OPEN

The Impact of Family Environment on Language Development of Children With Cochlear Implants: A Systematic Review and Meta-Analysis

Daniel Holzinger,[1–3] Magdalena Dall,[1,2] Susana Sanduvete-Chaves,[4] David Saldaña,[5] Salvador Chacón-Moscoso,[4,6] and Johannes Fellinger[1,2,7]

Objectives: The authors conducted a systematic review of the literature and meta-analyses to assess the influence of family environment on language development in children with cochlear implants.

Design: The Pubmed, excerpta medica dataBASE (EMBASE), Education Research Information Center, cumulative index to nursing and allied health literature (CINAHL), Healthcare Literature Information Network, PubPsych, and Social SciSearch databases were searched. The search strategy included terms describing family environment, child characteristics, and language development. Studies were included that (a) assessed distal family variables (such as parental income level, parental education, family size, and parental stress) with child language outcomes, and/or more proximal correlates that directly affect the child (such as family engagement and participation in intervention, parenting style, and more specifically, the quantity and quality of parental linguistic input) on child language; (b) included children implanted before the age of 5 years; (c) measured child language before the age of 21 years with standardized instruments; (d) were published between 1995 and February 2018; and (e) were published as peer-reviewed articles. The methodological quality was assessed with an adaptation of a previously validated checklist. Meta-analyses were conducted assuming a random-effects model.

Results: A total of 22 study populations reported in 27 publications were included. Methodological quality was highly variable. Ten studies had a longitudinal design. Three meta-analyses on the correlations between family variables and child language development could be performed. A strong effect of the quality and quantity of parental linguistic input in the first 4½ years postimplantation on the child's language was found, $r = 0.564$, $p \leq 0.001$, 95% confidence interval (CI) = 0.449 to 0.660, accounting for 31.7% of the variance in child language outcomes. Results demonstrated high homogeneity, $Q(3) = 1.823$, $p = 0.61$, $I^2 = 0$. Higher-level facilitative language techniques, such as parental expansions of the child's utterances or the use of open-ended questions, predicted child language skills. Risk of publication bias was not detected. The results on the impact of family involvement/participation in intervention on child language development were more heterogeneous. The meta-analysis included mainly cross-sectional studies and identified low to moderate benefits, $r = 0.380$, $p \leq 0.052$, 95% CI = –0.004 to 0.667, that almost attained significance level. Socioeconomic status, mainly operationalized by parental level of education, showed a positive correlation with child language development in most studies. The meta-analysis confirmed an overall low and nonsignificant average correlation coefficient, $r = 0.117$, $p = 0.262$, 95% CI = –0.087 to 0.312. A limitation of the study was the lack of some potentially relevant variables, such as multilingualism or family screen time.

Conclusions: These data support the hypothesis that parental linguistic input during the first years after cochlear implantation strongly predicts later child language outcomes. Effects of parental involvement in intervention and parental education are comparatively weaker and more heterogeneous. These findings underscore the need for early-intervention programs for children with cochlear implants focusing on providing support to parents for them to increase their children's exposure to high-quality conversation.

Key words: Cochlear implantation, Child, Family, Family involvement, Hearing loss, Language, Pediatric, Meta-analysis, Parental education, Parental linguistic input, Socioeconomic status, Systematic review.

(Ear & Hearing 2020;41;1077–1091)

INTRODUCTION

Cochlear implants (CIs) have significantly improved speech and language development of children with profound hearing loss. However, on average, children with CIs are delayed in spoken language development compared with children with normal hearing (Geers et al. 2009; Niparko et al. 2010; Lund 2016; Yoshinaga-Itano et al. 2018). Research on predictors of language development in children with CIs has identified many child characteristics (such as nonverbal intelligence, residual hearing, sex, additional disabilities, or residual hearing preimplantation), implant-related variables (such as age at implantation, duration of implant use, bilateral versus unilateral implantation, implant technology, or surgical factors), and intervention characteristics (such as communication mode or school setting), but leaves a high proportion of unexplained variance (Geers et al. 2007; Wu et al. 2011; Geers & Sedey 2011; Pisoni et al. 2017).

In research on typical child development, effects of the family on child language have been demonstrated extensively and consistently (Hoff 2006; Rowe 2012). Besides more distal variables such as socioeconomic status (SES) (Whitehurst 1978; Rowe et al. 2005), measures of variables representing the proximal environment have been shown to add significantly to the prediction of child language development. These

[1]Konventhospital Barmherzige Brüder, Institut für Sinnes- und Sprachneurologie, Linz, Austria; [2]Research Institute for Developmental Medicine, Johannes Kepler University Linz, Linz, Austria; [3]University of Graz, Institute of Linguistics, Graz, Austria; [4]Departamento de Psicología Experimental, Universidad de Sevilla, Seville, Spain; [5]Laboratorio de Diversidad, Cognición y Lenguaje Departamento de Psicología Evolutiva y de la Educación, Universidad de Sevilla, Seville, Spain; [6]Departamento de Psicología, Universidad Autónoma de Chile, Santiago, Chile; and [7]Division of Social Psychiatry, Medical University of Vienna, Vienna, Austria.

Supplemental digital content is available for this article. Direct URL citations appear in the printed text and are provided in the HTML and text of this article on the journal's Web site (www.ear-hearing.com).

<zdoi; 10.1097/AUD.0000000000000852>

include the style of parenting, for example, parental sensitivity and positive regard, and parental language, such as language input quantity and quality (Hart & Risley 1995), and home literacy environment. More recent findings demonstrate the role of early developing neural mechanisms that underlie the relationship between children's language exposure and their language development. As an example, Garcia-Sierra et al. (2016) found a specific impact of the amount of child-directed language on young children's brain functioning by showing significant correlations between higher quantity of language input and speech perception at 11 to 14 months of age as measured by event-related potentials, thus demonstrating that early neural reorganization is dependent on input (also referred to as neural commitment to language). Beyond that, Romeo et al. (2018) found significant correlations between the amount of early adult–child conversation and children's brain structure, specifically the strength of connectivity in the left hemisphere dorsal white matter language tracts. In both studies, children's real-world language exposure had been assessed with the Language Environment Analysis System (Gilkerson et al. 2017).

Research on the role of specific family characteristics and behaviors in child language development is further justified by converging evidence on the effectiveness of parent-implemented language interventions for different populations, such as children with primary and secondary language impairments (meta-analysis by Roberts & Kaiser 2011) or children with low SES (Hoff 2006; Hirsh-Pasek et al. 2015). As a consequence of the high and partly unexplained variance of language outcomes in children with CIs on the one hand, and the well-documented effects of the family variables in populations with normal hearing on the other, the role of family environment has gained interest in more recent studies (Niparko et al. 2010; Holt et al. 2013; Geers et al. 2017). But, so far, no systematic review and meta-analysis of the effects of family environment on language development in children with CIs have been carried out.

Purpose

The aims of the current review were to assess the evidence showing an impact of family environment on child language development in children with early cochlear implantation by conducting a systematic narrative review. In addition, meta-analyses on the correlations between family environment and language outcomes were contemplated, dependent on a sufficient number of studies with sufficient data.

To reduce the number of confounding auditory variables, the review was restricted to children with CIs (versus children with hearing aids or other technologies). Only studies referring specifically to spoken language outcomes (versus written or signed language development) were included.

Based on research results for typical language development, we included distal variables (such as parental SES, parental education, and family size), as well as more proximal factors, such as parenting style (e.g., parental sensitivity, emotional availability, and provision of control and structure), parental engagement in intervention, and the amount and kind of parental language input. Distal family variables may be less modifiable clinically than proximal variables, the latter of which may be more susceptible to the influences of interventions. Proximal family variables were expected to have a stronger influence on child language than distal family variables.

MATERIALS AND METHODS

A systematic review of the literature was carried out in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) (Moher et al. 2009; see Supplemental Digital Content 1, http://links.lww.com/EANDH/A623) and the Meta-analysis Of Observational Studies in Epidemiology (MOOSE) (Stroup et al. 2000) checklists, and registered on the international PROSPEctive Register Of systematic reviews (PROSPERO) (Holzinger et al. 2017). Before commencing the review, the authors specified, with internal protocols, the search strategy, selection criteria, procedures for data extraction, and a catalog of criteria for quality assessment. At the time of registration, the use of meta-analyses was considered subject to the availability of comparable data on family environment and child language outcomes. Evidence for policy and practice information reviewer 4 software was used for data handling.

Search Strategy

The following electronic databases were searched: Pubmed, EMBASE, Education Research Information Center, CINAHL, Healthcare Literature Information Network, PubPsych, and Social SciSearch. The search strategy included terms describing the child, language development, and family environment. The search string was adapted to each database in combination with database-specific filters and search terms (for the search string adapted to Pubmed, see Supplemental Digital Content 2, http://links.lww.com/EANDH/A643). The last update of the search results was on February 6, 2018. Articles published in non–peer-reviewed journals or unpublished literature were not included in this review. The reference lists of the papers that met the inclusion criteria after title-and-abstract screening (see later) were also searched for potentially eligible articles. This procedure was replicated until no more studies of interest were found.

