Association of Sibling Presence with Language Development from Infancy to Early School Age Among Children with Developmental Difficulties: A Longitudinal Study

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Research

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Abstract

Background

Siblings are crucial familial-ecological factors in children's language development. However, it is unclear whether sibling presence is associated with language development among young children with developmental difficulties. The aim of this study was to assess the association between sibling presence and changes in language trajectories of children with developmental delay before early school age.

Methods

We performed a retrospective longitudinal cohort study from December 2008 through February 2016. The medical records of the participants were collected from an official institution designated by Taiwan's Ministry of Health and Welfare for assessing and identifying young children with developmental difficulties. A total of 174 participants who had developmental difficulties and at least three-waves of evaluations were included in the analysis. Participants' age ranged from 10 to 90 months. The primary outcomes were receptive and expressive language delays evaluated by board-certified speech-language pathologists.

Results

Of the 174 participants (131 boys; at the first evaluation: mean [standard deviation (SD)] age, 31.74 [10.15] months) enrolled, 64.94% (*n*=113) had siblings and 35.06% (*n*=61) did not. At the age of approximately 10 months, the probability of receptive and expressive language delays was lower in participants with siblings than in those without (adjusted odds ratios, 0.19, 0.18; 95% confidence interval [CI], 0.06-0.64, 0.04-0.80; *P*=0.006, 0.024, respectively). However, at 10–90 months old, this probability of language delay became gradually higher in participants with siblings than in those without, exceeding that of participants without siblings (adjusted odds ratios, 1.04, 1.04; 95% CI, 1.01-1.07, 1.01-1.07; *P*=0.014, 0.020, respectively)

Conclusions

Having siblings does not necessarily have a positive association on the language development of children with developmental difficulties. Clinicians should consider the association of sibling presence with language development for these children in a broader familial-ecological context.

Background

Children with developmental difficulties (DDs) are those children with signs and symptoms of arrest, alteration, delay, and disability in any of the following developmental areas in early childhood spanning from infancy to 8 years of age: cognitive, language, emotional, behavioral, social, and fine and gross motor.[1,2] In pediatric populations, DD prevalence ranges from 5–15%.[2,3] In Taiwan, a recent study reported that DD prevalence in children aged < 6 years was 11.36%,[4] and the incidence of DD in this age

group showed an increasing trend from 2003 to 2016, ranging from 7.0 to 16.3 per 1000 person-years.[5] Furthermore, in children with DD, speech and language delays are the most common disabilities,[2] with a prevalence of as high as 42.19% in Taiwan.[4] Understanding the risk factors of the language developmental changes in children with DD in early childhood is important.

Siblings are children's most constant social companions who provide a proximal and long-lasting context for development, and siblings can advance one another's development in the context of their relationship.[6] For children with typical language development (TLD), their siblings also motivate them to practice their language skills and teach them various language domains, thereby facilitating language development in a familial-ecological context.[7,8] However, sibling presence may not be advantageous for children with DD. As an investment-type model would suggest, parents allocate more resources to the child with better developmental skills, instead of allocating resources aimed at reducing the developmental differences among siblings.[9,10] Additionally, studies on early intervention have reported that only trained siblings may be among the potential and powerful peer interventionists at home and in other community settings who can promote language acquisition in children with autism.[11,12] Thus, in households with sibling pairs, children with DD may receive fewer resources from their parents due to the competition from the other sibling. The association of sibling presence with language development among young children with DD may, therefore, not be positive.

To the best of our knowledge, no study has examined whether having siblings is associated with language development in children with DD before school age. In this study, we hypothesized that if siblings—as socialization agents—facilitate language development in children with DD, the probability of language delay should be lower in children with DD with siblings than in those without. However, if siblings were not advantageous for children with DD in a household, the probability of language delay would be higher in children with DD who have siblings. Additionally, the progression of language development in children is coupled with a relatively fixed time course and stages,[13] which are related to the facets of biological characteristics, including a child's sex,[14] speech development (e.g., oral function and swallowing/feeding)[15], history of medical diagnosis[16], and language exposure in ecological contexts, such as the home learning environment predicted by the maternal educational level.[17,18] To avoid confounders, we controlled for children's sex, oral function and swallowing problems, and early medical diagnosis as well as maternal educational level.

Methods

Data Source and Participants

Participants of this retrospective longitudinal cohort study were from an early developmental evaluation center in a medical center between December 2008 and February 2016 in New Taipei City, Taiwan. This early developmental evaluation center is among the official institutions designated by Taiwan's Ministry of Health and Welfare for assessing and identifying children with DD before the age of 7. According to

the definition of Taiwan's Ministry of Health and Welfare[19], participants with DD were those children with 1 SD (percentile 15) below the normative population mean in at least one of the following developmental domains: gross or fine motor, speech/language, cognition, social/personal, or activities of daily living. All study procedures were approved by the Research Ethics Review Committee (IRB ID: 105090-F) in Taiwan.

Participants were recruited if they had (1) at least three waves of language evaluations of at least 1 year apart, and (2) completed data on the measures listed in the next subsection. 174 participants with DD were included in the final analysis.

