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Duration of unilateral auditory deprivation is associated with reduced speech perception after cochlear implantation: A single-sided deafness study

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Abstract

Objective: Examine the relationship between duration of unilateral deafness and speech perception outcomes in adults with single-sided deafness after cochlear implantation.

Methods: A systematic review of PubMed articles containing individual speech perception and duration of deafness data from single-sided deaf adults. Studies selected for detailed review, and duration of deafness and speech perception outcomes were extracted, with speech scores reported as percent correct. A linear regression as a function of study and length of deafness was performed.

Results: A statistically significant negative effect of duration of unilateral deafness on speech perception was found, but there was substantial uncertainty regarding the strength of the effect.

Discussion: Existing data make it difficult to either support or reject a hard 5- or 10-year unilateral auditory deprivation limit on cochlear implant candidacy for patients with single-sided deafness. This is because the totality of available data are consistent with a very small effect, perhaps negligible in practical terms, and just as consistent with a very large effect. Regardless of effect size, the present results have important basic implications. They suggest that unilateral sound deprivation may have a deleterious effect on auditory processing even though more central parts of the auditory system have continued to receive input from a contralateral normal ear.

Conclusions: Speech perception scores in SSD patients are negatively correlated with duration of deafness, but the limited amount of data from cochlear implant users with long term single-sided deafness leads to substantial uncertainty, which in turn precludes any strong clinical

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Disclosure Statement

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recommendations. Further study of SSD CI users with long term deafness will be necessary to generate evidence-based guidelines for implantation criteria in this population.

Keywords

cochlear implant; single sided deaf; speech perception

Introduction

In recent years, it has become more common for patients with single-sided deafness (SSD, which means having severe to profound deafness in one ear and normal hearing in the contralateral ear) to receive a cochlear implant. The most common motivation is to alleviate severe tinnitus in the deaf ear (Van de Heyning et al., 2008; Vermeire & Van de Heyning, 2009), but cochlear implantation in SSD patients has also shown improvement in speech perception (Arndt, Aschendorff, et al., 2011), as well as some improvement in sound localization and in subjective report measures.

In many clinics as well as in clinical trials, SSD cochlear implantation has been limited to patients with a duration of unilateral hearing loss of less than 5 or 10 years due to the concern that patients with longer durations of deafness may have poorer speech perception outcomes. This concern stems from extensive literature showing negative correlations between duration of bilateral sound deprivation and speech perception outcomes after cochlear implantation in adults. In other words, results from patients with bilateral deprivation has been implicitly extrapolated to the case of patients with unilateral deprivation. However, it is not well established whether the effect of auditory deprivation on speech perception after cochlear implantation is central and occurs only after bilateral deprivation (in which case it would be irrelevant for SSD patients), or whether there is also a peripheral auditory degradation component such that sound deprivation in an individual ear will affect future cochlear implantation in that ear even in the absence of central auditory deprivation (Boisvert, McMahon, & Dowell, 2012; Lundin, Stillesjö, & Rask-Andersen, 2014). Some studies cast doubt on the existence of any peripheral effect (Boisvert, et al., 2012; Boisvert, McMahon, Dowell, & Lyxell, 2015; Boisvert, McMahon, Tremblay, & Lyxell, 2011), whereas other studies still recommend against implantation of ears that have been deprived of sound for more than ten years (Balkany et al., 2002; Connell & Balkany, 2006). Others make more nuanced recommendations based on a predictive model that takes preoperative speech perception and duration of profound deafness in each ear into account (UK Cochlear Implant Study Group [UKCISG], 2004a, 2004b, 2004c). Given these facts, it seems prudent to restrict initial clinical trials of SSD cochlear implantation to patients with shorter durations of unilateral deafness, as most SSD clinical trials have done to date (Peters et al., 2015). However, it is important to analyze the relevant data as they are published to ascertain whether the duration-of-deafness exclusion criterion remains valid, or whether it may be possible to relax or even eliminate it. This is one main aim of the present study: to perform a systematic review of speech perception data in adult SSD patients with CIs as a function of duration of deafness.

As described above, there is a clear clinical motivation for this study, but there is also a more basic motivation. It is well established in the animal literature that auditory deprivation affects the auditory system, depending on many factors including age at deprivation, extent of deprivation, and timing of deprivation (Kral & Eggermont, 2007; Sanes & Woolley, 2011). From that perspective, we are interested in how the auditory system may or may not be affected by unilateral deprivation, because this information will help refine hypotheses about the underlying mechanisms. In rodents it has been shown that monaural auditory deprivation distorts tonotopic maps and weakens the representation of the deprived ear. Plasticity upon reintroduction of auditory input exists, governed by critical periods (Knudsen, Esterly, & Knudsen, 1984; Popescu & Polley, 2010). In humans it is ethically impossible to conduct a long-term trial to study the effect of profound unilateral auditory deprivation, of course, but the existence of a novel clinical population (SSD cochlear implant users) presents the opportunity to address this question as a natural experiment of sorts. During the course of regular clinical practice as well as clinical trials, SSD patients with different durations of deafness have received CIs, and their speech perception scores may provide important information about how prolonged unilateral auditory deprivation may affect the human auditory system.

