

Original Article

Long-Term Outcome of Cochlear Implantation in Post-meningitic Deafness

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BACKGROUND: This study was planned (1) to evaluate long-term outcome after cochlear implantation in patients with post-meningitic deafness and (2) to compare the outcome measures with patients implanted for deafness due to other causes.

METHODS: Records of 54 patients deafened as a sequel of bacterial meningitis and implanted at the largest university-based cochlear implant program in Turkey were retrospectively reviewed. Fifty-four age- and sex-matched patients with a similar interval of implant use were selected for controls. Surgical and long-term audiological outcome (in terms of categories of auditory performance-II scores) was assessed and compared.

RESULTS: Twenty-seven (52%) patients had some degree of labyrinthitis ossificans and 19 of them had full electrode insertion via basal turn cochleostomy. Patients with and without labyrinthitis ossificans in the post-meningitic group had no difference in final categories of auditory performance-II scores (P = .559). Median categories of auditory performance-II scores were 6 for post-meningitic group and 7 for controls, with a significant statistical difference (P < .001). Partial or full insertions did not differ in outcome (P = .938). Mean time to implantation was not correlated with the final categories of auditory performance-II score for the post-meningitic group (P = .695).

CONCLUSION: Cochlear implant recipients deafened due to meningitis have a worse long-term hearing and speech performance as measured by categories of auditory performance-II than patients implanted for congenital deafness. The presence of labyrinthitis ossificans or the limited extent of electrode insertion produced overall results that were comparable with other cases.

KEYWORDS: Cochlear implant, hearing loss, meningitis, labyrinthitis ossificans

INTRODUCTION

There are unique challenges to cochlear implantation (CI) in this patient group. It is established that labyrinthitis ossificans (LO) resulting from meningitis may obliterate cochlear spaces and render CI unfeasible. Scala tympani, the usual target for electrode insertion, is primarily affected in its basal segment by LO² due to its intimate connection with the subarachnoid space via the cochlear aqueduct, and LO in this location may produce an obstruction to basal turn cochleostomy. The auditory neural pathway may also be adversely affected by meningitis, and post-implant rehabilitation may be hampered by additional central nervous system (CNS) sequelae.³ In spite of these obstacles, CI without delay remains the sole recourse short of an auditory brainstem implant for rapidly developing LO and profound hearing loss after meningitis. The aim of this article is to review a series of patients implanted for post-meningitic deafness, evaluate surgical success and auditory performance outcome in this cohort, and compare these results with a control group that had CI due to profound hearing loss due to congenital deafness.



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MATERIALS AND METHODS

Patient Characteristics

The medical records of patients who underwent cochlear implant surgery due to bilateral severe to profound SNHL in our tertiary referral center from January 1999 to December 2015 were retrospectively reviewed. Cases with meningitis recorded as the etiology of deafness were selected for the post-meningitic implant group (PMG). A control group (CG) matched with the PMG with respect to age and interval of cochlear implant use was assembled of cases implanted due to congenital hearing loss of hereditary or unknown etiology that had onset at birth. Post-meningitic implant group records were analyzed for the presence of labyrinthine ossification in preoperative high-resolution temporal computerized tomography (CT) and magnetic resonance imaging (MRI). Ossification in imaging was classified into 3 groups: gross, partial, and no ossification as described by Axon et al (Table 1).4 Indicators of operative success (possibility of a basal turn cochleostomy, extent of electrode insertion, alternative techniques such as circummodiolar drillout (CMD) or double electrode (DE) insertion if performed) were evaluated for each patient. Patients with poor adherence to follow-up, unrevised device (hard) failure, and insufficient chart records were excluded from the study. Patients with neurological comorbidities were not included in the CG group. The research was evaluated and approved by the institutional ethical committee.