Selection Criteria

The same criteria were used for the title-and-abstract and full-text screening phase. Research in English, German, or Spanish language and published after the year 1995 was included (to ensure that single-channel technology was excluded). The selection criteria were defined in line with Population, Intervention, Comparator, Outcome, Study Design (Schardt et al. 2007). The participants were children with hearing loss who received their cochlear implantation before the age of 5 years. Studies that included children implanted at an older age or children supplied with hearing aids were included only if they reported separate data on the population of interest. Family characteristics had to be mentioned in the abstract. The outcome had to be measured before the age of 21 years. Both prospective and retrospective studies were included in the review. Systematic reviews were excluded. Unspecified information in the abstract on any of the criteria was not a reason for exclusion at the title-and-abstract phase. In those cases, the full text was screened before deciding if it was included or not.

Study Selection

The first 202 abstracts (20% of the total obtained) were reviewed independently by 2 of the authors with high level of expertise in the field of pediatric hearing loss (D.H. and M.D.). Any disagreements were resolved by consensus. Afterward, the same 2 authors independently coded another 100 abstracts

and reached an inter-rater reliability of over 90%. Finally, M.D. reviewed the rest of the abstracts.

For the publications that met the inclusion criteria in title-and-abstract, the full texts were retrieved and valued by D.H. In this case, it was required that all information included in the criteria was present in the text.

Most of the studies included after the full-text screening did not contain sufficient data for a meta-analysis. The authors of these primary studies were contacted for additional information via e-mail.

Description of the Variables

The variables used for data extraction were agreed on by D.H., M.D., and D.S. Information was extracted on the following: study characteristics, language outcomes (dependent variables), family characteristics (considered independent variables [IV], and potential moderator variables that can influence the relationship between other family characteristics and language outcomes), and confounders (other potential moderator variables). A full list of variables included within each group can be found in the Supplemental Digital Content 3, http://links.lww.com/EANDH/A644.

Language variables extracted from the literature exclusively referred to spoken language (rather than signed or written), expressive or receptive communication, and the dimensions of speech sounds (phonology and speech perception), vocabulary (lexicon), or grammar (morpho-syntax). In addition, social (pragmatic) communication, including narrative skills, was considered.

Family variables included general sociodemographic characteristics, such as parental SES (parental income level, neighborhood index, parental education level), family size, and family-system characteristics (e.g., relationships within the family, parenting style, and parental stress). Other family variables referred to the families' involvement in intervention and their self-efficacy. Finally, family behaviors assumed to be more directly related to child language outcomes were considered, such as parenting style (e.g., sensitivity, emotional availability, warmth, and regard), and parental language input quality and quantity. Although quantity and quality measures are strongly associated with one another (Hoff & Naigles 2002), research suggests that both the amount and type of caregiver–child interaction play a significant role in language development and were considered distinct concepts in the systematic review. In a number of studies (Hirsh-Pasek et al. 2015; Gilkerson et al. 2018; Romeo et al. 2018), quality variables such as vocabulary sophistication and diversity, grammatical complexity, the use of facilitative language strategies, and interactional features (contiguous and contingent back-and-forth conversation) were shown to be stronger predictors of later language ability than the caregivers' total number of words or utterances during interaction. In a study by Rowe (2012), quality emerged as a distinct source of variability in language performances even after controlling for quantity of language input. Adult reading time, frequency of dialogic book reading, and parental teaching or tutoring literacy skills (e.g., the alphabet, phoneme awareness, reading of words) were categorized as home literacy environment (Sénéchal & LeFevre 2014). Parental communication with the child who is deaf or hard-of-hearing was usually assessed with time-consuming transcriptions and ratings of videotaped interactions, whereas other family environment variables were collected by the use of standardized or nonstandardized questionnaires from parents or practitioners.

Children's sex, intelligence quotient (I.Q.), chronological age, age at implantation, age at hearing loss, residual hearing, and unilateral/bilateral hearing loss were included as potential confounders.

Methodological Quality

The methodological quality of each study was assessed with a selection and adaptation of items relevant to our research question from a tool proposed by Chacón-Moscoso et al. (2016). Methodological quality assessment was based on (1) adequate inclusion and exclusion criteria for the participants, (2) the study design, (3) proportion and inclusion or not of attrition rates, (4) occasion of measurement, more than one measurement occasion (concurrent and post intervention) or only one measurement occasion (concurrent or postintervention only), (5) for longitudinal studies, whether the outcome measurements at time 1 also appeared at time 2, (6) the use of standardized instruments for the measurement of language outcomes (dependent variables), (7) family input and environment (IV), (8) use of control techniques such as double blinding (language and family variables assessed by different evaluators), (9) replicability of the construct definition of outcome, (10) family variables, (11) proportion of participants contacted who actually responded, (12) participant representativeness, (13) imputation of missing data, and (14) perspective (prospective or retrospective) (see Supplemental Digital Content 4, http://links.lww.com/EANDH/A645). The degree of methodological quality was considered an indicator of the level of credibility of the final results.

Data Extraction

Two coders, D.H. and M.D., piloted the data extraction on 5 full texts and discussed the disagreements with the arbitration of DS, a third researcher with high level of expertise in the area of child development. Afterward, data extraction was performed by D.H. and M.D independently, double-coding all papers.

A meta-analysis was performed on the data from the papers which, after following up with the authors, provided enough data. For the meta-analysis, only one language outcome variable was produced. Because there are high correlations between lexical, grammatical, and global language, as well as between receptive and expressive language development in children with hearing loss (Geers et al. 2009; Holzinger et al. 2011; Lund 2016), all those variables were regarded as representations of a common language category. When a study included several language measures, only one was selected, according to the following sequential criteria: first, global over specific language measures were preferred; second, expressive over receptive language measures; and third, language measures available for the largest sample were chosen.

Intercoder reliability was calculated using Cohen's k coefficient. Disagreements were solved through consensus.

Statistical Methods

Calculations were made with the Comprehensive Meta-Analysis v. 3 software (Borenstein et al. 2013). The individual and average effect sizes were calculated as Pearson's correlation coefficients. Three average effect sizes were obtained separately, depending on the IV measured: (a) parental linguistic input, (b)

family involvement, and (c) parental SES. Two coauthors (S.S.-C. and S.C.-M.) calculated the individual and average effect sizes independently, obtaining an intercoder reliability of 0.992, using the intraclass correlation coefficient.

For each average effect size, a random-effects model was assumed. This type of model is recommended in this case, given the representativeness of the studies found as a result of the exhaustive systematic review carried out, and the diversity of such studies in characteristics of the samples, the scenarios where they were conducted, and the results obtained (Borenstein et al. 2009). Individual effect sizes were converted to the Fisher's z scale, with its statistical significance and confidence interval. Then, the summary of Fisher's z was calculated. Finally, this result was transformed into an r scale (Borenstein et al. 2009), with values around 0.1 considered low effect sizes, around 0.25, medium, and around 0.4, high (Cohen 1988). Confidence intervals and statistical significance (p) are also reported. A $p < 0.05$ was interpreted as a statistically significant average effect size.

Heterogeneity was calculated with the Q statistic, where $p < 0.05$ would imply a possible statistically significant heterogeneity between effect sizes. In addition, given that Q is sensitive to the number of studies included, I^2 was also calculated. Values around 25% were interpreted as a low heterogeneity; around 50%, medium heterogeneity; and around 75%, high.

When heterogeneity was found, all the potential moderator variables that presented enough information to carry out the analyses were studied assuming a mixed-effects model. Meta-regression was used, given that all the moderated variables with available information to be included were quantitative. Z values with associated p values were obtained. A statistically significant influence of the moderator variable over the effect size was detected when $p < 0.05$.

Finally, publication bias was analyzed using Duval and Tweedie's Trim and Fill (Borenstein et al. 2009). When the observed point estimate (represented by an open diamond) is close to the imputed point estimate (shown as a filled diamond), we interpreted that there was no risk of publication bias. When we found differences between the observed and the imputed point estimate, we calculated Egger regression test for bias (Borenstein et al. 2009). A risk of bias was considered significant when p values were below 0.05.

RESULTS

Study Selection

In total, 1012 individual publications were identified and included into the first of 3 selection steps. After screening on title-and-abstract, 172 publications remained and the full texts were retrieved. The screening of full texts excluded 145 publications that either did not meet the inclusion criteria or referred to the same study sample without reporting on an additional family variable, which led to 27 studies remaining to be included in the review. These 27 studies refer to 22 different study samples (Fig. 1 and Supplemental Digital Content 5, http://links.lww.com/EANDH/A646).

Specifically, the same study samples in full or partly were Cruz et al. (2013) and Quittner et al. (2013). The studies Geers et al. (2003, 2011) were looking at the longitudinal effects of the same study population. The studies Holt and Svirsky (2008) and Holt et al. (2013) investigated subpopulations from a larger longitudinal study. The study from Szagun and Schramm (2016) included the study sample from Szagun and Stumper (2012). The studies Sarant and Garrard (2014) and Sarant et al. (2014) were most likely sharing at least some of the study participants.

Methodological Quality

The results for intercoder reliability for each item of the quality assessment are presented in Supplemental Digital Content 6, http://links.lww.com/EANDH/A647. Seven of 14 items had a very good kappa (above 0.8). The other seven items had a substantial kappa (values between 0.6 and 0.8).

