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Autism risk status and maternal behavior: impacts on infant language and communication development from 6 to 36 months of age

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AUTISM RISK STATUS AND MATERNAL BEHAVIOR: IMPACTS ON INFANT LANGUAGE AND COMMUNICATION DEVELOPMENT FROM 6 TO 36 MONTHS OF AGE

by

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DEDICATION

I would like to dedicate this work to my fantastically patient and supportive spouse George and to Baby Thombott.
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I have been incredibly fortunate to be involved with the Infant Sibling Project during my time here at Boston University. My dissertation work and training would not have been possible without the dedication and support of the project staff, the many students who have helped with my coding projects including Suzanne Schiavone and Chelsea Cohen, the support of Drs. Alice Carter and Charles Nelson, and my advisor, Helen Tager-Flusberg. I would like to thank Helen in particular for all of her guidance, advice, and encouragement over the last few years.

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AUTISM RISK STATUS AND MATERNAL BEHAVIOR: IMPACTS ON INFANT LANGUAGE AND COMMUNICATION DEVELOPMENT FROM 6 TO 36 MONTHS OF AGE

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ABSTRACT

This study explores the language and communication development of infant siblings of children with autism, who are at increased risk for impairments in these domains, over the first year of life (Jones et al., 2014). Additionally, maternal communicative input and background factors unique to this population (e.g. mothers’ concerns about their infants’ development and experience with a previously diagnosed child) were also examined to determine how these parental and family factors interact with infants’ early language and communication development.

These issues were examined in the context of a longitudinal study of high risk infant siblings using data collected from 89 high risk infant siblings and 76 low risk infants, with the sample varying across each of three studies. Group differences in maternal and infant communication, scored from both home-based written and video diaries collected over the first year of life, were analyzed using repeated measures ANOVA and non-parametric analyses; correlations analyses compared these scored behaviors to standardized measures collected in the laboratory. The number of infants
diagnosed with autism ranges from 5 – 19 infants per study; analyses address both this subset of infants and the larger group of high risk siblings as a whole. Results show that a) mothers of high risk infants have consistent and early-appearing concerns about their infants’ development, but these concerns are poorly related to infant symptoms before 9 months, b) delays in language, as evident in consonant production are not readily apparent at 9 months of age, and high and low risk mothers respond equivalently to these early vocalizations, c) at 12 months, high and low risk mothers use similar social-communicative prompting strategies but for high risk mothers these strategies are associated with autism-related concerns about their infant and the symptom severity of the older diagnosed child.

These findings support a transactional account of early dyadic interactions, with infant language delays emerging over the first year of life and parental behavior reflecting both these emerging symptoms and unique background factors. These results are discussed in terms of the larger literature on language and communication in early infancy, as well as implications for intervention practices.
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CHAPTER 1: INTRODUCTION

Language Development in Infant Siblings of Children with Autism

Language and communication impairments are some of the earliest appearing and most pervasive features of autism spectrum disorders (ASD), with core deficits in communication and social interaction emerging towards the end of the first year of life (Baranek, 1999; Osterling, Dawson, & Munson, 2002; Osterling & Dawson, 1994; Ozonoff et al., 2010; Zwaigenbaum et al., 2005). Despite the presence of these early appearing behavioral symptoms, the average age of diagnosis for ASD remains around four years of age (CDC, 2014). Investigations of children’s language and communication development prior to a formal diagnosis initially relied on retrospective methods - interviewing parents about their child’s earlier development or scoring behavior from family home videos (De Giacomo & Fombonne, 1998; Osterling et al., 2002; Osterling & Dawson, 1994; Werner, Dawson, Osterling, & Dinno, 2000). Over the last decade or so, prospective investigations have more systematically studied the early language and communication development of infants later diagnosed with ASD by monitoring the development of infants at increased risk over the first few years of life until a diagnosis can be made. The high risk samples used in these prospective investigations have primarily consisted of infant siblings of children with ASD, who are at increased familial risk for ASD relative to the general population (CDC, 2014; Ozonoff et al., 2011; see Zwaigenbaum et al., 2007 for a discussion of this methodology).

These longitudinal, prospective investigations of high risk infant siblings have provided more detailed descriptions of the early language and communication abilities of
infants later diagnosed with ASD. Importantly, they have revealed that difficulties with language and communication are present not only in infants later diagnosed with ASD, but are also present to some degree in large numbers of non-diagnosed high risk infants (Gamliel, Yirmiya, Jaffe, Manor, & Sigman, 2009; Ozonoff et al., 2014; Rogers, 2009). As a group, high risk infant siblings lag behind low risk infants in the production of late consonants (those that emerge latest developmentally), produce canonical syllables less frequently at 9 months of age, achieve the milestone of reduplicated babbling significantly later and produce fewer gestures in the first and second years of life than low risk, typically developing infants, (Iverson & Wozniak, 2007; Mitchell et al., 2006; Paul, Fuerst, Ramsay, Chawarska, & Klin, 2011). The extent to which these differences characterize the entire sample of high risk infants or are limited to a smaller subset is not fully understood, particularly for differences observed around 12 months of age. By three years of age, there is more substantial support for the existence of specific subgroups of high risk infants. These subgroups consist of infants who meet criteria for ASD on both gold standard behavioral assessments and expert clinical judgment, infants who are judged to be typically developing, and a third group of infants who exhibit difficulties with language or social communication similar to those exhibited by diagnosed infants, but who do not meet full criteria for ASD. The characteristics exhibited by this latter group are thought to represent early-emerging feature of the broader autism phenotype (BAP) - sub-clinical but qualitatively similar features of ASD that have previously been observed in parents and first-degree relatives of children with ASD (Gamliel et al., 2009; Ozonoff et al., 2014; Piven, Palmer, Jacobi, & Childress, 1997; Ruser et al., 2007). One
approach to studying the early emergence of these broader phenotype symptoms has been to classify infants into one of these three subgroups using data collected when infants were 36 months or older and use these groupings to model infants’ earlier language, communication, and cognitive development (Gamliel et al., 2009; Ozonoff et al., 2014; Yirmiya et al., 2006). These analyses have revealed that while infants ultimately diagnosed with ASD demonstrate the most significant impairments beginning at 12 months of age, infants classified into the BAP group also exhibit steady and significant language impairments, but not cognitive impairments, from 12 to 54 months relative to both low risk infants and high risk infants classified as typically developing (Gamliel et al., 2009; Ozonoff et al., 2014). Notably, Ozonoff and colleagues (2014) found that at 36 months of age, even high risk infants classified as typically developing had significantly lower scores on standardized language measures than low risk, typically developing infants. Group differences in language and communication are not present at 6 months of age, suggesting that these difficulties begin to emerge over the first few years of life, beginning around 12 months of age, in both later-diagnosed and non-diagnosed high risk infants (Gamliel et al., 2009; Ozonoff et al., 2010, 2014; Zwaigenbaum et al., 2005).

Together, the literature on high risk infant siblings suggests that language delays are quite variable, with infants later diagnosed with autism exhibiting more significant impairments at earlier ages, but delays or atypicalities present to some degree across much of the high risk group as a whole. Although more distinct subgroups of high risk infants emerge over the second year of life, there is immense heterogeneity and
substantial overlap in the language abilities of high risk infants, particularly at 12 months of age (Gamliel et al., 2009; Ozonoff et al., 2010, 2014; Rogers, 2009).

The presence of language and communication delays in non-diagnosed infant siblings is thought to reflect some aspect of their increased familial risk, but the specific factors that contribute to this risk remain poorly understood. While familial risk is generally considered to primarily reflect genetic risk, there is evidence to suggest that environmental influences, maternal linguistic input in particular, may also play a role. This evidence comes from two lines of research: a) the role maternal input plays in shaping the language development of typically developing infants and b) reports that mothers of high risk infants demonstrate behavioral differences in relevant language and social communication domains. The literature on these two domains is summarized below.

**The Role of Maternal Input in the Language Development of Typically Developing Infants**

A substantial body of research has examined the extent to which environmental input influences the language development of typically developing infants. Much of this research has demonstrated that although the vast majority of infants will develop language given adequate environmental input (levels of which vary greatly by culture and in some cases is quite minimal), variations in the quality and quantity of input, at least for some domains, have measurable effects on children’s language abilities (Hoff, 2006).

The impact of environmental input on children’s language ability is most clearly illustrated in the domain of vocabulary acquisition, where wide variation in the amount of
speech children hear is associated with corresponding variation in children’s vocabulary size (Hart & Risley, 1995; Pan, Rowe, Singer, & Snow, 2005; Rowe, 2012). Some of these effects may be driven partly by children’s own individual abilities. For example, although the amount of overall maternal speech is directly related to children’s later vocabulary size, maternal gestures and speech also contribute to children’s vocabulary indirectly though children’s own gesture use (Goldin-Meadow, Goodrich, Sauer, & Iverson, 2007; Iverson & Goldin-Meadow, 2005; Rowe & Goldin-Meadow, 2009; Rowe, Ozçalişkan, & Goldin-Meadow, 2008). Verbal input is also associated with infants’ phonemic perception and production. Live, contingent interactions also help infants to re-learn non-native phonemic contrasts they have lost as a result of perceptual narrowing (Kuhl, Tsao, & Liu, 2003). Experimental studies have shown that manipulating contingent maternal verbal responses to consist solely of either vowel or consonant-vowel responses to 9-month-old infants’ vocalizations results in specific increases in infants’ production of the same category of vocalizations they received as responses (Goldstein & Schwade, 2008).

The degree to which environmental influences impact children’s language abilities in meaningful, stable ways remains a matter of debate and may vary by domain (Hoff, 2006). For example, while contingent interactions influence the timing and content of infants’ phonemic perception, the amount of input required for typical acquisition is fairly minimal and most children will learn to distinguish the phonemes of their native language at roughly the same ages (Elsabbagh et al., 2013; Kuhl et al., 2003; Nittrouer & Burton, 2005). However, diminished early exposure to language (e.g., for infants with
chronic ear infections or from low socioeconomic status, or SES, backgrounds, both of whom hear less speech) is associated with difficulties with speech perception and phonological awareness that persist well into childhood (Nittrouer & Burton, 2005; Nittrouer, 1996). Differences in children’s vocabulary size also persist throughout early childhood and seem to be driven primarily by SES-related differences in early exposure to speech (Hart & Risley, 1995; Pan et al., 2005; Rowe, 2012). The impact of varying environmental input on infants’ language abilities seems to be particularly significant for infants receiving relatively diminished environmental input, though the factors that predict such diminished environmental input vary considerably. Some of the factors associated with reduced input include maternal depression, low SES (including maternal education levels and knowledge of child development), and chronic ear infections or other issues that impede infants’ perceptual abilities (Bettes, 1988; Hart & Risley, 1995; Pan et al., 2005; Rowe, 2008). These findings are relevant to the study of variation in the language development of high risk infant siblings because both high risk infants and their mothers exhibit characteristics that may contribute to diminished maternal input or impeded infant use of such input that may contribute to high risk infants’ variable language and communication development. While the relationships between maternal input, the factors that may influence maternal input, and the relations between maternal input and high risk infants’ language and communication development have not been explicitly studied, the following section reviews the existing literature on changes in the behavior of mothers of high risk infant siblings that may contribute to the observed variability in these infants’ language and communication development.
Behavioral Differences in Mothers of Infant Siblings of Children with ASD

There is emerging evidence that mothers of high risk infants demonstrate behavioral differences during dyadic interactions with their infants that may influence their infants’ language and communication abilities. A retrospective analysis of family home videos of children with ASD, developmental delays, or typical development found that mothers of children later diagnosed with autism use significantly more regulating behaviors (e.g. behaviors that increase infants’ emotional arousal) than mothers of typically developing infants as early as 6 months of age. By 12 months, differences in regulating behaviors distinguish mothers of infants later diagnosed with ASD from mothers of both typically developing infants and those with other developmental delays (Saint-Georges et al., 2011). Parents of high risk infant siblings as a whole (both those later diagnosed with ASD and those who are not) are rated as being more directive and showing less sensitive responding during parent-child interactions filmed when infants are between 6 and 10 months of age (Wan et al., 2012). By 12 months, group differences in maternal directiveness are driven by parents of infants later diagnosed with ASD (versus high risk infants not diagnosed), who were rated as significantly more directive than mothers of low risk infants. Simultaneously, infants later diagnosed with ASD were rated as less attentive to their parents and scored lower in dyadic mutuality than both non-diagnosed high risk and low risk infants (Wan et al., 2013). Talbott and colleagues (2013) examined the frequency and diversity of gestures used by 12-month old high risk infant siblings and their mothers. They found that compared to mothers of low risk, typically developing infants, mothers of non-diagnosed high risk infant siblings used significantly
more gestures during these interactions. Mothers of infants later classified as meeting criteria for ASD produced more gestures than mothers of low risk infants, but fewer gestures than mothers of non-diagnosed high risk infants, and did not differ significantly from either group. Only infants later diagnosed with ASD demonstrated significantly decreased rates of gesture production.

The underlying sources and extent of these differences in maternal behavior have not been explicitly studied. One possibility is that changes in maternal behavior reflect responses to infants’ emerging symptoms, even if mothers are not conscious of such changes (Dunst, Lowe, & Bartholomew, 1990; Leezenbaum, Campbell, Butler, & Iverson, 2013; Talbott, Nelson, & Tager-Flusberg, 2013). This interpretation is supported by the finding that mothers of infants’ who gesture less frequently (and who are later diagnosed with ASD) do not exhibit the same increase in gesture production as mothers of non-diagnosed high risk infants (Talbott et al., 2013). Indeed, decreased rates of gesture production by infants later diagnosed with ASD provide fewer opportunities for mothers to ‘translate’ these gestures verbally, responses which provide rich language-learning opportunities for infants, regardless of diagnosis (Goldin-Meadow et al., 2007; Leezenbaum et al., 2013). Another possibility is that in addition to reflecting differences in infant behavior, changes in maternal behavior may also reflect the unique background and personal characteristics of mothers of high risk infant siblings. Importantly, some of these characteristics are hypothesized to predict enhanced maternal input and others are hypothesized to predict diminished maternal input. Factors that may lead to enhanced maternal input include high risk parents’ knowledge of early autism symptoms, exposure
to intervention strategies through their experiences with an older diagnosed child, and the increased rates of concern reported by high risk parents if those elevated concerns reflect an increased attention and prompting of infants’ social communication. Factors that may lead to diminished maternal input include mothers own broader autism phenotype characteristics and depressive symptoms, both of which are elevated in parents of children with ASD and are associated with reduced pragmatic language skill and impoverished linguistic input, respectively (Bailey, Golden, Roberts, & Ford, 2007; Ingersoll & Hambrick, 2011; Lindgren, Folstein, Tomblin, & Tager-Flusberg, 2010; Ruser et al., 2007). Elevated levels of early concerns may also contribute to diminished, rather than enhanced, maternal input if those early concerns are driven primarily by increased anxiety and less sensitivity to infants’ concurrent abilities.

While previous investigations have analyzed high risk mothers’ concerns about their infants’ development, they have focused exclusively on the clinical utility of these concerns in terms of predicting infants’ diagnostic outcomes (Hess & Landa, 2012; Ozonoff et al., 2009). The relationship between these maternal concerns and maternal behavior has not been examined. Mothers of high risk infantsiblings report increased levels of concern early as 6 months of age - significantly earlier than mothers of children with ASD identified via community screenings (Ozonoff et al., 2009; Veness et al., 2012). Importantly, these very early concerns are poorly correlated with infants’ concurrent developmental level or eventual diagnostic outcome. Around 12 months of age, maternal concerns are moderately associated with infants’ concurrent functioning and eventual diagnostic status, but they are not a robust predictor of diagnostic outcome
(Hess & Landa, 2012; Ozonoff et al., 2009). These findings suggest that rather than fully reflecting infants’ symptoms or developmental level, maternal concerns in the first year of life likely reflect more general maternal hypervigilance. This increased hypervigilance has been offered as an explanation for high risk mothers’ increased gesture production and directiveness, but these hypotheses have not been tested (Talbott et al., 2013; Wan et al., 2012, 2013).

Together, the existing data on the parent-child interactions of infant siblings of children with ASD and their mothers suggest that infant and maternal characteristics each contribute to early dyadic interactions. Data from non-clinical populations highlight the importance of better understanding how high risk parents’ unique experiences, psychological profiles, and other family factors may shape maternal input and dyadic interactions with their high risk infants. If either maternal or infant characteristics result in mothers of high risk infants providing diminished linguistic input to their infants, this would have important implications for intervention practices, which could focus on enhancing the frequency or quality of maternal linguistic input.

**Current Studies**

The overarching goals of the current studies was to better understand the factors that contribute to maternal language and communication input to their infants and the relations between maternal input and infants’ language and communication abilities. In addition to any effects of infants’ emerging symptoms on mothers’ behavior, other non-infant maternal factors examined includemothers early concerns about their infants’ development, broader autism phenotype characteristics, education, depression symptoms,
and experience with an older diagnosed child in terms of that child’s symptom severity across the first year of life. The relations between each of these factors, including parents’ concerns, and parental behavior at 9 and 12 is also explored. Rather than focusing on maternal and infant behavior in the laboratory, the individual studies conducted here utilize data collected via more naturalistic, home-based diary procedures. This home-based data will help to determine how representative maternal input measured in the laboratory is of the everyday interactions of the home, not only for infants at risk, but for typically developing infants as well. While many studies have highlighted the importance of early social interactions in shaping the language development of typically developing infants, virtually nothing is known about how these maternal factors contribute to the language acquisition of high risk infants (Elsabbagh et al., 2013; Goldstein & Schwade, 2008; Hart & Risley, 1992; Rowe et al., 2008). Additionally, we know from both typically developing and other non-ASD clinical samples that maternal behavior itself is influenced by both unique maternal and family characteristics, but the impact of these characteristics on the behavior of mothers of infant siblings of children with ASD has not been explored. Understanding both the influences on maternal behavior and the influence of maternal behavior on the language and communication of infants at risk for autism will have important implications for our understanding of the environmental influences on the language development of this high risk population.

