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REVIEW ARTICLE

Cochlear implantation for single-sided deafness: the outcomes. An evidence-based approach

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Abstract The aim of the present paper is to critically review the current evidence on the efficacy of cochlear implantation as a treatment modality for single-sided deafness (SSD), and/or unilateral tinnitus. Systematic literature review in Medline and other database sources was conducted along with critical analysis of pooled data. The study selection includes prospective and retrospective comparative studies, case series and case reports. The total number of analyzed studies was 17. A total of 108 patients with SSD have been implanted; 66 patients due to problems associated with SSD, and 42 primarily because of debilitating tinnitus. Cochlear implantation in SSD leads to improved sound localization performance and speech perception in noise from the ipsilateral side with an angle of coverage up to (but not including) 90° to the front, when noise is present in the contralateral quartile (Strength of recommendation B). Speech and spatial hearing also subjectively improve following the insertion of a cochlear implant (Strength of recommendation B); this was not the case regarding the quality of hearing. Tinnitus improvement was also reported following implant placement (Strength of recommendation B); however, patients need to be advised that the suppression is mainly successful when the implant is activated. The overall quality of the available evidence supports a wider use of cochlear implantation in SSD following appropriate selection and counseling (overall strength of recommendation B). It remains to be seen if the long-term follow-up of large number of patients in well conducted high quality studies will confirm the above mentioned results.

Keywords Cochlear implant · Deafness · Unilateral · Tinnitus · Single-sided hearing loss · Sound localization · Speech perception in noise · Quality of life

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Introduction

Cochlear implants represent one of the most important achievements of modern medicine, as for the first time in history an electronic device is able to restore a lost or never existed sense—hearing [1]. More than 2,00,000 have been implanted so far, and this number is steadily increasing despite the related cost [2].

Significant advances in technology and better knowledge of the outcomes are constantly changing the criteria for cochlear implant candidacy. This is mainly because implanted patients are now obtaining increasing amounts of open-set word recognition with the available devices [3]. Hence, changes in patient selection have included implanting adult candidates with residual hearing, and children at younger ages, or with additional disorders.

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On the other side, historically, patients with single-sided deafness were rehabilitated either with contralateral routing of the incoming sound using specialized hearing aids (CROS amplification), or, with the advent of bone-anchored hearing aid (BAHA) technology, through the contralateral transmission of skull vibrations to the hearing ear [4]. The situation was further perplexed by the frequent presence of tinnitus, often debilitating, in the deafened ear for which rehabilitation techniques based on acoustic input (i.e., retraining, masking) were not possible.

Although in the first decades of cochlear implantation such an intervention would seem outrageous in unilaterally deaf people, the option of cochlear implantation gradually emerged as a possible method of rehabilitation in single-sided deafness with or without tinnitus, predominantly in adults, but also including pediatric patients [5–10].

The aim of the present paper is to critically review the current evidence on the efficacy of cochlear implantation as a treatment modality for single-sided deafness. The effect of cochlear implantation on unilateral tinnitus will also be explored.

Materials and methods

An extensive search of the literature was performed in Medline and other available database sources until May 2013, having three primary end-points: (a) to assess the effect of cochlear implantation in the speech comprehension in noise of unilaterally deafened patients, (b) to assess the effect of cochlear implantation in the sound localization ability of unilaterally deafened patients, and (c) to assess the clinical effectiveness of cochlear implantation as a treatment option for unilateral tinnitus. In addition, the clinical effectiveness of pediatric cochlear implantation for single-sided deafness was also assessed.

The inclusion criteria consisted of ipsilateral cochlear implantation in the deafened ear, in the presence of normal (or at least functional) hearing in the contralateral ear. Implantations due to unilateral tinnitus were also included, if the absence of a retrocochlear lesion had been excluded by means of MRI imaging. Only postlingually deafened adults and children with speech acquisition were included in the study population.

