

Cochlear Implantation and Single-sided Deafness: A Systematic Review of the Literature

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Abstract

Introduction Current data show that binaural hearing is superior to unilateral hearing, specifically in the understanding of speech in noisy environments. Furthermore, unilateral hearing reduce onés ability to localize sound.

Objectives This study provides a systematic review of recent studies to evaluate the outcomes of cochlear implantation in patients with single-sided deafness (SSD) with regards to speech discrimination, sound localization and tinnitus suppression.

Data Synthesis We performed a search in the PubMed, Cochrane Library and Lilacs databases to assess studies related to cochlear implantation in patients with unilateral deafness. After critical appraisal, eleven studies were selected for data extraction and analysis of demographic, study design and outcome data.

Conclusion Although some studies have shown encouraging results on cochlear implantation and SSD, all fail to provide a high level of evidence. Larger studies are necessary to define the tangible benefits of cochlear implantation in patients with SSD.

Keywords

- ▶ unilateral hearing loss
- ▶ cochlear implantation
- ▶ single-sided deafness
- ▶ speech discrimination
- ▶ sound localization
- ▶ tinnitus

Introduction

Single-sided deafness (SSD) may cause many problems involving communication between people. Permanent acquired unilateral several-profound hearing loss has been estimated to affect between 12–27 persons in every 100,000 among the general population, with the majority of losses being sudden or idiopathic.¹ The most common impairment is difficulty in hearing sounds in the affected side due to the head shadow effect, which attenuates the high-frequency components of sounds at the ear contra-lateral to their source.² Other problems involved are: prejudice in word discrimination, difficulty in understanding speech particularly in noisy environments; constantly adjusting head to try and compensate for the handicap; restriction of onés ability to localize sounds; and, in some cases, leading to social isolation.³

The cochlear implant (CI) is one of the more recent treatment options for such cases. However, there is a

concern about the ability of the brain distinguish acoustic and electric stimuli and concern that the hearing from the cochlear implant would interfere with acoustic signal processing from the good ear. Contralateral routing of sound (CROS) and osseointegrated implants are also devices used as rehabilitative options for SSD, although they are not able to provide binaural hearing⁴ or improve sound localization.⁵

People with binaural hearing enjoy certain advantages. The first advantage is better speech-to-noise ratio (SNR), which improves speech understanding in noisy environments. A second advantage results from the processing of the input sound signal by the brain from both ears. The brain is able to separate noise and speech from different locations using distinct interaural timing, spectral cues and level, thus, refining intelligibility. A third possible advantage is related to the summation effect, responsible for improved speech perception through the identification of identical signals arriving in both ears.⁶

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Based on these facts, the use of rehabilitation methods that can restore bilateral auditory input could lead to an improvement in spatial hearing and speech perception in patients with SSD.

Review of Literature

Methods

In this article, we present some studies and reviews up to February, 2015, that analyze the influence of cochlear implantation in patients with SSD with regards to (a) sound localization, (b) speech perception, (c) tinnitus, and, (d) quality of life.

Search Strategy

A systematic search was performed in the PubMed, Embase Cochrane Library, and Lilacs databases leading up to February, 2015, using the following terms: SSD (or synonyms – see ►Table 1) and cochlear implantation.

Study Selection

While screening titles and abstracts, the authors excluded any duplicates, review articles, animal studies, case reports and articles written in languages other than English or Spanish. Studies published only in abstract were also excluded.

The inclusion criteria consisted of studies that analyzed patients with unilateral deafness that had undergone ipsilateral cochlear implantation, in the presence of normal or functional hearing in the contralateral ear. Implantations due to unilateral tinnitus were also included. Asymmetric hearing loss was not used as a keyword, as there is no international consensus on the definition.

Results

The search in PubMed, Cochrane Library, and Lylacs retrieved a total of 228 articles, but only 17 met the inclusion criteria and were included in the study. Next, the respective studies were appraised, according to evidence-based guidelines of categorization of medical studies (►Table 2), and systemati-

cally analyzed. None of the studies were conducted as a randomized controlled trial and only one evaluated a control group.⁷ Furthermore, blinding was not observed in any study selected. Only prospective comparative studies and case series were to be analyzed in this review.

