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Speech Comprehension in Children and Adolescents After Sequential Bilateral Cochlear Implantation With Long Interimplant Interval

Angelika Illg, Alexandros Giourgas, Andrej Kral, Andreas Büchner, Anke Lesinski-Schiedat, and Thomas Lenarz

Department of Otolaryngology, Hannover Medical School, Hannover, Germany

Objective: Identify likely predictors for the outcome after contralateral cochlear implantation with a long interimplant delay.
Study Design: Retrospective case reviews.
Setting: Outpatient cochlear implant (CI) center.
Patients: Seventy-three children and adolescents who underwent sequential bilateral cochlear implantation with an interval between both implantations of 5 years or longer. The mean age of the patients at the first and second cochlear implantations was 2.72 ± 1.52 and 11.57 ± 2.9 years, respectively. The mean duration of experience with both implants was 4.01 ± 1.57 years.
Intervention: Rehabilitative.
Main Outcome Measures: All 73 patients underwent periodic speech perception testing in quiet and noise. The most recent unilateral data for each side were statistically analyzed. The speech test results were evaluated by the age at first implantation, the interval between both implantations, the duration of hearing aid use in the second side, and the duration of the bilateral CI use.
Results: A statistically significant difference for speech test results was obtained between the early-implanted ears and the late-implanted ears for all children. These results were dependent on the interimplant interval. All age groups demonstrated significant differences ($p > 0.05$) for the second side between the speech test results and the interval between both cochlear implantations. In addition, statistically significant differences influenced by the duration of hearing aid use were found for speech test results for the second side. Experience was also a factor for the second CI, yielding significantly higher speech test scores with longer use.
Conclusion: The development of hearing abilities in a second-implanted side depends on the interimplant interval, the hearing aid use, and the duration of the second CI use.
Key Words: Adolescents—Bimodal—Children—Cochlear implant—Interimplant interval—Sequential bilateral.

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In our clinic, most children receive 2 cochlear implants (CIs) within 1 surgical procedure, although some years ago, children were implanted predominantly in 1 ear only. As adolescents, many of those unilateral CI users requested a second, contralateral CI. The expectation for some of these patients and their parents was that the second side would deliver comparable results to those experienced from the first implanted side. This is, however, not always the case.

The age at which children receive a CI is one of the strongest predictors of hearing and speech skills after cochlear implantation (1–4). Long-term studies have also shown that performance improves slowly over time and requires years to approach the final outcome (5,6). This long-term progress likely reflects the complexity in the development of the auditory system that is largely dependent on auditory input. For example, during the first 2 to 4 years after implantation, a massive reorganization of dendritic branches in the cerebral cortex occurs (7). The cortical development continues until adulthood (8), with extensive developmental changes both at the cellular and at the microcircuitry levels (9). Absence of hearing experience in animal experiments alters these developmental processes extensively and demonstrates their dependence on auditory input (10).

Investigations in children with unilateral CIs demonstrate that auditory input provided by a CI has a

Address correspondence and reprint requests to Angelika Illg, Ph.D., HNO-Klinik der MHH – Deutsches HörZentrum Karl-Wiechert-Allee 3 30625 Hannover; E-mail: illg@hoerzentrum-hannover.de
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modifying influence on the auditory system and induces auditory maturation (10); however, at an implantation age of 6.5 to 7.0 years, the developmental maturation is less likely to be initiated. Sharma et al. (11) assumed that a second, 'late' implant would stimulate cortical areas but that neither would have normal connections within the cortical layers or to higher-order auditory and language areas and, thus, possibly lead to inferior outcomes for the later-implanted ear. Nonetheless, speech recognition improves in children who undergo sequential bilateral cochlear implantation. Systematic studies on the effects of stimulation of the second ear are rare and include only limited numbers of individuals (12,13).

Steffens et al. (14) described the effect of the binaural advantages after sequential bilateral cochlear implantation. The authors emphasized that both those children with high- and low-level monaural speech recognition scores for the first implanted ear had significant binaural advantage after the second implantation. Other studies (15–25) describe similar results in lateralization or for speech recognition in quiet or noise (26). More data on the effectiveness of a second CI as a function of the time delay between implantations are required to determine the optimal schedule for cochlear implantations in the second ear (26).

