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Associations between Caregiver Stress and Language Outcomes in Infants with Autistic[1] and Non-Autistic Siblings: An Exploratory Study

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[1]In accord with the current recommendations of autism researchers and autistic people, identity-first language will be used throughout this manuscript (see Bottema-Beutel et al., 2021 for further detail re: present guidance for use of terminology in referencing persons on the autism spectrum).

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Abstract

Purpose: Caregivers of autistic children present with high stress levels, which have been associated with poorer child outcomes in several domains, including language development. However, prior to the present study, it was unknown whether elevated caregiver stress was associated with language development in infant siblings of autistic children, who are at increased likelihood of receiving a future diagnosis of autism and/or language impairment compared to infant siblings of non-autistic children. The present study explored the degree to which, as well as the mechanisms by which, caregiver stress was linked with later language outcomes of infant siblings of autistic and non-autistic children (Sibs-autism and Sibs-NA).

Method: Participants were 50 infants (28 Sibs-autism; 22 Sibs-NA) aged 12-18 months at the first timepoint in this study (Time 1). Infants were seen again 9 months later, at 21-27 months of age (Time 2). Caregiver stress was measured via a validated self-report measure at Time 1. Caregiver language input, the putative mechanism by which caregiver stress may influence later language outcomes, was collected via two daylong recordings from digital recording (LENA) devices worn by the child at this same timepoint. Child language outcomes were measured via standardized and caregiver report measures at Time 2.

Results: Several models testing hypothesized indirect effects of caregiver stress on later child language outcomes through caregiver language input were statistically significant. Specifically, significant indirect effects suggest that (a) caregivers with increased stress tend to speak less to their infants, and (b) this reduced language input tends to covary with reduced child language outcomes later in life for Sibs-autism and Sibs-NA.

Conclusions: This study provides new insights into links between caregiver stress, caregiver language input, and language outcomes in Sibs-autism and Sibs-NA. Further work is necessary

to understand how to best support caregivers and optimize the language learning environments for infants.

Associations between Caregiver Stress and Language Outcomes in Infants with Autistic and Non-Autistic Siblings: An Exploratory Study

Autism is a neurodevelopmental condition characterized by differences in social communication, and by the presence of restricted, repetitive patterns of behavior, interests, or activities that impact an individual's ability to function in daily life (American Psychiatric Association, 2013). Features of autism typically emerge during the first few years of life and persist across the lifespan, potentially producing pervasive effects on the long-term outcomes of affected individuals (American Psychiatric Association, 2013). A large literature has shown, however, that the acquisition of language early in life is associated with increased social, educational, and vocational success for persons on the autism spectrum (e.g., Billstedt et al., 2005; Eaves & Ho, 2008).

The Role of the Caregiver in Language Development

Language development is shaped by a child’s early language environment and experiences (e.g., Gilkerson et al., 2018; Golinkoff et al., 2019; Hart & Risley, 1995; Vihman, 2014). The transactional model of language development posits that language skills are built upon “dynamic interactions” between a child and their caregiver, wherein the caregiver scaffolds communication bids and shapes language development around the infant’s experiences in their environment (e.g., Goldberg, 1977; Sameroff, 2009; see Woynaroski et al., 2014). Within this interactive process, as an infant gains language and communication skills, they engage in communicative exchanges to a greater extent and can thereby influence their caregivers to respond in a manner that further facilitates the infant’s language development (Fogel & Lyra, 1997; Hoff, 2006). Thus, this model emphasizes the bidirectional and interdependent effects of the child and their caregivers in language acquisition (Sameroff, 2009; Tamis-LeMonda et al., 2014).

Given the importance of early language in long-term outcomes of autistic children, a growing body of research has focused on elucidating how caregiver input may impact language development in children on the autism spectrum (e.g., Bang & Nadig, 2015; Haebig et al., 2013). Researchers have shown that caregiver language input is moderately associated with later vocabulary and broader spoken language skills of autistic and neurotypical children (e.g., Choi, Nelson et al., 2020; McDuffie & Yoder, 2010; McGillion et al., 2017; Yoder et al., 2015; for review, see Heidlage et al., 2020).

Evidence that Caregiver Stress is Linked with Child Language

Caregiver stress has been linked with language outcomes in autistic children and children with developmental conditions other than autism (e.g., Blank et al., 2020; Quittner et al., 2010; Roberts; 2019; Sarant & Garrard, 2014). Caregivers of autistic children experience high levels of stress relative to caregivers of non-autistic children, with one study finding that over 80% of caregivers of children on the spectrum feel “stressed beyond their limits” (Bitsika et al., 2013; Bonis, 2016). The stress that caregivers of autistic children experience is also significantly increased as compared to caregivers of children with other developmental conditions, such as Down syndrome (Bitsika & Sharpley, 2004; Bitsika et al., 2013; Estes et al., 2009; see Hayes & Watson, 2013 for a review). The impact of caregiver stress on child outcomes may be the greatest in the early stages of life, when parents have concerns about their child’s development (e.g., Bonis, 2016; Karp et al., 2017) and when children are most malleable and experiencing large qualitative changes in language development (Shonkoff & Phillips, 2000). A challenge to testing this hypothesis, however, is that autism cannot always be reliably diagnosed in the earliest stages of life (i.e., in infancy and toddlerhood; Ozonoff et al., 2015, 2018; Woolfenden et al., 2012).

Rationale for Focusing on Infant Siblings of Autistic Children

As a potential solution to the aforementioned problem, researchers often prospectively follow infants who are known to be at increased likelihood for a future diagnosis of autism based on having an autistic older sibling (Sibs-autism). Approximately one in five of these infant siblings will go on to be diagnosed with autism (Messinger et al., 2015; Ozonoff et al., 2011). Additionally, Sibs-autism who are not diagnosed with the condition are more likely to present with a language disorder, display below-average developmental functioning, and/or present with subclinical autistic features (e.g., Charman et al., 2017; Landa et al., 2012; Messinger et al., 2013). Researchers have not yet explored the degree to which caregiver stress is associated with language in Sibs-autism, despite multiple studies pointing to caregiver stress as an unexplored variable in the language outcomes of this population (Wan et al., 2012; Yirmiya et al., 2006).

Caregiver Language Input as a Putative Mechanism by Which Caregiver Stress May Influence Language Outcomes

At present, the mechanisms by which caregiver stress is associated with early language development and outcomes are not well-understood. Researchers have suggested, however, that caregivers who experience elevated stress may provide less language input to their child early in life, possibly contributing to poorer child language outcomes (e.g., Berryhill, 2016; Ellwood-Lowe et al., 2020; Wan et al., 2012; Yirmiya et al., 2006). Previously, there were limited options for measuring caregiver language input in a child’s everyday settings and, thus, for testing this hypothesis regarding how caregivers’ psychological and linguistic factors may collectively influence child language outcomes. The advent of day-long recording technology such as the

Language ENvironment Analysis (LENA) device and accompanying software, however, has now provided researchers with a tool that can be utilized to quantify the amount of adult language input occurring in the home environment (Gilkerson et al., 2017). A rapidly-growing body of literature has linked caregiver language input, as measured by LENA, to language development in typically-developing infants and children (e.g., Gilkerson et al., 2018; Leung et al., 2020; McGillion et al., 2017; Ramírez et al., 2020; Romeo et al., 2018).

