

Cochlear Implantation Improves Localization Ability in Patients With Unilateral Deafness

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Objectives: One of the major complaints of people with a single-sided deafness is the inability to localize sound sources. Evidence suggests that subjects with a hearing loss can benefit from the use of a cochlear implant (CI) in sound localization. This study aimed to determine the effect of CI use on localization ability in unilaterally deafened subjects.

Design: Sixteen adult subjects with postlingual unilateral deafness, fitted with a CI on the deaf side, were included in this study. The auditory speech sounds evaluation (AŞE) localization test was used to determine localization with a CI on (binaural) and a CI off (monaural). The root mean square error was used as a measure of the subject's localization performance. Stratified analyses were performed to test the influence of gender, age of implantation (<55 years and >55 years), and the duration of deafness (<10 years and >10 years) on localization ability.

Results: Subjects with a CI on localized significantly better than without a CI. Gender, age, and the duration of deafness had no effect on the localization ability of the subjects.

Conclusions: Cochlear implantation is effective in improving localization abilities in subjects with unilateral deafness. The root mean square error dropped significantly with binaural hearing compared to monaural hearing.

Key words: AŞE, Bilateral, Binaural, CI, Hearing loss, Localization, Monaural, Unilateral deafness.

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INTRODUCTION

Binaural hearing provides the benefit of sound localization in the normal auditory system and allows better understanding of speech in noisy situations when the source of speech and the noise are presented from different locations in the horizontal plane (reviewed by Grothe et al. 2010 and Litovsky et al. 2012).

Sound localization in the horizontal plane relies on two binaural cues: interaural level difference (ILD) and interaural time difference (ITD). The aforementioned cues help to localize high frequency and low frequency sounds respectively (Grothe et al. 2010). Several studies have demonstrated that cochlear implant (CI) users rely mostly on ILDs to localize sounds (e.g., Laback et al. 2004; Senn et al. 2005; Grantham et al. 2007). It is conceivable that this is also true for unilaterally deafened CI users. In view of that, it was thought that a localization test that only uses ILDs as a cue would be suitable to verify whether unilaterally deafened CI users can integrate the two, acoustic and electrical, signals to identify the sound location. This study was performed using the auditory speech sounds evaluation (AŞE) localization test (PJ Govaerts, Antwerp, Belgium) that creates an ILD in an artificial way.

Numerous reports over the last decade have argued that one of the benefits of bilateral cochlear implantation over

unilateral usage is sound localization in both pediatric and adult populations (e.g., Van Hoesel & Tyler 2003; Nopp et al. 2004; Grantham et al. 2007; Litovsky et al. 2009).

Cochlear implantation in the treatment of unilaterally deafened subjects is a relatively new treatment modality (Arndt et al. 2011). Traditionally, patients were offered either: no treatment, a conventional contra-lateral routing of signal (CROS), or bone-anchored hearing aid (Baha) (Arndt et al. 2011). Our clinical experience with unilateral deafened patients is that the possibility of acquiring localization skills can be the driving force that triggers some of them to explore the possibility of cochlear implantation. For this group of patients, Baha or CROS may not be satisfactory options, as both devices are limited to transferring the signal of the deaf ear toward the good hearing ear, therefore not providing binaural advantage for localization ability.

Arndt et al. (2011) compared the outcomes of cochlear implant, CROS, and Baha in a group of 11 subjects. Localization tests were performed using seven loudspeakers positioned at intervals of 30 degrees between –90 and 90 degrees. Localization results showed a significant improvement with CI when compared with the unaided condition, and also when compared to performance with the CROS or Baha devices.

Recently published reports include data on a very small number of CI subjects with unilateral deafness of short duration: Firszt et al. (2012) reported that localization was significantly better with bilateral hearing in all three subjects included in her study. Likewise, Hassepas et al. (2013) showed improved localization skills in two out of three children, 12 months after cochlear implantation. To our knowledge, there is no literature that solely addresses the localization ability in a large number of participants with unilateral deafness and CI. A novelty of this study is the inclusion of patients with long deafness duration.

In view of the increasing literature demonstrating the hearing benefits of cochlear implantation in unilateral deafness (Vermeire & van de Heyning 2009; Buechner et al. 2010; Arndt et al. 2011; Stelzig et al. 2011; Firszt et al. 2012; Hansen et al. 2013; Hassepas et al. 2013; Távora-Vieira et al. 2013a), we hypothesized that cochlear implantation could re-establish the benefits of binaural hearing in postlingual unilateral deafness, significantly improving the patients' ability to localize sound sources. The present study addresses this hypothesis by examining the localization ability in 16 adult subjects with postlingual unilateral deafness who received a CI within the last 18 months.

