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Long-term results of the cochlear implant in patients with otosclerosis

Abstract

Introduction: Otosclerosis is a bone dysplasia of the optic capsule that promotes progressive metabolic derangement, and can lead to profound hearing loss.

Aim: Compare the postoperative results in patients undergoing cochlear implant with otosclerosis compared with patients with other causes of deafness.

Design: Retrospective cohort study.

Materials and methods: Compare cochlear implant results in otosclerosis patient to those to matched pair control group within five years with cochlear implant program - Comfort and Threshold, speech test sentences, monosyllabic and disyllabic, audiometry - gender, age at implantation, duration of deafness.

Results: 17 with otosclerosis and 36 with other causes. Patients with otosclerosis had a mean age of 50.2years and the control group, mean age of 40.8years at the time of implantation ($p < 0.05$). The duration of deafness until the time of implantation showed no significant difference between the groups of patients. When assessed for the speech sentences, monosyllabic and disyllabic test found no statistical difference was found between the otosclerosis and other losses' groups. Observed greater stimulation of the facial nerve in otosclerosis patients were used when straight electrodes.

Conclusion: Otosclerosis patients' implanted showed good surgical results, despite the greater number of complications presented the stimulation of the facial nerve. These results are comparable to the study of patients in the control group with statistical difference between them, despite the progressive feature of otosclerosis disease

Keywords: otosclerosis, cochlear implant, cochlear otosclerosis

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Angela Rubia Oliveira Silveira, Alexandre Caixeta Guimarães, Silvia Fernanda Brihi Badur Curi, Arthur Menino Castilho, Paulo Rogério Cantanhede Porto, Agrício Nubiato Crespo

Department of Otorhinolaryngology and Head and Neck Surgery State University of Campinas, Brazil

Correspondence: Angela Rubia Oliveira Silveira Department of Otorhinolaryngology and Head and Neck Surgery State University of Campinas Faculty of Medical Sciences UNICAMP FCM Campinas SP, Brazil, Email Arubiasilveira@gmail.com

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Introduction

Otosclerosis is a primary focal osteodystrophy of the optic capsule that affects genetically predisposed individuals and promotes the metabolic derangement of the endochondral layer of the labyrinthine bone. Characterized by reordering and disordered bone neoformation.[1]

Only 23% of patients with otosclerosis observed by histological changes also present clinical manifestations [2], because in most cases otosclerosis reaches small regions of the optic capsule without causing clinical symptoms (histological form).[3] It is estimated that between 9 and 10% Of patients with otosclerosis will develop deep sensorineural hearing loss due to the evolution of the disease.[4,5]

The clinical symptoms of cochlear otosclerosis depend mainly on the progression of disease and involvement of the optic capsule. These patients may have sensorineural or mixed hearing loss, tinnitus and vertigo less frequently.[6]

The diagnosis is made mainly by the complaint of hearing loss with onset in adult life that is associated with family history in 40 to 60% of the cases and a tomographic image with signs of otospongiosis.

Due to the progressive characteristics of otosclerosis, some authors decided to compare patients implanted with otosclerosis with other patients implanted by other losses, and in general the results in patients with otosclerosis regarding speech tests and auditory thresholds are similar to those of patients with other.[7–14] A higher incidence of facial nerve stimulation is reported, which may be present in [2] to 75% of the implanted patients,[9–11,13,15,16] especially when straight-type electrodes are used, a decrease in the number of active electrodes over the years After the cochlear implant,[7,11] and rarely the insertion of the implant by cochlear ossification.[5,12]

One study reported the occurrence of new bone formation in the tympanic ramp present in two implanted patients with otosclerosis but without impairing the performance of the cochlear implant.[17]

Two studies have described cases of patients with otosclerosis implanted with a lot of facial stimulation that after the replacement of the cochlear implant with the implant Nucleus 24 Contour®, showed decreased facial stimulation and improved hearing results.[18,19]

It is known that the cochlear implant has provided good hearing results in several aetiologies of hearing loss, including in patients with otosclerosis. It is established that hearing loss caused by otosclerosis improves with cochlear implant treatment. However, considering the progressive characteristic of otosclerosis, reports of cases with poor post-implant auditory outcome and the shortage of studies with a long time of follow-up, we conducted this study that proposed to evaluate the hearing results and complications in patients with otosclerosis implanted after a period of five years and to compare the results with those of patients implanted by other etiologies.