Measures

Data on siblings, language delay, oral-motor function and swallowing/feeding problems, medical diagnosis, sex, and maternal educational level of each participant were collected. Data on language delay and oral-motor function and swallowing/feeding problems were gathered at each evaluation, and the others were obtained from the first evaluation.

Siblings

The information on sibling presence was obtained from parent reports and categorized binarily as presence of siblings (coded as 1) or absence of siblings (coded as 0).

Language Delay

The receptive and expressive language skills of each participant were evaluated using standardized tests by board-certified speech-language pathologists. For the standardized tests, the Mandarin-Chinese version of the MacArthur-Bates Communicative Developmental Inventories Toddler Form[20] was used in children aged < 3 years, and the Mandarin-Chinese version of Preschoolversion[21] and School Version[22] of Child Language Disorder Scale-Revised were used in children aged between 3 and 5 years and those aged > 6 years, respectively. Furthermore, receptive or expressive language abilities were dichotomous and coded as normal (coded as 0; scores on the standardized tests were > 15th percentile) or delayed (coded as 1; scores on the standardized tests were ≤ 1 SD of the normative population mean, i.e., at the ≤ 15th percentile). Receptive language delay refers to the delay in the developmental milestone to understand words, instructions, and stories, whereas expressive language delay refers to the delay in the developmental milestone related to pre-linguistics and word and sentence production.

Confounders

Oral-Motor Function and Swallowing/Feeding Problems

Speech-language pathologists also assessed participants' oral-motor function and swallowing/feeding problems. These skills were coded dichotomously as normal (coded as 0) or delayed (coded as 1).

Medical Diagnosis

The diagnosis of neurological (e.g., cerebral palsy or epilepsy) and neurodevelopmental (e.g., autism and attention deficit hyperactivity disorder) disorders was made by board-certified child neurologists and child psychiatrists with ≥ 5 years of experience. According to their medical diagnosis and language skills, the participants with DD were further categorized into children (1) with language delay alone, (2) with normal language development and no neurological or neurodevelopmental disorders, and (3) with language delay and neurological or neurodevelopmental disorder.

Sociodemographic Characteristics

(1) Participants' sex was categorized as male (coded as 1) and female (coded as 0). (2) Participants' maternal educational level was categorized as junior high school (coded as 0; reference group), senior high school, and college and above. This sociodemographic information was obtained from parent reports.

Data Analysis

Multilevel logistic regression models (MLRMs) are used to estimate models that include both estimates of intra-individual change over time and inter-individual variability in the trajectories.[23] In a two-level MLRM, the Level 1 model describes within-person repeated measures (i.e., linear time and time-varying variables) and the Level 2 model describes between-person variability in the trajectories, as defined by Level 1 model parameters, i.e., time-varying variables. Multilevel regression models providing a flexible and powerful method for modelling developmental changes in longitudinal data were used with normally distributed data (e.g., IQ, scores). For assessing developmental changes in longitudinal dichotomous data, including presence or absence of language delay, multilevel regression models can be extended using a logit function (i.e., MLRMs).

In the current study, participants' oral-motor function and swallowing/feeding problems, sex, maternal educational level, and medical diagnosis were controlled. We ran MLRMs separately for receptive (Models 1 and 2) and expressive (Models 3 and 4) language development. Moreover, each MLRM comprised the following two hierarchical levels: within-person repeated measures (Level 1) and inter-individual variance (Level 2). Linear time was included in the Level 1 model as predictor; oral-motor function and swallowing/feeding problems were included in the longitudinal structure of the data as time-varying predictors. The Level 2 model first included only sibling presence on intercept and linear time to examine whether the interaction between sibling presence and linear change rate of likelihood of language delay differed across groups (Models 1 and 3). Next, the final model (Models 2 and 4) included adjustments for confounders known to impact a child's language development, including sex, maternal educational level, and medical diagnosis.

The software package HLM (version 7.03)[24] was used to fit MLRMs. The models were fitted using restricted maximum likelihood estimations. For MLRMs, a population-average model with robust standard errors is reported. Analyses were conducted from October 2018 to April 2020.

Results

Sample Description

The participants' mean age at the first and final evaluations were 31.74 (SD_{age}=10.15, range = 10–58) and 63.90 (SD_{age}=10.32, range = 37–90) months, respectively. In the total sample, participant age over time—from infancy to early school age—spanned from 10 to 90 months, fitting a growth curve covering 7 years. Altogether, 113 (64.94%) participants had siblings. The mean age of participants with and without siblings at the first and final evaluations showed non-significant difference ($P > .05$) (Table 1). Of the 174 participants, 6, 45, and 123 had five, four, and three waves of evaluations, respectively. The sample size in the current study is significantly large enough for MLRM analysis.[24] Moreover, the waves of evaluations during the study period did not differ between participants with and without siblings ($P > .05$).