LENA additionally has been used in research with autistic children, as well as in studies of infant siblings of autistic children (e.g., Seidl et al., 2018; Swanson et al., 2018, 2019; Warren et al., 2010; Woynaroski, 2014; Yoder et al., 2013). It has been demonstrated, for example, that adult word count (AWC), a quantitative estimate of caregiver language input as measured by LENA, correlates with concurrent expressive language skill in autistic children (Warren et al., 2010). The AWC score, therefore, provides an automated metric for testing caregiver language input as a putative mechanism of theorized relations between caregiver stress and future language outcomes in Sibs-autism.

Mediation Analysis as an Approach to Statistically Testing Putative Mechanisms

One approach to evaluating the mechanisms that may influence hypothesized relations between caregiver stress and future language outcomes is mediation analysis. In a mediation analysis, both direct and indirect effects on the dependent variable are assessed (Hayes, 2009). See Figure 1 for a conceptual model that depicts how our specific questions can be answered through mediation analysis. The direct effect of interest in this study is the potential relation between caregiver stress and future language outcomes (i.e., the c′ path). Through mediation analysis, the indirect effect – of caregiver stress on future language outcomes via caregiver language input – can also be evaluated. This indirect effect comprises (a) the relation between caregiver stress and caregiver language input (i.e., the a path), and (b) the relation between caregiver language input and future language outcomes, covarying caregiver stress (i.e., the b path). If the product of the a and b paths is found to be significant in our test of mediation (i.e., if the confidence interval for a*b does not include 0), we can conclude that the relation between caregiver stress and future language outcomes is mediated, or explained at least in part, by caregiver language input.

The Possibility That Associations of Interest May Differ for Sibs-autism versus Sibs-NA

Notably, Sibs-autism may differ from infants at general population-level likelihood for autism (Sibs-NA, i.e., infant siblings of non-autistic children) in language development as early as 12 months of age (Bryson, 2007; Choi, Shah, et al., 2020; Choi, Nelson, et al., 2020; Elison et al., 2013; Hazlett et al., 2017; Meera et al., 2020), regardless of the eventual presence or absence of an autism diagnosis. A growing body of literature documents that caregiver–child interactions also often differ between Sibs-autism and Sibs-NA, which may influence language development (e.g., Choi, Nelson et al., 2020; Choi, Shah et al., 2020; Leezenbaum et al., 2014; Northrup & Iverson, 2015; Wan et al., 2013; Yirmiya et al., 2006; but see Tager-Flusberg, 2016).

Further, caregivers of Sibs-autism may present with greater levels of stress, given their child's increased likelihood for later autism diagnosis and differences in language development when compared to Sibs-NA (e.g., Karp et al., 2017; Tager-Flusberg, 2016). As mentioned above, prior evidence suggests there is differential stress directly related to having an older autistic versus non-autistic older child (Bitsika et al., 2013; Bonis, 2016). Levels of stress may be further heightened among caregivers concerned by their infant exhibiting behaviors similar to their older autistic child, resulting in increased stress for caregivers of Sibs-autism (e.g., DesChamps et al., 2020; MacDuffie et al., 2020). However, we do not yet know how this relates to the stress associated with raising a younger infant sibling, or how such stress could influence concurrent caregiver language input as well as the infant's later language outcomes. Thus, it is important to consider that hypothesized associations between caregiver stress and child language outcomes, through caregiver language input, may differ for Sibs-autism versus Sibs-NA.

An approach to assess whether associations between caregiver stress and child language outcomes differ by group is moderation. Moderation allows for assessment of whether relations between two variables (e.g., caregiver stress and child language) significantly differ based on a third variable (e.g., sibling group). Additionally, a mediation model testing effects of interest (e.g., Figure 1) can be assessed for moderation in order to evaluate whether indirect effects vary based on factors such as sibling group. We hypothesized that associations of interest to the present study may be stronger in Sibs-autism due to the potential for increased heterogeneity in caregiver stress and child language outcomes in this population (e.g., Hayes & Watson, 2013; Messinger et al., 2013). Notably, such differential effects have been observed in several prior studies testing associations between theorized predictors and child outcomes in Sibs-autism and Sibs-NA (e.g., Bruyneel et al., 2019; Choi, Nelson et al., 2020; Romeo et al., 2021).

Purpose

The present study, therefore, explored the degree to which, and the mechanisms by which, caregiver stress is associated with later language outcomes of Sibs-autism and Sibs-NA. The specific research questions include:

1. Are there between-group differences (Sibs-autism versus Sibs-NA) in the degree of stress that caregivers report experiencing?

2. Is caregiver stress negatively associated with caregiver language input in Sibs-autism and Sibs-NA? Are these associations moderated by sibling group?

3. Is caregiver language input positively associated with later child language outcomes in Sibs-autism and Sibs-NA, covarying caregiver stress? Are these associations moderated by sibling group?

4. Does caregiver language input mediate the associations between caregiver stress and later child language outcomes in Sibs-autism and Sibs-NA? Are these associations moderated by sibling group?

Methods

Data for this study were drawn from a larger longitudinal investigation, the **S**ensory **P**roject in **I**nfant/Toddler **S**iblings of Children with Autism (Project SPIS; PI Woynaroski). All procedures were approved by the Vanderbilt University Institutional Review Board.

Participants

Participants were 28 Sibs-autism and 22 Sibs-NA, recruited for Project SPIS. The sample entirely overlaps with a previous report from our laboratory (Santapuram et al., 2022). All infants were between 12-18 months (±30 days) at study entry and were living in a primarily English-speaking household. Infants were excluded from participation if they had adverse neurological history, a known genetic condition, and/or preterm birth (gestation < 37 weeks). To be included in the Sibs-autism group, infants were required to have at least one older sibling who was diagnosed with autism (i.e., via a research-reliable administration of the Autism Diagnostic Observation Schedule [ADOS]; Lord et al., 2012). To be included in the Sibs-NA group, participants were required to have only non-autistic older siblings; non-autistic status in the older siblings was confirmed by screening below the threshold for autism risk on the Social Communication Questionnaire (Rutter et al., 2003). Additionally, infants in the Sibs-NA group were required to have no first-degree relatives with an autism diagnosis per caregiver report. All primary caregivers reported their highest level of formal education attained as a proxy for socioeconomic status (SES). Groups were matched on biological sex and chronological age. Sibling groups did significantly differ on cognitive, language, and adaptive behavior scores at Time 1. See Table 1 for a detailed summary of participant characteristics.