Materials and methods

Retrospective cohort study approved by the Research Ethics Committee of the Faculty of Medical Sciences, with the number 132/2011, CAAE: 0091.0.146.000-11.

The medical records of all adult patients submitted to cochlear implantation from the otology clinic of the Otorhinolaryngology discipline of the Hospital das Clínicas of the State University of Campinas (Unicamp) from 2002 to 2009 were analyzed.

Age data were collected at the date of surgery, gender, implanted ear laterality and deafness time, defined as the time elapsed from hearing loss to the activation of the cochlear implant.

Complications related to the surgical site, such as bleeding, suture dehiscence, otitis, gusher, facial nerve palsy, electrode cracking, stimulation of other cranial nerves by electrodes and other complications have also been reported.

The following tests were performed in the preoperative period with and without AASI and in the first, third and fifth postoperative years. The Clinical Audiometer AC 30 audiometer was used.

i. Tonal audiometry in the frequencies of 0.25, 0.5, 1, 2, 3, 4, 6 and 8 Khz. Made with headphones and 0º of azimuth.

ii. Speech recognition test with monosyllabic, disyllabic and open set sentences (Annexes 1, 2 and 3). Made live. (From the list: Bevilacqua MC, Multichannel Cochlear Implant, 1998)

The patients were evaluated with the implanted device programming in:

i. Maximum level of electric current with sound comfort, Comfort.

ii. Minimum level of electrical current to generate acoustic stimulus, Threshold.

iii. Impedanciometry, with transverse section, in the fifth year after cochlear implant surgery. These tests were performed at the Otology outpatient clinic of the Otorhinolaryngology discipline of the Faculty of Medical Sciences - FCM (Unicamp).

iv. All the implants used were Cochlear® multichannel implants, being Nucleus 24k, Nucleus 24M and Nucleus Contour. All of them have an ACE-type processing strategy.

v. The Cochlear® Custom Sound 3.2 program was used to evaluate objective data, Comfort, Threshold and immitance measurements. Data were obtained in four different combinations of Comfort and Threshold.

vi. Subjective speech tests were performed in each of the four combinations of the program and it was determined in which circumstance there was a better adaptation to the patient. The programming chosen was the one the patient used until the next consultation.

vii. The data were evaluated in the preoperative period, after one, three and five years after the cochlear implant surgery.

viii. Study group: 17 patients operated on and diagnosed with otosclerosis.

ix. Control group: 37 patients operated without otosclerosis.

Inclusion criteria

Study group: patients with otosclerosis undergoing cochlear implant surgery. The diagnosis of otosclerosis was made by tomographic findings, history of previous surgery, stapedotomy or clinical evaluation.

Control group: all adult patients undergoing cochlear implant surgery, Cochlear® brand, with a different etiology from otosclerosis.

Exclusion Criteria

Patients who did not present complete information in the medical record about pre and post-surgical hearing evaluation of the cochlear implant.

Patients who underwent cochlear implants from other brands were also excluded because of the lower frequency of use of other implants. The surgeries were performed by the team composed of four otologists.

Data analysis

Descriptive analysis with presentation of position and dispersion measurements for numerical variables and frequency tables for categorical variables.

The chi-square test was used to compare proportions. Mann-Whitney test was used to compare numerical measures between two groups. To compare numerical measures between two groups over time, Anova was used for repeated measures with transformation by stations. The significance level adopted for the statistical tests was 5%.

Result

139 adults underwent cochlear implantation until December 2012 (Table 1).

Table 1 Etiology of hearing loss in adult patients undergoing cochlear implantation

Etiology	Number	%
Idiopathic	37	26.61
Advanced otosclerosis	25	17.98
Meningitis	25	17.98
TCE	11	7.91
Congênita	9	6.47
Medicamentosa	6	4.32
Rubéola gestacional	4	2.88
Meniere	3	2.14
Sind Pendred	2	1.44
Parotidite	2	1.44
OMC	2	1.44
HIV	2	1.44
Familiar Progressiva	2	1.44
Sind Usher	1	0.71
Sind Mondini	1	0.71
Sind de Cogan	1	0.71
Prematuridade	1	0.71
Osteogenese Imperfecta	1	0.71
Mutação conexina 26	1	0.71
Hiperbilirrubinemia	1	0.71
Fístula Perilinfática	1	0.71
Exerese de neuroblastoma bilateral	1	0.71
Total	139	100

Table 1 - Etiology of hearing loss in adult patients undergoing cochlear implantation Twenty-five patients had otosclerosis. Two were excluded because they had undergone cochlear implant from another brand, and six were excluded because they were implanted after 2009. Thus, the study group consisted of 17 patients.