Table 1
Characteristics of the study sample (*N* = 174)

	Presence of siblings		**Absence of siblings**		***P***
Characteristics	**(*n* = 113)**		**(*n* = 61)**		
	M (*SD*)	*Range*	*M* (*SD*)	*Range*	
Age (months)					
First evaluation	30.93 (10.26)	10-58	33.23 (9.84)	37-90	.154[a]
Final evaluation	63.56 (10.59)	13-58	64.52 (9.86)	37-83	.557[a]
	n	%	*n*	%	
Waves of evaluations					.559[b]
Three	77	68.14	46	75.41	
Four	32	28.32	13	21.31	
Five	4	3.54	2	3.28	
First evaluation					
Sex					.692[c]
Male	84	74.34	47	77.05	
Female	29	25.66	14	22.95	
Maternal educational level					.317[c]
Junior high school	18	15.93	5	8.20	
Senior high school	63	55.75	35	57.38	
College and above	32	28.32	21	34.43	
Medical diagnosis					.645[c]
NLD	30	26.55	15	24.59	
LD	65	57.52	39	63.93	
LDNN	18	15.93	7	11.48	

NLD: child with normal language development and without any neurological or neurodevelopmental disorders with other. LD: child with language delay alone. LDNN: child with language delay with neurological disorders or neurodevelopmental disorders. [a]t-test. [b]Fisher's exact test. [c]Chi-square test.

In the first evaluation, the maternal educational level for 30.46% (*n* = 53), 56.32% (*n* = 98), and 13.22% (*n* = 23) of the participants were college and above, senior high school, and junior high school, respectively. No significant differences in maternal educational level were observed between participants with and without siblings (all *P* > .05).

Additionally, 59.77% (*n* = 104), 14.37% (*n* = 25), and 25.86% (*n* = 45) of the participants were categorized as having language delay alone, language delay with neurological disorders or neurodevelopmental disorders, and normal language development and without any neurological or neurodevelopmental disorders, respectively. No significant differences in the proportions of patients with these diagnoses were observed between participants with and without siblings (*P* > .05).

Trajectories of Language Development and Sibling Presence

The intraclass correlation coefficients of receptive and expressive language trajectories were 0.22 and 0.19, respectively. For the trajectory of receptive language development (Table 2), in Model 1, the odds of receptive language delay at 10 months of age were lower in participants with siblings than in participants without siblings (OR = 0.16; *P* < .001). However, from 10 to 90 months, the odds of receptive language delay became gradually higher in participants with siblings than in participants without siblings (OR = 1.04; *P* = 0.001). The trend remained statistically significant after adjusting for confounders in Model 2 (aOR = 1.04; *P* = 0.014). Figure 1 displays the adjusted trend of receptive language development between participants with and without siblings. The probability of receptive language delay for participants with siblings increased gradually from the age of 10 to 90 months and exceeded that of participants without siblings after 50 months of age. By contrast, the probability of receptive language delay in participants without siblings remained stable from the age of 10 to 90 months.

Table 2
Summary of MLRM for the trajectory of receptive language delay (*N* = 174)

	Model 1				Model 2			
	Coff	ORs	95% CI	*P*	Coff	ORs	95% CI	*P*
Log odds at CA = 10 months (intercept)	3.66	38.84	15.59-96.78	< .001	4.31	74.33	5.55-995.88	.001
Male (Ref = female)					1.07	2.93	0.96-8.89	.058
Presence of siblings (Ref = absence of siblings)	-1.85	0.16	0.05-0.46	< .001	-1.65	0.19	0.06-0.64	.008
Maternal educational level (Ref = junior high school)								
Senior high school					-0.44	0.64	0.07-6.35	.703
College and above					-0.64	0.53	0.06-5.04	.575
Medical diagnosis (Ref = LD)								
NLD					-1.98	0.14	0.04-0.46	.001
LDNN					1.30	3.66	0.67-20.09	.134
Linear time (monthly change)	-0.07	0.93	0.91-0.95	< .001	-0.05	0.95	0.90-1.01	.095
Male (Ref = female)					-0.04	0.96	0.94-0.99	.010
Presence of siblings (Ref = absence of siblings)	0.04	1.04	1.02-1.07	.001	0.04	1.04	1.01-1.07	.014
Maternal educational level (Ref = junior high school)								
Senior high school					-0.00	1.00	0.95-1.05	.909
College and above					-0.02	0.98	0.93-1.03	.367
Medical diagnosis (Ref = LD)								

CA: chronological age; NLD: child with normal language development and without any neurological or neurodevelopmental disorders; LD: child with language delay alone; LDNN: child with language delay with neurological disorders or neurodevelopmental disorders; Ref: Reference group; Coff: coefficient.

	Model 1				Model 2			
NLD					0.02	1.02	0.99-1.06	.175
LDNN					-0.01	0.99	0.96-1.03	.595
Time-varying confounders								
Oral-motor function	0.17	1.18	0.79-1.76	.407	0.29	1.34	0.85-2.10	.202
Swallowing/feeding problem	0.52	1.68	0.84-3.39	.145	0.11	1.11	0.47-2.66	.808

CA: chronological age; NLD: child with normal language development and without any neurological or neurodevelopmental disorders; LD: child with language delay alone; LDNN: child with language delay with neurological disorders or neurodevelopmental disorders; Ref: Reference group; Coff: coefficient.

For the trajectory of expressive language development, in Model 3 (Table 3), the odds of receptive language delay at 10 months of age were lower in participants with siblings than in participants without siblings (OR = 0.14; *P* = 0.002). However, from the age of 10 to 90 months, the odds of expressive language delay were gradually higher in participants with siblings than in participants without siblings (aOR = 1.05; *P* = 0.001). This trend in the change rate (aOR = 1.04; *P* = 0.020) also remained statistically significant after adjusting for confounders included in Model 4. As shown in Fig. 2, the probability of expressive language delay was lower in the participants with siblings than in participants without siblings. Furthermore, after 50 months of age, the probability of expressive language delay was higher in the participants with siblings than in the participants without siblings.