**Overall Study Design**

Data for these studies was collected as part of a longitudinal study of infants at risk for ASD conducted jointly at Boston Children’s Hospital and Boston University. All
infants were at least second-born (i.e. all infants had at least one older sibling). Infants with at least one older sibling with ASD were enrolled as high risk infants; infants whose older siblings were typically developing were enrolled as low risk control infants. Detailed information on the subject demographics of each study sample is included in the relevant chapters. As part of the larger longitudinal study, infants were seen in the laboratory several times from 3 to 36 months of age where they participated in a range of standardized behavioral assessments and neurophysiological paradigms. In addition to these laboratory visits, families were asked to provide both written and home diary measures from 6 to 18 months of age. The video diaries were filmed monthly and consisted of semi-structured interactions between infants’ and their primary caregivers, lasting approximately 20 minutes. Because the vast majority of primary caregivers were mothers, the referent mother is used throughout. Mothers were instructed to present infants with a series of toys, play social games, attempt to elicit vocal imitation and smiles, read a picture book, and play for several minutes ‘in whatever way makes [them] feel most comfortable.’ Written diaries were completed weekly, and consist of 8 items. Mothers were asked to report on new sounds, words, or gestures their infant made that week, to describe infants’ play with their parents, a sibling, and alone, and describe any concerns about their infants’ development. These home diaries were analyzed using novel coding schemes (described in detail in each accompanying chapter), and were compared to standardized behavioral measures collected in the laboratory or via questionnaire in order to address the following specific aims:
Aim 1: Describe mothers’ home-based concerns over the first year of life and relations between these concerns and maternal and family characteristics. Previous studies on the early concerns of mothers of infant siblings of children with ASD have focused exclusively on the clinical utility of these early concerns in predicting infants’ diagnostic outcomes (Hess & Landa, 2012; Ozonoff et al., 2009). While parents of high risk infants report concerning behaviors much earlier than parents without older children with ASD, these concerns are poorly correlated with infants concurrent abilities and later diagnostic status, suggesting that maternal concerns in the first year of life reflect more than infant characteristics (Herlihy, Knoch, Vibert, & Fein, 2013; Ozonoff et al., 2009). One goal of this dissertation work was to better describe mothers’ early concerns in terms of their daily experiences as the parent of a high risk infant sibling, and the non-infant factors that are associated with their concerns in the first year of life. This was accomplished by analyzing both the frequency of maternal concerns reported via weekly open-ended home diaries and the associations between these diary-based concerns and maternal and family characteristics hypothesized to underlie the increased levels of maternal concern we expected to find.

Aim 2: Determine whether the language and communicative input mothers provide to their infants differs in mothers of high risk and low risk infants. Infant siblings of children with ASD demonstrate difficulties with early language and communication over the first few years of life that are thought to reflect their increased familial risk for ASD (Ozonoff et al., 2010, 2014; Rogers, 2009; Zwaigenbaum et al., 2005). One possibility that has not yet been explored is that some of this variation in
early language and communication ability is the result of diminished environmental input, and in particular, the language and communication mothers’ of high risk infants provide. This possibility is supported by previous findings that diminished maternal input associated with low SES, maternal depression, chronic infant ear infections and a range of other factors is associated with relatively impoverished linguistic input and consequently, reductions in infants language and communication abilities (Bettes, 1988; Nittrouer & Burton, 2005; Rowe, Pan, & Ayoub, 2005; Stein, Malmberg, Sylva, Barnes, & Leach, 2008). The possibility that mothers of infant siblings of children with ASD demonstrate similar reductions in linguistic input was explored by examining maternal behavior during home-based parent-child interactions filmed when infants were 9 and 12 months of age.

**Aim 3: Describe the relations between infant, family, and maternal characteristics, including early concerns, and maternal language and communicative input.** The final goal of this dissertation work was to better understand the relations between parental concerns and other background factors and maternal input and between maternal input and infant language and communication abilities. These goals were accomplished by examining maternal and infant behavior during home-based interactions filmed when infants were 9 and 12 months of age. At 9 months, this included analysis of mothers’ spontaneous responses to infants’ early vocalizations and relations between these early infant vocalizations, maternal responses and infant language abilities at 12 months of age. This particular task was selected for several reasons. The first is that previous work with typically developing dyads has suggested that maternal responses to
infants’ vocalizations at 9 months of age are related to infants’ language development. The second is that high risk infant siblings demonstrate reduced rates of consonant production at 9 months that may be driven by reduced frequency of maternal reinforcement of their vocalizations. Neither of these hypothesis have been explored previously. At 12 months, the relations between maternal prompting and infants’ social communication rate were examined. This task was selected because although high risk infant siblings demonstrate difficulties responding to social bids and mothers of both high risk infant siblings and toddlers diagnosed with ASD exhibit changes in attention-regulating behaviors, the extent to which these attention regulating behaviors relate to high risk infants’ social communicative abilities has not been explored. The work presented here fills these gaps in the literature by closely examining not only the relations between maternal and infant behavior in these domains, but the factors that predict maternal behavior in each of these contexts as well.

**Significance**

There is significant variation in the early developmental trajectory of ASD symptoms, particularly for infant siblings of children with autism, but the environmental influences on the language development of infants at increased familial risk for ASD remain poorly understood. Longitudinal, prospective investigations of high risk infant siblings of children with ASD have helped to overcome barriers to examining the early language and communication development of these high risk infants, but there remain significant limitations in our understanding of the factors that promote early language development, particularly within the everyday interactions of infants and their caregivers.
The studies presented in this dissertation fill these gaps in the literature by closely examining factors that contribute to variation in the environmental input they receive (i.e. maternal language and communication) and how this environmental input is related to infants’ language and communication abilities. Better understanding how risk status influences the early dyadic interactions of high risk infant siblings and their mothers will have important implications for our understanding of language development in this population, and may point to avenues to target for future intervention efforts. If mothers of high risk infants provide impoverished input to their high risk infants either as a result of infants’ own emerging symptoms or as a result of other unique maternal risk factors (i.e. depression), improving maternal input may help to promote language development in high risk infant siblings.
CHAPTER 2: STUDY 1: MATERNAL HOME-BASED CONCERNS

Parents are often the first person to report concerns about their child’s development and therefore serve as the first step in the screening and diagnostic process for autism spectrum disorders (ASD) that enable access to intervention services. Because of this important role in gaining access to treatment, several studies have examined the clinical utility of parents’ early concerns in predicting diagnostic outcomes. In general, parents of children with ASD report elevated levels of concern beginning in the second year of life (De Giacomo & Fombonne, 1998; Veness et al., 2012). Because infant siblings of children with ASD are at increased risk for developing ASD themselves, a limited number of investigations have examined whether parents of infant siblings report earlier and more clinically meaningful concerns about these later-born infants (Herlihy et al., 2013; Ozonoff et al., 2009; Ozonoff, Young, et al., 2011). Parents of high risk infants do in fact report concerns as early as 6 months of age, but their concerns do not begin to predict long-term diagnostic outcomes until the end of the first year of life (Ozonoff et al., 2009). While determining when and how parent concerns have the greatest clinical utility is of great importance, the fact that concerns reported by parents of high risk infant siblings of children with ASD are not well correlated with infant abilities suggests that these very early concerns reflect more than early symptom emergence. Previous investigations have hypothesized that parental concerns in the first year of life, primarily reported by mothers, reflect an overall increase in hypervigilance regarding their infants’ development, and that this hypervigilance underlies some of the changes in maternal behavior that have also been reported in high risk samples (Ozonoff et al., 2009; Talbott
et al., 2013; Wan et al., 2012). The goal of the current study is to better describe the day to day concerns of mothers of high risk infant siblings in the first year of life and some of the maternal and family factors that may contribute to these early concerns. The existing literature on parental concerns will be reviewed before outlining the specific goals of the current study.

**Clinical Utility of Parent Concerns**

Both retrospective and prospective investigations have demonstrated that despite the presence of behavioral differences in key domains beginning around 12 months of age in infants later diagnosed with ASD, parents typically do not report concerns until the second year of life (Chawarska et al., 2007; De Giacomo & Fombonne, 1998; Herlihy et al., 2013; Mitchell et al., 2006; Osterling & Dawson, 1994; Veness et al., 2012; Werner et al., 2000). On retrospective measures, parents for whom the diagnosed child is the second child with ASD in the family tend to report first becoming concerned about their infants’ development around 10 months of age, suggesting some benefit of prior experience with ASD in detecting early symptoms (Herlihy et al., 2013). Despite these findings, the clinical utility of these studies is limited by their retrospective nature; parent reports become less accurate as more time elapses between the time of the child’s diagnosis and the time of reporting, and retrospective reports in general are poorly correlated with objective measures of infant symptoms (Ozonoff, Iosif, et al., 2011).

Prospective investigations reduce some of the issues with retrospective methods by monitoring infant behavior and parental reports as they occur, yet only three prospective studies have examined the relationship between early parent concern and
diagnostic outcome, and all have focused on their clinical utility. The first is a community-based investigation that reported on 114 children (18 with ASD, 16 with Developmental delays, 18 with Specific Language Impairment, and 60 typically developing controls) who had been followed longitudinally from 8 months through 4 years as part of a large, population based study and were identified as meeting criteria for one of these diagnosis at 4 years of age (Veness et al., 2012). Parent concerns and a variety of standardized parent report measures of infant language and social communication abilities that had been collected at 8, 12, and 24 months of age were analyzed to determine whether parents of any of these groups of children had reported significantly more concerns regarding their child’s development at any of these ages. Parents of children with ASD reported significantly more concerns than the other groups at 24 months of age, but not at either 8 or 12 months. Parental concerns were not included in logistic regression analyses predicting diagnostic classification (only standardized behavioral measures were included), so the extent to which parental concerns at 24 months predict diagnosis in this population based sample is not clear (Veness et al., 2012). The other two prospective investigations involved parents of infant siblings of children with ASD. Ozonoff and colleagues (2009) collected parent concerns through open-ended queries during study visits in the laboratory at 6, 12, and 18 months of age. They found that although parents of high risk infants report increased levels of concern beginning as early as 6 months of age, these early concerns were poorly related to concurrent infant symptoms and did not help to predict infants who would later be diagnosed with autism from high risk infants and typically developing infants who were
not diagnosed. By 12 months of age, the presence of 1 or more ASD-related concerns (rather than more general developmental concerns) demonstrated moderate sensitivity and specificity in predicting diagnostic outcomes between high risk infants who were and were not diagnosed with autism. Hess and Landa (2012) followed a similar group of high risk infant siblings, collecting parent concerns at 14, 24 and 36 month laboratory visits. They compared high risk infants who were and were not diagnosed with ASD at 36 months of age (there was no low risk comparison group). Beginning at 24 months, the presence of one or more concerns was associated with an ASD outcome at 36 months of age. While specificity of parental concern was acceptable, sensitivity was low, with many parents of children demonstrating significant impairments not reporting concerns.

Together, the existing literature on early parent concerns suggests that concerns are most accurate when reported close to the time the concerning behavior first occurs (rather than retrospectively reporting on infants’ early behavior), and that the clinical utility of these concerns improves around 12 months of age in terms of predicting diagnostic outcome. Parents of high risk infants may be particularly well-positioned to report on autism symptoms as a result of their experience with ASD; they report concerning behaviors earlier than parents without such experience. What remains unclear is how these clinically-reported concerns reflect parents’ daily experiences and behavior. The poor association between parental concerns and infant behavior at 6 months of age suggests that parental concerns in the first year of life reflect more than infant symptoms (Ozonoff et al., 2009).
The Current Study

One explanation for the elevated levels of concerns reported by parents of high risk infant siblings, particularly in the first year of life, is that they reflect more general hypervigilance and anxiety regarding their infants’ development (Hess & Landa, 2012; Ozonoff et al., 2009). This hypervigilance is not unwarranted given the elevated recurrence risk for ASD in this population and the high numbers of infants who demonstrate difficulties with language and communication even if not diagnosed (Gamliel et al., 2009; Mitchell et al., 2006; Ozonoff et al., 2014; Ozonoff, Young, et al., 2011). The source of this increased hypervigilance is not entirely clear, but may simply reflect parents’ knowledge of their infants’ increased risk – their likely motivation for enrolling in these prospective investigations in the first place. It is also possible that maternal concerns in the first year of life, particularly those occurring prior to the onset of overt behavioral differences in infants themselves, may reflect specific characteristics of mothers of high risk infant siblings. One of these characteristics is the increased prevalence of depressive symptoms amongst mothers of children with ASD (Bailey et al., 2007; Ingersoll & Hambrick, 2011). Since mothers of high risk infant siblings all have at least one older child with ASD, they may exhibit increased levels of depressive symptoms that are associated with their daily experiences of concern. Other experiences related to parenting a child with ASD may also play a role, particularly for parents of more severely affected older children who report higher levels of parenting stress (Estes et al., 2009). Parents of more severely affected older children may also be more sensitive
to early behavioral symptoms in their high risk infants. Each of these factors may manifest in increased concern about the high risk infant sibling.

Better understanding the role of risk status in shaping mothers’ early concerns is important for several reasons. The first is that it may help to improve early identification and access to services within high risk infant samples. It will also shed light on the day to day experiences of mothers of infant siblings with ASD. While previous investigations have focused on maternal concerns reported in a laboratory or clinic setting, the current study investigates concerns recorded in weekly home-based measures. While these home-based measures may be less accurate in predicting infants’ diagnostic outcomes they are more likely to reflect maternal and family characteristics that impact the daily experiences and interactions of high risk infants and their mothers.

In this study, we use home-based diary methods to capture mothers’ weekly concerns regarding their infants’ development in a sample of high risk infant siblings of children with ASD and low risk infant siblings of typically developing infants. These home-based concerns are analyzed at three time points across the first year of life: 6, 9, and 12 months. Maternal concerns at each of these ages are compared to both concurrent infant behavior and maternal and family characteristics hypothesized to contribute to mothers’ home-based concerns, particularly for the high risk sample. These maternal and family factors include the presence of maternal depressive symptoms and education level and the severity of the older diagnosed child.
Methods

Participants

Participants included 89 infant siblings of children with autism and 87 low risk control infants and their mothers. Data for these families were collected as part of a larger longitudinal study of infants at risk for autism conducted jointly at Boston Children’s Hospital and Boston University. For the larger project, interested families were contacted by the study coordinator, who conducted a detailed telephone eligibility interview. All infants were screened for exclusionary criteria (prematurity, extended stays in the neonatal intensive care unit, maternal drug or alcohol use during pregnancy, family history of genetic disorders associated with ASD, and primary languages other than English). Infants were enrolled as high-risk infant siblings of children with autism (HRA) if they had an older sibling with a clinical diagnosis of Autism, Asperger Syndrome, or Pervasive Developmental Disorder-Not Otherwise Specified (PDD-NOS), confirmed by expert community diagnosis. Infants were enrolled into the low risk control (LRC) group if they had at least one older sibling who was typically developing and no first-degree relatives diagnosed with an ASD or other neurodevelopmental disorder. The sample had a balanced gender ratio, with 51% of the total sample being male. Twenty-three (13%) infants were from a racial or ethnic minority group. There were no group differences in gender ratio, racial background, or family income (all p’s > .24). Descriptive information on these subject characteristics is presented in Table 2.1. Informed consent was obtained from parents prior to participation.
Measures

Infants were seen in the laboratory several times across the first three years of life, where several standardized behavioral and diagnostic and autism symptom assessments were collected.

**Mullen Scales of Early Learning.** At the 6 and 12 month visit, infants’ developmental abilities were assessed using the Mullen Scales of Early Learning (MSEL; Mullen, 1995). The MSEL provides age-equivalent scores for 4 cognitive subscales (expressive and receptive language, visual reception, and fine motor) as well as an overall Cognitive T Score for the 4 cognitive domains. Verbal and non-verbal developmental quotients (VDQ and NVDQ) were calculated from the two verbal (Receptive and Expressive Language) and two non-verbal (Fine Motor, Visual Reception) cognitive subscale scores by averaging the two relevant subscale age equivalents, dividing by the child’s chronological age, and multiplying by 100. These two scores were used as measures of verbal and non-verbal abilities at 6 and 12 months of age.

**Autism Observation Scale for Infants.** Infants’ autism symptoms were assessed at 9 and 12 months using the Autism Observation Scale for Infants (AOSI; Bryson, McDermott, Rombaugh, Brian, & Zwaigenbaum, 2000). The AOSI is an 18-item assessment that measures a range of autism-related behaviors (visual attention and
tracking, social interest and reciprocity, affect, atypical sensory and motor behaviors, etc.) during a brief semi-structured interaction between a trained examiner and the infant, who is seated on their parents’ lap. Individual items are scored from 0 to 2 or 3, with higher scores indicating greater atypicality. The scale yields two final scores: the total number of items endorsed, and the total raw score (out of a possible 50). AOSI total scores were used here as a measure of autism symptoms at 9 and 12 months of age.

**Autism Diagnostic Observation Schedule.** Infants’ autism symptoms were assessed at 18, 24, and 36 months using the Autism Diagnostic Observation Schedule (ADOS; Lord et al., 2000). The ADOS measures children’s language, social communication, and restricted or repetitive behaviors during a semi-structured interaction with a trained examiner. Empirically derived cutoffs can be used to classify children as meeting criteria for an ASD. ADOS scores from infants’ final study visit were used to classify infants into diagnostic outcome groups along with clinical best estimates, when available.

