and .99. For the global rating scales, ICC's ranged between .73 and .84.

## Data-analysis

Outliers were defined as values higher or lower than 3 standard deviations above or below the mean. Since outliers were not considered to be random but characteristic of our sample, outliers were replaced by the highest/lowest value allowed (mean +/- 3sd) rather than deleted.

As assumptions for parametric testing were not met (deviation from the normal distribution), non-parametric analyses were used to analyse differences in sibling interaction characteristics between the HR and LR group (i.e., Mann-Whitney U). To compare younger and older siblings within each group and compare the sibling and peer interaction, the Wilcoxon signed-rank test for two related samples was used.

To evaluate the association between the sibling and peer interaction, and between ASD characteristics (SRS-2, SCQ) and the sibling or peer interaction, hierarchical regression models were tested. However, correlation analyses revealed high intercorrelations between several social interaction variables, causing multicollinearity in the regression model. Both during the sibling and peer interaction, positive initiations and positive responses of both children were significantly ( $p < .05$ ) intercorrelated as well as negative initiations and negative responses. To address the problem of multicollinearity, positive initiations/responses of both children on the one hand and negative initiations/responses of both children on the other hand were summed to form two composite scores: positive behaviour and negative behaviour. First, since sibling interactions preceded peer interactions, regression models were tested to evaluate whether sibling interactions predicted peer interactions. Second, regression models were tested to evaluate the predictive value of ASD characteristics for the sibling and peer interaction.

## RESULTS

### Sibling interaction characteristics

First, sibling interaction characteristics were compared between the LR and HR group. As shown in Table 2, both the youngest and oldest child in the LR group showed more positive responses (youngest:  $U=114.50$ ,  $p=.005$ ; oldest:  $U=125.00$ ,  $p=.010$ ) and positive initiations (youngest:  $U=146.50$ ,  $p=.042$ ; oldest:  $U=132.00$ ,  $p=.017$ ) compared to

the HR group. In addition, ASD-sibs spent more time interacting with a parent than TD-sibs ( $U=118.00, p=.004$ ).

**Table 2**

*Descriptives (mean (standard deviation)) and group differences (LR vs. HR group; Mann-Whitney U) of the sibling interaction*

	Low-risk (n=31)	High-risk (n=15)	U
Frequency coding - Youngest sibling			
Negative initiations <sup>a</sup>	7.17(5.69)	6.49(4.00)	227.50
Positive initiations <sup>a</sup>	8.05(6.36)	5.71(8.25)	146.50*
Negative responses <sup>a</sup>	8.44(7.63)	8.32(7.25)	228.00
Positive responses <sup>a</sup>	9.32(8.18)	4.13(6.28)	114.50**
Mutuality <sup>b</sup>	92.17(78.38)	85.51(107.92)	184.00
Interaction with experimenter <sup>b</sup>	76.39(69.22)	69.49(115.27)	157.00
Interaction with parent <sup>b</sup>	19.97(34.92)	64.56(114.59)	194.00
Orientation to sibling <sup>b</sup>	58.16(52.42)	47.39(51.10)	210.00
Play <sup>b</sup>	928.21(159.38)	895.13(156.36)	193.00
Frequency coding - Oldest sibling			
Negative initiations <sup>a</sup>	11.22(8.30)	10.23(11.25)	208.00
Positive initiations <sup>a</sup>	16.41(12.72)	10.50(15.84)	132.00*
Negative responses <sup>a</sup>	6.82(6.10)	5.82(5.48)	208.50
Positive responses <sup>a</sup>	5.17(5.08)	2.00(1.73)	125.00*
Mutuality <sup>b</sup>	92.17(78.38)	85.51(107.92)	184.00
Interaction with experimenter <sup>b</sup>	62.23(51.57)	53.50(79.52)	172.50
Interaction with parent <sup>b</sup>	7.15(14.92)	47.62(66.05)	118.00**
Orientation to sibling <sup>b</sup>	39.15(48.57)	42.69(68.31)	223.00
Play <sup>b</sup>	980.24(136.81)	953.51(190.67)	216.00
Global rating			
Interference <sup>c</sup>	1.28(0.40)	2.00(1.06)	139.50*
Proximity <sup>c</sup>	4.45(0.53)	3.77(1.18)	166.00
Repetitive behaviour youngest <sup>c</sup>	1.01(0.06)	1.11(0.35)	208.50
Repetitive behaviour oldest <sup>c</sup>	1.00(0.00)	1.29(0.59)	170.50**
Imitation youngest <sup>c</sup>	1.06(0.16)	1.07(0.14)	225.00
Togetherness <sup>c</sup>	2.05(0.74)	1.71(0.97)	173.00

Note. \* $p<.05$ ; \*\* $p<.01$ ;  $U$ =Mann-Whitney U; <sup>a</sup>results reflect absolute frequencies; <sup>b</sup>results reflect total duration (in seconds); <sup>c</sup>results reflect global rating (1-5)

Concerning the global rating scales, results revealed no significant differences in terms of *proximity* between both children ( $U=166.00$ ,  $p=.113$ ), *imitation* of the youngest child ( $U=225.00$ ,  $p=1.000$ ) or *togetherness* ( $U=173.00$ ,  $p=.159$ ). There was however a significant group difference in *interference* of the parent ( $U=139.50$ ,  $p=.020$ ), and the level of *repetitive behaviour* of the oldest child ( $U=170.50$ ,  $p=.008$ ). Parents interfered more frequently in the HR group than in the LR group and ASD-sibs showed more repetitive behaviours than TD-sibs.

Second, within-group differences (youngest vs. oldest sibling) were evaluated in each group separately. In the *LR group*, the oldest child was more dominant than the youngest sibling, expressed in higher levels of positive ( $z=-3.622$ ,  $p<.001$ ) and negative ( $z=-2.099$ ,  $p=.035$ ) initiations. The youngest sibling adopted a more following position with higher levels of positive ( $z=-2.851$ ,  $p=.003$ ) and negative ( $z=-2.566$ ,  $p=.009$ ) responses than the oldest child. In the *HR group*, there was only a significant difference in negative responses, with ASD-sibs showing more negative responses than HR-sibs ( $z=-2.430$ ,  $p=.013$ ). Both siblings showed equal levels of positive ( $z=-.711$ ,  $p=.497$ ) and negative ( $z=-1.512$ ,  $p=.141$ ) initiations and positive responses ( $z=-.446$ ,  $p=.660$ ).

Finally, it was evaluated whether these role patterns differed between groups. To determine the degree to which the oldest sibling was more dominant, the difference score between initiations of the oldest sibling and initiations of the youngest sibling was calculated. To evaluate to which extent the youngest sibling was more following, the difference score between responses of the youngest sibling and responses of the oldest sibling was calculated. These difference scores were compared between groups (Mann-Whitney  $U$  test). Results revealed no significant group differences in terms of positive initiations ( $U=168.00$ ,  $p=.130$ ), negative initiations ( $U=211.50$ ,  $p=.622$ ), positive responses ( $U=195.50$ ,  $p=.424$ ), or negative responses ( $U=199.50$ ,  $p=.436$ ).

### **Sibling vs. peer interaction**

In the HR group, sibling interactions were first compared with peer interactions (see Table 3). HR-sibs showed significantly more negative responses in interaction with their sibling than in interaction with their peer ( $z=-2.029$ ,  $p=.043$ ). Concerning the comparison between the child with ASD (mean age: 7.7 years) and the peer (mean age: 3.5 years), there were no significant results. The evaluation of the global rating scales revealed that

the proximity between HR-sibs and peers was higher than between HR-sibs and ASD-sibs ( $z=-2.148, p=.033$ ).

**Table 3**

*Descriptives (mean (standard deviation)) and group differences (sibling vs. peer interaction; Wilcoxon signed-rank) for the high-risk group*

	Sibling interaction	Peer interaction	Z
Youngest sibling			
Negative initiations <sup>a</sup>	6.49(4.00)	4.79(4.76)	-.692
Positive initiations <sup>a</sup>	5.71(8.25)	10.00(12.63)	-1.050
Negative responses <sup>a</sup>	8.32(7.25)	4.36(3.39)	-2.029*
Positive responses <sup>a</sup>	4.13(6.28)	6.79(9.61)	-.804
Mutuality <sup>b</sup>	85.51(107.92)	67.85(87.52)	-.094
Interaction with experimenter <sup>b</sup>	69.49(115.27)	70.14(57.33)	-1.036
Orientation to sibling/peer <sup>b</sup>	47.39(51.10)	43.53(50.29)	-.345
Play <sup>b</sup>	895.13(156.36)	998.74(141.94)	-1.412
Oldest sibling			
Negative initiations <sup>a</sup>	10.23(11.25)	7.57(8.05)	-.769
Positive initiations <sup>a</sup>	10.50(15.84)	8.21(8.82)	-.699
Negative responses <sup>a</sup>	5.82(5.48)	4.00(3.80)	-1.258
Positive responses <sup>a</sup>	2.00(1.73)	5.07(8.25)	-1.252
Mutuality <sup>b</sup>	85.51(107.92)	67.85(87.52)	-.094
Interaction with experimenter <sup>b</sup>	53.50(79.52)	99.02(95.63)	-1.475
Orientation to sibling/peer <sup>b</sup>	42.69(68.31)	69.10(136.61)	-.874
Play <sup>b</sup>	953.51(190.67)	922.76(223.21)	-.345
Global rating			
Proximity <sup>c</sup>	3.77(1.18)	4.57(0.70)	-2.148*
Repetitive behaviour high-risk sibling <sup>c</sup>	1.11(0.35)	1.21(0.34)	-.557
Repetitive behaviour oldest sibling/peer <sup>c</sup>	1.29(0.59)	1.19(0.62)	-.677
Imitation high-risk sibling <sup>c</sup>	1.07(0.14)	1.14(0.22)	-.905
Togetherness <sup>c</sup>	1.71(0.97)	2.05(0.93)	-.749

Note.  $n=14$ ; \* $p<.05$ ; \*\* $p<.01$ ; Z=Wilcoxon signed-rank z-value; <sup>a</sup>results reflect absolute frequencies; <sup>b</sup>results reflect total duration (in seconds); <sup>c</sup>results reflect global rating (1-5)

Next, it was evaluated whether sibling interaction characteristics (positive/negative composite) predicted the peer interaction. The regression model for *positive* peer interactions was not significant ( $R^2=.15$ ,  $F(2,11)=1.002$ ,  $p=.398$ ) and neither positive nor negative behaviours during the sibling interaction significantly predicted positive peer interactions (positive:  $\beta=-.25$ ,  $t=-.765$ ,  $p=.460$ ; negative:  $\beta=.46$ ,  $t=1.415$ ,  $p=.185$ ). The regression model for *negative* peer interactions was marginally significant ( $R^2=.36$ ,  $F(2,11)=3.099$ ,  $p=.086$ ), and negative sibling interactions significantly predicted negative peer interactions ( $\beta=.69$ ,  $t=2.444$ ,  $p=.033$ ). The association between positive sibling interactions and negative peer interactions was not significant ( $\beta=-.25$ ,  $t=-.872$ ,  $p=.402$ ).

### **Association with ASD characteristics**

Preliminary analyses revealed multiple medium to strong ( $r>.30$ ; Cohen, 1992) correlations between ASD characteristics of the HR-sib and ASD-sib and social behaviours during the sibling/peer interaction. Consequently, regression models were tested to determine to what extent ASD characteristics predicted the sibling or peer interaction. Due to the small sample size, not all measures of ASD characteristics could be added in the regression model (SRS vs. SCQ; parent- vs. teacher-report). Moreover, not all correlations between ASD characteristics and social behaviours were significant. Therefore, only significant associations between ASD characteristics and the peer or sibling interaction were tested by means of regression analyses. In addition, to evaluate whether ASD characteristics would predict social interaction, regardless of its nature (positive vs. negative), a *total interaction* composite score was created by adding all positive and negative behaviours. Results are presented in Table 4.

**Sibling interaction.** There were two significant associations between ASD characteristics of the HR-/ASD-sib and social behaviours during the sibling interaction (positive, negative, total). First, the SCQ of the ASD-sib correlated negatively with positive behaviours during the sibling interaction ( $r=-.63$ ,  $p=.012$ ). Second, the SRS of the HR-sib (teacher-report) was negatively correlated with negative behaviours during the sibling interaction ( $r=-.578$ ,  $p=.039$ ). Next, two hierarchical regression models were tested (positive and negative composite) including the SCQ of the ASD-sib and the SRS of the HR-sib as predictors. The model for *positive* behaviours was marginally significant ( $R^2=.45$ ,  $F(2,10)=4.075$ ,  $p=.051$ ), with only ASD characteristics of the ASD-sib as a



significant predictor ( $\beta=-.63$ ,  $t=-2.701$ ,  $p=.022$ ). More ASD characteristics were associated with fewer positive behaviours. The model for *negative* behaviours was significant ( $R^2=.46$ ,  $F(2,10)=4.337$ ,  $p=.044$ ), with ASD characteristics of the HR-sib as a significant predictor ( $\beta=-.59$ ,  $t=-2.535$ ,  $p=.030$ ). More ASD characteristics of the HR-sib were related to fewer negative behaviours during the sibling interaction.

**Peer interaction.** There was one significant correlation between ASD characteristics of the HR-sib and the peer interaction. The HR-sib's SRS (parent-report) significantly correlated with the *total interaction* composite of the peer interaction ( $r=-.61$ ,  $p=.022$ ). Since only the HR-sibs' SRS scores significantly correlated with the peer interaction, a simple linear regression model was tested with the HR-sib's SRS scores as predictor for the total peer interaction. The regression model was significant ( $R^2=.37$ ,  $F(1,12)=6.930$ ,  $p=.022$ ), with ASD characteristics of the HR-sib as a significant predictor ( $\beta=-.61$ ,  $t=-2.633$ ,  $p=.022$ ). More ASD characteristics of the HR-sib were associated with fewer social behaviours during the peer interaction.

**Table 4**

*Association between ASD characteristics and the sibling or peer interaction*

		Sibling interaction			
		<i>B (SD)</i>	$\beta$	$R^2$	<i>p</i>
<b>Positive behaviour</b>	(constant)	68.66(17.38)		.45	.051
	SCQ ASD-sib	-2.09(.78)	-.63		.022
	SRS HR-sib	-.19(.19)	-.23		.345
<b>Negative behaviour</b>	(constant)	72.57(15.80)		.46	.044
	SCQ ASD-sib	-.1.10(.71)	-.36		.150
	SRS HR-sib	-.44(.17)	-.59		.030
		Peer interaction			
		<i>B (SD)</i>	$\beta$	$R^2$	<i>p</i>
<b>Total behaviour</b>	(constant)	89.38(17.05)		.37	.022
	SRS HR-sib	-1.02(.39)	-.61		.022

*Note.* SCQ=Social Communication Questionnaire; SRS=Social Responsiveness Scale

## DISCUSSION

The goal of this study was threefold: 1) To compare sibling interactions between high- and low-risk sibling pairs; 2) To investigate the association between sibling and peer relationships of HR-sibs; and 3) To determine whether characteristics of the sibling and peer interaction of HR-sibs were associated with ASD characteristics of one of the interaction partners. This study was the first to compare sibling and peer interactions of HR-sibs and included the direct observation of HR-sibs' interaction with both a sibling and a peer in a naturalistic setting.

### Sibling interaction characteristics

Consistent with our expectations and previous studies (e.g., Knott et al., 1995, 2007), we found lower levels of positive initiations and responses in the HR group. Also, ASD-sibs showed more repetitive behaviours than TD-sibs, which is consistent with the diagnostic criteria of ASD (APA, 2013). Finally, parents interfered more often in HR sibling pairs and children with ASD interacted more frequently with their parent. However, interference of the parent often involved the child with ASD and both scales are strongly intercorrelated. Thus, either parents interfered more frequently in the HR group, hereby eliciting more interaction from the ASD-sibs with the parent, or ASD-sibs more frequently oriented towards their parent causing parents to interfere during the play session.

Second, we compared the social behaviour of younger and older siblings within each dyad. For the LR group, results were comparable to results found in typically developing children (Abramovitch et al., 1979; Lamb, 1978). Older siblings more frequently led the interaction (i.e., more initiations) while younger siblings were more following (i.e., more responses). However, in the HR group neither HR-sibs nor ASD-sibs assumed a dominant position. Only the number of negative responses differed between HR- and ASD-sibs, with ASD-sibs showing more negative responses than HR-sibs. This was not in line with the findings of Knott and colleagues (1995) who found that typically developing HR-sibs took over the dominant position. However, the age range in their sample was wide (e.g., HR-sibs: 1;1-12;5 years) and the participants were both younger and older siblings of children with ASD. In the current study, HR-sibs were very young ( $M_{age}=37.77$  months)

and consistently younger than the children with ASD. In addition, the studies of Knott et al. (1995, 2007) involved *typically developing* siblings of children with ASD while the HR-sibs in this study showed elevated ASD characteristics compared to LR-sibs. Both the age of HR-sibs and difficulties in social communication might have influenced their abilities to assume a dominant position during the sibling interaction. Nevertheless, even though the difference in role pattern between younger and older siblings was only found in the LR group, the group comparison was not significant. The higher level of negative responses of the ASD-sib compared to the HR-sib could be the beginning of the role reversal suggested by Knott et al. (2007). It is possible that, as both children get older, this role reversal becomes more pronounced.

Since positive sibling interactions are important for the development of younger siblings (e.g., Tucker, Updegraff, McHale, & Crouter, 1999), reduced levels of positive interactions in HR sibling pairs may have implications for the development of HR-sibs. Fewer positive interactions also means fewer learning opportunities for younger siblings to learn adequate social behaviours from their older sibling. In addition, the absence of a clear leader may also result in fewer social exchanges during the sibling interaction (Knott et al., 1995). Although older siblings often function as powerful role models, the combination of reduced positive/nurturing exchanges and the lack of leadership/dominance in older siblings with ASD could weaken their position as a role model (Bandura, 1977).