The quality assessment for the 27 publications can be found in the Supplemental Digital Content 7, http://links.lww.com/EANDH/A648. In total, 16 (59.3%) studies gave clear inclusion/exclusion criteria. Only 10 (37%) studies had a longitudinal study design. In 23 (85.2%) studies, attrition was mentioned. Eight (29.6%) studies measured the outcome variable and the family variable concurrently and had post measurement of the family variable. Eight (29.6%) studies collected all outcome and family measures at every time point. All studies (100%) used standardized measurements for language outcome measures. Twenty-three (85.2%) studies used at least one standardized measurement for the family variable. In none of the studies (0%), control techniques were reported. The construct definitions for the language outcome variables as well as family variables were described in all studies (100%), although in 2 of them, the definition of the family variables was vague. Within this review, only 11 (40.7%) studies mentioned a responder rate. Eight (29.6%) studies mentioned representativeness of the samples. In those that did, the sample was highly representative in 6 and low in 2. From the 23 studies that presented attrition, only 2 (8.7%) imputed missing data statistically. Twenty-one publications had a prospective design (77.8%). Inclusion of a study in a meta-analysis was not determined by its methodological quality but exclusively by the availability of correlational data between the family and child language variables of interest.

Study Characteristics

Table 1 provides an overview of the most relevant study characteristics. In addition, the number of studies with sufficient information available to be included in one of the meta-analyses is reported. The availability of data is reported separately for the three meta-analyses. Finally, the number of the most recent studies, published since 2015, is shown.

Thirty-five percent of all studies were conducted in North America; there is a complete lack of literature from African and Asian countries in the final selection of studies, except for China. Therefore, the available data mostly pertain to high-income countries. Of the studies reporting family SES, there is a tendency to disproportionally include families with high SES. As expected, more distal family characteristics such as family education level are reported in many publications (22), although there is also a significant number including family involvement (8) or parental linguistic input (9). Other family variables such as parenting style, family values, relationships, or parental stress are investigated rather rarely. For the language measures, global language variables measured by comprehensive language tests as well as measures of expressive vocabulary are used most often. There is a dearth of studies with a focus on social language use or narrative language skills. Age at implantation is the most commonly reported confounding variable. Surprisingly, other variables known to be significantly

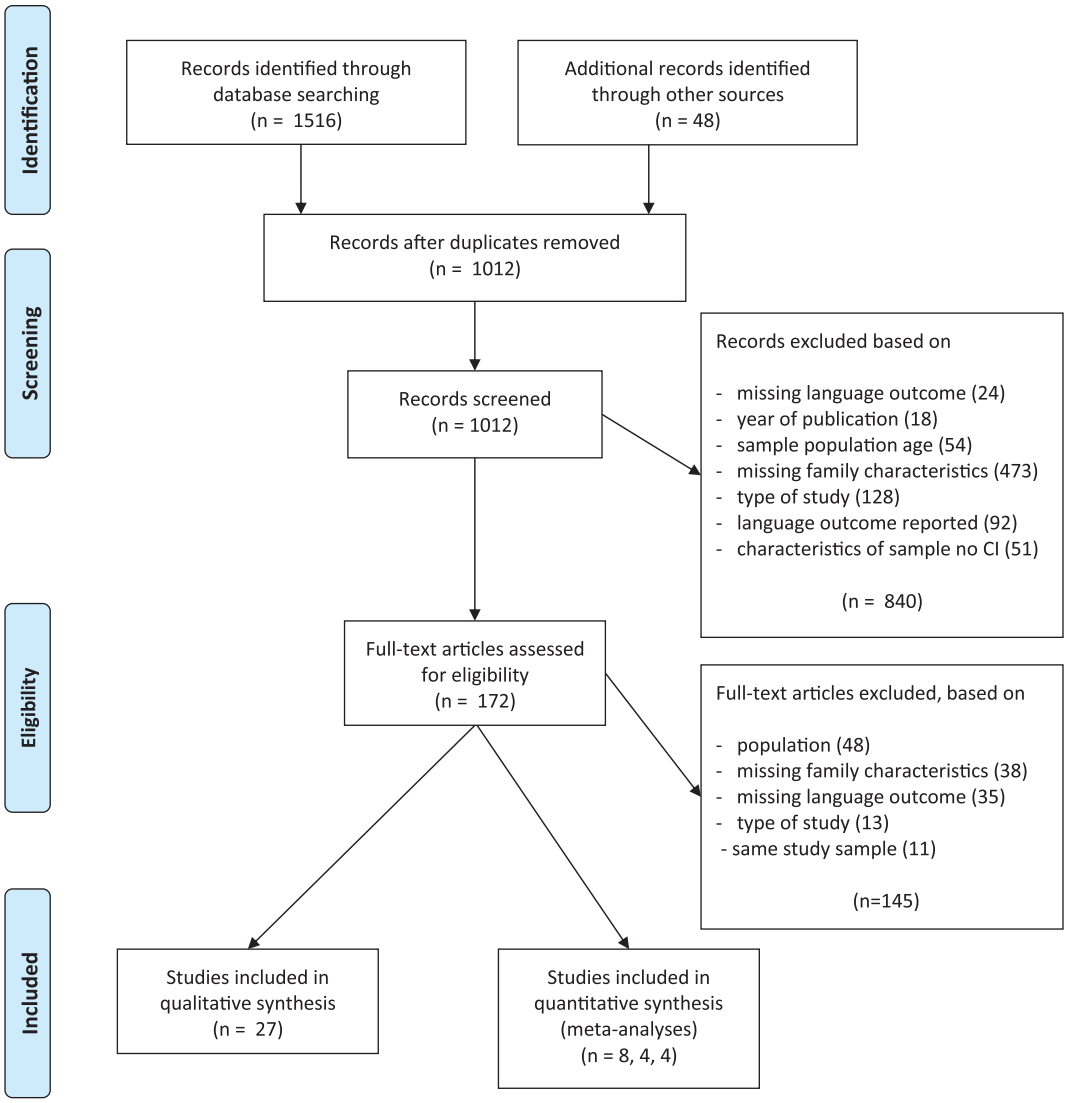

Fig. 1. Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) flow chart of the process of study selection. Retrieved from Moher et al. (2009).

associated with language development, such as child sex or I.Q., are only reported in about one-third of the studies.

Narrative Syntheses and Meta-Analyses

The studies investigating child speech and social communication skills and family variables could not be included in any meta-analyses due to insufficient correlational data. Based on the availability of correlations with same/similar dependent and IV, three meta-analyses for the influence of different family characteristics on child language were conducted. The presentation of results begins with the more proximal variables. For each of the three family variables, a narrative description of the studies not included in the meta-analysis is presented first and followed by the results of the meta-analysis (of only studies with sufficient data).

Parental Linguistic Input

A direct and significant correlation between parental language input and child language development was anticipated. Seven studies reported relationships between child-directed parental language and child language outcomes; for 4 of those studies, data were available to perform a meta-analysis. All of the studies that could be included in a narrative review or meta-analysis referred to quality rather than quantity of parental language.

With the exception of a single-case study (Szagun 1997), the studies only included in the narrative review were quite recent (2012 to 2013) and related to children implanted at a young age (range = 1.25 to 1.7 years, total n = 66) (Table 2). The German study with the smallest sample size (Szagun & Stumper 2012) and the case study (Szagun 1997) were the only longitudinal ones. Szagun & Stumper (2012) found significant correlations between the structural complexity of maternal language, that is, mean length of utterance (MLU) and the frequency of maternal expansions (reacting to a child by adding linguistic information to his/her utterance) 12 months postimplantation, and the child's MLU at 24 and 30 months postimplantation. The correlations demonstrated a significant moderate effect of quality aspects of maternal language, even after partialling out age at implantation and the child's MLU at 12 months, which indicates a specific

TABLE 1. Characteristics of studies included in the review (N = 27)

Variable	N	Data Available				Published After 2015
		Total	Meta-Analysis			
			Ling*	Involvement†	SES‡	
Type of study						
Cross-sectional	17	12	0	3	6	6
Longitudinal	10	6	4	1	2	2
Perspective						
Prospective	21	15	4	3	8	7
Retrospective	6	3	0	1	0	1
Country						
United States + Canada	9	6	2	1	2	1
Australia	4	2	0	1	3	2
China	1	0	0	0	0	3
Germany	4	2	2	0	0	1
Italy	3	2	0	0	1	2
Belgium	2	2	0	1	0	0
Other countries	4	4	0	1	2	3
Distribution of SES/parental education						
Mixed	11	8	2	3	5	3
High	5	4	0	1	2	2
Not reported	11	6	2	0	1	4
Family variables						
Parental income level	9	5	2	1	2	1
Parental education level	16	13	2	3	8	7
Family size	2	2	0	1	0	0
Family relationships	1	1	0	0	0	0
Family values	1	1	0	0	0	0
Parental stress	1	1	0	1	1	0
Family involvement	8	6	0	4	2	2
Parental self-efficacy	0	0	0	0	0	0
Parenting style	2	1	0	0	1	1
Parental linguistic input quality	5	3	4	0	0	1
Parental linguistic input quantity	4	2	0	0	0	1
Home literacy environment	2	1	1	0	1	0
Language outcomes						
Speech perception	3	1	0	0	1	1
Speech production	3	3	0	1	3	2
Size of receptive vocabulary	6	4	0	1	3	1
Size of expressive vocabulary	12	9	2	2	5	5
Grammar receptive	0	0	0	0	0	0
Grammar expressive	8	6	2	2	2	1
Grammar expressive length	3	1	0	0	0	1
Grammar composite expressive/ receptive	1	1	0	0	0	1
Global receptive	11	8	1	1	4	2
Global expressive	11	8	1	2	4	2
Global expressive/receptive combined	5	3	0	1	2	0
Social communication skills	4	4	0	1	1	3
Confounders						
Chronological age	5	3	0	2	0	1
Sex	11	7	1	2	3	2
I.Q.	7	5	1	1	3	1
Age at implantation	19	13	2	4	7	7
Age at onset of hearing loss	5	2	0	1	1	1
Residual hearing/PTA	5	2	0	0	2	3
Communication mode	6	4	0	1	2	2
Unilateral/bilateral	5	4	0	2	2	0

**Meta-analysis parental linguistic input.*
†Meta-analysis family involvement.
‡Meta-analysis parental socioeconomic level.
I.Q., intelligence quotient; PTA, pure tone average; SES, socioeconomic status.