During the search, the keywords "unilateral," "deafness," "tinnitus," "cochlear," "implant," and "pediatric" were utilized. The keywords "unilateral," "deafness," and "cochlear" were considered primary, and were either combined to each of the other keywords individually, or used in groups of three. Reference lists from the retrieved articles were also manually searched. Language restrictions limited the included literature into English- and German-speaking articles.

Table 1 Levels of evidence regarding the primary research question in studies that investigate the results of a treatment (http://www.cebm.net/index.aspx?o=1025)

Category of evidence	Study design
Level I	• High quality randomized trial with statistically significant difference, or no statistically significant difference but narrow confidence intervals
	• Systematic review of Level I randomized control trials (and study results were homogenous)
Level II	• Lesser quality randomized control trial (e.g., <80 % follow-up, no blinding, or improper randomization)
	• Prospective comparative study
	• Systematic review of Level II studies or Level 1 studies with inconsistent results
Level III	• Case control study
	• Retrospective comparative study
	• Systematic review of Level III studies
Level IV	• Case series
Level V	• Expert opinion

Table 2 Strength of recommendation by category of evidence for guideline development [11]

Strength of recommendation	Category of evidence
A	Directly based on category I evidence
B	Directly based on category II evidence or extrapolated recommendation from category I evidence
C	Directly based on category III evidence or extrapolated recommendation from category I or II evidence
D	Directly based on category IV evidence or extrapolated recommendation from category I, II or III evidence

Results

Twenty-seven studies met the inclusion criteria and were initially included in study selection. They comprised 11 prospective and four retrospective comparative studies, two case series, four case reports, one cross-sectional study, four systematic reviews and one book. Overlapping patient populations were found in a total of 11 studies. The demographics, audiometric and tinnitus data of the operated patients were carefully examined to avoid double counting of cases. Eleven centers which had performed cochlear implantation in unilaterally deafened patients with or without tinnitus were finally identified. Using this framework of results, the respective studies were then critically appraised, according to evidence-based guidelines for the categorization of medical studies (Tables 1, 2) [11]. Overall, ten prospective and three retrospective comparative studies, two case series and two case reports which had performed cochlear implantation as a treatment for single-sided deafness and/or tinnitus in unilateral deafness were systematically analyzed (Table 3).

A total of 108 patients with single-sided deafness have received a cochlear implant. The implantation was performed due to problems associated with the unilateral deafness in 66 patients, and primarily because of debilitating tinnitus in 42 cases. Among the implant recipients, four children were identified. None of those was implanted because of tinnitus.

Eight research groups had investigated the effect of cochlear implantation in treating unilateral tinnitus in the deafened ear. Five studies represented Level II, one Level III, and two Level IV evidence. Outcomes were available for 85 patients, although tinnitus alone was not the primary reason for implantation in all patients. Tinnitus improvement was reported in 81 implantees (95.3 %), with complete suppression of tinnitus occurring in 15 of the 44 patients for whom sufficient data were available (34.1 %). Despite the observed improvement in the vast majority of cases, statistically significant results were reported in only one Level II study ($n = 26$ patients).

Six research groups assessed the sound localization abilities of implant recipients with unilateral deafness. Three studies represented Level II, one Level III, and two Level IV evidence. The respective results referred to 63 operated patients. All researchers reported improved competence following the insertion of the cochlear implant. Statistically significant results were reported in one Level II and one Level IV study ($n = 25$ patients). The remaining studies did not have any statistical analysis.

Speech perception in noise was investigated by seven implant groups. Five studies represented Level II, one Level III, and one Level IV evidence. The respective results referred to 85 operated patients. Six research groups concluded that the insertion of the cochlear implant leads to improved perception of speech from the deafened ($n = 4$) or front side ($n = 3$) (both angles were tested in one study), when noise is coming either from the front ($n = 1$), or from the normally hearing ear ($n = 5$). The remaining study (Level III evidence) found neither improved nor impaired perception of speech from the deafened side when noise was coming from the front. Statistically significant results were reported in four Level II studies ($n = 50$ patients). The results regarding speech perception in noise in all other signal–noise configurations were contradictory, and as such inconclusive.