Studies in patients using both a CI and a hearing aid (HA; bimodal listening) report improved localization or speech recognition in the bimodal condition (27–30). The influence of bimodal hearing on speech comprehension after a second CI has not been described in detail in the literature to date (28,31).

The aim of our study was to find predictors for the outcome after additional cochlear implantation in the contralateral side with a long interimplant delay. For this purpose, the data of 73 young patients were analyzed according to the following possible predictors: 1) age of first implantation, 2) interimplant interval, 3) duration of HA use for the second side, and 4) duration of bilateral CI use.

MATERIALS AND METHODS

Patients

Seventy-three children and adolescents who underwent sequential bilateral CI in our clinic between 2003 and 2010 were included in the retrospective study. The minimum interimplant interval was 5 years. The implantations in the first side were performed between 1995 and 2005. Mean age at the first CI intervention was 2.72 ± 1.52 and that at the second CI was 11.57 ± 2.9 years. The patients were required to have experience with the second CI for at least 1 year. The mean duration of experience with the first implant was 12.86 ± 1.99 years and the mean experience with both implants was 4.01 ± 1.57 years.

It is standard procedure and care at our clinic to provide at least a 3- to 6-month HA trial before implantation. The decision to proceed with CI surgery was delayed for 8 of the 73 children accepted into this study because they had gained additional useful hearing benefit to develop auditory-verbal communication skills. Consequently, their parents chose to postpone their decision for a second implant until the initial improvements in speech and language development ceased. All others received a CI after it was clear that HAs provided little to no useful hearing that would support further speech and language development. All children were diagnosed with profound hearing loss below the age of 2. Significant additional cognitive handicaps were not observed among the group of children.

The cause of deafness for the 73 subjects was unknown (n = 59), genetic (n = 6), cytomegalovirus (n = 2), ototoxic influence (n = 1), otitis media (n = 1), Waardenburg syndrome (n = 2), enlarged vestibular aqueduct syndrome (n = 1), and meningomyolocele (n = 1). All patients received multichannel CIs from the companies Cochlear (Sydney, Australia) or Advanced Bionics (Valencia, California).

Speech Comprehension Tests

All 73 patients were periodically tested using the FET (the German Freiburger Monosyllabic Word test) in quiet and the HSM (the German Hochmair-Desoyer, Schulz, Moser Sentence Test) in quiet and in noise (10 dB S/N ratio, S0N0). All tests were performed in free field at 65 dB sound pressure level, and each side was tested separately. The most recent data for each implanted side were evaluated.

Evaluation and Statistics

The speech test results were evaluated according to 4 predictors: the age at first implantation (P1), the interval between both implantations (P2), the duration of HA experience in the second side (P3), and the duration of the bilateral CI use (P4). To evaluate different dependencies, we divided the patients into different subgroups of relatively equivalent group sizes for each predictor (P1–P4) category.

To evaluate Predictor 1, data for all patients were first divided into 3 age groups: P1G1, P1G2, and P1G3. The age groups show the age ranges of the first implantation in years (Table 1). In addition, the 3 age groups were further subdivided into interimplant interval groups. As such, every age group was divided into 3 subgroups: age group P1G1 was divided into P1G1a, P1G1b, and P1G1c; age group P1G2 was divided into P1G2a, P1G2b, and P1G2c; and age group P1G3 was divided into P1G3a, P1G3b, and P1G3c (Table 2).

To evaluate Predictor 2, data of all patients were divided into 3 interimplant interval groups: P2G1, P2G2, and P2G3. The interimplant interval shows the duration of the interval between both implantations in years (Table 3).

To evaluate Predictor 3, data of all patients were first divided into 3 subgroups according to the duration (years) of the HA experience in the second side before sequential implant. These groups are P3G1, P3G2, and P3G3 (Table 4). In addition, data were analyzed to determine the effect of the duration of unilateral hearing. Therefore, from all 73 patients, the difference between the interimplant interval and the bimodal (CI + HA) time was calculated.