Duration of deafness has been used in studies on bilateral severe-to-profound hearing loss as a predictive factor for the outcomes of cochlear implantation in postlingually deaf patients (UK Cochlear Implant Study Group 2004; Dunn et al. 2008). We have previously looked at a small cohort of five patients with more than 25 years of deafness duration and did not find that deafness duration was a determinant factor in the CI outcomes in postlingual unilateral deafness (Távora-Vieira et al. 2013b). There is some evidence that if unilateral deafness occurs after maturation

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of bilateral pathways, the lateralization of contralateral activation of the auditory areas is not altered (Langers et al. 2005) and thus it was thought that localization could be achievable by this group of subjects if cochlear implantation was provided to the deaf ear.

Younger age at implantation has been well accepted as essential for improved outcomes of cochlear implantation of children with congenital bilateral deafness (e.g., McConkey et al. 2004; Dettman et al. 2007; Van Deun et al. 2010). For adult with postlingual bilateral deafness, there is no evidence that age of implantation is a key factor for CI outcomes (Herzog et al. 2003; Olze et al. 2012). However, it is possible that older adults with unilateral deafness would find it more difficult to adapt to a new combined acoustic and electrical inputs.

This study also aimed to investigate whether deafness duration and age at implantation played a role in the localization performance of unilaterally deafened CI users.

MATERIALS AND METHODS

Subjects

Sixteen adult subjects (nine male, seven female) with postlingual unilateral deafness were recruited for this study. All subjects had opted for a CI after a wireless CROS hearing aid and Baha trial. The mean age at implantation was 55 (range: 36 to 73). The mean duration of deafness was 12 years (range: 3 months to 39 years). Table 1 shows more detailed demographic data.

In the subject's good hearing ear, the pure tone average at 0.5, 1, 2, and 4 kHz was ≤ 30 dB. All subjects were implanted with the FLEX²⁸ electrode array and received an OPUS 2 speech processor (MED-EL, Innsbruck, Austria). They used their speech processors on a full-time basis. Bilateral loudness balancing was performed for each patient's program settings. All the subjects in the present study received intensive auditory training for 3 to 6 months. The auditory training involved listening activities through the direct audio input of the speech processor. The adult subjects in the present study had 6 to 18 months experience with their CI at the time of data collection.

This study was designed and conducted in accordance with the Helsinki Declaration. Ethical and institutional review committee approval was obtained.

Sound Localization

Localization testing was performed using the A&E localization test software. This presents a narrow band noise of 1/3rd octave centered around 4000 Hz simultaneously through two loudspeakers that were placed at -60 and 60 degrees from the listener. The noise from the speakers was correlated with an interaural time difference of zero. The presentation level differs from each speaker simulating an ILD, hence creating the illusion of a sound source being localized somewhere on the azimuth between the two loudspeakers (<http://www.otoconsult.com/asse/Localization.aspx>).

Thirteen loudspeakers numbered -6 (left) to 6 (right) were setup on a semicircle, at 10-degree intervals. Using this setup, 11 loudspeakers were "sham" loudspeakers, which were numbered from -5 to 5 , and were placed in between two real loudspeakers, which were numbered as -6 and 6 (Fig. 1). Testing was performed in a soundproof booth. The subject sat directly facing speaker number 0 and did not move their head.

For each presentation the software randomly picks an ILD from the following series: -30 , -20 , -10 , -4 , 0 , $+4$, $+10$, $+20$, and $+30$ dB. The presentation level is 60 dB HL at one loudspeaker and 60, 56, 50, 40, or 30 dB at the other, depending on the ILD chosen.

The number of test items is 33. The stimuli with intensity differences of -30 , -20 , -10 , 10 , 20 , and 30 dB are presented three times each; the stimuli with intensity differences of -4 , 0 , and 4 dB are presented five times each.

