Effects of age at cochlear implantation on vocabulary and grammar: A review of the evidence

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Abstract

Purpose: The increasing prevalence of pediatric cochlear implantation over the past 25 years has left little doubt that resulting improvements in hearing offer significant benefits to language development for many deaf children. Further, given the documented importance of access to language from birth, there has been strong support for providing congenitally deaf children with implants as early as possible. Earliest implantation, in many ways, has become the "gold standard" in pediatric cochlear implantation, on the assumption that it is the key to language development similar to that of hearing children. Empirical evidence to support this assumption, however, appears more equivocal than generally is believed. This article reviews recent research aimed at assessing the impact of age of implantation on vocabulary and grammatical development among young implant users.

Method: Articles published between 2003 and 2018 that included age of implantation as a variable of interest and in which it was subjected to statistical analysis were considered. Effect sizes were calculated whenever possible; we conducted a multivariate meta-analysis to compare outcomes in different language domains.

Results: Taken together, findings from 49 studies suggest that age of implantation is just one of a host of variables that influence vocabulary and grammatical development, its impact varying with several factors including whether age at implantation is treated as a dichotomous or continuous variable. Results from a meta-analysis showed significant differences across language domains.

Conclusion: The pattern of results obtained indicates the importance of considering various child, family, and environmental characteristics in future research aimed at determining how early

"early implantation" needs to be and the extent to which age of implantation, duration of implant use, and other factors influence language and language-related outcomes.

Effects of age at cochlear implantation on vocabulary and grammar: A review of the evidence

Research over the past 25 years in the cochlear implant (CI) field has demonstrated clear benefits of the constant lowering of age of implantation in congenitally deaf children in several domains. CI teams generally advise parents of children diagnosed with severe to profound hearing loss to proceed with the procedure as early as possible. The rationale for "the earlier the better" argument is largely based on the *critical period hypothesis* that suggests the existence of an early, optimal timing for language learning (Lenneberg, 1967), but it also emerged from results of physiological studies and extrapolation of data obtained from children with hearing loss who use hearing aids (Bruijnzeel, Ziylan, Slegeman, Topsakal, & Grolman, 2016). Others have discussed the idea of a "heightened sensitivity for language learning" (Szagun & Schramm, 2015, p.3), based on studies of second language learning and sign language acquisition (e.g., Mayberry, 2009; Pichler & Koulidobrova, 2015). Applied to the context of a profound, congenital hearing loss, the existence of a critical or sensitive period would entail that if cochlear implantation does not take place before a certain age, spoken language acquisition might be seriously compromised (Campbell, MacSweeney, & Woll, 2014; Connor, Craig, Raudenbush, Heavner, & Zwolan, 2006).

Over a period of nearly three decades of pediatric CI research, the critical age of implantation "not to go over" went from 5 (e.g., Brackett & Zara, 1998; El-Hakim et al., 2001; Robbins, Svirsky, & Kirk, 1997) to 3 (e.g., Miyamoto, Svirsky, & Robbins, 1997; Kirk et al., 2002; Miyamoto, Kirk, Svirsky, & Seghal, 1999), to 2 (e.g., Boons et al., 2012; Holt & Svirsky, 2008; Kirk et al., 2000; Manrique, Cervera-Paz, Huarte, & Molina, 2004; Svirsky, Teoh, & Neuburger, 2004). More recently, some researchers have suggested that cochlear implantation at age 1 year or below enables children with significant, congenital hearing loss to develop their language similarly to hearing peers (Cuda, Murri, Guerzoni, Fabrizi, & Mariani, 2014; Dettman et al., 2016). Others advocate for cochlear implantation as soon as a profound hearing loss is confirmed (Colletti, Mandalà, Zoccante, Shannon, & Colletti, 2011).

Past research has repeatedly demonstrated that age at implantation has a considerable influence on the development of basic auditory-alone speech recognition skills (Govaerts et al., 2002; Harrison, Gordon, & Mount, 2005; Hassanzadeh, Farhadi, Daneshi, & Emamdjomeh, 2002; Nikolopoulos, O'Donoghue, & Archbold, 1999; Robbins, Green, & Waltzman, 2004; Tajudeen, Waltzman, Jethanamest, & Svirsky, 2010). "Children with CIs have demonstrated progress in speech identification within a single year of implantation and approached testing levels seen in normal hearing controls" (Markman et al., 2011, p. 392). Regarding language abilities, results of several studies have led their authors to conclude that there is a clear advantage in a younger age at implantation, but other studies failed to demonstrate such an advantage. Perhaps the most consistent observation is that "variance in observed results is notoriously high" (Niparko et al., 2010, p. 1498). Possible explanations for such variability are related to the various ranges of age at implantation found in studies and to what is considered "early implantation" as well as to a variety of child and family factors.

Whereas many personal, family and environment, and timing factors (i.e., duration of use as well as age at implantation) have been mentioned as potential sources of influence, research to date has not resolved the issue of which factors can best predict linguistic and cognitive outcomes after cochlear implantation (Black, Hickson, Black, & Perry, 2011; Marschark, Duchesne, & Pisoni, in press). Nevertheless, age at implantation is generally viewed as one of the most accurate predictors of language development among pediatric CI users. A close examination of research results from the past 15 years, however, reveals that the evidence regarding the influence of that variable on language outcomes might not be as consistent or conclusive as we think it is (Szagun & Stumper, 2012). For example, Tomblin, Barker, Spencer, Zhang, and Gantz (2005), found that age at implantation accounted for 14.6% of the variance in expressive language growth rate in 29 children who received a CI between 10 and 40 months of age. In contrast, in a study involving 153 children who received CIs before the age of 5, Geers, Moog, Biedenstein, Brenner, and Hayes (2009) found age at implantation accounted for only 2.5% of independent variance on receptive and expressive language scores.

Such findings suggest that it is likely that age at implantation is only one of many sources of influence on language development in this population. Moreover, some aspects of pediatric cochlear implantation are in constant evolution (e.g., mapping, surgical techniques). Consequently, the literature on age at implantation does not seem entirely consistent due to a variety of factors that become evident with a thorough examination of studies. Thus, the purpose of the present study was to investigate the extent to which a relationship between age at implantation and language development is supported by existing literature. A thorough and objective analysis of the literature on the influence of age at implantation on spoken language development will benefit health-care professionals and early interventionists involved with children with CIs and their families and can influence clinical practice. With that in mind, we undertook a review of peer-reviewed research from the past 15 years in order to examine the extent of effects of age at implantation on spoken vocabulary and grammar. Beyond language acquisition per se, these language components play an essential role in academic achievement and social participation (Tomblin, Spencer, Flock, Tyler, & Gantz, 1999).

Method

For the purposes of this review, we entered various combinations of relevant keywords into CINAHL, Pubmed, Embase, and Linguistics and Language Behavior Abstracts (LLBA) to identify articles published between 1998 and 2018, either in English or French. We used variations on the terms "cochlear implant","child/children", "age at/of implant", "vocabulary", "grammar/syntax", "communication skills/language ability", "oral/sign language". We also hand-searched tables of contents from relevant journals (e.g., *Journal of Deaf Studies and Deaf Education*, *International Journal of Pediatric Otorhinolaryngology*, *Ear & Hearing*, *Cochlear Implants International*). The result was the identification of 250 studies at this first search stage. We then applied the following inclusion criteria: 1) studies that explicitly mentioned examining the effects of age at implantation on receptive and /or expressive vocabulary and grammar; 2) studies that involved groups of children with CIs (we excluded single case studies and studies that included but did not distinguish both children with hearing aids and CIs); 3) studies in which statistical analyses were conducted to support the existence (or non-existence) of associations between age at implantation and language achievement, and 4) studies that were published in the last 15 years (i.e., from 2003 to 2018). This time frame allowed to maximize the number of articles including a majority of children implanted before age 3 and to minimize the potential effects related to the use of older implant technologies or mapping techniques. At this stage, we examined 52 relevant empirical studies and one meta-analysis that met our inclusion criteria. The review of the reference lists of the included articles did not result in additional studies that met inclusion criteria.

Although our approach was systematic, we did not establish levels of evidence for the included studies and did not appraise the overall quality of the studies. This choice was made mainly because studies examining language development in children with CIs traditionally yield relatively low levels of evidence according to biomedical research standards (e.g., Phillips et al., 2001). In a systematic review on the potential additional benefit on auditory, speech, and language development of cochlear implantation within the first year of life compared to later implantation (i.e., after the age of 12 months), Bruijnzeel et al. (2016) concluded that current research "lacks level 1 evidence studies and consists mainly of cohort studies with a moderate to high risk of bias" (p. 113). Vlastarakos et al. (2010) also deplored the quality of the studies included in their systematic review.

To ensure that overall quality and validity of the studies were adequate, this review was limited to peer-reviewed articles. We also included one meta-analysis. At this final stage, five studies were excluded: three reported intermediate results, one did not specify which aspect of language was assessed, and one was a systematic review that did not include statistical analyses. Our aim was to obtain a broad picture of the current evidence regarding the effect of age at cochlear implantation on spoken language abilities so as to guide future research. Although some of the studies included in the present review reported outcomes related to other domains (e.g., speech intelligibility, phonological processing), we only considered outcomes related to vocabulary and grammar skills. The primary information from the 49 reviewed studies is summarized in three supplementary tables, including the calculation of effect sizes wherever possible, using the "Practical meta-analysis effect size calculator" (Lipsey & Wilson (2001; https://cebcp.org). For the purposes of that tool, we entered the younger implanted group as the "treatment" group and the older group as the "control" group.

Finally, although many of the articles reviewed were missing sufficient information to calculate effect sizes (see Supplementary tables) we conducted a meta-analysis on 32 effect sizes (based on either means, standard deviations, and sample sizes, or on correlations). When more than two age groups were compared, we selected only the younger groups (i.e. the earliest comparisons) and we focused on language outcomes measured at the time of school entry (i.e. 4-5 years of age, depending on the studies). We used R software (Metafor and club Sandwich packages). Effect sizes are expressed as Glass's *d* values.

Results

Findings from the reviewed studies related to age at implantation are presented in three different sections, according to the way it was treated as a variable. In the first section, we review studies in which children were separated into several (i.e., more than two) age at implantation categories. The second section includes studies in which the age of implantation variable was dichotomized. Finally, in the third section, we review studies that treated age at implantation as a continuous variable. This trichotomization was not planned a priori, but resulted from our observation that the way in which age at implantation was treated as a variable for statistical purposes appeared to influence results across studies (see, e.g., Geers & Nicholas, 2013). In the supplementary material, studies in which age at implantation was treated as a discrete variable were regrouped in a single table (See Supplementary Table 1).

Sample Stratification According to Age at Implantation (n > 2 groups)

Eleven studies examined differences among several groups of children. These studies generally included relatively large numbers of participants (n = 73 to n = 403) distributed among 3, 4, or 5 groups, and used a variety of outcome measures (e.g., rates of growth, language quotients) as well as different analyses and statistical procedures (e.g., developmental trajectory analyses, logistic regression).

Connor, Craig, Raudenbush, Heavner, and Zwolan (2006) investigated receptive vocabulary growth curves and rates of growth over time in a group of children (n = 100) who received a CI between 1 and 10 years old (mean = 61 months)[1]. Four subgroups were formed according to age at implantation: from 1 to 2.5 years of age (n = 21); from 2.6 to 3.5 years (n = 15); from 3.6 to 7 years (n = 20); and from 7.1 to 10 years (n = 44). Mean duration of CI use was 4 years. Results showed that children in the youngest subgroup had greater rates of vocabulary growth according to Peabody Picture Vocabulary Test (PPVT: Dunn & Dunn, 1997) raw scores than children in all the other groups for the first 3 years after implantation. Rates of receptive vocabulary growth for the four subgroups were similar after 3 to 4 years of implant use (although they still differed when compared by chronological age).