To evaluate Predictor 4, data of all patients were divided into 3 subgroups according to the duration of the bilateral CI use in years. These groups are P4G1, P4G2, and P4G3 (Table 5).

TABLE 1. *Evaluation groups for Predictor 1: Age at first implantation*

Predictor	Age Group 1 (G1)	Age Group 2 (G2)	Age Group 3 (G3)
Age at first implant (P1)	**P1G1**	**P1G2**	**P1G3**
	1–2 yr n = 25	2–3 yr n = 27	3–9 yr n = 21

TABLE 2. *Combined groups for Predictors 1 and 2: Age at first implantation and interval between both implantations*

Predictor	Age Group 1 (G1)			Age Group 2 (G2)			Age Group 3 (G3)		
Age at first implant (P1)	**P1G1** 1–2 yr n = 25			**P1G2** 2–3 yr n = 27			**P1G3** 3–9 yr n = 21		
Groups combined with P2 interimplant interval	**P1G1a**	**P1G1b**	**P1G1c**	**P1G2a**	**P1G2b**	**P1G2c**	**P1G3a**	**P1G3b**	**P1G3c**
	>5–7 yr n = 7	>7–9 yr n = 8	>9–15 yr n = 10	>5–7 yr n = 6	>7–9 yr n = 7	>9–15 yr n = 14	>5–7 yr n = 6	>7–9 yr n = 6	>9–15 yr n = 9

All data were analyzed statistically using IBM SPSS Statistics, version 19. The tests performed were one-factor analysis of variance (ANOVA) and then the Scheffé post hoc test to compare the means of the different subgroups by evaluating each predictor (P1–P4). The Wilcoxon test followed by Bonferroni correction was used to statistically compare the mean of the data between the first and second side by evaluating the first predictor (P1).

To correlate 2 variables, Spearman ρ correlation coefficient was applied (32). Statistical significance was set at $p < 0.05$ (*$p < 0.05$, **$p < 0.01$, ***$p < 0.001$).

RESULTS

Speech Comprehension Based on Age Groups at the Time of First Implantation

The age-dependent category for first implantation (P1) was divided into 3 groups: 1 to 2, 2 to 3, and 3 to 9 years old (Table 1). In each age group, improvement in speech comprehension for the first side and lower performance for the second, HA side ($p < 0.001$) was observed, regardless of the test used (Fig. 1).

No significant differences were found between the first side and the bilateral mode in quiet. In the bilateral mode, the HSM results in noise were significantly better than those from the first side tested but only for Group P1G2 with children who received their first implant at an age between 2 and 3 years ($p = 0.039$).

Comparison between the scores obtained for bilateral monosyllabic words between the 3 different age groups revealed statistically significant differences for Groups P1G1 and P1G2 ($p = 0.047$) and Groups P1G2 and P1G3 ($p = 0.008$); no other comparisons were significantly different. The test results for monosyllable words and sentences in quiet showed that the youngest age group achieved the highest results and then the second and third groups. Speech performance on HSM sentences in noise was similar for all 3 groups.

Speech Comprehension Based on the Interimplant Interval

The groups were subsequently ordered by the interimplant interval (Table 3) divided into intervals of 5 to 7, 7 to 9, and 9 to 15 years between implantations. Using ANOVA and then a post hoc test (Scheffé procedure), the speech test scores in quiet for the second side with the monosyllabic word test and the HSM sentence test showed statistically significant differences with respect to the duration between implantations in all 3 groups (Fig. 2A).

