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Variability in Receptive Language Development Following Bilateral Cochlear Implantation

Angelika Illg,[a] **Doris Adams,**[a] **Anke Lesinski-Schiedat,**[a] **Thomas Lenarz,**[a] **and Andrej Kral**[a]

[a] Department of Otolaryngology, Medical University Hannover, Germany

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ABSTRACT

Objectives: The primary aim was to investigate the variability in language development in children aged 5–7.5 years after bilateral cochlear implantation (CI) up to the age of 2 years, and any impact of the age at implantation and additional noncognitive or anatomical disorders at implantation.

Design: Data of 84 congenitally deaf children that had received simultaneous bilateral CI at the age of ≤ 24 months were included in this retrospective study. The results of language comprehension acquisition were evaluated using a standardized German language acquisition test for normal hearing preschoolers and first graders. Data on speech perception of monosyllables and sentences in quiet and noise were added.

Results: In a monosyllabic test, the children achieved a median performance of 75.0 ± 12.88%. In the sentence test in quiet, the median performance was 89 ± 12.69%, but dropped to 54 ± 18.92% in noise. A simple analysis showed a significant main effect of age at implantation on monosyllabic word comprehension ($p < .001$), but no significant effect of comorbidities that lacked cognitive effects ($p = .24$). Language acquisition values correspond to the normal range of children with normal hearing. Approximately 25% of the variability in the language acquisition tests is due to the outcome of the monosyllabic speech perception test.

Conclusions: Congenitally deaf children who were fitted bilaterally in the 1st year of life can develop age-appropriate language skills by the time they start school. The high variability in the data is partly due to the age of implantation, but additional factors such as cognitive factors (e.g., working memory) are likely to influence the variability.

The primary objective of auditory habilitation in deaf children with cochlear implantation (CI) is to increase educational and employment opportunities through the development of functional listening skills for continued language learning and enhanced communicative interactions (Fryauf-Bertschy et al., 1997; Kral et al., 2019; Manrique et al., 1999; McConkey Robbins et al., 2004; Sharma et al., 2002). But the variability in recognizing speech and understanding spoken language is still unexplained in CI (Lazard et al., 2012; Pisoni et al., 2017). In pediatric CI, a shift in the implantation age below 3 years improved overall CI performance, such as speech and language development and understanding of spoken language, but did not reduce variability in these outcomes (Niparko et al., 2010).

During the 1st year of life, the child acquires the sound repertoire of the mother tongue (Johnson & White, 2020; Kuhl, 2004; Kuhl & Rivera-Gaxiola, 2008). In the process, a universal ability to limit oneself to the phonetic contrasts used in the native language develops (Werker & Tees, 1992). The 2nd year of life is characterized by the development of comprehension, the lexicon and its organization, and that of word production, so that multiword utterances emerge (Goldfield & Reznick, 1990; Reznick & Goldfield, 1992; Segbers & Schroeder, 2017). During the 3rd and 4th years of life, words are placed in semantic relationships to one another and coded according to morphological and syntactic rules; thus an abstract linguistic system of rules is present, with which the internal representation of language begins. These learning processes are primarily

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Correspondence to Angelika Illg: illg.angelika@mh-hannover.de. ***Disclosure:*** *The authors have declared that no competing financial or nonfinancial interests existed at the time of publication.*

characterized by the interplay of auditory perception, and cognitive and social skills, and continue through interactions with other developing linguistic and nonlinguistic skills (Bertoncini & Cabrera, 2014).

In hearing children raised in social deprivation beyond a critical period of 8–10 years, language acquisition (mainly grammar skills) was compromised, yet the accumulation of lexical and phonetic imitational skills was less affected (Curtiss, 1978). However, in addition to language, social isolation ("global neglect") may affect overall brain size and intelligence (Nelson et al., 2007; Uylings, 2006) and thus has a more severe impact than hearing loss alone.

Optimal hearing and speech and language development after CI during childhood depends on the developmental plasticity of the brain at the time of CI (Kral & Sharma, 2012). Studies indicated that CI in children under the age of 2 years is most effective and that speech and language development is possible in an age-appropriate manner when there are no additional disabilities (Archbold et al., 2008; Colletti et al., 2005; Lesinski-Schiedat et al., 2006; Sarant et al., 2014; Verhaert et al., 2008). Spoken language development also occurs in an age-appropriate manner when deaf children are implanted at ages younger than 1 year (Dettman et al., 2016, 2021; Karltorp et al., 2020; Nicholas & Geers, 2013; Wie et al., 2020). Published reviews concluded that the age of implantation is one of the main predictors with regard to language development after CI implantation and that an implantation age under 12 months favors the development of speech and language (Ching et al., 2013; Duchesne & Marschark, 2019; Ruben, 2018; Streicher et al., 2015). However, these studies often only included a small number of subjects, and many methodological differences exist, for example, for evaluation of the outcomes. Therefore, the summaries often remain cautious, and calls are made for further collection and evaluation of data on the spoken early language development of deaf children with CI.

In the assessment of the language skills of hearing-impaired children during language acquisition, normed, standardized test procedures according to the subjects' ages are used to describe the developmental status in comparison to age-matched normal hearing children and to decide on appropriate support measures. The comprehension of speech can only take place once spoken language has been made accessible to the child, and is usually tested with age-appropriate word material and/or sentences in quiet or in noise. However, in addition to standard speech perception tests (like word and sentence recognition), language abilities also include vocabulary, syntax, and semantics.

In this study, the test data from 84 children aged 5–7.5 years, who had been bilaterally and simultaneously fitted with CI at the age of ≤ 24 months, were retrospectively analyzed and statistically evaluated. The primary aim of the present retrospective study was to investigate the variability in language development and the relationship to the age at implantation and special etiology or comorbidities that did not impact cognition at implantation in children. The secondary goal was to compare phonetics, vocabulary, and comprehension-related linguistic abilities.

Method

Ethic Statement

This retrospective study was reviewed by the Ethics Committee of the Hannover Medical School and approved with the number 1897–2013.