One hundred and fourteen patients with post-lingual deep hearing loss caused by other etiologies were implanted by the year 2012. All patients operated after 2009 and those with incomplete medical records were excluded.

Citation: Silveira ARO, Guimarães AC, Curi SFBB, et al. Long-term results of the cochlear implant in patients with otosclerosis. *J Otolaryngol ENT Res.* 2017;9(4):636–641. DOI: 10.15406/joentr.2017.09.00297

The control group consisted of 36 patients (Table 2).

Table 2 Patients submitted to cochlear implant until 2009 selected for the control group

Etiology	Number	%
Idiopática	14	38.89
TCE	5	13.89
Meningite	5	13.89
Familiar progressiva	4	11.11
Medicamentosa	3	8.33
OMC	2	5.55
Meniere	2	5.55
Sind Mondini	1	2.77
Total	36	100

Table 2 - Patients submitted to cochlear implant until 2009 selected for the control group. The frequencies between the male and female patients in the two groups and the laterality of the operated ears in the two groups were compared (Table 3). There was no statistical difference between the study and control groups. The mean age of patients was compared at the time of cochlear implantation. Patients with otosclerosis had a mean age of 50.2years, a minimum of 23 and a maximum of 72years. Patients with hearing loss due to other causes had a mean age of 40.8years, a minimum of 18 and a maximum of 65years. There was a significant difference between the ages, p: 0.0263 (Table 4).

Table 3 Comparison of the variables sex and number of right or left ears between groups

	Male	Female	Right Ear	Ear Left
Otosclerosis	8	9	9	8
Others	17	19	27	9
Total	25	28	36	17
Chi-Square Test	p=0.9911		p=0.1083	

Table 4 Comparison between the age of the pre-implant patients and the time of deafness until the implant

	Pre-Implant Age (Years)	Deafness Time (Years)
Otosclerose		
Average	50.2	8.3
Minimum	23	5

The duration of deafness until the date of cochlear implantation was compared with an average of 8.3years for the otosclerosis group and 5.9years for the control group. There was no statistically significant difference (Table 5).

Table 5 Comparison between the groups regarding the time of implant with respect to the Perceptual Speech Test (TPF) for sentences (percentage of correct%)

Maximum	72	20
Others		
Average	40.8	5.9
Maximum	18	1
Maximum	65	28
	p:0.0263*	p:0.1602*

*Mann-Whitney test comparing the groups, using as standard deviation calculation.

Descriptive analyzes were performed comparing the Speech Percentage Test (TPF) between the groups in the pre-implant period and the post-implant period of one, three and five years. There was no statistically significant difference between them (Table 6).

Table 6 Comparison between the groups regarding the time of implant with respect to the Speech Perceptual Test for Monosyllables - TPF M - (percentage of % hits)

	TPF for (%)	TPF 3 Years (%)	TPF 3 Years (%)	TPF 5 Years (%)
Otosclerose				
Average	0,8	85,4	89,3	92,6
Minimum	0,0	50,0	50,0	50,0
Minimum	14,0	100,0	100,0	100,0
Otosclerose				
Average	1,7	88,4	91,4	92,5
Minimum	0,0	30,0	50,0	52,0
Minimum	20,0	100,0	100,0	100,0

p: 0.730 between the otosclerosis and other groups; p: 0.0001 for time comparison within the group;

p: 0.999 for comparison between groups and time (Mann-Whitney)

Descriptive analyzes were performed comparing the results of the Percent of Speech for Monosyllables between study and control groups in the period Pre-implant and post-implant periods of one and five years. There was no difference Statistic between them (Table 7).

Table 7 Comparison between the groups regarding the time of implantation with respect to the Speech Perceptual Test for Dissyllables - TPF D - (percentage of correct answers%)

	TPF M Pre (%)	TPF M 1 Year (%)	TPF M 5 Year (%)
Otosclerose			
Average	0,0	72,9	84,6
Minimum	0,0	50,0	60,0
Minimum	0,0	100,0	100,0
Otosclerose			
Average	1,4	71.2	79,8
Minimum	0,0	32,0	52,0
Minimum	20,0	94,0	100,0

p: 0.493 between the otosclerosis and other groups; p: 0.0001 for comparison of time within the group;

p: 0.130 for comparison between groups and time (Mann-Whitney test)

Descriptive analyzes were performed comparing Perceptual Speech Test for Dissyllables between the groups in the pre-implant period and post-implant periods of one and five years. There was no statistically significant difference between them (Table 8).

