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RESEARCH ARTICLE



## Evolution of speech perception in patients with ossified cochlea and short array cochlear implant

Guilherme Adam Fraga<sup>a</sup>, Rhaissa Heinen Peixoto<sup>a</sup>, Julia Speranza Zabeu<sup>b</sup> and Luiz Fernando Manzoni Lourenço<sup>c,d</sup>

<sup>a</sup>Hospital for Rehabilitation of Craniofacial Anomalies (HRAC), University of São Paulo, Bauru, Brazil; <sup>b</sup>Cochlear Implant Section, Hospital for Rehabilitation of Craniofacial Anomalies (HRAC), University of São Paulo, Bauru, Brazil; <sup>c</sup>Cochlear Implant Section and Department of Otolaryngology, Hospital for Rehabilitation of Craniofacial Anomalies (HRAC), University of São Paulo, Bauru, Brazil; <sup>d</sup>Bauru School of Dentistry, University of São Paulo, Bauru, Brazil

### ABSTRACT

**Background:** Short array cochlear implant is indicated as rehabilitation in patients with severe to profound deafness, especially when there is cochlear ossification. In these cases, with reduced intracochlear patency, total insertion becomes more difficult, requiring the use of this type of electrode (15mm). Few studies have been published to evaluate auditory performance, presenting controversial audiological results.

**Aims/Objectives:** To report the speech perception of users of cochlear implants (CI) with short array.

**Material and Methods:** A retrospective analysis of medical records of patients who underwent surgery for cochlear implantation with a short array, between 2009 and 2020, at the Hospital for Rehabilitation of Craniofacial Anomalies, University of São Paulo (HRAC-USP) was carried out.

**Results:** There was performance evolution in the speech perception tests in the data analysis. Meningitis and congenital hearing loss were the main indications for CI in the sample.

**Conclusion.** CI with a short array is an alternative in the management of patients with a history of cochlear ossification and severe or profound sensorineural hearing loss.

**Significance:** To demonstrate the evolution of speech perception tests with short array cochlear implant in patients with or without ossified cochlea and its characteristics for application in clinical practice.

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Cochlear implant; speech perception; short array; ossified cochlea

## Introduction

Cochlear Implant (CI) is considered one of the possible treatments for deafness, being indicated mainly for severe and profound bilateral sensorineural loss, since deafness is the most common sensory disorder, affecting 1 in every 700–1000 live births [1]. The first cochlear implant surgery was performed in 1961 by House and Edgerton, and since then, devices, technologies and surgical techniques have been developed [2].

The number of inserted electrodes is a significant factor in determining post-implant performance, especially with regard to speech and language skills. Each commercially available cochlear implant system contains a specific number of electrodes. Ideally, all electrodes should be inserted, even if they are not subsequently activated, as this allows greater flexibility in programming [3].

Standard electrode arrays are designed for a normal cochlea, therefore they may not be suitable for cochlea with anatomical changes. To contemplate the atypical cochlear anatomy, special sizes were developed, as is the case of the compressed array, from the company Med-El (Innsbruck, Austria), which compressed the same twelve electrodes

existing in its standard implants (26.4mm) in a space of 12.1mm.

Cochlear ossification was once considered a contraindication for implantation [4], as the obliteration of the cochlea by bone neoformation or fibrous tissue does not allow standard electrodes to be properly inserted from the basal turn, in addition to causing potential complications, such as perilymphatic fistula [5]. Some options for surgical techniques are available to solve this problem. If ossification or fibrosis is found in scala tympani, for example, an alternative is the insertion of electrodes in scala vestibuli. The oldest technique used in fully ossified cochleae is to drill the basal turn as deep as possible and insert the number of electrodes that fit inside it. In these cases, an average insertion length of 8.5 to 14.3mm can be achieved [6].

Studies have been done on the performance of cochlear implant users with cochlear ossification and partial electrode insertion and controversial results have been reported. Kirk et al. [7] did not find significant differences when comparing the speech perception tests in the partial or total insertion of the electrodes. Croghan et al. [8] analyzed speech performance with a competitive speaker, using 4, 8, 12 or 22

electrodes, with significant improvement as the number of active electrodes increased from 12 to 22 electrodes.

A major challenge for cochlear implant surgery is ossified cochleae, in which electrode insertion may be incomplete, with potential loss of auditory performance. In these patients, with short arrays, the evaluation of speech perception is still scarce in the literature. This study was motivated due to the parameters observed in the scores of these patients in our clinical practice, so the aim is to report the speech perception of users of cochlear implants (CI) with short array.