### **Sibling and peer interaction**

First, we found that HR-sibs responded more negatively to their older sibling with ASD than to their peer. Given the social-communicative impairments associated with the disorder, children with ASD could experience difficulties in adequately approaching their younger sibling. In addition, children with ASD more frequently show behavioural problems (e.g., Dominick, Davis, Lainhart, Tager-Flusberg, & Folstein, 2007). As there was no difference in social behaviour between ASD-sibs and peers, the difference in negative responses of HR-sibs is not solely attributable to a higher level of negative initiations in ASD-sibs. Through bidirectional processes, negative or inadequate social approaches (e.g., unclear request) of ASD-sibs combined with higher levels of ASD characteristics in HR-sibs could result in more negative responses of the HR-sibs towards

their sibling with ASD compared to a typically developing peer. Proximity was also higher between HR-sibs and their peer, suggesting that HR-sibs' peer relationships might be more close or intimate than HR-sibs' sibling relationships. The combination of social-communicative impairments in both children and a more negative interactional pattern could lead to lower levels of intimacy or closeness in the sibling interaction. However, two other factors could have influenced these results. First, peers included in the current study were selected based on the closeness of their relationship with the HR-sib. Second, given that proximity did not differ between the HR and LR group, both groups may show higher levels of closeness in interaction with a peer compared to a sibling. Higher physical proximity could be an expression of learned behaviour in the preschool classroom, a context in which children are frequently engaged in structured activities in small groups. At home, children are typically more free to choose how and with whom they play.

The social behaviour of the ASD-sib and peer was only compared because of the possible influence on the HR-sib's behaviour. It needs to be noted that older siblings with ASD ( $M_{age}=7.7$  years) were considerably older than the HR-sibs' peers ( $M_{age}=3.5$  years). In addition, given their ASD diagnosis, ASD-sibs show increased social-communicative impairments. As a result, ASD-sibs and peers were not comparable as interaction partners.

Second, more negative behaviours during the sibling interaction were associated with more negative behaviours during the peer interaction. Studies comparing sibling and peer interactions in preschool-aged children report both higher levels of positive as well as negative interactions during peer relationships (Abramovitch, Corter, Pepler, & Stanhope, 1986; McElwain & Volling, 2005). To our knowledge, there are no studies comparing sibling and peer relationships including high-risk siblings. Repeated negative interactions between HR- and ASD-sibs (i.e., more negative responses) may result in a learned, more negative interaction style in HR-sibs. In turn, HR-sibs might generalise this interaction style to other social contexts such as the peer relationship. The latter is in line with previous studies stating that individuals generalise their interaction style across relationships (e.g., Lockwood et al., 2001). Since peer interactions also influence children's early development (Denham et al., 2011), the generalisation of a negative

interaction style from sibling to peer relationships could influence HR-sib's early development.

### **Association with ASD characteristics**

Both HR- and ASD-sibs show increased levels of ASD characteristics. However, there is no research investigating how increased ASD characteristics relate to social behaviours during sibling or peer interactions. In general, it seems that increased levels of ASD characteristics lead to fewer social behaviours. First, ASD characteristics of ASD-sibs were associated with fewer *positive* behaviours during the sibling interaction. This was in line with our expectations since ASD is characterised by social-communicative impairments and lower levels of social engagement and social interest/motivation (e.g., Kasari et al., 2011). In addition, results from the group comparison showed that positive behaviours were less frequent in the HR group compared to the LR group. Second, surprisingly, ASD characteristics of the HR-sibs were associated with fewer *negative* behaviours during the sibling interaction and fewer *total* social behaviours during the peer interaction. We also did not find an association between the HR-sib's ASD characteristics and *positive* behaviours during the sibling interaction. First, because younger siblings are more likely to assume a following position whereas older siblings are more frequently responsible for leading the interaction, HR-sibs' ASD characteristics are less likely to influence overall positive interactions. In contrast, as the ASD symptoms of the ASD-sib increase, the child's capability to lead the interaction decreases, resulting in lower levels of positive interactions. Second, regarding the negative behaviours, the answer might be found in the combination of ASD in the older sibling and ASD characteristics in the younger sibling. It is possible that ASD in the older sibling leads to more negative interactions when the younger sibling does not show ASD characteristics because of a *mismatch* between both children. More socially skilled HR-sibs might interrupt the older sibling more, trying to initiate the interaction. In addition, ASD characteristics of the older sibling might lead to frustrations or confusions in HR-sibs. However, when *both* children show ASD characteristics this might lead to a more peaceful or balanced situation, better matched in terms of social expectations. For example, both children might avoid each other, leading to less conflict.

### **Limitations and future directions**

This study had several limitations. First, the sample size was small, especially in the HR group, affecting the generalizability of the findings and the power to detect significant results. Second, older children with ASD were on average older than older typically developing children and peers. This could have influenced the comparison between sibling interactions in the HR and LR group and the comparison between sibling and peer interactions. Unfortunately, the small sample size and distribution of our data excludes more elaborate statistical analyses such as controlling for the effect of age. Third, due to practical constraints, we were unable to evaluate peer interactions in the LR group. Therefore we were unable to compare the peer relationships and association between sibling and peer relationships of HR-sibs with a LR control group. Finally, the sibling and peer interaction observation were not counterbalanced. As a result, HR-sibs were already familiar with the toys and procedure at the time of the peer interaction, which could have affected their behaviour. For example, because the toys were no longer new, HR-sibs could have been less interested in or attracted to these toys during the peer interaction, leading to less conflict. However, results showed that the majority of the children were strongly involved during the peer interaction.

Three HR-sibs screened positive for ASD on the SRS-2, of whom two also screened positive on the SCQ. After research-reliable ADI-R and ADOS administrations, these three HR-sibs were classified as HR-sibs with ASD, whereas the other 12 HR-sibs were classified as HR-sibs without ASD. Comparison of the results based on the sample with the three HR-ASD siblings and without the three HR-ASD siblings revealed little differences, therefore all HR-sibs were included in further analyses. Unfortunately, the HR-ASD group was too small for more elaborate analyses. Future research should further evaluate sibling and peer interactions of HR-sibs based on ASD outcome.

### **Conclusion**

This study provides meaningful insights into the early social relationships of younger siblings of children with ASD. First, positive sibling interactions were less frequent in HR sibling pairs. More ASD characteristics of the older sibling with ASD were also associated with lower levels of positive behaviour, thus reduced positive interactions seem to be characteristic for HR sibling pairs. Second, compared to peer relationships, sibling

relationships of HR-sibs were somewhat more negative and characterised by less proximity. Moreover, the results suggest that HR-sibs may generalise a more negative interaction style from sibling relationships to their peer relationships. Although further research is needed to determine the association with developmental outcomes, it seems valuable to include these early social relationships in studies evaluating the (atypical) developmental trajectories of HR-sibs.

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In this doctoral dissertation we focused on the early social environment of younger siblings of children with ASD (i.e., high-risk siblings). The first aim was to broaden our knowledge on the parent-child, sibling, and peer interactions of these high-risk siblings. Second, the predictive value of early social interactions for the concurrent and later development of high-risk siblings was explored. This last chapter provides an overview and discussion of the main findings as well as a reflection on the possible methodological, theoretical, and clinical implications. Finally, the most important limitations and directions for future research are discussed.

## RECAPITULATION OF THE RESEARCH GOALS

With this dissertation, we aimed to gain insight into the early social experiences of younger siblings of children with ASD (i.e., high-risk siblings; HR-sibs) as well as to explore the association between these early experiences and HR-sibs' developmental trajectories. Although an association between the early social environment and later outcome has been suggested (Dawson, 2008; Inguaggiato, Sgandurra, & Cioni, 2017; Mandy & Lai, 2016), this is yet to be supported by empirical findings.

The first objective was to study the characteristics of HR-sibs' social interactions with parents and siblings and evaluate whether these interactions significantly differed from social interactions of younger siblings of typically developing children (i.e., low-risk siblings; LR-sibs). To address the limitations of existing studies, social experiences were observed during the first years of life and by means of a naturalistic, observational method. First of all, the quality of the *parent-child interaction* was evaluated in a within-family study (Chapter 2) including a sample of mothers with both a child with ASD (mean age: 68.06 months) and without ASD (mean age: 47.75 months). In addition, the parent-child interaction of HR- and LR-sibs was assessed in a between-family study at the ages of 5 and 10 months (Chapter 3). Given the lack of consensus regarding the coding method best suited to code social interactions, the two most commonly used coding methods (i.e., frequency vs. global coding) were compared and discussed (Chapter 2). Next, *sibling interactions* were evaluated using a between-family design at the ages of 18 months (Chapter 4), 24 months (Chapter 5), and 36 months (Chapter 6). Finally, at 36 months, the association between the sibling and peer interactions of HR-sibs was explored (Chapter 6).

A second objective was to explore whether early social experiences were associated with child development. To this end, the predictive value of these social experiences for the development of HR-sibs and the expression of the ASD phenotype was evaluated. Given that HR-sibs' developmental trajectories may be characterised by impairments in different domains (e.g., Brian et al., 2014; Toth, Dawson, Meltzoff, Greenson, & Fein, 2007; Zwaigenbaum et al., 2005), we explored the association with cognitive functioning, motor development, language development, and social-communicative functioning.

## OVERVIEW AND INTEGRATION OF THE MAIN FINDINGS

Throughout this dissertation we have emphasised the importance of the early social environment for the development of young children. The results from the studies included in this dissertation provided further evidence for differences in the early social environment of HR-sibs during the first years of life.

### Parent-child interaction

Studies have stressed the potential influence of parent-child interactions on child development (e.g., Feldman, Bamberger, & Kanat-Maymon, 2013; Houck & LeCuyer-Maus, 2002; Russel, 2011). In addition, it has been shown that parent-child interactions including children with ASD or HR-sibs differ from parent-child interactions including typically developing children (e.g., Adamson, Bakeman, Deckner, & Nelson, 2012; Freeman & Kasari, 2013; Wan et al., 2013). In this dissertation, we evaluated the parent-child interaction of HR-sibs in two different samples with both a between-family design (Chapter 3), comparing parent-child interactions of HR- and LR-sibs, and a within-family design (Chapter 2), comparing the parent-child interaction of typically developing HR-sibs and children with ASD.

**Characteristics of the parent-child interaction.** The comparison of HR- and LR-sibs' parent-child interaction (*between-family*) during the first year of life revealed little significant differences. First, LR- and HR-sibs did not differ in the social-communicative behaviours shown during the parent-child interaction. These results are in line with previous studies studying the parent-child interaction in HR-sibs during the first year of life and contribute to the growing consensus that the emergence of early ASD characteristics mainly occurs during the second year of life (Rozga et al., 2011; Szatmari et al., 2016; Wan et al., 2012; Yirmiya et al., 2006). However, in the Belgian subsample LR dyads were characterised by more positive affect at 10 months. In addition, positive affect of the parent and child were strongly correlated ( $r=.58, p=.012$ ), suggesting that a positive disposition in or positive behaviours of LR-sibs may elicit more positive affect in their parents and vice versa. Early differences in positive affect were also found in previous studies evaluating HR-sibs' temperament during the first year of life (Clifford, Hudry, Elsabbagh, Charman, & Johnson, 2013; Zwaigenbaum et al., 2005). Thus

temperamental profiles might be better suited to characterise HR-sibs during the first year of life and to distinguish HR-sibs who develop ASD from those who do not. As HR-sibs grow older and social-communicative deficits become more pronounced, differences in social-communicative behaviours during the parent-child interaction may arise more clearly, as also demonstrated by the findings of Rozga et al. (2011) and Wan et al. (2013) studying the parent-child interaction between 12-24 months.

Second, the interaction style of parents in the HR group strongly resembled the interaction style of parents in the LR group, which may be partly explained by the lack of differences in social-communicative behaviours of HR- and LR-sibs. This suggests that parents of children with ASD do not automatically generalise the interaction style adopted in interaction with their child with ASD to the interaction with younger siblings. Instead of using one interaction style with all their children, it seems that parents alter and adapt their behaviours in response to each individual child. The higher level of parental directiveness in HR dyads, as reported by Wan et al. (2012), was not replicated. However, their sample included 31.1% HR-sibs with a later ASD diagnosis, which is higher than the recurrence rate reported in other studies (e.g., Ozonoff et al., 2011). Combined with the fact that their follow-up study at 12-15 months showed that parent directiveness was higher in HR-sibs who later developed ASD compared to HR-sibs without an ASD diagnosis, this suggests that the increased level of parent directiveness at 6 months might be related to the higher prevalence of HR-sibs with ASD. The recurrence rate in the Belgian subsample was remarkably lower (18%) and more in line with previous studies, which could explain why the increased parental directiveness was not found in this subsample. To generalise this hypothesis to the European sample, the re-evaluation of results based on diagnostic outcome of the entire sample is needed.

The comparison of the parent-child interaction of typically developing HR-sibs and children with ASD (*within-family*) in a slightly older sample revealed differences in both the child's and parent's behaviour. First, HR-sibs (without ASD) more frequently used declarative initiatives whereas children with ASD showed more imperative initiatives (Meirsschaut, Warreyn, & Roeyers, 2011). Given the social-communicative deficits associated with ASD, this finding was not surprising. Second, the within-family study provided evidence for a more structuring (i.e., quality of instruction, structure and limit setting), supporting, and responsive parenting style in interaction with typically



developing HR-sibs compared to children with ASD during a task situation. It is possible that children with ASD are less receptive for and less responsive to structuring and supporting behaviours of their parent. Children with ASD experience more difficulties in following instructions, including responding to verbal information, shifting their attention between the parent and the task and language comprehension (Charman, Drew, Baird, & Baird, 2003; Quill & Institute, 1997). In addition, they are less sensitive to social rewards such as praise (e.g., Demurie, Roeyers, Baeyens, & Sonuga-Barke, 2011). A lower response to structuring and supporting behaviours might discourage parents to use these strategies. Moreover, in interaction with their child with ASD, mothers may experience certain difficulties or frustrations that lead to a decrease in emotional support. Children with ASD are for example more likely to withdraw from social interactions or ignore their parent (Doussard-Roosevelt, Joe, Bazhenova, & Porges, 2003). In addition, mothers of children with ASD more frequently experience parenting stress or negative emotions related to their child's diagnosis (e.g., unwillingness to accept the diagnosis) (Davis & Carter, 2008; Wachtel & Carter, 2008). Interacting with a child who does not show social-communicative difficulties and does not have an ASD diagnosis might be a relief for parents, triggering more enthusiasm and support. The age difference between HR-sibs and the children with ASD could have influenced the group differences as well. However, the scales quality of instruction and structure and limit setting were only correlated with the child's chronological age in the ASD group. In addition, parents' responsiveness and supportive presence were not related to the child's age at all. Thus, it is unlikely that group differences are better explained by the age difference between both children.

**Association with development.** The results from Chapter 3 provided some support for the association between the parent-child interaction and later child development. At 5 months, results were somewhat counterintuitive and given the small sample size, they should be interpreted with care. In the HR group, but not the LR group, lower levels of negative affect of the child during the parent-child interaction predicted better later gross motor skills whereas parents' sensitive responsiveness predicted lower receptive language abilities. At 10 months, associations between the parent-child interaction and development were only found in the LR group. More specifically, less negative affect of the child positively predicted the child's receptive language skills, whereas more

adaptive parenting (i.e., more sensitive responsiveness, scaffolding, positive affect) predicted better expressive language skills. The latter results are consistent with previous studies in typical development reporting positive associations between positive parenting behaviours and child development (e.g., Russel, 2011). As to the child's negative affect, children who show fewer signs of frustration or discomfort might elicit more verbal responses from parents. In turn, increased levels of verbal communication/input can stimulate children's language abilities (Kuhl, 2004).

**Conclusion.** At 5 months, possible ASD characteristics and differences between HR- and LR-sibs are not yet clear and parents from the HR group interact with their child in the same way as do parents from the LR group. At 10 months, differences are noticeable in terms of positive affect, suggesting that differences start to emerge between HR- and LR-sibs. For the purpose of another project not included in this dissertation, the videos from the Belgian sample were also coded by means of the Coding Interactive Behavior manual (CIB; Feldman, 1998), which only revealed lower levels of dyadic reciprocity in the HR group at 10 months (but no differences at 5 months). Since warmth and positive affect are an important aspect of dyadic reciprocity, this result was partly in line with the results found by the coding scheme used in this dissertation. As differences between HR- and LR-sibs increase from 10 months onwards, it is possible that parents in the HR group will adapt or change their interaction style and that similarities between the HR and LR group will gradually decrease. Although follow-up of the current sample beyond the first year of life is needed, this hypothesis is supported by the findings of Rozga et al. (2011) and Wan et al. (2013) studying the parent-child interaction during the second year of life. Thus, based on our results we can conclude that the parent-child interaction of HR-sibs strongly resembles the parent-child interaction of LR-sibs at the ages of 5 and 10 months. We hypothesise that, instead of just generalising the interaction style used with their child with ASD to other siblings, parents adapt their behaviours to the specific child characteristics. A differentiation in interaction style based on child characteristics was also found in the within-family study (see Chapter 2). In interaction with their typically developing child, parents provided more structure, emotional support and were more responsive, which may resemble the interaction style of parents without a child with ASD. In interaction with their child with ASD, parents adapted their behaviours to the abilities of their child (e.g., nonverbal mental age and word comprehension).