Table 3
Summary of MLRM for the trajectory of expressive language delay (*N* = 174)

	Model 3				Model 4			
	Coff	ORs	95% CI	*P*	Coff	ORs	95% CI	*P*
Log odds at CA = 10 months (intercept)	4.50	89.92	32.89-245.79	< .001	5.22	185.45	6.52-5271.76	.002
Male (Ref = female)					0.02	1.02	0.27-3.83	.973
Presence of siblings (Ref = absence of siblings)	-1.95	0.14	0.04-0.47	.002	-1.74	0.18	0.04-0.80	.024
Maternal educational level (Ref = junior high school)								
Senior high school					0.39	1.48	0.13-17.01	.753
College and above					0.06	1.06	0.08-13.70	.962
Medical diagnosis (Ref = LD)								
NLD					-1.96	0.14	0.04-0.48	.002
LDNN					2.42	11.28	1.77-71.71	.011
Linear time (monthly change)	-0.09	0.92	0.90-0.94	< .001	-0.08	0.92	0.86-0.99	.018
Male (Ref = female)					-0.00	1.00	0.96-1.03	.824
Presence of siblings (Ref = absence of siblings)	0.05	1.05	1.02-1.08	.001	0.04	1.04	1.01-1.07	.020
Maternal educational level (Ref = junior high school)								
Senior high school					-0.01	0.99	0.94-1.04	.563
College and above					-0.03	0.97	0.93-1.03	.302

CA: chronological age, NLD: child with normal language development and without any neurological or neurodevelopmental disorders, LD: child with language delay alone, LDNN: child with language delay with neurological disorders or neurodevelopmental disorders, Ref: Reference group, Coff: Coefficient.

	Model 3				Model 4			
Medical diagnosis (Ref = LD)								
NLD					0.02	1.02	0.98-1.05	.322
LDNN					-0.03	0.97	0.94-1.01	.163
Time-varying confounders								
Oral-motor function	0.17	1.19	0.80-1.75	.391	0.35	1.42	0.93-2.19	.107
Swallowing/feeding problem	0.47	1.60	0.78-3.30	.197	0.03	1.03	0.44-2.38	.947

CA: chronological age, NLD: child with normal language development and without any neurological or neurodevelopmental disorders, LD: child with language delay alone, LDNN: child with language delay with neurological disorders or neurodevelopmental disorders, Ref: Reference group, Coff: Coefficient.

Discussion

To the best of our knowledge, this is the first study to examine whether having siblings is associated with the language development in young children with DD in the long term. The longitudinal design provides a thorough investigation of the association between the familial-ecological factors, presence of siblings, and language developmental trajectories from a young age and across a critical period of development. Another strength of our study was the high validity of the medical diagnosis and language evaluation, which were all made by board-certified clinical child experts with the standard clinical diagnostic procedure in the developmental evaluation center designated by Taiwan's Ministry of Health and Welfare for assessing and identifying young children with DD, as compared to other community studies collecting data from parent-report questionnaire only.[1,2,4]

Are siblings associated with language development among young children with DD? We found that the probability of language delay in young children with DD who had siblings became gradually higher, exceeding that of children who did not have siblings. Furthermore, the magnitude of the association remained significant even after adjusting for well-measured potential confounders. Our study supported that having siblings was associated with an increasing probability of language delay before school age. This finding obviously contradicted the findings of previous studies showing that siblings facilitate language development in children with TLD.[7,8] The results can be explained by two reasons.

One reason is that children with DD may not be able to benefit from natural social learning environments provided by their siblings. To become an effective language facilitator to children with DD, siblings may need to learn and receive training. Efficacy studies on sibling-mediated interventions that include siblings

as active treatment implementers, rather than as passive bystanders, have reported positive outcomes.[25,26] Training programs might be a prerequisite for siblings to be successful in helping children with DD.

The other reason is the theory of intra-household resource allocation. Some studies reported that parents may invest more on children with DD to equalize the ability gap among their children ("compensating strategies").[27,28] However, in a household with finite resources, parents may invest more on the higher-achieving child, thus increasing the ability gap among children ("reinforcing strategies"),[9,10] and these parents would not adopt compensating strategies to equalize the ability gap among their children. Our finding supports that parents of children with DD might have an infinitesimally small chance to adopt compensating strategies to reduce the ability gap between children with DD and their siblings. In other words, siblings are likely to compete for the parents' attention and resources; therefore, children with DD who had siblings would receive less resources than those without siblings, placing those with siblings at a relative disadvantage.

Our study attempted to consider having siblings only at the first evaluation because we wanted to identify the red flag for language development for pediatricians and other clinical child experts at the first encounter with children with DD. However, children with DD might have upcoming siblings after the first evaluation. The information on sibling presence, regardless which time points, will be collected and considered again at each evaluation, with the estimated probabilities of language delay re-calculated and modified accordingly. Hence, our models can still explain the trajectories of language development in children with DD even under the condition of time-varying changes in sibling presence.