The operated patients' demographics and audiometric data were carefully examined to avoid double counting of cases. Three studies presented data which were also showed in more recent articles, including this review^{8–10}; thus, they were discarded. Two studies scored low in patient population and did not provide suitable follow-up (patients had missed follow-up)^{11,12}. Some studies presented incomplete data¹³ and were excluded for further analysis.

Therefore, after quality assessment and ruling out those failing to meet inclusion criteria, only 11 studies remained for data extraction and analysis (►Table 3). All studies accepted evaluated the effect of cochlear implantation on at least one outcome of interest.

Data Extraction

A total of 137 patients with single sided-deafness have been submitted to a cochlear implant. Pooling of data was not possible due to clinical heterogeneity among the studies. Furthermore, distinct parameters were used regarding duration of deafness, indication of cochlear implant, outcome measures, and follow-up time. As described above, data are summarized in ►Table 3. Several *p* values are missing because they were not reported.

Sound Localization

In individuals with two functioning cochleae, the auditory pathway uses interaural timing and intensity variation to calculate the coordinates and localize sound correctly. For frequencies below 800Hz, the auditory system relies mainly on phase delays caused by interaural time differences¹⁴; whereas for frequencies greater than 1600Hz, it primarily relies on interaural level differences. Both phenomena are used in the transition zone from 800Hz to 1600Hz.¹⁵

Table 1 Search databases and filters

Database	Search	Terms	Results
PubMed	#1	Single-sided OR one-sided OR unilateral OR monoaural	162
	#2	Deafness OR hearing loss OR loss of hearing	–
	#3	Cochlear implant	–
	#4	#1 AND #2 AND #3	–
Cochrane	#1	Single-sided deafness	8
	#2	Unilateral hearing loss	–
	#3	Cochlear implant	–
	#4	#1 AND #3 OR #2 AND #3	–
Scielo	#1	Search strategy designed for Scielo in Title/abstract fields – “unilateral hearing loss,” “single-sided deafness,” cochlear implant	58

Table 2 Levels of evidence in medical research in studies that investigate therapy

Level	Study design
Level I	- Systematic review (with homogeneity) of randomized control trials (RCT) - RCT with statistically significant difference or narrow confidence intervals
Level II	- Low quality RCT (e.g., <80% follow-up) - Systematic review of cohort studies - Prospective comparative study
Level III	- Case-control study - Systematic review of case-control study - Retrospective comparative study
Level IV	- Case series - Poor quality case-control studies
Level V	- Expert opinion

Source: <http://www.cebm.net/oxford-centre-evidence-based-medicine-levels-evidence-march-2009/>

There are several studies examining the effectiveness of cochlear implants and other treatments in rehabilitating sound localization. Localization error is commonly used as an outcome measure to assess localization. Localization error is the mean difference (in degrees) between sound source localization and the source pointed out by the patient. Three studies reported sound localization,^{16–18} a sum of 26 patients.

Arndt et al¹⁶ compared sound localization using CROS, osseointegrated implant devices or cochlear implants six months after implantation in a cohort of 11 patients. Seven loudspeakers were placed in a semicircle in front of the patients, which were then asked to identify the speaker that was delivering the sound. Patients who received cochlear implants showed significantly less localization error compared to those in an unaided condition ($p = 0.003$), and patients with osseointegrated implant ($p = 0.002$), and patients with CROS hearing aid devices ($p = 0.001$).

Recently, Firszt et al¹⁷ reported that seven out of ten adults that had undergone cochlear implantation showed improvement in sound localization in the bimodal condition (CI plus hearing aid (HA) in better ear) compared with the HA-only condition ($P \leq 0.05$). Interestingly, these same seven had postlingual deafness in contrast to three who did not exhibit any improvement in sound localization and who presented with either prelingual or perilingual deafness.

