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Original article

Sequential bilateral cochlear implants in children and adolescents: Outcomes and prognostic factors

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ABSTRACT

Objectives: Cochlear implants (CI) have enabled an increasing proportion of deaf children to develop oral communication. Despite the well-known benefits of bilateral implantation, many of these children and teenagers have only a unilateral implant. The aim of this study was to evaluate the benefits of sequential bilateral CI and the influence of relevant factors on outcome.
Material and methods: A single-center retrospective study included 109 children and adolescents who received a second sequential CI between 2008 and 2016. Subjects were evaluated before sequential implantation and subsequently at 3, 12 and 24 months, on Speech Intelligibility Rating and speech perception tests: Categories of Auditory Performance, word and sentence recognition in silence and in noise. The influence of inter-implant interval and performance with the first CI were analyzed.
Results: In the majority of patients, sequential CI provided significant improvement in speech and intelligibility perception. These benefits were seen not only for short but also for long inter-implant intervals. Some subjects with poor performance with their first implant showed significant progression after sequential bilateral implantation.
Conclusion: In view of the benefits of sequential bilateral CI, we suggest that a second CI should be proposed to all unilaterally implanted children and adolescents, regardless of inter-implant interval and initial performance with the first CI. Further studies need to be conducted to identify prognostic factors for success in sequential contralateral implantation.

1. Introduction

Hearing loss is the most common sensory disability in children worldwide. The development of cochlear implants (CI) since the late 1970s has enabled a growing proportion of deaf children to acquire oral communication and language, improving social integration.

CI indications are in constant progression. They were defined in an international consensus statement in 1995, then again in 2007. The French Health Authority (HAS) updated them in 2011 [1], notably as concerns bilateral implantation. The issue of a second CI mainly concerns patients with profound bilateral hearing loss who received unilateral CI before 2011, but also patients with asymmetric hearing loss with CI in the profoundly impaired ear and contralateral aggravation.

Bilateral CI improves perception of speech in noise [2,3] and sound localization [4] in some cases, enhancing quality of life [5].

Post-implantation linguistic performance varies according to internal and external factors [6]. Due to wide inter-subject variation in results, expectations also vary. Thus, to be able to inform patients and families of expected impact, many studies have sought to analyze prognostic factors for sequential implantation: hearing loss duration and etiology [6], age at implantation, and interval between the 2 implantations [7].

The present study aimed to assess the benefit of a sequential second CI in a patient cohort, and to identify factors influencing outcome.

2. Material and methods

A single-center retrospective study included 109 patients (51 girls, 58 boys) with severe to profound bilateral sensorineural hearing loss receiving sequential bilateral CI between 2008 and 2016. Inclusion criteria comprised: age < 20 years at first CI (CI-1), and minimum 12 years' follow-up after second CI (CI-2).

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Table 1
Hearing loss etiologies.

	n (%)
Connexin gene mutation	25 (22.9)
Usher's syndrome	13 (11.9)
Waardenburg's syndrome	7 (6.4)
CMV fetopathy	6 (5.5)
Pendred's syndrome	3 (2.8)
Post-meningitis	3 (2.8)
Mitochondrial	3 (2.8)
Fetopathy	1 (0.9)
Otoferlin gene mutation	1 (0.9)
Unknown/negative genetic analysis	47 (43.1)

Table 2
Demographic characteristics.

	Mean (years)	Range (years)	Median (years)
Age at CI-1	3.4	0.6–19.2	2.1
Age at CI-2	7.2	1.2–23.6	5.4
Inter-CI interval	3.8	0.2–18.1	2.8

Tables 1 and 2 show etiologies and demographic data.

On June 1st, 2016, mean follow-up after CI-2 was 42.7 months (range, 12–100 months; median, 39.6 months). During the interval between CI-1 and CI-2, 101 patients (92.7%) wore a hearing aid (HA) in the non-implanted ear, with mean hearing thresholds of 78.3 dB with and 110.9 dB without HA.

Throughout post-CI follow-up, speech perception was assessed on the Categories of Auditory Performance (CAP) scale, from 0 to 7, and on open word recognition lists at 60 dB in silence (Saussus and Boorsma, Fournier 2-syllable lists) and sentence recognition lists at 60 dB in silence and in noise and with a signal-to-noise ratio of +6 dB in noise (lists for young children, HINT, MBBA). Correct responses were counted and reported as percentages. Speech intelligibility was assessed on the Speech Intelligibility Rating (SIR) scale, from 1 to 5.

