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## The role of caregiver speech in supporting language development in infants and toddlers with autism spectrum disorder

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### Abstract

Parents play an essential role in supporting child development by providing a safe home, proper nutrition, and rich educational opportunities. In this article we focus on the role of caregiver speech in supporting development of young children with autism spectrum disorder (ASD). We review studies from typically developing children and children with autism showing that rich and responsive caregiver speech supports language development. Autism intervention studies that target caregiver speech are reviewed as are recent scientific advances from studies of typical development. The strengths and weakness of different techniques for collecting language data from caregivers and children are reviewed, and natural language samples are recommended as best practice for language research in autism. We conclude that caregivers play a powerful role in shaping their children's development and encourage researchers to adapt parent-mediated intervention studies to acknowledge individual differences in parents by using a personalized medicine approach.

### Keywords

autism spectrum disorder; ASD; caregiver speech; high familial risk; home language environment; language

### Introduction

Parents play an essential role in supporting child development by providing a safe home, proper nutrition, and rich educational opportunities. In this article we focus on the role of caregiver speech in supporting development of young children with autism spectrum disorder (ASD). Autism is a neurodevelopmental disorder with a strong, but complex genetic basis, that is typically diagnosed after 4 years of age (Baio et al., 2018). Children with autism have deficits in social interaction and communication, and have restrictive or repetitive patterns of behaviors, interests, or activities (American Psychiatric Association, 2013). In this article, the history of researching parents of children with autism will be reviewed, and results from modern studies will be synthesized. To highlight future directions

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for autism research, we will review recent scientific advances from studies of typical development. We conclude that caregivers play a powerful role in shaping their children's development. We also advocate that individual differences in caregivers should be taken into consideration if early intervention efforts are going to be maximally effective.

## Early History of Research on Parents of Children with Autism

Before examining current research on caregiver speech to children with autism one must first acknowledge the historical context and sensitive nature of this topic. Early descriptions by Leo Kanner described parents of children with autism as displaying “coldness” and “obsessiveness” (Kanner, 1949). These descriptions led others to popularize the term “refrigerator mother” and to propagate the theory that a lack of maternal warmth causes autism in children. It is important to note that Kanner himself viewed autism as innate, and later in his career he was an outspoken advocate against blaming parents for their children's autism diagnoses. Regardless, the “refrigerator mother” theory took hold in the 1950s and 1960s, facilitated primarily by Bruno Bettelheim who was instrumental in spreading the theory. Bettelheim himself has long been a controversial figure with accusations that he engaged in plagiarism, abusive treatment of students, and misrepresentation of credentials (he did not have a degree in child psychology, as claimed) (Pollak, 1998).

In the 1960s the prevailing theory of the cause of autism in both the public and the medical community was cold parenting. Bettelheim recommended that children with autism be removed from their homes and live in a therapeutic milieu institutional setting (Bettelheim & Sylvester, 1948). He also supported early research on the parents of institutionalized children which concluded that parents were not loving their children “right” (e.g., Henry, 1973). The popularization of this theory had profound effects on families of children with autism. Many parents experienced intense guilt and shame and were left trying to convince professionals that their children were indeed wanted and loved. Along the way, many parents lost trust in the professionals charged with helping their children. Belief in this misguided theory also meant that many researchers and psychotherapists were failing to intervene on actionable treatment targets.

By the mid 1960s critics of Bettelheim and the “refrigerator theory” of autism began to gain traction. Bernald Rimland was an early outspoken critic of Bettelheim, challenging his psychoanalytic perspective on autism and instead proposing a neurodevelopmental perspective (Rimland, 1964). Rimland's neurodevelopmental perspective, informed by the observation of co-occurring seizures in many children, asserted that autism was caused by aberrant growth and development of the brain very early in life. This theory is directly supported by recent magnetic resonance imaging (MRI) studies showing atypical brain growth in young infants who go on to develop autism (Hazlett et al., 2017).

Rimland was an advocate for families affected by autism, a natural alliance since he was a parent of a child with autism himself. In 1965 Rimland, with the help of Ruth Sullivan and group of parents, went on to form the advocacy group National Society for Autistic Children (now called Autism Society of America). Since the founding of the National Society for

Autistic Children and continuing today, parent advocacy groups have played an instrumental role in informing public policy, funding autism research, and shaping public perception.