Subjects

Eighty-four children who underwent simultaneous or sequential (with a maximum interval of 6 months) bilateral CI at the age of ≤ 24 months between June 26, 2006, and November 18, 2014, were included in this retrospective study, because they were able to complete age-appropriate language development tests. All children were preoperatively diagnosed with congenital deafness and had German as their first language. However, 13 of the participants showed etiology or comorbidities (e.g., meningitis, cytomegaly virus, inner ear malformation, or syndromes) that may have affected language development (see Table 1). Since these children were also able to complete the age-appropriate language development test, they are considered separately. After implantation, the children received different rehabilitation interventions such as auditory-verbal therapy and speech and listening therapy. The frequency and intensity of these therapy sessions varied, depending on the choices of parents and therapists. All children attended regular follow-up visits in our clinic, which included standardized hearing, speech, and language acquisition tests aiming to assess the development of hearing, speech recognition, and spoken language abilities. The study group was divided into two groups: Group A (*N* = 71) without any comorbidities or special etiology and Group B (*N* = 13) with special etiology or comorbidities but without cognitive impact. All demographic data are given in Table 1. In addition to evaluation in terms of comorbidities, implantation age groups were classified for group A (see Table 2).

Speech Perception Tests

Speech-perception tests were carried out using the German-language Freiburg MST (Hahlbrock, 1957) and

Table 1. Demographic information summarizing the study population divided into Groups A and B and showing the effect of special etiology and comorbidities in the overall population.

Variable	**Group A (*n* = 71)**	**Group B (*n* = 13)**
Gender		
Female	30	8
Male	41	5
Age at first implantation (months)		
Mean (*SD*)/median (range)	11.56 (4.4)/11 (5–23)	16.92 (6.4)/19 (5–24)
Age at MSVK (months)		
Mean (*SD*)/median (range)	72.96 (6.5)/72.00 (59–89)	74.38 (7.5)/71 (63–86)
Age of CI use at MSVK (months)		
Mean (*SD*)/median (range)	60.72 (7.1)/61.00 (44–80)	56.92 (8.5)/58 (46–77)
Age at speech perception test (months)		
M (SD)/median (range)	83.38 (10.3)/83.50 (62–107)	84.69 (11.2)/85 (67–105)
Type of implant		
Advanced Bionics total	**42**	**2**
Hires 90 K Helix	38	2
Hires 90 K Advantage HiFokus Mid-Scala	4	0
Cochlear total	**72**	**20**
Nucleus CI 512	11	6
Nucleus CI 24 RE (CA)	25	7
Nucleus CI 24 RE (ST)	0	2
Nucleus CI 422	32	4
Nucleus CI 24 RE Hybrid-L	4	1
MedEl total	**28**	**4**
Concerto Flex EAS 28	9	0
Concerto Flex EAS 24	1	0
Concerto Flex EAS 20	2	4
Concerto Standard Electrode Array	8	0
Sonata TI 100	8	0
Etiology		
Genetic: connexin 26	11	0
Genetic: others	14	0
Cytomegaly virus	0	2
Meningitis	0	3
Ototoxica	1	1
Inner ear malformation	0	3
Unknown	44	2
Noonan syndrome	0	1
Waardenburg syndrome	0	1
Infection	1	0
Auditory neuropathy	0	1
Comorbidities		
Motor skills	0	1
Short bowel syndrome	0	1
Balance problems	0	1
Visual impairment	0	1

Note. Group A (*n* = 71) corresponds to children without comorbidities in the etiology and group B (*n* = 13) to those with special etiology or comorbidities without cognitive impact. MSVK = Marburger Sprachverständnistest für Kinder; CI = cochlear implantation; CA = contour advanced; ST = straight; EAS = electric acoustic stimulation.

the German-language Hochmair-Desoyer–Schulz–Moser Sentence Test (HSM) in quiet and in noise (10 dB S/N ratio, S0N0; Hochmair-Desoyer et al., 1997). All tests were conducted in the free field at 65 dB SPL and with bilateral listening. The test time was that of the first speech perception test after that of the language acquisition test. Since the analysis was retrospective, not all speech-perception data were available. In Group A,

Table 2. Number of children compared among implantation age groups.

Implantation age group (months)	Number of children
5–7	14
8–10	22
11–13	20
14–16	6
17–19	10
≥ 20	12
Total	84

N = 66 results were available in monosyllables (MST), N = 56 in sentences in quiet (HSM), and N = 60 in noise. In Group B, N = 11 were available in MST and HSM sentences in quiet and N = 9 in HSM sentences in noise.

Language Acquisition Test

The language acquisition test used ("Marburger Sprachverständnistest für Kinder," MSVK) evaluates receptive language comprehension skills in semantics, syntax, and pragmatics and is standardized for German, normal-hearing preschoolers and first graders (Lohaus & Elben, 2000). The child works with his own workbook and responds by marking black-and-white pictures with a pen.

The first subtest in the field of semantics deals with the passive vocabulary (24 items) for all three principal word classes: nouns (16 items), adjectives (two items), and verbs (six items). The child is encouraged to choose the right picture among four choices, during the presentation of two added nouns with semantic distractions and one with phonological similarity, three added verbs with phonological similarity, and three added adjectives without any qualitative differentiation.

The second subtest gathers data on the understanding of the meaning of words (10 items) by matching three to four of five pictures to a generic term (three items), or to narrower terms (seven items) with regard for example to characteristics like shape, function, or surface. In the field of syntax, the third subtest assessed the comprehension of singular and plural in twelve sentences, present and perfect tenses in two sentences, and active and passive tense in four sentences.

In addition to the correct image, there was one with a distractor showing a syntactic alternative and one with an alternative sentence content. While the results of this subtest allowed conclusions about knowledge of syntactic rules, the fourth subtest examined the ability to translate instructions of varying complexity into action. Specifically, the instructions included understanding prepositions, conjunctions, and forms of comparison (superlatives). The actions are easy to perform and require little cognitive or motor effort. Each of the eight lines in the workbook consisted of five images showing one of the following creatures in different sequences: boy, girl, fattest dog, thinnest dog, baby, fattest cat, thinnest cat, mother, and father. The understanding of prepositions (from … to, around, beneath, through) and conjunctions (instead of, and, or, either, neither . . . nor) in the verbal instructions was tested by translation into drawing either a cross, a circle, or a line.

In addition to linguistic comprehension, the fifth subtest assessed the ability to interpret the personal context of situations. For each of 12 tasks, there was one situational picture and only one of several persons on it was the one to whom a statement, a question, or a request could be correctly assigned.

The sixth subtest measured the ability to assign a verbal statement, request, or question of a person to a corresponding situation. For eight tasks in the workbook, the one correct situation image had to be marked from a selection of three possible ones.