Finally, five research groups had also assessed the outcome of the operation from the patients' perspective, using a self-assessment questionnaire. The respective results referred to 57 operated patients. Four studies represented Level II, and one Level IV evidence. Statistically significant results were obtained in four Level II studies ($n = 43$ patients). Among the three dimensions of the questionnaire (speech, spatial, and qualities of hearing), the speech and spatial components were unanimously reported as improved. The qualities of hearing component were reported as not improved in one Level II, and one Level IV study.

The insertion of the cochlear implant was followed by consistent use in 83 of the 86 patients for whom data were available. The median follow-up period of the implanted patients was 2 years (Fig. 1), but the reported results in eight of the 11 studies were based on follow-up intervals of 1 year or less (Fig. 2). No perioperative or postoperative complication was reported in any of the operated patients.

Discussion

The criteria for cochlear implantation have broadened overtime, as substantial benefits for both adult and pediatric patients along with a safe surgical technique have been established, the implant technology has improved, and our familiarity with the devices has increased [4, 24–27]. However, every change in the selection criteria of cochlear implantation, especially in the era of evidence-based medicine needs to be based on accumulated data which are systematically and critically assessed, so that they can be approved by the medical community and the respective bodies of administration. This was the aim of the present study, which sought to examine the effectiveness of cochlear implantation with regard to sound localization and speech perception parameters of patients with single-sided deafness, as well as its effectiveness in treating the ipsilateral tinnitus from which such patients may suffer.

Drawing on the audiologic parameters, the sound localization performance of unilaterally deafened implantees improves after the insertion of the cochlear implant. Although statistical analysis was lacking in some of the related studies, based on the quality of the studies and the unanimity of the reported results, the strength of the respective recommendation can be graded as B. Indeed, while unilaterally deaf listeners rely on the head shadow (and to a lesser degree on the pinna) cue when localizing the sound in the horizontal plane [28], binaural hearing allows accurate azimuthal localization, due to the simultaneous utilization of multiple psychoacoustic phenomena from the two independent acoustic sensors [4]. By implanting a patient with unilateral deafness, the difference between the true and perceived azimuth angle of the sound stimulus is significantly reduced, thereby producing a binaural advantage for the respective localization ability.

Table 3 Cochlear implantation in single-sided deafness (patient series)