Microlevel sibling experiences and their meaning in children's development are embedded in macrocontexts[6]; that is, having a sibling has an impact on the ability development of children with TLD, which is intricately linked to family context, such as birth order of the children, sex composition in siblingships, and family size; however, this was not reported in our study. Based on the findings that sibling presence was associated with language development among young children with DD, future studies to determine the association between the siblingship characteristics in macrocontexts and language development in children with DD are warranted. Next, to consider alternative associations for language development among children with DD, most notably, family history of speech and/or language disorders, impact of intervention, and language development cascaded by other domains of functioning should be integrated to be examined. By focusing on longitudinal transactional models[29] to measure brain maturation in the context of endogenous characteristics (i.e., language competence associated with hereditary or cascaded by other domains of functioning[30]) and exogenous characteristics (i.e., environmental exposures), we can understand the mechanisms by which these constructs act together in shaping brain plasticity in children with DD from early ages. Additionally, only the impairment level (i.e., language delay) was examined in this study. To further comprehend the role of siblings in the trajectories of language development in children with DD, these trajectories could be traced using the International Classification of Functioning, Disability and Health for Children and Youth model as a framework,

specifically by using indexes, such as activity, participation, quality of life, and psychosocial health as outcome variables.

Limitation

Although this study provides the first longitudinal examination of siblings and trajectories of language development in young children with DD, the results need to be considered in light of selection bias. First, the sex ratio was uneven. It was approximately 3:1 (male:female), which was similar to that of a previous report in Taiwan[31]; hence, it might only represent the natural sexual distribution of DD, rather than a selection bias. Furthermore, we controlled the sex effect in our analytic models (Models 2 and 4) on the association between siblings and language development trajectories in young children with DD. Second, we were limited by the inclusion of participants from a medical center in a metropolitan region with high recruitment rates. This inclusion potentially limits the generalizability of our findings to children with DD in a rural area.[5]

Conclusion

Having siblings does not necessarily have a positive association on the language development of young children with DD. In children with DD who have siblings, the probability of language delay would become gradually higher from infancy to early school age. This is an important area of study that has implications for early screening, etiology, and early intervention. Emphasizing the need for a family-centered developmental evaluation in the language evaluation of children with DD, pediatricians and other clinical child experts not only should focus on assessing language skills per se, but also should consider the association between sibling presence and language development, to optimize language development outcomes in these children. Increasing sibling participation as language facilitators is imperative to strengthen language development of young children with DD in a broader familial-ecological context. Furthermore, clinicians should assist parents maximize resource availability for these children and improve their early learning environments.

Abbreviations

DD, developmental delay

TLD, typical language development

CI, confidence interval

SD, standard deviation

MLRM, multilevel logistic regression models

NLD, child with normal language development and without any neurological or neurodevelopmental disorders

LD, child with language delay alone

LDNN, child with language delay with neurological disorders or neurodevelopmental disorders

Declarations

Ethics approval and consent to participate:

All study procedures were approved by the Research Ethics Review Committee (IRB ID: 105090-F) in Taiwan.

Consent for publication:

Not applicable

Availability of data and materials:

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

Competing interests:

The authors declare that they have no competing interests.

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Authors' contributions:

HHL conceptualized and designed the study, carried out the initial analyses and interpretation of data, obtained funding, and drafted the initial manuscript. WCC carried out the initial analyses and interpretation of data, and drafted the initial manuscript. YJL supervised the data collection, obtained funding, and drafted the initial manuscript. JSL conceptualized and designed the study, supervised data collection, obtained funding, and drafted the initial manuscript. All authors approved the final manuscript as submitted and agreed to be accountable for all aspects of the work.