On the basis of a large sample, norm values were calculated from the raw values, which are the benchmark for age-matched, normal-hearing children. In our evaluation, T values were used as norm values and have a fixed range of values between 40 and 59 points. The mean corresponds to a T value of 50, and the SD is 10 T value points. According to the usual conventions, T values below 40 (mean minus 1 SD: $50 - 10 = 40$) were considered below average. T values above 60 (mean plus 1 SD: $50 + 10 = 60$) were considered to be above-average performance. The normalization test was performed by the test developers on a set of 1,045 normal-hearing children. The methodological construction followed the guidelines of test theory (Lohaus & Elben, 2000). The retest reliability (interval of 3 months) of the subtests is between $r = .35$ and $r = .88$; for the overall test, the retest reliability is $r = .67$. The internal consistencies (Cronbach's alpha) are between $\alpha = .51$ and $\alpha = .82$ for the subtests and $\alpha = .89$ for the overall test.

Evaluation and Statistics

All data were analyzed statistically using MATLAB R2021b (Mathworks). In individual comparisons, we used the paired Wilcoxon test and a two-way analysis of variance (ANOVA) with post hoc Fisher least significance difference procedure. When medians are presented, the variance is given as the maximum absolute deviation from the median. A Pearson correlation coefficient was calculated for the assessment of age at implantation dependence. Statistical significance level was always set to $p < .05$.

Results

In these early bilaterally implanted children, the speech-perception outcomes in quiet reached high values (see Figure 1). In the monosyllabic test, they achieved a median performance of 75.0 ± 12.9% (see Figure 1A). In the German HSM sentence test in quiet, the median performance was again high (89.0 ± 12.7%), in contrast to 54.0 ± 18.9% in noise. Language acquisition test results (MSVK) are shown in Figure 1B.

In all these subtests, the implanted children showed mean speech and language values that corresponded to the range of children with normal hearing. The overall value of 50.0 ± 6.4 (median ± maximum absolute deviation of the median) demonstrates that bilateral CIs can effectively provide access to language in this group of prelingually deaf children.

However, because of the high variability, we were further interested in the children that underperform. Two factors were focused on: age at implantation and suspected comorbidities due to the etiology of these children.

Speech Perception Tests

The outcomes of the monosyllabic test correlated significantly with the age at implantation (see Figure 2A; $r = -.4$; $p < .001$). The performance in children implanted at the oldest implantation age group was poorer,

Figure 1. Overall outcomes of speech perception (A) and language acquisition (B) in prelingually deaf children cochlear-implanted before 25 months of age. Shown are medians ± maximum absolute deviation of the median. The gray background is the value of 50 (± 10) and corresponds to the mean value of normal-hearing children. MST = German-language Freiburg monosyllabic word test; HSM = Hochmair-Desoyer–Schulz–Moser Sentence Test.

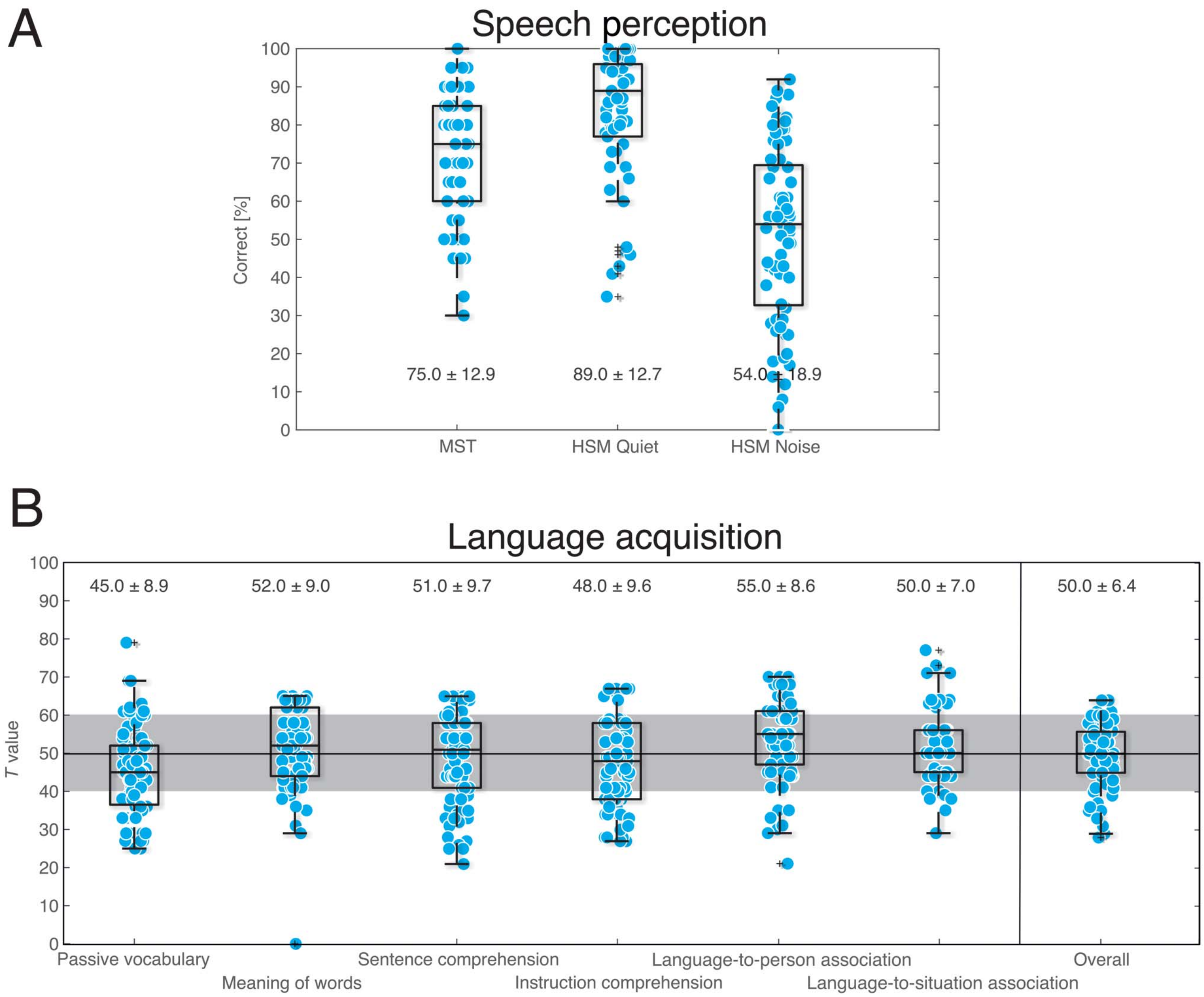
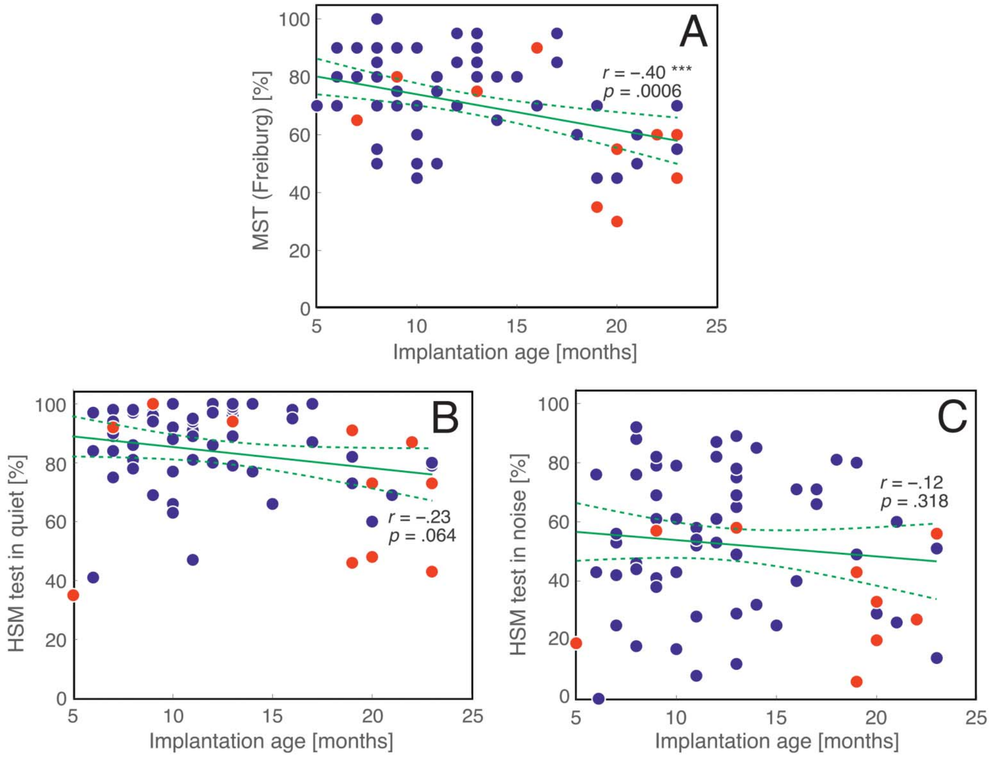