Regarding the *association with later development*, we only found limited support for the link between early parent-child interactions and HR-sibs' developmental trajectories. However, this was only the first study attempting to evaluate the association with development and we were limited in terms of the power of our study as well as the type of analyses being used. Given that we did find some evidence for the association with later development indicates that further research including a larger sample and more elaborate analyses might reveal more significant associations. Moreover, it is again possible that as ASD characteristics in HR-sibs increase throughout the first years of life, the predictive value of the parent-child interaction increases as well.

### **Sibling interaction**

The possible social-communicative impairments of children with ASD/HR-sibs are likely to influence the nature of their sibling interactions. There are several studies reporting on the quality of these sibling interactions (e.g., Kaminsky & Dewey, 2001; Orsmond & Kuo, 2009; Walton & Ingersoll, 2015), but, with the exception of Knott and colleagues (1995, 2007), only by means of self-report or parent-report rather than observations in a naturalistic context. Also, their sample included school-aged children or adolescents. The studies included in this dissertation (i.e. Chapters 4-6) report on the early sibling interactions of HR-sibs with their older sibling with ASD (HR group), using a naturalistic and observational study design. Moreover, these studies reflect a first attempt to *longitudinally* evaluate HR-sibs' sibling interactions compared to sibling interactions of LR-sibs and their older typically developing sibling (LR group) during the first years of life. In this chapter, we first provide a detailed description of HR-sibs' sibling interactions. Second, we elaborate upon the changes in these sibling interactions over time. Third, the association with HR-sibs' concurrent development is discussed.

**Characteristics of the sibling interaction.** Even though in both groups the total amount of interaction between the siblings was limited and children spent the majority of their time playing alone, several meaningful group differences emerged. Across all three time points, only two differences were found in the total amount of social interaction. The total amount of responses of the youngest child at 18 months was higher in the HR group, whereas the total amount of initiations of the youngest child at 24 months was higher in the LR group. Thus, the mere frequency of sibling interactions

was similar in both groups. When looking at the distinction between the amount of positive and negative behaviours, differences were more pronounced. At 18 months, HR-sibs' sibling interactions were more negative in nature whereas their sibling interactions at 24-36 months were characterised by lower levels of positive behaviours. Furthermore, there were differences in the ratio of positive versus negative behaviours (i.e., positive composite vs. negative composite). At 18 months, the ratio of positive versus negative behaviours was similar in both groups (LR: 57% HR: 53%;  $U=275.00$ ,  $p=.569$ ), but at 24-36 months there was a higher relative frequency of positive behaviours in the LR group (24 months: 63% (LR) vs. 51% (HR),  $U=263.50$ ,  $p=.066$ ; 36 months: 54% (LR) vs. 36% (HR),  $U=130.00$ ,  $p=.015$ ). Regarding the sibling interaction at 18 months, an equal ratio of positive vs. negative behaviours in both groups combined with increased negativity in the HR group suggests that positive behaviours were also higher in the HR group, though not significantly. Next, to gain insight into the interactional patterns between younger and older siblings in each group, both siblings were compared for each group separately. At all three time points, the LR group showed the asymmetrical pattern we expected based on studies including typically developing children (i.e., the older child as the leader, the younger child in a more following position; Abramovitch, Corter, & Lando, 1979; Lamb, 1978). At 18 and 24 months this asymmetrical pattern was also observed in the HR group. At 36 months, however, this asymmetrical pattern shifted to a more symmetrical pattern. There was no clear leader and the child with ASD was even slightly more following.

Even though the ratio of positive vs. negative behaviours did not differ between the LR and HR group at 18 months, Chapter 4 did show that the absolute frequency of negative behaviours was higher in the HR group. However, since the study described in Chapter 4 was the first to compare sibling interactions between HR- and LR-sibs at the age of 18 months, there was no other empirical evidence to support this increased negativity in HR dyads. Starting from 24 months, results were more comparable with previous studies reporting fewer prosocial behaviours and lower levels of involvement or closeness in HR sibling pairs (Kaminsky & Dewey, 2001; Knott et al., 1995, 2007; Walton & Ingersoll, 2015). However, these studies included older children and also reported differences in negative behaviours. For example, in a group of school-aged children (mean age range: 5;2 – 6;6 years), Knott et al. (1995) observed lower levels of

positive as well as negative initiations in children with ASD compared to children with Down Syndrome. Also, in somewhat older children (mean age range: 9.35 – 11.67 years), Walton and Ingersoll (2015) and Kaminsky and Dewey (2001) provided evidence for lower levels of negativity/conflict in HR sibling pairs. It is possible that, in HR dyads, lower levels of positive behaviours may only become clear from 24 months onwards and that the increased negativity at 18 months might change into a pattern of decreased negativity as HR-sibs grow older.

Methodological differences between the current studies and existing research (e.g., Kaminsky & Dewey, 2001; Knott et al., 1995, 2007; Walton & Ingersoll, 2015) limit the comparability between studies. First, the current study only included younger siblings whereas previous studies consistently included both younger and older siblings of children with ASD. This might have influenced the role patterns and behaviours observed during the sibling interaction. Walton and Ingersoll (2015) concluded that more developmental problems were observed in older siblings of children with ASD compared to younger siblings. Moreover, older siblings were more likely to show teaching behaviours. In addition, Petalas et al. (2012) report lower levels of conflict when the typically developing sibling is older than the child with ASD. Second, for the studies reported in this dissertation, the age of the younger sibling was standardised at 18, 24, and 36 months. In contrast, the defined age range in previous studies was very wide, ranging from infancy/preschool to middle childhood and adolescence. Third, the siblings of children with ASD included in the studies of Knott and colleagues (1995, 2007) and Kaminsky and Dewey (2001) were typically developing, whereas our sample included all HR-sibs, regardless of their diagnostic status.

**Sibling interactions from 18 to 36 months.** To better understand the change in group differences and role patterns over time, behaviours were re-evaluated across all three time points. Descriptives of the three time points are presented in Table 1 and a visual representation of the evolution over time is displayed in Figure 1. Because only nine HR-sibs were seen at 18, 24, and 36 months, the evolutions from 18 to 24 months (27 LR-sibs, 18 HR-sibs) and from 24 to 36 months (30 LR-sibs, 15 HR-sibs) were analysed separately. Given the small sample size, we rely on both the test statistics and visual inspection of the data to provide a detailed description of the sibling interactions.

**Table 1***Sibling interaction characteristics (mean(standard deviation)) from 18 to 36 months*

	<b>Low-risk group</b>		
	18m (n=29)	24m (n=32)	36m (n=31)
<b>LR-sib</b>			
Positive initiations <sup>b</sup>	5.54(6.02)	5.48(4.51)	8.05(6.36)
Negative initiations <sup>b</sup>	2.75(2.52)	4.04(5.03)	7.17(5.69)
Positive responses <sup>ab</sup>	13.66(9.85)	16.49(8.93)	9.32(8.18)
Negative responses	5.06(4.67)	6.73(5.93)	8.44(7.63)
<b>TD-sib</b>			
Positive initiations <sup>b</sup>	11.08(11.79)	10.95(8.72)	16.41(12.72)
Negative initiations	11.00(6.46)	10.13(5.93)	11.22(8.30)
Positive responses <sup>ab</sup>	5.98(6.22)	7.93(6.36)	5.17(5.08)
Negative responses	3.48(3.69)	5.48(5.99)	6.82(6.10)
	<b>High-risk group</b>		
	18m (n=22)	24m (n=24)	36m (n=15)
<b>HR-sib</b>			
Positive initiations	4.36(4.20)	3.38(3.52)	5.71(8.25)
Negative initiations <sup>b</sup>	3.45(3.05)	2.44(2.66)	6.49(4.00)
Positive responses <sup>b</sup>	14.72(10.74)	12.14(10.83)	4.13(6.28)
Negative responses	10.37(7.63)	7.83(6.69)	8.32(7.25)
<b>ASD-sib</b>			
Positive initiations <sup>a</sup>	14.01(17.11)	9.75(14.06)	10.50(15.84)
Negative initiations <sup>a</sup>	12.77(9.76)	9.48(6.16)	10.23(11.25)
Positive responses	6.68(8.01)	5.70(6.86)	2.00(1.73)
Negative responses	6.00(4.58)	5.11(4.98)	5.82(5.48)

Note. <sup>a</sup>Significant change from 18 to 24 months, <sup>b</sup>Significant change from 24 to 36 months

In the LR group there was a (marginally) significant increase in positive responses of both children from 18 to 24 months (youngest:  $z=-1.945$ ,  $p=.051$ ; oldest:  $z=-2.118$ ,  $p=.033$ ). From 24 to 36 months, there was a significant increase in positive ( $z=-2.311$ ,  $p=.020$ ) and negative ( $z=-3.009$ ,  $p=.002$ ) initiations of the youngest child and in positive initiations of the oldest child ( $z=-2.121$ ,  $p=.033$ ). In addition, there was a (marginally) significant decrease in positive responses of both children (youngest:  $z=-2.963$ ,  $p=.002$ ;

oldest:  $z=-1.767$ ,  $p=.078$ ). In contrast, in the HR group the positive and negative initiations of the child with ASD significantly decreased from 18 to 24 months (positive:  $z=-1.969$ ,  $p=.049$ ; negative:  $z=-2.040$ ,  $p=.040$ ). In addition, from 24 to 36 months there was a significant increase in the HR-sibs' negative initiations ( $z=-3.299$ ,  $p<.001$ ) and a significant decrease in HR-sibs' positive responses ( $z=-2.475$ ,  $p=.011$ ).

When looking at the changes over time, two patterns were noteworthy. The first pattern concerns the change in negative behaviours. As shown in Figure 1, there was a descending trend for negative initiations and responses of both siblings in the HR group from 18 to 24 months. Moreover, the decrease in negative initiations of the child with ASD was significant. In contrast, with the exception of negative initiations of the oldest child, all negative behaviours in the LR group showed an ascending trend. Regarding the change in negative behaviours from 24 to 36 months, there was a significant increase in negative initiations of the youngest child in both groups. Negative initiations of the oldest child and negative responses of both siblings (slightly) increased in both groups. Thus, while the change in negative behaviours in the LR group seems linear, with low levels of negative behaviour at 18 months and a gradual increase towards 24/36 months, the change in negative behaviours in the HR group seems more quadratic, with somewhat higher levels at 18 months, a decrease from 18 to 24 months, and a (small) increase towards 36 months. The second pattern relates to the positive initiations of the oldest child. From 18 to 24 months, there is a remarkable decrease in the positive initiations of the child with ASD whereas the positive initiations of the typically developing older child remain stable. In contrast, the frequency of positive initiations of the typically developing older child significantly increased from 24 to 36 months, with little to no change in positive initiations of the child with ASD. Whereas the positive initiations of the older children in the LR group are highest at 36 months, positive initiations of the child with ASD are highest at 18 months.

When looking at the role patterns, the evolution from 18 to 36 months also seemed to be somewhat different in both groups. Based on visual inspection of the data we can conclude that, in the LR group, the difference in initiations of the oldest and youngest child and the difference in responses of the oldest and youngest child appears to be similar across all three time points. As a result, role patterns remained similar over time.

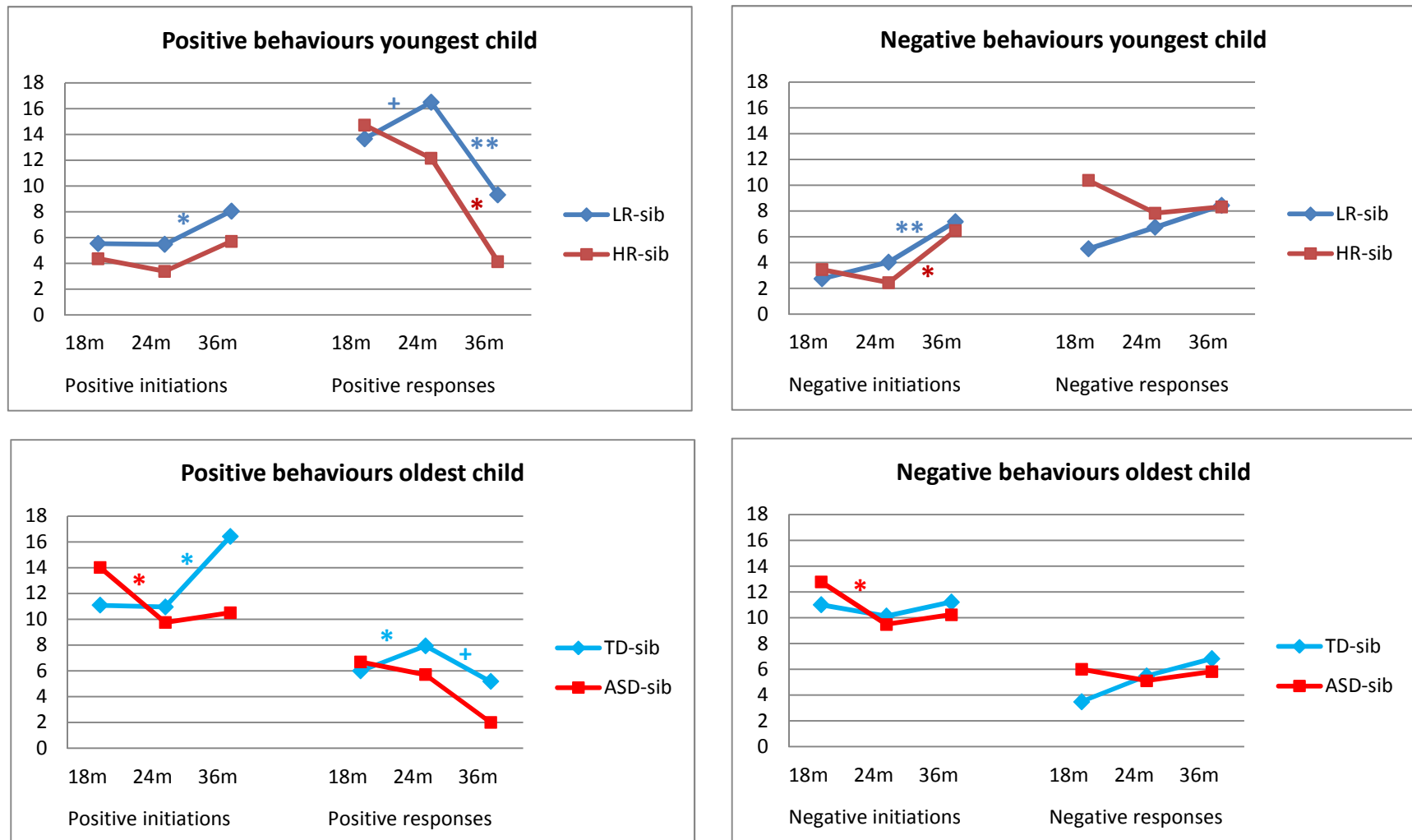


Figure 1. Frequency of positive and negative behaviours of both siblings in the HR and LR group at 18, 24, and 36 months ( $^+p<.10$ ,  $*p<.05$ ,  $**p<.01$ )



In the HR group, however, the difference in initiations as well as responses of both siblings seemed to decrease from 18 to 36 months. Consequently, the role asymmetry also decreased from 18 to 36 months, resulting in a more symmetrical pattern at 36 months. In both groups, younger siblings showed more initiations at 36 months compared to 18 and 24 months. Whereas older siblings in the LR group also showed higher levels of initiations at 36 months, this was not the case for the children with ASD who showed a decrease in initiations from 18 to 36 months. The role (a)symmetry in terms of total initiations and responses is presented in Figure 2.

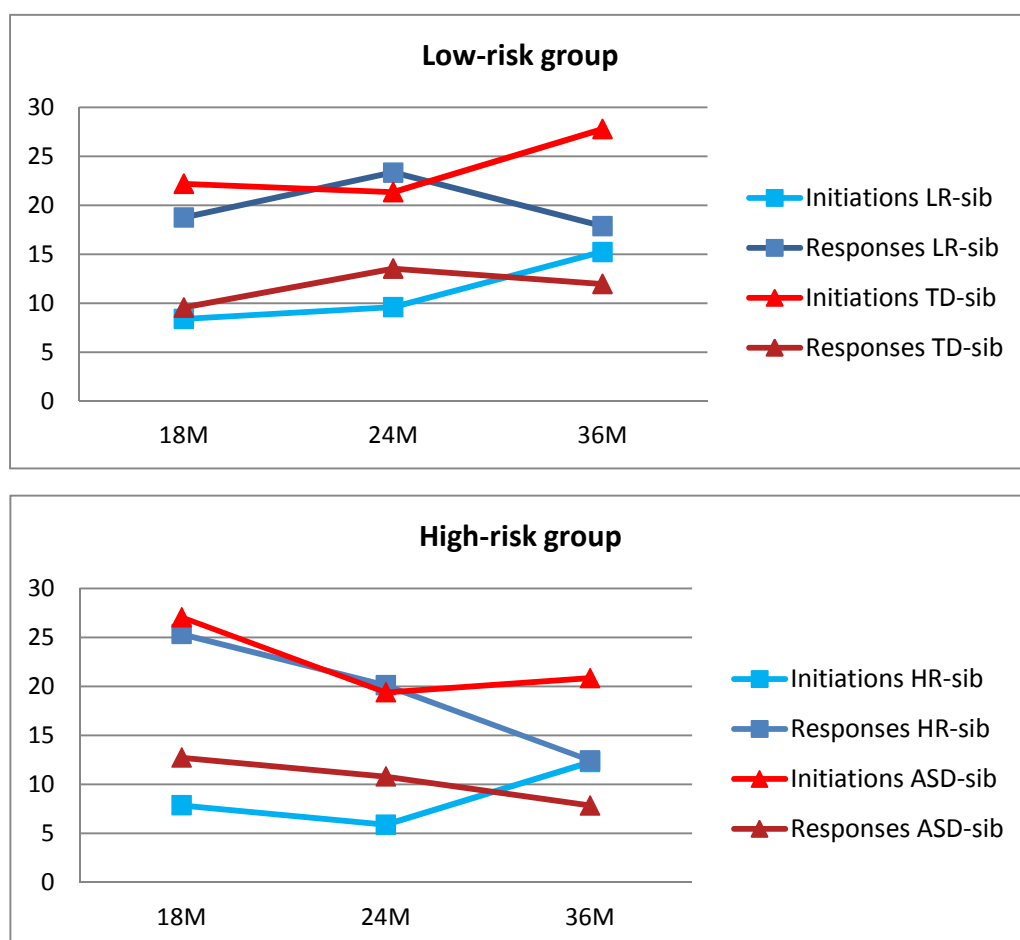


Figure 2. Frequency of total initiations and responses of both siblings in the HR and LR group at 18, 24, and 36 months – Evaluation of role (a)symmetry

If HR-sibs' initiations continue to climb while ASD-sibs' initiations decrease, it is possible that the roles will reverse and HR-sibs will take over the dominant position, as suggested in studies with older children (Knott et al., 1995; Smith, 2010). It is possible

that, at 18 months, HR-sibs lacked the social motivation and/or social-communicative skills to take over the lead of the interaction (Howes & Matheson, 1992; Lamb, 1978). As HR-sibs get older and acquire more social-communicative skills, they may be increasingly able and motivated to assume a more dominant position. In the transition from infancy to toddlerhood to early childhood, role patterns between children with ASD and their younger sibling may evolve from asymmetrical in favour of the child with ASD, to asymmetrical in favour of the HR-sib.