TABLE 2. Studies reporting effects of parental linguistic input not included in the meta-analysis

Narrative Synthesis												
First Author	Year	Country	D	N	Age in Years (SD)*	Age at CI in Years (SD)	Gender % Female	Family Variable	Measure Family Variable	Language Variable	Measure Language Variable	Main Results
Ceh et al.	2013	United States	C	39	5.78 (1.44)	1.25 (0.3)	56	Quality	Use of open-ended questions while book reading	Global receptive	RDLS or OWLS	Associated sig. pos. with language outcomes
Szagun	1997	Germany	L	2	3.10	2.9	50	Quality	Mothers degree of exaggerated intonation; degree of clarity of segmentation distribution of pragmatic categories	Grammar expressive Social conversational	MLU morphological marking, distribution of pragmatic categories	Substantial child language differences associated with concurrent maternal use of language
Szagun and Stumper	2012	Germany	L	25	n.a.	1.7 (0.92)	48	Quality	CHILDES maternal MLU + maternal expansions	Vocabulary expressive Grammar expressive Grammar receptive	CDI questionnaire vocabulary CHILDES transcriptions number of word types	Maternal MLU associated sig. pos. with child MLU at 24 mo Maternal expansions associated sig. pos. with child MLU at 24 and 30 mo

*Age at baseline/testing T_1.

C, cross-sectional; CDI, communicative development inventories; CHILDES, Child Language Data Exchange System; CI, cochlear implantation; D, design; L, longitudinal; MLU, mean length of utterance; OWLS, Oral and Written Language Scales; pos, positive; RDLS, Reynell Developmental Language Scales; sig, significant.

causal influence. Ceh's et al. (2013) findings indicated positive effects of the use of open-ended questions during book reading encouraging the child's more active participation in linguistic interaction. In Szagun's (1997) case study, longitudinal data of mother–child interactions demonstrate substantial differences in language development between the 2 cases as well as in the language of their mothers (speech and pragmatic functions). However, an influence of maternal language on the children's linguistic development cannot be inferred, because the aspects of parental language expected to be related to child language variables did not chronologically precede these.

All 4 studies included in the meta-analysis had a longitudinal design (Table 3). Two of them were conducted in the United States, the other 2 in Germany. Even though the size of the total number of participants was limited (n = 176), the data were considered as valuable being based on time-consuming transcriptions of videos of parent–child interactions. All the family measures pertained to facilitative language techniques (FLT), that is, qualitative language input. In 2 of the studies (DesJardin et al. 2009; Cruz et al. 2013), parental higher-level FLTs significantly predicted growth in children's expressive language. Higher-level FLTs included parental reactions to a child's linguistic utterance with expansions or recasts (restating the child's utterance in a question format), reactions to their child's current interests by describing and commenting on them, and the use of open questions encouraging the child's use of more complex language. Szagun & Schramm's (2016) study related to the use of parental expansions. Rüter (2011), in addition, referred to their grammatical complexity. Three of the studies measured parental language use with their children at 6 to 24 months postimplantation; DesJardin et al. (2009), at 53 months postimplantation. Child language outcomes were assessed 1 to 3 years later.

The meta-analysis demonstrated a strong effect ($r = 0.564$; $p \leq 0.001$; 95% CI = 0.449 to 0.660), explaining a variance of 31.7% of the children's global expressive language development, expressive grammar, or vocabulary size (Fig. 2). The results demonstrated high homogeneity among effect sizes, $Q(3) = 1.823$, $p = 0.61$, $I^2 = 0$. A study of moderator variables was thus not considered necessary.

The funnel plot showed no apparent risk of publication bias (Fig. 3).

Family Involvement

The relationship between family involvement and child language development was investigated in a total of 6 studies.

Due to insufficient data, 2 of the studies could not be included in the meta-analysis (Table 4). One of them was cross-sectional and was conducted in Belgium (Boons et al. 2012). Another was a longitudinal Australian study (Yanbay et al. 2014). Both studies demonstrated statistically significant positive correlations between family involvement and at least some of the language measures. However, for both studies, there were serious methodological constraints. Boons et al. (2012) used a nonvalidated binary scale to classify parental motivation and ability to fulfill their commitments in rehabilitation from information in the child's file. In Yanbay et al.'s study (2014), Moeller's Family Participation Rating Scale was filled in by educators/therapists working directly with the families and thus not blinded for the children's language development. Moeller's concept of family involvement includes parental adjustment to the child's hearing impairment,

TABLE 3. Studies reporting effects of parental linguistic input included in the meta-analysis

First Author	Year	Country	D	N	Age in Years (SD)*	Age at CI in Years (SD)	Gender % Female	Family Variable	Measure Family Variable	Language Variable	Measure Language Variable	Main Results
Meta-Analysis												
Cruz et al.	2013	United States	L	93	1.22 (0.48)	1.38 (0.4)	46	Quality at 24 mo post-CI	High level FLTs	Global expressive at 36 mo post-CI	RDLS expressive language scale	Associated sig. pos. with language outcomes
DesJardin et al.	2009	United States	L	16	4.42 (1.05)	2.16 (0.58)	56	Quality at 53 mo post-CI	Parental use of recasts during story book reading	Vocabulary expressive 89.6 mo post-CI	OWLS	Associated sig. pos. with oral vocabulary
Rüter	2011	Germany	L	21	n.a.	2.5	52	Quality at 5 mo post-CI	Parental expansions	Grammar expressive at 18 mo post-CI	Child MLU	Associated sig. pos. with language outcomes
Szagun and Schramm	2016	Germany	L	48	n.a.	2.0 (0.1)	50	Quality at 12 mo post-CI	Parental expansions	Grammar expressive 30 mo post-CI	Child MLU	Associated sig. pos. with language outcomes

*Age at baseline/testing T_1.
CI, cochlear implantation; D, design; FLTs, facilitative language techniques; L, longitudinal; MLU, mean length of utterance; OWLS, Oral and Written Language Scales; pos, positive; RDLS, Reynell Developmental Language Scales; sig, significant.

regular attendance and active participation in sessions, and advocating for the child. In addition, qualitative aspects of parental linguistic input (like those reported earlier) were included in the concept of family involvement; that is, the ability of families to become effective conversation partners with their children, the way they function as language models, the use of FLT, and their facility in the child's communication mode.

The meta-analysis included 4 studies referring to a total sample of 335 children in 4 different countries (Table 5). The study with the smallest sample size (Moreno-Torres et al. 2016) was the only longitudinal one. Most studies (except Geers et al. 2003) had been published recently and included children who, on average, had been implanted in their second year of life. Again, there were severe methodological limitations related to the construct validity of family involvement and the measures used in all the studies, which limited the validity of the results. Boons et al. (2013) asked audiologists or speech-language therapists to complete a self-constructed nonvalidated questionnaire with seven mostly indirect indicators of parental involvement (such as parental knowledge of their child's abilities, understanding how the CI works, attending appointments, or contacting professionals for help). The nonvalidated questionnaire used by Geers et al. (2003) asked parents to report the frequency with which they participated in activities to stimulate auditory and speech development in the home. The 2 remaining studies used Moeller's scale.

Three of the studies found positive weak to strong correlations between parental involvement and mainly expressive language development, whereas in Geers et al. (2003), a nonsignificant negative relationship was reported. Overall, a high-moderate mean correlation (r = 0.380, p = 0.052, 95% CI = −0.004 to 0.667), which almost reached significance was found (Fig. 4).

We found there was heterogeneity between the different effect sizes, $Q(3) = 30.639$, $p < 0.001$, $I^2 = 90.208$. However, the available moderator variables, that is, sex ($z = -1.01$, $p = 0.313$, 95% CI = −0.0984 to 0.0315) and age at implantation ($z = 1.39$, $p = 0.164$, 95% CI = −0.4306 to 2.5372) were found to be nonsignificant confounders.

Based on Figure 5, a certain degree of publication bias could be interpreted. However, the nonsignificant results in Egger regression test for bias, $t(2) = 2.7588$, $p = 0.11$, 95% CI = −3.4639 to 15.8440, indicated the risk of bias was not significant.