Figure 2. Scatterplot of speech perception based on a monosyllabic test (German-language Freiburg monosyllabic word test [MST]) in quiet (A), Hochmair-Desoyer–Schulz–Moser Sentence Test (HSM) Sentence Test in quiet (B), and HSM Sentence Test in noise (C). Green lines show linear regressions on the data, dashed lines show the 95% confidence interval of the regression. Purple shows children without and red those with special etiology and comorbidities. ***$p < .001$.



with an apparent age cutoff between 15 and 20 months. While the age at implantation explained only 16% of the variance, the correlation was significant, confirming that early implantations, particularly if performed before the age at implantation of 15 months, may improve outcomes.

As noted above, in the HSM sentence test, the performance approached the ceiling in the youngest-implanted children (see Figure 2B), and the linear trend was consequently flatter than in the monosyllabic test. In the HSM test in noise (see Figure 2C), however, the results were substantially more variable, and the dependence on age at implantation was again not well discernible. Neither the HSM in quiet nor that in noise showed a significant correlation with age.

Subsequently, to perform a two-way ANOVA, ages at implantation were grouped into 3-month periods from 5 months to 24 months (see Table 2). The ANOVA showed no interaction between the effects of age at implantation and comorbidities on monosyllabic word comprehension, $F(5, 59) = 1.4168$; $p = .23$. ANOVA showed a significant main effect of age at implantation on monosyllabic word comprehension ($p < .001$), but no significant effect of comorbidities ($p = .24$). This further demonstrates that even within the critical period, the age at implantation has an effect on speech acquisition. It is of particular interest that the children that were clinically defined with specific etiology or comorbidities without cognitive impact performed in the same range as that of the children without comorbidities. Post hoc statistical testing revealed that the oldest children at the time of implantation had significantly lower performance in monosyllabic word tests (MSTs) compared to all age groups up to the 14–16 months group ($p < .05$).