At 18 months (see Chapter 4), HR-sibs' sibling interactions were characterised as being more *negative* than LR-sibs' sibling interactions. In addition, at 24 and 36 months (see Chapter 5 and 6, respectively) HR-sibs' sibling interactions were less *positive*. Different patterns of change over time in both groups could help us understand why the groups differences change from differences in negative behaviours to differences in positive behaviours. In line with the longitudinal study of Abramovitch, Corter, Pepler, and Stanhope (1986) in a (slightly older) typically developing sample, positive and negative sibling interactions in the LR group increased over time. However, in the HR group negative behaviours of both siblings seemed to be more frequent at 18 months than at 24 months. The young age of the HR-sibs might entail certain challenges for the sibling interaction that are specific to the HR group. On the one hand, at 18 months, the social-communicative skills needed to actively interact with others are still developing, including empathy and perspective-taking abilities (Howes & Matheson, 1992; Lamb, 1978; Nilsen & Graham, 2009). Therefore, the social approaches of HR-sibs might lack quality (e.g., taking something instead of requesting) and HR-sibs may be more likely to disturb the play of their sibling with ASD. Given the social-communicative impairments (i.e., difficulties initiating and responding to social interaction) of children with ASD combined with lower levels of flexibility and an insistence on sameness (American Psychiatric Association, 2013), children with ASD may be more likely than typically developing children to respond negatively to these social approaches. On the other hand, given these social-communicative deficits and sometimes behavioural difficulties of children with ASD (American Psychiatric Association, 2013; Fitzpatrick, Srivorakiat, Wink, Pedapati, & Erickson, 2016), negative approaches of the child with ASD are to be expected as well. Moreover, at 18 months, HR-sibs are probably not fully capable of showing understanding or tolerance, again resulting in more negative exchanges. Results

further suggested that *positive* behaviours of the child with ASD were more frequent at 18 months, also reflected in the approximately 50-50 ratio in positive-negative behaviours in the HR group at 18 months. Thus, even though the challenges associated with interacting with a child with ASD more frequently resulted in conflict or negative exchanges, it might also be easier for children with ASD to approach their sibling when they are very young and not yet capable to lead or dominate the interaction.

**Association with development.** Up to now, we provided evidence for early differences in the sibling interactions of HR-sibs, both in the characteristics of social interactions (e.g., positive vs. negative) as in the observed role patterns. Given the importance of sibling interactions for the early development of children (Bank, Patterson, & Reid, 1996; Brody, 2004; Buist & Vermande, 2014; Harrist et al., 2014), these altered sibling interactions are likely to influence HR-sibs' social experiences needed for beneficial developmental outcomes. However, to date there are no studies evaluating the association between HR-sibs' sibling interactions and their development. Two studies included in this dissertation (Chapter 5-6) aimed to explore the cross-sectional association between the sibling interaction and the language and social-communicative development. Regarding the language development, results initially showed significant associations between the sibling interaction and receptive language/word comprehension as well as expressive language/word production at 24 months. However, these associations were better explained by pre-existing language abilities at 14 months. Thus, for the time being, we did not find support for the link between early sibling interactions and concurrent language development. Next, we evaluated the association between sibling interactions and the presence of ASD characteristics. At 24 months, the total amount of initiations and responses (i.e., positive and negative combined) was associated with more ASD characteristics in HR-sibs. At 36 months, ASD characteristics of both HR-sibs and children with ASD were taken into account. In contrast with the results at 24 months, the total amount of initiations and responses during the sibling interaction at 36 months was negatively associated with ASD characteristics. More ASD characteristics of the child with ASD were associated with fewer positive sibling interactions, whereas more ASD characteristics of the HR-sib were associated with fewer negative sibling interactions.

The cross-sectional nature of our study design inhibited us to evaluate the direction of these associations. Moreover, the relationship between social-communicative behaviours during the sibling interaction and ASD characteristics is presumably bidirectional. The deficits related to ASD are likely to influence the sibling interaction, but the sibling interaction could in turn impact on the expression of ASD characteristics (e.g., through social learning processes). Furthermore, how sibling interactions influence the expression of ASD characteristics and vice versa may depend on the developmental stage of the child (Inguaggiato et al., 2017). For example, given that children are usually not diagnosed before the age of three years (Sheldrick, Maye, & Carter, 2017), ASD characteristics are probably not yet fully expressed at the age of 24 months. Perhaps in this stage of development, sibling interactions are more likely to influence the ASD phenotype than vice versa. Through social learning (i.e., modelling, (deferred) imitation), HR-sibs may learn or imitate ASD characteristics from a sibling with ASD, especially if that sibling is older (Petalas et al., 2012). A stepwise regression model was tested to evaluate which behaviours (i.e., initiation vs. response, positive vs. negative, youngest vs. oldest) were associated with ASD characteristics at 24 months. Only the amount of negative responses of the child with ASD positively predicted ASD characteristics in the HR-sib ( $R^2=.31$ ,  $\beta=.559$ ,  $t=2.234$ ,  $p=.047$ ), which suggests that behaviours of the child with ASD could affect the ASD phenotype in HR-sibs.

At 36 months, on the other hand, ASD characteristics might be more stable and less susceptible to environmental influences. Thus at this age, it seems more likely that ASD characteristics (of both children) influence the sibling interaction rather than the other way around. Given that social-communicative impairments are core features of ASD (American Psychiatric Association, 2013) and that positive behaviours occurred less frequently in the HR group, the association between ASD characteristics in the child with ASD and fewer positive behaviours was in line with our expectations. More specifically, stepwise regression analysis revealed a significant negative association between ASD characteristics and positive initiations of the child with ASD ( $R^2=.39$ ,  $\beta=-.622$ ,  $t=-2.863$ ,  $p=.013$ ). In contrast, ASD characteristics of the HR-sib were associated with fewer negative behaviours. When interacting with a sibling with ASD, HR-sibs with fewer ASD characteristics might experience feelings of frustration, incomprehension, etc. In addition, they may interrupt the routines or play activities of their sibling with ASD. In

turn, this may lead to more negativity during the sibling interaction. When the HR-sib shows ASD characteristics as well, their capabilities during and expectations about the sibling interaction might be more matched to those of their older sibling with ASD, leading to less conflicting sibling interactions.

**Conclusion.** Even though the total amount of mutual interaction between both siblings was low in both groups, the sibling interactions of the LR and HR group significantly differed at all three time points. While it remains plausible that these differences in the early learning context will influence HR-sibs' development (e.g., language development), this was not fully substantiated by the current findings. The association between ASD characteristics and the sibling interaction, however, does indicate that sibling interactions could have the potential to significantly impact on the expression of the ASD phenotype in HR-sibs. Given the limitations of the current studies, more elaborate studies are needed to fully assess the potential impact of sibling interactions on HR-sibs' developmental trajectories. Moreover, it needs to be noted that there were various similarities as well (e.g., no differences in positive behaviours at 18 months and no differences in negative behaviours at 24-36 months). This could imply that not all aspects of the sibling interaction differ between groups and that children with ASD do provide learning opportunities for HR-sibs. Therefore, future studies should include the evaluation of differences (or weaknesses) in the sibling interaction of HR-sibs as well as the evaluation of similarities (or strengths/learning opportunities). Perhaps these similarities combined with other social influences (e.g., parents, day care, peers) will compensate for potential deficits or weaknesses in the sibling interaction. Lastly, it is possible that HR-sibs (temporarily) acquire ASD characteristics by modelling or imitating their sibling with ASD. Especially in the absence of other social experiences such as day care, social learning processes might lead to 'autistic-like' behaviours in younger HR-sibs. If other social relationships with peers are present as well (e.g., day care, school), HR-sibs' opportunities to imitate/copy and acquire adequate social-communicative and play behaviours increase and the expression of ASD characteristics might decrease. It is therefore important to separate inherent ASD characteristics from those ASD characteristics that might be learned in interaction with a sibling with ASD.

## Peer interaction

Research suggests that sibling interactions influence later peer interactions (Roskam, Meunier, & Stievenart, 2015). Therefore the association between sibling and peer interactions of HR-sibs was further investigated in Chapter 6. First, the comparison of sibling and peer interactions in the HR group revealed meaningful findings. More specifically, HR-sibs responded somewhat more negative to their older sibling with ASD than to their peer. Moreover, physical proximity was higher during the peer interaction than during the sibling interaction which could indicate that peer relationships are closer/more intimate. Given that proximity did not differ between the HR and LR group, physical proximity might be characteristic for peer relationships in general (and not only in the HR group). Also, physical proximity could be an expression of learned behaviour in the preschool classroom, a context in which children are frequently engaged in structured activities in small groups. In contrast, sibling interactions are more unstructured and children are more free to choose whether they play alone or with their sibling. Nevertheless, HR-sibs' sibling interactions appear to be more negative than their peer interactions. The study of Abramovitch, Corter, Pepler, and Stanhope (1986), comparing sibling and peer interactions in typical development, reported higher levels of prosocial behaviours during the peer interaction. However, they did not find any differences in negative behaviours. Although the comparison with a typically developing control group is needed to confirm this, these results suggest that the higher frequency of negative behaviours during the sibling interaction may be characteristic for HR-sibs.

Next, negative sibling interactions were associated with higher levels of negative peer interactions. This study was the first to evaluate the association between sibling and peer interactions of HR-sibs, therefore there is no other empirical support for these findings. We hypothesise that HR-sibs learn a specific, perhaps more negative interaction style in interaction with their sibling with ASD and generalise this interaction style to other social contexts such as the peer interaction. This is in line with other studies suggesting that individuals generalise their interaction style across contexts (e.g., Lockwood, Kitzmann, & Cohen, 2001). Based on results from a typically developing sample, Yucel and Downey (2015) state that sibling interactions provide the foundation for later peer interactions and that negative interactional patterns learned during the sibling interaction might persist during peer interactions. As a result, negative sibling

interactions may lead to negative peer interactions. Because sibling relationships precede peer relationships, it is more plausible to assume that sibling relationships influence peer relationships than vice versa.

On the other hand, Yucel and Downey (2015) also hypothesised that peer relationships have the potential to influence sibling relationships. In interaction with a peer children could acquire social-communicative skills they can translate to the sibling relationship (Howe, Ross, & Recchia, 2011; Yucel & Downey, 2015). As shown in Figure 1, HR-sibs' social initiations increased from 24 to 36 months whereas this was not the case from 18 to 24 months. It is possible that differences in HR-sibs' early sibling interactions (i.e., at 18-24 months) influenced the social approach behaviours of HR-sibs at 24 months. However, between 24-36 months HR-sibs start school, meaning that sibling interactions are supplemented with social interactions with peers, possibly promoting their social initiations. When looking at HR-sibs' day care experiences, we for example found a trend for a positive association between the hours spent in day care at 18 months and HR-sibs positive initiations at 18 months ( $p=.395$ ,  $p=.069$ ). However, not all HR-sibs attended day care (i.e., only 64%) whereas all HR-sibs started school before the age of 36 months.

**Conclusion.** These results demonstrate that HR-sibs' behaviours are somewhat more negative in interaction with a sibling compared to a peer. At the same time, HR-sibs generalise a more negative interaction pattern from sibling interactions to peer interactions. However, the fact that sibling interactions remain more negative than peer interactions suggests that especially the interaction with a sibling with ASD triggers negative responses in HR-sibs. It is possible that, due to prior experiences, HR-sibs are less tolerant towards their sibling with ASD, or that they have learned to stand up to them more. Although this was beyond the scope of the current dissertation, it is also possible that HR-sibs acquire social-communicative abilities in interaction with a peer which could benefit their sibling interactions (e.g., Howe et al., 2011; Kramer & Kowal, 2005).

## IMPLICATIONS OF THE RESEARCH FINDINGS

### Theoretical and methodological implications

On a theoretical level, this dissertation provided evidence for differences in HR-sibs' early social environment, especially in terms of altered sibling interactions. In addition, there was some support for the association between early social interactions and child development. This provides evidence for the hypothesis that environmental factors including early experiences may influence the development of neural circuits (i.e., brain plasticity) and shape individuals' physical as well as mental development (Dawson, 2008; Inguaggiato et al., 2017; Mandy & Lai, 2016). Brain plasticity is present throughout the lifespan, but appears to be most prominent in early life (Barbaro & Dissanayake, 2012; Elsabbagh & Johnson, 2010; Inguaggiato et al., 2017). In addition, Mandy and Lai (2016) concluded that early social experiences could modify a pre-existing susceptibility to ASD. Nevertheless, the studies discussed in this dissertation were the first to include early sibling interactions as a part of the early social environment and evaluate the association between early social interactions and HR-sibs' ASD characteristics as well as broader developmental domains (e.g., language). Despite the limitations of these studies, significant differences emerged in the early learning context of HR-sibs, which are likely to influence the HR-sibs' development. To some extent we also confirmed the association between early social interactions and (later) development, but further studies including a longitudinal design, larger samples matched on gender, age and dyad constellation, and more elaborate measures of child development are needed to fully explore the association between the early interactions and subsequent development. Moreover, given that the current results indicated that negative sibling interactions are at least partly generalised to the peer context, HR-sibs' peer interactions need to be further explored and compared with a low-risk control group.

Methodologically, Chapter 6 revealed that the informant used to report ASD characteristics of HR-sibs requires further consideration. The correlations between parent- and teacher-report were rather low (for HR-sibs' SRS:  $r=.22$ ,  $p=.480$ ), suggesting a low consensus between parents and teachers. It is possible that ASD characteristics are expressed differently in the home context than at school. As stated in the manual of the Social Responsiveness Scale (SRS-2; Constantino & Gruber, 2012), it is likely that children



(have to) display different behaviours in a school context. The interrater reliability between parents and teachers reported in the manual was also low ( $r=.28$ ) Teachers might be more suited to report on the expression of ASD characteristics in the classroom while parents provide more reliable information regarding ASD characteristics at home. Another possibility is that parents and teachers have another frame of reference. Parents of HR-sibs have a child with ASD, which may influence their perception of what is to be expected of a typically developing child. Teachers on the other hand interact with typically developing children on a daily basis, perhaps giving them a more accurate view on typical development. Consequently, a combination of multiple informants might be needed to provide a thorough description of ASD characteristics in young children. The added value of teacher-report to identify children with ASD is also emphasised in the manual of the SRS-2 (Constantino & Gruber, 2012).

Another important aspect that requires further discussion is the choice of coding method (i.e., global vs. frequency). The evaluation of coding methods in Chapter 2 as well as previous studies shows that each method has its merits, depending on the level that is being examined (macro vs. micro; e.g., Russel, 2011). If sufficient resources are available, a combination of both could be desirable to provide a thorough description of the interaction being evaluated. Global ratings are especially suited to address questions of quality and enable the rater to integrate information from multiple sources (e.g., different contexts, multiple interaction partners; Bakeman & Quera, 2011; Grotevant & Carlson, 1989). To get a qualitative description of the course of the interaction and because we wanted to evaluate a broad range of behaviours in a time-efficient manner, a global coding scheme was used to assess the parent-child interaction. In contrast, to evaluate sibling interactions we were more interested in the specific dynamics and interactional patterns that constitute the early learning environment of children. To this end, a more detailed, frequency coding scheme was used (supplemented by several global rating scales). Thus, in preparation of an observational study it is important to reflect on the most adequate coding method. If researchers are interested in a rather broad and qualitative description of social interactions, a global coding scheme seems more appropriate. This allows the researcher to include constructs for both interaction partners that are proven to be important within child development. However, given that global ratings most commonly reflect how the interaction partners behave *on average*,

information is lost regarding the specific interactions or dynamics. For example, within an average score of sensitivity, it is possible that parents are sensitive half of the time and miss child signals during the other half, but it is also possible that parents show a combination of sensitive and insensitive behaviours. If researchers are interested in collecting very detailed information, it might be better to opt for a frequency coding method. To continue the example of parent sensitivity, researchers could code how often parents respond to a child signal in a sensitive way, in an insensitive way, or how often they fail to respond to a child signal. If needed, the child's signals could also be classified into more subgroups (e.g., positive vs. negative). However, this requires moment-by-moment coding and can be very time-consuming. As a result, applying the frequency coding scheme to multiple behaviours might not be feasible.