## Methods

### Study design

This is a retrospective and descriptive study, with a two-year longitudinal follow-up, of data from the medical records of all patients who underwent surgery for cochlear implantation with a Compressed electrode from the company Med-El® (Innsbruck, Austria), between 2009 and 2020, in Cochlear Implant Section of the Hospital for Rehabilitation of Craniofacial Anomalies, University of São Paulo (HRAC-USP), in Bauru (São Paulo), Brazil. Upon submission of the research project to the Research Ethics Committee of HRAC/USP, the informed consent form was waived.

### Participant eligibility

Patients with severe or profound sensorineural hearing loss, with an indication for CI according to GM/MS Ordinance No. 2776 of the Brazilian Ministry of Health; all patients operated with a Med-El® cochlear implant, Ti100 internal device, compressed electrode, between 2009 and 2020, who had imaging tests with signs suggesting ossification or cochlear fibrosis, regardless of the etiology of the hearing loss; use of CI for at least two years after surgery, with daily use of the device; and cognitive ability to respond to audiological tests performed pre- and postoperatively.

Free field tests were collected at frequencies of 0.5, 1, 2, 3, 4 e 6kHz in all patients when cochlear implant was indicated, before surgery. At 0.5 and 1kHz, the mean was 118.3dB and the median was 120dB; at 2, 3, 4 and 6kHz, the mean and median were 120dB.

The Med-El Ti100 device is made of silicone and titanium, with a single electrode array and ground wire located in the unit.

The indications of the Brazilian Ministry of Health are severe and/or profound bilateral sensorineural hearing loss; use of a personal sound amplification device (PSAD) prior to surgery, with a result equal to or less than 60% of sentence recognition in an open set with the use of PSAD in the better ear and equal to or lower than 50% in the ear to be implanted; the presence of favorable indicators for the development of oral language measured by standardized protocols; psychological adequacy; access to speech therapy; and commitment to care for the components of the cochlear implant.

Patients with auditory neuropathy or who did not allow adequate audiological evaluation were excluded from the study.

### Data collection

The data collected and analyzed by the study were: age, presence of associated syndrome, clinical indication, complete or partial insertion of electrodes, presence of cochlear ossification and pre-surgical speech perception tests and 3, 6, 12, and 24 months after implantation.

The tests collected and described are: IT-MAIS (Infant Toddler – Meaningful Auditory Integration Scale) [9], MUSS (Meaningful Use of Speech Scale) [10], hearing category [11], language category [12], word recognition [13] and sentences recorded in silence and in noise [14], according to what was done in each analyzed patient.

Based on the questionnaires and assessments, children were classified into one of the following six hearing categories [11]: Category 0- Does not detect speech; Category 1- Detects speech; Category 2- Differs words based on suprasegmental cues; Category 3- Begins closed set identification (identical words in duration, but with multiple spectral differences); Category 4- Identifies words by recognizing vowels in a closed context; Category 5- Identifies words by recognizing consonants in closed contexts; Category 6- Recognizes words in open sets.

Based on responses to MUSS [10], the therapist's perception and observation, and the parents' report, the children were classified into one of the following five language categories [12]: Category 1- The child does not speak and may present undifferentiated vocalizations; Category 2- The child speaks only isolated words; Category 3- The child builds sentences with two or three elements; Category 4- The child builds sentences with four or five words and begins using connectives; Category 5- The child builds sentences with more than five words and conjugates verbs, uses connectives, and is fluent in oral language.

Speech perception was assessed using an open-set list of 20 disyllable words (CVCV; C=consonant, V=vowel) [13] in quiet and three open-set lists with 20 sentences each in quiet and noise [14]. The speech signal was presented in an acoustic booth by using the Madsen Astera audiometer (Otometrics; Natus, Medical Denmark) connected to an amplifier in a free field at 0° azimuth at 60dB SPL. Words were presented by trained audiologists using live voice while monitoring the VU meter to keep the intensity at 60dB SPL. Sentences were conducted by applying a recorded list of 20 affirmative Portuguese sentences with each sentence having three to seven words for a total of 100 words without repetition. The lists were balanced according to the Brazilian Portuguese phonetic inventory [14]. For the test in noise, a cocktail party noise at 50dB SPL (+10dB signal-to-noise ratio, SNR) was presented simultaneously through the same loudspeaker. For both the word and sentence recognition tests, participants were asked to repeat the speech stimuli and each word repeated correctly was scored, resulting in a final score range from 0 to 100%.

