PARENTING STRESS AND LANGUAGE DEVELOPMENT IN CHILDREN

Associations in Angelman Syndrome and Down Syndrome

Abstract

One of the defining characteristics of neurogenetic syndromes such as Angelman syndrome (AS) and Down syndrome (DS) is delayed language development. Although it is commonly reported that parenting stress is associated with language development, these associations have not been widely studied in AS and DS despite other research showing elevated stress levels in the parents of these children. To fill this gap in research, the present study examined how parenting stress relates to language production in children with AS and DS. Daylong recordings were obtained from 72 participants using a Language Environment Analysis recording device, which was then processed through an online program to produce vocal productivity scores. Parents of these participants filled out the Parenting Stress Index, Fourth Edition Short Form to report their stress levels. Using linear regression, data was analyzed to determine if there is an association between language levels and parenting stress in a low-risk (LR) group and if the magnitude of that association was stronger in either the AS group or the DS group than in the LR group. Results suggested trends toward negative associations between parenting stress and language levels, though the association was not statistically significant. The magnitude of these associations did not differ significantly between groups. This negative association should be examined more with larger sample sizes, as it is important to the future development of early intervention techniques for children with AS and DS and their families.

Keywords

language development, psychology, child development, neurodevelopmental disorders, Angelman syndrome, Down syndrome

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INTRODUCTION

Language is an important indicator of long-term functional and social development, which helps build communication skills and affects emotional and behavioral needs as well (Helland & Helland, 2017). Children with neurogenetic disorders are more likely to experience deficits in language development than typically developing children. Down syndrome (DS) and Angelman syndrome (AS) are two such neurogenetic disorders that impact cognitive functioning, including language development.

Previous research has suggested that child language development is affected by environmental factors such as socioeconomic status and parenting stress (Noel, Peterson, & Jesso, 2007). Although having a child with a genetic syndrome is associated with increased levels of parenting stress (Adams et al., 2018; Almogbel, Goyal, & Sansgiry, 2017) and parenting stress has been shown to have a considerable effect in the development of language levels in children with other developmental conditions such as hearing loss (Blank, Frush Holt, Pisoni, & Kroenenberger, 2020), little is known about the effect of parenting stress on language development in genetic syndromes. This study aims to examine the association between parenting stress and language development in children with neurogenetic syndromes, specifically DS and AS.

Parenting Stress and Language Levels

While previous research shows that environmental factors such as socioeconomic status (Kim, Puranik, & Otaiba, 2015) and parental depression (D'Souza, Lathan, Karmiloff-Smith, & Mareschal, 2020) affect children's cognitive development, the association between parenting stress and cognitive outcomes in AS and DS is unclear. Studies of other neurodevelopmental groups suggest, however, that parenting stress may moderate developmental outcomes in high-risk youths (i.e., those who are at high risk for developmental delays). For example, Almogbel et al. (2017) studied the associations between parenting stress and functional impairment in children with neurodevelopmental disorders of mixed etiologies. The authors recruited 150 participants diagnosed with a variety of neurodevelopmental conditions, including autism, AS, and ADHD, and found that

higher functional impairment in children was associated with higher parenting stress. However, "functional impairment" is a broad term, and the authors did not specify whether this term included any aspects of language development. Therefore, although this study suggests that parenting stress may be relevant to child development in high-risk groups, the specific nature of these associations remains unclear.

A number of studies have also examined how parenting stress relates to language in other neurodevelopmental groups. For example, Blank et al. (2020) examined how parenting stress affects language comprehension and inhibitory control in children with and without hearing loss. They found that higher parenting stress was associated with lower language comprehension and inhibitory control in children with hearing loss but not in children without hearing loss. Importantly, this finding suggests that associations between parenting stress and clinical outcomes may differ in high- and low-risk populations, although further studies are needed to determine whether these findings would extend to groups such as DS and AS. Additional work is also needed to understand whether these patterns extend to language development more broadly. Language comprehension differs from language development, as language comprehension focuses on whether the individual understands language. Language development, on the other hand, is focused on whether an individual actively produces speech; therefore, it is unclear whether language development and comprehension are similarly related to parenting stress.