Today, the “refrigerator theory” of autism has been widely disproven; however, the damage of this era of autism research is enduring. It is because of this history that some in the autism community feel that mothers of children with autism should not be the focus of research. For a discussion on the topic see Tager-Flusberg (2016). In this article, we discuss how caregiver behaviors support child development. This article does not subscribe to the notion, nor promote the idea that caregiver behavior can *cause* autism. Rather, this article promotes a strength-based approach to understanding individual differences in caregiving. We also adopt an integrated resilience framework, in a broad sense, where parents promote positive infant development through the way they nurture, socialize, and communicate with infants (Lai & Szatmari, 2019; Masten, 2018). In the traditional resilience framework, resilience is demonstrated when adaptive outcomes are observed in the face of adversity. We endorse a broader framework where autism is not viewed as an adversity (Lai & Szatmari, 2019), while acknowledging that the genetic underpinning of autism contributes to risk of higher rates of language delay (Marrus et al., 2018; Swanson et al., 2017). In this framework children with autism who go on to have positive outcomes in terms of language and communication have demonstrated resilience. We present data showing that, much like how good nutrition supports physical growth, rich and responsive caregiver speech contributes to resilience by supporting language and communication development in young children with autism.

## Early Language Skills are a Harbinger of Later Development in Typically Developing Children

Supporting early language skills is crucial because these early skills are related to school readiness (Fiorentino & Howe, 2004) and long-term development (Pace, Alper, Burchinal, Golinkoff, & Hirsh-Pasek, 2019). Early language skills are the best predictor of later academic outcomes across broad domains (Pace et al., 2019). For example, preschool language skills are positively associated with school-age language, reading, and mathematics skills (Pace et al., 2019). Early language skills also predict gains in subsequent language skills. Children who enter elementary school with the best language skills have the largest gains in reading between 1st and 3rd grade (Pace et al., 2019).

These cascading developmental events can be disrupted by exposure to a variety of social risk factors, like poverty, maternal depression, and low parental education. For example, exposure to multiple risk factors is related to lower levels of reading and mathematics skills (Burchinal, Roberts, Zeisel, Hennon, & Hooper, 2006). However, the negative effects of social risk factors can be mitigated by exposure to protective factors such as responsive and sensitive parenting. Research has shown that a stimulating childcare environment can mitigate the negative effects of being exposed to social risk factors (Burchinal et al., 2006). Early language skills also play a significant role in the predictive association between social risk factors and school readiness: early language skills mediate the relationship between

social risk factors and academic achievement (Burchinal et al., 2006; Forget-Dubois et al., 2009).

## The Role of Caregiver Speech in Supporting Typical Child Development

In the 1990s two seminal studies were published showing that hearing more caregiver speech during infancy results in better language skills later in life (Hart & Risley, 1995; Huttenlocher, Haight, Bryk, Seltzer, & Lyons, 1991). For both studies research staff went into the families' homes and recorded mothers interacting with their children. They then transcribed the audio files, a time-consuming process that yields estimates of the quantity of caregiver language exposure and estimates of infant vocabulary growth. Huttenlocher et al. (1991) found that girls and boys experienced similar levels of caregiver speech, but girls had faster vocabulary growth when compared to boys. These sex differences in vocabulary growth started to decline after 20 months and were no longer observable by the end of the second year of life. For both sexes, those children that heard the most caregiver speech also had the largest vocabulary growth.

Hart and Risley (1995) generated rich profiles of the language environment experienced by children of varied socioeconomic backgrounds. Every month for over two years they collected home language samples that were transcribed and annotated, generating a remarkable data set. Results indicated that parent's socioeconomic status (SES), assigned based on occupation of both parents, was related to the amount of caregiver speech an infant experienced with high SES parents talking to their children more than parents from lower SES households. Hart and Risley famously coined the term "word gap," espousing that by 3 years of age, low SES children heard 30 million fewer words than high SES children (Hart & Risley, 2003). In addition to disparities in quantity of caregiver speech, they also found SES differences in qualitative aspects of caregiver speech. Higher SES caregivers used more encouraging and less discouraging words when compared to lower SES caregivers. These early differences in experience had lasting developmental impacts. Children who heard the most caregiver speech as infants had the best vocabularies when they were 3 years old. Discussion of this study often focuses on the amount of speech, but Hart and Risley's data actually showed that the richness of caregiver speech was more strongly related to later child development than the amount of caregiver speech.

All of the children in these seminal studies by Huttenlocher et al. (1991) and Hart and Risley (1995) learned to walk and talk, and by all accounts they developed typically. The lasting contribution of this work is the understanding that parents varied greatly in how much they talked to their children, and this variation was significantly associated with later performance on vocabulary and IQ tests. Hart and Risley (1995) concluded that rather than providing early intervention services to children who experience less caregiver speech, we should rather support and help parents. This call to action spurred decades of research and wide-scale public policy campaigns aimed to increase caregiver speech (e.g., Providence Speaks, Too Small to Fail).