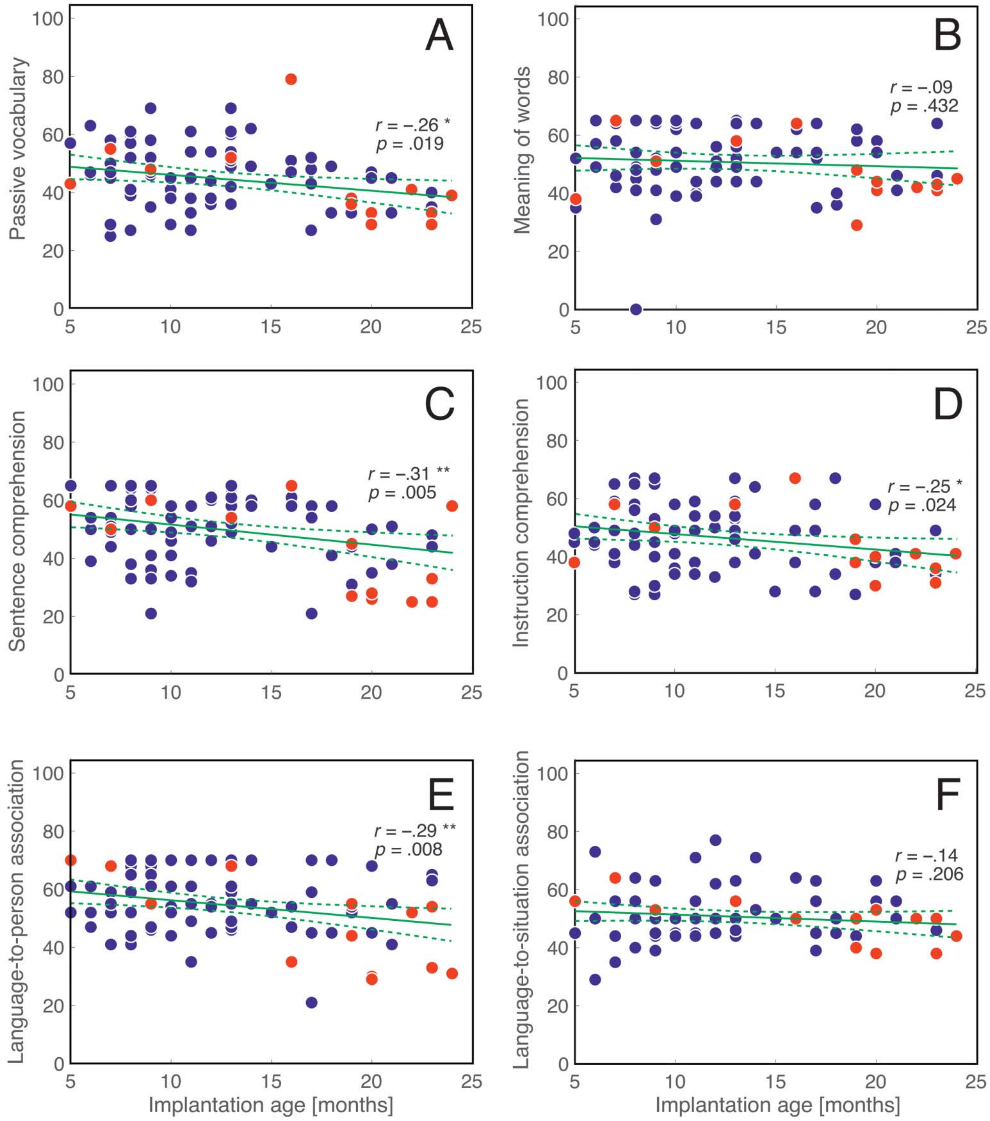
For the HSM sentence test in quiet, the overall results were similar: Again, there was no significant interaction between the effects of age at implantation and etiology on HSM sentence comprehension in quiet, $F(4, 56) = 1.1089$; $p = .362$. A simple analysis showed that there was a significant main effect of age at implantation ($p = .043$) on sentence comprehension in quiet. Missing values precluded testing the main effect of etiology on HSM sentence comprehension in quiet. Post hoc statistical testing did not reveal significant differences between the age groups (see Table 2) in HSM. For the HSM in noise (see Figure 2C), there were no significant effects in the two-way ANOVA in any measure (interaction or main effects, all $p > .29$).

Language Acquisition Tests

A similar analysis of the language acquisition tests (see Figure 3) showed that for passive vocabulary (see Figure 3A), the individual outcomes correlated significantly with the age at implantation ($r = -.257$; $p = .019$). A two-way ANOVA was performed to analyze the effect of age at implantation and etiology on passive vocabulary, and showed no significant interaction between the effects of age at implantation and etiology, $F(5, 71) = 1.6421$, $p = .160$. A simple main effect analysis showed that the age at implantation ($p = .001$) had a significant effect on passive vocabulary, but there was no effect of etiology ($p = .216$).

A test for the meanings of words (see Figure 3B) did not correlate with age at implantation ($r = -.09$; $p = .432$). In a two-way ANOVA, there was no significant interaction between the effects of age at implantation and etiology on

Figure 3. Scatterplots of the results of the language acquisition tests, shown as *T* values normalized to 100. Data are shown for passive vocabulary (A), meaning of words (B), sentence comprehension (C), instruction comprehension (D), language-to-person association (E), and language-to-situation association (F). The green line is the linear regression, while dashed lines are the 95% confidence intervals of the regression. Shown are correlation coefficients with their significance. Several measures show a statistically significant reduction of performance with increasing age. Purple data points correspond to children without specific etiology and comorbidities, while red data points correspond to children with specific etiology and comorbidities. *$p < .05$. **$p < .01$.



the comprehension of the meanings of words, *F*(5, 71) = 0.6140; *p* = .690. A simple main effect analysis showed that there was no significant effect of age at implantation (*p* = .308) nor etiology (*p* = .753) on the meanings of words.

Sentence comprehension (see Figure 3C) correlated with age at implantation (*r* = −.31; *p* = .005). In a two-way ANOVA, there was no significant interaction between the effects of age at implantation and etiology on sentence comprehension, *F*(5, 72) = 1.2030; *p* = .317. A simple main effects analysis showed that there was an effect of age at implantation (*p* = .002), but no main effect of etiology (*p* = .932) on sentence comprehension.