Parental SES

SES was conceptualized as the social standing or class of an individual or group, often measured as a combination of education, income, and occupation (American Psychological Association, Task Force on Socioeconomic Status 2007). Parental SES was commonly reported in the publications (22 publications, 16 study samples). From an overall of 16 studies, SES was measured using the maternal/main carer's or family's highest education (n = 12) and/or an income/neighborhood index (n = 6).

Eight studies could not be included in the meta-analysis. Five of these studies reported statistically significant but rather weak positive correlations between SES and child language, specifically with expressive/receptive vocabulary or global language (see Supplemental Digital Content 8, http://links.lww.com/EANDH/A649).

Eight studies pertaining to a total of 512 children and with good geographical variation could be included in a meta-analysis on the relationship between parental education level and

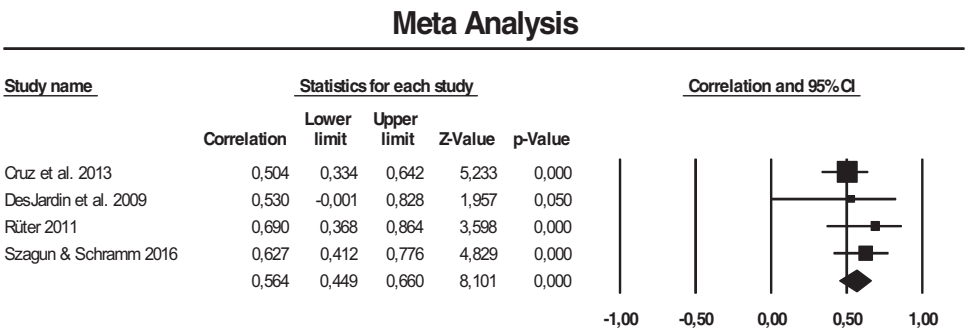

Fig. 2. Forest plot of parental linguistic input. CI indicates confidence interval.

child language (see Supplemental Digital Content 9, http://links.lww.com/EANDH/A650). All except for one study (looking at parental income) looked at parental/maternal education level. The meta-analysis resulted in an overall low- and nonsignificant average correlation coefficient, $r = 0.117$, $p = 0.262$, 95% CI = −0.087 to 0.312, so there was no statistical evidence of the relationship between parental SES and the language development of the child (Fig. 6).

There was a high level of heterogeneity between studies, $Q(7) = 28{,}598$, $p < 0.001$. $I^2 = 75.523$. The I.Q. of the child was a positive significant moderator variable, $z = 3.2080$, $p = 0.001$, 95% CI = 1.0069 to 4.1693. Sex and age at implantation were nonsignificant moderators, $z = 0.0569$, $p = 0.955$, 95% CI = −1.7194 to 1.8222 and $z = -0.1861$, $p = 0.852$, 95% CI = −0.5170 to 0.4273, respectively.

The funnel plot did not present indications of publication bias (Fig. 7).

Other Family Variables

In addition to the three family variables reported earlier, some additional aspects of family environments deserve mentioning, even though meta-analyses could not be performed due to a lack of data (Table 1).

Two studies (Ceh et al. 2013; Sarant et al. 2014) reported significant benefits of the frequency of the child's exposure to books in the home (home literacy environment) for his/her global language development. In Sarant's et al. (2014) study, time spent reading books even predicted oral language development more strongly than an extra 10 I.Q. points.

Another 2 studies (Geers et al. 2003, 2011) provided information on the relationship between family size and child speech and language development. Both of them showed statistically significant negative correlations, demonstrating possible advantages of smaller families for speech-language acquisition of a child with hearing loss.

Parenting style was found to be significantly related to child language development in a number of studies. Quittner et al. (2013) reported positive effects of maternal sensitivity and cognitive stimulation on the growth of oral language. In the study of Ketelaar et al. (2017), a negative and uninvolved parenting style was found to be negatively correlated with child language development. Holt et al. (2013) demonstrated statistically significant correlations indicating that lower family self-reported levels of control, implying less rule emphasis and less obvious hierarchy of power, and higher levels of organization (more planning,

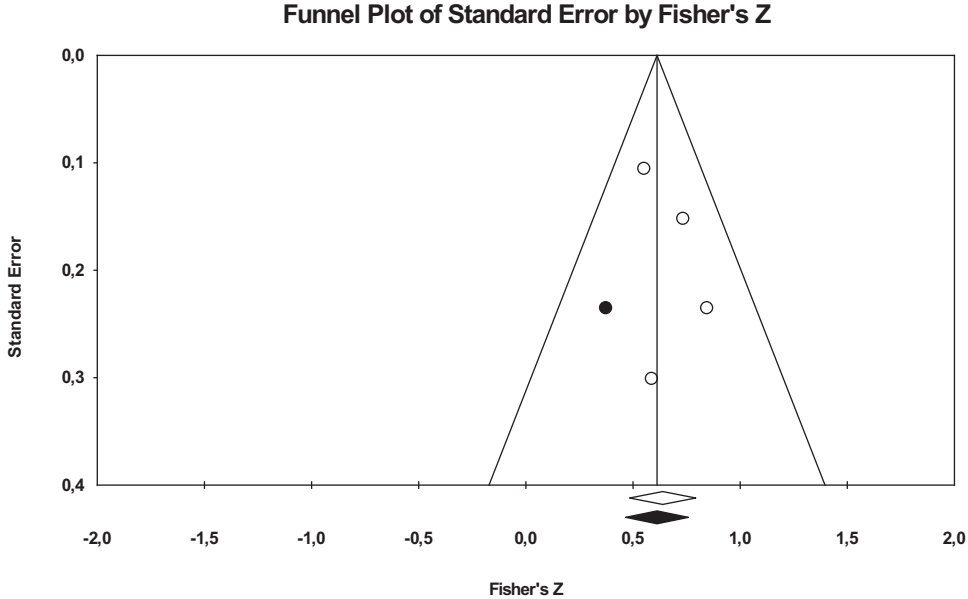

Fig. 3. Funnel plot of parental linguistic input.

TABLE 4. Studies reporting effects of family involvement not included in the meta-analysis

Narrative Synthesis												
First Author	Year	Country	D	N	Age in Years (SD)*	Age at CI in Years (SD)	Gender % Female	Family Variable	Measure Family Variable	Language Variable	Measure Language Variable	Main Results
Boons et al.	2012	Belgium	C	288	n.a.	2.16 (1.08)	47	Family involvement	Binary scale: parental involvement in rehabilitation	Vocabulary expressive Grammar expressive Global receptive	SELT word development SELT sentence development RDLS receptive	Associated sig. pos. with receptive language and word development
Yanbay et al.	2014	Australia	L	42	2.77–6.63 (range for subgroups)	0.53–3.30 (range for subgroups)	55	Family involvement	Moeller Family Participation Rating Scale	Global expressive Global receptive Vocabulary receptive	PLS-4 PPVT-4	Associated sig. pos. with language outcomes

*Age at baseline/testing T.
C, cross-sectional; CI, cochlear implantation; D, design; L, longitudinal; PLS-4, Preschool Language Scale Fourth Edition; pos., positive; PPVT-4, Peabody Picture Vocabulary Test Fourth Edition; RDLS, Reynell Developmental Language Scales; SELT, Schlichting Expressive Language Test; sig., significant.

TABLE 5. Studies reporting effects of family involvement included in the meta-analysis

Meta-Analysis												
First Author	Year	Country	D	N	Age in Years (SD)*	Age at CI in Years (SD)	Gender % Female	Family Variable	Measure Family Variable	Language Variable	Measure Language Variable	Main Results
Boons et al.	2013	Belgium/The Netherlands	C	70	8.2 (median)	1.8 (median)	56	Family involvement	Expert questionnaire (Calderon 2000; 7 indicators)	Vocabulary others (morphology)	CELF (formulating sentences + word structure)	Associated sig. pos. with all language outcomes
Geers et al.	2003	United States/ Canada	C	181	8.11 (0.6)	3.5 (0.10)	50	Family involvement	Parent questionnaire (26 items)	Global expressive	Composite score IPSyn (bound morphemes lexical diversity utterance length)	Associated non sig. pos. with language outcome
Moreno-Torres et al.	2016	Spain	L	14	n.a.	1.43 (0.18)	36	Family involvement	Moeller Family Participation Rating Scale	Vocabulary expressive	Spanish Version of the MacArthur parental questionnaire	Associated sig. pos. with language outcomes
Sarant and Garrard	2014	Australia	C	70	6.6 (1.4)	1.55 (0.8)	49	Family involvement	Moeller Family Participation Rating Scale	Global expressive/ receptive combined	PLS-4 or CELF-4	Associated sig. pos. with language outcomes

*Age at baseline/testing T.
C, cross-sectional; CELF, Clinical Evaluation of Language Fundamentals; CI, cochlear implantation; D, design; IPSyn, index of productive syntax; L, longitudinal; PLS-4, Preschool Language Scale Fourth Edition; pos., positive; sig., significant.