The finding that early caregiver speech is associated with child language skills has been replicated many times since the initial studies were published (Cartmill et al., 2013; Hirsh-

Pasek et al., 2015; Hoff, 2003; Hurtado, Marchman, & Fernald, 2008; Huttenlocher, Waterfall, Vasilyeva, Vevea, & Hedges, 2010; Pan, Rowe, Singer, & Snow, 2005; Rowe, 2008, 2012; Weisleder & Fernald, 2013), as have the findings that there are SES differences in caregiver speech (Cartmill et al., 2013; Hoff, 2003; Huttenlocher et al., 2010; Rowe, 2008, 2012). A recent paper challenged the size of the word gap (Sperry, Sperry, & Miller, 2018); however, there are noteworthy methodological distinctions between the Sperry et al. (2018) report and the original work by Hart and Risley (1995). For example, Sperry et al. (2018) do not include a high SES group, and their definition of language input is much broader than the original work by Hart and Risley (1995). Given the methodological differences between studies, claims that the Sperry et al. (2018) study refutes the findings of Hart and Risley (1995) should be evaluated with caution.

More recent studies have aimed to understand what types of caregiver speech best support language skills across development. There is now an understanding that caregiver speech patterns change as infants develop new communication skills, but our understanding of this developmental sequence is still being refined. Rowe and Zuckerman (2016) put forward a conceptual framework for how caregiver speech changes as infants develop. In this framework infants 0–6 months benefit the most from infant-directed speech, a speech style characterized by slower speech, elongated vowels, and wide ranges in pitch. Between 6 and 18 months infant's gesture repertoire greatly expands and language learning is supported when parents point with their infants and contingently respond with object labeling to points by the infant. During the toddler years the use of diverse vocabulary, especially the use of wh-questions supports children's expanding vocabularies. After the age 3 years, caregivers can support language skills by using more sophisticated language and using speech that includes the past and present tense. Several studies support this general shift from speech quantity to speech quality as the "key ingredients" to supporting infant language skills (Jones & Rowland, 2017; Ramírez-Esparza, García-Sierra, & Kuhl, 2014, 2017; Rowe, 2012).

### **Children with Autism Show Early and Persistent Difficulties with Language**

Many children with autism show early and persistent difficulties with language. However, language skills in children with autism are highly variable, as are early trajectories of language development. Children with autism may show delays in reaching early language milestones such as canonical babbling and first word acquisition (Iverson & Wozniak, 2007; Mayo, Chlebowski, Fein, & Eigsti, 2013; Patten et al., 2014). When group-level language skills are explored, children with autism show deficits in language at 12 months of age, with these deficits becoming more pronounced at 24 months of age (Estes et al., 2015; Hudry et al., 2014; Swanson et al., 2017). These early difficulties with language persist for a substantial proportion of children with autism. About 29% of school-age children with autism display minimal language and another 24% produce words but not sentences (Anderson et al., 2007; Tager-Flusberg & Kasari, 2013). Language difficulties may also be more subtle, presenting as difficulties in semantic and pragmatic language (for a review of this literature see Tager-Flusberg, Edelson, & Rhiannon, 2011).

Language difficulties aggregate in families of children with autism, even for siblings without an autism diagnosis themselves. Siblings of children with autism have been shown to have higher rates of language delay (Gamliel, Yirmiya, & Sigman, 2007; Iverson & Wozniak, 2007; Marrus et al., 2018), and lower language scores (Gangi, Ibañez, & Messinger, 2014; Miller et al., 2015; Mitchell et al., 2006; Paul, Fuerst, Ramsay, Chawarska, & Klin, 2011; Seery, Tager-Flusberg, & Nelson, 2014; Swanson et al., 2017; Toth, Dawson, Meltzoff, Greenson, & Fein, 2007) than children with no familial risk for autism. Further, early language skills in children with autism represent a reliable predictor of later skills. A younger age of first word acquisition is significantly associated with better cognitive and adaptive skills (Kover, Edmunds, & Ellis Weismer, 2016; Mayo et al., 2013).