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Figures

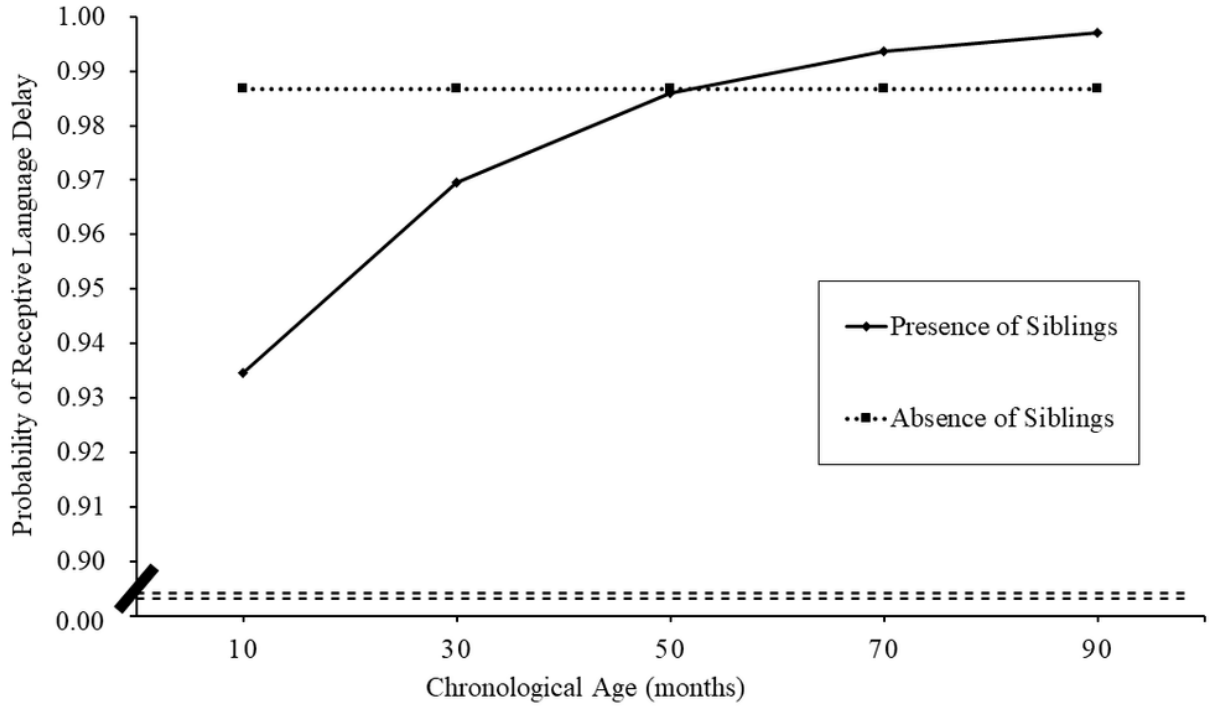


Figure 1

Predicted receptive language delay trajectories for the presence and absence of siblings after controlling for confounders.

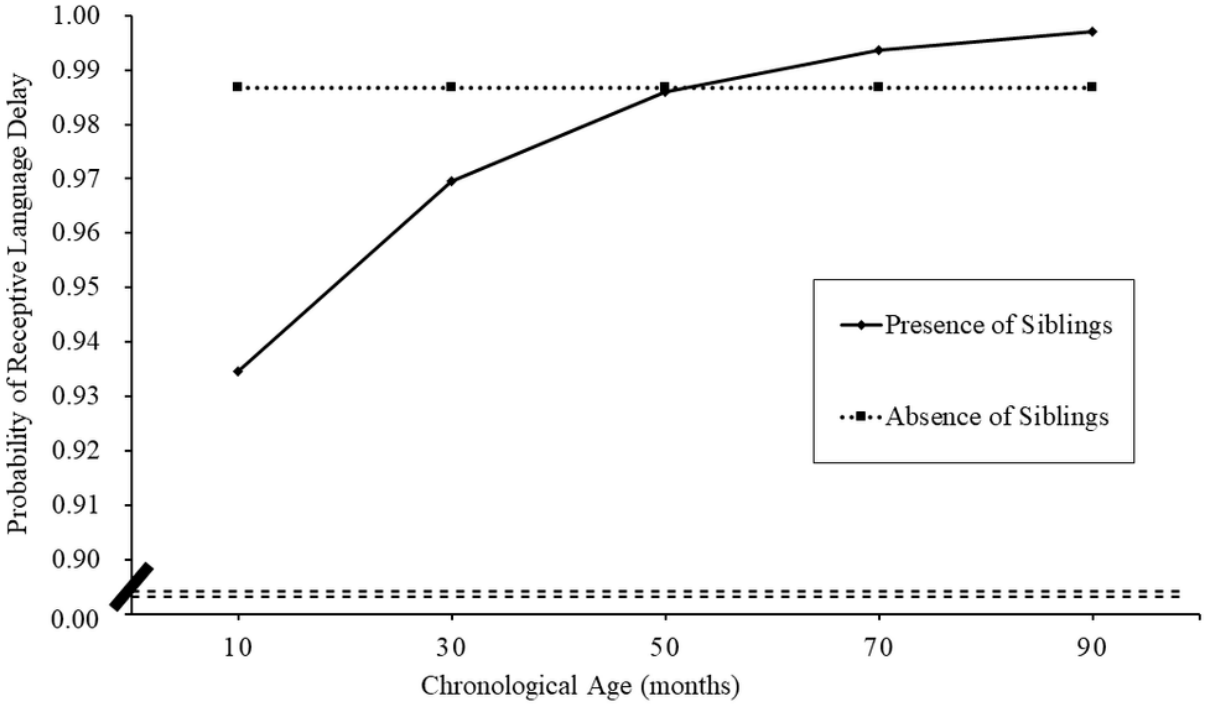
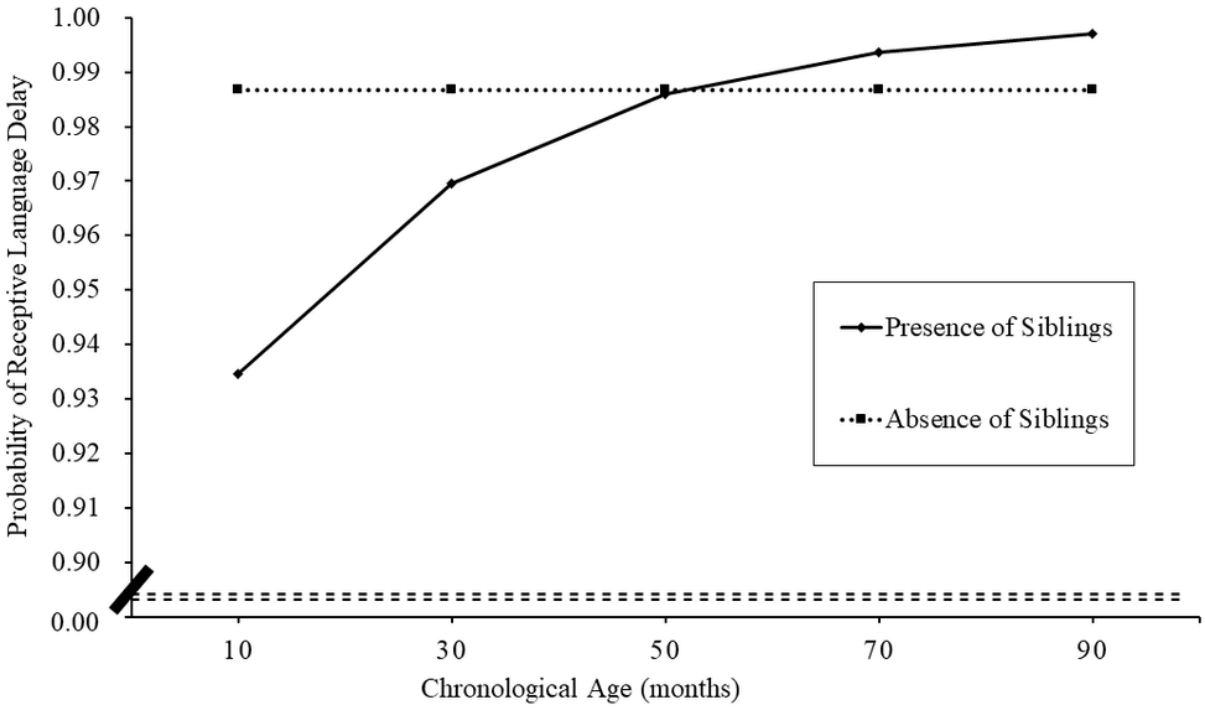