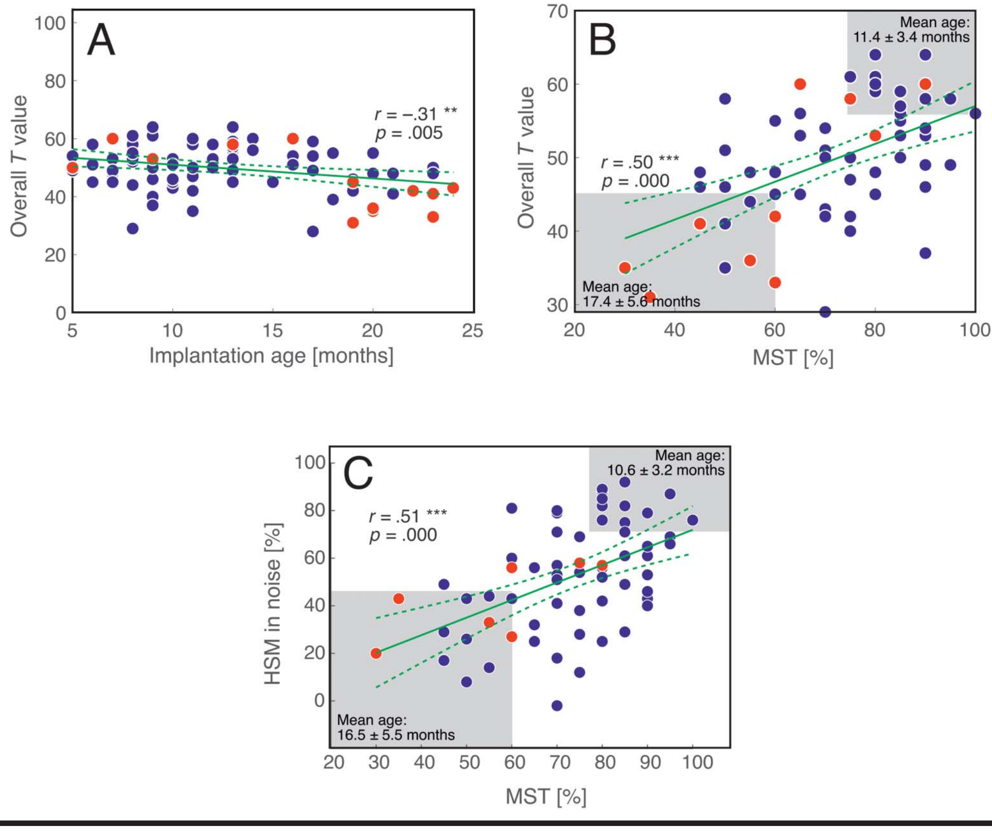
In addition, the comprehension of instructions (see Figure 3D) correlated significantly with age at implantation (*r* = −.25; *p* = .024). In a two-way ANOVA, there was no significant interaction between the effects of age at implantation and etiology on instruction comprehension, *F*(5, 72) = 1.0323, *p* = .405. There was no main effect of age at implantation (*p* = .121) nor etiology (*p* = .405) on instruction comprehension.

For language-to-person associations (see Figure 3E), the outcomes correlated significantly with age at implantation (*r* = −.29; *p* = .008). In a two-way ANOVA, there was a significant interaction between the effects of age at implantation and etiology on language-to-person associations, *F*(5, 72) = 3.4007; *p* = .008. There was a significant main effect of age at implantation (*p* = .021), but no main effect of etiology (*p* = .475) on language-to-person associations.

The association of language to the situation (see Figure 3F) did not correlate significantly with age at implantation (*r* = −.14; *p* = .206). In a two-way ANOVA, there was no significant interaction between the effects of age at implantation and etiology on language-to-situation associations, *F*(5, 72) = 0.8606; *p* = .512. There was neither a main effect of age at implantation (*p* = .477) nor etiology (*p* = .781) on language-to-situation associations.

Finally, the overall *T* value (see Figure 4) correlated with age at implantation (*r* = −.309; *p* = .005). In a two-way ANOVA, there was no significant interaction between

Figure 4. Scatterplots of (A) the dependence of the overall *T* value on the age at implantation, (B) language-acquisition tests and the results in monosyllables, and (C) results of Hochmair-Desoyer–Schulz–Moser Sentence Test (HSM) sentences in noise and the monosyllables. The green line is the linear regression, while dashed lines are the 95% confidence intervals of the regression. Shown are correlation coefficients with their significance. Purple data points correspond to children without specific etiology and comorbidities, while red data points correspond to children with specific etiology and comorbidities. Gray squares show the areas for best and poorest performers. ***p* < .01. ****p* < .001.



the effects of age at implantation and etiology on the overall *T* value, *F*(5, 71) = 0.1772; *p* = .130. There was a significant main effect of age at implantation (*p* = .002), but no significant main effect of etiology (*p* = .916) on the overall *T* value. In all language acquisition analyses with significant main effects in ANOVA in the absence of interactions, post hoc testing did not reveal any significance between different implantation ages at implantation groups.

We were subsequently interested in whether the results of the monosyllabic test related to the outcomes of the receptive language acquisition tests. Given that monosyllabic tests are most strongly dependent on phonetic analysis, and given that phonetic analysis is a precondition for speech and language acquisition, it appears likely that there will be common variability between other language measures and monosyllabic tests.

When we correlated the outcomes of MSVK subtests with the monosyllabic test, the passive vocabulary correlated significantly with the MST (*r* =.49; *p* < .001), similarly to word meaning (*r* =.35; *p* = .003), sentence comprehension (*r* =.48; *p* = .000), instruction comprehension (*r* =.358, *p* = .002), and language-to-person associations (*r* = .24, *p* = .044). The highest correlation was found with the overall *T* value (*r* = .50, *p* < .001; see Figure 4B), between passive vocabulary and sentence comprehension. Taken together, this suggests that approximately 25% of the variability of the overall language acquisition *T* value is related to monosyllabic speech perception. Subsequently, best performers were defined those that achieved results of MST > 75% and *T* value > 55%, and poor performers were MST < 60% and *T* value < 45%. These border values were selected to include true good and poor performers, having sufficient number of subjects in each group. The best performers had significantly lower implantation age than poorest performers (best performers: 11.4 ± 3.4 months, *N* = 18; poorest performers: 17.4 ± 5.6 months, *N* = 7; Wilcoxon–Mann–Whitney two-tailed test, *p* = .029).

We examined correlation between the results of the HSM test in noise and the results of the MST (*r* =.51; *p* = .000; see Figure 4C). This comparison is interesting, since (similarly to the overall *T* value) the HSM in noise also includes high-level aspects of language and cognition. The relationship informs about how much of the HSM in the noise test is determined by phonetic function. The result shows that in prelingually deaf children, there is 25% of common variability in MST and HSM in noise. Subsequently, best performers were defined as having achieved results of MST > 75% and HSM test in noise > 75%, and poor performers were MST < 60% and HSM test in noise < 50%. These border values were selected to include true good and poor performers, having sufficient numbers of subjects in each group. The best performers had significantly lower implantation age than poorest performers (best performers: 10.6 ± 3.2 months, *N* = 12; poorest performers: 16.5 ± 5.5 months, *N* = 11; Wilcoxon–Mann–Whitney two-tailed test, *p* = .019).