## Caregiver Speech and Later Language Skills in Toddlers and Children with Autism

In 2002 Michael Siller and Marian Sigman published a study examining how mothers communicated with their young children with autism (Siller & Sigman, 2002). The study had two major findings. First, the authors reported that mothers of children with autism provided similar amounts of responsive verbal utterances as mothers of typically developing children. Second, the study was the first to report that responsive verbal utterances were significantly associated with better language 10 and 16 years later for children with autism (Siller & Sigman, 2002). Responsive verbal utterances, or “synchronized caregiver speech” were defined as instances when the caregiver makes a comment or reinforcing statement related to the child’s ongoing activity. Verbal responsiveness supports early child language skills by providing labels for the object or activity at the center of the child’s attention. This style of communication is responsive to the child’s focus of attention and does not require them to shift attention. This strategy may be especially effective for children with autism who may have relative deficits in social communication, including joint attention. Siller and Sigman went on to replicate their findings in an independent sample of children with autism (Siller & Sigman, 2008). These studies played a role in spurring others to investigate the role of caregivers in supporting language and cognitive development in children with autism.

Recent studies support early reports by showing that responsive caregiver communication is associated with gains in language skills across a 6-month time period when children with autism were 3–4 years old (McDuffie & Yoder, 2010). However, the potential benefit of responsive caregiving may rely on the child’s cognitive skills. Haebig, McDuffie, & Weismer (2013) found that children with autism with minimal language skills had larger language gains from responsive caregiver communication when compared to children with autism with fluent language skills. The engagement state of children also seems to influence potential benefits. Responsive caregiver language during times when the caregiver and child are playing collaboratively seems to support later language skills more so than when responsive caregiver language is used when the child and caregiver are not playing collaboratively (Bottema-Beutel, Yoder, Hochman, & Watson, 2014).



## Caregiver Speech and the Home Language Environment in Infants with a Family History of Autism Spectrum Disorder

The advent of the “baby-sibling” research design where infant siblings of older children with autism are prospectively followed was another paradigm shifting moment for the autism field. For families with one child with autism, the risk for autism is higher in subsequently born infants. Population-based cohort studies estimate recurrence risk to be around 10%, whereas baby-sibling convenience samples estimate recurrent risk to be closer to 20% (Ozonoff et al., 2011; Sandin et al., 2014). These recurrence risk rates indicate that most families impacted by autism are simplex families, meaning there is one child with autism in the family. Multiplex families, where there is more than one child with autism in the family, represent the minority of families.

The baby-sibling research design provided a window into the development of infants who would later be diagnosed with autism. For the first time researchers could describe autism during infancy, before clinically reliable diagnoses are available. Over the last decade, these studies have documented a pre-symptomatic period early in infancy where the defining behavioral features of autism are not yet present (Estes et al., 2015; Ozonoff et al., 2010; Wolff et al., 2014). However, atypical neurodevelopment does begin during this period (Shen et al., 2017; Wolff et al., 2012), and pre-symptomatic brain features can accurately predict later autism diagnoses (Emerson et al., 2017; Hazlett et al., 2017). The results of these studies could drastically lower the age of identification and age of entry into treatment for children from multiplex families. Further research using population-based sampling strategies will be needed to determine if these findings extend to children with autism from simplex families.

The baby-sibling design has been used to examine early parent–child interactions and to quantify aspects of the home language environment. Overall, mothers of high- and low-risk infants provide equally rich linguistic environments (Campbell, Leezenbaum, Mahoney, Day, & Schmidt, 2015; Leezenbaum, Campbell, Butler, & Iverson, 2014; Swanson et al., 2018; Talbott, Nelson, & Tager-Flusberg, 2015). However, there are subtle differences in parent–infant vocal and play interactions when comparing high- and low-risk dyads. For example, among 9-month-old high-risk infants and their mothers, a greater amount of simultaneous speech was associated with later language delay (Northrup & Iverson, 2015). Simultaneous speech was defined as times when the infant and parent were talking at the same time.

Several studies have reported that high-and low-risk mothers show statistically equivalent levels of parental responsiveness (e.g., appropriate engagement to the infant that is contingent and developmentally appropriate) both when high risk is compared to low risk, and when infants who go on to have autism are separated into their own group (Wan et al., 2013, 2012). These studies show that mothers of high-risk infants are just as sensitive when responding to their infant as low-risk mothers. However, levels of non-directiveness do seem to differ by group, with high-risk parents being rated as more directive than their low-risk peers (Wan et al., 2013, 2012). Non-directive behaviors focus on the infant’s experience and current activity, while directive behaviors include prohibiting infant behavior, vocal

demands, and requesting the infant to shift attention from a preferred activity. This finding that parents of high-risk infants are more directive may be a spillover effect learned from interacting with an older child with autism. The older child with autism may have limited functional play skills and in response the parent may have adapted a more directive play style.