Artières, Vieu, Mondain, Uziel, and Venail (2009) also formed four subgroups of children in their sample according to age at implantation: below 2 years of age (group 1), between 2 and 3 years (group 2), between 3 and 4 years (group 3), and between 4 and 5 years (group 4), totalling 74 children who received CIs before the age of 5. Children were tested annually on a set of speech and language measures from age 4 up to 8 years old. Significant differences between adjacent groups at each data collection point were few for the earlier-implanted groups. For receptive vocabulary, children implanted below age 2 (group 1) outperformed those implanted between the ages of 2 and 3 (group 2) only at 4 and 5 years of age, and for expressive language, only at 4 years of age. The calculated effect size (see Figure 1) of the difference between the two younger ages at CI groups was .7 for both receptive vocabulary and expressive language.

Between children implanted from age of 2 to 3 (group 2) and those implanted from age 3 to 4 (group 3), significant differences favouring earlier age at CI were only noted for expressive language at 4 and 5 years of age. Differences between the two oldest groups (groups 3 and 4) were more consistent over time. Children in group 3 outperformed those in group 4 at 5, 6, and 8 years of age for receptive vocabulary, and at 5, 6, and 7 years of age for expressive language. Glass's *d* values of differences between older groups went from .8 to 1 (See supplementary Table 1). Further results from a logistic regression model showed that the PPVT "equivalent lexical scores" they calculated were significantly associated with age at implantation, as well as with duration of use and preoperative hearing thresholds. Expressive language, assessed on a 5-point scale, was only associated with age at test, duration of CI use, and preoperative hearing thresholds.

Svirsky, Teoh, and Neuburger (2004) compared expressive language outcomes of three subgroups of children with CIs: 12 children had received a CI between 16 and 24 months of age, 34 children between 25 and 36 months of age, and 29 children between 37 and 48 months of age (total n = 75). Using a developmental trajectory analysis (DTA), Svirsky et al. found an expressive language advantage, assessed either with the Reynell Developmental Language Scales (RDLS: Reynell & Gruber, 1990) or the MacArthur-Bates Communication Development Inventory (MBCDI: Fenson et al., 1993) for children who received implants between 16 and 24 months of age.

Refining the DTA technique in a subsequent study, Holt and Svirsky (2008) compared four subgroups of a total of 96 children who had received a CI before the age of 4. Children in group 1 (n = 6) received their implant between 6 and 12 months of age, group 2 (n = 32) between 13 and 24 months, group 3 (n = 37) between 25 and 36 months, and group 4 (n = 21) between 37 and 48 months of age. Duration of implant use varied among groups, depending on age at implantation. Receptive language and expressive language (both assessed either with RDLS or MBCDI) were the language outcomes variables. DTA analyses aimed to determine which group of children demonstrated better outcomes throughout the entire follow-up period. Results showed that the expressive language performance of children implanted between 6 and 12 months of age (group 1) was not significantly better of that of those implanted between 13 and 24 months of age (group 2; although other group comparisons showed statistically significant differences favouring earlier ages at CI) and no additional variance was accounted for by any of the covariates. On the basis of group comparisons, the authors concluded that there is no clear added benefit on language development to implanting before the age of 12 months. Finally, multiple linear regression analyses for receptive language scores revealed significant differences between all groups, presumably due to age at implantation. Although the impact of age of implantation cannot be distinguished from that of duration of use, family income accounted for significant additional variance in the comparisons between groups 1 and 2 and groups 1 and 3.

Miyamoto, Hay-McCutcheon, Iler Kirk, Houston, and Bergeson-Dana (2008) investigated the issue of a possible language advantage for cochlear implantation under the age of 12 months. A group of 91 children was divided in three subgroups according to age at implantation: before the age of 12 months (n = 8); between 12 and 23 months (n = 38); and between 24 and 36 months (n = 45). Miyamoto et al. administered two language tests: the Preschool Language Scale (PLS: Zimmerman, Steiner, & Pond, 1992) was administered to children with 6 months to 1 year of CI experience (n = 13), and the RDLS was administered to children after 2-3 years of CI experience. Although the authors concluded that the language skills of earlier implanted children (i.e., before the age of two) were better than those of later implanted children, differences in receptive and expressive language scores (RDLS Language Quotients) among the three groups were not statistically significant. The effect size of the difference in receptive Language Quotients between children implanted before the age of 12 months and those implanted between 12 and 23 months was -.18 (Glass's *d*). Statistical analyses were not performed with the PLS data (standard scores) because the samples were too small.

May-Mederake (2012) stated that in a sample of 28 children those who received CIs before 12 months of age performed better than those implanted later (i.e., 12-18 months) on a set of grammatical tests, including the TROG-D (i.e., the German version of the Test for Reception of Grammar: Bishop, 1998) and different versions of the 'Sprachentwicklungstest fur Kinder' (SETK: Grimm, 2001), depending on age at testing. However, the authors reported only one statistically significant difference (n = 15) between those groups, on the SETK-3-5, a test of Sentence comprehension for 3- to 5-year-olds, out of a total of 6 different subtests. Surprisingly, the authors considered a *p* value of .076 to be statistically significant. No other group differences were found.

In contrast to Holt and Svirsky (2008) and Miyamoto et al. (2008), Colletti et al. (2011) concluded that there is a receptive language advantage when cochlear implantation is performed under the age of 12 months. Expanding on their previous investigations (Colletti, 2009; Colletti, Miorelli, Guida, Colletti, & Fiorino, 2005), they reported on receptive vocabulary (PPVT) and syntax (TROG) of 73 children who received a CI between the age of 2 months and 3 years over a period of 10 years post-implantation. Colletti et al. formed three groups according to age at implantation: 2 to 11 months of age (n = 19), 12 to 23 months of age (n = 21), and 24 to 35 months of age (n = 33). At the 10-year follow-up, the number of participants in each group was 10, 16, and 21, respectively (total n = 47). For both receptive vocabulary and syntax, the group of children who had received a CI before the age of 12 months performed better than the two older-implanted groups. Differences were all statistically significant.

Reporting data from a three-year follow-up study of pediatric implant recipients, Niparko et al. (2010) presented results from 188 children. Children were stratified into 3 groups by age at implantation: younger than 18 months (n = 72), 18 to 36 months (n = 64), and 36 months to 5 years (n = 52). All participants received the RDLS (receptive and expressive scales). Results showed that children implanted before 18 months had significantly higher rates of growth for both receptive and expressive language when compared with children in the other two groups. Results from multivariate analyses showed that better residual hearing, higher ratings of parent-child interactions, and a higher socioeconomic status (SES) were associated with higher rates of receptive and expressive language improvement. Nevertheless, there was large variability in the Niparko et al. results, and their Figure 1 (p. 1502) indicates that not all children benefited from their implants.

Percy-Smith and collaborators (2013) calculated odds ratio estimates of young CI users implanted at different ages performing at age-equivalent level on three language measures: receptive vocabulary (PPVT), receptive language (RDLS), and "active vocabulary" (tested with 'Viborgmaterialet,' a Danish test). A total of 83 children were tested at a mean age of 46.3 months (range: 17 months to 6 years). Mean age at implantation was 19.6 months (range: 5 months to 4 years, 7 months). Sixty-eight children received the PPVT, 71 received the RDLS, and 49 received the 'Viborgmaterialet.' Odds ratios were calculated in reference to a younger age at implantation (5 to 11 months at the time of CI) compared to older ages (12-17 months and 18 months and above). Results of logistic regression analyses revealed that age at implantation was a significant predictor of achievement on all three language measures: children who received a CI between the age of 5 and 11 months had higher odds ratios than children implanted between 12 and 17 months and after the age 18 months. A large set of additional predictive variables were also found to be significant, depending on the language measure, including age at hearing aid fitting, educational placement, and the region of residence. Despite the observed effect of age at implantation, the majority of the participants in this study did not perform at age-equivalent levels in any of the language measures.

Nicholas and Geers (2017) divided a sample of young CI users (n = 126) into five subgroups according to age at implantation: from 6 to 11 months (group 1), 12 to 18 months (group 2), 19 to 24 months (group 3), 25 to 30 months (group 4), and 31 to 38 months (group 5). Results from groups 1 and 2 had been reported in Nicholas and Geers (2013; see below). All children were tested at 3.5 and again at 4.5 years old. Spontaneous language data were collected during a 30-minute play session and measures of lexical (number of different root words) and grammatical development (mean length of utterance (MLU) in words and number of different bound morphemes) were derived from the language samples. Effect sizes were calculated using *z*-scores based on comparisons with typically-hearing group scores. For all language measures, effect sizes were large (i.e., Cohen's *d* greater than 0.8) when group 1 was compared to group 3 and when group 2 was compared to group 4. Effect sizes were smaller when age at implantation went beyond 18 months (i.e., Cohen's *d* between 0.31 and 0.48). Regression analyses conducted with age at implantation as a continuous variable indicated that pre-CI aided pure-tone average hearing level (PTA) and age at (first) implant significantly predicted the three language scores at age 4.5.

Finally, Dettman et al. (2016) divided a group of 403 children into four age at implantation subgroups: before 12 months (n = 151), 13-18 months (n = 61), 19-24 months (n = 66), 25-42 months (n = 82), and 43-72 months (n = 43). Different subsamples of children participated in language assessments: 95 children completed the PLS at school entry (mean age at test was 5.4 years and mean duration of CI use was 3.8 years), 207 children completed the PPVT at school entry (mean age at test was 5.6 years and mean duration of use was 3.4 years), and 122 completed the Clinical Evaluation of Language Fundamentals (CELF: Semel, Wiig, & Secord, 2003), at a mean age of 8.02 years and a mean duration of CI use of 6.03 years. Regression analyses were conducted for each language test. Results for the PLS showed that both age at implantation and cognitive ability accounted for 34% of the variance in PLS scores. Results for the PPVT and CELF showed a similar pattern: age at implantation and cognitive skills both accounted for 32% of the variance in PPVT scores and 26% of the variance in CELF scores. ANOVAs with pairwise comparisons showed that the youngest group obtained significantly higher scores on all language measures (and had longer CI experience at the time of PPVT and PLS testing). The effect size of the difference in PPVT scores between children implanted before the age of 12 months and those implanted between 13 and 18 months was .9 (Glass's *d*; Figure 1).

Interim Summary

The findings from the 11 studies that divided their samples into multiple groups according to age at implantation suggest an advantage for cochlear implantation either under the age of 24 months (Svirsky et al., 2004), 18 months (Niparko et al., 2010), or 12 months (Colletti et al., 2011; Dettman et al., 2016; Percy-Smith et al., 2013). Holt and Svirsky (2008) and Miyamoto et al. (2008) did not find an added value to cochlear implantation under the age of 12 months. Moreover, Miyamoto et al. (2008) found no effect of age at implantation on either receptive or expressive language. However, that was one of two studies in this set with the narrowest age range (all participants received CIs before 36 months of age). This restriction of range might explain why the authors did not find any effects of age at implantation in their data. The 19 effect sizes that could be calculated from data provided by authors (plus the three effect sizes reported in Nicholas & Geers (2017) ranged from -1.8 to 1.2 (See Supplementary Table 1 for statistical details) which indicate enormous variability. The same variability is found in the number of subgroups and their size, the various age ranges, and the different types of scores used to report language outcomes.