Cardieux et al¹⁸ researched five patients with SSD submitted to CI and reported a significant enhancement in bimodal scores in three patients compared with those in the HA-only condition ($p < 0.05$). These results are in accordance with the outcomes in Firszt et al.¹⁷

Effectiveness of Cochlear Implants in Improving Speech Perception

Redundant information received by two independent acoustic sensors allows for summation and squelch. Binaural summation occurs when the same acoustic stimulus presents in both ears. The higher order auditory processing of redundant information provides 2-6dB in signal threshold and is particularly beneficial in noisy environments. The squelch

effect, on the other hand, represents a different form of higher order auditory processing, which helps to sort out meaningful sound from background noise, given that it is able to reduce noise ratio by 2-3dB.^{6,16,19} Cochlear implants allow for both an acoustic sensor and an electrical input to individuals' deaf side. Thus, if the auditory system can effectively combine this electrical signal with acoustic hearing in the opposite ear, patients that have undergone cochlear implantation will theoretically benefit from summation and squelch.

Seven studies reported on speech perception in patients with SSD and CI ($n = 82$).^{6,16–18,20–22} Different configurations have been used to assess overall speech understanding. In the following section, we decided to abbreviate sound (S) and noise (N) followed by the direction: "HE" for sound or noise directed toward the hearing ear (better ear) and "CI" for the cochlear implant side. The "O" is for 0 (zero) degrees azimuth.

Arndt et al¹⁶ evaluated speech perception using three conditions and compared CROS and bone-anchored hearing aid (BAHA) device recipients: first, sound and noise directed at the front of the patient's head (S_0/N_0), second, sound directed at normal hearing and noise directed at the deaf side (S_{HE}/N_{CI}), and third, sound directed toward the deaf side and noise directed at the normal hearing ear (S_{CI}/N_{HE}). The results demonstrated that CI provided a statistically significant improvement in speech perception in the S_{HE}/N_{CI} and S_{CI}/N_{HE} configurations compared to those with CROS ($p = 0.031$ and $p = 0.03$) or osseointegrated implant devices ($p = 0.023$ and $p = 0.001$). The only configuration where CI was significantly superior to an unaided condition was S_{CI}/N_{HE} ($p = 0.001$). There was no significant difference in improvement between the groups when the noise was directed head-on.

In agreement with Arndt et al¹⁶, Vermeire et al⁶ didn't find a significant outcome in the S_0/N_0 configuration. By contrast, they found a significant improvement in the binaural condition in the S_{CI}/N_0 configuration in both subgroups: the contralateral HA ($p = 0.042$) and normal-hearing ($p = 0.003$) subgroups. Only the HA-subgroup experienced a significant benefit with the CI activation ($p = 0.031$) in the S_0/N_{CI} configuration.

Table 3 Studies characterization and outcomes

Authors	Study type	Evidence level	Implant type	Total no. of patients	Statistics	Follow-up	Outcomes	Speech perception	Tinnitus
Ramos et al. ²³	Prospective comparative	II	Nucleus	10	N/A	≥3 months	N/A	N/A	THI, VAS-distress
Arndt et al. ¹⁶	Prospective comparative	II	Nucleus	1-1	S ₀ /N ₀ : NS S ₀ /N _{IE} S _{CI} /N _{IE} p < 0.01 (CIxUnaided) p < 0.01 (CIxBAHA) p = 0.03 (CIxCRoS) S _{HE} /N _{CI} p = 0.02 (CIxBAHA) p = 0.03 (CIxCRoS) Sound localization: p < 0.01* Tinnitus: p < 0.01	6 months	7 loudspeakers in a 180° arch with intervals of 30°	HSM, OLSa	VAS-distress
Firszt et al. ¹⁷	Prospective comparative	II	Nucleus	10	Sound localization: p ≤ 0.05 (for all patients) Speech perception: TIMIT in quiet p ≤ 0.05 ^b	6 and 12 months ^a	15 loudspeakers arranged in an arc, 10° apart	CNC, HINT, TIMIT	N/A
Stelzig et al. ²¹	Case series	IV	Med-El	4	N/A	6 months	N/A	HSM, OLSa	N/A
Tavora-Vieira et al. ²² Tavora-Vieira et al. ⁹	Prospective comparative	II	Med-El	28	S ₀ /N ₀ : p < 0.001 S ₀ /N _{IE} : p = 0.003 S _{CI} /N _{IE} : p < 0.001 Tinnitus: p = 0.011	12-24 months ^d	N/A	BKB-SIN	TRQ
Vermeire et al. ⁶	Prospective comparative	II	Med-El	20	S ₀ /N ₀ : NS S ₀ /N _{CI} : p = 0.03 (only HA group) ^e S _{CI} /N ₀ : p = 0.04 (HA) p = 0.003 (HE) VAS: p = 0.04 ^f TQ: p = 0.04 ^f	12 months	N/A	LIST	N/A
Punte et al. ⁷	Prospective comparative	II	Med-El	7	VAS: p = 0.04 ^f TQ: p = 0.04 ^f	1,3 and 6 months	N/A	N/A	VAS:loudness TQ
Mertens et al. ²⁴	Prospective comparative	II	Med-El	15	VAS: p < 0.01 ^g	12 months	N/A	N/A	VAS:loudness TQ
Buechner et al. ²⁰	Prospective comparative	II	HiRes	5		12 months	N/A	OISa	VAS:loudness and distress
Van de Heyning et al. ²⁵	Prospective comparative	II	Med-El	22	VAS: p < 0.01 TQ: p < 0.01	24 months	N/A		VAS:loudness TQ
Cardieux et al. ¹⁸	Case series	IV	Nucleus HiRes (3 patients)	5	**	6 months	15 loudspeakers arranged in an arc, 10° apart	CNC, HINT, BKB-SIN	