Subtle group differences in caregiver speech may also emerge as high-risk infants display difficulties with the development of social-communicative skills. Choi et al. (Choi, Nelson, Rowe, & Tager-Flusberg, 2020) found that parents of high-risk infants and low-risk parents used similarly complex sentences when speaking to their 12-month-old infants, but high-risk parents used sentences with simpler complexity when speaking to their 18-month-old infants. It may be the case that parents of high-risk infants adjust their language to match their infant's communication skills (Choi et al., 2020).

A recent report took a different approach and examined caregiver speech to infants using day-long home language recordings (Swanson et al., 2019). Home language recordings were collected when infants were 9 and 15 months of age and language skills were assessed when infants were 24 months of age. Across all infants in the study, including those with autism diagnoses, infants who heard more caregiver speech at 9 or 15 months had better language skills later in life. This pattern of results held true for both measures of the quantity and quality of early caregiver speech. This study also showed that caregiver speech mediated the relationship between maternal education and later child language skills. Mothers in the study with higher educational attainment talked more to their children, and in turn their children had better language skills (Swanson et al., 2019).

Results also revealed that there are dynamic changes in parent-child communication across the first two years of life, and these patterns may differ in typically developing children and children with autism (Swanson et al., 2019). Typically developing children experience a decrease in adult words and a compensatory increase in conversational turn counts over the 9–15-month time period. These findings likely reflect the expanding communication repertoire of typically developing children during this time frame. Swanson et al. (2019) saw a different pattern of results in children later diagnosed with autism. These infants also experienced a decrease in adult words counts across 9–15 months, but they did not experience an increase in conversational turn counts. These results could be reflecting an early disruption in the social feedback loop (Warlaumont, Richards, Gilkerson, & Oller, 2014). In summary, caregiver behaviors during infancy have a significant impact on development and caregiver speech may be a critical medium to support development in infants who go on to receive an autism diagnosis.

## Caregiver Speech as an Intervention Target

Promising results from studies of typically developing children and children with autism showing the effects of responsive caregiving on child language skills contributed to the development of intervention studies targeting responsive caregiving. To date, at least 27 studies have been published outlining results of parent-training interventions on language development of children with autism (Roberts, Curtis, Sone, & Hampton, 2019). On average,



these studies report large effect sizes on parent outcomes and medium effect sizes on child language skills (Roberts et al., 2019). These intervention studies provide evidence that, with instruction, parents can learn responsive communication strategies in the course of a short intervention, and that these changes support child language skills. We outline a selection of notable studies below.

Based on their early results Michael Siller and Marian Sigman designed Focused Playtime Intervention, a parent-mediated intervention for toddlers and young children with autism designed to support family capacity building. They tested the approach first in minimally verbal preschoolers with autism and results indicated positive treatment effects for parent responsive verbal behaviors (Siller, Hutman, & Sigman, 2013) and parents' capacity for reflection and self-evaluation (Siller et al., 2018). There were also positive treatment effects for child language gains (Siller et al., 2013) and child attachment behaviors (Siller, Swanson, Gerber, Hutman, & Sigman, 2014). Results from a subsequent multisite study of toddlers showing early signs of autism (identified in the community) indicated positive treatment effects for responsive verbal behaviors, but these effects were not maintained at the one-year follow-up (Kasari et al., 2014).

Green and colleagues carried out the Preschool Autism Communication Trial (PACT), a large and rigorous randomized study of toddlers with autism, employing trial registration, double-blind assessment procedures, an independent statistician, and long-term follow-up. Children who had participated in PACTS had positive treatment effects for autism severity and responsive verbal behaviors, but insignificant treatment effects for child language skills (Green et al., 2010). Follow-up analyses indicated that the treatment effects on autism severity were mediated by responsive verbal behaviors (Pickles et al., 2015). Children who participated in PACT had reduced autism symptoms at a long-term follow-up assessment over 6 years later, but the treatment and control groups no longer differed in responsive verbal behaviors (Pickles et al., 2016). One interpretation of these findings is that responsive verbal behaviors have a larger impact early in development when pivotal skills are emerging when compared to later in development.

Another large randomized trial of preschoolers with autism echoes the PACT results, with positive treatment effects for responsive parental behaviors, but no significant effects for child language skills (Solomon, Egeren, Van Egeren, Mahoney, Huber, & Zimmerman, 2014). These null results for language skills could be a result of many causes, including issues related to the outcome measure (e.g., insufficient dimensionality, floor effects) and the intervention (e.g., approach not specific enough, low fidelity).