Monosyllabic word recognition scores were significantly higher in the first group (P2G1, the 5- to 7-yr interval) who had the shortest interimplant interval when compared to the third group with the longest interval ($p = 0.001$). The results of the HSM test showed significant differences ($p = 0.015$) between the first and second groups (P2G2), as well as between the first and third groups ($p < 0.001$). No statistical difference was revealed between the second (P2G2) and third group (P2G3). A trend analysis between the first and the second side for the monosyllabic words showed that the difference became greater with increasing interimplant interval; for example, test results for the first and second sides in patients with interimplant intervals of 9 to 15 years were extremely different (Fig. 2B). The monosyllabic word test results for the second side of each individual patient showed a negative correlation to the interval length between both implantations using a linear trend line with the Spearman ρ correlation coefficient ($r = -0.505$; Fig. 2C).

Speech Comprehension Based on the Interimplant Interval and the Age at First Implantation

The 2 predictors of age (P1) and interval between implants (P2) were compared. The 25 children and adolescents receiving their first implant at the ages of 1 to 2 years were sorted into 3 groups by the interimplant intervals (Groups P1G1a, P1G1b, and P1G1c; Table 2).

TABLE 3. *Evaluation groups for Predictor 2: Interval between both implantations*

Predictor	Interval Group 1 (G1)	Interval Group 2 (G2)	Interval Group 3 (G3)
Cochlear implantation interval (P2)	**P2G1** >5–7 yr n = 19	**P2G2** >7–9 yr n = 21	**P2G3** >9–15 yr n = 33

TABLE 4. *Evaluation groups for Predictor 3: Duration of hearing aid (HA) experience in the second side*

Predictor	HA Group 1 (G1)	HA Group 2 (G2)	HA Group 3 (G3)
HA usage (P3)	**P3G1** 0–12 mo n = 30	**P3G2** 13 mo–36 mo n = 27	**P3G3** >3–16 yr n = 16

TABLE 5. *Evaluation groups for Predictor 4: Duration of the bilateral cochlear implant (BICI) use*

Predictor	BICI Group 1 (G1)	BICI Group 2 (G2)	BICI Group 3 (G3)
Second CI duration (P4)	**P4G1** 1–3 yr n = 19	**P4G2** 3–5 yr n = 31	**P4G3** 5–9 yr n = 23

Although statistically not significant, there was a tendency for the scores for monosyllable words and sentences in quiet in the second side to decrease nominally with increasing interval between implantations (Fig. 3A). Test results for the 27 subjects (Groups P1G2a, P1G2b, and P1G2c; Table 2) receiving their first implant between the ages of 2 and 3 years, however, showed significant differences between the groups and similar trends (Fig. 3B). For the monosyllable word test, significant differences between groups with the smallest and largest intervals (p = 0.003) were demonstrated. Results of the sentence test revealed significant differences (p = 0.031) between the smallest interval group and the other 2 longer-interval groups. Findings in the 21 cases receiving their first implant after the age of 3 years (Groups P1G3a, P1G3b, and P1G3c; Table 2) again showed no statistically significant differences, but a similar trend in the data for poorer scores with increasing interval (Fig. 3C).

Speech Comprehension Based on Duration of HA Use in the Second Side

Traditionally, patients in Germany were encouraged to continue HA use in their nonimplanted, contralateral ear. Many of the children and adolescents continued to wear their HAs before their second implantation, although the residual hearing provided them with only minimal auditory cues. All 73 children and adolescents had continued HA use (P3) after unilateral implantation. They were divided into 3 groups: short (>1 yr), 1 to 3 years, and 3 to 16 years of experience (see Table 4 for P3, Groups 1–3).

No statistically significant differences were found between the 3 groups for mean monosyllable word test scores in the second CI: 19.23%, 24.25% and 33.1% for Groups P3G1, P3G2, and P3G3, respectively. The results of the HSM in quiet and noise were comparable. If the HAs were used 0 to 1 year, the mean was 15.36% in quiet and 0.94% in noise. If the HAs were worn 1 to 3 years, the HSM results were 24.5% in quiet and 0.73% in noise. However, if the HAs were worn 3 to 16 years, the results were 41.23% in quiet and 8.19% in noise, yielding a significant difference in scores in the HSM between the short and long duration of HA use (p = 0.033). The sentence test in noise scores also resulted in significant differences between the shortest (P3G1) and longest (P3G3) duration of HA use (p = 0.024) and between the 1- to 3-year group (P3G2) and the longest (P3G3) use group (p = 0.018). Depending on the duration of the bimodal time, the score for speech comprehension increased. On average, the patients of Group P3G1 spent 4.7% of the interimplant delay with HA use for bimodal listening; it was 18.5% for P3G2 and 78.9% for P3G3.