## Statistical analysis

The Friedman test was used to assess the effect of the five periods (pre-implantation, 3 months, 6 months, 12 months and 24 months) on the score of each test performed on the individual. The use of the Friedman test is justified because the observations are characterized by repeated measurements on the same individuals, and despite the number of observations ( $n = 53$ ), the normal approximation may be unfeasible given the nature of the scale of the response variables.

The Friedman Test is a non-parametric test analogous to the analysis of variance with repeated measures. The significance level adopted was  $\alpha = 0.05$ . In case of statistical significance of the Friedman test, post-hoc analysis was performed with the Conover Test.

## Results

In a total of 1713 patients undergoing implantation, 70 users of the MED-EL Cochlear Implant, Sonata Ti 100 internal device, short array, who underwent surgery between 2009 and 2020 at the Hospital for Rehabilitation of Craniofacial Anomalies, University of São Paulo (HRAC-USP). Twenty-five patients were excluded: 10 who underwent internal device replacement, 10 who used another type of contralateral electrode, and 5 who did not have sufficient data for analysis, due to transfer to another service or unjustified absences from follow-up appointments.

Among the 45 patients eligible for the study, 53 ears implanted unilaterally or bilaterally were analyzed (Table 1). Thirty-eight ears were from male patients (72%) and 15 from female patients (28%). All patients underwent unilateral or bilateral cochlear implantation using a surgical technique with a standardized retro auricular approach, through posterior tympanotomy, performed by the same surgical team.

In this total of 53 ears, 37 (70%) were from patients aged between 1 and 12 years old and 16 (30%) from patients over 12 years old. The most frequent indications were post-meningitis (66%) and congenital hearing loss (24%). In addition, cases of otosclerosis and sudden deafness were also observed. Among the congenital causes, there were idiopathic ones, Branchio-Oto-Renal Syndrome, Cardiofacial Syndrome and Dandy-Walker Syndrome.

Ossification was observed at the opening of the cochlea in 25 ears (47%), while cochlear patency was found in 28

ears (53%). All patients were successful in completely inserting the array, except for 11 cases (21%). These patients with partial insertion, due to severe cochlear ossification, consist of 8 cases of post-meningitis, 1 case of sudden deafness, 1 case of otosclerosis and 1 case of congenital deafness.

Among patients who reported a medical history of meningitis as the etiology of deafness, the mean age at implantation was 1.8 years (standard deviation 2.06). The youngest patient was implanted at 1 year old, while the oldest underwent the procedure at 59 years old. Cochlear ossification was found in 18 ears (51%): 7 had partial insertion of electrodes, and 11 were complete. The average implantation time after the diagnosis of meningitis was 6.97 years, with a median of 2 years. The shortest time interval between diagnosis and CI was 30 days, and the longest was 38 years.

## Performance outcomes

Patients with ossification, regardless of the etiology of hearing loss ( $n = 25$  ears), were evaluated in five periods with eight tests performed at each time. In Language Category, stability of scores was observed in the first 12 months of follow-up and an improvement after 24 months of CI use ( $p = .001$ ). Word and Sentence Recognition in Silence and Noise also tended to increase scores at 24 months, but without statistical significance ( $p = .09$ ,  $p = .09$  and  $p = .24$ , respectively). IT-MAIS showed worsening scores over time, but also without statistical significance ( $p = .64$ ) (Table 2).

Patients without ossification were also evaluated in the same period, regardless of the etiology of the hearing loss ( $n = 28$  ears). IT-MAIS kept the scores stable over time ( $p = .03$ ). In Language Category, scores improved at 12 months, with a new increase at 24 months ( $p = .00$ ). Word and Sentence Recognition in Silence remained stable from the preoperative period up to 12 months of CI use, showing improvement at 24 months (both  $p = .00$ ). Sentence Recognition in Noise tended to improve over time, but no statistical difference was observed in the Friedman Test for this variable ( $p = .14$ ) (Table 2).