Figure 1

Predicted receptive language delay trajectories for the presence and absence of siblings after controlling for confounders.



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Predicted receptive language delay trajectories for the presence and absence of siblings after controlling for confounders.

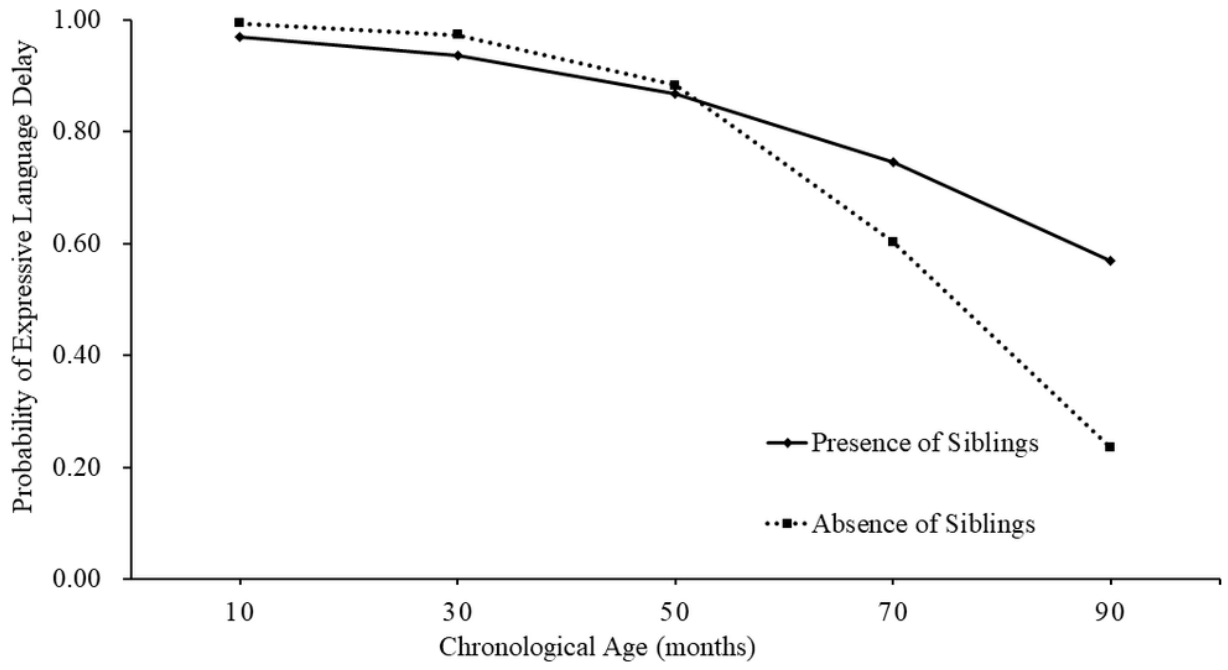

Figure 2

Predicted expressive language delay trajectories for the presence and absence of siblings after controlling for confounders.