Audiological Evaluation

Both groups were evaluated for hearing outcome as measured by categories of auditory performance-II (CAP-II) and sentence recognition test scores at the final follow-up. Categories of auditory performance-II^{5,6} is a standardized score of 10 categories of increasing sound/speech awareness and capability of spoken communication (Table 2). Patients are assigned to a category ranging from 0 ("No awareness of environmental sounds or voice") to 9 ("Use of telephone with an unknown speaker in unpredictable context). The highest CAP-II categories attainable depend on the age of the patient (as telephone use or group conversation are skills achieved later in life).

Statistical Testing

For statistical comparison across groups, Mann–Whitney *U*-test was used for non-parametric data or non-normal distributions, and student's *t* test was used for parametric data. Correlation between non-parametric variables was tested with Kendall's τ -b. SPSS version 23 for Mac OS X (IBM, Armonk, NY, USA) was utilized for statistical testing.

RESULTS

The PMG comprised of 54 patients (24 females, 30 males) implanted due to post-meningitic deafness, and the CG included 54 patients

Table 1. Axon Classification for Labyrinthine Ossification

Degree of Ossification	Finding
None	No ossification
Partial	Ossification localized to the basal turn of the scala tympani
Gross	Gross ossification of the scala tympani and variable amounts of the scala vestibuli

(24 females, 30 males) implanted for deafness not related to meningitis. All patients had bilateral severe to profound SNHL preoperatively. The mean age at implantation was 12.5 years for the PMG and 12.9 years for the CG with no statistically significant difference between groups (P=.891, t test). Mean interval of implant use was 10.3 ± 3.8 years for the PMG and 8.3 ± 3.1 years for controls.

Out of 54 patients in the PMG, there were 27 (50%) with some degree of LO in temporal CT or cochlear fibrosis in temporal MRI. Three cases (5%) had evidence of gross obliteration of cochlear spaces in preoperative imaging, while the remaining 24 (44%) had partial ossification. Partial ossification almost always included the cochlear basal turn in this patient group. Twenty-seven patients (50%) had no evidence of LO in preoperative CT or MRI. All 27 patients with no evident LO in imaging were fully inserted via a basal turn cochleostomy. Of the 3 patients with gross total LO in temporal imaging, 1 required a CMD procedure as described by Gantz et al⁷ and 2 were implanted with DE arrays with an additional second turn cochleostomy. Nineteen of 24 patients with findings of partial ossification in imaging were found to have an available cochlear lumen after clearing basal turn fibrous/osteoid tissue and fully inserted via a basal turn cochleostomy. Five were partially inserted, 2 of which required an ascending turn cochleostomy drilled inferior to the cochleariform process, anterior to the oval window to identify a cochlear lumen. A total of 6 revision procedures were required in PMG patients. Notably, 1 patient with a DE array had early revision due to electrode malposition. One case was revised due to skin complications, and 4 eventually had their implants replaced due to device failure. Patient characteristics for the PMG cases are presented in detail in Table 3. Patients in the CG had no labyrinthine ossification in preoperative imaging and were all fully inserted via a basal turn cochleostomy with no complications.

In the PMG, 13 of the 54 patients (24%) had co-morbid conditions that may have altered outcome, such as visual impairment in 3 cases, hydrocephalus in 3 cases, learning disability and/or attention-deficit hyperactivity disorder in 4 cases, autism spectrum disorder in 1 case, seizures in 1 case, and global developmental delay in 1 case.

Median CAP-II score for the PMG was 6 (Table 4). There was no statistically significant difference between the distribution of CAP-II scores

 Table 2. Categories of Auditory Performance II (CAP-II) Scale

CAP-II Score	Corresponding Skill
0	No awareness of environmental sounds or voice
1	Awareness of environmental sounds
2	Response to speech sounds
3	Identification of environmental sounds
4	Discrimination of speech sounds without lip reading
5	Understanding of common phrases without lip reading
6	Understanding of conversation without lip reading
7	Use of telephone with known speaker
8	Follows group conversation in a reverberant room or where there is some interfering noise, such as a classroom or restaurant
9	Use of telephone with an unknown speaker in unpredictable context

 Table 3.
 Patient Characteristics for Cases Implanted Due to Post-meningitic Deafness