The participants were asked to say the number of the speaker they thought the sound was coming from. The subjects' responses were entered into the computer and the software calculated the median values and the test error. The root mean square (RMS) error was used as a measure of the subject's

TABLE 1. Subject demographic data

ID	Duration of Deafness (Years)*	Age at Implantation (Years)	Experience With CI (Months)	Ear	Etiology	Pure Tone Average	Pure Tone Average
						(0.5, 1, 2, and 4 kHz)—Nonimplanted Ear in dB*	(0.5, 1, 2, and 4 kHz)—Implanted Ear in dB*
S1	2	56	14	L	Head trauma	18	90
S2	35	40	18	L	Unknown	7	>110
S3	0.6	70	12	R	Unknown	25	84
S4	20	55	14	L	Meniere's disease	18	>110
S5	30	36	16	L	Unknown	8	>110
S6	39	51	8	R	Mumps	4	>110
S7	0.9	48	8	L	Unknown	8	72
S8	0.6	53	10	R	Unknown	14	72
S9	12	41	17	L	Unknown	5	72
S10	0.3	62	6	R	Sudden hearing loss	7	72
S11	0.4	72	6	L	Sudden hearing loss	24	82
S12	40	55	14	L	Unknown	13	79
S13	5	71	7	R	Unknown	30	74
S14	1.5	73	7	L	Unknown	21	>110
S15	1	57	12	L	Sudden hearing loss	25	76
S16	0.3	39	6	R	Fistula	27	>110

*Before implantation.

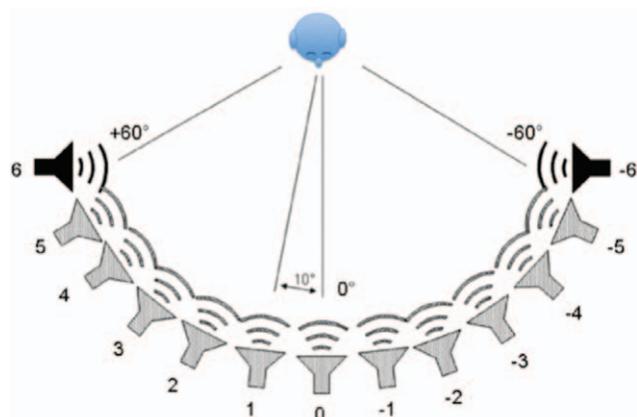


Fig. 1. Position of the subject and the loudspeakers. The test person is facing loudspeaker 0. The loudspeakers 6 and -6 (in black) are real functioning loudspeakers, and the other speakers (in gray) are sham loudspeakers. The 2 real loudspeakers are positioned at -60° and $+60^\circ$ in front of the test person, and the sham loudspeakers are at 10° angles between -60 to $+60^\circ$.

localization performance. A lower RMS error represents better localization skills.

Each subject performed the localization testing in two listening conditions: monaural hearing (normal acoustic hearing alone) and binaural hearing (acoustic hearing and CI activated). The order of the test was randomized. All subjects had at least 6 months experience with their CI. Testing was performed with the subject's everyday program settings, which accounts for subjective loudness balancing between the normal ear and the CI stimulation.

Normative Data of the A ξ E Localization Test

The developers of the software carried out a normative study to obtain reference data using 30 normal hearing subjects. The subjects were recruited at the Eargroup (Antwerp, Belgium). They had an average age of 27 years (SD: 6.8 years; range: 15 to 41 years) with normal hearing (hearing thresholds of 20 dB HL or better in the frequency range from 0.25 to 4 kHz) and an uneventful otological history. The subjects underwent the localization test as described above. Each ILD thus yielded 30 responses, namely the loudspeakers as identified by the 30 listeners. The distribution of these responses was summarized by the average and standard deviation. Table 2 shows the normative data.

Analyses

Paired sample t tests were used to examine the differences between CI off and CI on in localization. Stratified analyses using paired sample t test was performed to test the influence of gender, age at implantation (36 to 55 years; and 56 to 73 years), and the duration of deafness (3 months to 10 years; and 10 to 39

years) on localization. The Kolmogorov-Smirnov test was used to determine the data distribution before data analyses. The independent-samples t test was used to determine a significant effect of age at implantation, gender and the duration of deafness between age groups. A p value of <0.05 was considered statistically significant.

RESULTS

Table 3 displays the results of Kolmogorov-Smirnov test, which confirmed that the data are approximately normally distributed and parametric statistical methods could be used.

Figure 2 shows the localization test results of the subjects ($n = 16$) tested with their CI on and with their CI off. The difference in localization ability, as determined using the A ξ E, was significantly better with the CI on (mean RMS error = 22.8; SD = 11.58) than the CI off (mean RMS error = 48.9; SD = 11.86) (Paired samples t test: $t = 6.979$; $df = 15$; $p < 0.001$).