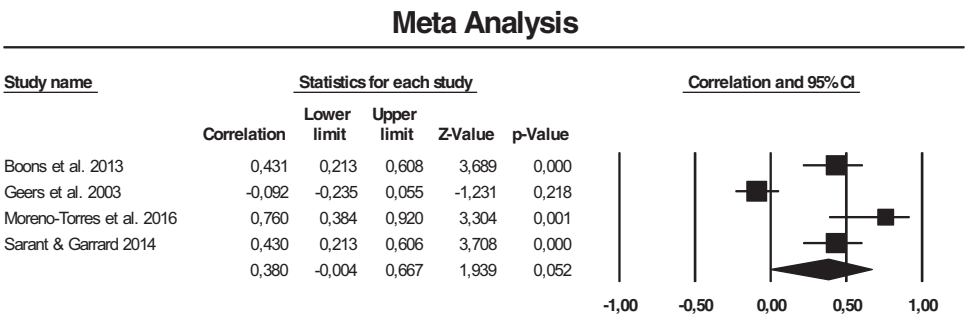

Fig. 4. Forest plot of family involvement. CI indicates confidence interval.

clearer expectations, and neatness without the control power) related to larger receptive vocabularies in their children.

Lower levels of parental stress were significantly correlated with children's speech-language development in one study (Sarant & Garrard 2014).

DISCUSSION

To our knowledge, this study is the first systematic review of the literature on the effects of family environment on language outcomes in children with CIs. The review was performed to decrease the unexplained variability in language outcomes in children with CIs by investigating the role of family variables. Twenty-seven studies that contained information on family variables as related to child language development were identified. Available data permitted the conduction of 3 meta-analyses on the influence of parental linguistic input (4 studies; DesJardin et al. 2009; Rüter 2011; Cruz et al. 2013; Szagun & Schramm 2016), family involvement (4 studies; Geers et al. 2003; Boons et al. 2013; Sarant & Garrard 2014; Moreno-Torres et al. 2016), and parental SES (8 studies; Geers et al. 2009, 2011; Huber & Kipman 2012; Sarant & Garrard 2014; Cupples et al. 2016; Guerzoni et al. 2016; Moreno-Torres et al. 2016; Ketelaar et al. 2017) on child linguistic skills. Our findings demonstrated strong and homogenous effects (r = 0.536) of the amount of high-quality early parental language input explaining variance of almost 32% of child language development after cochlear implantation. The use of parental expansions, such as reacting to the child's utterance by "playing it back" to the child in a linguistically correct form and with some new information, proved to be a highly effective facilitative language strategy. Another one was the use of open-ended questions. A common feature of both strategies is that they stimulate the child's active participation in linguistic interaction. Other characteristics of high-quality parental linguistic input were grammatical complexity (maternal MLU) and lexical diversity (type-token ratio). Frequency of a child's exposure to books in the home (home literacy) was found to predict child language development in 2 studies included in the narrative systematic review.

However, despite strong correlations between parental linguistic input and child language development documented in the longitudinal studies, it could be argued that these correlations could also reflect the influence of child characteristics on parental behavior. For example, there is evidence from twin studies (Dale et al. 2015) that, in addition to causal influences of parental language on child development, there are also child-to-parent effects. Children who were more talkative or advanced in language development elicit parental speech with

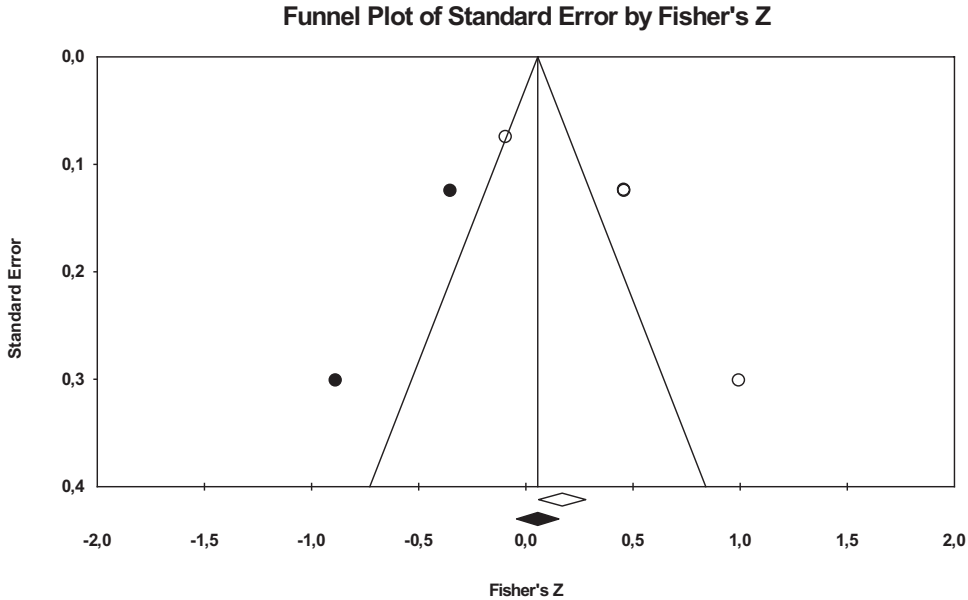

Fig. 5. Funnel plot of family involvement.

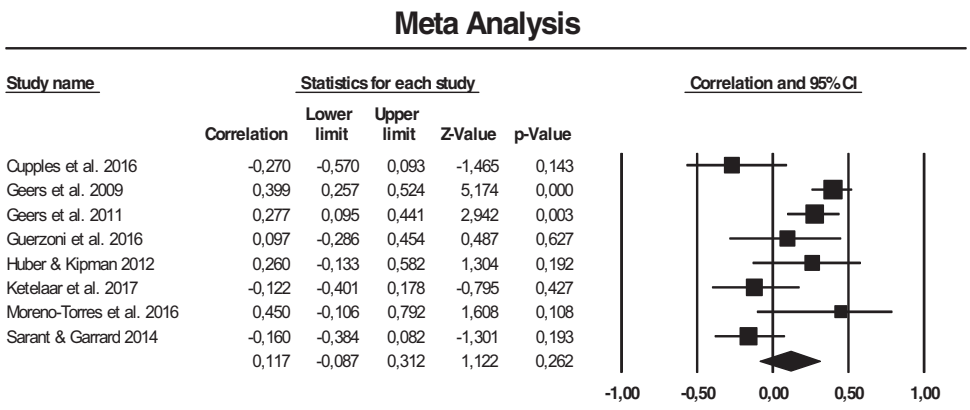

Fig. 6. Forest plot of parental socioeconomic status. CI indicates confidence interval.

more advanced language facilitating features, thereby creating their own language environment.

But the data from the primary studies in our review support the view that even considering the influence of child variables, parental input has an impact on language development. Two of the studies included in the meta-analysis (Rüter 2011; Szagun & Schramm 2016) controlled for the influence of the child's language level at baseline in their analysis of the correlation between early parental language input and later child language outcomes. Rüter (2011) demonstrated strong effects of parental expansions on different aspects of child expressive grammar outcomes (r of 0.42 to 0.70) even after partialling out child language at baseline (and age at implantation). In Szagun and Schramm's (2016) study, variance of child MLU at 24 months postimplantation explained by parental expansions at 11 to 12 months postimplantation ($R^2 = 0.048$) was still 12% (and 15% for early child MLU) when child and parental variable were inserted simultaneously into a multiple-regression model. In another study (Cruz et al. 2013), the authors conducted analyses to separate these unidirectional and bidirectional effects, examining whether parents' use of higher-level FLT led to increases in child language, and simultaneously, whether children's expressive or receptive language skills led to increases in parents' use of higher-level facilitative language strategies. They found a bidirectional association between higher-level parental FLT and expressive language only within the first year of cochlear implantation. Parents of recently implanted children with more spoken language before implantation may have been reinforced in interactions with their child to use higher-level strategies. In contrast, for receptive language, there was only a unidirectional effect between the number of different word types used by parents and receptive language development. In summary, despite possible bidirectional influences of parental language and child language, the role of parents to promote linguistic interactions is supported by the data. Children learn through interactions with their parents. They also use their language skills to elicit and expand their interactions.

The results for children with CIs and their families are in line with what is known about predictors of language development in children with normal hearing. Many studies have substantiated empirical evidence for a strong connection between early rich language exposure and developmental outcomes (Huttenlocher et al. 1991; Hart & Risley 1992, 1995; Hoff 2003; Landry et

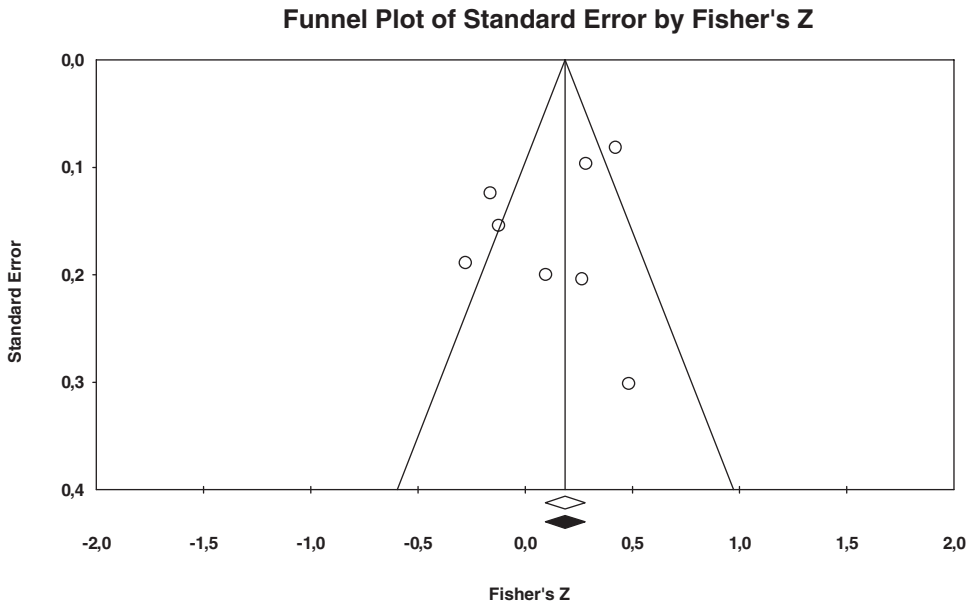

Fig. 7. Funnel plot of parental socioeconomic status.