## Caregiver Speech as an Intervention Target for Pre-symptomatic Infant Studies

Parent-mediated interventions including components of responsive caregiver behaviors have been downward extended into infancy using the baby-sibling research design. The overarching goal of many of these “pre-emptive interventions” is to mitigate risk and modify developmental trajectories, not to eliminate the disorder or condition (Insel, 2007). Reliable diagnostics are not available for autism until the second year of life so the infant intervention

studies carried out to date have randomized across high-risk participants, with no additional selection criteria (with one exception, see Rogers et al., 2014 below), regardless of eventual diagnostic status. Since the intervention is then applied to infants who will go on to have autism as well as those who go on to be typically developing, ethical considerations require that the intervention be beneficial to all participants (and not only those with autism). It should be noted that parents find these approaches acceptable and enjoyable, and they find that the intervention provides tools to better understand their infants' behavior (Green et al., 2013).

The iBASIS trial was a low-intensity randomized control trial testing a social communication intervention for infants at high familial risk for autism (Green et al., 2015, 2017). Results for this trial indicated significant intervention effects for parental synchrony and infant attentiveness to caregiver (Green et al., 2015). However, they did not find intervention effects for diagnostic outcome or child language skills (Green et al., 2017). In a different study of similar intensity, the "Promoting First Relationships" intervention for high-risk infants showed intervention effects on neurocognitive metrics including reduction in habituation time to faces during an eye-tracking task and increased frontal theta power during an electroencephalography (EEG) task (Jones, Dawson, Kelly, Estes, & Webb, 2017). The use of two "low-risk" control samples allowed the researchers to determine that the intervention resulted in more normalized neurocognitive metrics. Rogers and colleagues used yet a different approach and tested an infant version of the Early Start Denver Model called *Infant Start* in a small pilot study ( $n = 7$  in the treatment group). In *Infant Start*, infants either had an older sibling with autism *or* were identified through the community (e.g., parents or clinicians were concerned about early signs of autism and referred the infant) (Rogers et al., 2014). Results of this pilot study found lower rates of autism and higher developmental functioning in infants who had received the treatment when compared to infants in the control group.

The studies reviewed have several limitations to consider, but the preliminary results are promising and warrant further investigation. Of the studies reviewed, two of the three were randomized control trials (Green et al., 2015, 2017; Jones et al., 2017), and all have yet to be replicated. The samples of all studies were small, with the largest study treating 28 infants (Green et al., 2015, 2017). Last, the samples in these studies were overwhelmingly white and from middle and upper SESs. All of the studies were either parent-mediated or included parent coaching studies. However, only one study directly tested the effects of the intervention on parent behaviors, reporting that parents in the intervention decreased their directiveness during play (Green et al., 2015).

Future infant intervention efforts will also likely use parent-mediated approaches. Parent-mediated approaches are advantageous as they make infant intervention feasible, ecologically valid, scalable, and cost-effective. These approaches are also in line with recommended practice for early intervention services for toddlers with autism (Schertz, Baker, Hurwitz, & Benner, 2011) and the guiding principles from Part C (Individuals with Disabilities Education Improvement Act of 2004, IDEIA). For example, key principles for early intervention for autism include, in part, that (a) families play a central role in the intervention, (b) the resources, priorities, and concerns of the family should be taken into

consideration, as should the family's cultural and ethnic context, (c) the intervention should be carried out in the child's natural environment, and (d) the intervention should promote child-initiated learning and supporting functional skills (Schertz et al., 2011). Taking a developmental perspective, commencing intervention before an infant shows delays in reaching language milestones and before difficulties in social interaction are present would have the largest impact on later development (Adamson, Kaiser, Tamis-LaMonda, Owen, & Dimitrova, 2020).

Questions about the design of infant intervention are still being debated, but there is also the question of who should be the target population. It would certainly be unfeasible to provide infant intervention to all younger siblings of children with autism. Studies by the Infant Brain Imaging Study (IBIS) Network showing that brain development in the first year of life can independently predict later autism diagnoses at an individual level (Emerson et al., 2017; Hazlett et al., 2017) highlight how future studies could leverage neurobiology to target only those at ultra-high risk for later autism. A criticism of this approach, using MRI-based screening of high-risk infants to determine who should receive treatment, is the cost associated with MRI brain scans. A recent report counters this claim, with analyses showing that MRI-based screening is a cost-effective strategy for determining which high-risk infants should receive intensive early intervention (Williamson, Elison, Wolff, & Runge, 2020). Early intensive behavioral intervention is a costly endeavor, but these upfront costs are offset by long-term societal and economic savings (Jacobson, Mulick, & Green, 1998; Penner et al., 2015).