Table 8 Comparison of the mean of the minimum electric current to generate stimulus between the groups regarding the time of implantation

	TPF D Pre (%)	TPF D 1 Year (%)	TPF D 5 Year (%)
Otosclerose			
Average	0,0	79,9	89,5
Minimum	0,0	52,0	60,0
Minimum	0,0	100,0	100,0
Otosclerose			
Average	5,6	78,1	86,4
Minimum	0,0	60,0	66,0
Minimum	44,0	100,0	100,0

p: 0.5617 between the otosclerosis and other groups; p: 0.0001 for comparison of time within the

group; p: 0.161 for comparison between groups and time (Mann-Whitney test)

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The following analysis was made by dividing the electrodes into series of three groups. O First group with electrodes of numbers 1 to 7. The second group with electrodes of Numbers 8 to 15 and the third group with electrodes of numbers 16 to 22.

Analyzes were performed between the groups comparing the minimum current threshold to generate auditory stimulus (Threshold) - T and maximal electrical stimulation with Comfort (Comfort) - C.

Although Threshold and Comfort had higher averages for the otosclerosis group in all groups of electrodes, there was no statistically significant difference between the groups (Tables 9 & 10).

The groups were compared for pre-implant tonal audiometry with and without Sound Amplification Apparatus (AASI) and post-implant with one, three and five years.

Table 9 Comparison of Comfort (Maximum electric current without generating discomfort auditory) between the groups regarding the time of implantation

	Comfort First Year			Comfort Third Year			Comfort Fifth Year		
Series of Electrodes	1 a 7	8 a 15	16 a 22	1 a 7	8 a 15	16 a 22	1 a 7	8 a 15	16 a 22
Otosclerose	184.1	185.4	182.9	188.5	188.3	187	191.6	196.3	195.6
Outros	181.2	180.7	182.3	186.6	187.2	190.1	187.5	187.5	190.8

P value for each series of electrodes, from 1 to 7 p: 0.0625; from 8 to 15 p: 0.1191; 16 to 22 p: 0.3161 *Bruno*

Table 10 Mean of audiometric results of patients submitted to cochlear implant with otosclerosis (study group) with hearing loss from other causes (control group) regarding the time after implantation

	Preoperative Without AASI		Preoperative With AASI		Preoperative 1st Year		Preoperative 3rd Year		Preoperative 5th Year	
	Study	Control	Study	Control	Study	Control	Study	Control	Study	Control
0,25 Khz	97.9	100.7	81.2	98.5	28.8	28.3	25.6	27.1	23.2	31.8
0,5 Khz	108.8	115.4	91.8	83.6	33.2	26.3	29.1	28.8	28.2	30.8
1 Khz	112.4	111.8	92.1	107.6	30.6	32.8	26.5	28.8	25.6	26.3
2 Khz	114.4	108.8	97.4	95	29.1	31.9	25.6	24.6	24.1	25.4
3 Khz	114.7	115.1	103.8	108.5	30	30.4	28.2	28.3	27.6	24.3
4 Khz	115.3	106.8	108.2	104.6	33.5	37.2	31.8	30.3	30	28.2
6 Khz	112.1	114.6	106.2	89.7	35.9	36.1	28.8	26.7	30.3	29.9
8 Khz	107.1	114.6	104.1	109.4	44.7	39.3	36.5	30.7	37.1	28.5

Tonal audiometric evaluation by frequency: At 0.25 Khz, when comparing the results between the study group and the control group, with pre-implantation hearing aid, better results were observed in the otosclerosis group, p: 0.0096. After five years of implantation, the group with otosclerosis had better results, with a mean of 23.2 db and in the control group with 31.8 db (p: 0.009).

For the frequency of 0.5 Khz there are better results in the group with otosclerosis (p: 0.0114) preoperatively without AASI.

In the 1Khz evaluation, the results with pre-implant hearing aids were better in the otosclerosis group, with a mean of 92.1 db, and a control group with 107.6 db, p: 0.0134.