Individual RMS data is presented in Figure 3. The data show that subjects S2 and S5 did not show any improvement in localization ability.

In female participants, the localization ability with the CI on was significantly better than with the CI off ($p = 0.003$). In male participants, the localization ability with the CI on was significantly better than with the CI off ($p = 0.001$). There was no significant difference with the CI on between female subjects and male subjects (Independent-samples t test: $t = 0.377$; $df = 14$; $p = 0.712$; CI: -10.547 to 15.047). Also, no significant difference between genders was found for CI off ($t = -0.389$; $df = 14$; $p = 0.703$; CI: -15.473 to 10.723).

Localization ability with the CI on was significantly better than with the CI off in subjects <55 years of age ($p < 0.001$; $n = 10$), and in subjects >55 years of age ($p = 0.012$; $n < 6$). There was no significant difference with the CI on ($t = 0.357$; $df = 14$; $p = 0.727$; CI: -11.023 to 15.423) between subjects <55 years of age and subjects >55 years of age. Likewise, there was no significant difference with the CI off ($t = -0.058$; $df = 14$; $p = 0.955$; CI: -13.965 to 13.232) between subjects <55 years of age and subjects >55 years of age.

Localization ability with the CI on was significantly better than with the CI off in subjects implanted after a duration of deafness of <10 years ($p = 0.001$; $n = 9$) and in subjects with a duration of deafness >10 years ($p = 0.010$; $n = 7$). There was no significant difference with the CI on ($t = -0.375$; $df = 14$; $p = 0.713$; CI: -15.153 to 10.645) or with the CI off ($t = 1.187$; $df = 14$; $p = 0.255$; CI: -5.651 to 19.651) between subjects with duration of deafness <10 years and subjects with a duration of deafness >10 years.

Additionally, the effect of gender, duration of deafness, and age at implantation on the difference between the CI-off and CI-on conditions was examined. No significant difference

TABLE 2. Results of the A ξ E localization test in 30 normal hearing subjects. For each interaural level difference (column headers), the average responses (number of the sham loudspeaker) are given, together with the lower (Min) and upper (Max) limit of the 95% confidence interval

ILD (dB)	-30	-20	-10	-4	0	4	10	20	30
Min	-6.0	-6.0	-4.6	-2.6	-1.0	0.0	2.3	3.4	3.7
Average	-5.5	-5.2	-3.2	-1.2	0.2	1.7	3.5	4.9	5.1
Max	-4.0	-3.3	-1.6	0.7	1.6	3.4	5.0	6.0	6.0

TABLE 3. Kolmogorov-Smirnov test

	CI Off	CI On
N	16	16
Kolmogorov-Smirnov-Z	0.607	1.024
<i>p</i> Value (Two-sided)	0.855	0.245

was found between female and male subjects (Independent-samples *t* test: $t = -0.606$; $df = 14$; $p = 0.554$; CI: -20.993 to 11.743), between subjects <55 years and those >55 years of age (Independent-samples *t* test: $t = -0.323$; $df = 14$; $p = 0.752$; CI: -19.628 to 14.495), and between subjects with <10 years of duration of deafness and those >10 years of duration of deafness (Independent-samples *t* test: $t = -1.252$; $df = 14$; $p = 0.231$; CI: -6.594 to 25.102).

DISCUSSION

This is the first study to solely investigate the localization ability in a large number of unilaterally deafened CI users, including subjects with long-term deafness duration (up to 39 years). The results confirmed our hypothesis that localization would improve significantly after cochlear implantation. This study also investigated some factors that potentially could influence the outcomes.

This study investigated localization skills in unilaterally deafened CI users using A \S E test localization software, which simulates an ILD, therefore generating the impression of the sound originating from the azimuth between the two loudspeakers. This software was developed to assess subjects using bilateral CIs and is used for the first time in this study to determine localization ability in unilateral deafness.