Procedures

All infants were seen at two time points. At the first visit, all infants were between 12-18 months. The second visit occurred nine months later (i.e., when participants were 21-27 months).

Measure of Caregiver Stress

Caregivers were asked to complete the Parenting Stress Index Short Form, fourth edition (PSI) at the first time point of the study (Abidin, 2012). The PSI is a validated, 36-item caregiver report measure that yields an overall raw score, a standardized T-score, as well as scores for various subscores (e.g., Haskett et al., 2006; Whiteside-Mansell et al., 2007; Zaidman-Zait et al., 2011). In responding to questions pertaining to the caregiver–child relationship, caregivers were instructed to focus on their interactions with the infant participating in the study. Prior work by Zaidman-Zait et al. (2011) on caregiver stress in families with autistic children suggested that the standardized, three-subscore version of the PSI was not optimal for characterizing caregiver stress in this population. Thus, in the present study we derived the overall score and the five subscores from Zaidman-Zait et al. (2011) for measuring caregiver stress in families with autistic children (i.e., general distress, parenting distress, rewards parent, child demandingness, and difficult child). The general distress subscore measures the broad stressors that a caregiver experiences. The parenting distress subscore measures the distress that a caregiver experiences that is specifically related to the caregiving role. The rewards parent subscore examines child characteristics that foster positive caregiver–child interactions. The child demandingness subscore measures caregiver perceptions that taking care of their child is unexpectedly difficult. Finally, the difficult child subscore measures child characteristics, such as emotional dysregulation and difficulty with adaptability, that could contribute to caregiver stress (for additional information on the five subscores, see Zaidman-Zait et al., 2011). These subscores have not been previously utilized in studies of Sibs-autism. However, the internal reliability of each subscore for the participants in this study was good to excellent (Cronbach's α range = .81–.85). The five PSI subscores were also moderately to strongly intercorrelated for the present sample (*r*s ranged from .37 to .79; see Table 2), providing empirical support for the notion that they tap the same superordinate construct (i.e., caregiver stress) but are not redundant in our population of interest. Ninety-six percent of PSI respondents in the Sibs-autism group were mothers, and 95% of PSI respondents in the Sibs-NA group were mothers (i.e., one father served as the primary caregiver in each sibling group). To index individual differences in caregiver stress for analyses, we derived the overall raw score, as well as the five raw subscores supported by Zaidman-Zait et al. (2011).

Measures of Caregiver Language Input

At the first time point in the study, families were additionally provided with two LENA recording devices and a specialized garment (i.e., vest) for the infant to wear during day-long recordings collected in each infant's everyday settings. Devices were worn for 16 hours, the maximum recording time for LENA processors, in the infant's home and community environments for two days (see Feldman et al., 2022 for more information). Caregivers were instructed to turn the device on when their child woke up in the morning and to leave the device on and in the garment pocket for the duration of the 16 hours. Recordings were analyzed using LENA Advanced Data EXtractor (ADEX) software to derive AWC scores. The scores were averaged across the two recording days to increase stability, and thus potential predictive validity, of the AWC variable (Feldman et al., 2022; Rushton et al., 1983).

There are some limitations inherent to the use of LENA hardware and software that should be noted. First, though LENA provides an estimate of adult words spoken in the presence of an infant (e.g., Oller, 2010), it cannot capture more fine-grained linguistic measures such as mean-length of utterance (MLU), MLU in morphemes, or linguistic diversity. Additionally, LENA does not capture other differences that may be of interest in the infant's language environment (e.g., primary caregiver versus other adult talk, infant-directed versus other types of adult talk), and therefore, one cannot assume that all adult words estimated by LENA in the AWC metric necessarily reflect a precise count of actual words spoken directly to the child by their caregiver.

Measures of Language Outcomes

To assess child language outcomes, the Mullen Scales of Early Learning (MSEL), the Vineland Adaptive Behavior Scales, second edition (VABS-2), and the MacArthur-Bates Communicative Development Inventories: Words and Sentences (MCDI) were collected at the second time point (ages 21-27 months). The MSEL is a norm-referenced assessment that evaluates language, as well as motor and cognitive (visual reception) abilities (Mullen, 1995). The receptive and expressive language age equivalency scores were derived for use in analyses. The VABS-2 is a norm-referenced assessment that measures adaptive behavior in several domains, including communication, via a semi-structured interview (Sparrow et al., 2005). The receptive and expressive communication age equivalency scores were derived for use in analyses. The MCDI is a parent-report measure of the words and sentences that a child can say

(Fenson et al., 2007). The raw number of words spoken (i.e., expressive vocabulary) was derived for use in analyses.

Analytic Plan

Prior to conducting analyses, data were imported into R (R Core Team, 2020) to assess normality. Any variables that were not normally distributed (i.e., skew > |1| or kurtosis > |3|) were transformed, and missing data (ranging from 0-16% missingness across all scores) were then imputed using the *missForest* package (Stekhoven & Bühlmann, 2012).

Aggregate receptive and expressive language scores were derived for each participant by averaging the relevant z-scores from the MSEL, VABS-2, and MCDI to increase the stability and, thus, the potential construct validity of the language outcomes (Feldman et al., 2021; Rushton et al., 1983). By aggregating assessments that capture various aspects of receptive and expressive language (e.g., the MCDI measures vocabulary, while the MSEL measures broader language), we can more comprehensively index language level in our sample (e.g., Feldman et al., 2021; Rogers et al., 2021; Santapuram et al., 2022). The expressive aggregate was the average of the z-scores for: (a) the age-equivalency score from the expressive language scale of the MSEL, (b) the age equivalency score for the expressive communication scale of the VABS-2, and (c) the expressive vocabulary raw score from the MCDI. The receptive aggregate was the average of the z-scores for: (a) the age equivalency score from the receptive language scale of the MSEL, and (b) the age equivalency score from the receptive communication scale on the VABS-2 (Table 3). Scores from the MSEL, VABS-2, and MCDI were sufficiently intercorrelated across participants to warrant aggregation (all *r*s > .79).

To answer the first research question, independent samples t-tests were conducted to evaluate whether stress varied between caregivers of the Sibs-autism and Sibs-NA groups. To answer the second and third research questions, a series of multiple regression models was carried out (a) to evaluate associations between caregiver stress as indexed by the PSI overall score and the five PSI subscores, and caregiver language input (i.e., a paths relevant to hypothesized indirect effects) and (b) to evaluate associations between caregiver language input and later child receptive and expressive language outcomes, covarying caregiver stress (i.e., b paths relevant to hypothesized indirect effects) as well as (c) to test whether the aforementioned associations of interest were moderated by sibling group.