The association between parenting stress and language development in DS and AS may also be informed by prior studies of other high-risk groups. For example, Noel, Peterson, and Jesso (2007) looked at how parenting stress and child temperament affects language levels among preschool children from low socioeconomic status backgrounds. Expressive and receptive vocabulary of 56 preschool children were tested using the Peabody Picture Vocabulary Test and the Expressive Vocabulary Test, while parenting stress was measured through the Parenting Stress Index—short form, 3rd edition. These authors found that low levels of parenting stress were related to higher levels of child expressive and receptive vocabulary. These findings further suggest that parenting stress may be a salient moderator of language development in high-risk samples.

Characterizing Associations in Neurogenetic Groups

Children with rare disorders are also a high-risk group in terms of language development and parenting stress. Here, we focus on two specific neurogenetic subgroups: DS and AS. DS is a well-known neurogenetic disorder, present in approximately 1 out of 800 individuals globally (Bull, 2020). It is caused by a third copy of chromosome 21, which leads to the overexpression of more than 500 genes and an excess of protein development (Ruparelia & Mobley, 2015). This affects physical, social, and cognitive development (Bull, 2020). Sokol and Fey (2013) found that with regard to speech development, children with DS actually outperform those who have similar overall cognitive ability at age 2, suggesting that deficits in language development seen in individuals with DS, such as vocabulary development, may be more attributed to auditory delays than speech-production skills.

AS is a neurogenetic syndrome that presents severe deficits in cognitive, motor, and language development, affecting about 1 in 15,000 individuals. The effects of AS are usually seen within the first year of life, and depending on the severity of the disorder, children develop fewer than 20 words to no language at all. Clayton-Smith (1993) found that 30% of individuals with AS do not use any spoken language to communicate. Furthermore, children with AS incorporate fewer gestures into their communication (Grieco et al., 2018). In typical development, utterances are often combined with gestures that enhance language development, as gesturing is used to communicate and learn new words. This suggests that language delays may be part of a broader pattern of communication delays in AS. While both AS and DS present deficits in intellectual and language development, AS tends to have more profound effects in both areas (Margolis, Sell, Zbinden, & Bird, 2015).

Previous research has demonstrated that the associations between parenting stress and child development may vary between high- and low-risk groups (Blank et al., 2020). Characterizing the unique experiences of families affected by rare syndromes is important for several reasons. First, these families typically report higher levels of stress than parents of children without disabilities (Craig et al., 2016), likely reflecting that children with these conditions demonstrate high rates of challenging

behaviors (Craig et al., 2016), comorbid medical conditions (Alabaf et al., 2019), and mental health needs (Morgan, Leonard, Bourke, & Jablensky, 2008). Families of children with rare disorders are also often resilient to these challenges. For example, most caregivers report that they have more open communication as a result of having a child with a rare disorder, which leads to faster adaptation (Hall et al., 2012). As such, it is possible that the relationship between child language and caregiver stress may function differently in these groups relative to families of children with hearing loss or those with lower socioeconomic status backgrounds. Without characterizing these specific associations, it will remain unclear how to best support families to optimize their child's development and parental well-being.

Current Study

There is little research on whether parenting stress is associated with the development of language levels in children with neurogenetic disorders. This study examines how parenting stress specifically affects the development of language levels in children with AS and DS compared to a low-risk (LR) control group. We aim to answer the following questions: (1) Do the AS and DS groups differ from the LR group in vocal productivity (VP)? (2) Does the association between VP and parenting stress differ in the AS and DS groups compared to the LR group? We predicted that the AS group and the DS group will have significantly lower VP than the LR group and that the magnitude of the association between VP and parenting stress will be stronger in both the AS group and the DS group than in the LR group.

METHODOLOGY

Participants

Participants included 72 children: 34 LR, 20 DS, and 18 AS, aged 6.37 to 46.85 months. Of these 72 participants, 7 were Hispanic (9.7%), and 65 were non-Hispanic (90.3%). Sixty-four of the participants were white (88.9%), 7 were multiracial (9.7%), and 1 was Asian (1.4%) (Table 1). Participants were recruited via posts on social media as well as through research registries including the Global Angelman Registry and the Down Syndrome Connect Registry.