Except for Colletti et al. (2011), the large majority of the above studies reported on relatively short follow-up periods, typically after 2-3 years of implant experience. Two studies mentioned that differences between groups dissipated after approximately 2 years (Artières et al., 2009) to 4 years of CI use (Connor et al., 2006). In Svirsky et al. (2004), only 25% of children in the earlier implanted group had follow-up data after age 4. In the oldest age at implantation group, apparently only 24% of children had follow-up data after the age of 6. Lack of follow-up data on most of the children makes the Svirsky et al. results difficult to interpret. Similarly, Colletti et al. (2011) lost 35% of their participants (26/73) at the 10-year follow-up, potentially representing children who did not benefit or benefited less from their implants. Finally, in studies that reported results of regression analyses, age at implantation, along with additional variables, predicted language achievement: family income (Holt & Svirsky, 2008; Niparko et al., 2010), cognitive skills (Dettman et al., 2016), and residual hearing (Nicholas & Geers, 2017; Niparko et al., 2010).

Age at Cochlear Implantation as a Dichotomized Variable

Nine studies reviewed examined differential effects of age of implantation between two subgroups of children with CIs. The number of children in these studies ranged from 15 to 160. Different cut-offs were used to separate groups according to age at implantation. Five studies examined differences between children who received CIs before and after the age of 12 months. In the remaining four studies, the cut-off was established either at 18 months, 2 years, 2.5 years, or 4 years old. The majority of the studies used *t*-tests to establish whether differences in outcomes between groups were statistically significant. Age at implantation ranged from 5 months to 14 years.

Expanding on a previous investigation (Dettman, Pinder, Briggs, Dowell, & Leigh, 2007), Leigh, Dettman, Dowell and Briggs (2013) included a larger sample (n = 120) of children who received CIs between 6 and 12 months of age (n = 35) and between 13 and 24 months of age (n =

85). Receptive language (Rossetti Infant-Toddler Language Scale, RITLS: Rossetti, 1990) and vocabulary (PPVT) were assessed at different time intervals. The PPVT was administered to 21 younger-implanted children and 40 later-implanted children. PPVT data at 3 years of CI use were analyzed. Results showed that age at implantation was not significantly associated with receptive RITLS scores. This was confirmed by our calculation of the effect size of the difference in RITLS scores mean growth: Glass's *d* value was .32. Differences between children who received their CI by 12 months of age and those who were implanted between 13 and 24 months of age were also not significant. PPVT scores showed significant negative correlations with both age at implantation and age at hearing aid fitting.

In a small-scale study, Houston and Miyamoto (2010) compared the performance of two subgroups of early implanted children on receptive vocabulary (PPVT) – used as a measure of word learning skills – at two intervals: after 2 to 2.5 years of CI use and after 3 to 4 years of use. Each subgroup included seven participants at interval 1 and respectively five and six participants at interval 2 (total n = 15). They found that at both intervals, the earlier implanted subgroup (age at implantation between 7 and 13 months) performed significantly better than the later-implanted subgroup (age at implantation between 16 and 23 months) on the receptive vocabulary measure. The calculated effect size was 1.25 at 2-2.5 years of use and 1.44 at 3-4 years of use (See Supplementary Table 1).

Expanding on findings of Niparko et al. (2010), Markman et al. (2011) reported on 116 children who received a CI between 6 months and 5 years of age. Two groups were formed, based on the presence of spoken language skills before implantation. Participants who had not developed any spoken language (n = 96) were then divided according to age at implantation: before 18 months of age (n = 34) vs. after 18 months of age (n = 62). The research team used the Comprehensive Assessment of Spoken Language (CASL), composed of 15 subtests that assess four language subdomains: lexical-semantic abilities, syntax, supralinguistics, and pragmatics (receptive and expressive). Children performed four tasks, one for each specific subdomain, after 4-5 years of CI use. Results revealed that the earlier implanted subgroup (CI before 18 months) performed significantly better than the later-implanted subgroup (CI after the age of 18 months) in each language subdomain. Multivariable-adjusted modeling analyses also showed that maternal sensitivity (i.e., warmth, positive regard, and respect for autonomy in the parent–child relationship) significantly predicted the "core composite standard score" in both groups.

Tobey et al. (2013) reported on 160 children who completed language assessments after 4, 5, and 6 years of CI use. Groups were divided according to age at implantation: before 2.5 years of age (n = 98) versus 2.5 to 4.9 years of age (n = 62). The same four core subtests described in Markman et al. (2011) were administered. Age at testing ranged from 4.8 to 11.5 years old. Results showed that trajectories of the core composite standard scores at 4, 5, and 6 years post-implantation did not significantly differ as a function of age at implantation. However, multivariable-adjusted analyses with age at implantation as a continuous variable revealed that age at implantation was significantly associated with core composite scores at each follow-up time. Core composite scores also were significantly associated with the level of language comprehension at baseline (RDLS scores), parent-child interaction, and speech recognition index.

Uziel et al. (2007) reported on a 10-year follow-up of 82 children who received a CI between the age of 1.9 and 14 years. They compared the performance of children who received their CI by age 4 and after age 4 on measures of speech perception and production, as well as receptive vocabulary. At 12 to 24 years of age, a higher proportion of individuals who had received a CI before the age of 4 scored above the 50th percentile on the PPVT compared to children who received CIs after age 4. The presence of other disabilities and educational placement in a school for the deaf were both significantly associated with scores below the 50th percentile. The calculation of odds ratios showed that an older age at implantation increased the risk of a PPVT score below the 50th percentile by a factor of 2.6 (compared to an increased risk by factors of 9.43 on speech recognition, 12.7 on speech intelligibility, and 15.15 on speech tracking). Overall, odds ratios suggested that receptive vocabulary seemed less influenced by age at implantation than by school placement and that speech perception and production abilities seemed far more influenced by age at implantation than receptive vocabulary.

Dunn et al. (2013) conducted a retrospective study involving 83 children grouped according to age at implantation: below age 2 (n = 38) and between 2 and 3.9 years of age (n = 45). Children participated in periodic language assessment using the CELF; mean duration of CI use was 7.8 years for the earlier-implanted group and 12.2 years for the later-implanted group. Raw scores from one receptive language subtest (Concepts and Directions), one expressive subtest (Formulated Sentences), and one reading subtest were used for analyses. Data were processed using a linear mixed-model, and two-sample *t*-tests subsequently were performed. CELF results included either 38 (receptive) or 39 (expressive) children: 13 from the earlier-implanted group and 25 and 26 from the later-implanted group. Results showed that at age 7, children implanted earlier had better receptive language scores than those implanted later. From 8 years old onward, the differences between groups were no longer statistically significant. Calculated effect sizes were .72 at age 7 and .59 at age 9 (Supplementary Table 1). Expressive language showed a similar trend: At 7 years of age, children who were implanted before age 2 performed significantly better. At 10 and 11 years of age, the advantage of early implantation was no longer significant. Calculated effect sizes were .92 at age 7, .63 at age 10, and .44 at age 11 (Supplementary Table 1). These findings suggest that the influence of age at implantation on language abilities seems to gradually decrease as chronological age increases, as do benefits of CIs more generally (Marschark & Knoors, 2019).

The Italian version of the MBCDI was used by Rinaldi, Baruffaldi, Burdo, and Caselli (2013). Twenty-three children were divided according to age at implantation: before 12 months of age and between 13 and 26 months of age. The number of words used, the number of sentences produced, and the percentage of complex sentences, were transformed into *z*-scores. Group comparisons (*t*-tests) revealed that differences in mean *z*-scores between the groups were not statistically significant for any of the three measures after 7 to 25 months of CI experience. The calculated effect sizes, respectively, were .5 (number of words), .23 (number of sentences) and .52 (percentage of complex sentences).

Wie (2010) also examined receptive and expressive language abilities in a group of 20 very young (bilaterally) implanted children. Age at implantation ranged from 5 to 18 months of age. Language was assessed with the Mullen Scale of Early Learning (MSEL: Mullen, 1995) and the Minnesota Child Development Inventory parent questionnaire (MCDI: Ireton & Thwing, 1974) at 8 intervals (3, 6, 9, 12, 18, 24, 36, and 48 months post-implantation). Except for two children, all participated in at least 6 follow-up evaluations. When the group was divided according to age at implantation (before and after 12 months of age), children who received their implants before 12 months of age (n = 13) had significantly higher receptive and expressive language scores than later-implanted children (n = 7). When age at implantation was treated as a continuous variable, earlier age at implantation was moderately associated with better receptive and expressive language scores (associations were stronger for expressive language), especially during the first 18 months of CI use. The strength of associations diminished over time and were no longer significant after 36 months of CI use.

Finally, in a study involving a subsample of children also tested by Nicholas and Geers (2017), Nicholas and Geers (2013) compared the two youngest age groups: children implanted from 6 to 11 months of age (n = 27) vs. 12 to 18 months of age (n = 42). All children were tested at 4.5 years of age on receptive vocabulary (PPVT) and receptive and expressive language (PLS). For all three measures, mean scores of the earlier implanted group were statistically higher (*t*-tests) than those of the later implanted group. The calculated effect sizes of the difference in test scores were respectively .72 (PPVT), .79 (receptive PLS), and .61 (expressive PLS).

When analyzed as a continuous variable, age at implantation was significantly associated with an average standard score derived from the three language measures. Regression analyses showed a significant linear effect of age at implantation on all three language measures at 4.5 years of age.

Interim Summary

Findings from the nine studies in which the age at implantation variable was dichotomized are suggestive of better language performances in groups of children who received a CI as early as 12-13 months of age (Houston & Miyamoto, 2010; Nicholas & Geers, 2013), between 18 to 24 months (Markman et al., 2011) and up to 4 years of age (Uziel et al., 2007) compared to groups that received CIs later. Two studies found neither receptive (Leigh et al., 2013) nor expressive (Rinaldi et al., 2013) differences in language skills of children implanted before 12 months of age and between 13 and 26 months of age, after up to 3 years of implant experience. In one study, differences between groups were found in only one of six language subtests (May-Mederake, 2012). Tobey et al. (2013) found that trajectories of core composite standard scores at 4, 5, and 6 years post-implantation were not significantly different between children implanted before 2.5 years of age and from 2.5 to 5 years of age. In the majority of the studies, authors explained their results in terms of the existence of a sensitive period for the auditory system, matching their chosen cut-off age. The 15 effect sizes that could be calculated from data provided by authors show two main trends. First, effect sizes for receptive vocabulary were large: except for one moderate effect size of .71 in Nicholas and Geers (2013), effect sizes ranged form 1.09 to 1.44 (See Supplementary Table 1), at 2 to 4 years of CI use. Second, the effect sizes for receptive and expressive language seem to be decreasing over time: the longer the follow-up period is, the smaller is the effect size (e.g., .44 when children were tested for expressive language at 11 years old (Dunn et al., 2014). Finally, as in studies that compared several age at implantation groups, additional factors were consistently found to be associated with language achievement including parent-child interaction variables (Markman et al., 2011; Tobey et al., 2013), pre-implantation language skills (Tobey et al., 2013), and school placement (Uziel et al., 2007).

Age at Cochlear Implantation as a Continuous Variable

In this section, we present the findings of 20 empirical studies and of one meta-analysis that explored the influence of age at implantation on vocabulary and grammatical skills. Sample sizes varied from 9 to 288 participants. Age at implantation ranged from 4 months to 15 years.