Paired sample *t* tests showed that in the binaural condition (CI on) subjects performed significantly better on the A \S E localization test than in the monaural condition (CI off). The mean RMS error dropped from 49 degrees (CI off) to 22.8 degrees (CI on). If S2 and S5, the poorer performers on the localization test, were excluded from the analysis, the mean RMS error

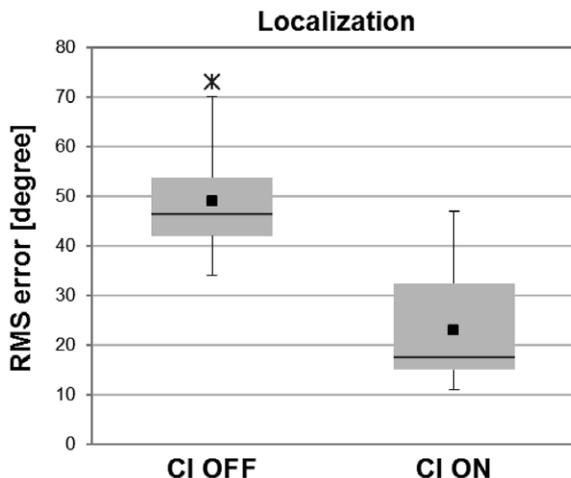


Fig. 2. Mean root mean square (RMS) error with CI on and CI off ($n = 16$). Lower RMS represents better localization skills. Median values are displayed as horizontal line, mean values as black squares. Length of the whiskers corresponds to the range of the data. The black asterisk represents an outlier (1.5 to $3 \times$ box height above the 75th percentile).

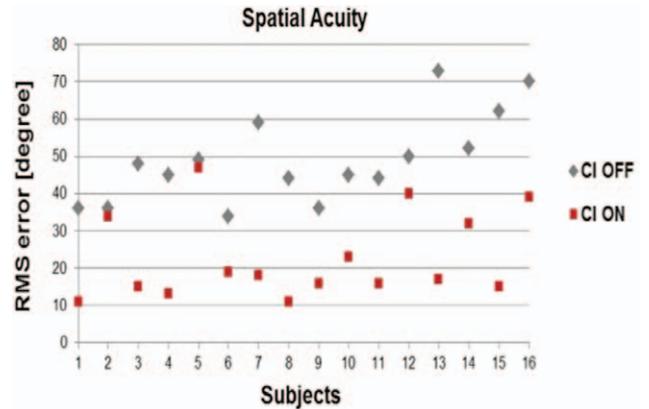


Fig. 3. Per subject root mean square (RMS) error with CI on and CI off. Lower RMS represents better localization skills.

would be 20.0 degrees. Based on the normative data, the RMS error for 30 normally hearing adults using A \S E ranged from 5 to 11 (median = 8) degrees. The RMS error for the group of subjects in this study ranged from 11 to 47 (median = 17.5) degrees in the binaural condition. Although the results shown by the unilaterally deafened group is worse than the normal hearing subjects tested in the same condition, the outcomes are very encouraging. While normal hearing individuals make use of interaural matched stimuli to localize sound sources, unilaterally CI users combine two distinct inputs (acoustic and electrical) and consequently their brain needs to create a spatial map with a substantial interaural dissimilarity.

There are very few studies that investigated the localization ability of unilaterally deafened adults CI recipients (all with duration of deafness < 10 years). Arndt et al (2011) investigated the localization ability of 11 adult with unilateral deafness and CI using a setup of seven loudspeakers positioned at intervals of 30 degrees between -90 and 90 degrees. Speech was used as the stimuli. The localization error was significantly reduced with CI on (median = 15.0 degrees) compared with the CI off condition (median = 33.9 degrees). This study showed that the RMS error in the CI-off and CI-on conditions were higher than what was reported by Arndt et al. (2011). This is true even if we included only the patients with less than 10 years of deafness. It is possible that, as observed with bilateral CI users, the use of speech signal as stimuli (Verschuur et al. 2005; Grantham et al. 2007) and fewer loudspeakers facilitated sound localization in that study. Similarly to our results, Firszt et al (2012) reported that a small group of three adults performed significantly better in the binaural condition with the RMS error being 18, 19, and 25 degrees. Although a speech signal was used as the stimuli, localization testing was performed using 15 loudspeakers located 10 degrees apart making the setup more comparable to ours.

Several studies of children and adults with bilateral deafness have demonstrated the superiority of localization skills with bilateral CIs over unilateral (e.g., Van Hoesel & Tyler 2003; Nopp et al. 2004; Litovsky et al. 2006a, b, 2009; Grantham et al 2007). Litovsky et al. (2009) reported the localization test results of 17 adult bilateral CI users. The mean RMS error was 56.6 to 60.4 degrees with monaural hearing and 28.4 degrees with binaural hearing. Grantham et al. (2007) reported that the average RMS localization error of 30.8 degrees for 22 CI users compared to 6.7 degrees for normal hearing subjects. This study

showed that the RMS error in the CI-off and CI-on conditions were lower than what was found with bilateral cochlear implant users in monaural and bilateral listening conditions. Although bilateral CI users can localize sound significantly better when using both devices, they perform worse than normal hearing individuals. This is explained on the basis that they rely on a binaural input that is likely to be dissimilar due to potentially different innervation pattern of the cochlea, and different depth of electrode insertion with consequent mismatch of anatomical place of stimulation (Kan et al. 2013). The challenge for the unilaterally deafened CI users is not lesser as their brain works with an interaural divergence to create a spatial map as mentioned earlier.