To answer the final research question, mediation models were evaluated using the PROCESS macro (Hayes, 2017) in R to assess whether indices of caregiver language input, as measured by AWC, significantly mediated associations between caregiver stress as measured by either the PSI overall score or any of the five PSI subscores and later child receptive and expressive language. Sibling group was also evaluated as a moderator in models where prior analyses indicated that sibling group was a significant moderator of paths comprising the indirect effect to test whether the hypothesized mediation relations varied according to higher versus lower likelihood for a future autism diagnosis. Prior to running analyses relevant to our stated research questions, SES, as indexed by primary caregivers' level of formal education, was considered as a covariate. We did not correct for multiple comparisons, given the exploratory nature of the study and relatively small sample size (and, thus, limited power to estimate effects of interest).

Results

Consideration of SES as a Covariate in Analyses

Our proxy index for SES (i.e., caregiver level of formal education) was not significantly associated with caregiver stress (rs ≤ |.15|, p ≥ .29 across all indices of stress) or with child language outcomes; therefore, this variable was not included in subsequent statistical models.

Between-Group Differences in Caregiver Stress

Caregivers did not significantly differ by group, on average, on the overall raw score on the PSI, t = 1.85, p = .070, Cohen's d = 0.51, or on the additional five subscores (see Table 4). However, small to moderate effect sizes for between-group differences, in the anticipated direction, were observed across indices of caregiver stress.

Relations Between Caregiver Stress and Caregiver Language Input

A series of multiple regression models was run to assess relations between caregiver stress and concurrent caregiver language input (i.e., the a paths relevant to hypothesized indirect effects; Baron & Kenny, 1986; Hayes, 2009). Caregiver language input was unconditionally associated with the overall PSI score (zero-order correlation = -.28, p = .046; see Figure 2A), the rewards parent subscore (zero-order correlation = -.33, p = .018), and the child demandingness subscore (zero-order correlation = -.35, p = .012). These relations were not moderated by sibling group (see Table 5 for a summary of results relevant to a paths).

Relations Between Caregiver Language Input and Child Language Outcomes, Covarying Caregiver Stress

Another series of regression models was run to assess the relations between caregiver language input and later child language outcomes, covarying caregiver stress (i.e., the b paths relevant to hypothesized indirect effects; see Table 6). All relations between caregiver language input and later child language were statistically significant, covarying for all caregiver stress measures (p values < .05; see Figure 2B for a representative scatterplot); in all cases, higher caregiver language input was associated with greater child language at the second timepoint. These relations were not moderated by sibling group (see Table S1).

Indirect Effects of Caregiver Stress on Child Language Outcomes Via Caregiver Language Input

To assess whether caregiver stress was indirectly related to child language outcomes, through caregiver language input, a series of mediation analyses was conducted. Models were constructed to assess the relations between PSI scores (independent variables) and receptive and expressive language outcomes (i.e., receptive and expressive language aggregate scores; dependent variables) via AWC (putative mediator).

Mediation Models with Overall PSI Score as Independent Variable

The first series of mediation models employed the overall PSI score as the independent variable. These models revealed that caregiver language input as indexed by AWC significantly mediated the relations (a) between caregiver stress as indexed by the overall PSI score and expressive language, 95% CI [–0.0128, –0.0004] and (b) between caregiver stress as indexed by the overall PSI score and receptive language [–0.0161, –0.0009]. The direct effect of caregiver stress on later receptive and expressive language was not statistically significant when covarying for [the indirect effect of] AWC (i.e., the mediation model was complete; Hayes, 2009).

Mediation Models Evaluating Additional PSI Scores as Independent Variables

We ran additional, exploratory analyses to evaluate whether relations between other indices of caregiver stress derived from the PSI and later child language outcomes were significantly mediated by AWC. Several models were statistically significant. Specifically, the model assessing the indirect effect of the parenting distress subscore on receptive language, the models assessing the indirect effect of the rewards parent subscore on receptive and expressive language, and the models assessing the indirect effect of the child demandingness subscore on receptive and expressive language all yielded confidence intervals for the indirect effect that did not include zero (see Table 7 for a summary of relevant statistics; see S2 for additional mediation analyses that were run post hoc using alternative LENA variables likely to reflect language input specific to each infant's reported primary caregiver). These significant mediation models were complete, meaning that the direct effects of the aforementioned PSI subscores on later child language were non-significant when covarying AWC. Figure 3 depicts results of a representative mediation model. The direction of effects was similar across all significant indirect effects, such that increased caregiver stress was associated with reduced caregiver language input, which covaried with lower child language at outcome assessment, covarying caregiver stress as measured at study entry.

Moderated Mediation Models

We did not conduct moderated mediation models due to prior analyses indicating that moderated effects in the a (i.e., the effects of caregiver stress on AWC) and b (i.e., the effects of AWC on later language, covarying entry-level caregiver stress) paths in our regression models were not moderated by sibling group.

Discussion

This study sought to evaluate hypothesized associations between caregiver stress, caregiver language input, and later language outcomes in younger siblings of autistic and non-autistic children (i.e., Sibs-autism and Sibs-NA). Our results suggest that caregivers of Sibs-autism and Sibs-NA may not significantly differ on mean levels of reported stress. However, findings indicate that several aspects of caregiver stress may indirectly influence later child language outcomes, not only in Sibs-autism, but also in Sibs-NA.

Caregiver Language Input Mediates Associations between Caregiver Stress and Language Outcomes

Mediation analyses indicated that caregiver stress influenced later child receptive and expressive language indirectly through caregiver language input as indexed by Adult Word Count (AWC). In all significant mediation models, the indirect effect of caregiver stress on future child language via caregiver language input as indexed by AWC did not vary according to sibling group. This finding supports prior work demonstrating that caregiver language input is a salient factor for language learning across risk groups (e.g., Bang & Nadig, 2015; Choi, Nelson et al., 2020; Gilkerson et al., 2018; Hirsh-Pasek et al., 2015; Hoff, 2006; Rowe, 2012), and furthers our understanding of how caregiver stress may influence later language outcomes in young children (Swanson et al., 2019).

Caregiver Stress Does Not Differ by Sibling Group

To our knowledge, this is the first study to explore caregiver stress in infant siblings of autistic children. Caregivers of Sibs-autism did not report experiencing statistically significantly more stress than caregivers of Sibs-NA, a result that is seemingly inconsistent with previous work in caregivers of autistic children (Bitsika et al., 2013; Bonis, 2016; for review, see Hayes & Watson, 2013). However, caregiving stress in the Sibs-autism population is complex in nature (e.g., DesChamps et al., 2020; MacDuffie et al., 2020). Though some caregivers may experience more stress due to the knowledge that there is a higher occurrence of autism in Sibs-autism, other caregivers of Sibs-autism may feel less stress regarding their relationship with their infant because they feel more equipped to identify developmental concerns should they arise. Notably, our analyses may have simply been underpowered to detect true between-group differences, given that the direction and size of effects were in the expected direction and were small to moderate in magnitude. Larger sample sizes are necessary to ascertain, with a higher level of confidence, whether caregivers of Sibs-autism, on average, experience higher levels of stress. Finally, the use of the PSI only allowed for examination of stress specifically related to caregiving. Other stressors, unrelated to caregiving, should be examined in future work to determine whether additional life stressors differ between caregivers of Sibs-autism and Sibs-NA.