**Maternal education.** Information in maternal education levels was collected at study intake along with other demographics measures. There were 9 education level options ranging from ‘some high school’ through ‘doctorate’. These education levels were ordered from 1-9, with higher numbers representing more years of formal education.

**Proband autism symptoms.** Proband ASD symptoms were measured during the telephone screen using the Social Communication Questionnaire (SCQ; Rutter, Bailey, & Lord, 2003). The SCQ is a 40-item parent report screening measure that covers
communication, social interactions, and restricted and repetitive behaviors. There are two different versions of the SCQ: a ‘current’ version for children under the age of 5 and a second ‘lifetime’ version for children 5 years or older. Total score is out of 39, with higher scores indicating greater impairment.

**Maternal depression.** Maternal depression was measured at 6 and 12 months using the Center for Epidemiologic Studies Depression Scale: Revised (CESD-R; Eaton, Smith, Ybarra, Muntaner, & Tien, 2004). The CESD-R is a 20-item questionnaire designed to measure depressive symptomology. A revised version, with two items related to death and self-harm removed, was used as a self-report measure of maternal depressive symptoms.

**Parent-Reported Diary Concerns.** On a weekly basis from 6 through 18 months, parents were given the opportunity to report about their child’s development across a range of language and social domains in home-based diaries. The final diary item was an open ended question asking if parents had any concerns about their child. Diaries could be recorded on physical paper diaries provided by project staff and returned via pre-stamped envelopes, or via an online system set up with an individual id number and personal password. Parents’ responses to the concern item were coded for content, and placed into 1 of 16 categories. For many of the diaries, the concerns field was left blank. If the parents had answered earlier questions (describing their child’s language or play for that week), these blank responses were considered ‘no concerns’. Diaries with all fields left blank were discarded and not included in these or any other analyses.
From these total responses, four concerns subscales of interest were created: Language, Social Communication, Restricted or Repetitive Behaviors (RRB), and General/Medical Concerns. An Autism Concerns subscale was created by summing the Language, Social Communication, and RRB subscales. Descriptive information on each of these categories is provided in Table 2.2.

**Table 2.2 Home-Based Diary Concerns Categories**

<table>
<thead>
<tr>
<th>Autism Concerns</th>
<th>Restricted or Repetitive Behaviors</th>
<th>General/Medical</th>
</tr>
</thead>
<tbody>
<tr>
<td>Language</td>
<td>Social Communication</td>
<td></td>
</tr>
<tr>
<td>• babbling</td>
<td>• eye contact</td>
<td>• teething</td>
</tr>
<tr>
<td>• words</td>
<td>• gestures</td>
<td>• feeding</td>
</tr>
<tr>
<td>• language loss</td>
<td>• social interactions</td>
<td>• sleeping</td>
</tr>
<tr>
<td>• odd sounds</td>
<td></td>
<td>• motor milestones</td>
</tr>
<tr>
<td></td>
<td></td>
<td>• illnesses</td>
</tr>
<tr>
<td></td>
<td></td>
<td>• activity level</td>
</tr>
</tbody>
</table>

Parents’ concerns that explicitly mentioned ASD were included in the relevant category of concern (i.e. a parent reporting they were concerned because their child stopped saying ‘ball’ and they knew language loss was a red flag for autism would be coded as a language-related concern).

**Results**

**Overview**

A total of 3,003 individual diaries were returned (1458 HRA, 1545 LRC). There were no significant differences in the average number of diaries completed by each group \((t(175) = -.638, p = .53)\). In order to address our primary goal of describing the
relationship between parents diary-reported concerns and infant’s concurrent
developmental ability and autism symptoms, three time periods that overlapped with our
laboratory-based measures were selected for analysis: 6-8 months, 9-10 months, and 11-
13 months. Scores from all diaries completed during each window were averaged for
each of the concerns categories (Language, Social Communication, RRB, Autism, and
General/Medical). Information on the number of subjects and the number of diaries
contributed to each of these three analysis windows is included in Table 2.3. There were
no significant differences between the groups at any age in the average number of diaries
contributed by each group of infants, either by risk group or diagnostic group (all p’s >
.05).

### Table 2.3 Average Number of Diaries Completed at Each Age, by Outcome Group

<table>
<thead>
<tr>
<th>Age</th>
<th>Outcome Group</th>
<th>LRC</th>
<th>HRA-N</th>
<th>ASD</th>
</tr>
</thead>
<tbody>
<tr>
<td>6 Months</td>
<td>Number of Diaries (mean, SD)</td>
<td>2.21 (.82)</td>
<td>2.42 (.73)</td>
<td>2.5 (.67)</td>
</tr>
<tr>
<td></td>
<td>Number of Subjects</td>
<td>n = 81</td>
<td>n = 54</td>
<td>n = 12</td>
</tr>
<tr>
<td>9 Months</td>
<td>Number of Diaries (mean, SD)</td>
<td>1.74 (.44)</td>
<td>1.77 (.42)</td>
<td>1.67 (.49)</td>
</tr>
<tr>
<td></td>
<td>Number of Subjects</td>
<td>n = 65</td>
<td>n = 44</td>
<td>n = 9</td>
</tr>
<tr>
<td>12 Months</td>
<td>Number of Diaries (mean, SD)</td>
<td>2.40 (.79)</td>
<td>2.36 (.79)</td>
<td>2.21 (.80)</td>
</tr>
<tr>
<td></td>
<td>Number of Subjects</td>
<td>n = 53</td>
<td>n = 42</td>
<td>n = 14</td>
</tr>
</tbody>
</table>

### Diagnostic Outcomes

Infants were classified as meeting criteria for ASD (hereafter, the ASD group) if
they met ADOS criteria for ASD on their most recent study visit and received clinical
judgments of ASD by an expert clinician if the most recent visit was their final study
visit. Seventeen infants met these classification criteria and were included in the ASD
group. Thirteen of these infants met both ADOS criteria and received clinical judgments of ASD (10 from 36 month data, 3 from 18 month data). An additional 4 infants met ADOS criteria at their most recent visit (3 at 24, 1 at 18) but have not yet received clinical judgments. HRA infants who did not meet on the ADOS or who were judged to be typically developing are hereafter referred to as HRA-N.

**General Analytic Approach**

Data are presented within age, with respect to both initial risk group and separately by diagnostic outcome. Due to the relatively large number of ‘no concerns’ reported by all parents on the diary measures and significant skew in the diary measures as well as several of the behavioral measures, non-parametric analyses are used throughout. A Bonferroni-Holm correction was applied to correct for multiple comparisons within age for pairwise Chi-square analyses (Holm, 1979). Within each age and group analysis, we first examined group differences in the proportion of parents reporting at least one concern and whether infants’ ASD symptoms, cognitive, and language abilities were worse for infants whose parents had reported such concerns. We were also interested in describing the associations between infant behavior, maternal and family characteristics, and maternal concerns when concerns occur. To answer these questions, correlational analyses were conducted for the subset of HRA parents who reported at least one concern of any type during each of the three analysis windows.
Within Age Analyses

6 month data.

**Family risk group comparisons.** Chi-square analyses were used to assess whether the proportion of parents reporting concerns differed between parents of high risk infants as a whole (including those later diagnosed) and parents of low risk infants. Significant group differences were observed for the Language, Social Communication, RRB, and Autism Concerns, with parents of high risk infants reporting more concerns than parents of low risk infants. There were no significant group differences for General/Medical Concerns. Results are summarized in Table 2.4, including the percentage of parents in each group reporting concerns and relevant odds ratios, which represent the probability of a parent in the high risk group reporting a concern in a given subscale relative to the probability of a parent in the low risk group reporting a concern of the same type (e.g. at 6 months, HRA parents are 4.27 times more likely to report an Autism concern than LRC parents).

We next examined HRA infants’ concurrent VDQ and NVDQ scores (available for 60 HRA infants) to determine whether infants whose mothers had reported at least one concern during this period had lower scores on these standardized measures than infants whose mothers had reported no concerns. There were no significant differences in the VDQ and NVDQ scores for these two groups of infants (VDQ: Mean difference = 3.98, \( p = .34 \); NVDQ: Mean difference = 1.96, \( p = .67 \)). Finally, for HRA mothers who had reported at least one concern (38 HRA infants), Spearman correlations were used to
<table>
<thead>
<tr>
<th>Concern Type</th>
<th>6 Months</th>
<th>9 Months</th>
<th>12 Months</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>df = 1</td>
<td>df = 1</td>
<td>df = 1</td>
</tr>
<tr>
<td>N</td>
<td>150</td>
<td>124</td>
<td>109</td>
</tr>
<tr>
<td>Languages</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Contrast</td>
<td>$\chi^2 = 7.41$</td>
<td>$\chi^2 = 7.99$</td>
<td>$\chi^2 = 12.59$</td>
</tr>
<tr>
<td>Reporting</td>
<td>HRA = 23.2%</td>
<td>HRA = 18.6%</td>
<td>HRA = 35.7%</td>
</tr>
<tr>
<td>Percent</td>
<td>$p = .010$</td>
<td>$p = .007$</td>
<td>$p = .000$</td>
</tr>
<tr>
<td>OR</td>
<td>OR = 3.77</td>
<td>OR = 7.22</td>
<td>OR = 6.81</td>
</tr>
<tr>
<td>Social</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Communication</td>
<td>$\chi^2 = 8.35$</td>
<td>$\chi^2 = 8.17$</td>
<td>$\chi^2 = 6.58$</td>
</tr>
<tr>
<td>Reporting</td>
<td>HRA = 13%</td>
<td>HRA = 11.9%</td>
<td>HRA = 16.1%</td>
</tr>
<tr>
<td>Percent</td>
<td>$p = .006$</td>
<td>$p = .005$</td>
<td>$p = .017$</td>
</tr>
<tr>
<td>OR</td>
<td>OR = 12.0</td>
<td>-</td>
<td>OR = 10.0</td>
</tr>
<tr>
<td>RRB</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Contrast</td>
<td>$\chi^2 = 6.13$</td>
<td>$\chi^2 = 5.67$</td>
<td>$\chi^2 = 6.53$</td>
</tr>
<tr>
<td>Reporting</td>
<td>HRA = 13.05%</td>
<td>HRA = 15.3%</td>
<td>HRA = 19.6%</td>
</tr>
<tr>
<td>Percent</td>
<td>$p = .024$</td>
<td>$p = .025$</td>
<td>$p = .016$</td>
</tr>
<tr>
<td>OR</td>
<td>OR = 5.9</td>
<td>OR = 5.7</td>
<td>OR = 6.2</td>
</tr>
<tr>
<td>Medical</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Contrast</td>
<td>$\chi^2 = 2.41$</td>
<td>$\chi^2 = .998$</td>
<td>$\chi^2 = .183$</td>
</tr>
<tr>
<td>Reporting</td>
<td>HRA = 47.8%</td>
<td>HRA = 37.3%</td>
<td>HRA = 37.5%</td>
</tr>
<tr>
<td>Percent</td>
<td>$p = .14$</td>
<td>$p = .37$</td>
<td>$p = .70$</td>
</tr>
<tr>
<td>OR</td>
<td>OR = .60</td>
<td>OR = .70</td>
<td>OR = .85</td>
</tr>
<tr>
<td>Autism</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Contrast</td>
<td>$\chi^2 = 12.17$</td>
<td>$\chi^2 = 13.90$</td>
<td>$\chi^2 = 17.56$</td>
</tr>
<tr>
<td>Reporting</td>
<td>HRA = 34.8%</td>
<td>HRA = 32.2%</td>
<td>HRA = 48.2%</td>
</tr>
<tr>
<td>Percent</td>
<td>$p = .001$</td>
<td>$p = .000$</td>
<td>$p = .000$</td>
</tr>
<tr>
<td>OR</td>
<td>OR = 4.27</td>
<td>OR = 7.24</td>
<td>OR = 7.29</td>
</tr>
</tbody>
</table>

Note: Bonferroni-Holm corrections were applied within age. Significant Chi-square results are bolded. RRB = Restrictive, Repetitive Behaviors; OR = Odds Ratio.
analyze the associations between each of the 4 ASD-related concerns subscales (Language, Social Communication, RRB, and Autism) and infants’ concurrent MSEL VDQ and NVDQ scores. None of these associations were significant.

**Diagnostic outcome group comparisons.** Group differences in the proportion of parents reporting concerns across the LRC, HRA-N and ASD groups were assessed across each of the concern categories, yielding 5 omnibus chi-square analyses. Significant results were followed up with pairwise contrasts between each of the three groups. These pairwise contrasts revealed that at 6 months, HRA-N parents were significantly more likely than LRC parents to report Social Communication and Autism Concerns. No other pairwise contrasts were significant. These results are summarized in Table 2.5, which also includes descriptive information on the percentage of parents reporting concerns for each category, and the odds ratios for each contrast in each category.

There were no significant differences in VDQ and NVDQ scores for infants whose parents had reported concerns and infants whose parents had reported no concerns for either HRA-N (VDQ: Mean difference = 5.89, \( p = .19 \); NVDQ: Mean difference = 1.07, \( p = .84 \)) or ASD (VDQ: Mean difference = -3.19, \( p = .71 \); NVDQ: Mean difference = 6.67, \( p = .33 \)). For infants whose parents had reported at least one concern during this time period (32 HRA, 6 ASD), Spearman correlations were used to analyze the associations between parents’ reported concerns across each of the 4 ASD-related concerns subscales and infants’ VDQ and NVDQ scores. The only significant
<table>
<thead>
<tr>
<th>Concern Type</th>
<th>Omnibus $df = 2, N = 150$</th>
<th>Percent Reporting Concerns</th>
<th>Omnibus $df = 2, N = 150$</th>
<th>Percent Reporting Concerns</th>
<th>Omnibus $df = 2, N = 150$</th>
<th>Percent Reporting Concerns</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>6 Months</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Language</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>HRA- vs. LRC</td>
<td>$\chi^2 = 7.91$ $p = .019$</td>
<td>ASD = 16.7% HRA- = 24.6% LRC = 7.4%</td>
<td>HRA- vs. LRC</td>
<td>$\chi^2 = 8.60$ $p = .014$</td>
<td>ASD = 13.3% HRA- = 20.5% LRC = 3.1%</td>
<td>HRA- vs. LRC</td>
</tr>
<tr>
<td>ASD vs. LRC</td>
<td>(1.14, .274) OR = 1.9</td>
<td>ASD = 4.1 ASD = 5.3</td>
<td>ASD vs. LRC</td>
<td>(2.70, .158) OR = 4.8</td>
<td>ASD vs. LRC</td>
<td>(3.74, .712) OR = 61</td>
</tr>
<tr>
<td>RRB</td>
<td>$\chi^2 = 6.61$ $p = .037$</td>
<td>ASD = 8.3% HRA- = 14.0% LRC = 2.5%</td>
<td>HRA- vs. LRC</td>
<td>$\chi^2 = 11.46$ $p = .003$</td>
<td>ASD = 0% HRA- = 20.5% LRC = 3.1%</td>
<td>HRA- vs. LRC</td>
</tr>
<tr>
<td>Autism</td>
<td>$\chi^2 = 13.0$ $p = .002$</td>
<td>ASD = 25% HRA- = 36.8% LRC = 11.1%</td>
<td>HRA- vs. LRC</td>
<td>$\chi^2 = 18.63$ $p = .000$</td>
<td>ASD = 13.3% HRA- = 38.6% LRC = 6.2%</td>
<td>HRA- vs. LRC</td>
</tr>
<tr>
<td>Social</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Communication</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>ASD vs. LRC</td>
<td>(1.50, .016) OR = 0</td>
<td>ASD = 0% HRA- = 15.8% LRC = 1.2%</td>
<td>ASD vs. LRC</td>
<td>(4.39, .187) OR = 0</td>
<td>ASD vs. LRC</td>
<td>(5.2, .666) OR = 61</td>
</tr>
<tr>
<td>RRB</td>
<td>$\chi^2 = 6.61$ $p = .037$</td>
<td>ASD = 8.3% HRA- = 14.0% LRC = 2.5%</td>
<td>HRA- vs. LRC</td>
<td>$\chi^2 = 11.46$ $p = .003$</td>
<td>ASD = 0% HRA- = 20.5% LRC = 3.1%</td>
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</tr>
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<td>HRA- vs. LRC</td>
</tr>
</tbody>
</table>

Note: Bonferroni-holm corrections were applied within age. Significant results are bolded. RRB = Restrictive, Repetitive Behaviors; OR = Odds Ratio.
associations were between Language Concerns and VDQ Scores \( (r_s = -.823, p = .044) \) for the ASD group.

**Summary of 6 month results.** At 6 months of age, mothers of high risk infant siblings are significantly more likely to report concerns about their infants’ development across all three ASD-related concerns categories, but not more likely to report non-ASD related concerns than mothers of low risk infants. Infants whose parents report concerns at this age do not demonstrate difficulty on standardized measures of language and cognitive abilities, nor is there an association between mothers’ level of concern and their infants’ language and cognitive abilities. These patterns are observed across the entire group of high risk infants, both those later classified as meeting criteria for ASD and those who are not.

**9 month data.**

**Family risk group comparisons.** Chi-square analyses were used to assess whether the proportion of parents reporting concerns differed between parents of high risk infants as a whole (including those later diagnosed) and parents of low risk infants. Significant group differences were observed for the Language, Social Communication, RRB, and Autism Concerns, with parents of high risk infants reporting more concerns than parents of low risk infants. There were no significant group differences for General/Medical Concerns. Results are summarized in 2.4, including the percentage of parents in each group reporting concerns and relevant odds ratios.