To analyze the additional effect of duration of unilateral hearing only with the first CI alone, the difference was calculated between the interimplant interval and the bimodal use. The data were sorted into 3 groups depending on the duration of unilateral use: 0 to 5 years (n = 18), 5 to 8 years (n = 27), and 8 to 16 years (n = 28). The results of the 3 tests (FET, HSM in quiet, and HSM in noise) show higher scores for the group with the shortest unilateral duration (0–5 yr; Fig. 4) in the second CI. Significant differences were found between the speech comprehension scores of the shortest (0–5 yr) and longest (8–16 yr) group.

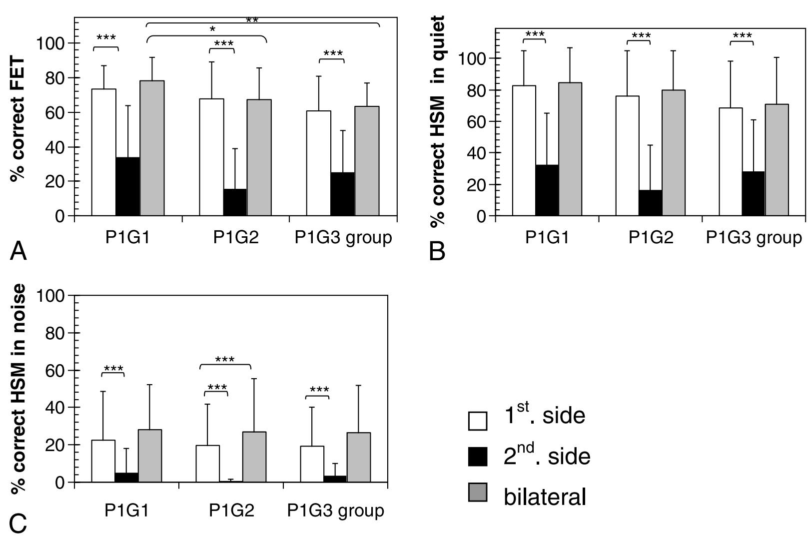

FIG. 1. Predictor 1 (P1): Speech comprehension based on age groups at the time of first implantation (P1G1, P1G2, P1G3) (Table 1): (*A*) results of the FET monosyllable word test, (*B*) results of the HSM sentence test in quiet, and (*C*) results of the HSM sentence test in noise.