During the observation of sibling interactions, different play materials were used to elicit different levels of play (i.e., parallel, associative, or cooperative play). It is therefore possible that each context elicited different behaviours. Because group differences were largely similar in all contexts, the different play contexts were combined when possible within this dissertation to present the results more clearly. Comparing all contexts did not reveal a consistent pattern, which inhibited us to make any specific predictions as to which context elicited which behaviours. However, the building blocks (Duplo™)/Playmobil™ seemed to elicit the least interactive behaviours (both positive and negative), whereas the marble run elicited both negative and positive behaviours. On the one hand, the building blocks (Duplo™)/Playmobil™ allowed for too much solitary play, which discouraged interactive behaviours. On the other hand, the keyboard did not allow for joint play, which often resulted in a short episode of conflict followed by solitary play. The marble run seemed to lead to a good balance of both solitary and joint play and was probably best suited to observe the sibling interactions. Moreover, both the youngest and oldest child still enjoyed the marble run, despite of the age difference between both children. Only providing one toy at a time also promoted the children's orientation towards each other, which was why we preferred this to offering all toys at once. Although the specific type of toy should be based on the children's age, a toy that allows for both joint and solitary play seems best suited to observe sibling interactions.

Finally, it needs to be noted that the quality and nature of social interactions strongly depend on the moment these interactions are measured. Repeated measures analyses revealed that, in parent-child as well as sibling interactions, the nature of interactions significantly change over time. It is therefore important, when measuring social interaction, to carefully consider at what age these interactions should be measured. This could have important implications for the behaviours that are being observed and the association with development. Although further research is needed to evaluate the predictive value of the social interactions at different time points, it is possible that observing social interactions during the first two years of life might be more valuable to assess the association with the emerging ASD phenotype. From 36 months onwards, the ASD phenotype might be more stable and less susceptible to environmental influences.

### **Clinical implications**

Although we were not yet able to distinguish HR-sibs with and without an ASD diagnosis, the results do suggest that HR-sibs as a group show differences in their early sibling interactions compared to the LR group. Studies including older children or adolescents/adults show that positive sibling relationships are important for siblings of children with ASD (Tomeny, Ellis, Rankin, & Barry, 2017). For example, a positive attitude towards the relationship with their sibling with ASD is important for their general life satisfaction. Moreover, positive sibling relationships benefit child development (e.g., Harrist et al., 2014). However, both our results and previous studies including older children (also see Beyer, 2009) suggest that, in some cases, sibling interactions including a child with ASD are characterised as less positive, either based on observations, parent-report, or self-report. Especially when combined with for example differential parental treatment (or parental favouritism towards the child with ASD), siblings of children with ASD perceive their sibling relationship as less positive (McHale, Sloan, & Simeonsson, 1986; Rivers & Stoneman, 2008). Furthermore, the current results showed that, at 36 months, negative behaviours were almost twice as frequent as positive behaviours, which is also likely to influence child and family functioning. Given the beneficial impact of positive sibling relationships on child outcome reported in previous studies (Brody, 2004; Buist & Vermande, 2014; Harrist et al., 2014), the promotion of positive sibling

relationships could benefit the development and well-being of siblings of children with ASD. One strategy for increasing positive interactions, is to teach play skills to both siblings during a game they can play together (Beyer, 2009). In addition, parental supervision might increase positive sibling interactions as well. For example, McHale et al. (2000) showed that more time spent by the sibling dyad in the company of parents was linked to better sibling relationships.

On a diagnostic level, there is insufficient evidence to conclude that the assessment of sibling interactions (or parent-child or peer interactions) might improve the diagnostic process of ASD. However, these observations might reveal possible strengths or weaknesses in the learning environment of HR-sibs as well as targets for intervention. If, for example, parents would characterise the early sibling relationship of their children as highly negative or indicate that positive sibling interactions do not occur regularly, specific strategies (e.g., teaching play skills to improve positive interactions) could be integrated in for example the intervention programs of home guidance services. For example, in a study including preschool-aged typically developing siblings of children with ASD it was demonstrated that these siblings were able to learn skills (e.g., joining the play of the child with ASD) to promote social play with their brother with ASD (Oppenheim-Leaf, Leaf, Dozier, Sheldon, & Sherman, 2012). Moreover, for some children this resulted in an increase of positive behaviours and a decrease of negative behaviours during play with their sibling with ASD.

Another clinical consideration involves the social experiences of HR-sibs outside of the sibling interaction. Somewhat against what we might have expected, sibling interactions were not characterised by more conflict at 24 or 36 months. Instead, there was a lack of positive social input. Nevertheless, the associations with HR-sibs' development were limited, suggesting these lower levels of social input in itself might not influence the HR-sibs' development as expected. In addition to the interaction with a sibling with ASD, other social experiences might have influenced HR-sibs as well (perhaps compensating for lower levels of input during the sibling interaction). For example, the hours spent in day care at 18 months were (marginally significantly) correlated with HR-sibs positive initiations at 18 months ( $\rho=.395$ ,  $p=.069$ ) and negatively correlated to HR-sibs' negative initiations at 24 months ( $\rho=-.47$ ,  $p=.044$ ). Social experiences during day care, whether or not in interaction with social experiences

during other social contexts such as the sibling interaction, may also influence the developmental trajectories of HR-sibs. It is possible that the general social context of HR-sibs, including all relevant social experiences, influences their development rather than one context in itself. Perhaps the experiences with typically developing children in day care protect HR-sibs against the negative effects of fewer positive experiences during the sibling interaction. In this regard it is important to note that HR-sibs less frequently attended day care than LR-sibs (18 months: 64% vs. 93%,  $\chi^2(1) = 6.89$ ,  $p=.013$ ; 24 months: 70% vs. 93%,  $\chi^2(1) = 5.22$ ,  $p=.031$ ). More research is needed, including the interaction between different social contexts, but it might be valuable to encourage day care attendance in (at least some) HR-sibs to provide them with opportunities to experience positive social interactions with peers.

Lastly, results showed that, in the HR group, negative sibling interactions might be generalised to the peer relationship. Although further research is needed to determine whether HR-sibs' peer relationships are also more negative than LR-sibs' peer relationships, negativity in the HR-sib's sibling relationship could adversely affect the peer relationship (Lockwood et al., 2001), in turn influencing the learning context in the preschool classroom. If indeed HR-sibs' peer relationships are more negative compared to those of LR-sibs, it could be important for teachers to monitor the peer relationships of HR-sibs and possibly play a role in the prevention of the generalisation of negative behaviours.

## STRENGTHS AND LIMITATIONS

### Strengths

**Prospective follow-up study of social interactions during the first years of life.** Atypicalities in the early developmental trajectories of HR-sibs emerge around the first year of life (Szatmari et al., 2016). In addition, the impact of early social experiences on development may be more pronounced during early developmental stages (Inguaggiato et al., 2017). Nevertheless, studies including siblings of children with ASD mainly focus on school-aged children or adolescents (e.g., Kaminsky & Dewey, 2001; Knott et al., 1995; Walton & Ingersoll, 2015). The studies included in this dissertation are the first to

include HR-sibs' early social experiences during the first years of life (parent-child interaction: 5-10 months; sibling interaction: 18-24-36 months), which enabled us to assess the social interactions at a time when these experiences are more likely to impact child development. Furthermore, we used a prospective study design to assess the development of HR-sibs over time. This allowed us to evaluate HR-sibs development at different time points in the same sample, including the exploration of changes over time. Finally, the prospective design allowed us to evaluate the predictive value of early parent-child interactions for child development at 24 months.

**Observational method.** Even though they provide valuable information, questionnaires entail certain disadvantages. Specifically when the focus is on early social interactions in a very young sample, questionnaires might not be the best option. First, self-report is not possible in infants and toddlers. In addition, there are studies reporting rater bias in parents and differences between parent- and child-report (Rivers & Stoneman, 2008; Stone, Hoffman, Lewis, & Ousley, 1994), thus parent-report might not provide an accurate description. Finally, whereas relationship quality can be translated into specific questions (e.g., "how much do your children quarrel with each other?"), this is not the case for specific interactional patterns. By using an observational method, we were able to obtain detailed information about the social interactions while avoiding these disadvantages. Then again, a possible disadvantage of our observational method lies in the fact that interactions are only observed at one occasion. However, in the vast majority of our observations parents indicated that the play observation was representative for a typical play episode at home. Future studies could also supplement an observational method with parent-report by means of questionnaires. Perhaps this would provide a more detailed and nuanced description of sibling interactions.

**Novel coding scheme.** For the studies in this dissertation, the choice of coding scheme was carefully considered. Both for the parent-child interaction and the interaction between siblings/peers, a novel coding method was developed based on existing research. This enabled us to include all constructs relevant for the research questions of this dissertation. Moreover, all behaviours/scales were described in detail, enabling students to reliably code the interactions. It needs to be noted that the development of a novel coding scheme required a lot of work and did not guarantee that it would be sensitive enough to detect group differences. To establish the value of a

coding scheme, the coding scheme needs to be validated in a typically developing population as well as the clinical population for which it is intended. To state with certainty that the coding schemes used in this dissertation are suited to code interactions of LR- and HR-sibs, further validation is needed. This is especially the case for the parent-child interaction coding scheme, for which group differences were limited. The sibling interaction coding scheme did detect significant group differences, which is a first step in assuring the coding scheme is suited to code sibling interactions of LR- and HR-sibs. Nevertheless, further replication of the results in other samples is needed. Researchers could also opt for a coding scheme that is already validated, but in that case the researcher is limited to the constructs defined in the coding scheme. Moreover, an intensive training would be needed to ensure that the coding scheme is adequately used, which is also time-consuming. Altogether, within this dissertation, the benefits of developing a new coding scheme (e.g., comprehensive, adapted to the research questions) outweighed the disadvantages (e.g., lack of validation).

### **Limitations**

Several specific limitations were already discussed in each of the separate chapters (and in the previous sections in this chapter). In this section, we would like to direct attention to some limitations referring to the dissertation in its totality.

As mentioned above, the longitudinal, prospective nature of the study design has clear advantages, but there are also some limitations that require further discussion. With the exception of Chapter 2, all chapters/studies relied on a subsample of children participating in the broader longitudinal follow-up study of younger siblings of children with ASD and a low-risk control group. This led to detailed information on the development of the high- and low-risk siblings, but limited the capacity to include measures for other interaction partners (e.g., older siblings, parents). The measurement of for example the cognitive functioning of the older children (with ASD) would have required an additional testing moment, which was not feasible within the longitudinal protocol. It is recommended that future studies include detailed measures of HR-sibs as well as of other interaction partners (e.g., parents, siblings with ASD). For example, children with ASD are likely to differ in terms of cognitive development, strengths and weaknesses, etc. (Meirsschaut et al., 2011), impacting on the nature of sibling

interactions. In addition, this dissertation was finalised when information regarding the HR-sibs' ASD outcome was not yet available for the entire sample. Therefore it was not possible to distinguish typically developing HR-sibs from HR-sibs with ASD or BAP. The nature of the parent-child, sibling, and peer interactions should be re-evaluated based on the diagnostic outcome of HR-sibs (i.e., TD, BAP or ASD). Further, at each age the parent-child or sibling interaction was observed on one occasion only during a short play observation. As a result, these observations were susceptible to random variations (e.g., parent/child being sick, child in a bad mood). Multiple observations or longer lasting observations were unfortunately not feasible.

A limitation related to the general sample, is the small sample size. First, this led to a decreased power and an increased probability of making Type II errors. Even though significant results were detected, it is possible that other real life differences did not reach level of significance due to the low power. To avoid a further reduction of statistical power, we chose not to correct for the Type I error rate due to multiple comparisons (Nakagawa, 2004; Perneger, 1998). Therefore, we also cannot exclude the possibility that some significant results were observed due to chance. Second, we were limited in the statistical analyses that could be used. Due to a lack of normal distribution, data were often analysed by means of nonparametric analyses. More elaborate, parametric analyses that could have included other factors such as the role of sample characteristics, were not possible. In addition, the number of predictors used in the regression model was limited. Even though small sample sizes are common in clinical populations and the sample size of the current study exceeded the sample size of the observational studies of for example Knott et al. (1995, 2007), replication of results in a larger sample is necessary. It should, however, be noted that collecting data on the sibling interaction at three time points during the first three years of life from both a LR and HR group is not evident and entails several challenges. For example, keeping the interest of their child with ASD in mind (e.g., need for structure/sameness), parents sometimes refused home observations. In addition, collecting data regarding the younger child's development as well as the sibling interaction is time-consuming for parents. As a result, not all parents are willing or able to engage in a longitudinal follow-up study.



The results of the studies included in this dissertation relied on correlational patterns or regression analyses. The main limitation regarding this analytic strategy is that it does not allow the researcher to infer causality. Even though some associations are theoretically more plausible than others (e.g., sibling interactions predicting peer interactions), we can't exclude the possibility that the reversed association exists as well. Furthermore, there was little exploration of alternative explanations. Due to the small sample size, more elaborate covariance or mediation analyses were not possible. In addition, the longitudinal protocol did not allow for the inclusion of more detailed information (e.g., parental functioning). Thus, even when an association was found, there may be other variables moderating or mediating this relationship. Therefore, statements in terms of causality were not possible.

**Sibling interaction.** The study design and sample of the sibling interaction studies entailed some limitations as well. First, this study focused on the *younger* siblings of children with ASD. However, because we could not divide the HR-sibs into subgroups (TD, BAP, ASD), this mixes two possible effects. Possible differences in the sibling interaction could be due to either characteristics of the older child with ASD or to characteristics of the younger HR-sib (or a combination of both). We were unable to determine the relative contribution of each sibling. One way to do this was to observe HR-sibs in interaction with an older, typically developing sibling. However, not all HR-sibs had an older typically developing sibling. In addition, this would have required additional testing moments, which was not possible due to pragmatic reasons.

Unfortunately, only nine HR-sibs were seen at all three time points (18, 24, 36 months). Due to various reasons, HR-sibs' sibling interactions were frequently not observed at one or two of the three time points (e.g., parents entered the study at a later time point, children were already older than 18 months when the study on sibling interactions started, or parents chose not to participate in the sibling interaction study). Consequently, longitudinal analyses studying the evolution from 18 to 36 months were not possible. Similarly, the sample was too small to predict development at 36 months based on previous sibling interactions at 18 or 24 months. There is a need for longitudinal studies that assess the impact of early social experiences on *later* development rather than the concurrent development

## DIRECTIONS FOR FUTURE RESEARCH

Several specific directions for future research were given after the discussion of the limitations. In this section, we would like to discuss two broader directions for future research.

Within this dissertation, parent-child, sibling, and peer interactions were mostly evaluated as separate interactional systems. In reality, these systems influence and interact with each other. The interplay between these familial and extrafamilial influences will contribute to the social development of young children (Russel, 2011). This is also in line with the bioecological theory of human development as proposed by Bronfenbrenner (1979). For example, through processes such as differential parenting and the mediation of sibling conflict, parent-child interactions are likely to influence sibling interactions (McHale, Updegraff, & Feinberg, 2016). In addition, peer relationships could buffer the effect of negative parent-child relationships or strengthen the influence of positive parent-child interactions (Reich & Vandell, 2011). Consequently, there is a need for studies that integrate these different social contexts and evaluate causal processes as well as reciprocal influences among different levels of the system. The studies included in this dissertation aimed to describe HR-sibs' early social interactions in detail and to provide a first exploration of the association with outcome. It was outside the scope and possibilities of this dissertation to include the interplay between different social contexts. Moreover, the small sample size did not allow for more elaborate analyses. In a larger sample, the individual influence of each interactional system as well as the relative value of each system to predict child development could be explored. This would require a longitudinal study design including the assessment of parent-child, sibling and peer interactions during the first years of life as well as outcome measures at a later time point.

There is an important distinction between a within- and between-family study design. The results reported in this dissertation are mainly based on between-family comparisons between families with a child with ASD and families with typically developing children. Although the between-family design was suited to answer the research questions, a within-family design could have merits as well. In a between-family design, both interaction partners that are compared differ, whereas in a within-

family design, one interaction partner is the same person in both groups (i.e, one parent in interaction with either a child with or without ASD, the high-risk sibling in interaction with a sibling with ASD and a typically developing sibling). Thus, a within-family design controls for variability in one of the interaction partners, which enables researchers to make more reliable conclusions. Consequently, future studies with a within-family design could provide valuable information on HR-sibs early social interactions.

### **FINAL CONCLUSION**

This doctoral dissertation provided a detailed description of the social environment of younger siblings of children with ASD during the first years of life. While the social experiences of these HR-sibs showed several similarities with the low-risk control group, especially in the parent-child interactions, meaningful differences emerged as well. With the exception of lower levels of positive affect in the HR group in the Belgian sample, the parent-child interaction did not differ from typical development at the ages of 5 and 10 months. In contrast, sibling interactions between HR-sibs and their older sibling with ASD significantly differed from sibling interactions between typically developing children. Regarding the possible influence of these early social experiences on child development, this dissertation provided some evidence for the potential of early social interactions to influence HR-sibs' (atypical) developmental trajectories. However, the evidence was not irrefutable and further research is needed to replicate these results and provide clarity regarding the direction of these associations as well as the association between social interactions and later developmental outcomes. If these associations are confirmed in longitudinal studies with larger samples, early social experiences are an important factor to consider when studying the ASD phenotype.