One of the first large-scale studies that focused on core language abilities in children who received CIs before the age of 5 years involved 181 children who were using either oral communication, or both speech and sign (Geers, Nicholas, & Sedey, 2003; Geers, 2004). Children were assessed with a battery of speech, language, and reading tests at a chronological age of 8-9 years. Mean age at implantation was 3 years and 5 months (Geers & Brenner, 2003). A "Total Language Score" included data from testing of receptive language using the Test for Auditory Comprehension of Language (TACL: Carrow-Woolfolk, 1985) and lexical and grammatical measures derived from a language sample (e.g., MLU in words and Index of Productive Syntax (IPSyn) elements). Regression analyses showed that age at implantation was not significantly associated with language achievement. Variables related to child and family characteristics (e.g., nonverbal IQ, gender, SES) significantly predicted language and reading outcomes (Geers et al., 2003; Geers, 2004).

Nicholas and Geers (2006, 2007, 2008) conducted a second set of studies with 76 children who were implanted between the age of 12 and 38 months and using oral communication. Geers and Nicholas (2013) reported on the same sample of children, as did Geers, Nicholas, and Moog (2007; n = 74). Children were tested at 3.5 years old (Nicholas & Geers, 2006), 4.5 years old (Geers et al., 2007; Nicholas & Geers, 2007, 2008), and 10.5 years old (Geers & Nicholas, 2013). Language tests included early expressive language measures (MBCDI, data from a 30-minute play session, and a teacher rating). Receptive and expressive vocabulary (PPVT and Expressive One-Word Picture Vocabulary Test, EOWPVT (Gardner, 2000); global language measures (PLS and CELF) were also administered. A multiple regression analysis using the early language measures as criterion variables (Nicholas & Geers, 2006) revealed that pre-implant aided hearing PTA threshold, duration of hearing aid use, post-implant PTA threshold, and duration of CI use accounted for 58% of the variance in language factor scores. Both pre-implant aided PTA threshold and duration of CI use were significant predictors. Similar analyses with data from the testing at 4.5 years of age (n = 74; Geers et al., 2007) showed that five variables, age at implantation, gender, parent education, age at hearing aid fitting, and age at test, accounted for 23% of the variance in PPVT scores. Only age at implantation was a statistically significant predictor in the regression model. Hierarchical linear modeling indicated higher grammatical scores (derived from a language sample) significantly associated with pre-implant residual hearing and age at implantation, as were the PLS-3 scores at 4.5 years of age. However, except for the number of different bound morphemes, the expected grammatical growth between 3.5 and 4.5 years old did not differ for any age at cochlear implantation (Nicholas & Geers, 2007). Linear and quadratic regression analyses (Nicholas & Geers, 2008) showed similar results with PPVT, MBCDI, and PLS scores at 4.5 years of age (i.e., age at implantation and the amount of residual hearing both predicted test scores). Finally, when the children were tested at 10.5 years of age using the CELF, PPVT, and EOWPVT, regression analyses showed that age at implantation, pre-CI aided PTA, parental education/income, and nonverbal skills together accounted for 38% of the language outcome variance (Geers & Nicholas, 2013).

In a third set of studies by Geers and colleagues, children who received a CI between the age of 11 and 59 months of age were tested at school entry (5-6 years old) by Geers et al. (2009) (n = 153) and Geers et al. (2007) (sample 2, n = 126). Characteristics of both samples are highly similar, hence presumably composed of children drawn from the original sample of 181 CI users who were 8 to 9 years old when tested between 1997 and 2000. Receptive vocabulary (PPVT) was assessed in both studies whereas expressive vocabulary (using the EOWPVT or Expressive Vocabulary Test, EVT: Williams, 1997) and receptive and expressive language (CELF) was assessed in Geers et al. (2009). Multiple regression analyses with PPVT data from children tested at 5-6 years of age (sample 2; Geers et al., 2007) showed that five variables, age at implantation, gender, parent education, age at hearing aid fitting, and age at test, accounted for 24% of the variance in PPVT scores. Age at implantation as well as age at test and parent education was statistically significant predictors. A different set of predictors was entered in regression analyses by Geers et al. (2009). For all four language measures, nonverbal IQ was the strongest predictor of outcomes, accounting for from 15% to 24% of the variance in vocabulary and language scores, followed by parent education level (from 4% to 10% of the variance). Age at implantation was a very small contributor to language outcomes, accounting for between 1.7% and 2.7% of the variance. The different influence of nonverbal IQ in both studies might be explained by the fact that Geers et al. (2009) included children with IQs of 70 and above whereas only children with IQs of 80 or higher were included in Geers et al. (2007). We were able to calculate a few effect sizes for differences in PPVT, EOWPVT, and PLS scores among the three sets of studies by Geers and her collaborators (See Supplementary Table 2). Depending on the language domain assessed and on the test used, effect sizes ranged from -.85 to .96.

Finally, in a similar study, Hayes, Geers, Treiman, and Moog (2009) administered the PPVT yearly to 65 children who received a CI before the age of 5. Mean PPVT standard scores for the whole group (mean age at implantation = 2.69) remained below 1 *SD* up to 6 years of CI use. Growth curve analyses revealed that children who received their implant at a younger age showed a faster receptive vocabulary growth rate than children who were older when they received their implant. Surprisingly, the expected vocabulary growth curves according to age at implantation showed a decline after 4 years post-implantation and that decline was more pronounced in earlier implanted children.

Taken together, the findings of this large set of influential studies by Geers and colleagues suggests that whereas age at implantation has some influence on various language outcomes, several other auditory, personal, and family factors are likely to have at least an equal influence on vocabulary and grammar development up to 10 years post-implantation. It also is worth noting that these studies have involved relatively homogenous samples that included mostly children who were enrolled in intervention programs and schools that strongly encouraged listening and spoken language as a means of instruction, who had no other disabilities, and who came from relatively advantaged socioeconomic environments (Geers & Nicholas, 2013).

In line with some questions that were raised in the studies by Geers and her collaborators, the issue of age at implantation compared to experience with language was examined in Schorr, Roth, and Fox (2008). They assessed 39 children who received a CI between 1; 3 and 8; 2 at a mean chronological age of 9 years and compared their performance on a spectrum of speech and language abilities with that of a matching group of 37 hearing children. Results showed that SES was significantly associated with both receptive and expressive vocabulary skills, with receptive morphosyntax, and with metaphonology. Age at implantation was significantly associated only with receptive vocabulary scores (17% of unique variance). Duration of implant use was a significant predictor of receptive morphosyntax scores (10% of unique variance), thus suggesting different influences depending on which language component is evaluated.

The question of age of implantation versus CI experience was the focus of a study by Szagun and Stumper (2012). Twenty-five children who received a CI between 6 and 42 months of age participated in the study. Various expressive language data based on the MBCDI and spontaneous speech samples were collected at 12, 18, 24, and 30 months after implantation. Analyses were conducted with the language scores apparently only at 30 months post-implantation. For each of the five language measures, maternal education level was significantly associated with language scores after 30 months of CI use, whereas age at implantation was not. Maternal child-directed input was also strongly associated with children's MLU. A different set of analyses was conducted in which children were grouped according to age at implantation: between 6 and 11 months of age (n = 7), between 12 and 23 months of age (n = 9), and after the age of 24 months (up to 42 months of age; n = 9). Results of two-way analyses of covariance with repeated measures showed that for both vocabulary and grammar, the age at implantation factor was not significant. No significant differences among the groups were found at any data collection point. Although the duration of follow-up was relatively short in this study, the different trajectories of language growth were influenced by language experience and linguistic environment rather than by age at implantation per se.

Szagun and Schramm (2015) further examined the relative influence of age at implantation, parental child-directed speech, and early language level on grammar development after 24 and 30 months of CI experience. Spontaneous language data from two separate samples (n = 22 and n = 26) of German children who received a CI between the age of 6 and 46 months were collected and analyzed at various time intervals. Two different regression models with both samples combined were conducted. Results showed that both parental expansions (i.e., parent utterances that expand incomplete or incorrect child utterances) and age at implantation significantly predicted MLU both at 24 and 30 months post-implantation. However, parental expansions accounted for 48% and 43% of the unique variance of MLU, at 24 and 30 months after implantation, with age at implantation adding 9% and 10% of unique variance, respectively. When early language level (i.e., MLU at 11-12 months after implantation) was added as a potential predictor, all three variables remained statistically significant predictors of MLU both at 24 and 30 months post-implantation. Early language level explained the largest part of unique variance (15% and 17% respectively), whereas age at implantation and parental expansions uniquely accounted for 9% to 12% depending on the duration of CI experience. In both regression models, age at implantation was not significantly associated with any other predictor variable. Effect sizes of the difference in MLU were .46 at 24 months of use and .5 at 30 months of use. Correlational analyses showed significant associations between a series of language measures (e.g., use of determiners and number of word types) at earlier data points (from 6.5 up to 20 months post-implantation), but correlations with age at implantation were all non-significant. These findings indicate that parental input and early linguistic competence, rather than age at implantation, most strongly influence subsequent linguistic development.

The influence of early language skills on later abilities and the role of age at implantation were examined in Hay-McCutcheon, Kirk, Henning, Gao, and Qi (2008). Thirty children participated in repeated language testing either with the RDLS (administered until children were 7 years old) or the CELF (administered to children aged from 7 up to 18 years). Mean age at implantation was 4.48 years (range: 1.4 to 7.7). Analyses using mixed-effects models showed that age at implantation was significantly associated with both RDLS receptive and expressive age equivalent scores at ages 2 to 7 years. Null results were found for CELF scores at ages 9 to 13 years old (with a few participants aged up to 17 years at the time of testing). That is, associations between CELF core percentile rank scores and age at implantation were not statistically significant, suggesting that the early impact of age at implantation diminishes over time.

Willstedt-Svensson, Löfqvist, Almqvist, and Sahlén (2004) examined receptive and expressive grammar in 15 Swedish children who received a CI between 2 and 6 years of age (mean = 3 years, 11 months). Children were aged between 5;4 and 11;5 years (mean = 7 years, 7 months) at the time of testing. Mean length of CI experience was 4 years, 1 month. Receptive grammar was assessed with the TROG and expressive grammar was assessed with the Lund Test of Grammar. Age at implantation was significantly correlated with both receptive and expressive grammar scores, whereas duration of CI use was correlated only with receptive grammar. When only *timing* variables (i.e., age at implantation, duration of CI use, and age at testing) were entered in a regression analysis, results showed that age at implantation predicted 43% of the receptive grammar and 36% of the expressive grammar variance. However, when working memory tasks (i.e., non-word repetition, non-word discrimination, and complex working memory) were added as predictor variables in a subsequent regression model, age at implantation did not account for significant variance. These findings raise challenging questions regarding the influence of underlying neurological and cognitive processes that are related to age and maturation but also to auditory deprivation (Kral, Kronenberger, Pisoni, & O'Donoghue, 2016).

More recently, Cuda et al. (2014) assessed early expressive language development in younger CI recipients. They tested 30 Italian children who received a CI between the age of 8 and 17 months with the MBCDI. Overall results when the children were 36 months old suggested that those who received a CI before the age of 12 months had slightly better early language skills than children who received a CI between 12 and 17 months of age. Results from a regression analysis showed that age at implantation and gender (females) were significantly associated with a higher number of words and a higher mean length of the three longest utterances. Sentence complexity was significantly associated with age at implantation, gender, and maternal education level.

Fagan (2015) examined the impact of age at implantation after a shorter follow-up period. Her study involved nine children implanted from 8.9 to 14.4 months of age. She found that age at implantation was significantly correlated with MBCDI scores after 12 months of CI use. Effect size was large (Glass's $d = 1.53$). However, participants' scores remained far below expected achievement according to chronological age despite the very young age at cochlear implantation.