TABLE 1. Group vocal productivity scores.*Participant Demographics*

Characteristic	Down Syndrome		Angelman Syndrome		Low Risk		Full Sample	
	<i>n</i>	%	<i>n</i>	%	<i>n</i>	%	<i>n</i>	%
Gender								
Female	10	50	10	55.6	16	47.1	36	50
Male	10	50	8	44.4	18	52.9	36	50
Race								
Asian	1	5	0	0	0	0	1	1.4
Multiracial	2	10	2	11.1	3	8.8	7	9.7
White	17	85	16	88.9	31	91.2	64	88.9
Ethnicity								
Hispanic	4	20	1	5.6	2	5.9	7	9.7
Non-Hispanic	16	80	17	94.4	32	94.1	65	90.3
Mother's education level								
Less than high school	0	0	0	0	0	0	0	0
High school diploma	0	0	2	11.1	0	0	2	2.8
Some college	2	10	5	27.8	5	14.7	12	16.7
2-year degree	1	5	0	0	3	8.8	4	5.6
4-year degree	8	40	5	27.8	10	29.4	23	31.9
Professional degree	6	30	6	33.3	14	41.2	26	36.1
Doctoral degree	2	10	0	0	2	5.9	4	5.6
Prefer not to answer	1	5	0	0	0	0	0	1.4
Father's education level								
Less than high school	0	0	1	5.6	0	0	1	1.4
High school diploma	0	0	4	22.2	1	2.9	5	6.9
Some college	2	10	2	11.1	8	23.5	12	16.7
2-year degree	0	0	1	5.6	4	11.8	5	6.9
4-year degree	10	50	5	27.8	12	35.3	27	37.5
Professional degree	3	15	5	27.8	8	23.5	16	22.2
Doctoral degree	3	15	0	0	0	0	3	4.2
Prefer not to answer	2	10	0	0	1	2.9	3	4.2
	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>
Time between recording and PSI (months)	13.0	5.2	31.4	10.2	16.3	6.2	19.2	10.1

Measures*Parenting Stress Index, Fourth Edition Short Form*

We used the Parenting Stress Index (PSI) to measure the stress a parent experiences in relation to parenting and their relationship with their child (Abidin, 2012). The

PSI is collected using a 36-item inventory filled out by a parent. It is divided into three domains: parental distress, parent-child dysfunctional interaction, and difficult child. Parental distress measures the extent to which parents feel competent, restricted, conflicted, supported, and/or depressed in their role as a parent. Parent-child dysfunctional interaction looks at the extent to which

parents feel satisfied with their child and their interactions with the child. Finally, the difficult child domain captures whether a parent perceives her or his child to be easy or difficult to care for. These three domains are combined to create a total stress scale. We used the total stress scale percentile in our analysis.

Language Environment Analysis

We gathered daylong audio recordings in the child's natural environmental setting using a Language Environment Analysis (LENA) recording device (Xu et al., 2008). The LENA device records up to 16 hours and captures both the child's vocalizations as well as sounds occurring in the child's immediate environment, such as adult speech or electronic noise. Participants wear the LENA device on their chest, secured in a pocket on the front of a child-sized vest (Figure 1). These audio recordings are then processed using an online program that produced several automated metrics characterizing

FIGURE 1. Child 12–18 months old wearing vest with LENA recording device.

various aspects of the child's speech and language development. For the present study, we used the VP percentile score. VP is a measure of vocal maturity that examines canonical syllables (syllables containing both a consonant and a vowel) used during back-and-forth vocal interactions (also known as a conversational turn) between the child and an adult.

Procedure

All study procedures were approved by the Institutional Review Board at Purdue University (IRB 1811021381, "Neurodevelopmental Natural History Study"). Data were obtained from several ongoing studies in the Neurodevelopmental Family Lab at Purdue University. For each study, participants' mothers completed a set of online forms that included demographic questions, questionnaires about various domains of their child's development, and self-report measures of their own experiences as a parent, including the PSI. Families were provided with all materials to complete the daylong audio recording with the LENA device, including the recorder, the vest, charging materials, and instructions to reference on the day of the recording.

Hypotheses and Analytic Plan

We conducted a regression analysis using IBM SPSS to test if syndrome group (i.e., AS or DS), PSI total stress percentile, and the interactions between PSI total stress percentile and each syndrome group were associated with participants' VP percentile scores. We included age in the regression model to control for the wide variability in age across participants and used LR as the reference group. We predicted that the AS and DS groups would both have significantly lower VP scores than the LR group, as indicated by significant main effects. We also predicted that the magnitude of the association between VP and PSI would be stronger in the AS and DS groups than in the LR group.

RESULTS

Descriptive statistics of PSI and VP scores are reported in Table 2. When age and PSI scores are held constant,

TABLE 2. Associations of vocal productivity with parenting stress.