Authors	Study type	Evidence level	Implant type	Total number of patients	Consistent use	Follow-up	Remarks
Tavora-Vieira et al. 2013 [12]	Prospective comparative	II	Med-EL	9	9/9	3 months	a) Implanted patients report integration of the implant sound with the hearing of the NH ear b) Hearing in the most challenging situations is improved
Plontke et al. 2013 [13]	Case report	IV	Nucleus	1	n.r.	6 months	CI in an acutely deafened child can lead to rapid development of good speech discrimination with the device and improvement in speech perception/sound localization
Hassepass et al. 2012 [10]	Prospective comparative	II	Nucleus	3	3/3	12 months	a) There were indications for the presence of binaural squelch b) There was improvement in sound localization acuity due to binaural listening in the CI-aided condition, compared with the unilateral listening before implantation
Firszt et al. 2012 [14]	Case series	IV	Nucleus	3	3/3	≥8 months	a) CI did not improve nor hinder sentence understanding or speech perception in noise b) Word recognition in quiet and sound localization was better after implantation compared with the NH alone
Arndt et al. 2011 [15] Arndt et al. 2011 [16] Arndt et al. 2010 [9][a]	Prospective comparative	II	Nucleus[b]	22	10/11[b]	≥6 months	a) Speech understanding in noise is significantly negatively correlated with the duration of deafness b) Short duration of deafness probably enables better binaural processing c) CI is superior to alternative rehabilitation options in patients with SSD, after careful preoperative diagnostic testing and patient selection
Jacob et al. 2011 [17] Jacob et al. 2011 [18] Stelzig et al. 2011 [19][a]	Retrospective comparative case series	III IV	Nucleus/Med-EL	25	25/25	≥8 months	a) CI is significantly better than CROS and BAHA amplification in patients with SSD b) The restoration of binaural hearing is associated with improved balance tests in most patients, and an increase in balance control of 10–30 % c) The hearing quality of the NH ear is not disturbed
Kleine Punte et al. 2011 [20] Vermeire and van de Heyning 2008 [7] van de Heyning et al. 2008 [5][a]	Prospective comparative	II	Med-EL	26[c]	20/20[c]	24 months[d]	a) All patients reported significant benefit in tinnitus when the CI was activated, both in occurance and in intensity b) The tinnitus reoccured in its original loudness in 92 % of patients when the CI was switched off c) There is no tinnitus adaptation to the electrical stimulation of the CI, or recurrence overtime
Ramos et al. 2011 [21]	Prospective comparative	II	Nucleus	10	10/10	≥3 months	Tinnitus suppression can be achieved with CI in SSD, even in the presence of preserved residual hearing
Roland et al. 2011 [22]	Retrospective comparative	III	Nucleus	3	n.r.	≥2 months	There is significant improvement in speech perception performance in quiet and noise in patients with SSD following CI

Table 3 continued

Authors	Study type	Evidence level	Implant type	Total number of patients	Consistent use	Follow-up	Remarks
Buechner et al. 2010 [23]	Prospective comparative	II	HiRes	5	3/5	12 months	CI in patients with unilateral tinnitus accompanying severe to profound HL may be beneficial, but needs to be examined on an individual basis
Kleinjung et al. 2009 [8]	Case report	IV	Med-EL	1	n.r.	3 months	There is no hearing conflict between implanted and NH ear

NH normal hearing, *CI* cochlear implant/cochlear implantation, *SSD* single-sided deafness, *CROS* contralateral routing of offside signal, *BAHA* bone-anchored hearing aid

[a] authors belong to the same investigating team

[b] data available for 11/22 patients

[c] data regarding consistent use and audiometric parameters based on 20 patients