It was thought that the ability to achieve good binaural integration might be time dependent, linked to (1) age at implantation, (2) duration of deafness, or (3) the age at onset of deafness. Age of implantation and duration of deafness were not supported by the results of our study. The age at onset of deafness was not analyzed in the study.

When looking if age at the time of implantation had an influence on the results, the difference between CI off and CI on was significant for both age groups. This finding is in line with several studies showing that the hearing outcomes achieved by the elderly CI users do not differ from the younger population (Herzog et al. 2003; Noble et al. 2009; Olze et al. 2012). Nevertheless, the sample size of both age-stratified groups was relatively small. It is possible that a larger sample would more reliably reflect the population mean and yield a different result. Additionally, gender did not influence the results.

Previous literature has reported only the outcomes of patients with less than 10 years of unilateral deafness duration. As the patients in this study had between 3 months and 39 years of deafness duration, it was decided to stratify the listeners into two groups with deafness duration $>$ or $<$ 10 years. The difference between CI off and CI on was significant for both groups and there was no significant difference between the groups in each listening condition. However, it is important to note that from the 6 patients with $>$ 10 years of deafness of duration, 4 lost their hearing after age of 12. Normal binaural hearing during the development of bilateral auditory pathways may explain why these patients had an improvement in spatial hearing even after a prolonged period of hearing deprivation (Grothe et al. 2010; Litovsky et al. 2010; Keating & King 2013). Electrophysiological studies have demonstrated that if unilateral deafness occurs after maturation of bilateral pathways the lateralization of contralateral activation of the auditory areas is not altered (Langers et al. 2005) supporting the idea that localization may be improved even if cochlear implantation occur after a long-term unilateral deafness.

In the present study, two subjects (out of the 16 that participated) did not demonstrate any improvement in localization ability with CI use. Interestingly, both subjects had a long duration of deafness before implantation and had lost their hearing at 6 and 7 years of age, the youngest ages of deafness onset in the group. Similarly, Nopp et al. (2004) found that two patients who lost their hearing in early childhood (one at birth and one at the age of 6 years) did not show improvement in localization skills. Johnstone et al. (2010) reported that localization ability was only improved in children with unilateral hearing loss who received a hearing aid before the age of 5. In the last few years, human and animal studies have demonstrated that there

is a critical period for bilateral auditory pathway development explained by neuronal reorganization of the auditory brain following prolonged monaural hearing input in bilateral congenital profound deafness (e.g., Gordon et al. 2011, 2013; Kral et al. 2013). Keating and King (2013) also suggested that spatial hearing might be affected permanently if an extended period of asymmetrical hearing input occurs at a young age. Age of onset of deafness was not analyzed in this study due to small sample size. However, it is conceivable that the two poorer performing patients in our study were unilaterally deafened prior to binaural hearing maturation. It is important to note that both subjects used their speech processor on a full-time basis and reported a substantial benefit. The subjects also reported that they could not localize sounds precisely in an everyday listening environment, but that they were able to lateralize correctly most of the time.

Litovsky et al. (2006b) reported that sound localization in children improves over time. It is thought that children develop experience-dependent skills because of auditory plasticity. It is possible that there are differences in the ability of children and adults with cochlear implants to improve their localization ability, which may depend on auditory plasticity. However, a long-term follow-up is needed to investigate if localization ability improves over time in the adult population.

Training probably facilitates adults' ability to adapt to new spatial information (Nawaz et al. 2014). Further research is needed to determine whether appropriate training is enough to re-establish the auditory pathways related to localization skills in postlingually unilaterally deafened individuals and whether the subjects that did not perform well in the present study would develop strategies to localize sound better with practice.

CONCLUSION

The data herein suggest that cochlear implantation is effective in improving localization abilities in unilaterally deafened subjects. This improvement occurred within the period from 6 to 18 months after cochlear implantation and the RMS error dropped significantly with binaural hearing compared to monaural hearing. Deafness duration and age at implantation do not seem to influence the results.

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