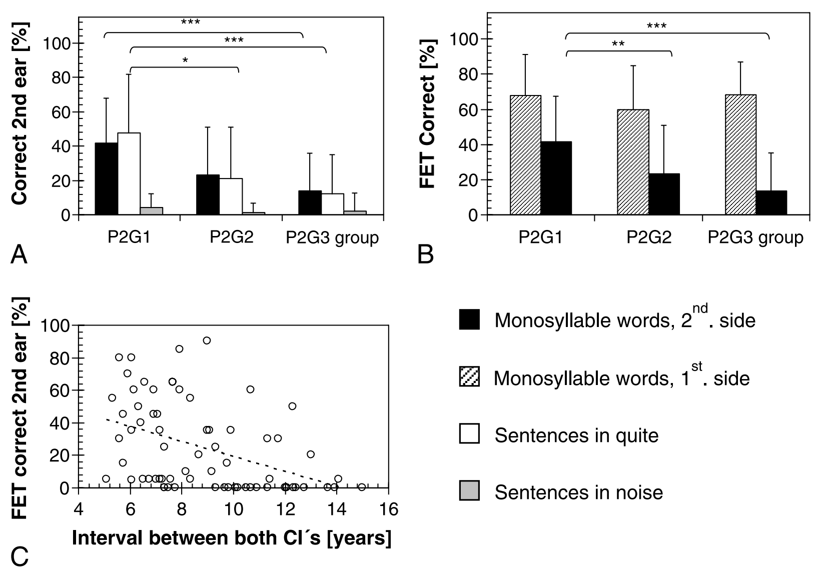

FIG. 2. Predictor (P2): Speech comprehension based on the interimplant interval (P2G1, P2G2, and P2G3) (Table 2): (*A*) results of the 3 speech tests for the second side, (*B*) results for the FET monosyllable word test from the first and second sides, and (*C*) results of the monosyllable word test from the second side for each patient and the linear trend line.

Speech Comprehension Based on Duration of Bilateral CI Experience

To evaluate test results based on duration of bilateral implant (P4), the 73 patients were again divided into 3 relevant groups: P4G1, 1 to 3 years; P4G2, 3 to 5 years; and P4G3, 5 to 9 years of bilateral experience (Table 5). Mean scores for the monosyllable word test were 11.11%, 25.68%, and 33.64%, respectively (Fig. 5). Mean scores for the HSM sentence test in quiet indicated the same trend. Results of the HSM in noise were not statistically significant at 3.03% (P4G1), 1.43%, (P4G2), and 3.82% (P4G3). One-factor ANOVA and then the Scheffé post hoc test revealed significant differences between shortest and longest duration for FET and HSM ($p = 0.029$ and $p = 0.013$).

DISCUSSION

Our results agree with the general finding for children with an interimplant interval longer than 5 years that speech perception scores obtained with the second implant are

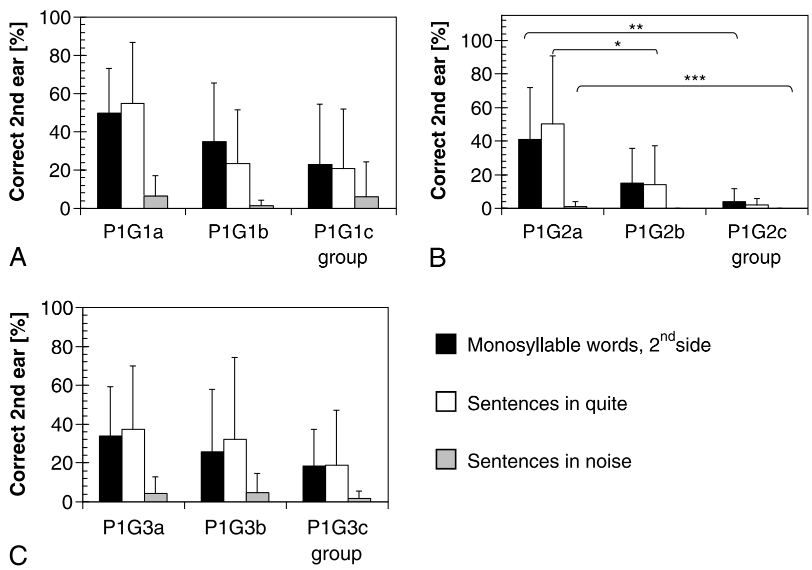

FIG. 3. Combined Predictor 1 and 2 (P1/P2): Speech comprehension based on the age at first implantation and the interimplant interval (Table 3): (*A*) results for age group P1G1 ordered by the interimplant interval, (*B*) results for age group P1G2 ordered by the interimplant interval, and (*C*) results for age group P1G2 ordered by the interimplant interval.