In the frequencies of 4 and 8 Khz, there were better results in the preoperative control group without the device (p: 0.008) and (p: 0.007), respectively.

At 6 Khz the results were worse when they used preoperative hearing aid in the otosclerosis group. The graphs of the respective audiometric analyzes can be found in the 4. The mean of the audiometric values can be visualized in Table 3.

Table 3 - Mean of the audiometric results of patients undergoing cochlear implant with otosclerosis (study group) with hearing loss from other causes (control group) regarding the time after implantation.

Surgical findings

All patients with otosclerosis had new bone formation. Ten patients had ossification of the round window and seven had ossification of the tympanic ramp. In the control group there was partial ossification of the tympanic ramp in four patients. The insertion of the electrodes was complete in all patients, except in one patient with otosclerosis.

Three patients with otosclerosis presented facial nerve stimulation, one in the immediate postoperative period (Nucleus 24K) and two patients after one year of surgery (Nucleus 24M). None of the Nucleus contour model. In the group with otosclerosis there was a patient with odynophagia, with pruritus in the pharynx. Symptoms disappeared after the electrodes six, seven and eight were disconnected.

All patients used ACE programming and miscellaneous processors. In the group with otosclerosis the electrodes turned off because they caused facial nerve stimulation were the numbers 13 to 15. The electrodes that caused odynophagia - pharyngeal stimuli are the numbers six, seven and eight. The remainder of the electrodes was switched off because of no auditory stimulus.

Discussion

In two patients with otosclerosis, there was deterioration of auditory perception, speech recognition test and audiometric tests.

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Toung et al.,[16] reported their experience with a 66-year-old male patient with advanced otosclerosis, initially with good results and with a gradual loss of benefit. After 13years it was decided to implant it again but without good results even after reimplantation in this case. 5, 8, 16.

We found in the group with otosclerosis a patient with a sensation of discomfort in the larynx, but after the withdrawal of electrodes number six, seven and eight, this sensation ceased.

Quaranta et al. 13 also exemplify patients with laryngeal discomfort, but do not report which electrodes were responsible for such sensation.

In our study, there was no significant difference in the speech perception tests between the groups before or after the cochlear implant, even after 5years of implantation. Other authors who studied patients with otosclerosis and Cochlear also report good results, but did not make a comparative study.[20]

Although otosclerosis is more frequent in women with prevalence (2:1),[21] the number of patients implanted with advanced otosclerosis in our sample showed that the number of men and women is similar.

There were nine women and eight men implanted. Castillo et al. [14], in a study on the results of cochlear implant surgery in patients with otosclerosis, show a greater tendency of implanted women, four men to 13 women. In this study, the authors show otosclerosis affecting more females due to a probable hormonal cause, but not related to the pregnancies.

Regarding age, one of the largest epidemiological studies on patients implanted with advanced otosclerosis, performed by Rotteveel et al. [5], analyzed 53 patients and showed that the age of the implanted patient ranged from 42 - 79years, mean age of 62 years. In our sample, we observed that the mean of patients implanted with otosclerosis was 50.2 years, minimum of 23 and maximum of 72 years, and in the control group. Mean was 40.8 years, minimum of 15 and maximum of 65 years, with $p < 0.0263$. In agreement with other studies in which the patients with otosclerosis have the most advanced age.

The mean deafness time was higher in the otosclerosis group, eight years, compared to the mean of the control group, five years but with no statistical difference between the groups. Matterson et al.,[10] in a retrospective study, evaluated 59 patients implanted with otosclerosis. They studied whether the deafness time would affect the outcome of the implant. It was found that after three months there was an initial advantage, but after six months the results were similar. Because the results of the speech test obtained resemble after six months, there is no privilege in implanting ears with recent hearing loss or long-term loss.

A study also compared the minimum electrical current threshold (T) and maximal electrical stimulus with comfort (C). 6 Similar to the previous study, the values for patients with otosclerosis were higher, but no statistical difference between the groups.

For the sound comfort levels (C), over time, the parameters within the otosclerosis group were increased for all electrode groups, and the values were significant. Over time, the level of sonorous comfort increased in the otosclerosis group.

Sainz et al.,[9] in a prospective, five-year study, compared 15 patients with otosclerosis and 30 implanted patients for another reason regarding the speech test and implant programming. The programming levels for T were higher in the otosclerosis group, but without significant difference in comparison to the control group. The level of T was higher in the basal turn, differently in the middle and with statistically significant values. For the levels of C, the behavior was similar to the T, higher levels, but with no statistical difference.