We next examined HRA infants’ concurrent AOSI total scores (available for 37 HRA infants) to determine whether infants whose mothers had reported at least one
concern during this period had higher scores on this measure of ASD symptoms than infants whose mothers had reported no concerns about their infants. MSEL data was not available at 9 months. There were no significant differences in the AOSI scores of these two groups of infants (Mean difference = .383, \( p = .73 \)). Within the group of HRA mothers who had reported at least one concern (19 HRA infants), Spearman correlations were used to analyze the associations between each of the 4 ASD-related concerns subscales and infants’ AOSI total scores. There were significant associations between infants AOSI total score and Language Concerns (\( r_s = .574, p = .01 \)), RRB Concerns (\( r_s = .50, p = .03 \)) and Autism Concerns (\( r_s = .688, p = .001 \)).

**Diagnostic outcome group comparisons.** Group differences in the proportion of parents reporting concerns across the low risk, high risk non-diagnosed, and diagnosed groups were assessed across each of the concern categories, yielding 5 omnibus chi-square analyses. Significant results were followed up with pairwise contrasts between each of the three groups. These pairwise contrasts revealed that at 9 months, HRA-N parents were significantly more likely than low risk parents to report Social Communication and Autism Concerns. No other pairwise contrasts were significant. These results are summarized in Table 2.5, which also includes descriptive information on the percentage of parents reporting concerns for each category, and the odds ratios for each contrast in each category.

There were no significant differences in AOSI scores for infants whose parents had reported concerns and infants whose parents had reported no concerns for either HRA-N (Mean difference = .243, \( p = .85 \)) or ASD (Mean difference = 1.0, \( p = .60 \)).
Spearman correlations between parents’ reported concerns across each of the 4 ASD-related concerns subscales and 9-month AOSI Total scores were conducted on the subset of infants whose parents had reported at least one concern in any category during the 9 month analysis window and who had available AOSI data (15 HRA, 4 ASD). For HRA infants, there were significant associations between their AOSI total score and Language Concerns ($r_s = .551, p = .03$), and Autism Concerns ($r_s = .672, p = .006$). Correlations could not be calculated for the ASD group due to insufficient non-zero data (e.g. only two parents of ASD infants reported an ASD-related concern).

**Summary of 9 month results.** At 9 months, mothers of high risk infants are significantly more likely to report concerns about their infants’ development across all three ASD-related concerns categories, but not more likely to report non-ASD related concerns than mothers of low risk infants. Infants whose parents report concerns at this age do not demonstrate increased symptoms of ASD on a laboratory-based measured. There were significant associations between mothers’ level of concern and their infants’ laboratory-based ASD symptoms, but this association was observed for the HRA and HRA-N, but not ASD, infants. Mothers of high risk infants later diagnosed with ASD were not more likely to report concerns about their infants than mothers of non-diagnosed HRA infants at 9 months of age.

**12 month data.**

**Family risk group comparisons.** Chi-square analyses were used to assess whether the proportion of parents reporting concerns differed between parents of high risk infants as a whole (including those later diagnosed) and parents of low risk infants. As observed
at both 6 and 9 months, significant group differences were observed for the Language,
Social Communication, RRB, and Autism Concerns, with parents of high risk infants
reporting more concerns than parents of low risk infants. There were no significant group
differences for General/Medical Concerns. Results are summarized in Table 2.4,
including the percentage of parents in each group reporting concerns and odds ratios for
each of the concerns variables.

We next examined HRA infants’ concurrent AOSI total scores (available for 55
HRA infants) and MSEL VDQ and NVDQ scores (available for 54 HRA infants) to
determine whether infants whose mothers had reported at least one concern during this
period had higher ASD symptom or lower cognitive scores than infants whose mothers
had reported no concerns about their infants. There were no significant differences
between HRA infants whose parents reported concerns for AOSI scores (Mean difference
= -.583, p = .39), VDQ (Mean difference = 2.0, p = .70), or NVDQ (Mean difference =
.65, p = .88). Within the group of HRA mothers who had reported at least one concern (34
HRA infants), Spearman correlations were used to analyze the associations between
each of the 4 ASD-related concerns subscales and infants’ AOSI, VDQ, and NVDQ
scores. There were significant associations between Autism Concerns and VDQ ($r_s = -
.372, p = .03$), and NVDQ ($r_s = -.341, p = .05$) and trend-level associations between AOSI
scores and Autism Concerns ($r_s = .330, p = .06$) and RRB Concerns ($r_s = .302, p = .08$)
and between Language concerns and VDQ ($r_s = -.327, p = .06$).

**Diagnostic outcome group comparisons.** Group differences in the proportion of
parents reporting concerns across the low risk, high risk non-diagnosed, and diagnosed
groups were assessed across each of the concern categories, yielding 5 omnibus chi-square analyses. Significant results were followed up with pairwise contrasts between each of the three groups. These pairwise contrasts revealed significant differences in the percentages of parents reporting concerns for the following pairs: ASD vs. LRC for Language Concerns, HRA-N vs. LRC for RRB Concerns, and HRA-N vs. LRC and ASD vs. LRC for Autism Concerns. None of the contrasts between ASD and HRA-N were significant. These results are summarized in Table 2.5, which also includes descriptive information on the percentage of parents reporting concerns for each category, and odds ratios for each contrast in each category.

We next examined HRA-N and ASD infants’ concurrent AOSI, VDQ, and NVDQ scores to determine whether infants whose mothers had reported at least one concern during this period had lower scores on these standardized measures than infants whose mothers had reported no concerns. There were no significant differences between the concerns and no concerns HRA-N infants for AOSI (Mean difference = -.297, p = .67), VDQ (Mean difference = -.134, p = .82), or NVDQ (Mean difference = 1.17, p = .83) or between the concerns and no concerns ASD infants for AOSI (Mean difference = -1.15, p = .55), VDQ (Mean difference = 9.17, p = .42), or NVDQ (Mean difference = -1.92, p = .81).

Spearman correlations between the 4 ASD-related concerns subscales and infants’ 12-month AOSI, VDQ and NVDQ Scores were calculated for the subset of infants whose parents had reported at least one concern in any category during the 12-month analysis window and who had available AOSI and MSEL data (24 HRA, 10 ASD). For HRA-
Ninfants, there were significant associations between RRB Concerns and MSEL Cognitive T Scores ($r_s = -0.419, p = 0.04$), MSEL VDQ ($r_s = -0.582, p = 0.003$), between RRB Concerns and AOSI Total Score ($r_s = 0.43, p = 0.04$) and trend-level significance was observed for the associations between Autism Concerns and MSEL NVDQ ($r_s = -0.359, p = 0.09$). None of the associations were significant for the ASD group.

**Summary of 12 month results.** At 12 months, mothers of high risk infant siblings are more likely to report concerns than mothers of low risk infants across all 3 ASD-related subscales. Although there were no overall differences in infants’ ASD symptoms, language, or cognitive abilities for high risk infants whose parents had reported at least one concern than infants whose parents had reported no concerns, within the group of parents reporting concerns, these concerns were correlated with infants’ behavior. These results were largely unchanged when dividing the high risk sample into infants who were later diagnosed with ASD from those who were not. Mothers of both these high risk groups were more likely to report at least one kind of ASD-related concerns than mothers of low risk infants. Mothers of infants later diagnosed with ASD were not more likely to report any type of concern than mothers of non-diagnosed high risk infants.

**Family Background Factors**

In addition to examining the relationships between parental concerns and both risk group and infant developmental abilities, we examined several family factors hypothesized to potentially relate to HRA parents’ reported levels of concern: maternal education, proband autism symptoms, and concurrent levels of maternal depression.
Group differences in background factors. There were no significant differences between HRA mothers who reported concerns and those with no concerns for the subsets contributing data at 6, 9, and 12 months for maternal education, or proband autism severity. Measures of maternal depression (CESD-R) were available at 6 and 12 months. There were no group differences in CESD-R levels for mothers who did and did not report concerns at 6 months. At 12 months, there was a significant difference in CESD-R scores, with mothers reporting concerns also reporting more depressive symptoms (Concerns: Mean = 14.41, SD = 10.7; No Concerns: Mean = 7.88, SD = 9.0). t(44) = -2.11, p = .041.

Relations between background factors and parent concerns. To better understand the associations between background family factors and parents’ ASD-related concerns when they occur, Spearman correlations were calculated for associations between parents’ ASD Concerns and Maternal Education, Proband Autism Symptoms (SCQ Score) at 6, 9, and 12 months and between ASD Concerns and Maternal Depression at 6 and 12 Months for parents who had reported at least one concern of any type at any age. The only significant correlation was between parents’ ASD Concerns and Proband SCQ at 12 months (r_s = .56, p = .004, n = 25), and at trend level at 9 months (r_s = .33, p = .08, n = 28). The correlation between ASD Concerns and Proband SCQ at 12 months remained significant even when controlling for infants’ concurrent ASD symptoms (AOSI total score; r = .65, p = .001, n = 22).
Discussion

The goal of this study was to describe the concerns reported by parents of infant siblings of children with ASD in the everyday setting of the home at 6, 9, and 12 months of age and to analyze the associations between these home-based concerns, concurrent infant behavior, and other maternal and family characteristics hypothesized to contribute to high risk mothers’ day to day concerns about their infants. We found that across all three ages measured, mothers of high risk infants were significantly more likely than mothers of low risk infants to report concerns about language, social communication, and restricted and repetitive behaviors, but were not more likely to report general, medically-based concerns. These findings mirror those obtained from laboratory-based parental reports of concern at 6 and 12 months, and extend this work by demonstrating that high risk parents’ concerns remain elevated at 9 months as well (Ozonoff et al., 2009).

Closer examination of parental concerns across the three groups of infants – low risk infants, high risk infants later meeting criteria for ASD, and high risk infants who are not diagnosed, reveal that at 6 and 9 months, group differences are observed primarily in the high risk non-diagnosed group, who differ significantly from the low risk group in the number of parents reporting both social communication and overall ASD-related concerns at both ages. At 12 months, significantly more mothers of non-diagnosed high risk infants than low risk infants report restricted and repetitive behaviors and ASD-related concerns; significantly more parents of diagnosed high risk infants than parents of low risk infants report language and ASD-related concerns. At no age do significantly
more parents of diagnosed infants report concerns than parents of non-diagnosed high risk infants.

The fact that elevated levels of concern were not significantly higher in the group of infants later diagnosed with ASD, even at 12 months, is also consistent with the existing literature on laboratory-based concerns in high risk samples. Although Ozonoff and colleagues (2009) found that the presence of at least one parental concern at 12 months of age helped to predict diagnostic outcome, 12-month concerns were not a robust predictor of later classification. Hess and Landa (2012) reported on parent concerns collected at 14, 24, and 36 months and found that parental concerns did not distinguish high risk diagnosed and non-diagnosed infants until 24 months of age.

The data presented here suggest similarly limited predictive value for mothers’ daily concerns in the first year of life. However, the primary goal of the home-based methodology used here was not to predict infants’ diagnostic outcomes, but to better understand the factors that contribute to mothers’ subjective experiences of concern on a day to day basis. Rather than solely reflecting infant behaviors, maternal concerns at 6, 9, and 12 months reflect both broad and specific characteristics of the high risk population. Across the entire high risk sample, we found that the number of high risk mothers reporting concerns in our home diary measures ranged from 32% to 48% at each of the three ages studied here. Although there were moderate correlations with infant behavior at 12 months of age, 12-month maternal concerns were also associated with the severity of the proband’s ASD symptoms. This is in contrast to previous analyses of laboratory-based concerns which found no association between parental concerns and
The association between maternal concerns and proband severity (Ozonoff et al., 2009). The kinds of concerns mothers report on home-based diary measures may be qualitatively different than the kinds of concerns than those reported in a laboratory or clinic setting (Hess & Landa, 2012; Ozonoff et al., 2009). Maternal concerns at 12-month were also associated with mothers’ concurrent depressive symptoms, such that mothers with more concerns also reported high levels of depressive symptoms. It is not clear from these data whether increased depressive symptoms result in increased concerns or increased concerns result in increased depressive symptoms, but our findings indicate that maternal psychological states are related in some way to their daily experiences as a parent of a high risk infant sibling.

At 6 and 9 months of age, maternal concerns appear to reflect more broad effects of risk status, rather than specific maternal or family characteristics. These broad effects likely represent parents’ knowledge of their infants’ increased risk for ASD and increased hypervigilance and anxiety regarding their development. Although this early hypervigilance does not seem to accurately reflect infants’ behavioral symptoms it is not unwarranted given both the high recurrence risk for ASD and the frequency of language and communication difficulties across the entire high risk sample (Gamliel et al., 2009; Ozonoff et al., 2014; Ozonoff, Young, et al., 2011). A limited number of investigations have reported on changes in maternal behavior during dyadic interactions in the first year of life, including increased gesture production and a more directive interaction style (Talbott et al., 2013; Wan et al., 2012, 2013). These changes were hypothesized to reflect
the increased hypervigilance observed in the home-based diaries analyzed here, but this hypothesis has not yet been explicitly studied. Future investigations will be needed to determine whether and to what extent maternal concerns result in meaningful changes in maternal behavior. It is important to note that the extent to which increased early concern characterizes families of high risk infant siblings who do not choose to enroll in prospective, longitudinal investigations may differ significantly from families that seek out participation. Further investigation into the characteristics of families who do not enroll in these studies will be needed to determine the generalizability of early hypervigilance in high risk mothers.

In sum, the findings presented here suggest that mothers of high risk infants are not only more frequently concerned about their infants' development, but that their experiences as parents of children with ASD influences the day to day concerns they experience during their infants’ first year of life. The relations between mothers’ early concerns and infant, maternal, and family characteristics highlight the need to examine high risk infants’ development in the family context as it is possible some of these same family factors, including parents’ concerns, may influence early parent-child dyadic interactions.
CHAPTER 3: STUDY 2: Maternal Prompting of Social Communication

Infant siblings of children with autism (ASD) are themselves at increased risk for the disorder relative to the general population (CDC, 2014; Ozonoff, Young, et al., 2011). In addition to elevated risk for ASD, as a group, high risk infants also display a variety of impairments in social attention and reciprocal communication during the first few years of life. At 12 months they are rated as less attentive to their mothers during laboratory-based parent-child interactions (Wan et al., 2013). In the second year of life they require more redundant prompts than low risk infants to use an experimenter’s social cues to successfully orient to a target (Presmanes, Walden, Stone, & Yoder, 2007). Mothers of high risk infants demonstrate differences in behaviors broadly related to attention regulation in the first year of life, including gesture production and overall directiveness (Talbott et al., 2013; Wan et al., 2013). These changes in maternal behavior have been hypothesized to potentially reflect maternal hypervigilance regarding their infants’ increased risk and reactions to infants’ emerging social difficulties. However, the extent to which these broad changes in maternal behavior are observed specifically in the prompting strategies mothers of high risk infants use to elicit social responses from their infants and whether any changes in maternal prompting behavior reflect responses to infants’ attentional difficulties or maternal characteristics has not yet been studied. The current study examines these issues by scoring maternal prompting of infant social communication during parent-child interactions filmed in the home when infants were 12 months of age. In addition to evaluating whether mothers of high risk infants rely on different prompting strategies for eliciting social communication from their infants,
associations between mothers’ prompts and both infant behavior and maternal characteristics hypothesized to influence their behavior were also assessed.

**Response to Social Bids in Young Children with ASD and High Risk Infant Siblings**

Both young children with ASD and infant siblings of children with ASD demonstrate difficulty in responding to a social partner’s bids for attention (for a review see Bruinsma, Koegel, & Koegel, 2004). These include difficulty with both responding to laboratory-based discrete trial joint attention tasks (e.g. following an experimenter’s point towards a target) as well as more global changes in social responsiveness.

These difficulties in both specific and broad measures of social responding have been observed in both later-diagnosed and non-diagnosed high risk infant siblings. At 12 months, high risk infants later diagnosed with ASD are rated as less attentive to their parents and score lower in ratings of dyadic mutuality during laboratory-based parent-child interactions than both non-diagnosed high risk and low risk infants (Wan et al., 2013). As a group (without differentiating between high risk infants who are and are not later diagnosed with ASD), 12-to-23-month old high risk infants are less successful at following an experimenter’s bids to locate a target amongst an array of objects when those bids are relatively simplistic (e.g. calling the infants’ name and turning the head in the direction of the target), but are just as successful as low risk control infants when bids include redundant clues (e.g. adding a point in addition to a name call and head turn; Presmanes et al., 2007). Impairments were also reported in a longitudinal study modeling infants’ abilities on discrete trial joint attention tasks from 8 to 18 months (Ibañez, Grantz, & Messinger, 2013). Infants’ initiation of joint attention (IJA), responses to joint
attention (RJA) and initiation of requesting behaviors (IBR) were measured at 5 points across this period. As a group, high risk infants exhibited significantly lower levels of both initiation of and responses to joint attention at 8 months. When the high risk infants who later were classified as meeting criteria for ASD were excluded, only responses to joint attention were significantly lower in the non-diagnosed high risk infants compared to the low risk controls (Ibañez et al., 2013).

In the second year of life, toddlers with ASD shift their attention less frequently between people and objects and ignore or actively reject their mothers’ bids for joint attention episodes more frequently than typically developing toddlers (Adamson, McArthur, Markov, Dunbar, & Bakeman, 2001; Swettenham et al., 1998). These difficulties are particularly noticeable for episodes of coordinated joint attention, when the child explicitly engages with a social partner and objects or events, versus joint attention episodes where the child is sharing in objects or events without explicitly engaging or acknowledging the social partner (Adamson, Bakeman, Deckner, & Romski, 2009).