Clinical Implications

The present results suggest that it may be important to intervene when caregivers of infants at high and low likelihood for autism are experiencing elevated stress, to mitigate potential indirect influences of such stress on child language acquisition. Fortunately, there are practices and interventions known to reduce caregiver stress. For example, in a review of stress in caregivers of autistic children, Bonis (2016) found that caregiver-led support groups were effective in remediating the stress that caregivers experience. Additionally, caregivers who utilized respite services reported lower stress levels (Bonis, 2016). Weitlauf et al. (2020) recently demonstrated, in the context of a randomized controlled trial, that mindfulness-based stress reduction provided in tandem with a parent-implemented naturalistic developmental behavioral intervention (NDBI) may reduce caregiver stress relative to receiving training in the use of NDBI strategies alone. Thus, there are a number of potential approaches that display promise for reducing stress in caregivers of infants and young children.

Use of the PSI with Infant Siblings

To our knowledge, this is the first study to use the PSI in a group design with infant siblings, despite prior use of this instrument for measuring stress in caregivers of autistic children (Zaidman-Zait et al., 2011). Given this novel use of the PSI, we conducted exploratory analyses to test whether different indices of caregiving stress were more strongly associated with outcomes of interest. We found that mediation models employing parenting distress, rewards parent, and child demandingness subscores as predictor variables were statistically significant. This suggests that the stressors in a caregiver–child relationship that may be driving associations between stress, caregiver language input, and child language may be more nuanced and specific than just caregiver stress, broadly speaking. Our post-hoc results, further, suggest that the PSI may be useful for identifying specific areas wherein caregivers of Sibs-autism (and Sibs-NA) are experiencing high amounts of stress that could cascade onto the developmental outcomes of their infants, and for providing targeted support and referrals to mental health services as necessary.

Further, there is ongoing discussion regarding the factor structure of the PSI and whether it is valid for measuring caregiver stress in families with a child/children who are not neurotypical. Though we used five subscores of the PSI as supported by Zaidman-Zait et al. (2011) in our analyses, to our knowledge no one has previously studied the factor structure of the PSI in caregivers of Sibs-autism. Thus, it is possible that the psychometric properties of this measure may vary across different clinical and at-risk populations, especially given the potential unique challenges of caregiving a child with or at high likelihood for autism (Bitsika et al., 2013; Bonis, 2016; Hayes & Watson, 2013).

Use of LENA with Infant Siblings

LENA has been used in many prior studies of infants to unobtrusively measure the home language environment, but few studies have investigated longitudinal links between early LENA variables and later language in Sibs-autism (i.e., Seidl et al., 2018; Swanson et al., 2018). Despite the limitations of LENA mentioned above, a growing body of research suggests that LENA may be an ecologically valid measure of infants' home language environments without great cost or time to researchers (e.g., Seidl et al., 2018; Swanson et al., 2018), and these novel findings for Sibs-autism and Sibs-NA provide a valuable starting point for researchers interrogating similar questions in the infant sibling population using daylong recordings. Nonetheless, further work should evaluate the use of AWC and daylong recordings as compared to other, laboratory-based measurements of caregiver language input; current studies of infant siblings typically use one of these measurement options but not both (e.g., Choi, Nelson et al., 2020; Choi, Shah et al., 2020; Romeo et al., 2021; Swanson et al., 2018).

Limitations and Future Directions

This study provides novel insights into the mechanisms by which caregiver stress may influence child language outcomes but has several limitations. First, we measured two constructs of interest, caregiver stress and caregiver language input, at the same time-point. Although we found support for associations between caregiver stress and concurrent caregiver language input, additional work is needed to determine whether higher levels of caregiver stress precede and predict reduced caregiver language input. A study design wherein caregiver language input is measured at a later timepoint, rather than concurrently, would establish temporal precedence for all constructs comprising theorized mediation relations and thereby increase our confidence in the indirect effects observed.

Additionally, the present study did not consider which infants in the Sibs-autism group went on to be diagnosed with autism. Our team is continuing to follow the participants in the present study longitudinally, with a plan to evaluate whether associations between caregiver stress, caregiver language input, and later child language outcomes do vary according to diagnostic outcome, versus simply familial likelihood for a future autism diagnosis. Subsequent studies may also include consider whether autism severity influences any effects of interest, such as the amount of caregiver stress that is reported or associations between stress and developmental outcomes.

Further, our participants were largely homogenous in race and ethnicity, with the majority of families being White and not Hispanic/Latinx. Future work would benefit from exploring these associations in more diverse samples, as the present results may not generalize to all families.

Finally, a limitation of this work is that most of our statistical analyses were run without correcting for multiple comparisons, which has increased the likelihood of Type I error. However, this research was designed in an exploratory manner to serve as a proof-of-principle for later studies. Our hope is that these results provide a foundation for future research wherein specific findings from this study can be leveraged to test a smaller number of a priori-specified hypotheses with larger sample sizes.

Conclusion

The findings of the present study advance our understanding of the links between caregiver stress, caregiver language input, and later language outcomes in infant siblings of autistic and non-autistic children. Our results indicate that caregiver stress may indirectly influence child language outcomes through caregiver language input in infants at higher and lower likelihood for a future diagnosis of autism. Additional research is necessary to understand how we can best support caregivers and optimize the early language learning environment for infants.

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Data Availability Statement

The datasets generated during and/or analyzed during the current study are available from the corresponding author on request.

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Supplemental File Descriptions

S1. Table S1 depicts associations between caregiver language input and later language, as moderated by sibling group. These associations covaried for caregiver stress.

S2. Table S2 describes additional mediation analyses that were run post hoc to examine LENA variables that were specific to each infant's reported primary caregiver.