The programming of the cochlear implant is very dynamic, requiring constant evaluations and scheduled appointments with the audiologist. In the cochlear implant group with otosclerosis, this requirement increases due to sclerotic changes of the bone and degeneration of the cortical organ and hyalinization of the spiral ligament.[22]

We also tested the results of speech test, and tests for monosyllables and disyllables between the two groups. There was a significant improvement when we compared the groups over time with significant values, $p < 0.001$. That is, post-implant results are better than pre-implants, but when we analyze the control group compared to the study group, the results are not statistically different, corroborating those of other authors.[9,13]

Facial nerve stimulation after cochlear implant activation is a noted condition in several surgical cases, according to Polak et al.,[19] This author did a retrospective analysis of his cases, observed two patients implanted with Nucleus [22] who had facial nerve stimulation. Initially, the number of active electrodes decreased, but the speech understanding decreased. When he chose to reimplant the patients with the Nucleus[24] contour model, one patient in the same ear and the other in the contralateral ear, there was elimination of the facial nerve stimulus and speech tests rose in a patient from 12% to 42% and in another 0% to 86%.

Battmer et al.,[18] published another study with four patients using the cochlear implant type Nucleus 22, which presented with facial nerve stimulation. It was necessary to disconnect several electrodes to eliminate the sensation of the stimulus of VII pair. Active 4, 11, 13 and 15 electrodes were only of the 22 possible for programming.

In all cases it was decided to reimplant another model, the Nucleus 24 contour. In all cases the elimination of the stimulus in the VII pair was observed and the programming levels changed, increasing the level of maximum auditory comfort, C, and decreasing the minimum current level to generate sound stimulus, T.

These data corroborate other previously published articles, such as de Rayner, Bigelow and Muckle et al.,[15,23,24] These authors show cochlear implants of the right type causing stimulation of the facial nerve in greater number than other models.

In this study we observed that of the 17 patients with otosclerosis, three had facial nerve stimulation, one patient in the immediate postoperative period (Nucleus 24K) and two patients after one year of surgery (Nucleus 24M) and none of the Nucleus contour model.

These models prior to Nucleus Contour have the conformation of straight electrodes. And the current model is perimodiolar. Frijins et al.,[25] show in the article a model computed tomography of the cochlea, facial nerve (labyrinth segment) and intracochlear electrodes. This study tries to explain if the type of contact of the electrode (circular or half-band) and position (perimodiolar or lateral wall) influence the stimulation of the facial nerve. The authors used previous studies of cochlear volume and auditory nerve model. Equivalent electrodes were used as Nucleus Straight, Nucleus Contour and Advanced Bionics HiFocus. The straight electrodes, which have their circular contact mode, are closer to the lateral wall, and the curves are perimodiolar. This means that the electrodes located on the lateral wall require a

greater electrical current to stimulate the auditory nerve, which consequently can stimulate the facial nerve in its labyrinth segment with greater ease. The electrodes of the contour model, because they are perimodiolar, are more.

Near the auditory nerve fibers, requiring less electrical current to stimulate them. Changes in otosclerosis change the intracochlear electrical conductivity leading to a greater chance of stimulating the facial nerve. Matterson et al.,[10] for example, in a retrospective study, evaluated 59 patients implanted with otosclerosis. They observed 35 patients implanted with straight electrodes, 14 of them had facial nerve stimulation, and 24 implants with curved, perimodiolar electrodes, without any stimulation of the facial nerve.

Electrode insertion difficulties were also reported in this study for the otosclerosis group, in ten patients there was ossification of the round window and in seven ossification of the tympanic ramp. In the control group, partial ossification of the tympanic ramp was observed in four patients. The difficulty of insertion of electrodes is another difficulty in cochlear implant surgeries in patients with otosclerosis reported by the great majority of authors.[5,7,9,13,14]

Conclusion

Patients with otosclerosis who underwent cochlear implant treatment had good hearing results even after five years after implantation, similar to those presented by patients with hearing loss due to other causes. In patients with implanted otosclerosis the number of complications as facial nerve stimulation was higher.

Considering the studied parameters of patients undergoing cochlear implantation in patients with otosclerosis, there was great auditory benefit with few surgical complications and little difficulty in programming the equipment.

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Conflicts of interest

Author declares there are no conflicts of interest.

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