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### INLEIDING

Autismespectrumstoornis (ASS) is een ontwikkelingsstoornis die wordt gekenmerkt door tekorten in de sociale communicatie en sociale interactie alsook door patronen van repetitief gedrag, interesses of activiteiten (American Psychiatric Association, 2013). De prevalentie van ASS wordt geschat op 60 à 70 per 10,000 kinderen, met een verhouding van 4 mannen op 1 vrouw (Elsabbagh et al., 2012; Fombonne, 2009). Onderzoek toont aan dat, in vergelijking met de algemene bevolking, ASS vaker vastgesteld wordt bij eerstegraadsverwanten (ouders, broers/zussen) van kinderen met ASS (Ozonoff et al., 2011; Sasson, Lam, Parlier, Daniels, & Piven, 2013). Meer specifiek krijgt 14-23% van de ouders en 18.7% van de broers/zussen (hierna *siblings*) een diagnose van ASS (Ozonoff et al., 2011; Sasson, Lam, Parlier, et al., 2013). Daarnaast vertonen eerstegraadsverwanten die geen ASS ontwikkelen vaker ASS-gerelateerde kenmerken die onvoldoende ernstig zijn om aan de criteria van ASS te voldoen. Deze subklinische kenmerken van ASS vallen onder het *brede autisme fenotype* (Bailey, Palferman, Heavey, & Le Couteur, 1998; Georgiades et al., 2013; Sucksmith, Roth, & Hoekstra, 2011). Vanwege het toegenomen risico op (subklinische kenmerken van) ASS worden jongere *siblings* van kinderen met ASS ook wel *hoog-risico siblings* (HR-sibs) genoemd. Reeds vanaf de eerste levensjaren vertonen HR-sibs tekorten op vlak van taal, wederkerige sociale interactie en cognitief functioneren, onafhankelijk van een latere ASS-diagnose (Brian et al., 2014; Gamliel, Yirmiya, & Sigman, 2007; Georgiades et al., 2013; Hudry et al., 2014; Landa & Holman, 2007). Daarom is het belangrijk om de ontwikkeling van deze HR-sibs op te volgen, ook indien er geen sprake is van een ASS-diagnose (Szatmari et al., 2016). Het brede autisme fenotype bij ouders uit zich eerder in tekorten in taalpragmatiek, minder vriendschappen of een verminderde interesse in sociale activiteiten (Sucksmith et al., 2011). Hoewel een betrouwbare diagnose van ASS reeds mogelijk is vanaf de leeftijd van 2 jaar, wordt slechts een minderheid van de kinderen (17-23%) gediagnosticeerd voor de leeftijd van 3 jaar (Barbaro & Dissanayake, 2009; Charman et al., 2005; Chawarska, Klin, Paul, Macari, & Volkmar, 2009; Sheldrick, Maye, & Carter, 2017). Gezien hun verhoogd risico op (subklinische kenmerken van) ASS,

zijn HR-sibs de afgelopen jaren meer en meer het onderwerp van longitudinale opvolgstudies. Door gedurende de eerste levensjaren de ontwikkeling op te volgen van HR-sibs die later ASS ontwikkelen, kunnen onderzoekers ASS-kenmerken op jonge leeftijd beter in kaart brengen. Dit kan op zijn beurt een vroegere diagnose van ASS mogelijk maken. Het opvolgen van HR-sibs, reeds vanaf enkele maanden na de geboorte, laat ook toe om kwetsbare HR-sibs zonder latere ASS diagnose (bv. HR-sibs die subklinische kenmerken van ASS vertonen) verder op te volgen en te ondersteunen (Szatmari et al., 2016).

De oorzaken van ASS zijn uiteenlopend, met onder meer een sterk aandeel van genetische factoren (Tick, Bolton, Happé, Rutter, & Rijdsdijk, 2016). Genetische factoren zijn echter onvoldoende om het volledige ASS-fenotype te verklaren. Zowel (epi-)genetische factoren, omgevingsfactoren (bv. leeftijd van de ouders, foliumzuur) alsook de gen-omgeving interactie kunnen het ontstaan en de ontwikkeling van ASS beïnvloeden (Mandy & Lai, 2016). Een belangrijke omgevingsfactor die vaak wordt verwaarloosd in de literatuur en tevens het onderwerp is van dit doctoraatsonderzoek, is de vroege sociale omgeving (bv. ouder-kind interactie, interactie tussen broers/zussen (sibling interactie); Dawson, 2008; Inguaggiato, Sgandurra, & Cioni, 2017). HR-sibs hebben immers niet alleen een genetische kwetsbaarheid, maar ook hun vroege sociale omgeving vertoont enkele belangrijke verschillen met deze van typisch ontwikkelende kinderen. Gezien de invloed van vroege sociale interacties, zoals de ouder-kind of sibling interactie, op de ontwikkeling van kinderen (bv. Clifford & Dissanayake, 2009; Harrist et al., 2014; Russel, 2011), is het belangrijk om met deze vroege relaties rekening te houden bij het in kaart brengen van de ontwikkeling van HR-sibs. Hoewel de sociale omgeving de mogelijke atypische ontwikkeling van HR-sibs niet *veroorzaakt*, kan ze wel mee bepalen hoe het ASS-fenotype zich uit gedurende de eerste levensjaren (Mandy & Lai, 2016). Desondanks houden studies slechts in beperkte mate rekening met de vroege sociale interacties van HR-sibs en hoe deze een rol zouden kunnen spelen bij de atypische ontwikkeling van deze kinderen. In wat volgt wordt dieper ingegaan op de bestaande literatuur omtrent de ouder-kind interactie, sibling interactie en interactie met leeftijdsgenoten bij kinderen met ASS en HR-sibs.

Als eerste toont onderzoek aan dat kinderen met ASS en hun gezin elkaar wederzijds beïnvloeden, wat een negatieve impact kan hebben op de gezinscontext en

op het welzijn van de ouders (bv. verhoogde stress; Karst & van Hecke, 2012). Ouders van kinderen met ASS hebben ook zelf een verhoogd risico op ASS of het breder autisme fenotype (Sasson, Lam, Childress, et al., 2013). ASS-kenmerken en een verminderd welzijn bij ouders, in combinatie met sociaal-communicatieve tekorten bij kinderen met ASS en HR-sibs, kunnen de **ouder-kind interactie** beïnvloeden. Zo toont onderzoek aan dat ouders van kinderen met ASS vaker directieve gedragingen stellen en minder responsief zijn in vergelijking met ouders van typisch ontwikkelende kinderen (Doussard–Roosevelt, Joe, Bazhenova, & Porges, 2003; Freeman & Kasari, 2013; Shapiro, Frosch, & Arnold, 1987). Anderzijds blijken ouders van kinderen met ASS even sensitief te zijn als ouders van typisch ontwikkelende kinderen en stemmen ze hun interactiestijl af op de noden van hun kind (bv. stimuleren van spel; Adamson, Bakeman, Deckner, & Nelson, 2012; Lemanek, Stone, & Fishel, 1993; van Ijzendoorn et al., 2007). In interactie met hun ouders zijn kinderen met ASS minder responsief, maken ze minder gebruik van oogcontact en zijn ze minder betrokken in de interactie dan typisch ontwikkelende kinderen (Dawson, Hill, Spencer, Galpert, & Watson, 1990; Dolev, Oppenheim, Koren-Karie, & Yirmiya, 2009; Doussard–Roosevelt et al., 2003). Ook de ouder-kind interactie van HR-sibs vertoont enkele verschillen in vergelijking met siblings van typisch ontwikkelende kinderen (i.e., laag-risico siblings; LR-sibs). Zo zijn ouders van HR-sibs vaker directief en minder sensitief ten opzichte van hun kind terwijl HR-sibs lager scoren op vlak van levendigheid, aandacht voor de ouder en positief affect (Wan et al., 2012, 2013; Yirmiya et al., 2006). Rozga et al. (2011) daarentegen vonden geen verschillen in de ouder-kind interacties van HR-sibs in vergelijking met LR-sibs.

Ten tweede is de kans groot dat ook de **sibling interactie** beïnvloed wordt door de sociaal-communicatieve tekorten bij kinderen met ASS en bij een proportie van de HR-sibs (bv. minder sociale uitwisselingen, meer negatief gedrag). Desalniettemin is onderzoek naar de sibling interactie tussen kinderen met ASS en hun siblings beperkt. Gezien de sibling interactie belangrijke leermogelijkheden biedt voor kinderen kunnen wijzingen in deze leercontext de ontwikkeling van beide kinderen beïnvloeden. Door elkaar te observeren en te imiteren alsook door het bekrachtigen van zowel positief als negatief gedrag wordt de relatie tussen siblings onderling gevormd (i.e., sociaal leren; Bandura, 1977; Whiteman, McHale, & Soli, 2011). De beschikbare literatuur ondersteunt de stelling dat sibling interacties tussen kinderen met ASS en HR-sibs verschillen van die

bij kinderen zonder ASS (bv. typisch ontwikkelende kinderen, kinderen met Down syndroom). Zo tonen kinderen met ASS minder sociale toenaderingen naar hun sibling en zijn ze minder responsief, terwijl typisch ontwikkelende HR-sibs minder betrokken en vaker vermijdend zijn gedurende de sibling interactie (Knott, Lewis, & Williams, 1995; Walton & Ingersoll, 2015). Daarnaast is er bij HR-sibs minder sprake van conflict in vergelijking met LR-sibs (Kaminsky & Dewey, 2001). Hoewel meer onderzoek nodig is om dit te bevestigen, is het mogelijk dat deze atypische sibling interacties de ontwikkeling van HR-sibs beïnvloeden.

Tot slot is de **interactie met leeftijdsgenoten** een derde belangrijke context waarbinnen kinderen sociale ervaringen opdoen. Tot op heden zijn er geen studies terug te vinden omtrent de interactie tussen HR-sibs en hun leeftijdsgenoten. Studies bij kinderen met ASS tonen wel aan dat deze kinderen vaker moeilijkheden vertonen en minder aansluiting vinden in interactie met een leeftijdsgenootje (Boyd, Conroy, Asmus, & McKenney, 2011; Kasari, Locke, Gulsrud, & Rotheram-Fuller, 2011). Ook hier is het mogelijk dat de aanwezigheid van sociaal-communicatieve tekorten bij HR-sibs hun interactie met leeftijdsgenoten tekent. Aanvullend stellen Roskam, Meunier, en Stievenart (2015) dat sibling interacties een langdurend effect hebben op latere interacties met leeftijdsgenoten. Zo is het mogelijk dat interactiepatronen, aangeleerd tijdens de sibling interactie, overgedragen worden naar de interactie met leeftijdsgenoten (Howe, Ross, & Recchia, 2011; Lockwood, Kitzmann, & Cohen, 2001). Daarnaast is het eveneens mogelijk dat kinderen die geconfronteerd worden met veel negatief gedrag of andere tekorten tijdens de sibling interactie dit zullen proberen te compenseren tijdens andere sociale interacties (Howe et al., 2011; Mendelson, Aboud, & Lanthier, 1994; Stocker, 1994).

Concluderend kan worden gesteld dat, binnen de typische ontwikkeling, zowel de ouder-kind interactie, sibling interactie als interactie met leeftijdsgenootjes een belangrijke impact hebben op de sociaal-communicatieve, taal-, emotionele, en cognitieve ontwikkeling van kinderen (bv. Cheah, Nelson, & Rubin, 2001; Denham et al., 2011; Feinberg, Solmeyer, & McHale, 2012; Feldman, 2010; Feldman, Bamberger, & Kanat-Maymon, 2013; Howe & Recchia, 2014; Russel, 2011). Indien deze drie sociale contexten anders verlopen voor HR-sibs, kan dit belangrijke implicaties hebben voor hun vroege leeromgeving en ontwikkeling alsook op de uitdrukking van het ASS-fenotype.

Het is daarom ook belangrijk om het verband tussen vroege sociale interacties en de ontwikkeling van HR-sibs na te gaan.

### **DOELSTELLINGEN VAN HET DOCTORAATSONDERZOEK**

De belangrijkste doelstelling van dit doctoraat was het in kaart brengen van de vroege sociale interacties van jongere siblings van kinderen met ASS (HR-sibs). Verschillende studies hebben de vroege ontwikkelingstrajecten van deze HR-sibs uitvoerig bestudeerd (bv. Hudry et al., 2014; Toth, Dawson, Meltzoff, Greenson, & Fein, 2007; Zwaigenbaum et al., 2005), maar er werd hierbij zelden rekening gehouden met de rol van de sociale omgeving. Ten eerste werden binnen dit doctoraat de vroege ouder-kind interacties van HR-sibs geëvalueerd en vergeleken met de ouder-kind interacties van kinderen met een typisch ontwikkelende oudere broer of zus (i.e., laag-risico siblings; LR-sibs). Gezien de inconsistente resultaten in de literatuur met betrekking tot het eerste levensjaar, werd deze ouder-kind interactie bestudeerd op de leeftijd van 5 en 10 maanden. Ten tweede werden de vroege sibling interacties van HR-sibs in kaart gebracht. Het onderzoek rond dit onderwerp is immers beperkt en omvat ook enkele beperkingen. Zo bestond bij voorgaand onderzoek de steekproef vooral uit schoolgaande kinderen, was er sprake van zowel jongere als oudere siblings van kinderen met ASS, focuste men vooral op typisch ontwikkelende HR-sibs (en niet op deze met ASS of andere tekorten), en maakte men bijna uitsluitend gebruik van vragenlijsten. Gezien er bij kinderen met ASS en HR-sibs al sociaal-communicatieve tekorten zichtbaar zijn tijdens de eerste levensjaren (Carpenter, Nagell, Tomasello, Butterworth, & Moore, 1998; Szatmari et al., 2016), is het belangrijk de sibling interactie reeds op jongere leeftijd in kaart te brengen. Vragenlijsten kunnen zeer relevante informatie bieden, maar worden onder meer beïnvloed door *response bias* (i.e., de vaststelling dat het gegeven antwoord niet accuraat is door bv. sociale wenselijkheid of inaccurate herinneringen; Stone, Hoffman, Lewis, & Ousley, 1994). Daarom werd binnen dit doctoraatsonderzoek de keuze gemaakt om de sibling interacties te observeren in een natuurlijke setting (i.e., thuis) en dit reeds op verschillende momenten gedurende de eerste levensjaren (op 18, 24, 36 maanden). Gezien eerdere sociale interacties (bv.

met siblings) een invloed kunnen hebben op latere sociale interacties (bv. met leeftijdsgenoten), werd tot slot ook de interactie tussen HR-sibs en hun leeftijdsgenootjes geëvalueerd op de leeftijd van 36 maanden.

Om na te gaan in welke mate vroege sociale interacties de ontwikkeling van HR-sibs kunnen beïnvloeden, werd het verband in kaart gebracht tussen vroege ouder-kind en sibling interacties enerzijds en de ontwikkeling van HR-sibs op 24 en 36 maanden anderzijds. Dit zou immers empirische ondersteuning kunnen bieden voor de vooropgestelde samenhang tussen de vroege sociale omgeving en de ontwikkelingstrajecten van HR-sibs (Dawson, 2008; Inguaggiato et al., 2017; Mandy & Lai, 2016).

## OVERZICHT EN BESPREKING VAN DE BELANGRIJKSTE ONDERZOEKSRESULTATEN

### Ouder-kind interactie

De ouder-kind interactie werd onderzocht aan de hand van een *between-family* studie (i.e., de vergelijking van de ouder-kind interactie tussen HR- en LR-sibs *tussen* gezinnen) alsook met een *within-family* studie (i.e., de vergelijking tussen de ouder-kind interacties van HR-sibs en deze van hun broer/zus met ASS *binnen* hetzelfde gezin). Op de leeftijd van 5 maanden (*between-family*) waren er geen verschillen op te merken tussen de ouder-kind interactie van HR- en LR-sibs, noch in het gedrag van het kind, noch in het gedrag van de ouder. Op de leeftijd van 10 maanden waren er eveneens geen verschillen in het sociaal-communicatief gedrag van HR- en LR-sibs. Dit ondersteunt de heersende consensus dat vroege kenmerken van ASS nog niet zichtbaar zijn gedurende het eerste levensjaar en pas toenemen na de leeftijd van 12 maanden (bv. Szatmari et al., 2016). Indien ASS-kenmerken duidelijker worden gedurende het tweede levensjaar, zullen HR- en LR-sibs ook in toenemende mate van elkaar verschillen. Vervolgens is het mogelijk dat ouders hun interactiestijl steeds meer zullen aanpassen aan (de tekorten van) hun kind, zoals geobserveerd bij kinderen met ASS. Bijgevolg zullen ook potentiële verschillen in de ouder-kind interactie tussen HR- en LR-sibs pas duidelijk worden vanaf de leeftijd van 12 maanden. Deze stelling wordt eveneens bevestigd door studies die de ouder-kind interactie bij HR-sibs evalueerden gedurende

het tweede levensjaar (Rozga et al., 2011; Wan et al., 2013). Er werd echter wel vastgesteld dat zowel ouders in de HR groep als HR-sibs minder positief affect vertoonden dan ouders en hun kind in de LR groep, wat suggereert dat vroege verschillen tussen HR- en LR-sibs op vlak van temperament wel reeds aanwezig kunnen zijn voor de leeftijd van 12 maanden. Er was een sterk verband tussen het affect van ouders en hun kinderen, wat betekent dat minder positief affect bij HR-sibs aanleiding kan geven tot minder positief affect bij hun ouders (en vice versa). Vroege verschillen in positief affect gedurende het eerste levensjaar werden eveneens gevonden in andere studies bij HR-sibs (bv. Clifford, Hudry, Elsabbagh, Charman, & Johnson, 2013; Zwaigenbaum et al., 2005).