Figure Captions

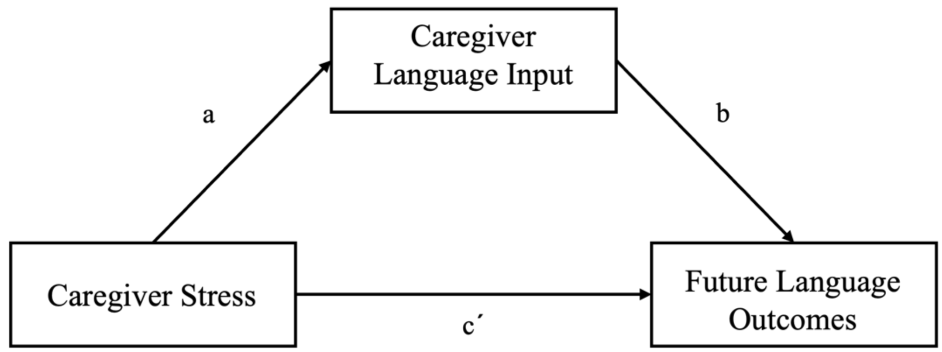

Figure 1. Figure depicts a conceptual mediation model designed to test hypothesized indirect effects of caregiver stress on future language outcomes through caregiver language input. a = the relation between caregiver stress and caregiver language input; b = the relation between caregiver language input and later child language, covarying caregiver stress; c′ = the direct effect of caregiver stress on later language, covarying caregiver language input.

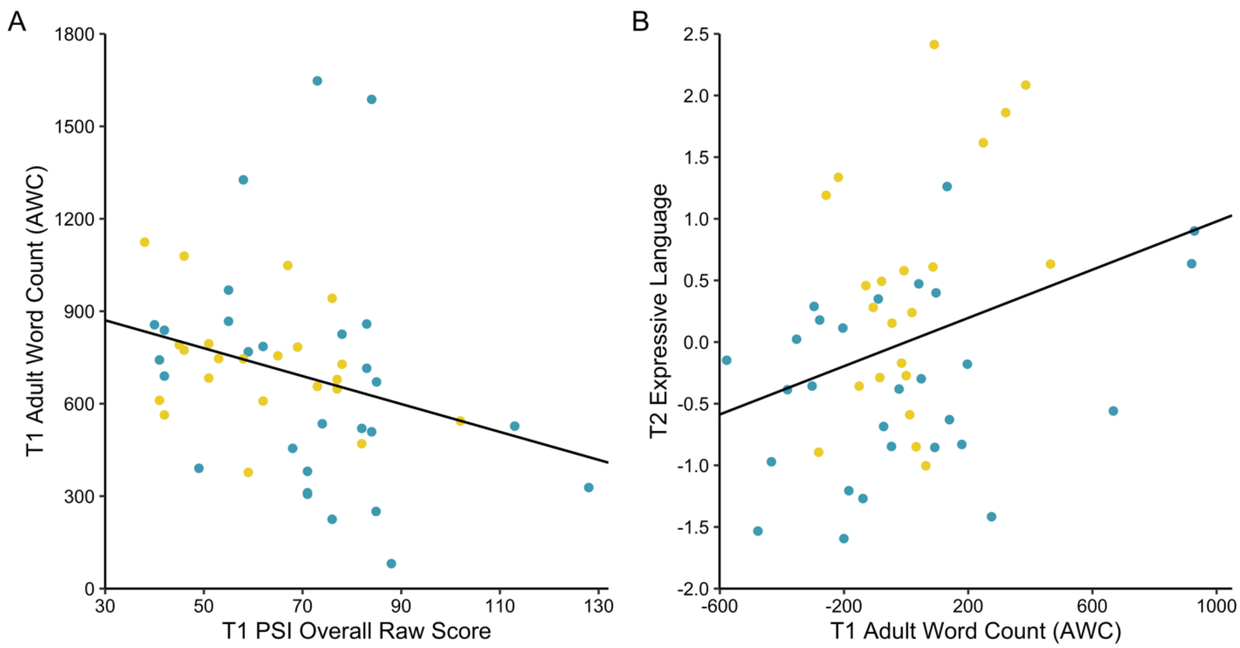

Figure 2. Figure depicts representative scatterplots for relations comprising indirect effects across infant siblings of autistic and non-autistic siblings (represented by blue and gold dots, respectively). T1 = Time 1 (12-18 mos), PSI = Parenting Stress Index Short Form, fourth edition (Abidin, 1995), T2 = Time 2 (21-27 mos), A = the relation between caregiver stress as indexed by the overall raw score from the PSI and caregiver language input as indexed by Adult Word Count (AWC), B = the relation between AWC and later child expressive language when covarying caregiver stress. Relations for all paths were in the anticipated directions, such that increased caregiver stress was associated with reduced caregiver language input, and reduced caregiver language input covaried with lower child language levels.

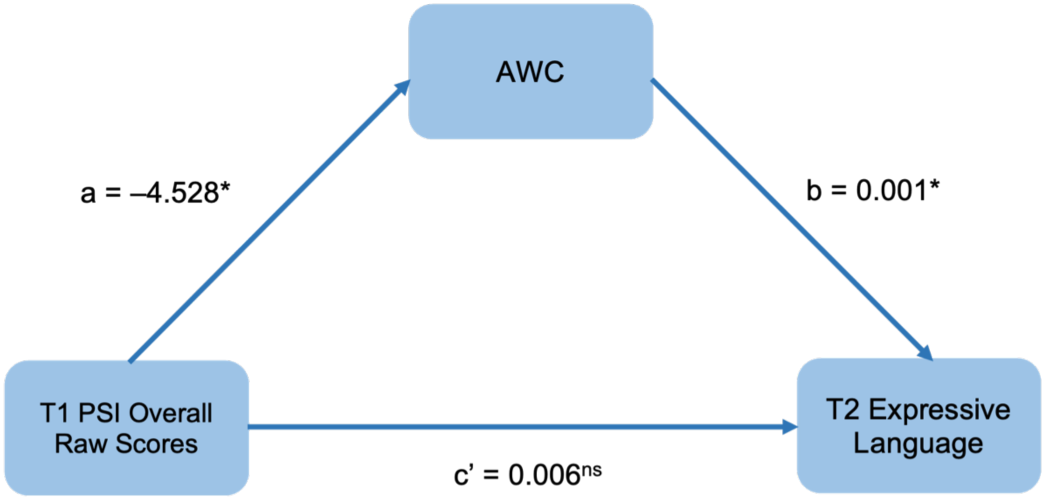

Figure 3. Figure depicts a representative mediation relation. T1 = Time 1 (12-18 mos), PSI = Parenting Stress Index Short Form, fourth edition (Abidin, 1995), T2 = Time 2 (21-27 mos), a = the relation between caregiver stress and caregiver language input as indexed by Adult Word Count (AWC), b = the relation between AWC and later child expressive language, covarying caregiver stress, c′ = the direct effect of caregiver stress on later expressive communication, covarying AWC. Note that c′ is non-significant, meaning that the association between caregiver stress and later child expressive communication is completely mediated by AWC. This indirect effect is not moderated by sibling group. All values are unstandardized coefficients.

*$p < .05$, ns = non-significant result.