Descriptive Statistics

	n	Min.	Max.	M	SD
Angelman syndrome					
VP percentile	18	1	47	7.06	11.50
PSI percentile	18	1	81	45.61	26.04
Down syndrome					
VP percentile	20	1	83	27.50	23.83
PSI percentile	20	4	76	27.90	21.97
Low risk					
VP percentile	34	4	96	46.65	28.07
PSI percentile	34	6	95	39.24	24.03

TABLE 3.

Regression Analysis Results

Variable	B	SE	β	t	p
(Constant)	61.394	10.214		6.011	0.000
PSI	−0.241	0.174	−0.207	−1.385	0.171
AS	−40.801	16.032	−0.623	−2.545	0.013*
DS	−27.582	11.976	−0.436	−2.303	0.024*
PSI*AS	0.168	0.285	0.139	0.589	0.558
PSI*DS	0.166	0.305	0.098	0.543	0.589
Age	−0.325	0.408	−0.115	−0.795	0.430

Note. * $p < .05$

the AS group had significantly lower VP scores ($M_{AS} = 7.06$, $SD_{AS} = 11.50$) than the LR group ($M_{LR} = 46.65$, $SD_{LR} = 28.07$; Table 2), as indicated by a significant main effect ($p = .013$) (Table 3). The DS group also had significantly lower VP scores than the LR group ($M_{DS} = 27.50$, $SD_{DS} = 23.83$, $p = .024$).

We observed a negative association between PSI scores and VP scores in the LR group, though it is not statistically significant ($\beta = -.207$, $p = .171$) (Figure 2). In the AS group, the magnitude of the association between PSI scores and VP scores did not statistically differ from that of the LR group ($\beta = .139$, $p = .558$), nor did the magnitude of the association between PSI scores and VP scores in the DS group ($\beta = .098$, $p = .589$).

Exploratory Post Hoc Analyses

Given the large sample sizes needed to detect significant interaction effects in multiple regression models (Shieh, 2009) and the nonsignificant effect of age in our primary models, we conducted supplemental Spearman correlations to test whether PSI related to VP within each group: LR, AS, and DS. These analyses did not compare the relative magnitude of effects but did probe whether each group demonstrated a significant association. Results indicated small (Cohen, 1969) nonsignificant associations between parenting stress and VP in AS ($\rho = -.182$, $p = .470$), DS ($\rho = -.155$, $p = .515$), and LR ($\rho = -.254$, $p = .148$).

DISCUSSION

While many parents of children with rare disorders are resilient, many have also historically reported high levels of caregiving-related stress (Blank et al., 2020; Almogbel et al., 2017). Although prior research suggests that caregiving stress relates to poorer developmental outcomes in children, there is some evidence that these associations vary across high- and low-risk groups. Although families affected by rare syndromes are at risk for both high parenting stress and child language delays, no studies to date have explicitly tested how these two features are related. Here, we conducted an initial study to examine whether VP—a measure of vocal maturity—is related to higher parenting stress in families affected by two neurogenetic syndromes. Our results suggest that children with AS and DS demonstrate lower VP than children at low risk for developmental delays. Similarly, there was a negative magnitude of association between parenting stress and VP for both the AS and DS groups, though these associations were not statistically significant.

Group Vocal Productivity

As anticipated, the AS and DS groups had significantly lower VP scores than the LR group when PSI scores and age were held constant. This means that the AS and DS group incorporated significantly fewer canonical

FIGURE 2. Associations of vocal productivity and Parenting Stress Index percentile scores by syndrome group.

syllables into conversational turns between the child and an adult. Canonical babbling is a precursor to speech development that emerges in typically developing children around 6 months in age; thus, a later onset to babbling is associated with a delay in meaningful speech (Nyman and Lohmander, 2018). These findings emphasize that language development in children with DS and AS is delayed when compared to typically developing children and warrants further study of how to best treat these delays to promote optimal outcomes.