We kunnen hieruit voorzichtig besluiten dat de ouder-kind interactie bij HR-sibs sterk lijkt op die van LR-sibs op de leeftijd van 5 en 10 maanden. Onze hypothese is dat ouders hun interactiestijl, aangeleerd in interactie met hun kind met ASS, niet zomaar veralgemenen naar hun andere kinderen, maar dat ze hun interactiestijl aanpassen aan de kenmerken van het specifieke kind. De idee van een aangepaste interactiestijl in functie van de kindkenmerken wordt verder ondersteund door de resultaten uit de *within-family* studie. In deze studie wezen de resultaten op een meer structurerende, ondersteunende en responsieve interactiestijl bij typisch ontwikkelende HR-sibs in vergelijking met hun broer/zus met ASS. Deze interactiestijl zou kunnen overeenkomen met de interactiestijl van ouders van typisch ontwikkelende kinderen.

### **Sibling interactie**

Als tweede luik binnen dit doctoraatsonderzoek werd de sibling interactie van HR-sibs in kaart gebracht op de leeftijd van 18, 24 en 36 maanden. Hoewel in zowel de laag-risico (LR) als hoog-risico (HR) groep de hoeveelheid effectieve interactie tussen beide kinderen beperkt bleef, waren er toch significante verschillen tussen beide groepen. Terwijl op de leeftijd van 18 maanden de sibling interacties in de HR groep vaker *negatief* waren (bv. iets afnemen, protesteren), werden deze op 24 en 36 maanden gekenmerkt door minder *positieve* gedragingen (bv. iets vragen, iets voortonen). Vervolgens werd de verhouding tussen het jongste en oudste kind binnen elke groep (i.e., rolpatronen/rol(a)symmetrie) nagegaan. Op de leeftijd van 18 en 24 maanden was er duidelijk sprake van rolasymmetrie in beide groepen. Dit betekent dat het oudste kind

een meer leidende, dominante positie (i.e., de interactie zelf initiëren) innam terwijl het jongste kind eerder volgde (i.e., de interactie minder vaak zelf initiëren, maar reageren op initiaties van de interactiepartner). In de HR groep, maar niet in de LR groep, veranderde dit asymmetrische patroon naar een meer symmetrisch patroon op 36 maanden. Er was niet langer sprake van een duidelijke leider en het kind met ASS (oudste kind) was zelfs iets meer volgend. Dit is in overeenstemming met studies bij oudere kinderen waar men vaststelde dat HR-sibs de dominante positie overnamen en kinderen met ASS eerder volgend waren (Knott et al., 1995; Smith, 2010).

Om inzicht te krijgen in hoe sibling interacties zich ontwikkelen doorheen de tijd, werd de evolutie over de drie meetmomenten heen meer in detail bestudeerd. Hieruit konden we afleiden dat ook de evolutie van de interacties doorheen de tijd anders verloopt bij de HR groep in vergelijking met de LR groep. De resultaten in de LR groep tonen aan dat de negatieve interacties stabiel bleven of toenamen van 18 naar 24 maanden en toenamen van 24 naar 36 maanden (i.e., lineair verband). In de HR groep daarentegen, kwamen negatieve gedragingen vaker voor op 18 maanden met een daling in negatief gedrag van 18 naar 24 maanden. Vervolgens bleef de frequentie negatief gedrag stabiel of was er een toename van 24 naar 36 maanden (i.e., kwadratisch verband). Aanvullend was het opvallend dat kinderen met ASS de interactie vaker op een positieve manier initieerden wanneer hun jongere broer/zus 18 maanden oud was dan wanneer deze ouder was. Dit in tegenstelling tot typisch ontwikkelende oudere kinderen die het meeste aantal positieve toenaderingen lieten zien op het laatste meetmoment, wanneer hun jongere sibling 3 jaar oud was. Deze verschillende evoluties doorheen de tijd kunnen mee verklaren waarom de LR en HR groep op 18 maanden verschilden op vlak van negatieve gedragingen terwijl er op 24-36 maanden verschillen geobserveerd werden in positieve gedragingen. Met betrekking tot de rol(a)symmetrie kunnen we besluiten dat de verhouding tussen het jongste en het oudste kind in de LR groep vrij stabiel bleef doorheen de tijd, met een duidelijke asymmetrie op de drie meetmomenten. In de HR groep daarentegen, was er aanvankelijk een groot verschil in het aantal toenaderingen tussen beide kinderen (in het voordeel van het kind met ASS), maar dit verschil werd gradueel kleiner naarmate HR-sibs ouder werden. Bijgevolg was er ook een verschuiving van rolasymmetrie naar rolsymmetrie. Het is mogelijk dat, naarmate HR-sibs ouder en meer sociaal vaardig worden, de rolpatronen verder zullen



verschuiven naar een nog duidelijkere rolasymmetrie met de HR-sibs in de leiderspositie.

Uit deze resultaten kunnen we hoofdzakelijk twee dingen afleiden. Ten eerste worden HR-sibs hun sibling interacties gekenmerkt door meer negatieve (18 maanden) of minder positieve (24-36 maanden) interactiepatronen. Ten tweede kunnen we vaststellen dat de rolasymmetrie die typisch ontwikkelende kinderen kenmerkt niet teruggevonden werd in de HR groep op de leeftijd van 36 maanden. Het is echter belangrijk op te merken dat zowel de kenmerken van de sibling interactie als deze rolasymmetrie, waarbij het oudste kind de interactie leidt en hierbij een voorbeeld stelt voor het jongste kind, een belangrijk onderdeel uitmaken van de vroege leeromgeving van kinderen. Wijzigingen hierin kunnen bijgevolg belangrijke implicaties hebben voor de ontwikkeling van deze HR-sibs.

### **Interactie met leeftijdsgenoten**

Naast de ouder-kind en sibling interactie werd in Hoofdstuk 6 dieper ingegaan op de interactie met leeftijdsgenoten. Als eerste kwam naar voren dat HR-sibs vaker negatief reageerden in interactie met hun broer/zus met ASS dan in interactie met een leeftijdsgenootje. Hoewel dit verder geëvalueerd moet worden in vergelijking met een typisch ontwikkelende controlegroep, suggereert dit dat sibling interacties van HR-sibs negatiever zijn dan hun interacties met leeftijdsgenoten. Ten tweede kon uit de resultaten afgeleid worden dat negatieve interacties gedurende de sibling interactie samenhangen met meer negatieve gedragingen tijdens de interactie met leeftijdsgenoten. Dit laatste zou kunnen betekenen dat HR-sibs de negatieve interactiestijl, aangeleerd in interactie met hun broer/zus met ASS, generaliseren naar de interactie met andere interactiepartners zoals leeftijdsgenoten. Deze stelling wordt ondersteund door onderzoek dat stelt dat sibling interacties aan de basis kunnen liggen van latere interacties met leeftijdsgenoten (bv. Yucel & Downey, 2015).

### **Invloed op ontwikkeling**

Bovenstaande resultaten wijzen op belangrijke verschillen in de vroege sociale interacties van HR-sibs, vooral op vlak van hun sibling interacties. Gezien het belang van

deze sociale ervaringen voor de ontwikkeling van kinderen (bv. Cheah, Nelson, & Rubin, 2001; Denham et al., 2011; Feinberg, Solmeyer, & McHale, 2012; Feldman, 2010; Feldman, Bamberger, & Kanat-Maymon, 2013; Howe & Recchia, 2014; Russel, 2011), is het aannemelijk te veronderstellen dat deze verschillen in de vroege leeromgeving van HR-sibs ook hun ontwikkeling zullen sturen. Zo is het mogelijk dat de vroege sociale omgeving inwerkt op de genetische kwetsbaarheid van HR-sibs, wat bijgevolg de ontwikkeling en manifestatie van ASS-kenmerken bij HR-sibs zou kunnen beïnvloeden (Mandy & Lai, 2016). Dit werd echter nauwelijks onderzocht in de wetenschappelijke literatuur.

Inzake de *ouder-kind interactie in het eerste levensjaar*, werd er in het huidige doctoraatsonderzoek slechts in beperkte mate ondersteuning gevonden voor de associatie tussen de ouder-kind interactie en de ontwikkeling van HR-sibs. Enerzijds is het mogelijk dat deze associatie, net zoals de groepsverschillen, duidelijker zal worden naarmate HR-sibs ouder worden en mogelijke ASS-kenmerken of tekorten in de ontwikkeling zichtbaar worden. Anderzijds had deze studie een aantal beperkingen en is uitgebreider onderzoek met grotere steekproeven nodig om te bevestigen of associaties al dan niet aanwezig zijn.

Het verband tussen de *sibling interactie* en de ontwikkeling van HR-sibs (bv. taal) werd eveneens niet bevestigd door de resultaten van dit doctoraatsonderzoek. Er werd echter wel een belangrijke samenhang gevonden tussen de sibling interactie en ASS-kenmerken bij HR-sibs. Deze bevinding ondersteunt de mogelijke link tussen de vroege sociale omgeving en het ASS-fenotype bij HR-sibs. Op de leeftijd van 24 maanden was meer sociaal gedrag van HR-sibs gedurende de sibling interactie (zowel positief als negatief) gerelateerd aan meer ASS-kenmerken bij HR-sibs. Op 36 maanden daarentegen, werd een negatief verband gevonden tussen ASS-kenmerken van beide kinderen (HR-sibs en kinderen met ASS) en de hoeveelheid sociaal gedrag tijdens de sibling interactie. Deze verschillende resultaten op 24 en 36 maanden kunnen mogelijk verklaard worden doordat het verband tussen kenmerken van de sibling interactie en ASS-kenmerken bij HR-sibs bidirectioneel is. Dit betekent dat er zowel een invloed is van ASS-kenmerken op de sibling interactie als van de sibling interactie op ASS-kenmerken (bv. via sociaal leren). Hoe en in welke mate sibling interacties het ASS-fenotype bij HR-sibs beïnvloeden, is ook mede afhankelijk van de ontwikkelingsfase waarin deze

kinderen zich bevinden (Inguaggiato et al., 2017). Het feit dat de diagnose van ASS doorgaans niet wordt gesteld voor de leeftijd van drie jaar (Sheldrick et al., 2017), betekent waarschijnlijk dat ASS-kenmerken nog niet volledig ontwikkeld zijn voor die leeftijd. Bijgevolg is de kans groter dat, op de leeftijd van 24 maanden, de sibling interactie het ASS-fenotype meer beïnvloedt dan andersom. Via sociaal leren (bv. imitatie) kunnen HR-sibs bepaalde ASS-specifieke gedragingen overnemen van hun broer/zus met ASS. Op de leeftijd van 36 maanden, wanneer ASS-kenmerken stabiel zijn en mogelijk minder beïnvloedbaar door de sociale omgeving, is het dan weer waarschijnlijker dat ASS-kenmerken de sibling interactie beïnvloeden.

## **IMPLICATIES VAN DE ONDERZOEKSBEVINDINGEN**

### **Theoretische implicatie**

Het huidige doctoraatsonderzoek toont aan dat de vroege sociale omgeving van HR-sibs belangrijke verschillen vertoont ten opzichte van typisch ontwikkelende kinderen, vooral op het vlak van vroege sibling interacties. Verder biedt het ook enige ondersteuning voor de associatie tussen de vroege sociale omgeving en de ontwikkeling van HR-sibs. Dit betekent dat, in combinatie met een aangeboren kwetsbaarheid, vroege sociale ervaringen een invloed zouden kunnen hebben op de ontwikkeling van HR-sibs, inclusief het ASS-fenotype (Dawson, 2008; Inguaggiato et al., 2017; Mandy & Lai, 2016). De studies binnen dit doctoraat zijn tot op heden de enige die de sibling interacties van HR-sibs hebben geëvalueerd gedurende de eerste levensjaren. Aanvullend waren deze studies de eerste die bij HR-sibs de associatie hebben onderzocht tussen de vroege sociale interacties en zowel ASS-kenmerken als andere ontwikkelingsdomeinen (bv. taalontwikkeling). Hoewel het doctoraatsonderzoek ook enkele beperkingen kent, werden er significante resultaten gevonden. Het is bijgevolg aangewezen om de vroege sociale omgeving verder te onderzoeken, vooral in verband met de atypische ontwikkelingstrajecten van HR-sibs. Longitudinale opvolgstudies met grotere steekproeven, uitgebreidere meetinstrumenten en meer geavanceerde analyses kunnen hieromtrent meer duidelijkheid bieden. Gezien de resultaten suggereren dat de interactiepatronen uit de sibling interactie tenminste gedeeltelijk overgedragen worden

naar de interactie met leeftijdsgenoten, is het ook aangewezen om de interactie van HR-sibs en hun leeftijdsgenoten verder te onderzoeken, inclusief de vergelijking met een typisch ontwikkelende controlegroep.

### **Methodologische implicaties**

De bevindingen uit Hoofdstuk 6 suggereren dat het belangrijk is om rekening te houden met wie de vragenlijsten over ASS-kenmerken bij kinderen invult. Wanneer de vragenlijsten van ouders en leerkrachten werden vergeleken, bleek dat de overlap tussen beiden eerder laag was. Dit komt overeen met wat gerapporteerd wordt in de handleiding van de *Social Responsiveness Scale* (SRS-2; Constantino & Gruber, 2012), één van de gebruikte vragenlijsten. Het is mogelijk dat kinderen zich anders (moeten) gedragen op school dan thuis. Het is eveneens mogelijk dat ASS-kenmerken op school anders tot uiting komen dan thuis. Bijgevolg zijn leerkrachten wellicht beter geschikt om te rapporteren over de uiting van ASS-kenmerken op school terwijl ouders beter geschikt zijn om te rapporteren over ASS in de thuiscontext. Verder hebben ouders en leerkrachten een verschillend referentiekader. Ouders van kinderen met ASS hebben in vele gevallen enkel ervaring met een kind met ASS, waardoor ze mogelijk moeilijker kunnen inschatten wat de verwachtingen zijn voor een typisch ontwikkelend kind. Leerkrachten, daarentegen, hebben dagelijks te maken met typisch ontwikkelende kinderen. Om een volledig en accuraat beeld te schetsen van de ASS-kenmerken van kinderen, is het nodig de informatie van zowel de ouder als de leerkracht te integreren (Constantino & Gruber, 2012).

Hoe sociale interacties omgezet worden in ruwe data (bv. frequenties), is afhankelijk van de codeerprocedure die gehanteerd wordt. Specifiek met betrekking tot het in kaart brengen van sociale interacties worden twee codeerprocedures vaak gebruikt: globale codeerschalen en een gedetailleerd, frequentie codeerschema. Zoals besproken in Hoofdstuk 2 hebben beide methodes voordelen. Indien de beschikbare middelen dit toelaten, kan een combinatie van globale en frequentie coderingen een grondig beeld schetsen van de sociale interactie die wordt bestudeerd. Gezien dit niet altijd mogelijk is en er vaak een keuze moet worden gemaakt tussen beide, is het belangrijk met de voor- en nadelen van elk codeerschema rekening te houden. Enerzijds zijn globale codeerschalen uitermate geschikt om de *kwaliteit* van een interactie te beoordelen

alsook om informatie van meerdere bronnen (bv. verschillende personen) te integreren (Bakeman & Quera, 2011; Grotevant & Carlson, 1989). Dus, indien de onderzoeker vooral interesse heeft in een brede, kwalitatieve omschrijving van de sociale interactie, dan zijn globale codeerscalen beter geschikt. Dit laat de onderzoeker immers toe om die gedragingen van beide interactiepartners te evalueren waarvan studies aangetoond hebben dat ze belangrijk zijn voor de ontwikkeling van het kind. Omwille van deze reden werden binnen het huidige doctoraat globale codeerscalen gebruikt om de ouder-kind interactie op een tijdbesparende wijze in kaart te brengen. Globale codeerscalen hebben echter als nadeel dat, gezien de interactie overkoepelend beoordeeld wordt, informatie over de specifieke dynamieken verloren gaat. Anderzijds is een gedetailleerd, frequentie codeerschema beter geschikt om de *kwantiteit* (bv. aantal/duur van een bepaald gedrag) en dynamieken van een sociale interactie in kaart te brengen. Gezien we voor de sibling interactie interesse hadden in de specifieke interactiepatronen die deel uitmaken van de vroege leeromgeving, ging de keuze uit naar een gedetailleerd, frequentie codeerschema. Deze methode biedt een zeer gedetailleerd beeld van de sociale interactie, maar heeft als nadeel dat het proces zeer tijdrovend is. Bijgevolg is het niet haalbaar deze methode toe te passen op een verscheidenheid aan gedragingen.

Om de sibling interactie te observeren, werd gebruik gemaakt van drie soorten speelgoed (i.e., ballenbaan, Duplo™/Playmobil™, keyboard) opdat verschillende spelniveaus aan bod zouden kunnen komen. In de huidige studies waren de resultaten gelijklopend in de drie contexten waardoor de verschillende contexten afzonderlijk niet in detail werden besproken. We stelden echter wel vast dat elk speelgoed ander gedrag uitlokte. Terwijl Duplo™/Playmobil™ te veel ruimte liet voor solitair spel, wat zich uitte in weinig rechtstreekse interactie tussen beide kinderen, bood het keyboard niet genoeg mogelijkheden om samen te spelen, wat zich doorgaans uitte in korte episodes van conflict gevolgd door opnieuw solitair spel. De ballenbaan leidde tot een goed evenwicht tussen solitair en gezamenlijk spel en binnen deze context werden de meeste interactieve gedragingen geobserveerd, zowel positief als negatief. Dus, om sibling interacties te observeren kiezen onderzoekers best voor een type speelgoed dat mogelijkheden biedt voor zowel solitair als gezamenlijk spel, zoals bijvoorbeeld de ballenbaan.

Tot slot is het belangrijk rekening te houden met het moment waarop de sociale interactie wordt geobserveerd en gemeten. Zowel de aard van de ouder-kind als van de sibling interactie veranderde doorheen de tijd en ook de associatie met de ontwikkeling van het kind varieerde. Gezien kenmerken van ASS nog niet verankerd lijken te zijn gedurende de eerste twee levensjaren, kan het waardevol zijn om de invloed van de sociale omgeving vooral dan te onderzoeken. Eens het ASS-fenotype stabiel is, is het mogelijk moeilijker om veranderingen in ASS-kenmerken teweeg te brengen.