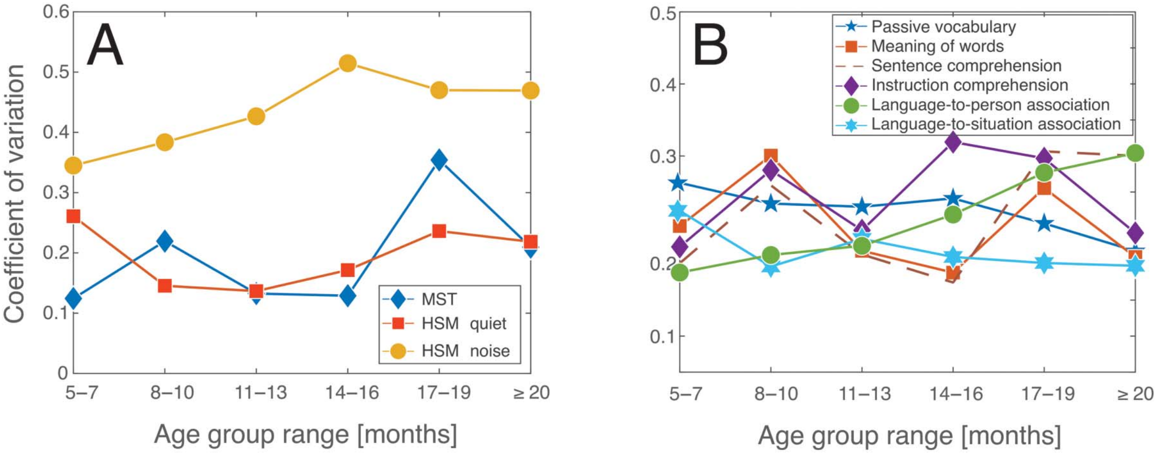
Finally, to study whether the age at implantation affected the variance in the different implantation age groups, we calculated the coefficient of variation for all measures tested and plotted it as a function of the age at implantation (see Figure 5). We found a consistent implantation-age effect in HSM sentence test in noise, the most difficult test, where the variance increased with increasing age at implantation (see Figure 5A). For the different language acquisition tests, it was the language-to-person association (that showed significant age effects in ANOVA; see Figure 3) that also showed a continuous, systematically increasing variance with increasing implantation age (nearly 3 times larger in the latest implanted group compared to the youngest implanted; see Figure 5B). This suggests that particularly the tests that involve a reference to cognitive function show increased variance with increasing implantation age.

All the other tests provided no indications of systematic changes (see Figure 5B). This might indicate that in addition to implantation age effects on overall (mean/median) performance, there are only very few effects on the variance of the results.

Discussion

The present retrospective study examined the variability of receptive language acquisitions in children aged 5 to 7.5 years after bilateral CI within the first 2 years of age. The majority of children with CIs reached age-appropriate linguistic performance comparable to normal hearing children. Outcomes were significantly related to the age at implantation (earlier providing better outcomes) despite all implantations having been performed within the currently accepted critical period for the therapy of deafness. When variability was compared between the different implantation age groups, only the more demanding HSM speech in noise and language-to-person associations showed a systematically increasing variability with increasing age at implantation. While in other aspects, the variability did not show a systematic trend, the variance in language-to-person association increased with increasing age at implantation (the coefficient of variation tripled with increasing age at implantation). In a recently published study (Busch et al., 2022), the variability of the receptive vocabulary of early bilaterally fitted CI children (< 3 years) showed no significant differences to that of

Figure 5. Coefficient of variation of the data as a function of age. (A) In the speech perception data, the only systematic effect was observed in the Hochmair-Desoyer–Schulz–Moser Sentence Test (HSM) in noise test, where the variation increased with increasing implantation age. (B) In language acquisition tests, only language-to-person association showed a systematic increase in variance with increasing implantation age. Note that the scales of the ordinate axes differ. MST = German-language Freiburg monosyllabic word test.



children with normal hearing (although implantations were not simultaneous).

Sequential implantations involve the issue of introducing an unbalance in the representation of the ears, compromising both speech understanding through the later-implanted ear and binaural fusion (Gordon & Kral, 2019; Illg et al., 2013, 2019; Kral et al., 2013). Therefore, the present cohort was provided with binaural symmetric input that allowed the extraction of more speech and language even in difficult conditions. The present study has the important advantage that all children were almost simultaneously bilaterally equipped with cochlear implants (inter implantation interval < 6 month), and thus had access to some acoustic localization cues. These are especially important in children that are very active, and bilateral implantation provides advantages for these children under difficult hearing conditions (Godar & Litovsky, 2010; Gordon et al., 2015; Litovsky, 2011).

A particular strength of the present study is the additional use of language acquisition tests that also evaluated higher order linguistic functions. Furthermore, the use of age-appropriate testing allowed comparisons of language competence across different developmental stages without the biasing factor of the age at testing. The language acquisition tests used here (German MSVK) offer the possibility of converting raw values into *T* values that were age-normalized and, without a comparison group, and indirect comparison to age-matched normal-hearing individuals.

The present study is the first to use the MSVK test on hearing-impaired children. In a study that tested language developmentally disabled children during 1st grade using the MSVK and math tests, 44% percent of children showed below-average language values and also below-average math skills (Berg, 2015). Compared to this group of children, the data evaluated here are closer to those from normal-hearing children.

Our outcomes are largely consistent with the conclusion of previous studies, in which CI before the end of the 2nd year of life was shown to provide a speech-perception trajectory in quiet that is close to normal-hearing children (Chweya et al., 2021; Hoff et al., 2019; Karltorp et al., 2020; Nicholas & Geers, 2013; Wie et al., 2020). Asynchronous development of the individual sensory systems (as in the case of CI) and their embedding into cognitive processing may cause cognitive scatter in cochlea-implanted children (Kral et al., 2016), and this is minimized by very early implantation. However, since the performance of children implanted before the 2nd year of life is excellent, the improvements of even earlier implantations do not show up with all tests, that is, some tests are likely not sensitive enough. For example, the results of speech understanding in noise show a lower performance with a large scatter, in contrast to the sentence test in quiet or monosyllable word comprehension. Children with CIs generally show the poorest performance in speech-in-noise perception, and the highest signal-to-noise ratio for speech comprehension compared to children with hearing aids, development language disorders, or even normal-hearing children (Torkildsen et al., 2019). Using more demanding language tests, potentially at even older ages, may require the quantification of additional "hidden" phenomena like listening effort (Wild et al., 2012; Winn et al., 2015). When test scores were related (see Figure 4), such as the MST requiring high phonetic competence and the HSM sentence test in noise requiring high cognitive ability, the children whose scores were highest were implanted at an average age of ~10 months. A very similar implantation age was reached when relating MST to the overall *T* value. Karltorp et al. (2020) refer to an implantation age limit of ~9 months for CI to allow following normal linguistic developmental trajectories. Speech development in deaf children with CI is not only influenced by implantation age or etiology (Geers et al., 2016; Niparko et al., 2010), it also has challenges during the development period, which are described around the time of school enrollment (Wie et al., 2020), the age range of the children of our evaluation.