Abbreviations: 0, 0 degrees azimuth; BAHA, bone anchored hearing aid; CI, cochlear implant; CNC, consonant-vowel nucleus-consonant test; CRoS, contralateral routing of signal; HA, hearing aid; HE, hearing ear (better ear); HINT, hearing in noise test; HSM, Hochmair-Schulz-Moser sentence test; LIST, Leuven Intelligibility Sentence Test; N, noise; NS, not significant; OLSa, Oldenburger sentence test; S, sound; THI, Tinnitus Handicap Inventory; TQ, Tinnitus Questionnaire; TRQ, Tinnitus Reaction Questionnaire; VAS, visual analogue scale.

*Cochlear implant improved sound localization compared with unaided, BAHA and CRoS conditions.

**Bimodal pros in CNC words were significantly better than HA-condition alone in quiet for 3 patients and in noise for only one patient (p < 0.05). Three patients demonstrate a significant improvement in localization in bimodal compared with HA-condition (p < 0.0001).

^a12 months evaluation available only for prelingual participants.

^bDiscrepancy between results mentioned in the text and the tables. The results in the tables are adopted. This p value showed (TIMIT in quiet) is related to postlingual patients with bimodal condition (CI and HA).

^cPatients belong to both studies and are put together to avoid double counting of cases.

^dSpeech perception follow-up was assessed 12 months after implantation; SSQ questionnaire and tinnitus distress were evaluated 24 months after CI surgery.

^eThe author separated the subjects in two groups: patients with hearing aid (HA) contralateral to the implanted ear and patients without any prosthesis in the hearing ear (HE).

^fData referent to 6 months after cochlear implantation.

^gDifference between the Cl-on and Cl-off condition.

Firszt et al¹⁷ evaluated 10 patients with SSD or asymmetric hearing loss (7 postlingually and 3 prelingually) and demonstrated a significant improvement in speech perception in the CI-only condition at the six-months interval for the following tests: Consonant-Vowel Nucleus-Consonant (CNC), $p \leq 0.001$; Hearing in Noise Test (HINT), $p \leq 0.001$; TIMIT in noise, $p \leq 0.01$; and TIMIT in quiet, $p \leq 0.001$. The bimodal condition (CI + HA) showed a significantly better performance only in TIMIT sentences in a quiet environment ($p \leq 0.05$). Only one of three pre-lingual subjects had open-set speech recognition with the CI.

Another study designed by Buechner et al²⁰ conducted with five patients showed that, for three patients ($p < 0.01$), CI led to a highly significant improvement when noise was presented from the normal hearing side and speed from the front.

A case series conducted by Stelzig et al²¹ showed an improvement in speech perception measured by the Hochmair-Schulz-Moser sentence test (HSM) and the Oldenburger sentence test (OISa). All the patients' HSM scores were higher in the binaural condition. Their performance in the OISa test was better when the noise signal was presented on the CI side and worse when the noise was presented on the normal hearing side. This study lacks statistic values.