Together, the literature on joint attention in both young children with ASD and high risk infant siblings suggests that impairments in initiating social interactions may be more specific to infants eventually diagnosed with ASD, while impairments in responding to a partner’s social bids may characterize high risk infants as a group, including those who are not later diagnosed with ASD. Notably, several studies have documented changes in the attention regulating behaviors of mothers of both toddlers with ASD and high risk infant siblings that have been hypothesized to reflect, at least in
part, mothers adaptations to infants’ difficulties in this area (Adamson et al., 2001; Talbott et al., 2013; Wan et al., 2012, 2013). The following sections review the existing literature on these maternal behaviors in the domain of joint attention, broadly defined to include not only discrete cues prompting shifts in attention (analogous to discrete laboratory-based joint attention trials) as well as more broad initiation and maintenance of joint engagement episodes, where mothers and infants are interacting with a shared focus of attention (Adamson et al., 2001).

**Attention Regulating Behaviors of Mothers of Children with ASD and High Risk Infant Siblings**

Attention regulating behaviors in mothers of children with ASD. Mothers of toddlers with ASD make bids for their child’s attention just as frequently as mothers of typically developing toddlers, but tailor the content of their bids for joint attention to be more concrete and perceptual (e.g. tapping on an object) rather than solely conventional (e.g. using gestures alone) (Adamson et al., 2001). Differences in the frequency of mothers’ attention bids are only seen in specific contexts – they produce fewer bids when joint engagement interactions revolve around commenting (i.e. looking at pictures), but not interactions centered around requesting (object-based interactions) or purely social interactions (turn taking) (Adamson et al., 2001). Mothers of young children with autism also use more touching and other regulatory behaviors compared to mothers of typically developing infants beginning in the first 6 months of life. These increased regulatory behaviors are observed particularly when mothers are attempting to up-regulate or stimulate their infants, rather than calm them (Saint-Georges et al., 2011).
Attention regulating behaviors in mothers of high risk infant siblings.

Mothers of high risk infant siblings also demonstrate behavioral changes in attention regulating behaviors. Talbott and colleagues (2013) found that mother of non-diagnosed high risk infants use significantly more gestures than mothers of low risk, typically developing infants. Notably, this was not the case for mothers of later-diagnosed infants, who themselves produced significantly fewer gestures than low risk infants. Mothers of high risk infants are rated as being more directive and showing less sensitive responding on global measures of laboratory-based parent-child interactions when infants are 6 to 10 months of age (Wan et al., 2012). At 12 months, group differences in directiveness are driven specifically by parents of infants later diagnosed with ASD (versus high risk infants not diagnosed), who were rated as significantly more directive than mothers of low risk infants. Simultaneously, infants later diagnosed with autism were rated as less attentive to their parents and scored lower in dyadic mutuality than both non-diagnosed high risk and low risk infants (Wan et al., 2013). These results suggest that early directiveness may be more characteristic of high risk parents as a group, but that beginning near the end of the first year of life, this directiveness may begin to reflect infants’ behavioral symptoms.

An outstanding question is the extent to which these observed changes in maternal behavior, particularly for mothers of high risk infants, reflect their sensitivity to infants’ difficulty responding to social bids. If this were the case we would expect that mothers of high risk infants’ who were exhibiting the most significant difficulties might increase their communicative support by producing the most frequent or redundant prompts,
similar to the type reported by Presmanes and colleagues (2007). The gesture findings of Talbott and colleagues (2013) suggest this is not always the case, as increased rates of maternal gesture production were observed only for mothers whose infants were not impaired in gesture production at 12 months of age. In the domain of gesture then, a boost in maternal gesture production seems to reflect more broad effects of risk status or other maternal characteristics, rather than reflecting infants’ communicative difficulties. The relative impacts of both maternal and infant characteristics on maternal behavior may vary not only depending on whether or not infant symptoms are present, but also across domains of social communication and at different points in time. For example, while mothers of non-diagnosed but not later diagnosed high risk infants gesture more frequently, the opposite pattern is observed for broad ratings of maternal directiveness at 12 months of age, where directiveness is increased in mothers of later-diagnosed infants, who themselves are demonstrating attentional difficulties.

Together, these initial studies on the behavior of mothers of high risk infants highlight the need to examine both infant and maternal characteristics that contribute to dyadic interactions in the first year of life. Given high risk infants’ particular difficulties in responding to social bids, the current study focuses on mothers’ strategies for eliciting social communication from their infants as a context for examining both infants and mothers unique contributions to these interactions. In addition to describing relations between infant and maternal behavior, we have examined additional maternal and family characteristics of high risk samples that may contribute to changes in maternal behavior. Previous investigations have suggested that some of the observed changes in the behavior
of high risk mothers (increased gesture production and directiveness) reflect maternal characteristics and other factors associated with their high risk status. These include the presence of depressive symptoms or broader autism phenotype characteristics, both of which are elevated in mothers of children with ASD, parenting strategies adapted from experiences parenting an older diagnosed child, or increased early hypervigilance and concern regarding infants’ elevated risk of ASD, but these possibilities have not been explicitly addressed (Ozonoff et al., 2009; Talbott et al., 2013; Wan et al., 2012, 2013).

The goals of the current study were to determine whether mothers of infant siblings of children with autism differed from mothers of low risk infants in the rate or types of prompts (attention bids) they provided to their infant during a brief semi-structured, home-based task at 12 months of age and whether the strategies employed by high risk mothers were related to infant behavior, mothers concurrent home-based concerns, depressive symptoms, maternal broader autism phenotype features, or the severity of the older diagnosed child’s ASD symptoms. This was accomplished in the context of an ongoing longitudinal study of infants at risk for autism, but focused on data collected in the naturalistic setting of the home, rather than the laboratory.

Methods

Participants

Participants included 27 infant siblings of children with autism and 32 low risk control infants and their mothers. These families were participating in an ongoing longitudinal study of infants at risk for autism conducted jointly at Boston Children’s Hospital and Boston University. For the larger project, interested families were contacted
by the study coordinator, who conducted a detailed telephone eligibility interview. All subjects were screened for exclusionary criteria (prematurity, extended stays in the neonatal intensive care unit, maternal drug or alcohol use during pregnancy, family history of genetic disorders associated with ASD, and primary languages other than English). Infants were enrolled into the high risk autism group (HRA) if they had an older sibling with a diagnosis of Autism, Asperger’s Syndrome, or Pervasive Developmental Disorder-Not Otherwise Specified (PDD-NOS), confirmed by expert community diagnosis. Infants were enrolled into the low risk control (LRC) group if they had at least one older sibling who was typically developing and no first-degree relatives diagnosed with an ASD or other neurodevelopmental disorder. The sample was well matched for gender (55% female) and was primarily Caucasian (8.5% non-Caucasian) and high SES, with the majority of mothers in each group having at least a college degree (8.5% less than a college degree) and an income over $75,000 (23.7% less than $75,000). There were no significant group differences in the gender ratio, infant race, maternal education or family income. Informed consent was obtained from parents prior to participation.

**Procedure**

As part of the larger longitudinal study, infants were seen in the laboratory several times from 3 to 36 months of age where they participated in a range of standardized behavioral assessments and neurophysiological paradigms. In addition to these laboratory visits, families were asked to provide both written and home diary measures from 6 to 18 months of age. Because these diary measures were completed primarily by mothers, all parent measures are hereafter referred to as maternal measures. Video diaries were filmed
monthly and consisted of semi-structured interactions between infants’ and their mothers which lasted approximately 20 minutes. Mothers were instructed to present infants with a series of toys, play social games, attempt to elicit vocal imitation and smiles, read a picture book, and play for several minutes ‘in whatever way makes [them] feel most comfortable.’ Written diaries were completed weekly, and consist of 8 items. Parents were asked to report on new sounds, words, or gestures their infant made that week, to describe infants’ play with their parents, a sibling, and alone, and describe any concerns about their infants’ development. Video and Written diary measures were scored by coders blind to group membership and trained extensively on the coding schemes (described below).

The focus of this study involves a subset of laboratory and home-based measures collected at 12 months of age:

**Laboratory Based Measures**

**Autism Observation Scale for Infants (AOSI; Bryson et al., 2008).** The AOSI is an 18-item assessment that measures a range of autism-related behaviors (visual attention and tracking, social interest and reciprocity, affect, atypical sensory and motor behaviors, etc.) during a brief semi-structured interaction between a trained examiner and the infant, who is seated on their parents’ lap. Individual items are scored from 0 to 2 or 3, with higher scores indicating greater atypicality. The scale yields two final score: the total number of items endorsed, and the total raw score (out of a possible 50). Here, infants’ AOSI Total Scores were used as a measure of emerging autism symptoms at 12 months of age.
Autism Diagnostic Observation Schedule (ADOS; Lord et al., 2000). The ADOS is a semi-structured play-based interaction designed to assess participants’ social and communicative abilities across a range of contexts which vary according to language ability. The presence of repetitive behaviors and restricted interests are also noted. Individual items are scored from 0-3, with higher scores indicating more profound impairment. The items in the scoring algorithm map onto DSM-IV criteria for ASD, and empirically-derived cut-offs can be used to categorize scores into those meeting criteria for Autism, Autism Spectrum, or non-spectrum. For the current study, ADOS scores are used to classify infants into preliminary diagnostic groups, along with a clinical best estimate judgment. The ADOS was administered at 18, 24, and 36 months of age.

Questionnaire Data

Family SES information. Basic demographic information was collected upon entry to the study and included: race and ethnicity for each parent, proband, and infant, maternal and paternal education, and family income.

Maternal broader phenotype characteristics. The presence of broader autism phenotype symptoms in mothers was assessed using the Broad Autism Phenotype Questionnaire (BAP-Q; Hurley, Losh, Parlier, Reznick, & Piven, 2007). The BAP-Q is a 36-item questionnaire that assesses behavior across three subscales: aloof, pragmatic language, and rigidity. It is collected once from mothers upon entry to the study. BAP-Q Total Scores were used here as a measure of broader phenotype features in mothers.

Maternal depression measures. The presence of depressive symptoms in mothers was assessed using a modified version of the Center for Epidemiologic Studies
Depression Scale – Revised (CESD-R; Eaton, Smith, Ybarra, Muntaner, & Tien, 2004)
The CESD-R was collected from mothers when infants were 12 months of age. The items related to suicidal thoughts and self-harm were removed, so final scores are the total number of items endorsed out of a possible 18 points. Here, mothers Total CESD-R scores were used as a global measure of symptom presence, rather than identifying clinical diagnoses.

**Video Diary Coding**

**Toy drop task.** From the larger video diary behavioral battery, the toy drop task was selected for analysis in the current study for several reasons. The first is that it provided a relatively standard context for assessing both infant and maternal behavior across families. Families were asked to film the entire toy play section of the video diaries with infants seated in a highchair or at a table, and filming tended to be of higher quality for this section than for the later social interaction portion of the video diary. Finally, rather than the entire toy play section of the diaries, the toy drop task was specifically designed to elicit both prompting from parents and social responses from infants, the goal of this study.

**Maternal scaffolding** Maternal behaviors hypothesized to promote infants’ social communication were scored during the toy drop task, beginning at the time the mothers held infants’ preferred toy out of reach and ending when the toy was returned to the infant or placed back in the bag (e.g., not returned). These categories of maternal responses were selected based on previous literature on mothers’ social responses and maintenance of joint attention episodes, and pilot coding of mothers’ spontaneous
prompts. Scored maternal behavioral prompting categories include: Questions, Requests, Comments, Gestures, and Gesture +Vocal Combinations, which included instances where gestures and any of the 3 verbal prompts co-occurred, either simultaneously or within a 2 second window. Examples of each of these codes are included in Table 3.1.

Table 3.1 Descriptions and examples of maternal contingent response codes

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<th>Code</th>
<th>Description</th>
<th>Examples</th>
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| Question| Maternal questions directly related to the toy or to the infants’ mental states (what the infant wants, sees, etc.). | • “Can you find the wand?”
|         |                                                                             | • “What do you want?”
|         |                                                                             | • “Do you want the car?”                                                                         |
| Request | Maternal verbal prompts for infant behavior                                 | • “Show me what you want.”
|         |                                                                             | • “Point to the car”                                                                               |
| Comment | Maternal verbalizations that are directly related to the object or the infant, but are neither a question nor a request | • “Oh, I see the car!”
|         |                                                                             | • “Yes, you are sad I took it away.”                                                                 |
| Gesture | Maternal gestures related to the dropped toy, including iconic gestures, deictic gestures, and signs. | • Points to the dropped toy
|         |                                                                             | • Makes a gesture representing the rattle                                                          |
|         |                                                                             | • Gestures to indicate the object was dropped or thrown from the tabletop.                         |

To account for differences in overall task length, a Total Prompting Score was created for each mother that represents the rate of all prompting behaviors per minute. A relative proportion score was created for each of the 5 prompt types (Question, Request, Comment, Gesture, Gesture +Verbal Combinations) by dividing the total number of prompts within a single category by the raw summed total number of prompts.

Infant social communication (toy drop task). Infant communicative behaviors were scored from the same toy drop task. The categories of scored behavior were selected
based on previous literature suggesting high risk infants’ may have particular difficulty with these social communicative behaviors at 12 months of age. Coded infant behaviors included:

1. Vocal (all verbal and vocal utterances, but not vegetative sounds like laughing or coughing)
2. Gesture (all points, reaches, or other communicative gestures)
3. Gesture + Vocal Combinations (instances of codable gesture and vocalizations which occurred simultaneously or within a 2 second window).

To account for differences in overall task length, a Total Communication Score was computed for each infant that represented the rate of all communicative behaviors per minute. Relative proportion scores for each of the 3 communication types (Vocal, Gesture, Gesture + Vocal Combinations) were calculated by dividing the total number of prompts within a single category by the summed raw total number of scored behaviors.

**Written Diary Coding**

**Maternal concerns.** Concerns reported in weekly home-based written diaries were scored across the following categories: General/Medical (illness, teething, etc.), Language, Social Communication, and Restricted and Repetitive Behaviors. The Language, Social Communication, and Restricted and Repetitive Behavior scores were collapsed into a single Total Autism Concerns Score. The coding scheme and procedures are described in detail in Chapter 2. For the current study, mothers Total Autism Concerns reported from 11-13 months were used as a measure of their concerns about their infants’ ASD symptoms at 12 months of age.
Results

Overview

Toy drop task sessions were an average of 35.3 seconds, and were filmed when infants were an average of 52.7 weeks of age. HRA infants were on average, 1.9 weeks younger than LRC infants, a difference that was statistically significant \( t(57) = -1.626, p = .03 \). Task sessions were an average of 12.6 seconds longer for HRA infants, which was also statistically significant, \( t(57) = 2.150, p = .04 \). In order to address our aims of describing the frequency and types of prompts mothers are providing to their infants, and the relationships between these prompts and both infant behavior and maternal characteristics, a series of analyses were conducted. The first set focused on describing the frequency and type of infant communication and any group differences in these features. The second set described these same features of maternal prompting strategies. Due to the significant positive skew exhibited by the coded maternal and infant variables, nonparametric analyses were used for these first two sets of analyses. The final set of analyses compared the relationship between these variables and parental concerns, infant autism symptoms, and other maternal and family characteristics hypothesized to contribute to maternal prompting strategies and rates. For these analyses, coded variables were transformed using a log transformation and evaluated using Pearson correlations.

Diagnostic Outcomes

Diagnostic group membership was determined using both ADOS scores at infants’ most recent visit and clinical judgments for infants for whom their most recent visit was their final visit (either at 36 months or if they had discontinued participation
after the 18 or 24 month visit). Inclusion in the ASD group required meeting criteria for ASD on the infants’ most recently completed study visit, and a confirmation of ASD from a clinical best estimate judgment made for infants at their final study visit. 6 HRA infants met these criteria for ASD. All met ADOS criteria, and 5 received clinical judgments of ASD (3 from 36 month data, and 2 from 18 month data). The last infant met ADOS criteria at their 24 month visit, but has not yet been seen at 36 months. Together, these 6 infants are hereafter referred to as the ASD group. HRA infants who did not meet ADOS criteria for ASD are hereafter referred to as HRA-N.

**Infant Communication Rates**

Across both groups, infants produced an average of 7.8 communicative acts per minute (range = 0 – 15.7). There were no significant differences between the groups for this overall rate ($t(57) = .385, p = .70$). To test whether high risk infants were slower to produce their first communicative act, the average latency (in seconds) to infants’ first communication was compared between the two groups. There were no significant differences between the groups (LRC: Mean= 8.10, SD= 7.0; HRA: Mean= 7.78, SD= 3.78; $F(1,56) = .041, p = .84$).

In order to determine whether infants differed in the distribution of behaviors used, we used non-parametric analyses (Mann-Whitney U) to compare the relative proportion of each communication type (Vocal, Gesture, Gesture+Verbal Combinations). These relative proportions are presented in Figure 3.1. The majority of communicative acts produced by both groups of infants were Vocal. There were no significant group
Figure 3.1 Relative proportions of infant social communicative acts, by type. Error bars represent standard error.
differences for Vocal (mean\textsubscript{LRC} = .48, SD = .33; mean\textsubscript{HRA} = .55, SD = .31, U = 337.0, \( p = .49 \)) or Gesture+Vocal Combinations (mean\textsubscript{LRC} = .16, SD = .18; mean\textsubscript{HRA} = .23, SD = .31, U = 356.5, \( p = .70 \)). There was a trend for HRA infants to produce relatively fewer Gesture acts (mean\textsubscript{LRC} = .36, SD = .31, mean\textsubscript{HRA} = .22, SD = .27, U = 486.5, \( p = .06 \)).