While many cognitive developmental milestones are normally achieved after the implantation age of 2 years, they may still be affected by earlier sensory deprivation ("sleeper effects"; Maurer et al., 2007). Subjective analysis of the data (e.g., by studying the file entries) of children without comorbidities who showed *T* values below 40 revealed three candidate factors potentially influencing outcomes: (a) a different (non-German) native language spoken in the family although the child has German as his mother tongue, (b) low frustration tolerance because of behavioral problems of the child, and (c) a problematic parenting behavior. Since there were only few very low performing children, statistical analysis was not possible. Due to the retrospective design of our study, no cognitive functions (see, e.g., Kronenberger et al., 2014) were systematically tested here and this is a clear limitation of this evaluation. We were also unable to examine the socioeconomic status of the families, which is thought to have an impact on language development.

Implanted prelingually deaf children typically show a pronounced acoustic–phonetic effect. If implanted within the first 2 years, this is early enough for the developmental period of the vocabulary "spurt" (Goldfield & Reznick, 1990; Reznick & Goldfield, 1992; Segbers & Schroeder, 2017) and early enough for the development of syntactic processing (Skeide & Friederici, 2016), but for phonetic development, it may be important to implant as early as possible. Electroencephalographic responses to CI stimulation in prelingually deaf children confirmed a critical period of up to 3.5 years in the early P1 (auditory cortex) component (Sharma et al., 2002, 2005). Corresponding critical periods with cochlear implants were found in the primary auditory cortex of deaf cats (Kral, 2013; Kral et al., 2002, 2019). Along with degraded speech and language competence, auditory discrimination skills were compromised if implantations were performed after the critical period, including reduced electrode discrimination and increased gap-detection threshold (Busby & Clark, 1996, 1999; Rousset et al., 2016; Wei et al., 2007), but also impaired auditory numerosity judgments (Busby et al., 1992). Taken together, all these hints and the present results suggest that in deaf children, it is the acoustic–phonetic aspect of speech that is primarily compromised by deafness. That then affects the acquisition of subsequent developmental steps of speech and language competence. The linguistics literature suggests that phonetic skills in a native language indicate the "brain commitment" to the given language (Kuhl et al., 2008). The outcomes of the MST showed a significant correlation with several of the other language-acquisition tests, particularly sentence comprehension and passive vocabulary. While this may sound rather obvious, since individual aspects of language do not exist in isolation, but are codependent, the fact that the monosyllabic test was at least similarly sensitive to age at implantation as the other tests (that depend on MST) suggests the acoustic/phonetic analysis is the lowest linguistic level affected by deafness in hearing-impaired children. It is therefore likely that it is the phonetic analysis that represents the bottleneck for speech acquisition in deaf children. Only this provides the brain with the inputs that can be used to further build up the linguistic system and provide the fast and automatic analysis that is the base of the low-effort speech listening and comprehension skills (Rönnberg et al., 2019). On the other hand, it was only 25% of variability that these MST and the other measures had in common, suggesting a significant participation of higher level cognitive components in the overall linguistic performance of the child.

This is consistent with the process of normal language acquisition that first builds up phonetic skills, with lexicon developing next and grammar skills developing years after (Kuhl, 2004; Kuhl & Rivera-Gaxiola, 2008). While sensory deprivation delays developmental steps in the cortex (Kral et al., 2005, 2019), the early specialization for phonetics may provide the first critical step that may be missed if implantations are delayed. This is likely reflected in the dependence of most parameters tested here on the age at implantation. Even within the critical period, earlier is better.

The phoneme-level of representation normally develops within the first 8 months of life (review in Werker & Tees, 1992). Thus, the critical period for CI in children likely involves better outcomes in even earlier implantation than the traditional time windows of 18–24 months (Niparko et al., 2010; Wie et al., 2020). First data indeed confirmed an additional benefit of such early implantations (Chweya et al., 2021; Hoff et al., 2019; Karltorp et al., 2020; Lesinski-Schiedat et al., 2006; Nicholas & Geers, 2013). However, complete sensory deprivation is known to delay such developmental processes by delaying, for example, synaptogenesis in deaf auditory cortex (Kral et al., 2005, 2019). This explains why implantation after the age of 8 months can still provide good results.

In the whole group of children, the median language acquisition T values were above or very close to 50 (± 10), suggesting that in this set of tests, the performance of deaf children with early bilateral CI closely approaches that of hearing controls. Again, earlier implantation provided better outcomes in the present study. Current literature demonstrates that the development of phonological and lexical knowledge, phonological working memory, and regular language are related (Rodrigues & Befi-Lopes, 2009). Further research should also examine the working memory in young children to identify other factors of variability in language development.

Our data also document that special etiology, like inner ear malformation, meningitis, CMV, Noonan or Waardenburg syndrome, auditory neuropathy, or comorbidities like motor skills, short bowel syndrome, balance problems, and visual impairment without suspected cognitive impact had no significant influence on language development in the present group of subjects. This is a promising result, recommending the implantation of deaf children with comparable etiology or comorbidities. On average, their results were comparable to those of other children (children with CI and even normal-hearing peers). Certainly, the prognosis of these children must be considered on a case-by-case basis, as there are also children with these etiologies who show poorer outcomes in hearing and language development (Bolduc et al., 2021; Chweya et al., 2021; Ciavarro et al., 2022; Daneshi et al., 2020; Fan et al., 2022; Helmstaedter et al., 2018; Hoff et al., 2019; Karltorp et al., 2020; Lesinski-Schiedat et al., 2006; Nicholas & Geers, 2013).

Conclusions

Congenitally deaf children fitted with CI bilaterally in the 1st year of life developed age-appropriate language skills when tested at 5–7 years, whereas the chances for bilaterally congenitally deaf children who were fitted with a bilateral CI in the 2nd year of life were lower. On average, at 5–7 years of age, all implantation age groups achieved a language development comparable to that of their normal-hearing peers. The outcome variability increased with implantation age for sentences in noise and language-to-person associations.

Data Availability Statement

The data table on which this study is based can be viewed at the MHH ENT link.

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