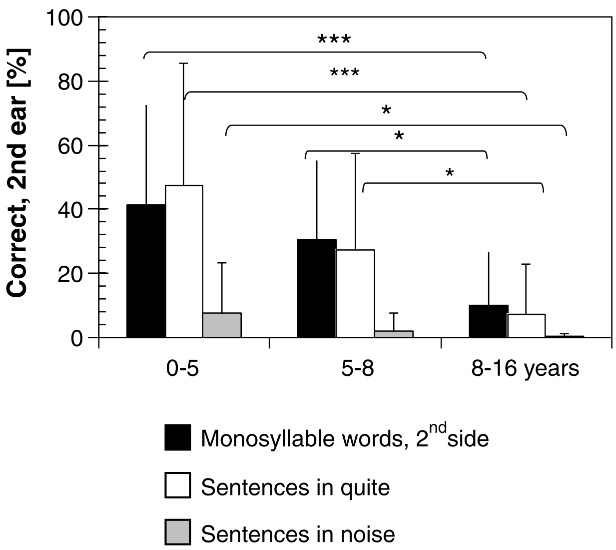

FIG. 4. Speech comprehension based on the duration of unilateral CI experience (unilateral time in years = interimplant interval minus bimodal interval).

poorer than the performance for the first implanted side (see our data, Fig. 1) (14,31). Speech comprehension in the first side may be influenced by the high degree of plasticity in central auditory pathways in early childhood and the ''sensitive period,'' a topic frequently described by different authors (1,2,9–11). Correspondingly, in the present study, results in speech comprehension tend to be poorer with increasing age at implantation when considering results obtained relative to the first implant alone. Speech test results for the first 2 P1 age groups (P1G1 and P1G2) are slightly better than in the third, older age group (P1G3). Although most children received HAs before first implantation, the speech perception results in the older group (P1G3) are poorer than in the younger age groups (P1G1 and P1G2). Also, with respect to those children who developed some degree of language skills after receiving HAs before, the younger implanted children showed higher speech comprehension results, which again speaks for early implantation.

Interestingly, in the present study, the speech test results of the second side show no relation (Fig. 1) between speech comprehension in quiet and age at the time of first-side implantation in contrast to findings reported by Zeitler et al. (19), a rare study in which data of first implant were available for comparison.

Instead, we see that results achieved by the second side are affected by the duration of the interimplant interval. Provided that areas of the afferent auditory system are differentially driven by both ears, ongoing deprivation for the second ear may have degenerative ear-specific consequences. Therefore, speech comprehension in noise may decrease in children and adolescents with increasing interimplant interval (Fig. 2). Even after longer experience with the second implant, the scores of the speech tests were very poor and no significant increase of scores over time were demonstrated.

Despite the differences between the 2 sides, all of these 73 young patients continue to wear both implants. The small input from the second implant does not interfere with the better results of the first side; rather it assists the bilateral hearing mode (Fig. 1). We observed no detrimental effects from wearing both implants as was reported by Ramsden et al. (33).

Not only can the interimplant interval between both implantations serve as a predictor for the degree of speech comprehension but also the usage of HAs before second implantation seems to influence speech understanding. Although most of our patients had minimal residual hearing, results demonstrate increasingly higher test scores if patients wore their HAs for a longer period. Further, if we compare the time of unilateral hearing with the speech test results after second implantation, the scores in the second CI are better after a short unilateral time (Fig. 4). This suggests that even minimal auditory input will have a positive effect on maturation and survival of the afferent neural activity (20).

Furthermore, a positive effect on speech comprehension with decreasing interimplant interval was clearly demonstrated. The speech perception scores of patients with the longest interimplant interval (9–15 yr) were significantly poorer than for the other 2 groups with shorter intervals (Figs. 2A, C). Although the mean experience with the second implant, especially in the third group (9- to 15-yr interval), was shorter by nearly 2 years than in the other groups, the speech comprehension scores for the second group with the longest, second mean implant experience were significantly poorer than the comprehension of the first group with similar mean experience. Therefore, the question arises for P2G3 as to whether the patients