Távora-Vieira et al²² showed a significant improvement in speech perception in noise scores when speech and noise were presented from the front and in the following arrangement: S_0/N_{HE} ($p = 0.003$) and S_{CI}/N_{HE} ($p < 0.001$). The speech perception improved the most in the S_{CI}/N_{HE} , coinciding with the outcomes found by Arndt et al¹⁶. There was no significant interaction between age at implantation and duration of deafness.²²

Cardieux et al¹⁸ identified a significant improvement in performance in localization in the bimodal compared with HA-only condition for three of five patients ($p < 0.001$) studied.

Tinnitus

Some studies have evaluated the suppression or release of tinnitus after cochlear implantation. Seven studies analyzed herein report on tinnitus ($n = 98$).^{7,16,19,22-25} Six used a Visual Analog Scale (VAS) to assess tinnitus and two used questionnaires. Among the studies using VAS, four demonstrated a significant reduction of tinnitus loudness or distress after CI.^{7,16,24,25}

Távora-Vieira et al,²² in a very recent study from 2015, tracked twenty-eight patients for 24 months and demonstrated a significant decrease on tinnitus disturbance ($p = 0.011$), which was measured using the Tinnitus Reaction Questionnaire (TRQ).

Ramos et al²³ reported that, from a cohort of ten patients, two reported tinnitus suppression, seven experienced reduction in tinnitus intensity and one presented no change after cochlear implantation. The mean score for tinnitus retraining therapy (THI) fell from 72.1%, preoperatively, to 14.3% at 3 months after cochlear implantation. The VAS showed a reduction from 7.9 points before surgery to 2.7 points at 3 months postoperatively.

Punte et al⁷ relayed tinnitus relief in patients with SSD after CI. The authors observed a significant reduction of average VAS from 8.21 points (SD = 1.22) to 4.36 (SD = 1.31) ($p = 0.027$) three months after activation of the CI. After six months, tinnitus loudness VAS decreases to 3.5 points ($p = 0.042$). Evaluation of tinnitus questionnaire (TQ) demonstrated a total score decrease (from 60.0 to 39.4) ($p = 0.041$) six months after implantation, denoting tinnitus improvement.

A significant reduction of tinnitus loudness was reported by Mertens et al,²⁴ when they compared the CI-on and CI-off conditions. The mean VAS score was 7.2 (SD = 2.6) in the CI-off condition and declined to 3.4 (SD = 2.5) in the CI-on condition ($p < 0.01$). This study also reported an improvement in speech reception threshold (SRT) in the non-tinnitus ear when the cochlear implant was switched on ($p < 0.01$).

In a study by Van de Heyning et al,^{6,25} twenty-two patients in the cohort had tinnitus, of which three experienced suppression, 18 reported improvement and one reported no change.

Arndt et al¹⁶ studied 11 patients with tinnitus in a cohort: six months after CI use, the median tinnitus intensity decreased significantly in the VAS score (from 5 to 0) when the CI was switched on ($p = 0.0078$). Five patients showed complete suppression of the tinnitus with the activated speech processor.

Three of five patients experienced suppression of the tinnitus in the study by Buechner et al²⁰. However, no statistical data are shown for this study.

Discussion

Currently approved treatment solutions for unilateral hearing loss (contralateral routing of sound and osseointegrated implants) are effective in addressing the head shadow effect, but fail to provide psychoacoustic information to the deaf side (squellch and summation, which help to improve speech perception in noise).

A cochlear implant is the only option that provides ear-specific information and, thus, potentially benefits SDD patients' bilateral listening. Furthermore, recognition in noise and sound localization are superior under binaural hearing.²⁶ To date, overall selection criteria for cochlear implantation in SSD have not yet been established and the factors that may affect outcomes are unknown. Nevertheless, as long as familiarity with the cochlear implant device increases, there is a broadening of selection criteria for the surgery.

Based on that, we sought to review the literature regarding the effects of cochlear implantation on clinical outcomes, such as speech recognition, sound localization, and tinnitus, in patients with single-sided deafness. This systematic review is characterized by a critical appraisal and clear synthesis of the selected studies.