Speech therapy data were recorded between 2 weeks and 6 months before CI-2, with CI-1 alone, and 3, 12 and 24 months after CI-2, using both implants. Post-CI-2 results with both implants at M3, M12 and M24 were compared versus pre-CI-2 results with CI-1 alone.

The impact of the CI-1 to CI-2 (inter-CI) interval was analyzed, as was the impact of CI-1 performance on performance after sequential implantation (post-CI-2).

Patients were divided into 2 groups according to inter-CI interval: < 3 years (Early Group), versus ≥ 3 years (Late Group); and also into 2 groups according to initial performance (with CI-1 only): word recognition < 85% (Weak Group), versus ≥ 85% (Strong Group).

Inter-group thresholds were set empirically to balance numbers and facilitate statistical analysis.

Statistical analysis comprised first descriptive analysis of qualitative variables expressed as percentages and quantitative variables as means and medians. Individual assessment used matched Wilcoxon signed ranks test. The significance threshold was set at $P < 0.05$. Tests were run on R 3.2.5 software by the center's biostatistics department.

3. Results

3.1. Benefit of second CI

On the CAP scale, 28.1% of patients showed improvement at 3 months post-CI-2, 47% at 12 months, and 51.9% at 24 months. Progression in CAP score between CI-1 and M3, M12 and M24 post-CI-2 was significant ($P < 0.05$).

Table 3
Demographic characteristics according to early (E) or late (L) second CI.

n	Group E 62	Group L 47	*P*
Percentage	56.8%	43.2%	
Mean age at CI-1	3.5	3.2	0.106
Mean age at CI-2	5.3	9.7	< 0.001
Mean inter-CI interval	1.8	6.5	< 0.001

On the SIR scale, 33.7% of patients showed improvement at 3 months, 45.4% at 12 months, and 52.6% at 24 months. Progression was significant ($P < 0.05$).

On word recognition, 47.4% of patients showed improvement at 3 months, 50.8% at 12 months, and 55% at 24 months. Progression was significant ($P < 0.05$).

On sentence recognition in silence, 66.6% of patients showed improvement at 3 months, 61.2% at 12 months, and 60.6% at 24 months. Again, progression was significant ($P < 0.05$).

Progression on sentence recognition in noise, on the other hand, was not significant: $P = 0.55$ at M3, $P = 0.4$ at M12 and $P = 0.05$ at M24. The small number of patients able to be assessed on the more demanding sentence recognition in noise test led to lack of statistical power.

3.2. Impact of inter-CI interval

Demographic data for the Early and Late groups are shown in Table 3. Mean age at CI-1 did not differ significantly ($P = 0.106$): i.e., the 2 groups were comparable independently of age.

In the Early group, CAP score improved in 44.4% of patients at M3, 72.4% at M12 and 76.1% at M24; progression was significant ($P < 0.05$). In the Late group, progression was not significant at M3 ($P = 1$) or M12 ($P = 0.06$) but was significant at M24 ($P < 0.05$) (Fig. 1a).

In the Early group, SIR score improved in 49.1% of patients at M3, 63.0% at M12 and 72.1% at M24. In the Late group, SIR score improved in 14.3% of patients at M3, 23.3% at M12 and 27.3% at M24. Improvement was significant in both groups at M3, M12 and M24 ($P < 0.05$) (Fig. 1b).

Word recognition improved in 50% of patients in the Late group and 60% of patients in the Early group at M24. Progression was significant at M3, M12 and M24 ($P < 0.05$) in both groups (Fig. 1c).

Sentence recognition in silence improved in 73.6% of patients in the Early group at M3, 66.7% at M12 and 80% at M24. In the Late group, rates were 60.9% at M3, 57.1% at M12 and 44.4% at M24. Progression was significant in the Early group ($P < 0.05$) at all 3 time points, but only at M3 in the Late group (Fig. 1d).

No significant differences in sentence recognition in noise emerged for either group, due to small sample sizes on this test.