In a large-scale retrospective study, Black, Hickson, Black, and Khan (2014) reviewed a total of 174 cases. Mean age at implantation was 44 months old (range: 4 months to 15 years old). Data on language outcomes were collected at 18-24 months post-implantation. Depending on the language measure, the number of participants varied from 38 to 89. Three different multiple regression models included a large set of potential predictors of receptive vocabulary (PPVT), receptive, and expressive language (PLS, CELF). Results showed that "family concern," the extent to which the family was able to accept and cope with their child's hearing loss (yes or no, dichotomized variable), was a significant predictor of achievement in all three language domains: Children with family concern had significantly lower scores than those without family concern. The presence of inner ear malformation was a strong predictor of expressive language scores. Finally, whereas a later age at implantation was associated with lower receptive and expressive language scores, the association did not reach significance.

Boons et al. (2012) also conducted a large-scale retrospective study. They assembled data from three language tests that were administered to children who received a CI before the age of 5 (total n = 288; depending on the language measure, *n* varied between 79 and 159). Standardized receptive (RDLS) and expressive (Schlichting Expressive Language Test, SELT: Schlichting, Van Eldik, Lutje Spelberg, & Van der Meulen, 1995) language tests scores were examined at 1, 2, and 3 years post-implantation. A linear regression analysis showed that age at implantation significantly predicted language quotients (LQ: age equivalent on the test divided by chronological age at the time of testing) over the three-year course of the study. Further *t*-tests for independent samples with two separate subgroups (implantation below age 2 and above age 2) confirmed that children implanted before the age of 2 had significantly higher LQs on all 3 tests than children who received their implant after the age of 2. A linear regression analysis with the subgroup of children implanted before the age of 2 revealed a weak to modest effect of age at implantation after 1 and 2 years of use and no effect after 3 years of use. Furthermore, additional disabilities and contralateral stimulation were consistently found to contribute to additional variance in all 3 LQs. Multilingualism (at all three data collection points) and parental involvement (at 2 and 3 years post-implantation) both were significant predictors of receptive language skills (RDLS) and expressive word development (SELT).

Boons et al. (2013) calculated LQs in a subsequent cross-sectional study with 70 Dutch children implanted before the age of 5 who were compared with a group of hearing children on four expressive language components (vocabulary, morphology, syntax, and narrative skills). Age at the time of test ranged from 5 to 13 years of age. A logistic regression model exploring the factors associated with good or poor performance on each language measure showed that age at implantation was not a significant predictor of any of the language components. The factors that best predicted vocabulary performance were, respectively, the presence of additional disabilities and multilingualism (i.e., more than one spoken language at home). The best predictors of morphology outcomes were chronological age and multilingualism (more than one spoken language at home was significantly associated with weaker syntax abilities).

Tomblin et al. (2005) examined expressive language growth rates in 29 children who received a CI between 10 and 40 months of age. Children participated in repeated language assessments with the Minnesota Child Development Inventory (MCDI) and the PLS at chronological ages ranging from 5 months to 78 months. Tests scores were converted into Expressive Language Quotients (ELQs). Both at 12- and 24-month follow-up assessments, age at implantation was significantly associated with ELQs. However, standard scores decreased over time, suggesting a decline in language abilities in relation to chronological age. Results from a HLM analysis of expressive language growth revealed that age at implantation accounted for 14.6% of the variability in expressive language growth rate.

Finally, some results and conclusions of a meta-analysis conducted by Lund (2016) are relevant here. In her study, which was limited to vocabulary development in pediatric CI users, the majority of the participants in 16 primary studies had received a CI before the age of 30 months (five of which are included in the present review). Results from meta-regression analyses showed that neither age at implantation, nor duration of use, nor age at the time of testing were associated with the magnitude of weighted effect sizes. Lund concluded that there is no evidence that early implantation allows children with a profound hearing loss to attain vocabulary levels similar to those of same-age hearing peers. It is likely that additional child-related factors contribute to vocabulary achievement for children with CIs, namely underlying learning mechanisms.

Interim Summary

Findings from several of the studies that treated age at implantation as a continuous variable show two main trends regarding the influence of age at implantation on core language skills. The global examination of the primary findings of the reviewed studies (See

Supplementary Table 2) shows that in eight studies, significant associations were found between age at CI and language scores. In five other studies, age at CI predicted language scores, along with other variables. In six studies, age at CI was not associated with language achievement. The 21 effect sizes that could be calculated from data provided by authors also illustrate a large variability, ranging from -1.8 to 1.75.

In many studies, the amount of variance uniquely explained by age at implantation was moderate, typically between 10% and 25%. Moreover, other variables were consistently found to influence language outcomes, for instance gender (Cuda et al., 2014; Geers et al., 2003), SES and maternal education (Cuda et al., 2014; Geers et al., 2003; Geers & Nicholas, 2013; Schorr et al., 2008), parental input and parental involvement (Boons et al., 2012; Szagun & Stumper, 2012; Szagun & Schramm, 2015), and residual hearing (Nicholas & Geers, 2006). Finally, whereas a number of studies found no effect of age at implantation on language outcomes (e.g., Black et al., 2014; Geers et al., 2003; Szagun & Stumper, 2012), other studies found an effect only for one or two specific language elements (e.g., Guasti et al., 2012) or only within the first few years of implant use (e.g., Boons et al., 2012; Hay-McCutcheon et al., 2008).

Some Additional Evidence

Beyond the studies described above, we briefly summarize the results of eight more studies that have reported on the possible influence of age at implantation even though it was not the main goal of the study. We did not calculate effect sizes and did not include these studies in the multivariate meta-analysis. All results were based on correlations/regressions. For example, among children who had received a CI between 8 and 28 months of age (n = 27), Duchesne, Sutton, and Bergeron (2009) found no significant correlation between receptive or expressive RDLS scores and age at implantation, duration of use, or chronological age at the time of testing.

Similarly, neither Ruffin, Kronenberger, Colson, Henning, and Pisoni (2013) nor Castellanos et al. (2014) found age at implantation to be significantly associated with either PPVT or CELF scores in children tested after longer-term follow-up periods. In a study on Dutch finite verb production, Hammer, Coene, Rooryck, Gillis, and Govaerts (2010) found a significant association between finite verb production and age at implantation at 4 and 5 years of age. In a study on lexical and grammatical achievement in Italian children with CIs (n = 33), Guasti et al. (2012) analyzed data from five different tasks related to receptive vocabulary and acquisition of grammar that were administered when children were aged between 50 to 82 months (mean: 63.9 months). A significant effect of age at implantation was found for one task (production of clitic pronouns). Geers et al. (2017) conducted a follow-up regression analysis and found that age at implantation significantly predicted spoken language, as measured by the CASL, in early and later elementary grades. Nittrouer, Sansom, Low, Rice, and Caldwell-Tarr (2014) collected data from a language sample in 21 children who received a CI at a mean age of 21 months and were tested at a mean age of 82 months. Results showed that age at implantation was significantly associated with only two of the five measures of lexical and grammatical abilities. Nittrouer, Lowenstein, and Holloman (2016), however, found no significant correlations between age at (first) CI and any of the morphosyntactic measures in 51 children tested when in second grade. In four of these studies, age at CI was not associated with language achievement; in two studies, age at CI was associated with language in some tasks but not in others; in one study, age at CI was significantly associated with expressive grammar, and in one last study, it was associated with a general measure of language.

Meta-analysis on Effect Sizes

In this final section, we report the results of a multivariate meta-analysis that was conducted on 32 effect sizes. Figure 1 displays a forest plot of effect sizes.

Figure 1. here

In figure 1, grey diamonds indicate the effect size for each dependent variable in each study. The meta-analytic effect sizes for each language domain appear under "meta-analysis effect size". The *p* value indicates whether the meta-analytic effect size is different from zero. Differences among meta-analytic effect sizes for each language domain were statistically significant (QM (df = 5) = 187.22; $p < .001$). Depending on the language domain, Glass's *d* values range from -.07 (expressive language) to .74 (receptive language). Glass's *d* for receptive vocabulary is .71, and .59 for expressive vocabulary. Meta-analytic effect sizes for expressive language and expressive grammar are small.

Discussion

The purpose of this review was to examine the extent to which age at implantation was associated with spoken language outcomes in individuals who received CIs as children, according to studies published from 2003 to 2018. We considered 49 peer-reviewed studies that included age at implantation as a variable of interest and conducted statistical analyses on that variable. One of the studies was a meta-analysis. Overall, the reviewed evidence suggests only a moderate influence of age of implantation on central components of language. It is worth noting that the majority of the studies that examined the influence of age at cochlear implantation on language development focused primarily on receptive vocabulary in children aged under age 6, but there were wide ranges in both age of implantation and age at testing. Consistent with our larger interpretation of the whole set of studies, meta-analysis effect sizes are moderate at best, depending on the language domain assessed. Meta-analytic effect sizes were larger for receptive than expressive skills. According to Hay-McCutcheon et al. (2008), expressive language skills "historically lagged behind" (p.375) the receptive domain. In their study, Hay-McCutcheon et al. suggested that the development of expressive language was slower (or more variable) compared to the development of receptive abilities. Meta-analytic effect sizes also suggest that vocabulary (both receptive and expressive) is more resilient than grammar in relation to age at implantation. This finding is confirmed in many studies on children who received CIs before age 3. Not only are discrepancies often found between domains (e.g. Geers et al., 2009; Geers & Nicholas, 2013), but also when vocabulary and grammar are both assessed, vocabulary scores are typically better that grammar scores (e.g. Caselli et al., 2012; Duchesne et al., 2009). Finally, within the vocabulary domain, effect sizes for receptive vocabulary were larger than for expressive vocabulary. This finding is in agreement with Lund (2016) who conducted a meta-analysis on vocabulary knowledge in children with cochlear implants. She also found that the weighted effect size average was larger for receptive than for expressive vocabulary. The present review expands on the findings of Lund (2016) in including grammatical skills as well as vocabulary skills. Conversely to Lund, we did not compare the performance of children with CIs with that of typically hearing groups; rather, we compared children with CIs who had different ages at the time of implantation.

Two main trends emerged from the present review. First, there is compelling evidence of short-term advantages from "earlier implantation." From as early as 3 to 12 months of CI use, up to 3 to 4 years of follow-up, age at implantation is generally associated with better language outcomes. The influence of age at implantation becomes more equivocal after 36 months of CI use. In some studies, authors concluded that there is a decreasing influence of age at implantation as chronological age increases. But, short-term follow-up periods generally represent early stages of language development. As Schorr et al. (2008, p. 208) stated: "it is possible that age at implant is most critical when examining growth of language during the most dramatic spurt stage (i.e., between 2 and 5 years of age).” The fact that very few studies have examined long-term linguistic outcomes limit our ability to draw any firm conclusions on the enduring influence of age at implantation. Geers and Nicholas (2013) found that spoken language skills were still correlated with age at implantation at 10.5 years of age, but studies by Sarchet et al. (2014) and Convertino, Borgna, Marschark and Durkin (2014) failed to find any effect of age of implantation on receptive vocabulary (PPVT) scores among deaf and hard-of-hearing college students. Because the current cohort of college-aged students (in the United States) received their CIs relatively late by current standards, and deaf and hard-of-hearing college students represent a group that might be expected to have better-than-average language skills (Dammeyer & Marschark, 2016), some caution is warranted in generalizing the results from only two studies. Nevertheless, other studies involving this population of CI users have shown them to have surprisingly diverse language skills (e.g., Spencer et al., 2018), and there do not appear to be other studies that have considered individuals who have reached this age/length of CI use. Obtaining similar data from more heterogeneous samples is definitely needed, although doing some will become more difficult as greater numbers of additional adolescent CI users enter postsecondary education.