After rigorous evaluation, three studies could be analyzed in terms of sound localization.¹⁶⁻¹⁸ All of them presented statistical data, proving that sound localization is better in bimodal condition than in unaided or CROS/BAHA conditions. It is important to note that these outcomes refer to

postlingual subjects. The only study that evaluated the outcomes of cochlear implantation in patients with prelingual onset of deafness was the one conducted by Firszt et al.¹⁷ They did not show any benefit from cochlear implantation in prelingual patients. This study suggests that, perhaps, there are some limitations in improving localization in patients with pre- or perilingual deafness.¹⁷ Our results on the outcomes described above are congruent with the results from Kamal et al²⁷, Vlastarakos et al²⁸, and Van Zon et al.²⁹

Several studies describe improvement in speech perception;^{6,16–18,20–22} however, only four have shown consistent statistical data.^{6,16,17,22} All four used divergent parameters to measure outcome. Two of the studies^{16,22} found a significant improvement in speech understanding when sound is introduced at the cochlear implanted side and noise in ear with normal hearing, considered the most challenging situation in daily life. Only Távora-Vieira et al²² found statistically superior performance with a configuration with sound and noise introduced to the subjects front (S_0/N_0). The abovementioned results are encouraging, in that they can be attributed to the squelch effect, meaning that the auditory system is able to process binaural signals after cochlear implantation. Recently, Vlastarakos et al²⁸ and Van Zon et al²⁹ reviewed speech perception in SSD patients, arriving at results consistent with our reports.^{28,29}

Duration of deafness is a well-known factor affecting auditory performance in postlingual patients submitted to cochlear implantation.³⁰ Nonetheless, this study is the first review to analyze a study concerning this topic. Távora-Vieira et al., in 2015, were the first to investigate whether duration of deafness and age at implantation have an effect on the outcomes in postlingual patients. The study showed that these variables do not seem to affect the speech perception in noise or the improvement of tinnitus.²² Furthermore, their results, combined with CI acceptance, suggest that subjects with SSD are probably able to integrate the acoustic and electrical signals.

Cochlear implantation was used first in 2008 to treat tinnitus in patients with unilateral hearing loss.²⁵ Reports of significant reduction of tinnitus after CI in patients with SSD confirm the effect of CI on treating unresponsive tinnitus (when tinnitus retraining therapy, sound therapy and drugs are not effective).^{7,16,22,24,25} The improvement of tinnitus can occur due several mechanisms: habituation, acoustic masking, direct stimulation of the cochlear nerve and organization of cortical pathways.²¹

Among seven studies that evaluated tinnitus relief or suppression, five presented statistically significant reductions of the symptom.^{7,16,22,24,25} The improvement of tinnitus distress or loudness after cochlear implantation supports the theory of auditory deafferentation,^{31,32} an outcome resulting from reafferentation by the restoring of auditory input.³³

In a recent meta-analysis analyzing only case series, Blasco and Redleaf³⁴ found that cochlear implants had a statistically significant improvement in the severity of tinnitus. Van Zon et al²⁹ analyzed six studies and reported a significant reduction of tinnitus distress in three of them.

Tokita et al³ and Vlastarakos et al²⁸ found similar outcomes. These studies corroborate our view.

Finally, we would like to point the awkwardness in performing this systematic review, considering the large degree of heterogeneity in outcomes and subject groups among the studies. There is great variation concerning the duration and onset of deafness, and in some cases, no mention thereof. Moreover, studies diverge in their follow-up and, especially, the tests and parameters used to assess outcomes. These inconsistencies largely impede a straight-forward comparison between the studies.

Final Comments

We conclude that there is a large clinical heterogeneity among the studies that evaluated cochlear implantation in patients with unilateral hearing loss. Furthermore, there has yet to be a high level-of-evidence study performed concerning this question.

Outcomes regarding enhancement of sound localization, speech perception, and, mainly, improvement of tinnitus are promising indications as well; however, high quality studies are required before standardizing cochlear implantation as a treatment for single-sided deafness. Nonetheless, the results obtained up to this point from cochlear implantation in patients with single-sided deafness are encouraging in deeming this procedure a reasonable treatment. Given that the cochlear implant seems to bring greater benefits than contralateral routing of sound (CROS) and osseointegrated implants, it should be the first choice of treatment for patients with SSD in that which pertains to satisfactory selection criteria.

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