This pattern of non-significance remained unchanged when comparing the total communication rate, latency to infants’ first communication, and each of the relative proportion scores when comparing infants’ in the ASD, HRA-N, and LRC groups. The trend-level significance observed for infants’ Gesture scores between the HRA and LRC infants dropped to non-significance when comparing the ASD, HRA-N, and LRC infants.

**Maternal Prompting Strategies**

Mothers produced prompts at an overall average rate of 14 prompts per minute (range = 2.7 – 40.0). There was no significant difference in the overall prompting rate between the two groups, (HRA: mean = 13.7, SD = 7.6; LRC: mean = 14.0, SD = 5.2; U = 504, \( p = .273 \)). To test whether mothers in the groups differed in the types of prompts used, we analyzed the distribution of each prompt type by comparing the relative proportion of each prompt type between the groups. Descriptive information on these relative proportions of each type is presented in Figure 3.2. There were no significant group differences in the mean proportions for Questions, Comments, Requests, Gestures, or Gesture + Verbal combinations (all \( p \)’s > .25).
Figure 3.2 Relative proportions of maternal prompts, by type. Error bars represent standard error.
To determine whether the two groups differed in the number of prompts mothers provided prior to the infants’ first communicative act, an ANOVA was used to compare the mean number of prompts used by each of the two groups. Due to significant positive skew, this variable was transformed prior to analysis. The two groups did not differ significantly in the total number of prompts produced prior to the infants’ first communicative behavior (LRC: mean = .45, SD = .27; HRA: Mean = .38, SD = .23, F (1, 56) = 1.305, p = .30.

These same analyses of maternal prompting behavior were conducted separating the groups by infants’ diagnostic outcome. This pattern of non-significant group differences in mothers’ Total Prompting rate, the relative proportion of each prompting type, and the average number of prompts produced prior to infants’ first communicative behavior remained unchanged.

**Interrelations Among Maternal Prompting, Infant Communication, and Maternal and Family Characteristics**

Coded variables (Maternal Total Prompts, Infant Total Communication, and Maternal Autism Concerns) were transformed using a logarithmic transformation prior to analyses. We first examined the relationships between Maternal Total Prompts and Infant Total Communication. Pearson’s zero-order correlations were calculated both for the entire sample and separately for the HRA and LRC groups. None of these associations were significant (all p’s > .44). We next focused on analyzing the relationship between maternal prompts and the maternal and family characteristics of interest. Interrelations between Maternal Total Prompts, Maternal Autism Concerns, Maternal Depression
Symptoms, Maternal Broader Autism Phenotype characteristics, Proband Autism Symptoms, and Infant Autism Symptoms were calculated for the HRA sample only.

These analyses focused on the entire group of high risk infants, rather than separately for the non-diagnosed and diagnosed infants. This approach was selected primarily because we were interested in associations with infants’ autism symptoms and separating the two groups of HRA infants into diagnostic groups would be redundant with autism symptoms. Of the 27 HRA infants included in the video diary analyses, written diary data (Maternal Autism Concerns) were available for 18 families, Maternal Depression Symptoms were available for 26 families, Broad Autism Phenotype Symptoms (BAP-Q) were available for 20 families, and Infant and Proband Autism Symptoms were available for 25 families. The only significant correlation was between Maternal Total Prompts and Maternal Autism Concerns ($r = -.57, p = .013$). There were trend-level associations between Maternal Total Prompts and Infant Autism Symptoms ($r = -.40, p = .06$) and Infant Autism Symptoms and Maternal Autism Concerns ($r = .45, p = .07$). The significant association between Maternal Total Prompts and Autism Concerns remained even after controlling for concurrent Infant Autism Symptoms ($r = -.50, p = .04, n = 15$). As a final check, the mean number of prompts produced by high risk parents who had reported at least one concern and parents who had not reported any concerns was compared. As a group, high risk mothers who had reported at least one concern produced significantly fewer prompts than mothers who had reported no concerns, $F(1,17)= 4.673$, $p = 0.046$. This data is presented in Figure 3.3.
Figure 3.3 Mean rate of overall prompts by mothers reporting at least one concurrent concern about autism symptoms in their infant and parents reporting no such concerns. Error bars represent standard error.

Discussion

The goals of this study were to describe the strategies mothers of infants at risk for autism use to elicit social communication from their infants and determine whether these strategies differed from parents of low risk infants in either their frequency or type. We were also interested in exploring the relations between the high risk mothers prompting strategies and maternal and family characteristics previously hypothesized to underlie some of the behavioral changes reported in other domains of parent behavior. These characteristics include infant behavior (responses to social attention bids) but also include maternal concerns regarding their infants’ development, depressive symptoms, broader autism phenotype characteristics, and the severity of their older diagnosed child.

We found that overall, mothers of high risk infants provided the same frequency of prompting to their infants as parents of low risk infants, and used the same repertoire
of strategies when doing so. However, within the group of high risk mothers, those who expressed concurrent concerns regarding their infants’ autism symptoms provided fewer prompts than parents reporting no concerns. This effect was not driven by the subset of high risk infants later diagnosed with autism, as mothers of these infants used just as many prompts as mothers of both non-diagnosed high risk infants and typically developing infants. These findings suggest that rather than increasing their scaffolding behaviors, mothers reduce the frequency of their prompting when they have concerns about autism symptoms.

It is possible that the decreases in the prompting behavior of high risk mothers with concerns reflect responses to infants’ decreased social responsiveness that we were simply not able to capture in this brief task. Infants may demonstrate difficulties on the kinds of discrete trial RJA tasks usually tested in the laboratory or other more global difficulties in responsiveness that mothers are picking up on. However, even if mothers were more sensitive to these kinds of difficulties than the coding scheme used here, we would expect these difficulties to be more significant in the group of later diagnosed high risk infants than infants who are not diagnosed. The fact that differences in maternal prompting rates were seen in mothers who expressed concerns, rather than mothers of infants later diagnosed with ASD provides further supports the interpretation that these decreases in maternal prompts were driven primarily by maternal, rather than infant, characteristics.

The results presented here have relevant clinical implications. If parents who believe their infants are less likely to respond (indicated by their concerns about the
presence of ASD symptoms) provide fewer opportunities for them to do so, they may inadvertently provide their infants with less opportunity for social interaction that contribute to later difficulties with social responding. While the differences between high risk mothers in our sample were quite minimal, we did not find any significant group differences in infant behavior. Infants who exhibit more overt difficulties may exert stronger influences on maternal behavior, leading to fewer maternally-initiated interactions and subsequently more striking difficulties in infants. To answer these questions, future investigations will need to examine these issues in infants demonstrating overt difficulties with social responding.

Overall, our results suggest that prior to the onset of infant symptoms in the domain of social responsiveness, mothers of high risk infants do not differ from mothers of low risk infants in the type or frequency of prompts they are using to elicit social communication from their infants. Similarly, high risk infants use the same type and frequency of social communication during these semi-structured home-based interactions. These findings suggest that changes in maternal behavior in this domain are not likely to drive some of the difficulties in social responding previously reported in laboratory-based investigations of high risk infants’ social communicative abilities. While future work is needed to determine if the kinds of maternal prompts studied here are altered after the onset of infant symptoms, the data presented here demonstrate that risk status may in some ways act as a protective factor, as mothers of high risk infants provide largely the same input and communicative contexts for their high risk infants as mothers of low risk infants.
CHAPTER 4: STUDY 3: MATERNAL FEEDBACK TO INFANT VOCALIZATIONS

Many studies have highlighted the importance of early social interactions in shaping the language development of young typically developing children. In particular, previous studies have demonstrated that contingent social feedback is associated with specific features of infants’ language abilities such as consonant production and phonemic discrimination. The extent to which laboratory-based measures of maternal contingent feedback reflect the daily interactions of the home, whether maternal input provided in the home is related to infants’ language development more broadly, and whether either maternal input or its relation to infants’ language development varies across clinical populations has not been well studied. The current study fills these gaps in the literature by examining links between maternal contingent responses to infants’ vocalizations and infants’ later language abilities, in both typically developing infants and those at increased familial risk for autism spectrum disorders (ASD). Infant siblings of children with autism are of particular interest because as a group, they are at risk for impairments in key domains of interest: early consonant production, broad delays in language development, and reciprocal social interactions. The extent to which maternal input influences high risk infants’ early language production has not been examined, though the current literature on typically developing dyads would suggest that changes in maternal input may contribute to some of these observed early delays. We begin by providing a summary of the relevant literature on maternal and environmental influences on the language development of typically developing infants.
The Influence of Social Feedback on Language Development in Typically Developing Infants

The influence of contingent social interactions on early language development is illustrated by the literature on perceptual narrowing, the process whereby infants become more adept at discriminating phonemes used contrastively in their native language while simultaneously losing the ability to discriminate between variants that are not used contrastively, such as those found in their non-native languages (Werker & Tees, 2002). This narrowing is thought to reflect infants’ ability to use statistical regularities in their linguistic environment to identify meaningful phonemic contrasts, but it is modulated by social interaction, such that mere passive exposure to non-native contrasts is not enough to maintain the ability to discriminate between them over the first year of life (Saffran, 2003; Tsao, Liu, & Kuhl, 2004). Rather than passive exposure, contingent, reciprocal social interaction is necessary to maintain the discrimination of phonemic contrasts.

Kuhl, Tsao, & Liu (2003) demonstrated this ‘social gating’ of language learning by exposing 9-month-old native English speaking infants to 5 hours of Mandarin Chinese via either live interaction with a native Mandarin speaker, or via auditory-visual or auditory-only DVDs. Only infants who received non-native exposure through live interaction regained the ability to perceive a Mandarin phonemic contrast. More recently, Elsabbagh and colleagues (2013) showed that the influence of environmental input on perceptual narrowing does not simply require live social interaction – the amount of linguistic exposure is related to the timing of this developmental process. They tested infants’ native and non-native phonemic discrimination at 6 and 10 months of age and
found that while all infants had lost the ability to discriminate the non-native contrasts by 10 months of age, infants whose mothers were rated as highly contingent during a laboratory-based mother-child interaction showed even earlier perceptual narrowing, failing to discriminate the non-native contrasts at 6 months of age. Infants who receive impoverished early language exposure demonstrate persistent difficulties with phonemic discrimination even in childhood (Nittrouer & Burton, 2005).

The facilitating effect of social interaction applies not only to infants’ phonemic perception (as outlined above) but is also related to infants’ vocal production in the first year of life. When provided with contingent social feedback (social smiles and touching), 8-month-old infants respond by increasing the complexity of their early utterances, producing more canonical syllables than infants who receive the same frequency of feedback in a non-contingent manner (Goldstein, King, & West, 2003). Goldstein and Schwade (2008) experimentally tested the specificity of infants’ sensitivity to contingent linguistic feedback by coding the vocalizations of 9-month-old infants and their mothers. After a baseline period, mothers were instructed to provide one of 4 types of feedback: contingent fully resonant vowel responses, contingent responses containing consonants, or yoked control versions of each of these two experimental manipulations. For the control conditions mothers were remotely instructed to provide feedback at the same rate as a mother in either the resonant or consonant conditions but this feedback was unrelated to the timing of infants’ vocalizations. Infants in both contingent groups had significant increases in the rate of high quality (fully vs. quasi resonant vowels and consonant vs. vowel only) utterances relative to infants in the control conditions, who received the same
rate of feedback but in a non-contingent manner. Infants in the two contingent conditions increased the proportion of vocalizations unique to the specific type of feedback they had been provided with. Infants receiving high rates of fully resonant contingent responses significantly increased the proportion of fully resonant utterances they produced but not the proportion of consonant utterances; the reverse was true for infants receiving high rates of contingent consonant feedback from their mothers. Notably, infants did not increase the proportion of fully resonant or consonant productions by imitating their mothers’ exact content, but rather by imitating only the phonological form. The authors suggest these results demonstrate how social feedback can guide infants’ use of statistical regularities. The close temporal relationship between infants’ and mothers’ utterances highlights for the infants the underlying phonological structure of mothers’ vocalizations and makes the discrepancy between their own utterances more salient, thus facilitating infants’ production of more mature utterance forms.

Gros-Louis, West, Goldstein, & King (2006) conducted a more detailed analysis of a subset of the 8-month-old infants who participated in the Goldstein, King, and West 2003 study. Spontaneous infant vocalizations and mothers’ spontaneous contingent responses to these vocalizations were scored from a free play section of the laboratory visit (rather than the period of experimental manipulation). Infant vocalizations were categorized as either vowel or consonant-vowel utterances and maternal responses were categorized across a range of vocal and non-vocal response types. The majority of mothers’ spontaneous contingent responses were vocal. These vocal responses were further categorized as responses hypothesized to either promote language development
(acknowledgements, imitations, labeling and questions) or not to promote language development (attributes, directives, and play). Results indicated that mothers differentially responded to infants’ vowel and consonant-vowel utterances, responding to consonant-vowel utterances with more language promoting than non-promoting responses, while responding equally with both types of responses to infants’ vowel vocalizations (Gros-Louis, West, Goldstein, & King, 2006).

Together, the literature on typically developing infants demonstrates that a) infants respond to social feedback by increasing the frequency and complexity of their vocalizations, particularly for the specific type of feedback they receive, and b) mothers’ spontaneously differentially reinforce infants’ consonant-vowel over vowel-only utterances. While this pattern of differential responding to more developmentally advanced infant vocalizations has been hypothesized to shape early language development by fostering increasingly advanced language production, the relationship between maternal responses and infants’ later language have not been assessed. One goal of the current study was to determine whether mothers’ spontaneous responses to infants’ vocalizations at 9 months of age are related to infants’ language ability at 12 months. Rather than relying solely on laboratory-based data, these relationships were assessed using more naturalistic, home-based data that likely more accurately reflect the day to day language environment and dyadic interactions of infants and their mothers. Furthermore, these relationships between maternal contingent responses and infants’ language ability are examined not only in typically developing dyads, but in a sample of infants at increased risk for ASD. The following section reviews the relevant literature on
the early language development of high risk infant siblings and the role maternal input may play in the language development of this high risk sample.

**Infants at Risk for Autism**

Infant siblings of children with ASD are at increased risk relative to the general population for both a diagnosis of autism and a variety of social and language impairments. Delays in early language are one of the most striking early symptoms in children later diagnosed with ASD, and have been reported by 12 months of age for both retrospective and prospective samples (Mitchell et al., 2006; Ozonoff et al., 2010; Zwaigenbaum et al., 2005; see Jones, Gliga, Bedford, Charman, & Johnson, 2013 for a recent detailed review). Delays in the achievement of early linguistic milestones and atypical language trajectories have also been reported across the group of high risk infant siblings as a whole, both those later diagnosed as well as a substantial minority of those who are not (Gamliel et al., 2009; Iverson & Wozniak, 2007; Ozonoff et al., 2014; Paul et al., 2011). Notably, high risk infant siblings demonstrate delays in early consonant production, lagging behind low risk infants in the production of late consonants (those that emerge latest developmentally), producing canonical syllables less frequently at 9 months of age, and achieving the milestone of reduplicated babbling significantly later than low risk, typically developing infants (Iverson & Wozniak, 2007; Paul et al., 2011).

In addition to delays in specific early language milestones, high risk infant siblings demonstrate more global difficulties with language that emerge towards the end of the first year of life and persist throughout early childhood (Gamliel et al., 2009; Ozonoff et al., 2014). Systematic prospective investigations of high risk infant siblings...
have reported that at 12 months of age, infants later diagnosed with ASD score significantly lower than low risk infants on standardized language measures. Importantly, high risk infants who are not later diagnosed with ASD but are classified as demonstrating similar sub-clinical features of ASD (the broader autism phenotype, or BAP) also score significantly lower than low risk infants on these 12-month measures. No such differences are present at 6 months of age, suggesting that these deficits begin to emerge over the first year of life (Ozonoff et al., 2010, 2014). From 12 to 36 months of age, these language impairments become even more striking (Ozonoff et al., 2014).

The current study explores the hypothesis that these early-appearing language delays may be related to altered maternal linguistic input amongst mothers of high risk infant siblings. These changes in maternal input could arise from either maternal or infant characteristics. For example, if high risk infants initially produce fewer consonants, they may receive fewer contingent responses to them, even if mothers are responding with the same frequency as mothers of typically developing infants. Alternatively, mothers of high risk infants may use a different repertoire of responses or respond at different rates than mothers of typically developing infants that result in less robust reinforcement of infant vocalizations, negatively impacting infants’ language development from 9 to 12 months. The possibility that mothers of high risk infants may provide different vocal feedback than mothers of low risk infants is supported by previous investigations that have reported changes in maternal behavior during dyadic interactions in the first year of life for global ratings of directiveness and sensitive responding, as well as in the domain of gesture (Talbott et al., 2013; Wan et al., 2012, 2013). These changes have previously been
hypothesized to reflect both responses to infants’ emerging symptoms as well as specific characteristics of high risk mothers: as elevated levels of concern and hypervigilance, the presence of broader autism phenotype characteristics, or depressive symptoms, or the adaptation of behavioral strategies used with an older diagnosed child (Hess & Landa, 2012; Ruser et al., 2007; Talbott, Nelson, & Tager-Flusberg, n.d.). These same maternal characteristics may influence high risk mothers’ spontaneous responses to their infants’ vocalizations, but this has not been examined.