[d] follow-up for audiometric data was 12 months

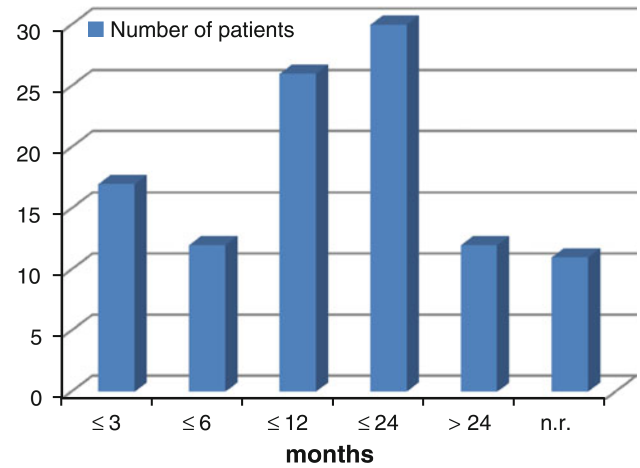

Fig. 1 Follow-up intervals of patients implanted for single-sided deafness

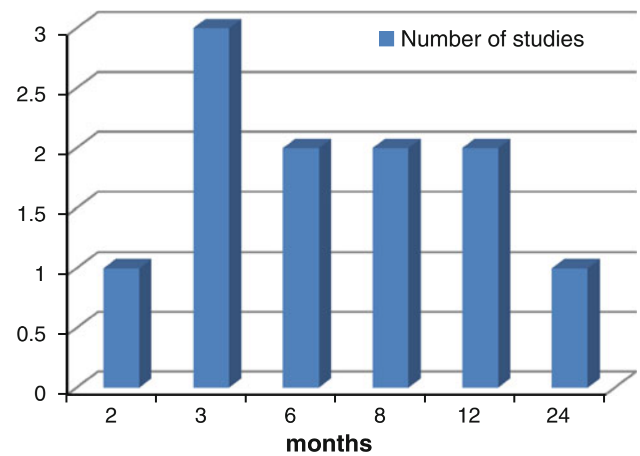

Fig. 2 Follow-up intervals of reported outcomes following cochlear implantation for single-sided deafness

With regard to speech perception in noise, the insertion of the cochlear implant leads to postoperative improvement from the ipsilateral side with an angle of coverage up to (but not including) 90° to the front, when noise is present in the contralateral quartile (strength of recommendation B). However, the observed responses in all other signal–noise configurations are highly variable across studies, and preclude us from drawing any conclusions, regarding the strength of the respective recommendations. Indeed, the advantage of binaural over monaural hearing with regard to the comprehension of speech, especially in noisy environments, is based on three parameters; the psychoacoustic effects of squelch and summation, and the effect of the volumetric size of the head on the intensity, timing, phase, and timbre of the sound, when it approaches biased to one ear over the other (head shadow effect) [4]. The observed improvement in the former abovementioned test conditions can be attributed to a combination of the head shadow, and squelch effects. The contradictory results in the latter test conditions indicate that the obtained central summation effect is not significant [6, 8]. This is probably because unilaterally deaf patients have clear interaural asymmetry, even after been aided by a cochlear implant [6, 29].

In addition to the audiometric workup, the utilization of self-assessment questionnaires to evaluate the outcome of the operation from the patients' perspective may add substantial information about daily performance, which may not be easily assessed in a laboratory setting. Indeed, five research groups had employed a speech, spatial, and qualities of hearing questionnaire to evaluate the subjective improvement from the use of the cochlear implant. Among the three dimensions of the questionnaire, the speech and spatial components were unanimously reported as deriving substantial benefit from the cochlear implant placement. Based on the quality of the respective studies, and the unanimity of the reported results, the strength of the respective recommendation can be graded as B. However, the presence of two studies, one representing Level II, and one Level IV evidence, which reported that the related qualities of hearing do not improve following cochlear implantation, although not indicating deterioration of the respective parameter, precludes us from drawing conclusions, regarding the strength of the respective recommendation.

With regard to the clinical effectiveness of cochlear implantation as a treatment option for unilateral tinnitus, marked improvement was reported in the vast majority of implanted patients (95 %), with complete suppression occurring in 34 % of cases for which sufficient data were available. However, one patient experienced temporary deterioration of tinnitus postoperatively, and another two deterioration of tinnitus when the implant was switched off, in comparison with the respective annoyance before placement of the implant. We should also take into account that only five of the 74 implanted patients, for whom there are available data, reported that they experienced the same improvement in their tinnitus, regardless of whether the implant was activated or not. Overall, and based on the quality of the respective studies, and the unanimity of the reported results, an improvement in debilitating unilateral tinnitus associated with single-sided deafness can be supported with a grade B strength of recommendation, although statistical analysis was performed in only one study (representing Level II evidence).

The electrophysiological base of the observed improvement lies upon the perceived deafferentation of the auditory nerve by the electrical stimulation of the cochlear implant, which theoretically reverses a cortical reorganization associated with peripheral deafferentation, which may be the cause of tinnitus [30, 31]. The ensuing increase in afferent information may also have inhibitory effects in the auditory nerve system, and in conjunction with the enhanced attentiveness to environmental sounds after the activation of the cochlear implant, may also result in less awareness and consequent reduction of tinnitus [20]. This approach differs considerably from habituation, coping, and sound enrichment strategies based on the Jastreboff neurophysiological model [32].