3.3. Impact of initial (CI-1) performance

Demographic data for the Strong and Weak groups are shown in Table 4. Mean ages at CI-1 and CI-2 did not differ significantly

Table 4
Demographic characteristics according to weak (W) or strong (S) performance with CI-1.

n	Group W 29	Group S 34	*P*
Percentage	46%	54%	
Mean age at CI-1	5	3.5	0.499
Mean age at CI-2	8.7	8.5	0.448
Mean inter-CI interval	3.7	5	0.029

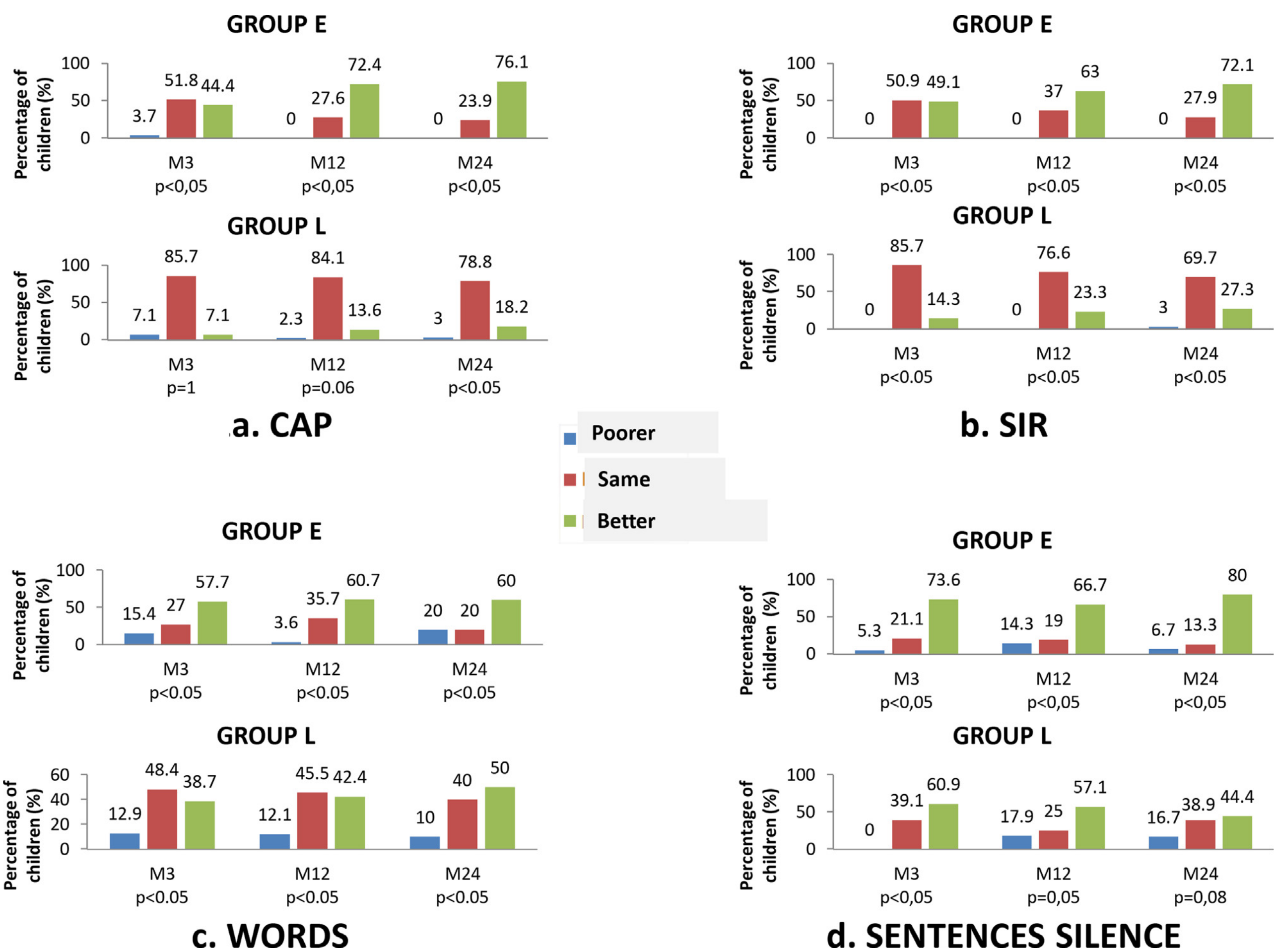

Fig. 1. CAP (a), SIR (b), Words (c) and Sentences in Silence (d) scores post-CI-2 in Early (E) and Late (L) inter-CI interval groups.

between groups: i.e., the 2 groups were comparable independently of age.