Tables

Table 1

Participant Demographics by Sibling Group

Time 1 Variables	Sibs-autism (n = 28) *M* (*SD*) Min-Max	Sibs-NA (n = 22) *M* (*SD*) Min-Max	*p*-value
Age in Months	13.71 (1.84) 11-18	14.05 (2.13) 11-18	.565
MSEL Early Learning Composite**	89.56 (13.28) 70-118	100.14 (8.02) 84-121	.001
MSEL Receptive Language	11.48 (2.08) 8-17	12.73 (3.38) 8-19	.145
MSEL Expressive Language*	11.92 (2.40) 6-16	13.95 (3.54) 8-20	.026
VABS-2 Receptive Communication*	12.07 (3.66) 1-21	16 (5.22) 12-30	.010
VABS-2 Expressive Communication***	12.11 (3.65) 4-18	15.90 (3.21) 9-22	<.001
VABS-2 Adaptive Behavior Composite*	93.23 (11.44) 73-116	103.4 (6.33) 94-115	.001
MCDI Expressive Vocabulary*	8.04 (7.03) 0-30	22.23 (18.28) 0-63	.002
	N	*n*	
Sex	15 Male 13 Female	11 Male 11 Female	.801
Race	28 White	20 White	.266

		1 Black/African-American 1 Multiple	
Ethnicity	1 Hispanic/Latino 27 Not Hispanic/Latino	1 Hispanic/Latino 21 Not Hispanic/Latino	.862
Primary Caregiver's Highest Level of Education	2 High School Diploma or GED 10 College/Technical (1-2 Yrs) 8 College/Technical (3-4 Yrs) 5 Graduate/Professional School (1-2 Yrs) 3 Graduate/Professional School (3+ Yrs)	3 College/Technical (1-2 Yrs) 7 College/Technical (3-4 Yrs) 5 Graduate/Professional School (1-2 Yrs) 7 Graduate/Professional School (3+ Yrs)	.146

Note. Time 1 = 12-18 months, MSEL = Mullen Scales of Early Learning (Mullen, 1995), VABS = Vineland Adaptive Behavior Scales, Second Edition (Sparrow et al., 2005); MCDI = MacArthur Communicative Development Inventories, Words and Sentences (Fenson et al., 2007).
*Groups differed at $p <.05$.
** Groups differed at $p < .01$
***Groups differed at $p < .001$.

Table 2

Intercorrelations Between PSI Factors

Factor	1	2	3	4
1. General Distress				
2. Parenting Distress	.67***			
3. Rewards Parent	.57***	.56***		
4. Child Demandingness	.37**	.53***	.66***	
5. Difficult Child	.51**	.54***	.74***	.79***

Note. PSI = Parenting Stress Index Short Form, Fourth Edition (Abidin, 1995). Subscores were derived using guidelines from Zaidman-Zait et al. (2011).
**$p < .01$, ***$p < .001$.

Table 3

Summary of Key Study Constructs, Measures, and Variables According to Research Question

Construct	Measure/s	Variable(s)	Role Per Research Question	Measurement Period
Sibling Group	Demographic form, parent report, ADOS, SCQ	Infant sibling of (a) autistic child/ren, as confirmed with the ADOS, or (b) only non-autistic, neurotypical child/ren, as confirmed via score below threshold for autism concern (i.e., score of 15) on the SCQ	IV (RQ 1), Moderator (RQ 2,3,4)	Time 1
Caregiver Stress	PSI	(a) overall PSI raw score (b) general distress subscore (c) parenting distress subscore (d) rewards parent subscore (e) child demandingness subscore (f) difficult child subscore	DV (RQ 1), Predictor (RQ 2,4) Covariate (RQ 3)	Time 1
Caregiver Language Input	LENA	Average of scores across two recordings for Adult Word Count	DV (RQ 2), Predictor (RQ 3), Mediator (RQ 4)	Time 1
Later Receptive Language	MSEL VABS-2	Average of z scores for Time 2: (a) MSEL receptive age equivalency (b) VABS-2 receptive age equivalency	DV (RQ 3,4)	Time 2
Later Expressive Language	MSEL VABS-2 MCDI	Average of z scores for Time 2: (a) MSEL expressive age equivalency (b) VABS-2 expressive age equivalency (c) # words "child says" on MCDI	DV (RQ 3,4)	Time 2

Note. RQ = research question, DV = dependent variable, IV = independent variable, ADOS = Autism Diagnostic Observation Schedule, Second Edition (Lord et al., 2012), SCQ = Social Communication Questionnaire (Rutter et al., 2003), PSI = Parenting Stress Index Short Form, Fourth Edition (Abidin, 1995), LENA = Language ENvironment Analysis, MSEL = Mullen Scales of Early Learning (Mullen, 1995), VABS-2 = Vineland Adaptive Behavior Scales (Sparrow et al., 2005), Second Edition, MCDI = MacArthur Communicative Development Inventories, Words and Sentences (Fenson et al., 2007).

Table 4

Comparison of Caregiver Stress by Sibling Group

Variable	Sibs-autism (n = 22)	Sibs-NA (n = 28)				
	M (*SD*)	*M* (*SD*)	*t*	*p*	Cohen's *d*	95% CI
PSI Overall Raw Score	73.04 (20.37)	64.27 (15.16)	1.85	.070	0.51	[–1.12, 20.52]
PSI General Distress	17.43 (6.44)	14.45 (5.05)	1.83	.073	0.51	[–0.39, 6.34]
PSI Parenting Distress	11.21 (4.27)	10.14 (3.48)	0.98	.331	0.27	[–1.18, 3.34]
PSI Rewards Parent	13.19 (4.25)	11.27 (3.15)	1.82	.073	0.50	[–0.27, 4.10]
PSI Child Demandingness	8.25 (3.43)	7.55 (2.87)	0.79	.433	0.22	[–1.13, 2.54]
PSI Difficult Child	14.29 (5.49)	12.27 (3.83)	1.54	.130	0.42	[–0.72, 4.75]

Note. PSI = Parenting Stress Index Short Form, Fourth Edition (Abidin, 1995). Subscores were derived using guidelines from Zaidman-Zait et al. (2011).

Table 5

Zero-Order and Moderated Associations between Caregiver Stress and Caregiver Language Input

	Zero-order correlation		Values from Full Multiple Regression Model Predicting Adult Word Count		
Caregiver Stress	*r*	95% CI	β PSI	β Sibling Group	β PSI* Sibling Group
PSI Overall Raw Score	–.28*	[–.52, .01]	–.37	–.10	.14
PSI General Distress	–.10	[–.37, .18]	.05	.20	–.16
PSI Parenting Distress	–.25	[–.50, .03]	–.34	–.04	.13
PSI Rewards Parent	–.33*	[–.56, –.06]	–.87*	–.63	.75
PSI Child Demandingness	–.35*	[–.57, –.08]	–.28	.12	–.09
PSI Difficult Child	–.22	[–.47, .06]	–.17	.10	–.06

Note. Adult Word Count was derived from LENA (Language ENvironment Analysis). PSI = Parenting Stress Index Short Form, Fourth Edition (Abidin, 1995), CI = Confidence Interval, β = Standardized correlation coefficients from multiple regression models. These results correspond to the "a paths" in our hypothesized indirect effects model.
*$p < .05$.