al. 2006) in typical development that remained strong even after controlling for parental SES (Rowe 2012; Weisleder & Fernald 2013). An outstanding recent study (Gilkerson et al. 2018) demonstrated that the amount of turn-taking interactions with children with normal hearing 18 to 24 months old measured by use of Language Environment Analysis software accounted for 32% of the variance of verbal comprehension even about 10 years later. The prediction remained strong after adjustment for parental SES (R^2 = 0.027) confirming the specific impact of parental talk and interaction on child language. Noteworthy, the prediction of child language by parent–child conversational turns was much stronger than by the number of adult child-centered words (quantity of parental language input). Our findings demonstrating the role of the frequency of exposure to books for child oral language development are in line with research on typical development, which shows an enhancement of language development by the informal literacy environment at home (National Early Literacy Panel (US) 2008; Sénéchal & LeFevre 2014).

Due to incomplete data, the specific influence of parental language in addition to family SES could not be analyzed as part of our meta-analysis. However, individual studies demonstrate the specific character of parental language as shown for typical development. In a model to predict child language including SES, initial child language and higher parental facilitative language strategies (Cruz et al. 2013), SES did not significantly affect changes in child language over time.

Family involvement was shown to correlate moderately high with child language development, explaining 14% of variance and almost attaining significance. However, heterogeneity between the studies' effect sizes was high. Heterogeneity was assumed to be a consequence of a missing unified construct and the use of nonvalidated instruments for measuring family involvement. Furthermore, the lack of data did not permit investigation of suspected moderator variables such as parenting style, which was included in some of the measures of family involvement. Therefore, the results on the role of family involvement must be interpreted with caution.

As hypothesized, the more distal variable of family SES, mainly operationalized by parental (mostly maternal) education, was shown to have weaker effects on child language development (R^2 = 0.054) compared with specific parental behaviors related to children's language exposure, such as parental linguistic input or involvement in intervention. The seminal study of Hart and Risley (1992, 1995) demonstrated higher correlations between parental SES and child language in typical development. Lower correlations in populations of children with deafness or profound hearing loss might be due to limited variability of SES in the study sample and/or a consequence of a leveling effect of early-intervention programs supporting parents in their use of facilitative language strategies. As described earlier (compare Cruz et al. 2013), in studies including both SES and parental language input, SES played a minor or even statistically insignificant role for their child's language trajectories, in accordance with the results obtained from the meta-analysis.

Implications for Intervention

The results speak strongly to the importance of high-quality parental child-centered language following cochlear implantation. Early-intervention programs should carefully respect the context of language learning that takes place in parent–child interactions, with responsive parents stimulating the child's active participation in conversational exchanges. Children profit from opportunities for language-rich interactions. Early-intervention programs need to be truly family-centered, supporting parents in the frequent use of FLT in everyday situations. Irrespective of the economic status or the educational level of families, all of them could be a positive influence in their children's language development.

Implications for Further Research

Studies on outcomes in children with CIs including family environment as a critical factor for language development are still scarce. Based on the quality assessment of the 27 studies, implications for further research were identified. The use of standardized instruments to measure language outcome variables can be regarded as a strength in the field. Furthermore, almost all studies give clear descriptions of the measures used for dependent and IV, which permit replication. But there is still a great need for longitudinal multicenter studies that follow large study populations over a longer period of time. The merging of databases as well as open access databases will be of great importance to increase reliability of results with substantial sample sizes.

Many studies failed to mention the representativeness of their study population. In most cases, samples were not representative: they included a selection of families with higher SES, monolingual and majority culture background and children without additional disabilities. To draw conclusions for the total population of children with CIs, it would be important to include children from the whole variety of SES backgrounds, as well as multilingual families and children with special needs.

Regarding the family variables, there is a demand for valid standardized measures for constructs such as family involvement. Also, future studies need to implement control techniques such as measuring language outcomes and family variables independently.

Regarding the presentation of the results of the individual studies, almost all of them publish exclusively multiple-regression analyses, which could not be used for the calculation of meta-analyses. In line with current open science recommendations, there is still the need to provide access to the raw data or at least to full correlational data of the main variables and confounders.

Limitations

Precisely because the availability of correlational data was limited, the number of possible meta-analyses and the respective number of included studies was reduced. Some additional variables that could be of interest were beyond the scope of the review, but could be of future interest, such as the level of oral and signed family multilingualism, family screen-viewing time, and written and signed language outcomes.

ACKNOWLEDGMENTS

We thank all authors who provided additional data for their studies necessary for inclusion in the meta-analyses.

This research was partly funded by MED-EL Elektromedizinische Geräte GmbH Innsbruck Austria and, in addition, by the Research Projects 1150096 and 1190945. FONDECYT. CONYCIT. Chile, COST IS1406, US-1263096, Junta de Andalucía, and PSI-2015-65656-P of the Spanish Ministry of Economy and Competitiveness. The research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest. Product brands were not included in this review.

D.H., M.D., D.S., and J.F. decided together on all variables important for the data extraction. D.H., M.D., and J.F. performed the literature search and data extraction. D.H. and M.D. performed the quality assessment of all studies included. S.S.-C. and S.C.-M. conducted statistical analysis and critical revision. All authors discussed the results and implications and were involved in writing the manuscript at all stages.

Systematic review registration number PROSPERO 2017 CRD42017060568.

The authors have no conflicts of interest to disclose.

Address for correspondence: Daniel Holzinger, Konventhospital Barmherzige Brüder, Seilerstätte 2, 4020 Linz, Austria. E-mail: daniel.holzinger@bblinz.at

Received February 27, 2019; accepted December 28, 2019.

REFERENCES

American Psychological Association, Task Force on Socioeconomic Status. (2007). *Report of the APA Task Force on Socioeconomic Status*. Washington, DC: American Psychological Association.

Boons, T., Brokx, J. P., Dhooge, I., et al. (2012). Predictors of spoken language development following pediatric cochlear implantation. *Ear Hear, 33*, 617–639.

Boons, T., De Raeve, L., Langereis, M., et al. (2013). Expressive vocabulary, morphology, syntax and narrative skills in profoundly deaf children after early cochlear implantation. *Res Dev Disabil, 34*, 2008–2022.

Borenstein, M., Hedges, L. V., Higgins, J. P. T., et al. (2009). *Introduction to Meta-Analysis*. Chichester, UK: John Wiley & Sons.

Borenstein, M., Hedges, L. V., Higgins, J. P. T., et al. (2013). *Comprehensive Meta Analysis version 3.0.* Retrieved April 4, 2018 from https://www.meta-analysis.com/downloads/Meta-Analysis%20Manual%20V3.pdf.

Calderon, R. (2000). Parental involvement in deaf children's education programs as a predictor of child's language, early reading, and social-emotional development. *Journal of Deaf Studies and Deaf Education, 5*, 141–155.

Ceh, K. M., Bervinchak, D. M., Francis, H. W. (2013). Early literacy gains in children with cochlear implants. *Otol Neurotol, 34*, 416–421.

Chacón-Moscoso, S., Sanduvete-Chaves, S., Sánchez-Martín, M. (2016). The development of a checklist to enhance methodological quality in intervention programs. *Front Psychol, 7*, 1811.

Cohen, J. (1988). *Statistical Power Analysis for the Behavioral Sciences (2nd ed.)*. Hillsdale, NJ: Lawrence Earlbaum Associates.

Cruz, I., Quittner, A. L., Marker, C., et al.; CDaCI Investigative Team. (2013). Identification of effective strategies to promote language in deaf children with cochlear implants. *Child Dev, 84*, 543–559.

Cupples, L., Ching, T., Button, L., et al. (2016). Language and speech outcomes of children with hearing loss and additional disabilities: Identifying the variables that influence performance at five years of age. *Int J Audiol*, 93–104.

Dale, P. S., Tosto, M. G., Hayiou-Thomas, M. E., et al. (2015). Why does parental language input style predict child language development? A twin study of gene-environment correlation. *J Commun Disord, 57*, 106–117.

DesJardin, J. L., Ambrose, S. E., Eisenberg, L. S. (2009). Literacy skills in children with cochlear implants: The importance of early oral language and joint storybook reading. *J Deaf Stud Deaf Educ, 14*, 22–43.

Garcia-Sierra, A., Ramírez-Esparza, N., Kuhl, P. K. (2016). Relationships between quantity of language input and brain responses in bilingual and monolingual infants. *Int J Psychophysiol, 110*, 1–17.

Geers, A. E., & Sedey, A. L. (2011). Language and verbal reasoning skills in adolescents with 10 or more years of cochlear implant experience. *Ear Hear, 32*(1 Suppl), 39S–48S.