In the Strong group, word recognition improved in 18.7% of patients at M3, 18.2% at M12 and 25% at M24. Progression was not significant: $P = 0.2$ at M3, $P = 0.9$ at M12 and $P = 1$ at M24.

In the Weak group, word recognition improved in 84% of patients at M3, 89.3% at M12 and 85% at M24. Progression was significant ($P < 0.05$) (Fig. 2).

4. Discussion

Analysis of the present data showed that sequential contralateral cochlear implantation provided significant benefit in terms of speech intelligibility and perception in silence as of 3 months after the second implantation (CI-2).

Sequential bilateral CI significantly improved speech intelligibility and perception in patients with both early and late second CI. Other studies reported that late CI-2 provided significant benefit after 1 year's use [2]. Zeitler et al. reported that speech perception continued to improve, whatever the inter-CI interval [3]. Another team found no significant correlation between inter-CI interval and sentence recognition in silence using either only the second CI or both [7]. Likewise, other studies reported no effect of inter-CI interval on sound localization [4,8].

However, other studies reported that longer inter-CI intervals were associated with poorer language development, comprehension and expression. Zeitler et al. and Steffens et al. [3,9] found negative correlations between inter-CI interval and speech perception in noise.

These discrepancies may be due to differences in the tests employed. There is no evidence of an inter-CI threshold beyond which the second CI ceases to show benefit [7].

Neurophysiologic studies reported that prolonged inter-CI interval could impair cerebral maturation, due to time difference in central auditory activity [10], and that minimizing the interval could avoid auditory cortical reorganization caused by prolonged unilateral hearing loss [11]. No such effects were found with simultaneous bilateral CI, due to the binaural effect [12].

Myhrum et al. [13] reported that a long inter-CI interval was predictive of poor speech perception results with the second CI, but only in the case of children not equipped with hearing aids during the interval; the study even found that prolonged inter-CI interval was predictive of non-use of the second CI in the long-term [13].

Sequential implantation entails acoustic stimulation with onset at a different age and stage of cerebral maturation. Age is a major factor in brain plasticity, and has to be taken into account as much as the inter-CI interval. Implantation before three and a half years of age respects the sensitive period of language development [14], corresponding to the critical period of central auditory pathway maturation. After a certain age, the inter-CI interval makes little difference, as seen in studies of adults [3,15] and adolescents [7].

In the present cohort, children showing weaker performance with their first CI progressed significantly, as of 3 months' bilateral implantation. Thus, a second CI can help accelerate language development in children with poorer initial performance.

Few studies analyzed pre-CI-2 performance as a predictive factor. Friedmann et al. [7] found that CI-1 performance was non-predictive of performance using the second implant alone, but did