Second, global trends in the results may slightly differ depending on the way age at implantation is managed as a variable. In studies that divided their sample into groups according to age at implantation, there is a general tendency for children in earlier implanted groups to demonstrate better language outcomes. This is also the case when age at implantation is treated a dichotomous variable. However, the various cut-offs makes it difficult to conclude that there is a “magical age” for cochlear implantation that should not be exceeded for greater language benefits. Several studies demonstrated that children who received CIs before the age of 24 months had better language scores, whereas studies that applied a cut-off at 18 or at 12 months also demonstrated an "earlier advantage." In a few other studies, the turning point was purported to be 2.5 years old, or even 4 years old. This variability resonates with the conclusion of Bruijnzeel et al. (2016), who found inconsistent evidence in favor of additional speech and language benefits of cochlear implantation before the age of 12 months. Similarly, Forli et al. (2011) in a systematic review on the effectiveness of pediatric cochlear implantation also concluded that "the data in the selected publications is insufficient to assess whether the advantages identified in children implanted in their first year of life is retained over time and to what extent they are influenced by a longer period of usage of the implant" (p.283).

This is not to say that age at implantation is not an important factor in predicting later language outcomes. Early cochlear implantation is undoubtedly beneficial and does carry predictive value on spoken language achievement. Moreover, early cochlear implantation benefits various developmental domains other than spoken language. For example, studies have shown that cochlear implantation improves quality of life (e.g. Archbold, Sach, O'Neill, Lutman, & Gregory, 2008) and self-esteem (e.g. Percy-Smith, Cayé-Thomasen, Gudman, Jensen, & Thomsen (2008), and decreases loneliness (Schorr, 2006). Nevertheless, there are a variety of child-, family-, and environment-related factors that can influence children's' progress following cochlear implantation.

The role of personal, familial, and environmental factors as sources of variability

In the present review, especially when age at implantation is considered as a continuous variable in studies considering its association with language achievement, there is striking evidence that other predictive factors play important roles. For instance, when children either with other disabilities or those from multilingual homes are included in study samples, those variables emerge as significant predictors of language outcomes (e.g. Boons et al., 2013). In many studies, these potential sources of variability are simply not present or not reported in the study samples. For example, when all children come from high SES backgrounds, have no other disabling conditions, and speak a single language at home and at school, it is more likely that age at implantation tends to emerge as a key variable in predicting language outcomes, because other sources of variability are not present. However, it is also likely that personal characteristics of deaf children beyond age at implantation are related to the variability in language outcomes. A variety of studies have indicated that 30 to 40% of deaf children present with additional disabilities (e.g., Fortnum, Marshall, Bamford, & Summerfield, 2002; Knoors & Vervloed, 2011). In a survey of 15 European cochlear implant centers, the proportion of children considered having complex needs ranged from 10% to 60% (Archbold et al., 2015). In recent years, there has been a substantial increase of children with additional disabilities receiving CIs (Meinzen-Derr, Wiley, Grether, & Choo, 2011). The question of whether children with additional disabilities should be included in CI studies along with more typical deaf children remains an important issue and the representativeness of the study samples should be re-examined if research is intended to better reflect clinical realities. Homogeneity of samples might be a good idea from a methodological point of view, as it decreases variability and increases the internal validity of studies, but such samples limit the generalizability of the results, from both clinical and experimental points of view.

In the body of research on typical language acquisition, the issue of variability, especially regarding vocabulary, has been discussed frequently (e.g., Bassano, 1998; Bates, Dale, & Thal, 1995; Dale & Goodman, 2005). The predictive factors of language growth in hearing children are also a critical issue with regard to pediatric CI users. For example, findings from many studies suggested that SES has an important effect on language development, and that parental input plays a central role in language growth during preschool years (Hoff, 2003; Huttenlocher et al., 2010; Rowe, 2008). In the CI field, and in research on children with hearing loss in general, it is only recently that researchers have started to consider environmental variables in children with CIs and rigorously document family variables and parental input. Since it is likely that age at implantation is really just one element among a large set of predicting factors rather than the only factor that counts for language achievement, future studies should focus more on environmental variables, thus following research designed to better understand typical language acquisition in children. Controlling for such variables is difficult, especially with the relatively small samples seen in most of the relevant research (and their diminution over time). As pediatric cochlear implantation becomes more frequent, studies should also be able to provide additional insights into the complex interplay of such factors in language and other developmental domains, for example, the onset, intensity and content of therapeutic intervention.

The role of language input

Does early access to sound or early access to language matter more for "good" language outcomes in children with severe to profound hearing loss? Naturally, spoken language acquisition is dependent on auditory input, but the construction of language relies on more than acoustics. The spoken language delays that often are observed in children with hearing loss are assumed to be primarily related to an auditory-perceptual deficit (Levine, Strother-Garcia, Golinkoff, & Hirsch-Pasek, 2016). However, in young children, impoverished sensory input also results in fewer opportunities to communicate and fewer linguistic interactions. This situation exists before cochlear implantation and can persist afterwards. For example, Fagan, Bergeson, and Morris (2014) found that mothers of children with CIs provided a reduced linguistic environment compared to that provided to same-aged hearing children. Campbell et al. (2014) suggested that a "secure first language" should be established as soon as possible to ensure "good" language outcomes after cochlear implantation. Consequently, the quantity and quality of the linguistic experience, pre-CI and with a CI, emerge as an important issue to address in future studies.

Limitations of this Review / Validity Issues

Although our initial search stage was rigorous, we did not conduct a genuine systematic review. Levels of evidence and overall quality of the reviewed studies were not appraised, largely because studies with high levels of evidence remain scarce in the CI domain – as well as in many areas of research in communication disorders and interventions for children with hearing loss. Consequently, the inclusion of studies with weaker designs (e.g. retrospective studies) might have influenced the conclusions we drawn from primary studies, as well as the way age at implantation is defined and operationalized, as a categorical or continuous variable.

Not only the methods, designs, analyses, and types of scores used to express outcomes varied (e.g. standard scores, raw scores, language quotients), but other timing factors (chronological age and duration of CI use) also were wide-ranging, somewhat limiting the interpretation of the results from primary studies. Similarly, in several studies, the influence of age at implantation is confounded with the length of experience with a CI (Nicholas & Geers, 2007) whereas in other studies, duration of CI use is confounded with chronological age (Lund, 2016).

In addition, the use of language quotients somewhat limits the generalizability of the findings in four of the reviewed studies (Boons et al., 2012, 2013; Miyamoto et al., 2008; Tomblin et al., 2005), as age-equivalent scores are ill suited to adequately monitor language progress (McCauley & Swisher, 1984). This caveat has also been reported by Lund (2016). Finally, several studies reported analyses of the same dataset. There are serious limitations of reusing data across multiple publications (van Raaij, 2018). Data overlap might suggest that earlier published studies constitute "corroborating proof for findings in a later publication" (van Raaij, 2018, p. 186) thus introducing a form of circular reasoning. We cannot rule out the possibility that the inclusion of overlapping data in the meta-analysis of effect sizes impacted the findings of the present review.

Despite these limitations and validity issues, we devoted effort to avoid selective reporting of the existing evidence. Our aim was to provide a general overview of the current evidence regarding the effect of age at implantation on central linguistic abilities.

A second limitation is that we have not addressed pragmatics and conversational and narrative skills in this review. Compared to vocabulary, very few studies have examined language use; yet, it is well recognized that more complex linguistic skills, especially conversations and narratives, are very important for school functioning and for peer-acceptance and social relations (Boons et al., 2013; Martin, Bat-Chava, Lalwani, & Waltzman, 2010). Boons et al. (2013) also pointed out that narrative abilities are predictors of reading and writing achievement and can provide insights on the effect of cochlear implantation on more complex language skills. Future research should examine language outcomes beyond standardized vocabulary scores and evaluate how children use their vocabulary in a functional manner and how early cochlear implantation influences natural interactions with the environment.

Conclusions

In many high-income countries, congenital hearing loss is viewed as a neurological emergency that needs to be managed urgently. With the advent of newborn hearing screening, babies are identified within their first weeks of life and families are oriented toward CI programs as soon as the hearing loss seems of significant degree. Early identification and the possibility of even earlier cochlear implantation – well before the first birthday – can impact family adjustment to their child's hearing loss and parental expectations (Young & Tattersall, 2007). The claim that cochlear implantation have to be performed before a certain age to prevent language delay clearly puts pressure on parents and creates the expectation that if (and perhaps only if) cochlear implantation takes place speedily, language will develop similarly to a typically developing hearing child. It is not hearing loss in itself that causes language delays but rather the lack of appropriate exposure to language during the first years of life.

To the extent that the impact of age of implantation varies with child and family characteristics, parents need to be informed of all the implications of early implantation in order to be able to make appropriate decisions for their children and their families. Is the family in a position to provide a child with the level of support necessary to succeed with a CI? This and a variety of other questions require a fuller understanding of the effects of age of implantation than currently appear to be assumed by practitioners and researchers in the field. Only when parents have complete, objective information concerning the likely needs and outcomes of their unique child will they be in a position to make an informed decision for all involved.

[1] Throughout this article, ages are reported in the formats found in the original sources.

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Description of Supplemental files

Supplementary Table 1:

Effects of age at implantation and language achievement with age as a discrete variable (age groups): Information provided by authors for CI users and calculated effect sizes.

Supplementary Table 2:

Effects of age at implantation and language achievement with age as a continuous variable: Information provided by authors for CI users and calculated effect sizes.

Supplementary Table 3:

Additional studies that reported associations between age at CI and language (effect of age at CI was not the main goal of the study). Information provided by authors.

Conflict of interest

The authors disclose no conflicts of interest. No financial interest has been provided to the authors as a result of this work.

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Supplementary Table 1. Effects of age at implantation and language achievement with age as a discrete variable (age groups): Information provided by authors for CI users and calculated effect sizes (NC = not calculable from information provided)

Authors	**Groups**	**N**	**Mean Age at CI (SD) [range]**	**Mean Age at Test (SD) [range]**	**Mean Duration of CI Use (SD) [range]**	**Language Domain: Assessment/ Task[2]**	**Statistical Analyses[1]**	**Primary Findings**	**Effect Size (Glass' d)**
N age groups									
Artières, Vieu, Mondain, Uziel, & Venail (2009)	Group 1	32	1.7 y [1.1-1.9]	3.9 [2.7-6.4]	2.7 [1.1-4.5]	Receptive vocabulary: PPVT Expressive language (5-levels scale of development)	*t* tests Logistic regression	Differences between groups were more consistent for groups of later-implanted children (mean age at CI 3.6 vs 4.5); Regression analysis: better receptive vocabulary scores were significantly associated with age at CI. No association with expressive language.	Group 1 vs group 2 PPVT at age 5: .69 Expressive language at age 4: .80
	Group 2	15	2.6 y [2.5-2.9]	8 [5.6-9.1]	5.3 [3.1-6.6]				Group 2 vs group 3 Expressive language at age 5: 1.001
	Group 3	14	3.6 y [3.3-3.7]	6.9 [5.8-10.2]	3.1 [2.5-6.3]				Group 3 vs group 4 PPVT at: age 5: .95 age 6: 1.2 age 8: .83 Expressive language at: age 5:

									1.0 age 6: .98 age 8: .81
	Group 4	13	4.5 y [4.3-4.6]	9.9 [7.2-11]	4.5 [2.7-6.2]				-
Colletti, Mandalà, Zoccante, Shannon, & Colletti (2011)	Group 1	19	6.4 mo (2.8) n=10	-	10-year follow-up	Receptive vocabulary: PPVT Receptive grammar: TROG	Wilcoxon-Mann-Whitney	Earliest-implanted group outperformed later implanted groups on both tasks.	NC
	Group 2	16	19.3 mo (3.8)	-					
	Group 3	33	30.1 mo (5.9) n=21	-					
Connor, Craig, Raudenbush, Heavner, & Zwolan (2006)	Group A1	21	21 mo	-	4 years [up to 13 years]	Receptive vocabulary: PPVT (raw scores)	Hierarchical linear modeling (HLM); Regression	Earliest implanted group had greater rates of vocabulary growth than children in other groups for the first 3 years after implantation.	NC
	Group A2	15	36 mo	-					
	Group 2	20	50 mo	-					
	Group 3	44	90 mo	-					
Dettman, Dowell, Choo, Arnott, Abrahams, Davis, ... & Briggs (2016)	Group 1	15 1	0.70 y (0.15)	PPVT: 5.6 y (0.87) (n=207) PLS: 5.4 y (1.0) (n=95) CELF: 8.02 y (2.22)	PPVT: 3.4 y (1.1) PLS: 3.8 y (1.0) CELF: 6.03 y (2.02)	Receptive vocabulary: PPVT Receptive and expressive language: PLS 4 and 5 Receptive and expressive language: CELF	Regression ANOVA	Group 1 outperformed other groups for all language measures (Group 1 had longer CI experience at the time of PPVT and PLS testing).	(PPVT at school entry) vs group 2: .89
	Group 2	61	1.24 y (0.14)						vs group 3: .91
	Group 3	66	1.75 y (0.13)						vs group 4: -1.8
	Group 4	82	2.60 y (0.43)						vs group 5: .03

	Group 5	43	4.45 y (0.69)	(n=122)					-
Holt & Svirsky (2008)	Group 1	6	10.2 mo	From 12 to 96 months	From 6 to 90 months	Receptive language: MBCDI or RDLS Expressive language: MBCDI or RDLS	Regression (on DTA) HLM	Receptive: differences between groups were significant throughout the entire follow-up period. Expressive: no differences between the two earlier-implanted groups (1-2).	NC
	Group 2	32	18.6 mo						
	Group 3	37	29.9 mo						
	Group 4	21	40.8 mo						
Miyamoto, Hay-McCutcheon, Iler Kirk, Houston, & Bergeson-Dana (2008)	Group 1	8	before 12 mo	-	PLS at 6 mo to 1 y of use (n=13); RDLS at 2-3 y of use	Receptive language: - PLS (standard scores) - RDLS (language quotients); Expressive language: - PLS (standard scores) - RDLS (language quotients)	ANOVA (RDLS)	Differences in language quotients between earlier-implanted and later-implanted groups were not statistically significant.	Group 1 vs group 2: - Receptive RDLS LQ: -.17 Expressive RDLS LQ: : .06 Receptive PLS: 1.05
	Group 2	38	from 12 to 23 mo	-					Group 2 vs group 3: Receptive RDLS LQ: .43 Expressive RDLS LQ: .42 Receptive PLS:

									.85
	Group 3	45	from 24 to 36 mo	-					-
May-Mederake (2012)	Group 1	total n = 28	> 12 mo	From 33.3 (7.3) to 72.6 (16.3) mo	From 1.77 to 4.45 y depending on the subtest	Receptive language: SETK (6 subtests depending on chronological age); Receptive grammar: TROG (n = 19)	Mann-Whitney	Children in group 1 had higher scores (*p* = .076?) than children in group 2 only for the Sentence comprehension subtest (n=15).	NC
	Group 2		12-18 mo						
	Group 3		18-24 mo						
Nicholas & Geers (2017)	Group 1	27	Total sample = 19.23 mo (8.51)	4.5 y	Total sample = 35.54 mo (8.47)	Expressive language measures (spontaneous samples): - NDRW - MLU-w - NDBM	Effect sizes (Cohen's *d*) on mean differences in z-scores; Pearson correlations	Effect sizes of mean differences in z-scores between groups were large for ages at CI below 18 months.	Cohen's *d* ranged between .31 and 1.02 (Table 4 in the article)
	Group 2	42							
	Group 3	24							
	Group 4	14							
	Group 5	22							
Niparko, Tobey, Thal, Eisenberg, Wang, Quittner, ... & CDaCI Investigative Team (2010)	Group 1	72	15.5 mo (3.2)	51.6 mo	Testing at 3 y of use	Receptive and expressive language: RDLS	Non-parametric regression	Children in group 1 (< 18 mo) had significantly higher rates of growth for both receptive and expressive language than children in other groups.	NC
	Group 2	64	29.4 mo (5.6)	65.7 mo					
	Group 3	52	48.5 mo (7.4)	85 mo					
Percy-Smith, Busch, Sandahl,	Group 1	28	Total sample = 19.6	Total sample = 46.3	Total sample = 25.9 mo	Receptive vocabulary: PPVT-4 Receptive	Fisher's exact tests Logistic	Children in group 1 (age at CI: 5-11 mo)	NC
	Group 2	19							

Nissen, Josvassen, … & Cayé-Thomassen (2013)	Group 3	36	mo	mo		language: RDLS "Active vocabulary" (Viborgma-terialet)	regression Odds ratios (Wald tests)	had higher odds ratios than children in other groups.	
Svirsky, Teoh, & Neuburger (2004)	Group 1	12	19.7 mo (1.9)	From 16 to 84 months	-	Expressive language: MBCDI or RDLS	*t* tests DTA	Children in group 1 had better expressive language skills (for various durations of use).	NC
	Group 2	34	29.8 mo (3.4)		-				
	Group 3	29	40.6 mo (2.5)		-				
Dichotomized age at implantation									
Dunn, Walker, Oleson, Kenworthy, Van Voorst, Tomblin, ... & Gantz (2014)	Group 1 (CI under age 2)	13	1.38 y (0.27)	7.8 y (2.7) [3.0 – 12.8]	Periodical testing at 7 to 11 years of age	Receptive language: CELF-3 (subtest Concepts and Directions) Expressive language: CELF-3 (subtest Formulated Sentences)	*t* tests	At 7 years of age, the younger implanted group had higher both receptive and expressive language scores. By 8 to 10 years of age, no difference was found.	Receptive language: at age 7: .72 at age 9: .59 Expressive language: at age 7: .92 at age 10: .63 at age 11: .44
	Group 2 (CI from 2 to 4 y)	25	2.99 y (0.55)	12.2 y (5.04) [3.2 – 22.4 y]					
Houston & Miyamoto (2010)	Group 1 (CI from 7 to 12 mo)	7	Total sample = 14.8 mo [7.6-22.6]	-	Testing at 2-2.5 and 3-4 years of CI use	Receptive vocabulary: PPVT	*t* tests	At both intervals, earlier-implanted group had better scores than the later-implanted group (n=14 at interval 1; n=11 at interval 2).	At 2-2.5 y of use: 1.25 At 3-4 y of use: 1.44
	Group 2 (CI from 16 to 23 mo)	8							
Leigh, Dettman,	Group	35	0.84 mo	-	Testing at	Receptive language:	Correlations	Receptive language:	RITLS mean

Dowell, & Briggs (2013)	1		(0.15)		1,2,3, and 5 years of use	RITLS Receptive vocabulary: PPVT (at 3 years of use)		average growth rate was not different between both groups; Receptive vocabulary at age 3: standard scores were correlated with both age at hearing aids fitting and age at CI.	growth: .32 PPVT at 3 years of use: 1.09
	Group 2	85	1.60 mo (0.25)	-					
Markman, Quittner, Eisenberg, Tobey, Thal, Niparko, & Wang (2011)	Group 1	34	1.15 y (0.17)	-	Testing at 4-5 years of use	Receptive and expressive language: CASL (core composite)	Regression	Children in group 1 had better scores in each of the four language tasks. Maternal sensitivity was a significant predictor of language outcomes in both groups.	NC
	Group 2	62	2.88 y (1.03)	-					NC
Nicholas & Geers (2013)	Group 1	27	9.6 mo (1.3)	54.4 mo (1.5)	44.9 mo	Receptive vocabulary: PPVT Receptive language: PLS Expressive language: PLS	*t* tests	Mean scores of the earlier implanted group were statistically higher than those of the later implanted	Receptive vocabulary: .71 Receptive language: .79 Expressive languag
	Group 2	42	14.7 mo (2.5)	54.8 mo (1.3)	40.1 mo				

								group.	e: .60
Rinaldi, Baruffaldi, Burdo, & Caselli (2013)	Group 1	11	total sample = 14.26 mo (4.69)	total sample = 28.78 mo (5.08)	total sample = 14.52 mo (5.08)	Expressive language: MBCDI - words produced - sentences produced - % complex sentences	*t*-tests on *z*-scores	No effect of age at CI (below 12 vs 13-26) on vocabulary size and grammatical skills	words: .50 sentences: .23 %complex: .52 (same as in Table 3 in the article)
	Group 2	11							
Tobey, Thal, Niparko, Eisenberg, Quittner, Wang, et al. (2013)	Group 1	98	total sample = 29 mo [6 mo – 4 y 11 mo]	From 4.8 to 11.5 y	Testing at 4,5, and 6 years of use	Receptive and expressive language: CASL (core composite)	Multivariate analyses Fisher's exact test	Trajectories of the core composite standard scores at 4, 5, and 6 years of CI use did not significantly differ as a function of age at CI.	NC
	Group 2	62							NC
Uziel, Sillon, Vieu, Artieres, Piron, Daures, & Mondain (2007)	Group 1	43	total sample = 4.8 y (2.3 y)	From 12 to 24 y	total sample = 11.7 y (1.7 y)	Receptive vocabulary: PPVT	Odds ratio Chi-square ANOVA	An older age at implantation increased the risk of a PPVT score below the 50th percentile by a 2.6 factor.	NC
	Group 2	39							NC
Wie (2010)	Group 1	13	total sample = 11.3 mo (3.9)	7 to 29 mo	total sample = 37 mo (10.4)	Receptive language: MSEL; MCDI Expressive language: MSEL; MCDI	*t* tests	Children in group 1 had higher language scores than children in group 2 at all times of	NC
	Group 2	7							

								testing.	

[1]Note: DTA: Developmental Trajectory Analysis; HLM: Hierarchical Linear Modeling.
[2]Note: CASL: Comprehensive Assessment of Spoken Language; CELF: Clinical Evaluation of Language Fundamentals; MCDI: Minnesota Child Development Inventory; MSEL: Mullen Scale of Early Learning; MBCDI: MacArthur-Bates Communicative Development Inventories; PPVT: Peabody Picture Vocabulary Test; PLS: Preschool Language Scale; RDLS: Reynell Developmental Language Scales; RITLS: Rossetti Infant-Toddler Language Scales; SETK: Sprachentwicklungstest fur Kinder ; TROG: Test for Reception of Grammar; MLU-w: Mean length of utterances-words; NDBM: Number of different bound morphemes; NDRW: Number of different root words.