Methods

Data Sources:

Studies were selected by searching on Pubmed for the terms “single sided cochlear implant,” “SSD cochlear implant,” and “single sided deafness” during the dates 3/9/2017-3/11/2017. These search terms revealed 137, 56, and 229 results, respectively.

Study Selection:

Studies were selected for analysis based on the following criteria: 1) article in English, 2) duration of deafness was reported for each individual, 3) score on a speech perception test that recorded responses as “percent correct” was reported for each individual, 4) date at testing was at least 6 months post-implantation, 5) subjects had normal hearing in one ear to qualify as single-sided deaf, 6) subjects were implanted at 18 years of age or older, and 7) only postlingually deaf subjects were included.

Initially, 27 studies relevant studies were identified. 5 were excluded because subjects were implanted at younger than 18 years old, 3 were excluded because the text was not in English, 9 were excluded because no individual data on speech perception scores and/or duration of deafness was reported, and 2 were excluded because speech perception scores were not reported as “percent correct”. The remaining 8 studies were selected for further analysis: 1) Friedman et al., 2016 (Friedmann et al., 2016), 2) Arndt et al. 2011a (Arndt, Aschendorff, et al., 2011), 3) Arndt et al. 2011b (Arndt, Laszig, et al., 2011), 4) Hansen et al., 2013 (Hansen, Gantz, & Dunn, 2013), 5) Rahne et al., 2016 (Rahne & Plontke, 2016), 6) Stelzig et al., 2011 (Stelzig, Jacob, & Mueller, 2011), 7) Kitoh et al., 2016 (Kitoh et al., 2016), 8) Aschendorff et al., 2017 (Aschendorff et al., 2017). Three data points that were duplicated across studies 2 and 3 were deleted from the study 2 data set.

Data Extraction:

The included studies used percent correct scores (N=78 subjects) as a measure of speech perception outcomes. This measure was used to determine whether speech perception with a CI is affected by the previous duration of peripheral deprivation. Whenever there was a choice among different testing conditions within a study we used the conditions that would best isolate the contribution of the CI ear. This was done in different ways in each study, as described in Table 1: presenting a speech signal only to the CI ear via direct connection to the speech processor; placing an earplug and an earmuff in the NH ear; or presenting spatially separated speech and noise signals to favor the CI ear.

All studies included data on duration of deafness in the implanted ear and speech perception scores in either the CNC words test (studies 1 and 4), the Hochmair-Schulz-Moser sentence test (studies 2 and 3), the Freiburger monosyllabic word tests (studies 5, 6, and 8), or the Japanese monosyllable test (study 7).

Data concerning duration of deafness were taken from tables, except for Stelzig et al. (taken directly from text) and Arndt et al., 2011b (transcribed from Fig. 3 of that article). Data concerning speech scores were taken from tables, except for Arndt et al., 2011b, Hansen et al., Stelzig et al., and Kitoh et al., in which case the scores were transcribed from figures in these articles. Jpg files were obtained from PDF versions of each paper, and data were transcribed from figures with the use of an automated plot digitizer (25).

Data Analysis:

The main analysis was a linear regression as a function of study and duration of deafness. We fit a single line to the data from each individual study, and calculated the 95% confidence interval for the slope corresponding to each study. Tests were then conducted to determine whether slopes were significantly different across studies, and the same was done for intercepts. Because the first test was negative (that is, slopes were not significantly different between studies within each one of the two data sets), an overall pooled slope was calculated. It is important to note that the overall pooled slope was not calculated by pooling all the data, which would have been at least questionable given that each one of the eight studies used different methods and different outcome measures. Instead, the overall pooled slope was calculated using a regression function that had a single parameter for the “slope” variable and eight different “intercept” parameters for each one of the eight data sets. This analysis method allows the estimation of a more precise slope estimate that could be obtained with any subset of studies. Linear regressions of outcomes as a function of duration of deafness were also conducted for each individual study. In studies where a significant effect of duration of deafness on outcomes was found, we examined the effect of outliers by finding the most extreme single data point and performing the linear regression both with and without that point. This was done to determine whether any given significant result for an individual data set was robust in the face of a single data point being removed. The same three outliers were removed one at a time to determine whether the overall regression remained statistically significant. Lastly, separate analyses were conducted by pooling studies that used the same outcome measure: studies 1 and 4, which used the CNC word test, and studies 5, 6, and 8, which used the Freiburger word test.