Post-op SRT	77	50	10	100	80	24	45	77	100	73	0	35	94	88	0	0	82	50
Post-op P CAP-II	5	5	4	9	9	4	5	9	9	5	0	5	7	7	-	3	9	5
Additional Disabilities	-	-	-	ADHD	-	-	-	ı	Cleft palate	-	Global developmental delay	1	-	ı	Learning disorders	Learning disorders	1	
Cl Brand	Nucleus	Nucleus	Medel	Nucleus	Clarion	Nucleus	Nucleus	Medel	Nucleus	Nucleus	Nucleus	Nucleus double array	Medel	Nucleus	Nucleus double array	Nucleus	Nucleus	Nucleus
Revision	No	No	No	No	No	No	No	No	No	No	8 0	No	No	No	No No	No	No	No
Active Electrode %	100	100	83	91	100	100	91	83	100	100	100	09	92	100	50	91	91	100
Laterality	Unilateral/right ear	Bilateral (simultaneously)	Unilateral/left ear	Unilateral/right ear	Unilateral/right ear	Unilateral/right ear	Unilateral/left ear	Unilateral/left ear	Unilateral/left ear	Unilateral/right ear	Unilateral/right ear	Unilateral/right ear	Unilateral/left ear	Unilateral/left ear	Unilateral/right ear	Unilateral/right ear	Unilateral/right ear	Unilateral/left ear
Extent of Insertion	Full	Full	Full	Drillout	Full	Full	Full	Full	Full	Full	Full	Partial	Full	Full	Partial	Full	Full	Full
Insertion Method				СМД								DE			DE			
MRI	Normal	Cochlear fibrosis	Cochlear fibrosis	Cochlear fibrosis	Normal	Normal	Cochlear fibrosis	Normal	Lateral SCC fibrosis	Cochlear fibrosis	Normal	Cochlear fibrosis	Normal	Normal	Cochlear fibrosis	Normal	Cochlear fibrosis	Cochlear
ΓΟ3	None	Partial	Partial	Gross	None	None	None	None	Partial	Partial	None	Gross	None	None	Gross	None	Partial	Partial
Pre- Postlingual	Prelingual	Prelingual	Prelingual	Perilingual	Perilingual	Prelingual	Postlingual	Perilingual	Prelingual	Prelingual	Prelingual	Postlingual	Postlingual	Postlingual	Prelingual	Prelingual	Prelingual	Prelingual
Duration of Implant Use (yr)	16	7	7	14	10	13	11	5	6	10	17	11	12	7	10	11	11	∞
Age at Implantation (mo/yr)	7 yr	5 mo	5 yr	4 yr	44 yr	12 yr	21 yr	12 yr	3 yr	12 yr	11 yr	10 yr	26 yr	57 yr	3 yr	3 yr	2.5 yr	7 yr
Age at Meningitis (mo/yr)	7 mo	4 mo	1 mo	3 yr	3 yr	6 mo	21 yr	3 yr	N/A	11 mo	3 mo	6 yr	7 yr	N/A	6 mo	N/A	om 6	N/A
Pt.	_	2	8	4	2	9	7	8	6	10	Ξ	12	13	4	15	16	17	18

 Table 3. Patient Characteristics for Cases Implanted Due to Post-meningitic Deafness (continued)

 Table 3.
 Patient Characteristics for Cases Implanted Due to Post-meningitic Deafness (continued)