The Current Study

The goals of the current study were as follows: 1) to determine whether mothers of infants at risk for autism differ in the frequency or content of their responses to infants’ early vowel and consonant-vowel vocalizations; 2) to describe the relations between high risk mothers’ contingent verbal responses and both infants’ concurrent language production and maternal characteristics hypothesized to influence high risk mothers’ behavior; and 3) the extent to which this contingent feedback is associated with both typically developing and high risk infants’ later language abilities. These questions are answered in the context of a longitudinal study of both typically developing infants and infant siblings of children with ASD by scoring parent-child dyadic interactions that take place in the home at 9 months of age. Scores from these early home-based interactions were compared to infants’ language abilities measured in the laboratory with standardized behavioral assessment at 12 months of age.
Methods

Participants included 30 infant siblings of children with autism and 30 low risk control infants and their mothers. These families were participating in a longitudinal study of infants at risk for autism conducted jointly at Boston Children’s Hospital and Boston University. For the larger project, interested families were contacted by the study coordinator, who conducted a detailed telephone eligibility interview. All subjects were screened for exclusionary criteria (prematurity, extended stays in the neonatal intensive care unit, maternal drug or alcohol use during pregnancy, family history of genetic disorders associated with ASD, and primary languages other than English). Infants were enrolled into the high risk autism group (HRA) if they had an older sibling with a diagnosis of Autism, Asperger’s Syndrome, or Pervasive Developmental Disorder-Not Otherwise Specified (PDD-NOS), confirmed by expert community diagnosis. Infants were enrolled into the low risk control (LRC) group if they had at least one older sibling who was typically developing and no first-degree relatives diagnosed with an ASD or other neurodevelopmental disorder. The sample was well matched for gender (52% male) and was primarily Caucasian (13% non-Caucasian) and high SES, with the majority of mothers in each group having at least a college degree (8.8% less than a college degree) and an income over $75,000 (20.5% less than $75,000). There were no significant group differences in the gender ratio, infant race, maternal education or family income. Informed consent was obtained from parents prior to participation.
Procedure

As part of the larger longitudinal study, infants were seen in the laboratory several times from 3 to 36 months of age where they participated in a range of standardized behavioral assessments and neurophysiological paradigms. In addition to these laboratory visits, families were asked to provide both written and home diary measures from 6 to 18 months of age. Because these diary measures were completed primarily by mothers, all parent measures are hereafter referred to as maternal measures. Video diaries were filmed monthly and consisted of semi-structured interactions between infants’ and their mothers which lasted approximately 20 minutes. Mothers were instructed to present infants with a series of toys, play social games, attempt to elicit vocal imitation and smiles, read a picture book, and play for several minutes ‘in whatever way makes [them] feel most comfortable.’ Written diaries were completed weekly, and consist of 8 items. Parents were asked to report on new sounds, words, or gestures their infant made that week, to describe infants’ play with their parents, a sibling, and alone, and describe any concerns about their infants’ development. Video and Written diary measures were scored by coders blind to group membership and trained extensively on the coding schemes (described below).

The focus of this study involves a subset of laboratory and home-based measures collected at 9 and 12 months of age:

Laboratory Based Measures

Mullen Scales of Early Learning (MSEL; Mullen, 1995). The MSEL is a standardized developmental assessment designed to be used with infants from birth
through 68 months of age. It measures skills in Gross Motor and four cognitive domains: Fine Motor, Visual Reception, Expressive Language and Receptive Language. Here, Expressive Language T scores were used as a measure of infants’ language ability at 12 months of age.

**Autism Diagnostic Observation Schedule** (*ADOS; Lord et al., 2000*). The ADOS is a semi-structured play-based interaction designed to assess participants’ social and communicative abilities across a range of contexts which vary according to language ability. The presence of repetitive behaviors and restricted interests are also noted. Individual items are scored from 0-3, with higher scores indicating more profound impairment. The items in the scoring algorithm map onto DSM-IV criteria for ASD, and empirically-derived cut-offs can be used to categorize scores into those meeting criteria for Autism, Autism Spectrum, or non-spectrum. For the current study, ADOS scores were used to classify infants into diagnostic groups, along with a clinical best estimate judgment. The ADOS was administered at 18, 24, and 36 months of age.

**Communication and Symbolic Behavior Scale** (*CSBS; Wetherby & Prizant, 1993*). The CSBS is a semi-structured interaction between an examiner and an infant, designed to assess infants’ communicative and symbolic behavior repertoires. Infants are presented with a series of tempting toys, snacks, and symbolic play opportunities in order to elicit requests and social initiations, including gesture. The CSBS as a standardized measure demonstrates excellent reliability and validity. The sounds subscale inventories both the number and frequency of consonants produced throughout the interaction. The CSBS consonant inventory includes: /m/, /n/, /b|p|, /d|t|, /g|k|, /w|, /l|, /y|, /s|, and /sh|. 
For the purposes of the present study, the total number of these consonants infants produced was used as a measure of consonant production at 12 months of age.

**Questionnaire Data**

**Family SES information.** Basic demographic information was collected upon entry to the study and includes: race and ethnicity for each parent, proband, and infant, maternal and paternal education, and family income.

**Maternal broader phenotype characteristics.** The presence of broader autism phenotype characteristics in mothers was assessed using the Broad Autism Phenotype Questionnaire (BAP-Q; Hurley, Losh, Parlier, Reznick, & Piven, 2007). The BAP-Q is a 36-item self-report questionnaire that assesses behavior across three subscales: aloof, pragmatic language, and rigidity. It was collected once from mothers upon entry to the study. BAP-Q Total Scores were used here as a measure of broader phenotype features in mothers.

**Video Diary Measures**

Of the total video diary session, maternal and infant vocalizations were scored from the toy and book reading sections of the home video diaries. These sections were selected because they provided a more consistent context across families (rather than the free play section, which varied in terms of both the activities and presence of siblings or other family members). For each infant, diaries closest in age to 9 months but within the range of 8-10 months were selected for coding. The coding scheme used to analyze infant and maternal vocalizations was adapted from Gros-Louis et al. (2006), who scored infant and maternal vocalizations at the same age, but in the laboratory. This coding scheme
was selected in order to determine whether maternal and infant vocalizations occurring
during dyadic interactions in the laboratory are representative of the daily interactions of
the home, and whether the types of maternal responses previously hypothesized to
underlie infants’ language development are associated with infants’ 12-month language
abilities.

**Infant vocalizations.** Infant vocalizations that occur during the toy and book
sections of each diary were classified as either vowel or consonant-vowel vocalizations.
For vowels, quasi- and fully-resonant utterances were included, but vegetative sounds,
laughter, and crying were not. In order to control for differences in session length, infant
vowel and consonant-vowel data are expressed as the number of vocalizations of each
type occurring per minute.

**Maternal contingent responses.** Maternal vocalizations that occurred during the
toy and book sections of each diary were classified as either non-contingent or
contingent. Vocalizations were categorized as contingent if they occurred within 2
seconds of an infant vocalization and were directed at the same object, involved imitation
of the same sound, provided the label for the infants’ object of focus, etc.). These
contingent vocalizations were scored across the following categories (adapted from Gros-
Louis et al., 2006): Language Promoting (Acknowledgement, Imitation, Label, and
Question) and Non-Promoting (Attribute, Directive, and Play). Descriptions and
examples of each of these responses are included in Table 4.1. Because the rate of each of
these maternal responses depends on the number of vocalizations produced by the infant,
scores for the 7 individual response types and 2 summary codes were calculated as the
proportion of infant vocalizations receiving each type of response. The total number of maternal vocalizations (both contingent and non-contingent) was also scored in order to provide a measure of overall talk. This Maternal Total Utterance score is expressed as the rate per minute, in order to control for differences in session length.

### Written Diary Measures

**Maternal concerns.** Concerns reported in weekly home-based written diaries were scored across the following categories: General/Medical (illness, teething, etc.), Language, Social Communication, and Restricted and Repetitive Behaviors. The Language, Social Communication, and Restricted and Repetitive Behavior scores were collapsed into a single Total Autism Concerns Score. The coding scheme and procedures are described in detail in Chapter 2. For the current study, mothers Total Autism Concerns reported between 9 and 10 months are used here as a measure of concurrent
maternal concerns. Due to significant positive skew, this variable was transformed using a logarithmic transformation before analysis.

**Results**

Infants were on average, 9 months of age at the time of filming for the diaries included in this analysis, which did not differ by group (HRA: Mean= 8.93, SD=.78, LRC: Mean=9.10, SD= .66; \( t(58) = - .889, p = .38 \)). There were no group differences in the total video diary session duration, which were an average of 9.8 minutes, \( t(58)= .795, p = .43 \).

Six infants met criteria for ASD on the ADOS at their most recent study visit. Five of these were at 36 months and one at 18 months, all of whom also received expert clinical judgments of ASD. Although limited by the small number of these outcome infants (hereafter referred to as ASD), analyses below consider them separately from the high risk infants who were not classified as ASD; these non-diagnosed infants are referred to as the high risk negative (HRA-N) group.

In order to address our specific study goals, we first analyzed infant data to determine whether infants differed in their overall vocalization rate or by utterance type. These analyses were followed up with more detailed analysis of maternal responses to these vocalizations in order to better characterize the distribution of response types across the three groups and to determine whether mothers differed in their pattern of responding to different infant vocalization types. Finally, within the high risk group, associations between maternal vocalizations and maternal and family characteristics were analyzed.
Infant Language and Communication

9 month video diary data. Descriptive information on infant vocalization rates are presented in Table 4.2. To determine whether the three groups of infants differed the rate of vocalizations or the relative frequency of each type, a 3 (Diagnostic Group) by 2 (Vocalization Type) repeated measures ANOVA was performed. There were no significant main effects of Group or a Vocalization Type x Group Interaction (both p’s > .40). There was a significant main effect of Vocalization Type, $F(1, 57) = 62.11$, $p < .001$, indicating that infants in all groups produced significantly more Vowel than Consonant-Vowel Vocalizations.

Chi-square analyses were also used to examine the relative percentages of infants in each group who did not produce any consonants. There were no significant differences between the groups, with 22% of the total sample (6 LRC, 6 HRA-N, 1 ASD) producing no consonants ($\chi^2 (1, N = 60) = .295$, $p = .86$).

Table 4.2 Infant Language Measures at 9 and 12 months, by Group

<table>
<thead>
<tr>
<th>Age</th>
<th>Language Measure (mean, SD)</th>
<th>LRC</th>
<th>HRA-N</th>
<th>ASD</th>
</tr>
</thead>
<tbody>
<tr>
<td>9 Months</td>
<td>Vowel</td>
<td>2.80 (1.3)</td>
<td>3.38 (2.0)</td>
<td>2.40 (2.0)</td>
</tr>
<tr>
<td></td>
<td>Consonant-Vowel</td>
<td>.68 (.70)</td>
<td>.76 (.84)</td>
<td>.65 (.55)</td>
</tr>
<tr>
<td>12 Months</td>
<td>Consonants Produced (CSBS)</td>
<td>3.90 (2.0)</td>
<td>3.96 (2.8)</td>
<td>3.17 (2.7)</td>
</tr>
<tr>
<td></td>
<td>Expressive Language T (MSEL)</td>
<td>49.30 (7.6)</td>
<td>46.57 (6.7)</td>
<td>38.67 (6.0)*</td>
</tr>
</tbody>
</table>

Note: * $p < .05$ for difference between ASD and both LRC and HRA-N, $\hat{I} p < .10$ for ASD vs. LRC

12-month language data. CSBS and MSEL scores were available for all but 1 HRA infant. Descriptive information for infants’ 12-month language scores are presented in Table 4.2. Although infants in the ASD group had smaller CSBS Consonant Inventories, this difference was not significant. There were significant group differences in infants’
Expressive MSEL T Scores \((F(2, 58) = 5.72, p = .01)\). Post hoc comparisons (Tukey’s HSD) indicated that infants in the ASD group had significantly lower Expressive MSEL T Scores than both LRC (mean difference: -.10.63, \(p < .001\)) and HRA (mean difference: -.7.90, \(p < .05\)).

**Interrelations between video and laboratory-based data.** In order to assess the relations between 9-month infant vocalizations coded from the home video diaries and infants’ language scores at 12 months Pearson’s zero-order correlations between infants’ 9-month vocalization and 12-month language measures were calculated for each of the three outcome groups: LRC, HRA-N and ASD. The results are presented in Table 4.3. The only significant association was between Infants’ 9-month Consonant-Vowel rate and 12-month Expressive Language T scores for ASD infants \((r = .95, p < .01)\).

<table>
<thead>
<tr>
<th></th>
<th>Expressive Language</th>
<th></th>
<th></th>
<th>Phoneme Inventory</th>
<th></th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>HRA-N</td>
<td>ASD</td>
<td>LRC</td>
<td>HRA-N</td>
<td>ASD</td>
<td>LRC</td>
</tr>
<tr>
<td>Vowel</td>
<td>-.22</td>
<td>-.01</td>
<td>.16</td>
<td>-.31</td>
<td>-.81 (^\dagger)</td>
<td>.21</td>
</tr>
<tr>
<td>Consonant Vowel</td>
<td>-.09</td>
<td>.84 (^*)</td>
<td>.20</td>
<td>-.30</td>
<td>-.06</td>
<td>.13</td>
</tr>
</tbody>
</table>

Note: \(^*\) \(p < .05\), \(^\dagger\) \(p < .10\)

**Maternal Vocalizations and Responses**

Mothers produced an average of 12.22 utterances per minute, which did not differ between the groups (LRC: \(M = 11.56, SD = 4.4\); HRA-N: \(M = 12.80, SD = 4.7\), ASD: \(M = 13.20, SD = 5.50\), \(F(2,59) = .624, p = .54\). The three groups also did not differ in the overall proportion of infant vocalizations they responded contingently to, with LRC
mothers responding to 46%, HRA-N mothers responding to 40%, and ASD mothers responding to 35% of infants’ total utterances, \( F(2, 59) = .92, p = .41 \).

**Distribution of maternal response types.** A 3 (Diagnostic Group) x 7 (Maternal Response Type) repeated measures ANOVA was utilized to analyze the distribution of individual maternal response types between the two groups. Due to significant positive skew within the individual response types, data were arcsine transformed prior to analysis. Descriptive information on the distribution of these individual response types (transformed data) is presented in Figure 4.1. There was a significant main effect of Maternal Response, \( F(4.93, 348) = 48.64, p = .000 \). Contrasts revealed that overall, mothers produced significantly more Acknowledgements than any other response type (all \( p \)’s > .000), higher Imitation than Attributes (\( p = .03 \)), Play than Attributes (\( p = .02 \)) and Questions than either Attributes or Directives (both \( p \)’s < .01). These main effects were qualified by a significant Diagnostic Group X Maternal Response Type Interaction, \( F(9.78, 342) = 1.88, p = .05 \), indicating some differences in the pattern of responses between the groups. Simple effects analyses revealed significant group differences only for mothers’ rate of Label responses, \( F(2,59) = 3.74, p = .03 \). Post hoc tests (Tamhane’s) revealed no robust differences between the groups, but a trend level difference for mothers of ASD infants to use more labels than mothers of LRC infants (\( p = .09 \)).

**Maternal responses to infant vowel and consonant-vowel vocalizations.** We were next interested in determining whether mothers’ responses differed to each the two infant vocalization types. Because of this interest in examining differential response
Figure 4.1. Response Rates for individual maternal responses, by group. Error bars represent standard errors.
patterns, these analyses were conducted using dyads whose infants had produced both vocalization types (24 LRC, 18 HRA, 5 ASD). Rather than investigating responses to infant vocalizations across all 7 individual maternal response types, we were primarily interested in determining whether mothers differed in their use of Language Promoting and Non-Promoting responses to infants’ vocalizations. To address this question, differences in maternal responses to vowel and consonant-vowel vocalizations between the risk groups were examined using a $2 \times 2 \times 2$ repeated measures ANOVA, with Infant Vocalization Type (VV and CV) and Maternal Response Type (Language Promoting and Non-Promoting) as the within-subjects factors and Group (Risk Status) as the between-subjects factors. Of these summary maternal response variables, only Maternal Non-Promoting Responses to Consonant-Vowels demonstrated significant positive skew, due to a large number of zeroes. Arcsine transformations were conducted to improve the normalization of these summary variables, but did not significantly improve the distribution shape. We proceeded with using the non-transformed values for ease of interpretation, and the pattern of results was unchanged when the analyses were conducted using the transformed variables. Descriptive information on means and standard deviations for these summary response variables are presented in Table 4.4.

For this ANOVA, there were significant main effects of both child vocalization type, $F(1,44) = 9.61, p = .003$, and maternal response type, $F(1,44) = 61.51, p = .000$, with infants overall producing more Vowel than Consonant-Vowel utterances, and mothers overall producing more Language Promoting that Non-Promoting Responses.
These main effects were modulated by a significant Infant Vocal Type X Maternal Response Type interaction, $F(1,44) = 22.45, p = .000$. There were no significant main or interaction effects involving group. Simple main effects analyses were conducted to determine the source of this significant interaction and revealed that Language Promoting responses occurred significantly more frequently in response to Consonant-Vowel than Vowel vocalizations ($F(1, 46) = 21.57, p = .000$), while the opposite pattern was observed for Non-Promoting responses, which occurred significantly more in response to Vowels than Consonant-Vowels ($F(1, 46) = 10.43, p = .002$). This interaction between Language- and Non-Promoting responses and infant vocalizations is displayed in Figure 4.2.