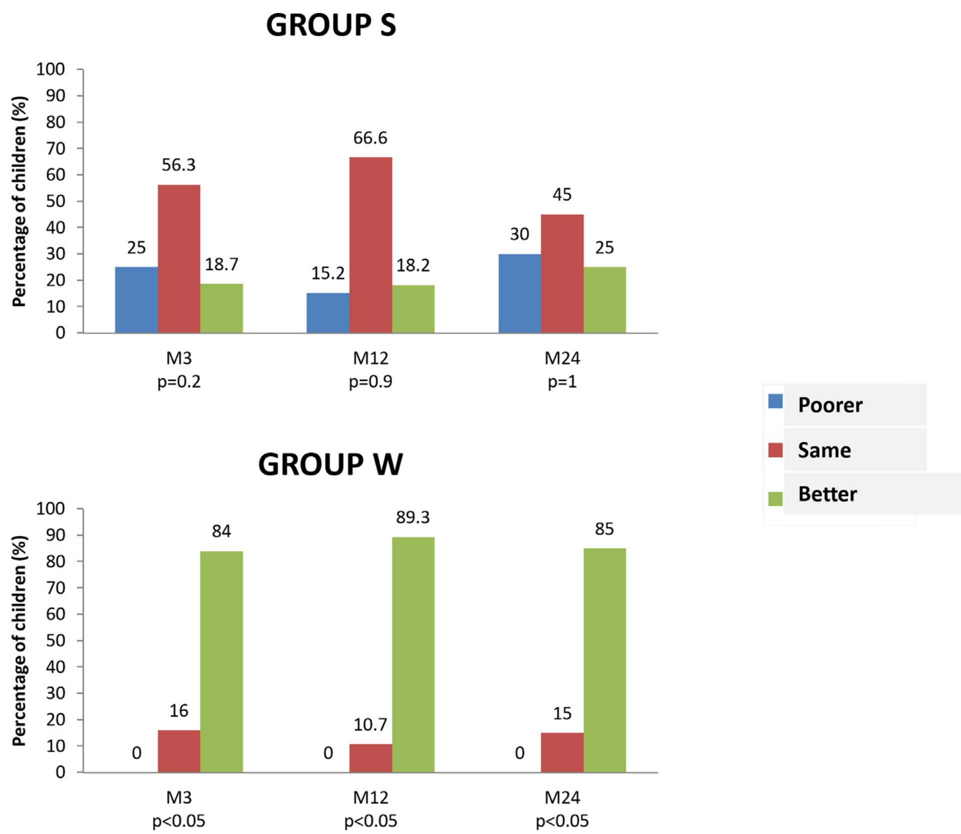

Fig. 2. Words scores post-CI-2 in Strong (S) and Weak (W) CI-1 performance groups.

show significant association with performance using the two CIs. Further studies are needed to confirm benefit of sequential CI in patients with less than optimal initial performance.

Assessing the benefit provided by CI is complex, involving several aspects: auditory perception, expression and language, behavior, school performance, and psychology. The benefit of a second CI is even harder to assess, as the range of benefit is narrower.

Assessment is based on hearing threshold results on audiography, various scales and calibrated speech therapy tests, so as to provide global assessment of perception, language, expression and behavior. However, the patient's and family's feelings and comfort are also important, and are more subjective and complex and possibly discordant with audiometric and speech therapy data. This can be quantified on quality of life scales [2,5,16,17].

The present study included a relatively large number of children, but in a cohort that was relatively heterogeneous, notably in terms of age. Other studies selected more homogeneous series by age: age at onset of hearing loss [18], at first implantation [19], or at sequential contralateral implantation [7].

Selecting a well-defined age group helps control for confounding factors such as linguistic maturation [18], but tends to result in small series: 21 for Grieco-Calub et al. [8] and 30 for Van Deun et al. [4].

In the present study, patient records at CI-2 were studied in a multidisciplinary team meeting. Children were selected for strong motivation, optimal postoperative speech therapy conditions and good psychological preparation; this may account for the good overall results, and means that extrapolation needs to be cautious.

The thresholds of 3-year inter-CI interval and 85% word discrimination were set empirically, and it would be interesting to explore other thresholds and the consequent differences in results. This could not be done here, due to the complexity of the statistical analyses that would be needed and to a certain number of missing data inherent to child assessment.

One particularity of pediatric studies is the importance of the fact that subjects' ages advance over the study period. Audiometric and speech-therapy tests are age-adapted, and were not necessarily the same at the various assessment time points; tests for older subjects are correspondingly more "difficult", so that speech therapy scores at 1 year post-CI-2 might be better than at 2 years, due to the nature of the respective tests. This biases assessment of individual progression over time.

Patients were implanted between 1.2 and 24 years of age. Speech therapy tests at M3, M12 and M24 thus differed between younger and older patients, introducing an inter-individual bias.

Moreover, certain factors were not taken into account, such as socioeconomic level, parental investment in the project, or associated behavioral, cognitive, psychomotor or sensory disorders, although these strongly impact CI results. They are, however, difficult to quantify, being subjective.

5. Conclusion

The number of CI candidates keeps increasing as indications widen. Health and social services have to face up to this. Given the socioeconomic stakes, cost/benefit analysis is critical for sequential bilateral implantation. Certain studies have therefore sought to assess the benefit of a second CI.

The present study found progress in terms of perception test results in children and adolescents receiving a second cochlear implant at a short interval. It is advisable to keep this interval short. There was also benefit after a long inter-implant interval: intervals of several years are not predictive of lack of benefit.

In some patients with poor performance on their first CI, a second CI can enable a leap forward in perceptual and linguistic accomplishment.

Benefit should not be assessed purely in terms of perception and intelligibility but also of auditory comfort, improved understanding in noise and progress in sound localization, especially in patients showing good performance with their first CI.

Thus, tests more specifically targeting the situation of these patients need to be developed, and further studies should be conducted to identify any prognostic factors for success or failure of sequential contralateral implantation, so as better to inform patients and families of the outcome that can be expected.

Disclosure of interest

The authors declare that they have no competing interest.

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