Although tinnitus may occur in children, particularly those with hearing impairment [33–35], and when intrusive can cause difficulties with sleep or concentration, and contribute to behavioral problems, none of the four children identified in the present analysis had been implanted because of tinnitus. In contrast, the children were implanted either due to upcoming cochlear fibrosis, difficulties in daily situations, or deterioration in academic performance. Unilateral deafness has indeed been associated with worse spoken language performance in school-aged children, compared with their normally hearing peers [36], whereas their socio-emotional development and health-related quality of life also seems to be affected [37, 38]. However, the speech perception in noise both from the ipsilateral and front sides improves in unilaterally deafened pediatric implantees compared to the preimplant unaided conditions, when noise is present in the contralateral quartile. This result seems to be better than the respective results of adult implantees. The sound localization performance and the speech and spatial components of the self-assessment questionnaire, along with the qualities of hearing domain also appear to be improved after the insertion of the cochlear implant, despite the presence of two different auditory stimulation modalities. It should be noted that the very small number of implanted children and the unavailability of statistical data regarding some of the studied parameters do not allow us to grade the respective recommendations.

In addition to the expected efficacy of cochlear implantation for the treatment of single-sided deafness, and/or unilateral tinnitus, appropriate preoperative informed consent requires that the patient (or parents) is aware of the potential risks which can be associated with this specific intervention (e.g., surgical complications). Nevertheless, cochlear implantation in unilaterally deafened individuals is a generally safe procedure; no perioperative or postoperative complications were reported so far in the operated patients.

A positive finding in our analysis was the highly consistent use of the device by the implant recipients. The fact that the respective percentage reached 96.5 % of patients, coupled with the seemingly harmonic integration of electrical and acoustic stimulations, the suppression of tinnitus, and the reported overall improvement in the speech and spatial components of hearing suggests that implanted patients are satisfied with the procedure. It should be noted, however, that statistical data were not always available in the included studies. Furthermore, the present meta-analysis did not identify any Level I studies regarding cochlear implantation in single-sided deafness. While the latter are very difficult or impossible to be conducted due to the nature of the surgical intervention, demonstrating statistical importance is not only desirable, but also necessary in the respective prospective and retrospective comparative studies (and if possible case series). Otherwise, the patient population which is used as a basis for the extracted recommendations may somehow limit the power of the obtained results.

We also have to take into account that the 108 implantees suffering from single-sided deafness, who were identified in the present meta-analysis, constitute a very selective group of patients, as they chose to proceed with a surgical procedure which is not yet widely accepted as a treatment modality in their condition. Therefore, a self-selection bias may be present in the reported outcomes, and the latter should be generalized with caution.

In addition, an appropriate follow-up period is of utmost importance for the assessment of treatment efficacy in cochlear implantation; this time-period may be up to 30 months for postlingually deafened children and adults [39–41]. Although the present analysis identified a median follow-up period of 2 years for the implanted patients (Fig. 1), the reported results in the majority of studies were based on follow-up intervals of 1 year or less (Fig. 2). There is a clear need of obtaining results based on longer follow-up periods, to delineate the indications, and further quantify outcomes and factors influencing the results of cochlear implantation in patients with single-sided deafness. This is because consistent and everyday use of the device by the implantees in the long-term is the most important outcome measure, as it verifies efficacy and, as such, justifies the intervention.

Conclusion

The criteria for cochlear implant candidacy are changing to include wider patient populations; however, the determination of implant candidacy is ultimately based on the best knowledge and judgment of the managing physician.

Although the outcomes of the 108 single-sided deaf implantees in the literature have certain weaknesses (e.g., short follow-up, various evaluation protocols, etc.,), the overall quality of evidence supports a wider use of cochlear implantation in single-sided deafness following appropriate selection and counseling (overall strength of recommendation B). It remains to be seen if the long-term follow-up of large number of patients in well conducted high quality studies will confirm the above mentioned results.

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