Our study used similar means of determining and analyzing VP as previous literature has done while filling gaps relating VP to parenting stress. For example, Grieco et al. (2018) analyzed vocalizations of children aged 34 to 126 months with AS and determined that the participants' vocalization ranges—including sounds of crying, discomfort, phonation, laughter, singular and glided

vowels, squeals, and babbling—were similar to typically developing children aged 0 to 6 months. Grieco et al. found that children with AS used an average of 5.67 canonical syllables, whereas the control participant (aged 16 months) used 50 canonical syllables during the play session. The increased use of canonical syllables by Grieco's control participant is consistent with findings in the LR group in our study. Another study by Nyman and Lohmander (2018) found that children with DS aged 12 to 22 months displayed lower rates of canonical babbling when compared to typically developing children. Because the participants in Nyman and Lohmander's study were older than those in previous studies, this emphasizes the progressive delay in speech and language development in these children. This highlights the necessity for early intervention techniques for children with DS and other neurodevelopmental disorders that affect language development. Sokol and Fey (2013)

completed a longitudinal study comparing the development of speech of children with DS to children with speech delays without DS. The participants had two assessments at 25 months and 43 months of age. Sokol and Fey discovered that children with DS had similar and sometimes greater consonant production than the children without DS during the first assessment but had less consonant production than the non-DS group at the second assessment. The study emphasized a pattern in slow development for children with DS between the first and second assessments distinguishable from the children without DS. Although the participants were not compared to a similar LR group as in our study, there were noted differences between consonant production and canonical syllable production in children with DS and the comparison groups. Overall, our findings are consistent with previous literature demonstrating that children with AS and DS have delays when compared to typically developing children, and further research should investigate treating these delays.

Associations between Vocal Productivity and Parenting Stress

Previous studies have shown that parenting stress is elevated in some genetic syndromes, but our study expands this by inquiring whether it relates to language development in groups that are understudied. We observed a small, nonsignificant association between parenting stress and VP in our LR group, and contrary to our hypotheses, we did not observe greater magnitude of associations in the AS and DS groups. The post hoc analyses also did not show a significant association between VP and PSI between the AS, DS, and LR groups, which implies that the relationship between VP and PSI is in general relatively small, particularly within the syndromic groups.

There are a few reasons why our findings may have differed from prior studies (e.g., Blank et al., 2020; Almogbel et al., 2017). First, it could be that at the ages included in our sample, there is not yet enough variability to detect these associations; indeed, children with AS had very low levels of VP, which constrains the ability to test for associations between VP and outcomes. It could also be the case that these associations emerge prospectively.

Thus, an important next step for our study would include looking longitudinally into how parenting stress impacts language development in children with neurogenetic syndromes as well as how parenting stress in early development can impact language development later in life. Our AS participants were older on average than the DS group and the LR group but were in similar age ranges to other studied AS groups. We also measured a different primary outcome—VP—than prior studies. For example, Almogbel et al. (2017) found associations between parenting stress and general developmental skills, and Blank et al. (2020) detected associations with language comprehension. In contrast, we examined a very specific metric of language development, which may differentially relate to stress. Continuing to probe the contexts in which parenting stress relates to language development could help clinicians and parents create more effective treatment plans for high-risk children and their families.

Limitations

Due to the smaller number of subjects in our syndrome groups in this study, the results should be considered preliminary and subjected to further testing in larger well-powered studies. The association between parenting stress and VP was negative in our LR group, and it is possible that with a larger sample size a statistically significant association could be detected. A larger sample size might also have more power to detect differences in associations between the three groups. We note that given the lack of information on syndromic groups such as AS in particular, these results can still inform the literature by documenting lower VP in AS and DS children. There were also several socioeconomic factors related to our sample—including race, parents' education, and parents' income—that could have impacted findings. For example, participants were recruited through social media postings and research registries, which could limit the diversity of participants due to limited access to those platforms. There is a limitation due to large differences in mean age ranges in the AS group compared to the DS and LR groups. Even though the study controlled for age, the association of VP and PSI might look different in younger children than in older children due to developmental differences and maturity. Another limitation is that there were no

experimental manipulations performed in the study, so there was no information gained about whether parenting stress directly causes changes in language development or vice versa.

Future Research

There are a number of potential expansions of this work. First, a larger sample size of children with neurogenetic disorders could potentially provide more variability and power to detect small associations between VP and parental stress. Second, future studies could expand these findings by probing the association between parents' mental health and the language levels of their children with a larger sample size. Finally, it will be important to test whether these associations differ across more diverse samples that are more representative of the population; there was little diversity in our sample size, with 88% of the participants self-identifying as white. In addition, longitudinal studies could inform how language development and parental stress intersect across development and how these associations vary across individuals. Such studies would inform how to best target interventions to individuals most at risk and deploy these interventions as early as possible.

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