Post-op SRT	0	55	45	06	77	43	40	06	65	88	45	42	06	20	0	80	32	y; SCC, semi-
Post-op CAP-II	m	5	5	7	9	5	5	9	9	9	5	5	8	9	4	80	4	cochleostom
Additional Disabilities	Epilepsy	ı	1	1	1	1	1	1	1	-	ı	1	1	Hydrocephalus	ı	ı	Vision impairment	AC, ascending turn
CI Brand	Nucleus	Nucleus	Nucleus	Medel	Nucleus	Nucleus	Nucleus	Medel	Nucleus	Medel	Nucleus	Nucleus	Medel	Nucleus	Nucleus	Nucleus	Nucleus	le electrode;
Revision	° N	No	No	ON.	ON	ON	ON.	O Z	No	Yes/ device failure	No	No	Yes/ electrode malposition	No	N O N	Yes/device failure	ON	ar drillout; DE, doub
Active Electrode %	06	91	95	100	91	80	100	100	100	20	100	91	42	16	%56	91	58	rcummodiol
Laterality	Unilateral/right ear	Unilateral/left ear	Unilateral/right ear	Unilateral/right ear	Unilateral/right ear	Unilateral/right ear	Unilateral/left ear	Unilateral/right ear	Unilateral/left ear	Unilateral/left ear	Unilateral/left ear	Unilateral/right ear	Unilateral/left ear	Unilateral/right ear	Unilateral/right ear	Unilateral/left ear	Unilateral/right ear	of auditory performance; SRT, sentence recognition test, CMD, circummodiolar drillout; DE, double electrode; AC, ascending turn cochleostomy; SCC, semi-
Extent of Insertion	Full	Full	Full	Full	Full	Full	Full	Partial	Full	Partial	Full	Full	Full	Full	Full	Full	Full	T, sentence re
Insertion Method								AC					DE					rformance; SF
MRI	Lateral SCC fibrosis	Normal	Cochlear fibrosis	Normal	Normal	Normal	Cochlear	Lateral SCC fibrosis	Normal	Cochlear fibrosis	Normal	Cochlear fibrosis	Cochlear fibrosis	Normal	Normal	Cochlear fibrosis	Cochlear fibrosis	f auditory pe
FO3	Partial	None	Partial	None	None	None	Partial	Partial	None	Partial	None	None	Partial	None	None	Partial	Partial	\ S
Pre- Postlingual	Perilingual	Prelingual	Prelingual	Prelingual	Postlingual	Prelingual	Prelingual	Prelingual	Postlingual	Prelingual	Postlingual	Postlingual	Postlingual	Prelingual	Prelingual	Postlingual	Prelingual	ificans; CAP-II, c
Duration of Implant Use (yr)	17	17	8	1	13	14	4	10	17	11	8	10	6	ю		2	10	yrinthitis oss
Age at Implantation (mo/yr)	7 yr	16 yr	11 yr	20 mo	12 yr	15 yr	3 yr	8 yr	26 yr	16 yr	22 yr	41 yr	5 yr	4 yr	4 yr	26 yr	7 yr	Pt, patient, mo, months; yr, years; LO, labyrinthitis ossificans; CAP-II, categories
Age at Meningitis (mo/yr)	3 mo	4 mo	N/A	N/A	5,5 yr	N/A	N/A	2 yr	7 yr	N/A	N/A	7 yr	5 yr	N/A	N/A	N/A	N/A	ent; mo, month
Pt. No.	38	39	40	14	45	43	44	45	46	47	48	49	50	51	52	53	54	Pt, patie

circular canal; ADHD, attention deficit-hyperactivity disorder; MRI, magnetic resonance imaging; CI, cochlear implant.

Table 4. FINAL CAP-II Scores, Age at Implant, Time to Implant, and Duration of Implant Use Among Subgroups

	CAP-II (Median (Range))	Age at Implant (Mean (Years))	Time to Implant (Mean \pm SD (Months))	Duration of Implant Use $(Mean \pm SD (Years))$
Post-meningitic group (overall)	6 (0-8)	12.5	67	10.3 ± 3.8
LO positive	6 (1-8)	7.8	50	10.3 ± 2.9
LO negative	5 (0-8)	17.5	84	10.3 ± 4.7
Control group	8 (5-9)	12.9		8.3 ± 3.1

CAP-II, categories of auditory performance; LO, labyrinthitis ossificans; SD, standard deviation.

among cases with or without LO (P=.559, Mann–Whitney U-test). A frequency distribution of CAP-II scores of patients with and without LO is presented in Figure 1. Median CAP-II score for the CG was 8, and the difference between the PMG and CG in CAP-II was statistically significant (P < .001, Mann–Whitney U-test). A visual comparison of CAP-II scores of patients in the PMG and CG is presented in Figure 2.