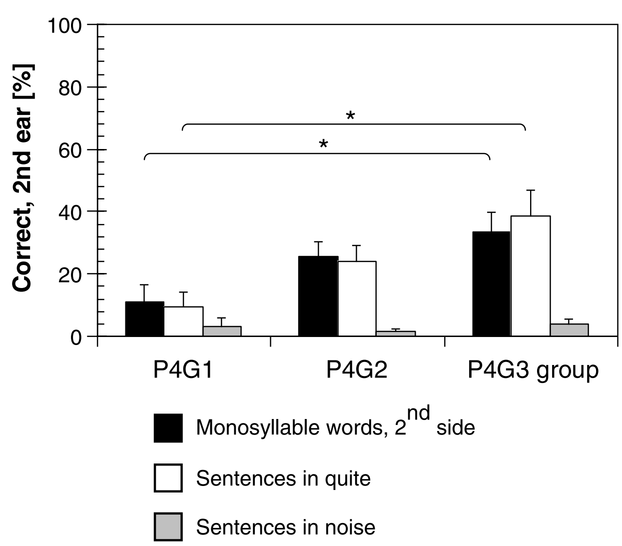

FIG. 5. Predictor 4 (P4): Speech comprehension based on the duration of bilateral CI experience (P4G1, P4G2, and P4G3) (Table 5).

would have performed better if they had had longer experience similar to the patients in Groups P2G1 and P2G2. The clearly significant relationship between speech performance and duration of interimplant interval leads to the conclusion that the speech comprehension of the group with the longest interimplant interval and even with a longer period of experience would perform no better than the patients in the group with shorter intervals.

Other authors (13,18) propose that interimplant intervals have a large influence on hearing and speech understanding but this could not be substantiated because of their small numbers of subjects and because there were only children with short interimplant intervals in those studies. Wolfe et al. (2007 [20]) concluded that an improvement in speech recognition seems to be possible for children who received their first CI before the age of 3 years and a second CI by the age of 10 years at the latest. This implies that the longest interimplant interval should be 7 years, which is in line with our results that also indicated a significant performance disadvantage if the interimplant interval was 7 years or longer (Fig. 2): between the Groups P2G2 and P2G3 years, no significant difference was indicated. Also, it should be noted that the difference between the outcome of the first and second side becomes even more apparent if the interimplant interval increases (Fig. 2B).

Further, in the combined evaluation of the speech perception scores based on the age at first implantation and the interimplant interval, a positive relation between speech comprehension and the interimplant interval was found and statistically verified (Fig. 3), thus confirming the importance of the predictor (P2) ''interimplant interval.''

Gordon and Papsin (12) also indicated that, for sequentially implanted children with short interimplant interval (<2 yr), interaural performance differences were observed on speech comprehension tests; however, the differences were not statistically significant. The results of our study indicated statistical significant differences between the speech comprehension of the first and second side given our large sample of patients and their more extreme interimplant intervals. Therefore, our analysis confirms the hypothesis of Gordon and Papsin that auditory deprivation proceeds in the contralateral side after unilateral cochlear implantation. The prevention of auditory deprivation may only be possible through simultaneous cochlear implantation or as a result of taking advantage of residual hearing by wearing HAs.

CONCLUSION

Both predictors of the age at first implantation (P1) and interimplant interval (P2) are interdependent. The interimplant interval is influenced by a third predictor, the use of a HA before second implantation (P3). Therefore, all patients should be encouraged to continue wearing their contralateral HA to maintain afferent neural activity. The fourth predictor is probably related to a learning effect associated with cognitive development. Longer experience (P4) with the second CI leads to significantly better scores as demonstrated by the subjects with 5 years or more of bilateral experience whose scores for both monosyllabic words and sentences in quiet were higher (second side; Fig. 5). All patients should be encouraged to intensively practice using their listening skills with the second CI alone and in combination with the first CI.

During the second implantation phase, most of our young patients underwent a difficult phase of life while growing up. Usually, they expected auditory information from their second CI to be similar to their first one. In addition, they often were not sufficiently motivated to practice suggested hearing and listening skills in their own time. Expectations and motivation are unbalanced in the time of decision for the second CI. It is important to consider these psychosocial effects during preoperative consultations and postoperative rehabilitation. The associated learning process to adapt to and use the additional auditory information after receiving a second implant, especially for those with a long interimplant interval, does not seem to be as easy as in early childhood, but our data show that improvement is possible.

Further research on the learning effects and interimplant intervals will certainly contribute to the discussion and should enable a better understanding of the continuous reorganization of the central auditory pathway.

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