Table 6

Associations between Caregiver Language Input and Language Covarying Caregiver Stress

	Expressive Language		Receptive Language	
	β PSI [95% CI]	β AWC [95% CI]	β PSI [95% CI]	β AWC [95% CI]
PSI Overall Raw Score	.12 [-.16, .39]	$.37^{*}$ [.08, .58]	.03 [-.25, .31]	$.53^{***}$ [.27, .69]
PSI General Distress	.09 [-.19, .36]	$.35^{*}$ [.07, .57]	.07 [-.21, .35]	$.52^{***}$ [.28, .70]
PSI Parenting Distress	.21 [-.06, .47]	$.39^{**}$ [.11, .60]	.17 [-.09, .45]	$.56^{***}$ [.31, .71]
PSI Rewards Parent	.11 [-.18, .38]	$.37^{*}$ [.08, .57]	.02 [-.26, .30]	$.52^{***}$ [.26, .68]
PSI Child Demandingness	.09 [-.19, .36]	$.37^{*}$ [.07, .57]	–.01 [-.28, .27]	$.52^{***}$ [.25, .68]
PSI Difficult Child	.05 [-.23, .32]	$.35^{*}$ [.07, .56]	–.06 [-.34, .22]	$.51^{***}$ [.26, .68]

Note. PSI = Parenting Stress Index Short Form, Fourth Edition (Abidin, 1995), *β* = Standardized correlation coefficients from multiple regression models, AWC = Adult Word Count derived from LENA (Language ENvironment Analysis), CI = Confidence Interval. These results correspond to the "b paths" in our hypothesized indirect effects model.

$^{*}p < .05$, $^{**}p < .01$, $^{***}p < .001$

Table 7

95% Confidence Intervals for Mediation Models Assessing the Indirect Effect of Caregiver Stress on Later Child Language as Mediated by Adult Word Count (AWC)

PSI Factor	Expressive Language 95% CI for Indirect Effect	Receptive Language 95% CI for Indirect Effect
Overall Raw Score	**[–0.0128, –0.0004]**	**[–0.0161, –0.0009]**
General Distress	[–0.0301, 0.0126]	[–0.0408, 0.0161]
Parenting Distress	[–0.0706, 0.0005]	**[–0.0786, –0.0013]**
Rewards Parent	**[–0.0912, –0.0096]**	**[–0.1149, –0.0252]**
Child Demandingness	**[–0.0829, –0.0127]**	**[–0.1012, –0.0229]**
Difficult Child	[–0.0403, 0.0032]	[–0.0513, 0.0044]

Note. Bolded values indicate 95% confidence intervals for the indirect effect that do not cross zero (i.e., significant mediation models). PSI = Parenting Stress Index Short Form, Fourth Edition (Abidin, 1995). Scores were derived using guidelines from Zaidman-Zait et al. (2011). Expressive language = aggregates generated from the Mullen Scales of Early Learning (MSEL; Mullen, 1995), Vineland Adaptive Behavior Scales, Second Edition (VABS-2; Sparrow et al., 2005) and MacArthur Communicative Development Inventories, Words and Sentences (MCDI; Fenson et al., 2007). Receptive language = aggregate score generated from the MSEL and VABS-2 (see Table 3 for more information on aggregate generation).

Table S1

Moderated Associations between Caregiver Language Input and Language Covarying Caregiver Stress

	Values from Full Multiple Regression Model Predicting Expressive Language				Values from Full Multiple Regression Model Predicting Receptive Language			
	β PSI	β AWC	β Sibling Group	β AWC * Sibling Group	β PSI	β AWC	β Sibling Group	β AWC * Sibling Group
PSI Overall Raw Score	0.26*	–0.23	–0.10	0.90	0.17	0.19	0.17	0.51
PSI General Distress	0.22	–0.27	–0.10	0.89	0.20	0.14	0.17	0.53
PSI Parenting Distress	0.29*	–0.23	–0.12	0.90	0.24*	0.18	0.15	0.53
PSI Rewards Parent	0.21	–0.10	0.03	0.69	0.14	0.27	0.26	0.37
PSI Child Demandingness	0.18	–0.29	–0.20	0.98	0.06	0.16	0.14	0.51
PSI Difficult Child	0.16	–0.24	–0.10	0.86	0.05	0.18	0.18	0.46

Note. PSI = Parenting Stress Index Short Form, Fourth Edition (Abidin, 1995), β = Standardized correlation coefficients from multiple regression models, AWC = Adult Word Count derived from LENA (Language ENvironment Analysis). These results correspond to the “b paths” in our hypothesized indirect effects model.

*$p < .05$.

S2

Additional mediation analyses were run wherein only the Adult Word Count (AWC) for the primary caregiver (i.e., the caregiver who filled out the Parenting Stress Index Short Form, Fourth Edition [PSI]) was used as the mediator by extracting the relevant Male Adult Near (MAN) and Female Adult Near (FAN) estimated adult word counts for each participant (MAN $n = 2$). We found significant indirect effects such that the language input of the primary caregivers significantly mediated the relations between the rewards parent and child demandingness subscores, respectively, and receptive language (rewards parent 95% CI = [-0.0629, -0.0041], child demandingness 95% CI = [-0.0718, -0.0088]). The indirect effect between caregiver stress and receptive language was complete in both models, meaning that the direct effect of caregiver stress on later receptive and expressive language was non-significant when covarying primary caregiver AWC.

There are a few factors that may have contributed to the lack of significant findings for several models when the putative mediator was the AWC for only the biological sex matching the primary caregiver. First, given that only two fathers were reported as the primary caregiver in this sample (1 Sibs-autism, 1 Sibs-NA), there may not be enough diversity in caregivers for this distinction to be meaningful. Second, the LENA recordings indicated that female speakers in the environment talked significantly more than male speakers, $t(44) = 9.35$, $p < .001$, $M_{\text{diff}} = 327.0$ words/hour. Closer inspection of the data shows that there were, further, more female than male adult words produced near the child for one of the two infants in the dataset whose father served as the primary caregiver. It is possible that this family may not have recorded truly representative days or that language input for this child comes predominantly from the mother or other adult females despite the fact that the father is considered the primary caregiver. Finally, by parsing the adult word count by primary caregiver’s sex, we may have altered the stability of our variables and, thus, our ability to detect effects of interest (see Feldman et al., 2022).