Among patients who required non-standard insertion techniques, the patient implanted via a CMD achieved a post-op CAP-II score of 6, while both patients who were implanted with DE arrays had a CAP-II outcome of 1 (sound awareness with no response to speech). In the PMG, 46 cases who were fully inserted had a median CAP-II score of 5, while 8 who had partial insertions had a median CAP-II score of 6. This difference in CAP-II score distributions between partial and full insertions was not statistically significant (P=.938, Mann–Whitney U-test).

Patients implanted due to post-meningitic deafness were evaluated for the impact of time from the onset of deafness to any intervention for auditory rehabilitation (either with a hearing aid or cochlear implant) on the outcome as measured by the CAP-II score. Mean time to implantation (TTI) was 67 months for this group, but no significant correlation was identified between TTI and CAP-II outcomes (Kendall's τ -b= -0.044, P=.695). A scatterplot of CAP-II outcome with regard to age at implantation is presented in Figure 3.

DISCUSSION

Cochlear implantation in the setting of post-meningitic deafness has been a controversial subject since the advent of implant surgery. Earlier reports suggest that due to lack of former auditory stimulation, congenitally deaf children would be outperformed by their counterparts

with acquired deafness.^{8,9} Particularly after the recognition of additional barriers posed on rehabilitation by neurologic sequelae of meningitis, this outlook reversed in favor of the congenitally deaf,¹⁰⁻¹² with occasional reports revealing equivalent results.¹³ The apparent contraindication has remained unsolved in current opinion and may be associated with numerous outcome-influencing factors: the presence of LO or additional CNS sequelae, insertion technique, age at implantation, or time elapsed from the onset of deafness to CI surgery.

The incidence of any extent of LO has been reported to range from 48.7% to 62% in recent series. 14-16 A comparison of CI outcome between subjects with and without LO has yielded worse results for LO-positive cases in earlier papers. El-Kashlan et al¹⁷ found decreasing mean speech perception categories (SPC) for patients with worsening LO with the gross ossification group obtaining a mean SPC of 3 ("beginning word identification") within 24 months postoperatively. The authors attribute this finding to a significantly better preoperative residual hearing in patients with patent cochleae. Philippon et al¹⁶ reported an inverse correlation between auditory performance and LO only if cases with stage III ossification according to Smullen and Balkany¹⁸ (>180° ossification of the basal turn) are included in the comparison. Recent investigations reveal a modest advantage favoring LO-negative cases: Nichani et al¹⁵ have found that 88% of LO-negative versus 74% of LO-positive CI recipients achieved open-set speech with a mean CAP score of 5.9 and 5.4, respectively. Liu et al.14 on the other hand, report no statistically significant difference between SPC outcomes of cases with and without LO. The present cohort has a 46% partial and 6% gross ossification rate that is consistent with previous literature, and our results confirm that if at least partially inserted, cases with LO have statistically equivalent outcome with that of LO-negative patients.

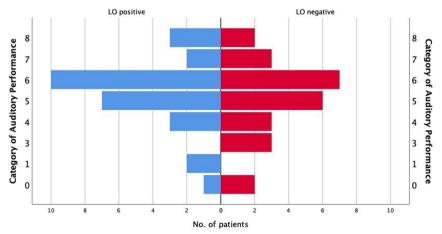


Figure 1. Comparison of final CAP-II scores of study group patients with and without LO. Mann–Whitney *U*-test, *P*=.559. CAP-II, categories of auditory performance; LO, labyrinthitis ossificans.

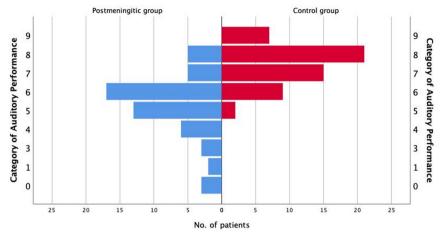


Figure 2. Comparison of final CAP-II scores for the post-meningitic and control groups. Mann–Whitney U-test, P < .001. CAP-II, categories of auditory performance.