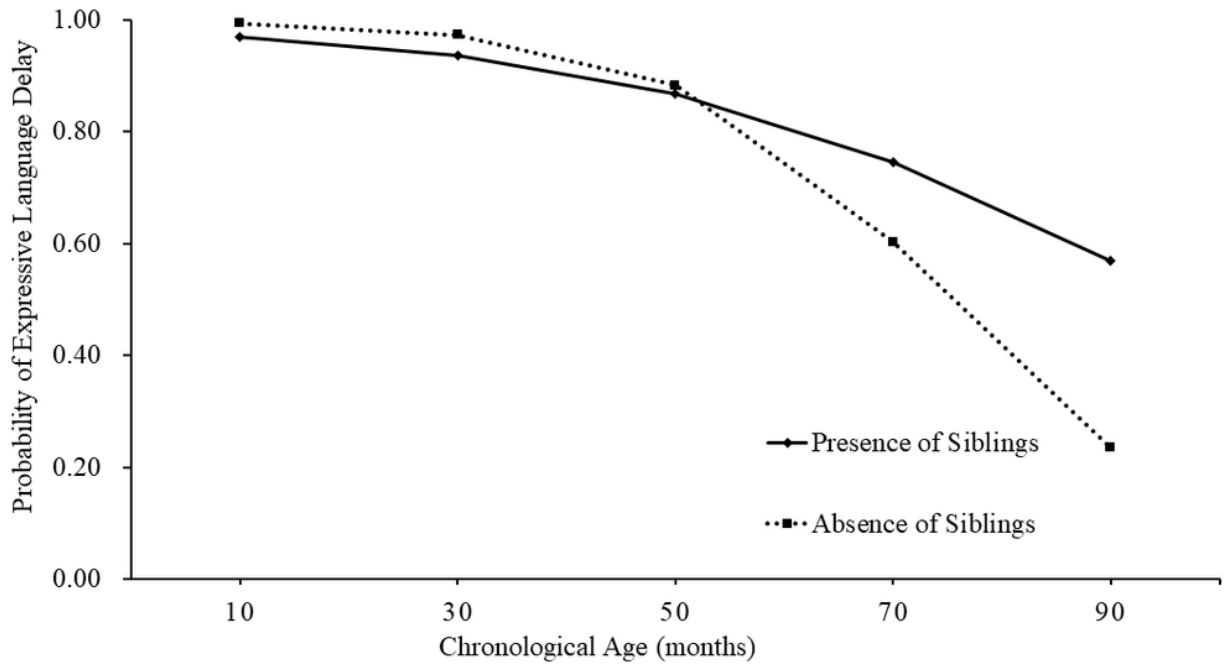

Figure 2

Predicted expressive language delay trajectories for the presence and absence of siblings after controlling for confounders.

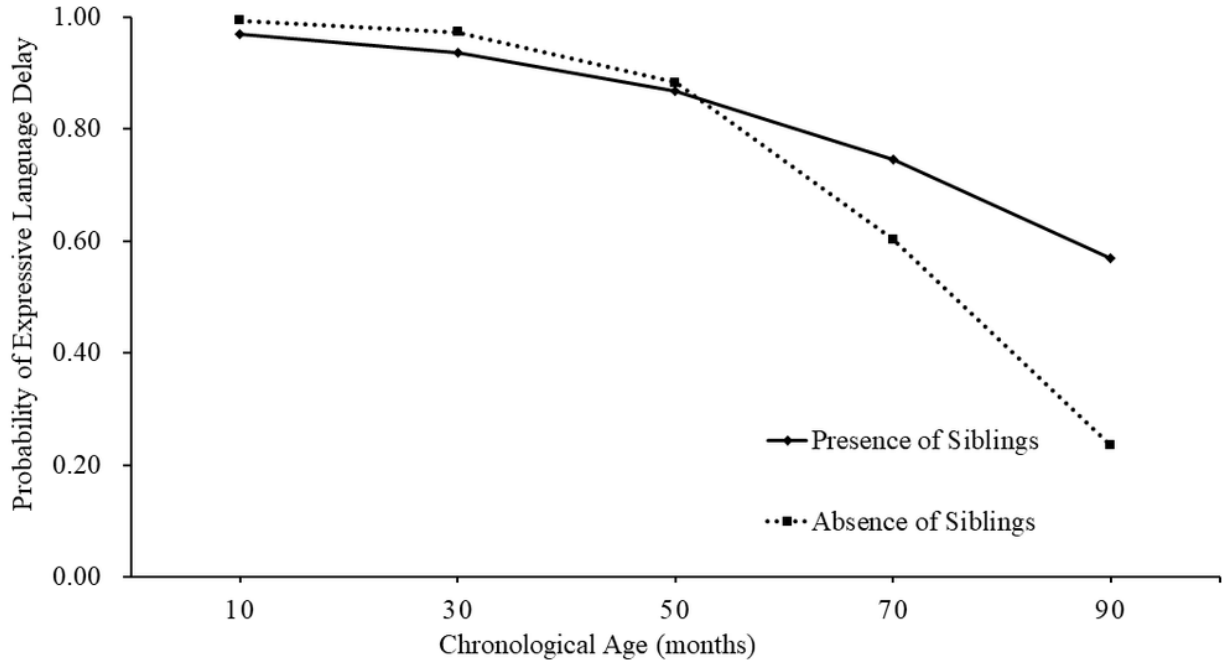

Figure 2

Predicted expressive language delay trajectories for the presence and absence of siblings after controlling for confounders.