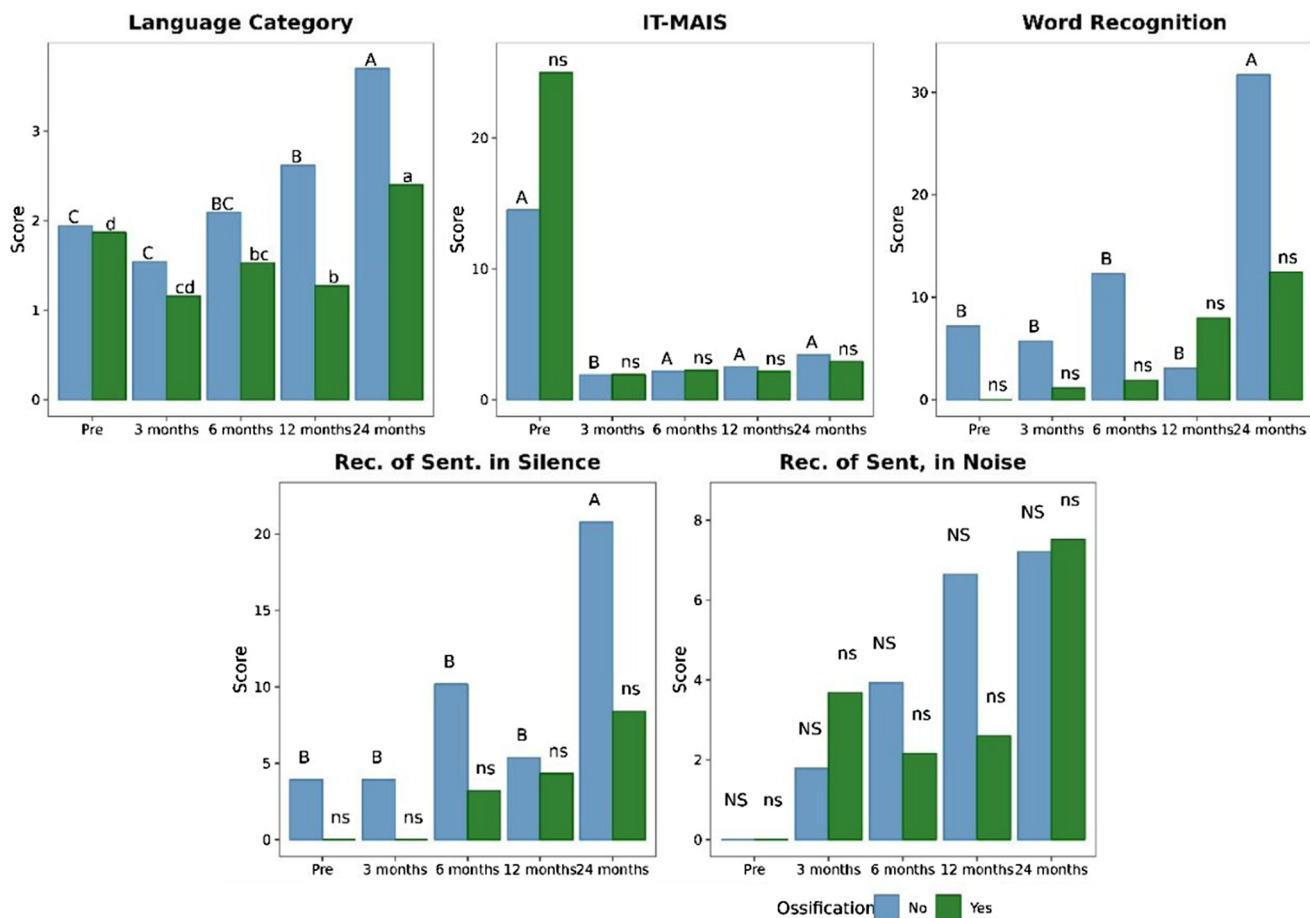
Auditory Category and MUSS tests had no patients in the five successive evaluations and therefore could not be evaluated. Comparing patients with and without ossification, an evolution was observed in all tests over 24 months, with

**Table 1.** Etiology x Ossification: short array patients data.

Etiology	Ossification	No ossification	Total of ears
Meningitis	18	17	35
Congenital hearing loss	4	8	12
Otosclerosis	1	1	2
Dandy Walker Syndrome	0	1	1
Cardiofacial Syndrome	0	1	1
Branchiootorenal Syndrome	1	0	1
Sudden hearing loss	1	0	1

**Table 2.** Evaluation of speech perception tests among groups of patients with ossified (Yes) and non-ossified cochlea (No). LC: language category; WR: word recognition; RSS: recognition of sentences in silence; RSN: recognition of sentences in noise.

Group	Test	Statistic	Degrees of Freedom	p-value
Yes	LC	17.8689	4	.0013
Yes	IT-MAIS	2.4762	4	.6489
Yes	WR	8.000	4	.0916
Yes	RSS	7.8049	4	.0990
Yes	RSN	5.4167	4	.2472
No	LC	25.7963	4	.0000
No	IT-MAIS	10.5618	4	.0320
No	WR	29.5260	4	.0000
No	RSS	26.3457	4	.0000
No	RSN	6.8454	4	.1443



**Figure 1.** Evolution of tests in patients with and without cochlear ossification (uppercase letters: group without ossification, with "A" being the best score found and "C" being the worst; lowercase letters: with ossification, with "a" being the best score and "d" the worst; NS/ns: not significant at a significance level of 5%; different letters in the same group represent statistically significant difference).

the exception of IT-MAIS. Better scores were observed in the follow-up of patients without cochlear ossification (Figure 1).