## Methodological Considerations Related to Measuring Infant Language Skills and Caregiver Speech

There are several methodological considerations related to measuring caregiver speech and infant language development that warrant discussion. The majority of infant autism studies of parent-child interactions measure caregiver speech and infant vocalizations from short, 10–13 min videos (Campbell et al., 2015; Chericoni et al., 2016; Heymann et al., 2018; Ozonoff et al., 2010; Schwichtenberg, Kellerman, Young, Miller, & Ozonoff, 2018; Walton & Ingersoll, 2015). Short video recordings produce a different account of the language heard by a child when compared to day-long audio recordings collected in the home (Bergelson, Amatuni, Dailey, Koorathota, & Tor, 2018). Specifically, assessments of short videos, compared to day-long audio recordings, revealed caregivers produced more questions, fewer declaratives, more nouns, and higher lexical diversity during short videos (Bergelson et al., 2018). Bergelson and colleagues concluded that short videos provide a dense sample of the infant's experience, rather than an account of the typical experience. In sum, short videos were similar to *peak* audio hours. Day-long audio recordings may generate more ecologically valid data because families may find it easier to behave freely when long-format audio recordings are collected versus short video recordings. Given these findings, day-long language recordings may be the optimal strategy if the goal of a study is to provide an account of what the infant typically experiences.

Measuring infant language and communication skills also requires a unique set of methodological considerations. Measures of expressive language skills are often an outcome measure for treatment studies, so the field requires language measures that are easy to obtain and are sensitive to change. Historically, treatment studies have used standardized assessments or parent-report language questionnaires as outcome measures. Standardized assessments provide a direct test of the child's language skills, but during infancy standard scores on these tests may be subject to floor effects that limit the dimensionality of the data (Klein-Tasman, Phillips, Lord, Mervis, & Gallo, 2009). Standardized tests are often administered before, during, and after an intervention, all within a brief time frame, making practice effects a potential issue. Practice effects may inflate scores on standardized assessments resulting in an inaccurate picture of the child's actual abilities. Both of these potential issues, floor effects and practice effects, can add noise to data and conceal treatment effects. An alternative to direct assessments is parent-report language measures, like the MacArthur-Bates Communication Development Inventories. A major limitation of parent-report measures is that they are susceptible to placebo effects in the context of an intervention (Guastella et al., 2015).

Both direct assessments and parent report measures provide normed summary-level data; however, they do not capture the multivariate aspect of pre-linguistic skills. Natural language samples, like day-long home language recordings, are an ecologically valid, multivariate approach that are not subject to many of the limitations of direct assessments or parent-report measures (Barokova & Tager-Flusberg, 2018; Tager-Flusberg, 2000). Day-long home language recordings capture spontaneous expressive language in an environment where the infant is comfortable. After data collection these recordings can be analyzed and coded for multivariate language and communication features. Coding can be tailored to the goals of the intervention. This approach is not susceptible to placebo effects or practice effects, and floor effects can be mitigated by selecting appropriate variables for coding. Importantly, day-long home language recordings capture the infant's actual abilities and how they communicate in everyday life. Day-long home language recordings may be an approach that combats the extreme sampling bias in developmental science. The approach has been used successfully with diverse samples including families that are low-income, from varied ethnic and racial backgrounds, and non-English speaking (Suskind et al., 2016; Weisleder & Fernald, 2013; Wood, Diehm, & Callender, 2016). The relative methodological advantages of natural language samples have led experts to recommend natural language samples as best practice for language research in autism (Barokova & Tager-Flusberg, 2018; Tager-Flusberg, 2000).

## **Considerations and Limitations Specific to Day-long Home Language Samples**

Day-long home language recordings provide the researcher with a window into the child's linguistic world. The benefits to this research strategy are outlined above, but there are also limitations and unique considerations to consider when using this approach. For a comprehensive guide to collecting day-long home language recordings and the ethics of

such data collection we refer the reader to Casillas and Cristia (2019) and Cychosz et al. (2020), respectively. We review some of these practical and ethical considerations below.