Supplementary Table 2. Effects of age at implantation and language achievement with age as a continuous variable: Information provided by authors for CI users and calculated effect sizes (NC = not calculable from information provided)

Authors	**N**	**Mean Age* at CI (SD) [range]**	**Mean Age at Test (SD) [range]**	**Mean Duration of Use (SD) [range]**	**Language Domain: Assessment/ Task[1]**	**Statistical Analyses**	**Primary Findings**	**Effect Size (Glass' d)**
Black, Hickson, Black, & Khan (2014)	174	44.02 mo (30) [4 – 180]	-	Retrospective data from outcome measures at 18 to 24 months of CI use	Various tests depending on chronological age (e.g. PPVT, PLS, CELF) (depending on the measure, n = 38 to 89)	Regression	Age at implantation was not significantly associated with language scores. Strongest predictors were family concern and the presence of an inner ear malformation.	NC
Boons, Brokx, Dhooge, Frijns, Peeraer, Vermeulen, et al. (2012)	288 (not all participants were tested at all times on all tests: n = 115 to 140)	26 mo (13) [6 – 60]	Up to 8 years old	Testing at 1, 2, and 3 years of CI use	Receptive language: RDLS Expressive Language: SELT (Language quotients (LQ)	Regression	Age at implantation was a significant predictor of language skills during the first 3 years after implantation.	NC
Boons, De Raeve, Langereis, Peeraer,	70	Median: 20 mo [6 – 60]	Median: 8 y 2 mo [5 – 13 y]	Median: 6 y 4 mo [1; 6 – 10; 6 y]	Expressive vocabulary: EOWPVT Expressive syntax: CELF	Logistic regression	Age at implantation was not a significant predictor for any of	NC

Wouters, & Van Wieringen (2013)							the language components. Strongest predictors were the presence of additional disabilities and multilingualism.	
Cuda, Murri, Guerzoni, Fabrizi, & Mariani, (2014)	30	11.8 mo (3.2)	24.2 mo (3.2)	Testing at 36 months of age	Expressive vocabulary and grammar: MBCDI	Regression	Age at implantation and sex (girl) were both associated with a higher number of words; sentence complexity was associated with age at implantation, sex, and maternal education level.	NC
Fagan (2015)	9	12.46 mo [8.9 – 14.4]	At 12 months of CI use: 25.7 mo (2.05)	Testing at 4 and 12 months of CI use	Expressive vocabulary: MBDCI	Correlations	Age at implantation was associated with MBCDI score at 12 months of use.	1.53 (at 12 months of use)
Geers, Moog, Biedenstein, Brenner, Hayes (2009)	153	2; 4 y (0; 11) [0; 11 – 5; 1]	5; 10 y (0; 6) [4; 11 – 6; 11]	3; 6 y (0; 11) [1; 0 – 5; 4]	Receptive vocabulary: PPVT Expressive vocabulary: EVT or EOWPVT	Regression	IQ was the strongest predictor of vocabulary and language scores, followed by parent education	Receptive vocabulary: .47 Expressive vocabulary: .50

							level.	
Geers, Nicholas, & Moog (2007)	sample 1: 74 sample 2: 126	28.21 mo (11.60)	70.04 mo (7.06) [60 – 83]	-	Receptive vocabulary: PPVT	Regression	Five variables (age at implantation, gender, parent education, age at hearing aid fitting, and age at test) accounted for 24% of the variance in PPVT scores.	sample 1: .96 sample 2: .54
Geers, Nicholas, & Sedey (2003) Geers (2004)	181	3; 5 y (0; 10) [1; 8 – 5; 4]	8; 11 y (0; 6) [7; 11 – 9; 11]	5; 6 y (0; 9) [3; 9 – 7; 6]	Receptive language: TACL Expressive language: lexical and grammatical measures converted in a Total Language Score	Regression	Age at implantation was not significantly associated with language achievement.	NC
Geers & Nicholas (2013)	60	22.7 mo (7.7) [12 – 38]	Testing at 10.5 years old	8.6 y (1) [7 – 11]	Receptive vocabulary: PPVT Expressive vocabulary: EOWPVT Receptive language: CELF Expressive language: CELF	Regression	Age at implantation and a set of additional variables (related to auditory, personal, and family factors) were associated with language outcomes at 10.5 years old.	PPVT: .67 EOWPVT: .95 CELF (receptive): .80 CELF (expressive): -.85
Hay-McCutcheon, Iler	30	4.48 y (1.61)	Regular testing up to 18	-	Receptive and expressive language:	Regression (mixed-	Age at CI was significantl	NC

Kirk, Henning, Gao, & Qi (2008)		[1.4 – 7.7]	years old		RDLS and CELF	effects)	y associated with early receptive and expressive language measures (from 2 to 7 years of age), but not the later language measures (from 9 years old onwards).	
Hayes, Geers, Treiman, & Moog, (2009)	65	2.69 y (0.90) [1.08 – 4.75]	5 y (at first test) longitudinal yearly testing	2.39 y (1.29) (at first test) [0 – 6.42]	Receptive vocabulary: PPVT	Multilevel regression models (growth curve analyses)	Children who received a CI at a younger age showed a faster receptive vocabulary growth rate than children who received an implant later.	NC
Lund (2015) Meta-analysis of 16 studies	34 to 158	16 to 46.5 mo	49 to 109 mo	-	Receptive and expressive vocabulary	Meta-regression	Neither age at implantation, nor duration of use, nor age at the time of testing were associated to the magnitude of weighted effect sizes.	-

Nicholas & Geers (2007; 2008; 2009)	76	23.16 mo (7.75) [12 – 38]	Testing at 3.5 and 4.5 years old	At 3.5 y: 19.76 mo (7.64) [7 – 32] At 4.5 y: 55.09 mo (1.15) [52 – 57]	Receptive and expressive vocabulary and language (various tests: PPVT, EOWPVT, PLS, CELF)	Regression (linear and quadratic)	Age at CI and a set of additional variables (related to auditory, personal, and family factors), were associated with language outcomes.	NC
Schorr, Roth, & Fox (2008)	39	[1; 3 – 8; 2 y]	9 y [5; 4 – 14; 11]	[1; 8-11; 8 y]	Various tests according to chronological age (e.g. PPVT, TOLD)	Regression	Age at implantation was associated only with receptive vocabulary scores.	NC
Szagun & Stumper (2012)	25	20.4 mo (11) [6 – 42]	-	Testing at 12, 18, 24, and 30 months of CI use - Analyses at 30 months of CI use	Expressive vocabulary: word types Expressive grammar: MLUm Expressive vocabulary and grammar: MBCDI	Correlations ANCOVA	At 30 months of use, only maternal education was significantly associated with language measures.	Language sample: -word types: .32 -MLUm: .34 MBCDI: -words: .56 -sentence complexity: .58 -inflectional morphology: .84
Szagun & Schramm (2015)	48	24 mo (10) [6 – 46]	-	Testing at regular intervals from 6 to 36 months of CI use	Language sample: -MLUm -Type and token frequencies of determiners -Type	Regression Correlations	Language measures from 6.5 up to 20 months of CI use were not associated	MLU at 24 months of use: .46 MLU at 30

					frequencies of lexical words		with age at implantation. At 24 and 30 months of use, age at CI added 9% and 10% of unique variance of MLU (parental expansions accounted for 48% and 43% of the unique variance of MLU).	months of use: .50
Tomblin, Barker, Spencer, Zhang, & Gantz (2005)	29	21 mo (7) [10 – 40]	5 to 78 mo	-	Expressive language: MCDI PLS (converted into an Expressive Language Quotient (ELQ)	Hierarchical linear model	Both at 12 and 24 months of CI use, age at implantation was significantly associated with ELQs.	PLS ELQ at 24 months of use: -1.8 MCDI ELQ at 12 months of use: .85 at 24 months of use: -1.35
Willstedt-Svensson, Löfqvist, Almqvist, & Sahlén (2004)	15	3 y and 11 mo [2 y – 6 y and 1 mo]	7 y and 7 mo [5 y and 4 mo – 11 y and 5 mo]	-	Receptive grammar: TROG Expressive grammar: Lund Test of Grammar	Regression	Age at CI was associated with both receptive and expressive grammar scores; with working memory added as a predictor, age at CI	Receptive grammar: 1.75 Expressive grammar: 1.5

							did not account for a significant proportion of variance.	

*mo: months; y: years

[1]Note: MCDI: Minnesota Child Development Inventory; TOLD: Test of Language Development; MLUm: Mean length of utterances (morphemes); SELT: Schlichting Expressive Language Test; CELF: Clinical Evaluation of Language Fundamentals; MBCDI: MacArthur-Bates Communicative Development Inventories; PPVT: Peabody Picture Vocabulary Test; PLS: Preschool Language Scale; RDLS: Reynell Developmental Language Scales; TACL: Test of Auditory Comprehension of Language; TROG: Test for Reception of Grammar; EVT: Expressive Vocabulary Test; EOWPVT: Expressive One-Word Picture Vocabulary Test.

Supplementary Table 3. Additional studies that reported associations between age at CI and language (effect of age at CI was not the main goal of the study).

Authors	N	Mean age at implantation [range] (SD)	Mean age at testing [range] (SD)	Mean duration of CI use [range] (SD)	Language domain: assessment/task[1]	Statistical analyses	Primary findings
Castellanos, Kronenberger, Beer, Henning, Colson, & Pisoni (2014)	35	25.47 mo [8.28 – 47.70] (10.77)	13.48 y [7.80 – 23.36] (10.77)	11.36 y [7.08 – 19.84] (3.40)	Receptive vocabulary: PPVT; Receptive and expressive language: CELF	Regression	Receptive vocabulary and speech intelligibility during preschool predicted later outcomes. Age at implantation did not add to the variance in language outcomes.
Duchesne, Sutton, & Bergeron (2009)	27	21.66 mo [8 – 28] (5.47)	68.4 mo [42 – 99] (17.76)	46.85 mo [23 – 71] (15.61)	Receptive and expressive language: RDLS	Correlations	Correlation between RDLS scores and age at implantation were not statistically significant.
Geers et al. (2017)	97	21.8 mo	early (5.0–7.9 y) and late (9.0–11.9 y) elementary grades	-	Receptive and expressive language: CASL	Regression	Age at implantation significantly predicted CASL scores, both in early and later elementary

							grades.
Guasti, Papagno, Vernice, Giuliani, & Burdo (2012)	33	21.7 mo [12 – 56] (10.4)	63.9 mo [50 – 82] (8.66)	42.7 mo [23 – 60] (9.5)	Receptive vocabulary: PPVT Receptive grammar: TCGB Elicitation of clitic pronouns	Mixed model analysis	A significant effect of age at implantation was found for the production of clitic pronouns.
Hammer, Coene, Rooryck, Gillis, & Govaerts (2010)	48	16 mo [5 – 43]	Between 4 and 7 years of age	-	Finite verb production (*z*-scores): spontaneous language sample	Regression	A significant association between finite verb production and age at implantation was found at 4 and 5 years of age.
Nittrouer, Lowenstein, & Holloman (2016)	51	22 mo (17)	103 mo (5)	Testing at 36, 48, and 72 months of age, and in second grade	Expressive grammar: - MLU - number of conjunctions - number of personal pronouns	Pearson product-moment correlations	No significant correlations we found between age at (first) CI and any of the grammatical measures.
Nittrouer, Sansom, Low, Rice, & Caldwell-Tarr (2014)	21	21 mo (13)	82 mo (5)	61 mo (13)	Expressive language (language sample): - MLU number of: - conjunctions - personal pronouns - final bound morphemes	Correlations	Age at (first) implant was significantly correlated with: - MLU - number of different words.

					- different words		
Ruffin, Kronenberger, Colson, Henning, & Pisoni (2013)	51	35.4 mo (19.9)	15.2 y (4.5)	12.2 y (3.6)	Receptive vocabulary: PPVT Receptive and expressive language: CELF	Correlations	Age at implantation was not associated with both PPVT and CELF scores.

[1] Note: CASL: Comprehensive Assessment of Spoken Language; CELF: Clinical Evaluation of Language Fundamentals; MLU: mean length of utterances; PPVT: Peabody Picture Vocabulary Test; RDLS: Reynell Developmental Language Scales; TCGB: Test di Comprensione Grammaticale per Bambini.

Figure 1. Meta-analysis of effect-sizes (n=32)