One case with gross ossification that achieved remarkable auditory performance with a CAP-II score of 6 was implanted via CMD. Split or DE arrays have given dismal performance, though, with both cases partially inserted with DEs achieving only sound awareness. This finding mirrors that of Nichani et al¹⁵ who reported 4 of the 7 split insertions in their series had a final CAP<5.

For a comparative evaluation of post-meningitic and congenital deafness cases, a majority of previous research reveals no significant difference in postoperative hearing outcome with regard to etiology of deafness¹9-21 Nikolopoulos et al²¹ report that 77% of congenitally deaf patients versus 73% of post-meningitic deafness cases have achieved a CAP score of ≥5. Both etiologic groups in the series of Bille et al.¹9 have a median CAP of 6 and speech intelligibility rating of 4. The findings of El-Kashlan et al¹7 are contradictory and demonstrate a markedly different mean SPC for post-meningitic cases and controls (3.7 vs. 5.1, respectively). Results of our cohort support the latter research, with PMG patients attaining significantly lower CAP-II scores compared to controls. This apparent difference may be due to alteration of central auditory processing capability after meningitis, of which there currently is no objective method of testing in CI recipients. Another factor of note may be the inclusion of 2 new categories

to the CAP score that measure previously untested skills, such as the ability to follow group conversation.

Time to implantation from onset of deafness is another important consideration for CI outcome. Durisin et al²² achieved significantly better results in post-meningitic patients implanted within 6 months from the onset of hearing loss. Our analysis did not reveal any significant correlation between the time to implant and final CAP-II scores for PMG patients. In the current medical era with wider access to healthcare, it may be surmised that children with meningitis have an expedited course to CI due to the perceived urgency of the illness. This temporal advantage does not translate into improved outcome, however, as evidenced by a trend toward equivalent or worse results in post-meningitic implant recipients.

Our series has one of the longest mean durations of implant use (over 10 years for PMG) hitherto reported in the English literature, and we believe it is reasonable to state that our results represent the final performance attainable by these patients. Certain limitations of this research are the inclusion of cases with a wide range of ages and prelingual/postlingual patients in the same group. Despite these shortcomings, our data indicate that post-meningitic implant recipients

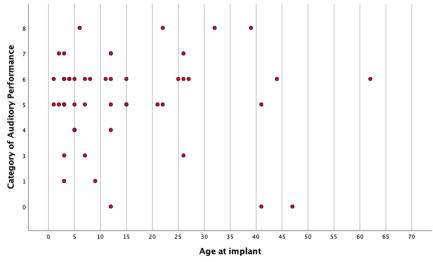


Figure 3. Final CAP-II score distribution in the post-meningitic group with regard to age at implantation. CAP-II, categories of auditory performance.

have poorer outcome than congenitally deaf patients, irrespective of the presence of labyrinthine ossification. The root cause of this finding may lie in a central auditory processing difficulty, co-morbid developmental/neurologic impairment, or a global CNS dysfunction as a sequel of meningitis. Whether central auditory processing is affected by an infectious process such as meningitis remains hitherto undefined in the literature.

CONCLUSION

In this study, cochlear implant recipients who were deafened as a sequel of meningitis had a worse long-term outcome than that of patients with deafness due to congenital causes. The final overall outcome is unaffected by the presence of labyrinthine ossification and the extent of electrode insertion. Central nervous system sequelae may also contribute to hearing loss in this patient group. Further research is needed to objectively assess the central auditory pathway in post-meningitic deafness.

Ethics Committee Approval: The research was evaluated and approved by the Hacettepe University institutional review board.

Informed Consent: Informed consent was obtained from all participants.

Peer Review: Externally peer-reviewed.

Author Contributions: Authors OMA and BÖ handled data collection and manuscript preparation. DB, GS and LS designed the study, reviewed and advised the manuscript. LS is the senior surgeon who operated a significant majority of the cases.

Conflict of Interest: The authors have no conflict of interest to declare.

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