The collection of day-long home language recordings poses complex ethical considerations related to privacy. If a child is wearing the recorder in public there is the potential to capture the voice of someone who has not provided informed consent. Researchers should be informed of state consent laws related to audio recordings that could impact their ability to record in public. One strategy to mitigate these privacy concerns is to have families complete the recording when they are at home for the day. It is more feasible to notify anyone coming into a participant's home of the recording than to notify everyone encountered while at the grocery store, for example. Similar privacy concerns would pertain to recordings collected when the child is at daycare or school.

When collecting home language recordings, the consent process should be given special attention. Researchers should ensure that families understand that they can withdraw from the research study at any time. Families can also be given the additional option to have a recording deleted but stay enrolled in the study. Consent forms should outline these options and include any relevant information on mandatory reporting laws.

Investigators should carefully consider the reliability and validity of the research strategy before collecting data. Day-long home language recordings are best suited to answer research questions about broad phenomenon (e.g., quantity of speech), and less well suited to ask questions about rare phenomenon or linguistic features without a salient acoustic signature (detection of *wh*-questions) (Casillas & Cristia, 2019). A priori research questions will help to ensure the approach is valid. For example, if a researcher is interested in capturing the daily experience of the child (e.g., how many adult words does the child hear during the day), then accounting for time periods when the infant is asleep may not be necessary for valid data. However, if researchers are interested in capturing how many words the child hears *per waking hour* then accounting for nap time and sleep time is necessary.

The recording parameters related to the environment should also be carefully considered. Researchers may limit recordings to the home if the research question relates to speech exclusively by parents. This approach comes with the limitation that environments outside of the home (e.g., daycare) are not being measured. To understand how representative the recording is of the child's environment parents can complete a childcare questionnaire. Some studies have limited recording sessions to weekend days (Thompson, Benítez-Barrera, Angley, Woynaroski, & Tharpe, 2020). However, this approach should be used with caution as the "work week" varies across sectors of employment. Low-income families are more likely to have individuals working non-standard hours than high-income families, so limiting recordings to weekend days has the potential to introduce unwanted bias in the data (Enchautegui, 2013).

Data from day-long home language recordings can be processed using automated analysis software or hand coded. Limitations of automated software include variability in accuracy and precision of available tools (Räsänen et al., 2019) and lack of information on the context of recorded speech. If the recorded data are to be annotated or transcribed, an appropriate

sampling procedure needs to be used as it is often not feasible to annotate the entire day-long recording (see Casillas & Cristia (2019) for a description of different sampling procedures).

## Future Directions and Conclusion

The research outlined in this report underscores the influential role caregivers play in children's long-term development, while also highlighting the first year of life as a time when parent training could substantially support child development. If caregiver speech is pursued as a target for pre-symptomatic intervention studies, further research is needed to identify modifiable factors that could increase intervention efficacy. For example, there is a growing appreciation that SES is a distal factor influencing caregiver speech. More modifiable and proximal factors include: parent knowledge of child development, views on teaching and learning, and the stimulation, support, and structure in the home (Rowe, 2008). Knowledge of child development fully mediates the relationship between SES and caregiver speech (e.g., the relationship between SES and caregiver speech was no longer significant when knowledge of child development was added to the model) (Rowe, 2008).

Expanding upon this work, Rowe & Leech (2019) administered a parent gesture intervention study that included growth mindset training. A growth mindset is the belief that intelligence is malleable. A fixed mindset, on the other hand, is the belief that intelligence is fixed. Parents who entered the study with a fixed mindset, but then received growth mindset training, had children with the largest language gains. Children had smaller language gains if they had parents with a fixed mindset that did not receive intervention (Rowe & Leech, 2019). In other reports, parents in a reading intervention with embedded growth mindset training who reduced their "fixedness" belief had children with the largest gains in reading scores (Andersen & Nielsen, 2016), and mothers primed to use a growth mindset were more supportive of children during a difficult task (Moorman & Pomerantz, 2010). This research highlights how individual differences in parental beliefs can impact child learning and development in meaningful ways.

Applying a personalized medicine approach to parent-mediated autism interventions could increase intervention efficacy, lower costs, and improve child outcomes. As the autism field advances toward first-year autism detection and pre-symptomatic intervention, an evidence base to inform these interventions is needed. Future research should aim to understand what specific features of caregiver speech make ideal treatment targets, and what individual differences in the parents are related to these features. Additionally, large, rigorous treatment studies are needed to determine if innovative personalized medicine approaches are feasible. We conclude with two recommendations. First, we encourage researchers to use long-format, natural language samples to measure caregiver speech and infant vocal behavior. Second, we encourage researchers to adapt parent-mediated intervention studies to acknowledge individual differences in parents by using a personalized medicine approach.

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