Geers, A., Nicholas, J., Sedey, A. (2003). Language skills of children with early cochlear implantation. *Ear Hear*, *24*, 46–58.

Geers, A. E., Nicholas, J. G., Moog, J. S. (2007). Estimating the influence of cochlear implantation on language development in children. *Audiol Med, 5*, 262–273.

Geers, A. E., Moog, J. S., Biedenstein, J., et al. (2009). Spoken language scores of children using cochlear implants compared to hearing age-mates at school entry. *J Deaf Stud Deaf Educ, 14*, 371–385.

Geers, A. E., Strube, M. J., Tobey, E. A., et al. (2011). Epilogue: Factors contributing to long-term outcomes of cochlear implantation in early childhood. *Ear Hear, 32*(1 Suppl), 84S–92S.

Geers, A., Mitchell, C., Warner-Czyz, A., et al. (2017). Early sign language exposure and cochlear implantation benefits. *Pediatrics, 140*, e20163489.

Gilkerson, J., Richards, J. A., Warren, S. F., et al. (2017). Mapping the early language environment using all-day recordings and automated analysis. *Am J Speech Lang Pathol, 26*, 248–265.

Gilkerson, J., Richards, J., Warren, S., et al. (2018). Language experience in the second year of life and language outcomes in late childhood. *Pediatrics*, *142*, e20174276.

Guerzoni, L., Murri, A., Fabrizi, E., et al. (2016). Social conversational skills development in early implanted children. *Laryngoscope, 126*, 2098–2105.

Hart, B., & Risley, T. (1992). American parenting of language-learning children: Persisting differences in family-child interactions observed in natural home environments. *Dev Psychol*, *28*, 1096–1105.

Hart, B., & Risley, T. (1995). *Meaningful Differences in the Everyday Experience of Young American Children*. Paul H. Brookes Publishing.

Hirsh-Pasek, K., Adamson, L. B., Bakeman, R., et al. (2015). The contribution of early communication quality to low-income children's language success. *Psychol Sci, 26*, 1071–1083.

Hoff, E. (2003). The specificity of environmental influence: Socioeconomic status affects early vocabulary development via maternal speech. *Child Dev, 74*, 1368–1378.

Hoff, E. (2006). How social contexts support and shape language development. *Dev Rev, 26*, 55–88.

Hoff, E., & Naigles, L. (2002). How children use input to acquire a lexicon. *Child Dev, 73*, 418–433.

Holt, R. F., & Svirsky, M. A. (2008). An exploratory look at pediatric cochlear implantation: Is earliest always best? *Ear Hear, 29*, 492–511.

Holt, R. F., Beer, J., Kronenberger, W. G., et al. (2013). Developmental effects of family environment on outcomes in pediatric cochlear implant recipients. *Otol Neurotol, 34*, 388–395.

Holzinger, D., Fellinger, J., Beitel, C. (2011). Early onset of family centred intervention predicts language outcomes in children with hearing loss. *Int J Pediatr Otorhinolaryngol, 75*, 256–260.

Holzinger, D., Fellinger, J., Dall, M., et al. (2017). The influence of family environment on language development in children with cochlear implants: a systematic review. *PROSPERO 2017 CRD42017060568.* Retrieved March 26, 2018 from http://www.crd.york.ac.uk/PROSPERO/display_record.php?ID=CRD42017060568.

Huber, M., & Kipman, U. (2012). Cognitive skills and academic achievement of deaf children with cochlear implants. *Otolaryngol Head Neck Surg, 147*, 763–772.

Huttenlocher, J., Haight, W., Bryk, A., et al. (1991). Early vocabulary growth: Relation to language input and gender. *Dev Psychol*, *27*, 236–248.

Ketelaar, L., Wiefferink, C. H., Frijns, J. H. M., et al. (2017). Children with cochlear implants and their parents: Relations between parenting style and children's social-emotional functioning. *Ear Hear, 38*, 321–331.

Landry, S. H., Swank, P. R., Smith, K. E., et al. (2006). Enhancing early literacy skills for preschool children: Bringing a professional development model to scale. *J Learn Disabil, 39*, 306–324.

Lund, E. (2016). Vocabulary knowledge of children with cochlear implants: A meta-analysis. *J Deaf Stud Deaf Educ, 21*, 107–121.

Moher, D., Liberati, A., Tetzlaff, J., et al.; PRISMA Group. (2009). Preferred reporting items for systematic reviews and meta-analyses: The PRISMA statement. *PLoS Med, 6*, e1000097.

Moreno-Torres, I., Madrid-Cánovas, S., Blanco-Montañez, G. (2016). Sensitive periods and language in cochlear implant users. *J Child Lang, 43*, 479–504.

National Early Literacy Panel (US). (2008). *Developing Early Literacy: Report of the National Early Literacy Panel: a Scientific Synthesis of Early Literacy Development and Implications of Intervention*. National Institute for Literacy.

Niparko, J. K., Tobey, E. A., Thal, D. J., et al.; CDaCI Investigative Team. (2010). Spoken language development in children following cochlear implantation. *JAMA, 303*, 1498–1506.

Pisoni, D. B., Kronenberger, W. G., Harris, M. S., et al. (2017). Three challenges for future research on cochlear implants. *World J Otorhinolaryngol Head Neck Surg, 3*, 240–254.

Quittner, A. L., Cruz, I., Barker, D. H., et al.; Childhood Development after Cochlear Implantation Investigative Team. (2013). Effects of maternal sensitivity and cognitive and linguistic stimulation on cochlear implant users' language development over four years. *J Pediatr, 162*, 343–8.e3.

Roberts, M. Y., & Kaiser, A. P. (2011). The effectiveness of parent-implemented language interventions: A meta-analysis. *Am J Speech Lang Pathol, 20*, 180–199.

Romeo, R. R., Segaran, J., Leonard, J. A., et al. (2018). Language exposure relates to structural neural connectivity in childhood. *J Neurosci, 38*, 7870–7877.

Rowe, M. L. (2012). A longitudinal investigation of the role of quantity and quality of child-directed speech in vocabulary development. *Child Dev, 83*, 1762–1774.

Rowe, M. L., Pan, B. A., Ayoub, C. (2005). Predictors of variation in maternal talk to children: A longitudinal study of low income families. *Parent Sci Pract*, *5*, 285–310.

Rüter, M. (2011). Influence of expansions on grammar acquisition in children with cochlear implants. [Einfluss von Expansionen auf den Grammatikerwerb von Kindern mit Cochlearimplantat]. *HNO*, *59*, 360–365.

Sarant, J., & Garrard, P. (2014). Parenting stress in parents of children with cochlear implants: Relationships among parent stress, child language, and unilateral versus bilateral implants. *J Deaf Stud Deaf Educ, 19*, 85–106.

Sarant, J., Harris, D., Bennet, L., et al. (2014). Bilateral versus unilateral cochlear implants in children: A study of spoken language outcomes. *Ear Hear, 35*, 396–409.

Schardt, C., Adams, M. B., Owens, T., et al. (2007). Utilization of the PICO framework to improve searching PubMed for clinical questions. *BMC Med Inform Decis Mak, 7*, 16.

Sénéchal, M., & LeFevre, J. A. (2014). Continuity and change in the home literacy environment as predictors of growth in vocabulary and reading. *Child Dev, 85*, 1552–1568.

Shadish, W. R. (2014). Analysis and meta-analysis of single-case designs: An introduction. *J Sch Psychol, 52*, 109–122.

Stroup, D. F., Berlin, J. A., Morton, S. C., et al. (2000). Meta-analysis of observational studies in epidemiology: A proposal for reporting. Meta-analysis Of Observational Studies in Epidemiology (MOOSE) group. *JAMA, 283*, 2008–2012.

Szagun, G. (1997). A longitudinal study of the acquisition of language by two German-speaking children with cochlear implants and of their mothers' speech. *Int J Pediatr Otorhinolaryngol, 42*, 55–71.

Szagun, G., & Schramm, S. A. (2016). Sources of variability in language development of children with cochlear implants: Age at implantation, parental language, and early features of children's language construction. *J Child Lang, 43*, 505–536.

Szagun, G., & Stumper, B. (2012). Age or experience? The influence of age at implantation and social and linguistic environment on language development in children with cochlear implants. *J Speech Lang Hear Res, 55*, 1640–1654.

Weisleder, A., & Fernald, A. (2013). Talking to children matters: Early language experience strengthens processing and builds vocabulary. *Psychol Sci, 24*, 2143–2152.

Whitehurst, G. (1978). The contributions of social learning to language acquisition. *Contemp Educ Psychol, 3*: 2–10.

Wu, C. M., Chen, Y. A., Chan, K. C., et al. (2011). Long-term language levels and reading skills in Mandarin-speaking prelingually deaf children with cochlear implants. *Audiol Neurootol, 16*, 359–380.

Yanbay, E., Hickson, L., Scarinci, N., et al. (2014). Language outcomes for children with cochlear implants enrolled in different communication programs. *Cochlear Implants Int, 15*, 121–135.

Yoshinaga-Itano, C., Sedey, A. L., Wiggin, M., et al. (2018). Language outcomes improved through early hearing detection and earlier cochlear implantation. *Otol Neurotol, 39*, 1256–1263.