### **Klinische implicaties**

Onderzoek toont aan dat positieve sibling interacties belangrijk zijn voor broers en zussen van kinderen met ASS (Tomeny, Ellis, Rankin, & Barry, 2017). Daarenboven kunnen positieve sibling interacties de ontwikkeling van beide kinderen positief beïnvloeden (bv. Harrist et al., 2014). Onze resultaten en deze van andere studies bij oudere kinderen (zie Beyer, 2009) wijzen er echter op dat sibling interacties bij kinderen met ASS vaak als minder positief worden bestempeld. Daarnaast kwamen negatieve gedragingen in de HR groep op de leeftijd van 36 maanden dubbel zo vaak voor als positieve gedragingen. Zowel een verminderd aantal positieve interacties als een overwicht aan negatieve interacties kunnen een invloed uitoefenen op het functioneren van het kind en het gezin. Gezien onderzoek aantoont dat positieve sibling interacties de ontwikkeling van kinderen positief kunnen beïnvloeden (Brody, 2004; Buist & Vermande, 2014; Harrist et al., 2014), zou het bevorderen van positieve sibling interacties tussen kinderen met ASS en HR-sibs de ontwikkeling en het welzijn van beide kinderen ten goede kunnen komen. Het is bijvoorbeeld mogelijk positieve interacties te verhogen aan de hand van een interventie die spelvaardigheden aanleert aan beide kinderen gedurende een gezamenlijke activiteit (Beyer, 2009). Ook ouderlijk toezicht gedurende de sibling interacties kan positief gedrag bevorderen. Zo vonden Mchale, Updegraff, Tucker, en Crouter (2000) een positief verband tussen de tijd die siblings doorbrachten in gezelschap van hun ouders en de kwaliteit van hun sibling interacties.

Er is te weinig evidentie om te stellen dat de evaluatie van sociale interacties kan bijdragen tot de diagnostiek of vaststelling van ASS. Het kan echter wel bepaalde sterktes of zwaktes in de gezinscontext in kaart brengen, wat op zijn beurt aangrijpingspunten kan bieden voor interventies. Indien ouders aangeven dat de sibling

interacties ofwel zeer negatief verlopen ofwel te weinig optreden, dan zouden bepaalde strategieën (bv. trainen van spelvaardigheden) geïntegreerd kunnen worden in de interventie van bijvoorbeeld thuisbegeleidingsdiensten. Zo werd bijvoorbeeld aangetoond dat jonge, typisch ontwikkelende siblings van kinderen met ASS vaardigheden kunnen leren (bv. aansluiting vinden bij het spel van het kind met ASS) die de sociale interactie met het kind met ASS bevorderen (Oppenheim-Leaf, Leaf, Dozier, Sheldon, & Sherman, 2012).

Hoewel de huidige studies zich doorgaans richtten op één sociale context, moeten zowel de ouder-kind interactie, sibling interactie en interactie met leeftijdsgenoten gezien worden binnen de ruimere context. De associatie tussen bijvoorbeeld de sibling interacties en de ontwikkeling van HR-sibs was eerder beperkt, wat erop kan wijzen dat louter de sibling interactie niet zo een grote impact heeft op de ontwikkeling als aanvankelijk gedacht. In plaats daarvan zou de sociale interacties met andere interactiepartners de ontwikkeling van HR-sibs eveneens kunnen beïnvloeden (en eventueel compenseren voor de verlaagde sociale input tijdens de sibling interactie). Zo vonden we bijvoorbeeld een positieve associatie tussen het aantal dagen dat HR-sibs naar de kinderopvang gingen op 18 maanden en het aantal positieve initiaties van HR-sibs op 18 maanden. Het is aannemelijk dat de combinatie van verschillende sociale contexten, eerder dan 1 sociale context op zichzelf, de ontwikkeling van HR-sibs beïnvloedt. Zo kan de interactie met typisch ontwikkelende kinderen in de kinderopvang HR-sibs eventueel beschermen tegen mogelijke nadelige effecten die ze zouden ondervinden van de sibling interactie.

Zoals aangetoond in Hoofdstuk 6, is het mogelijk dat negatieve interactiepatronen, aangeleerd tijdens de sibling interactie, (deels) worden gegeneraliseerd naar de interactie met leeftijdsgenoten. Hoewel we op dit moment niet kunnen nagaan of de interactie tussen HR-sibs en een leeftijdsgenootje verschilt van die van een typisch ontwikkelende controlegroep, zouden negatieve sibling interacties kunnen leiden tot meer negatieve interacties met leeftijdsgenoten (Lockwood et al., 2001), wat op zijn beurt een invloed heeft op de leeromgeving van HR-sibs. Indien de interactie met leeftijdsgenootjes effectiever negatiever is bij HR-sibs, zou het belangrijk kunnen zijn leerkrachten hierop te wijzen. Verhoogd toezicht en de gepaste ondersteuning van de leerkracht zou de generalisatie van negatief gedrag misschien kunnen beperken.

## CONCLUSIE

Met dit doctoraatsonderzoek werd een gedetailleerde omschrijving gegeven van de vroege sociale omgeving van jongere broers en zussen van kinderen met ASS gedurende de eerste levensjaren. Hoewel er een aantal gelijkenissen waren tussen de sociale omgeving van deze HR-sibs en een laag-risico controlegroep, werden eveneens belangrijke verschillen gevonden, vooral met betrekking tot de sibling interacties. Bovendien vonden we enige ondersteuning voor het verband tussen deze vroege sociale omgeving en kenmerken van het ASS-fenotype bij HR-sibs. De gevonden associaties tussen de sociale omgeving en ontwikkeling van HR-sibs waren echter beperkt en verder onderzoek is nodig om deze resultaten te ondersteunen. Als het verband tussen de vroege sociale omgeving en de latere ontwikkeling inderdaad verder wordt bevestigd in longitudinale studies met grotere steekproeven, dan benadrukt dit het belang van de sociale omgeving bij de atypische ontwikkelingstrajecten alsook de kenmerken van het ASS-fenotype bij HR-sibs.



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## SUMMARY

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Autism spectrum disorder is a neurodevelopmental disorder characterised by persistent deficits in social communication and social interaction across multiple contexts (American Psychiatric Association [APA], 2013). The prevalence of ASD is estimated around 60-70 per 10,000 children (Elsabbagh et al., 2012). In addition, with a recurrence rate of 18.7% the prevalence of ASD is significantly higher in younger biological siblings of children with ASD (hereafter high-risk siblings; HR-sibs; Ozonoff et al., 2011; Wheelwright, Auyeung, Allison, & Baron-Cohen, 2010). Of the HR-sibs who do not go on to meet the criteria for ASD, there is a substantial proportion that shows mild but qualitatively similar characteristics of ASD, also referred to as the 'broader autism phenotype (BAP)' (Bailey, Palferman, Heavey, & Le Couteur, 1998; Georgiades et al., 2013; Sucksmith, Roth, & Hoekstra, 2011). The aetiology of ASD is complex and both environmental and genetic influences need to be considered in the emergence (onset) and development (course) of the ASD phenotype in children with ASD and HR-sibs (Mandy & Lai, 2016). Because genetic factors cannot explain the full ASD phenotype, environmental factors and the gene-environment interaction should be considered as well, especially during critical periods of development when brain plasticity is high (Dennis et al., 2013; Elsabbagh & Johnson, 2010; Inguaggiato, Sgandurra, & Cioni, 2017). Different combinations of genetic and environmental factors can result in different ASD phenotypes (Elsabbagh & Johnson, 2010). Several potential environmental risk (e.g., hypoxia, maternal/paternal age) and protective (e.g., prenatal folate) factors have been identified, including the early social environment (Inguaggiato et al., 2017; Mandy & Lai, 2016). Therefore, the main objective of this dissertation was to gain more insight into the early social experiences (i.e., parent-child, sibling and peer interactions) of younger siblings of children with ASD (HR group) compared to younger siblings of typically developing children (low-risk (LR) siblings; LR group).

First, research on the **parent-child interaction** including HR-sibs during the first years of life suggests meaningful differences from parent-child interactions in typical development (Wan et al., 2012, 2013; Yirmiya et al., 2006). However, these differences are not consistently found (Rozga et al., 2011), especially before the age of 12 months. Therefore, we aimed to investigate early parent-child interactions including HR-sibs at

the ages of 5 and 10 months. At 5 months, possible ASD characteristics and differences between HR- and LR-sibs are not yet clear and parents from the HR group interact with their child in the same way as do parents from the LR group. At 10 months, differences are noticeable in terms of positive affect, suggesting that differences start to emerge between HR- and LR-sibs. Previous studies studying the parent-child interaction in HR-sibs during the first year of life show that the emergence of early ASD characteristics mainly occurs during the second year of life (Rozga et al., 2011; Szatmari et al., 2016; Wan et al., 2012; Yirmiya et al., 2006). As differences between HR- and LR-sibs increase, it is possible that parents in the HR group will adapt or change their interaction style and that similarities between the HR and LR group will gradually decrease. Although follow-up of the current sample beyond the first year of life is needed, this hypothesis is supported by the findings of Rozga et al. (2011) and Wan et al. (2013) studying the parent-child interaction during the second year of life. Thus, based on our results we can conclude that the parent-child interaction of HR-sibs strongly resembles the parent-child interaction of LR-sibs at the ages of 5 and 10 months. We hypothesise that, instead of just generalising the interaction style used with their child with ASD to other siblings, parents adapt their behaviours to the specific child characteristics. A differentiation in interaction style based on child characteristics was also found in the within-family study. In interaction with their typically developing child, parents provided more structure, emotional support and were more responsive, which may resemble the interaction style of parents without a child with ASD. In interaction with their child with ASD, parents adapted their behaviours to the abilities of their child (e.g., nonverbal mental age and word comprehension).

Regarding the association with later development, we only found limited support for the link between early parent-child interactions and HR-sibs' developmental trajectories. However, this was only the first study attempting to evaluate the association with development and we were limited in terms of the power of our study as well as the type of analyses being used. Given that we did find some evidence for the association with later development indicates that further research including a larger sample and more elaborate analyses might reveal more significant associations. Moreover, it is again possible that as ASD characteristics in HR-sibs increase throughout



the first years of life, the predictive value of the parent-child interaction increases as well.

Second, HR-sibs' **sibling interactions** with an older sibling with ASD were explored. Even though the possible social-communicative impairments of children with ASD/HR-sibs are likely to influence the nature of their sibling interactions, studies on sibling interactions including children with ASD are scarce. We aimed to evaluate early sibling interactions of 18-, 24-, and 36-month-old HR-sibs, irrespective of outcome at 36 months, using a naturalistic, observational method. Even though the total amount of mutual interaction between both siblings was low in both groups, the sibling interactions of the LR and HR group significantly differed at all three time points. At 18 months, HR-sibs' sibling interactions were more negative in nature whereas their sibling interactions at 24-36 months were characterised by lower levels of positive behaviours. There were also differences in terms of the role (a)symmetry between both siblings. At all three time points, the LR group showed the asymmetrical pattern we expected based on studies including typically developing children (i.e., the older child as the leader, the younger child in a more following position; Abramovitch, Corter, & Lando, 1979; Lamb, 1978). At 18 and 24 months this asymmetrical pattern was also observed in the HR group. At 36 months, however, this asymmetrical pattern shifted to a more symmetrical pattern. There was no clear leader and the child with ASD was even slightly more following. Next, to better understand the change in group differences and role patterns over time, behaviours were re-evaluated across all three time points. Two clear patterns emerged. First, the change in *negative behaviours* in the LR group seems linear, with low levels of negative behaviour at 18 months and a gradual increase towards 24/36 months. In contrast, the change in negative behaviours in the HR group seems more quadratic, with somewhat higher levels at 18 months, a decrease from 18 to 24 months, and a (small) increase towards 36 months. Second, *positive* initiations of the older children in the LR group are highest at 36 months, whereas positive initiations of the child with ASD are highest at 18 months.

While it remains plausible that these differences in the early learning context will influence HR-sibs' development (e.g., language development), this was not fully substantiated by the current findings. The association between ASD characteristics and the sibling interaction, however, does indicate that sibling interactions could have the

potential to significantly impact on the expression of the ASD phenotype in HR-sibs. At 24 months, the total amount of initiations and responses (i.e., positive and negative combined) was associated with more ASD characteristics in HR-sibs. In contrast, the total amount of initiations and responses during the sibling interaction at 36 months was associated with fewer ASD characteristics. The relationship between social-communicative behaviours during the sibling interaction and ASD characteristics is presumably bidirectional and how sibling interactions influence the expression of ASD characteristics and vice versa may depend on the developmental stage of the child (Inguaggiato et al., 2017). Given that children are usually not diagnosed before the age of three years (Sheldrick, Maye, & Carter, 2017), ASD characteristics are probably not yet fully expressed at the age of 24 months. Perhaps in this stage of development, sibling interactions are more likely to influence the ASD phenotype than vice versa. At 36 months, on the other hand, ASD characteristics might be more stable and less susceptible to environmental influences. Thus at this age, it seems more likely that ASD characteristics (of both children) influence the sibling interaction rather than the other way around.

Finally, at 36 months, the association between HR-sibs' sibling and **peer interactions** was evaluated. The results demonstrate that HR-sibs' behaviours are somewhat more negative in interaction with a sibling compared to a peer. At the same time, HR-sibs generalise a more negative interaction pattern from sibling interactions to peer interactions. This is supported by research suggesting that the pattern of interaction between siblings is carried over to the interaction between these siblings and their peers (Howe, Ross, & Recchia, 2011; Lockwood, Kitzmann, & Cohen, 2001). However, the fact that sibling interactions remain more negative than peer interactions suggests that especially the interaction with a sibling with ASD triggers negative responses in HR-sibs. It is possible that, due to prior experiences, HR-sibs are less tolerant towards their sibling with ASD, or that they have learned to stand up to them more.

This dissertation provided evidence for differences in HR-sibs' early social environment, especially in terms of altered sibling interactions. In addition, there was some support for the association between early social interactions and child development. This provides evidence for the hypothesis that environmental factors including early experiences may influence the development of neural circuits (i.e., brain

plasticity) and shape individuals' physical as well as mental development (Dawson, 2008; Inguaggiato et al., 2017; Mandy & Lai, 2016). Accordingly, Mandy and Lai (2016) also concluded that early social experiences could modify a pre-existing susceptibility to ASD. However, further studies including a longitudinal design, larger samples matched on gender, age and dyad constellation, and more elaborate measures of child development are needed to fully explore the association between the early interactions and subsequent development. Moreover, given that the current results indicated that negative sibling interactions are at least partly generalised to the peer context, HR-sibs' peer interactions need to be further explored and compared with a low-risk control group.

Studies including older children or adolescents/adults show that positive sibling relationships are important for siblings of children with ASD, both in terms of general well-being and developmental outcomes (e.g., Harrist et al., 2014; Tomeny, Ellis, Rankin, & Barry, 2017). However, both our results and previous studies including older children (also see Beyer, 2009) suggest that, in some cases, sibling interactions including a child with ASD are characterised as less positive. Thus, the promotion of positive sibling relationships could benefit the development and well-being of siblings of children with ASD. On a diagnostic level, there is insufficient evidence to conclude that the assessment of sibling interactions (or parent-child or peer interactions) might improve the diagnostic process of ASD. However, these observations might reveal possible strengths or weaknesses in the learning environment of HR-sibs as well as targets for intervention. Finally, the social experiences of HR-sibs outside of the sibling interaction need to be considered as well. The associations between HR-sibs' sibling interactions and development were limited, suggesting that differences in sibling interactions in themselves might not influence the HR-sibs' development as expected. In addition to the interaction with a sibling with ASD, other social experiences might have influenced HR-sibs as well (perhaps compensating for lower levels of input during the sibling interaction). For example, social experiences during day care, whether or not in interaction with social experiences during other social contexts such as the sibling interaction, may also influence the developmental trajectories of HR-sibs. It is possible that the general social context of HR-sibs, including all relevant social experiences, influences their development rather than one context in itself.

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## DATA STORAGE FACT SHEETS

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### DATA STORAGE FACT SHEET CHAPTER 2

Name/identifier study: Chapter 2 - PCI - coding schemes

Author: Chloè Bontinck

Date: 30/10/2017

#### 1. Contact details

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#### 2. Information about the datasets to which this sheet applies

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Bontinck, C. (2017). Parent-child interaction in children with autism spectrum disorder and their siblings: Choosing a coding strategy (Doctoral dissertation). Ghent University, Ghent, Belgium.

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**DATA STORAGE FACT SHEET CHAPTER 3**

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Author: Chloè Bontinck

Date: 30/10/2017

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- responsible ZAP
- all members of the research group
- all members of UGent
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## DATA STORAGE FACT SHEET CHAPTER 4

Name/identifier study: Chapter 4 - Sibling IA 18 months

Author: Chloè Bontinck

Date: 30/10/2017

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\* Reference of the publication in which the datasets are reported:

Bontinck, C. (2017). The early development of infant siblings of children with autism spectrum disorder: Characteristics of sibling interactions (Doctoral dissertation). Ghent University, Ghent, Belgium.

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## DATA STORAGE FACT SHEET CHAPTER 5

Name/identifier study: Chapter 5 - Sibling IA 24 months

Author: Chloè Bontinck

Date: 30/10/2017

### 1. Contact details

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## DATA STORAGE FACT SHEET CHAPTER 6

Name/identifier study: Chapter 6 - Sibling+Peer IA 36 months

Author: Chloè Bontinck

Date: 30/10/2017

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