Results

The entire data set is shown in the top panel of Figure 1., and each individual study in each panel of Figure 2. Three of the eight studies showed a statistically significant decrease in speech perception as a function of duration of unilateral deprivation. These were study 1 ($p=0.03$), study 3 ($p=0.017$), and study 5 ($p=0.048$). However, none of these results remain statistically significant once we remove a single outlier from each study (see top three left panels of Figure 2, outlier circled). No significant relationship between speech perception and duration of unilateral deprivation was found when pooling data across studies that used the same outcome measure (CNC words for studies 1 and 4, or Freiburger words for studies 5, 6, and 8).

Slopes of different studies were not significantly different ($F(7,62)=1.301$, $p=0.26$), so it was possible to calculate one representative slope for all the data. This slope was equal to -2.09 , and it was significantly different from zero ($t(68)=-2.92$, $p=0.0048$). The 95% confidence interval for the slope was $(-3.52, -0.66)$, indicating a substantial degree of uncertainty in our estimate. Nonetheless, the overall result (a negative association between speech perception scores and duration of deafness in the implanted ear) remains statistically significant even after removing the three outliers (from studies 1, 3, and 5) one at a time.

Discussion

The main result of the present systematic review is that duration of unilateral deafness seems to have a modest association with speech perception outcomes using the implanted ear. Taken individually, studies 1, 3, and 5 showed statistically significant decreases in speech perception as a function of duration of unilateral deafness, but that significance disappeared when a single outlier was removed from each study. None of the other individual study results were statistically significant. However, when taken as a whole, the main conclusion of these analyses is that there is a small but statistically significant decrease in speech perception outcomes ($P=0.0048$) as a function of duration of unilateral deafness, with a pooled slope of -2.1 percentage points per year of unilateral deprivation. This overall analysis was robust and remained significant when removing single outliers, which was not the case for any of the individual studies.

One important caveat is that there is a large amount of uncertainty regarding the estimate of 2.1 percentage point drop per year. The 95% confidence interval of the pooled slope goes from -0.66 to -3.52 percentage points per year of unilateral deprivation. It is important to remember that all analyses reported here are based on a relatively small N (78 subjects) and the number of data points at 10 or more years of duration of deafness is even smaller (only 5 subjects).

The analyzed data do not permit making strong clinical recommendations. If the estimated effect of duration of unilateral deafness is accurate, that means that patients with 10 years of deprivation would score on average 21 percentage points less than recently deafened patients. Remember also that the estimated drop of 2.1 percentage point drop per years is just the best estimate based on currently available data. The range of the 95% confidence

interval for the pooled slope means that the real drop in speech perception after ten years of unilateral sound deprivation might be as low as 6.6 percentage points (which is inconsequential in practical terms) or as high as 35.2 percentage points (which represents a very meaningful difference).

In summary, available evidence suggests that duration of unilateral deprivation has a modest but statistically significant negative effect on outcomes of cochlear implantation in SSD patients. The estimated drop of 21 percentage points after 10 years of unilateral sound deprivation should be considered when counselling SSD patients who are considering cochlear implantation. However, it seems insufficient for the purpose of recommending a hard 10-year unilateral auditory deprivation limit on cochlear implant candidacy - particularly in cases where there are reasons other than speech perception that might indicate cochlear implantation (such as the treatment of otherwise intractable tinnitus).

It is possible that future studies, with additional outcomes data from SSD patients with longer duration of auditory deprivation, may help refine the present estimates and conclusions. To that end, it will be particularly important to obtain data using methodology that is as uniform as possible across sites. It may also be important to obtain speech scores based on input from the CI only, preferably using methods like direct audio input or other types of direct delivery of the speech signal to the cochlear implant. This would allow a cleaner determination of speech perception performance with the CI only, while completely precluding any contribution from the normal hearing ear.

The present results, in addition to their clinical implications, also suggest that unilateral sound deprivation may have a deleterious effect on auditory processing even though more central parts of the auditory system have continued to receive input from a contralateral normal ear. This is a potentially important result that may be worthy of further investigation both in human studies and in animal experiments.

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Biographical Note:

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cochlear implantation, and the development of software/hardware based fitting tools to optimize cochlear implant fitting.

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FIG. 1.

(A) Raw speech perception score (percent correct) as a function of duration of unilateral deafness in eight separate studies. (B) 95% confidence interval of the linear regression slope for each study, and for all studies combined. Negative slope indicates deterioration in speech perception as a function of duration of unilateral deafness. *, $p < 0.05$; **, $p < 0.01$.

FIG. 2.

Raw speech perception score (percent correct) plotted against duration of unilateral deafness in eight separate studies (A-H). Specific test is indicated for each study on y-axis and linear regression is plotted. Studies 1, 3, and 5 have negative slopes that are statistically significant; circles indicate single data points that, if excluded, render the regression slope non-statistically significant for each study.

TABLE 1.

Speech presentation method for each study

Presentation method	Study Number
Direct connection to the CI	1 (6 subjects)
NH plugged and muffed	1 (4 subjects)
Speech presented to CI ear, noise presented to NH ear	2,3,6,8
Speech presented in front, noise to the NH ear	1,5,7

CI, cochlear implant; NH, normal hearing.

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