## Discussion

The most common indications described for the short array are post-meningitis and congenital hearing loss. One of the advantages is patient safety, once its insertion can be performed without a complete drill-out, which reduces the risk of injury to the facial nerve, as it is known that this technique is associated with its increase, in addition to injury to the carotid artery and the modiolus [15].

Sixteen patients had signs of fibrosis or cochlear ossification on preoperative imaging but had complete insertion of electrodes intraoperatively. Since their clinical conditions and complementary exams suggested ossification, the use of a short array was previously requested, without the possibility of changing to a longer array when the cochlear patency was verified, a fact also described in the population studied by Bauer (2004) [3]. Studies show that the sensitivity of Computed Tomography (CT) in detecting early ossification is low, approximately 33% to 73%. Magnetic Resonance Imaging (MRI) is considered to have better sensitivity and

specificity than CT in predicting ossification. The sensitivity of MRI combined with CT for detecting ossification can exceed 90% [4]. The ossification described in our study is the one found intraoperatively for each patient, regardless of imaging findings.

Wang et al. [16] showed that patients with partial insertion of electrodes had worse auditory performance than those with complete insertion. However, Tokat [17] and Nichani [18] affirm that patients with cochlear ossification may have auditory results as satisfactory as those without ossification, regardless of the array complete or partial insertion. These conflicting results are probably attributed to the clinical and surgical conditions of the patients in the different studies.

In our sample, among the 53 implants with a short array, in 25 (47%) ossification was found in the cochlear opening. In the global evaluation, the scores of the speech perception tests of these patients tended to stability in the first 12 months of use, showing significant changes from this period of time up to 24 months. Several factors can explain the level of performance observed. By compressing the electrode, the distance between the pairs of electrodes is decreased, which can lead to an overlap of electric fields. This would limit selective stimulation of the auditory nerve, which is associated with reduced pitch discrimination and worse speech understanding [6].



Among these patients with ossification, there are 11 with a history of meningitis (6 children between 1 and 11 years old and 5 adults between 15 and 53 years old), 3 with a diagnosis of congenital hearing loss (2-3 years old), 1 with otosclerosis (40 years old) and 1 with Branchio-Oto-Renal Syndrome (1-year-old). In these cases, it was not possible to exclude that sequelae resulting from the etiology of the hearing loss were responsible for the low performance in speech perception scores.

The etiology of hearing loss in our sample was meningitis in more than half of the cases, also being the most common cause of cochlear ossification reported in the literature. Infection of the subarachnoid space reaches the cochlea mainly through the cochlear aqueduct, which is the natural connection channel between the cerebrospinal fluid (CSF) and the inner ear [19], therefore, the ossification of the basal turn of the cochlea could be explained by the anatomical path itself. In these patients, there may still be a decrease in the number of functional spiral ganglion cells, in addition to the presence of fibrous tissue and new bone formation that may alter the cochlear nerve electrical impulse [19]. Damage to the central auditory system can occur, causing alterations in processing, as well as other cognitive deficits. In this sense, it is not surprising that this population has poorer objective outcome measures.

Cochlear ossification in meningitis develops rapidly, starting within 4 to 8 weeks of illness, and there may be complete ossification after 5 months. Auditory evaluation is recommended for all patients immediately after the diagnosis of meningitis [4], so that adequate auditory rehabilitation is carried out quickly and has a favorable outcome. The shortest time interval between the diagnosis of meningitis and CI was 30 days in our sample, with a median of 2 years among all patients with this etiology.

Despite the different tools for monitoring auditory performance in children and adults, the patients monitored had an evolution in speech perception with the time of use of the CI. In cases where these parameters were less satisfactory, further damage or reduction of spiral ganglion cells caused by inflammation or ossification and partial insertion of the electrode array should be considered. No complications were reported in the group of patients studied during the analyzed period.

## Conclusion

A short-array cochlear implant is an alternative in the management of patients with a history of cochlear ossification and severe or profound sensorineural hearing loss. The benefits for these patients are more clearly seen in subjective measures, showing evolution in auditory performance, especially with long-term effects.

## Disclosure statement

The authors declare no conflicts of interest. The authors alone are responsible for the content and writing of this manuscript.

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