In order to determine whether this pattern of maternal responses at 9 months was associated with infants’ later language, Pearson correlations were used to analyze the relationship between mothers’ Total Vocalization Rate, Language Promoting Responses to Consonant-Vowel utterances, and infants’ 12-month MSEL Expressive Language and

<table>
<thead>
<tr>
<th>Vocalization Type</th>
<th>LRC n = 24</th>
<th>HRA-N n = 18</th>
<th>ASD n = 5</th>
</tr>
</thead>
<tbody>
<tr>
<td>Promoting, Vowels</td>
<td>.36 (.18)</td>
<td>.31 (.16)</td>
<td>.16 (.21)</td>
</tr>
<tr>
<td>Non-Promoting, Vowels</td>
<td>.13 (.11)</td>
<td>.15 (.10)</td>
<td>.07 (.11)</td>
</tr>
<tr>
<td>Promoting, Consonant-Vowels</td>
<td>.53 (.27)</td>
<td>.50 (.36)</td>
<td>.50 (.24)</td>
</tr>
<tr>
<td>Non-Promoting, Consonant-Vowels</td>
<td>.10 (.14)</td>
<td>.05 (.06)</td>
<td>.00 (.00)</td>
</tr>
</tbody>
</table>
12-month CSBS Phoneme Inventories, by group. Zero-order Pearson correlation coefficients are presented in Table 4.5. None of the associations were significant.

Figure 4.2 Mean proportion of infant vocalizations receiving a maternal response for vowel and consonant-vowel vocalizations. Error bars represent standard errors.

Table 4.5 Zero-order Pearson Correlations between 9-Month Maternal Responses and 12-Month Infant Language Measures, by Group

<table>
<thead>
<tr>
<th></th>
<th>Expressive Language</th>
<th>Phoneme Inventory</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>HRA-N</td>
<td>ASD</td>
</tr>
<tr>
<td>Total Utterances</td>
<td>.20</td>
<td>.12</td>
</tr>
<tr>
<td>Language Promoting</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Responses to CV</td>
<td>-.13</td>
<td>-.55</td>
</tr>
</tbody>
</table>

Interrelations Amongst Maternal Language and Background Characteristics

Finally, to examine relationships between maternal vocalizations and maternal and family characteristics hypothesized to influence their vocalization patterns, Pearson correlations were calculated to assess the relations between Maternal Total Utterances and Total Contingent Responses and maternal and family background characteristics of
interest: maternal broader phenotype characteristics, the older diagnosed child’s symptom severity, and mothers’ concurrent ASD-related concerns. Zero-order Pearson’s correlation coefficients are presented in table 4.6. None of these associations were significant.

**Table 4.6** Zero-order Pearson Correlations Coefficients between Maternal Vocalizations and Responses and Family Background Factors, for High Risk Families

<table>
<thead>
<tr>
<th></th>
<th>BAP Total Score</th>
<th>ASD Concerns</th>
<th>Proband Severity</th>
</tr>
</thead>
<tbody>
<tr>
<td>Maternal Total Utterance Rate</td>
<td>.11</td>
<td>.01</td>
<td>.07</td>
</tr>
<tr>
<td>Maternal Total Contingent Response Rate</td>
<td>-.09</td>
<td>.08</td>
<td>-.36</td>
</tr>
</tbody>
</table>

Note: These correlations were conducted on the subset of HRA infants with available data; for BAP, \( n = 16 \); Concerns \( n = 24 \), Proband, \( n = 22 \).

**Discussion**

In this study, we examined infant vocalizations and maternal responses to those vocalizations at 9 months of age, and relations between these 9 month measures and infants’ 12 month language abilities in both typically developing infants and infants at risk for ASD. We also examined relations between high risk mothers’ pattern of responses to their infants’ vocalizations and maternal and family characteristics hypothesized to contribute to their behavioral responses.

We found no differences in infants’ vowel and consonant-vowel production rates at 9 months of age between low risk typically developing infants, high risk infants who were not diagnosed with ASD, and in the small subset of infants who later were classified as meeting criteria for ASD. In general, mothers in all three groups responded similarly to their infants’ early vocalizations, though mothers of infants’ later diagnosed tended to respond by labeling objects more frequently than the other two groups. The general
pattern of maternal responses reported here closely replicates the findings of Gros-Louis et al (2006), who also reported that mothers’ responses to their infants’ vocalizations were most frequently acknowledgements.

All three groups of mothers demonstrated significant differentiation in their responses to infants’ early vocalizations, responding with feedback hypothesized to promote language development significantly more frequently when infants produced consonant-vowel utterances rather than vowel-only utterances. This differential responding to consonants with higher quality maternal feedback is very consistent with previous laboratory-based analyses of maternal contingent responding at this age (Gros-Louis et al., 2006). Our results extend these laboratory-based findings to the home, and suggest that for this specific feature of dyadic interactions, laboratory-based interactions largely reflect the daily interactions of 9-month-old infants and their mothers.

Based on previous investigations that have found that experimentally manipulated maternal feedback significantly increases the complexity of infants’ proximal (within the same session) language production and that mothers spontaneously differentially respond to infants consonant-vowel vocalizations with responses hypothesized to promote language development, we expected that mothers’ language promoting responses to consonant vowels at 9 months would be associated with infants’ language abilities at 12 months of age. Across all three groups, we found no such associations. These findings suggest that while clearly exerting strong and specific influences on infants’ early language production, maternal feedback is more closely tied to proximal, rather than distal, features of infants’ language. Because the 9-month data presented here largely
replicates the previously reported literature, this lack of association between these 9 and 12-month measures is unlikely to reflect issues with accurately capturing the same phenomena. Instead it suggests that maternal contingent responses to infants’ vocalizations do not underlie broad language development during this period.

While this lack of association between maternal feedback at 9 months and infants’ language abilities at 12 months suggests a minimal role of maternal input in shaping language development, there are several important points to consider. The first is that infants’ demonstrate a tremendous increase in sophisticated social-communicative understanding and behaviors that emerge over the same period of 9 to 12 months (de Barbaro, Johnson, & Deák, 2013; Tomasello, Carpenter, Call, Behne, & Moll, 2005). This ‘social revolution’ represents a major shift in infants’ ability to engage in triadic interactions, behaviors that underlie many language-learning opportunities and individual differences in these abilities may dampen any effects of the relatively narrow behaviors of focus here. Second, many of the reported effects of environmental (maternal) input on infants’ language development have been most striking in cases of relatively impovershed input, as in the case of families from low socioeconomic status (SES), or in infants with physical hearing issues (i.e. chronic ear infections) (Nittrouer & Burton, 2005). The current investigation was not designed to answer questions about differences in maternal input related to SES, and the majority of families participating in this study were from high SES backgrounds and thus unlikely to provide relatively impovershed linguistic input. The current study was designed however, to determine whether mothers of high risk infants provided similarly impovershed input either in response to decreases
in infant vocal production or other unique characteristics of high risk mothers. If any of these factors resulted in mothers of high risk infants providing less frequent or lower quality feedback to their infants’ early vocal production, it may have helped to explain some of the delays in language ability amongst high risk infant siblings. Our results show that this is not the case. Mothers of high risk infant siblings provide nearly identical feedback to their infants’ 9-month vocalizations in terms of both frequency and content as mothers of low risk infants. These results suggest that risk status does not negatively influence maternal behavior in this domain. Importantly, these data do not eliminate the possibility that differences in maternal behavior emerge as a consequence of the language delays observed in the second year of life in both later diagnosed and non-diagnosed high risk infants (Mitchell et al., 2006; Ozonoff et al., 2014). For high risk infants, 9 to 12 months represents the period of development during which many atypical social and linguistic patterns are beginning to emerge (Ozonoff et al., 2010, 2014). Future investigations should determine whether intervening to increase high risk mothers’ frequency of contingent responses results in increased frequency of concurrent infant vocalizations or more rapid language development. Such studies would have clear and important implications for early intervention practices and are of particular interest for high risk infant siblings who are exhibiting overt delays in early language, as has been reported in other samples (Patten et al., 2014; Paul et al., 2011).
CHAPTER 5: GENERAL DISCUSSION

The goal of the dissertation work presented here was to better understand infant and maternal characteristics that influence the language and communication development in infant siblings of children with ASD. We were particularly interested in determining whether mothers of high risk infants differed from mothers of low risk infants in the language and communicative input they provide to their infants, as research with typically developing infants has demonstrated that environmental input contributes to infants’ language development. Here, we investigated the possibility that maternal and family factors associated with familial risk for autism would be associated with detrimental changes to maternal language and communication and that these changes could contribute to the language delays that emerge in high risk infants towards the end of the first year of life (Mitchell et al., 2006; Ozonoff et al., 2014). We investigated these issues across three studies, using data collected as part of a larger study of the development of both siblings of children with ASD and typically developing infants from 6 to 36 months of age.

We began by analyzing the day to day concerns reported by mothers of high risk infants in home-based written diaries at 6, 9, and 12 months of age, and later analyzed these home-based diary concerns as one of several maternal and family factors hypothesized to influence maternal behavior. In addition to providing important descriptive information about the daily experiences of mothers of high risk infant siblings, these home-based diary measures revealed several important findings. First, our home-based data expand on previous laboratory-based studies of parent concerns that
have reported elevated concerns at 6 and 12 months by demonstrating that across the first year of life, mothers of high risk infants report concerns about their infants’ development significantly more often than mothers of low risk infants. We found that at 9 months of age, mothers of high risk infants more frequently report concerns than mothers of low risk infants, and that these concerns are correlated with concurrent infant symptoms. At 12 months, maternal concerns were also moderately correlated with concurrent behavior. Importantly, we found no differences in the frequency of concerns reported by mothers of infants later diagnosed with ASD than mothers of high risk infants who were not diagnosed at either 9 or 12 months. These data support our hypothesis that maternal concerns in the first year of life reflect more than simply infants’ own behavior. While previous studies have suggested that parents’ elevated levels of concern in the first year of life broadly reflect parents’ knowledge and concern regarding their infants’ elevated risk status, here we examined whether specific characteristics of high risk families’ may contribute to this early hypervigilance and elevated levels of concern. We found that in fact, at 12 months of age, maternal home-based concerns are associated with both maternal depression and with the severity of the older diagnosed child’s ASD symptoms. These findings indicate that maternal concerns, particularly those experienced on a day to day basis, reflect several aspects of high risk mothers’ experience, rather than purely objective monitoring of their infants’ development.

Our primary focus was on determining whether these early home-based concerns, in addition to other maternal and family characteristics and infants’ own behavior, were related to mothers’ overt behavior during home-based parent-child interactions. We
found that for measures of both 9-month vocalizations and 12-month social responsiveness, high risk infants displayed no differences from low risk infants in the content or frequency of their communicative behaviors. Similarly, we found no overall group differences in mothers’ frequency or type of contingent responses to infants’ vocalizations at 9 months of age or in their frequency or type of prompting behaviors used to initiate social interactions at 12 months of age. We did find however, that at 12 months of age, mothers’ home-based concerns were related to their prompting behavior such that mothers who reported concerns about their infant produced fewer communicative prompts than high risk mothers with no concerns. These findings have several important implications for our understanding of the role of maternal input in the language development of both infant siblings of children with ASD and typically developing infants. First, these findings indicate that in the absence of infant language and communication difficulties, the language and communication of high risk mothers is largely unaffected and closely mirrors the language and communication input provided by low risk mothers. This suggests that while infant siblings of children with ASD are at increased familial risk for ASD and language difficulties, maternal input does not seem to contribute to this initial risk. Despite the presence of risk factors that would predict relatively more impoverished linguistic input in high risk mothers (e.g. depression, parenting stress), they are providing high quality linguistic input to their infants and thus, unlikely to contribute to high risk infants’ early language difficulties.

Although the lack of group differences in maternal behavior reported here may seem somewhat surprising given previous reports of increased maternal directiveness and
gesture use, the current studies help to form a more complete picture of the ways risk status influences early parent child interactions (Talbott et al., 2013; Wan et al., 2013). Together, the data on the behavior of high risk mothers indicates that risk status itself (broadly defined as parents’ awareness of their infants’ risk status and hypervigilance about their development) may contribute to some changes in maternal behavior, but these are observed more in the domain of behavior regulation (i.e. overall directiveness) than in the domain of language and communication (Wan et al., 2012). While there are some changes in maternal behavior in the domain of language and communication, they appear limited to the gesture domain, and are not observed in mothers’ contingent feedback to infants’ vocalizations or prompting strategies (Talbott et al., 2013). If anything, the influence of risk status on high risk mothers’ gesture use appears to be protective, rather than detrimental, as mothers of non-diagnosed high risk infants produce significantly more gestures than mothers of low risk infants (Talbott et al., 2013). The literature on high risk mothers’ gesture use and directiveness indicate that rather than predating infant communication delays, changes in maternal behavior likely arise as a result of infants’ emerging symptoms (Leezenbaum et al., 2013; Talbott et al., 2013; Wan et al., 2013). This interpretation is supported by the results presented here. As we did not find any differences in infant behavior, it is unclear whether such differences would lead to subsequent changes in maternal behavior. Future investigations with infants exhibiting early language and communication impairments are needed to answer this question, which would have important implications for intervention practices, a point to which we will return later.
A second important finding from this dissertation research is the lack of an association between maternal feedback to infant vocalizations at 9 months and infants’ language ability at 12 months of age, even for the typically developing group. While it is tempting to conclude that maternal input may not play a strong role in shaping infants’ language development in this domain, there are several important features of the samples investigated here that limit our ability to make strong claims in this regard. The first is that the measures of infants’ language ability at 12 months of age available for this sample may not be well-suited to capture the language skills most strongly influenced by our 9-month maternal measure. The measures included in the protocol for the larger study are fairly broad, and it is possible that maternal feedback to infants’ early consonant production may be more closely related to their development on a measure of language complexity or phonemic awareness. The second, and more important consideration, is that the sample studied in this investigation was primarily of high SES. The literature on maternal influences on infants’ language development strongly suggests that maternal influences are most apparent for infants receiving relatively impoverished environmental input; thus the typically developing infants in our study are not the ideal population to investigate these larger issues (Fernald, Marchman, & Weisleder, 2013; Hoff, 2006; Nittrouer & Burton, 2005; Rowe et al., 2005). However, while the current studies were not designed to determine effects of SES on maternal contingent feedback or prompting use, they were designed to investigate whether mothers of high risk infants’ differed in these domains. Here, we investigated the hypothesis that mothers of high risk infants might provide relatively impoverished input to their infants, not as a consequence of low
SES, but as a consequence of their risk status. Despite the presence of elevated depressive symptoms, increased hypervigilance, and parenting stress, mothers of infant siblings of children with ASD provide equally high quality input to their infants. Importantly, the negative impact of SES on maternal input is mediated by parental knowledge of child development, suggesting that the language input of high risk mothers from low SES backgrounds may be relatively protected as a result of their experiences with ASD and exposure to intervention programs. These findings have important implications for current early intervention efforts in high risk samples.

Recent studies implementing preventative, parent-led interventions for high risk infant siblings have found minimal treatment effects (Green et al., 2013). The results presented here suggest that parents of high risk infants may already be providing very high quality language and communication input to their infants that may minimize benefits from such treatment, at least in the first year of life. An important consideration is whether the mothers of high risk infant siblings described here are representative of mothers who choose not to enroll in similar intensive longitudinal studies. To the extent that increased hypervigilance contributes to both study enrollment and protective effects on maternal behavior, mothers who choose to enroll may demonstrate higher quality input than mothers who do not. Similarly, it will be important to determine whether and how maternal behavior is shaped by the emergence of overt behavior symptoms. These emerging symptoms are likely to negatively influence dyadic mother-child interactions, leading to atypical feedback and input from mothers and subsequently, amplification of infants’ language and communication difficulties (Dawson, 2008; Green et al., 2013).
The influence of infant symptoms on maternal behavior is observed not only in children with ASD, but in high risk infant samples as well. For instance, while mothers of non-diagnosed infants gesture more frequently than low risk mothers, mothers of high risk infants who are demonstrating reduced gesture rates do not exhibit this same boost in gesture production (Talbott et al., 2013). Even if mothers respond with the same type and frequency of feedback to their infants’ early communication, decreased frequencies of infant communication will lead to subsequent reductions in maternal input (Leezenbaum et al., 2013). It is possible that mothers of high risk infants demonstrating language delays or impairments are less negatively impacted by these symptoms than mothers without prior experience with ASD. Determining how maternal behavior is shaped by the emergence of infant symptoms will have important implications for how best to promote the kinds of maternal responsiveness that seem particularly beneficial for young children with ASD (Green et al., 2013; Haebig, McDuffie, & Ellis Weismer, 2013; Siller & Sigman, 2002, 2008).

Overall Conclusions

The data presented in this dissertation helps to refine the concept of familial risk in infant siblings of children with ASD. The role of maternal input in contributing to this familial risk has received little attention, despite evidence that environmental input plays a role in the language development of young children and that consequently, diminished quality maternal input may underlie some of the language delays observed in high risk infant siblings (Mitchell et al., 2006; Ozonoff et al., 2014). For instance, SES disparities...
are associated with reductions in the amount of speech young children are exposed to and consequently, children’s lower vocabulary knowledge (Hart & Risley, 1995; Pan et al., 2005; Rowe et al., 2005). While mothers of high risk infants enrolled in longitudinal investigations are not typically from low SES samples, they exhibit increased rates of depression and other characteristics that would predict reductions in linguistic input (Bettes, 1988; Stein et al., 2008). Despite these maternal risks, mothers of high risk infant siblings of children with ASD show no differences in the quality of linguistic input they provide to their infants at either 9 or 12 months. These findings suggest that while infant siblings of children with ASD are at increased familial risk, maternal environmental input does not contribute to increased initial risk. Instead, variability in high risk infants’ language and communication abilities are likely to reflect other genetic or environmental risk factors.
BIBLIOGRAPHY


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Training in the Autism Diagnostic Interview –Revised (ADI-R)
Reliable administration of the following measures (supervised by Dr. Alice Carter):
  i. Communication and Symbolic Behavior Scale – Developmental Profile (Behavior Sample)
  ii. Mullen Scales of Early Learning
  iii. Autism Observation Scale for Infants