Abnormal Voicing in Children Using Cochlear Implants

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Objectives: To measure acoustic voice outcomes in children with bilateral cochlear implants and to compare these with established norms, as well as to determine whether these acoustic measures were influenced by duration of cochlear implant use, age at implantation, and overall “time in sound.”

Design: Cross-sectional study.

Setting: Pediatric tertiary care cochlear implant center.

Patients: All children using bilateral cochlear implants who were followed up during a 4-month period at our implant center were invited to participate. Twenty-seven children (17 males and 10 females) aged 3 to 15 years were enrolled. Causes of deafness included congenital (n=8), genetic (n=8), meningitis (n=3), cytomegalovirus (n=2), and unknown (n=6). The interval between first and second implantations ranged from simultaneous to 7.8 years (mean, 4.2 years).

Main Outcome Measures: Children completed acoustic voice testing using a Computerized Speech Lab and a Multi-Dimensional Voice Program. Acoustic results were compared with those of children receiving unilateral implants and with normative data.

Results: Compared with established pediatric normative data, children with bilateral implants demonstrated poor control of long-term frequency perturbation and long-term amplitude perturbation when vocalizing sustained phonations ($P < .001$ for both). This finding was consistent with data previously reported in children using unilateral cochlear implants. Long-term control of frequency perturbation improved as children used their bilateral cochlear implants over time and was significantly influenced by overall time in sound ($P = .02$).

Conclusions: Similar to unilateral cochlear implant users, children using bilateral implants have difficulty controlling long-term frequency perturbation and long-term amplitude perturbation during sustained phonations. These measures improved as the duration of usable hearing increases.


Cochlear Implants provide the perception of sound through the conversion of sound stimuli into electrical impulses, which are received by the cochlear nerve and processed by the central auditory system. Although cochlear implants do not restore sound perception as experienced by an individual with normal hearing, the implant provides the user with auditory feedback in the domains of timing, intensity, and frequency of sound. These auditory feedback cues may be critical for the user to monitor his or her speech production and to make purposeful moment-to-moment adjustments in voicing.1,5,7 More specifically, adults using cochlear implants have been reported to show improvements in accuracy of fundamental frequency, reduced variability in voice intensity, and more normal speaking rates.1,6,7

The effect of cochlear implantation on voicing in children has not been studied extensively. Our group previously showed that children using unilateral cochlear implants have difficulties with long-term fre-
frequency perturbation (vFo) and long-term amplitude perturbation (vAm) control during sustained phonations compared with children having normal hearing; these parameters translate clinically into perceived abnormalities of pitch and loudness control, respectively, and perceptually contribute to the persistence of a deaf voice in these children. As the duration of cochlear implant use increased in our cohort, there was an objective improvement in these voicing parameters, up to 6 months after surgery. Based on these initial data, we hypothesized that voice parameters can be predicted by the duration of auditory experience.

Auditory experience during development is critical to acquisition of speech and language. Indeed, as the period of early-onset auditory deprivation in children persists, the potential to achieve age-appropriate speech and language decreases once hearing is provided (eg, through cochlear implantation). Similarly, the development of normal voice parameters may be negatively affected by the duration of auditory deprivation, and access to sound will be of positive benefit to voice production. In support of this argument, our group recently demonstrated that a subset of children with postmeningitic hearing loss and short intervals of auditory deprivation achieved normal voice outcomes following unilateral cochlear implantation.

In our cochlear implantation program, many children who have received cochlear implants at older ages had access to sound through residual hearing and hearing aid use that predated cochlear implantation. These children were able to acquire speech and language using their hearing aids but then experienced progressive deterioration of hearing and required cochlear implants to provide sufficient auditory input. In the present study, we explored the influence of auditory experience before and after cochlear implantation on the development of voice production and examined these issues in children who were using bilateral cochlear implants.

**METHODS**

With approval of the ethics review board at our institution, all children using bilateral cochlear implants who were followed up at a pediatric cochlear implant center during a 4-month period (March 1, 2008, through June 30, 2008) were invited to participate in this study. Informed consent was obtained from the parent or primary caregiver of each child. Exclusion criteria included the following: (1) an inability to complete the voicing tasks, usually because of young age and (2) a history of laryngeal or voice pathology. A detailed history of hearing experience (aided and unaided) was obtained, and all previous audiograms reviewed. Twenty-seven children (17 males and 10 females) aged 3 to 15 years (mean age, 7.3 years) were enrolled. Causes of deafness included congenital (n=8), genetic (n=8), meningitis (n=3), cytomegalovirus (n=2), and unknown (n=6). The interval between first and second implantations ranged from simultaneous (n=3) to 7.8 years (mean, 4.2 years). Total duration of cochlear implant use (age at testing minus age at first cochlear implant activation) ranged from 0.3 to 9.8 years (mean age, 7.3 years) and required cochlear implants to provide sufficient auditory input. In the present study, we explored the influence of auditory experience before and after cochlear implantation on the development of voice production and examined these issues in children who were using bilateral cochlear implants.
of cochlear implant use (range, 1.2-11.8 years; mean, 5.2 years). We defined usable hearing as a pure-tone average, aided or unaided, of 40 dB or less. This cutoff was chosen because it represents a mild degree of hearing loss, meaning that conversational speech sounds are audible. Table 1 gives demographic details for each child.

All acoustic voice samples were collected and analyzed using a commercially available program (Multi-Dimensional Voice Program [MDVP] of the Computerized Speech Lab [CSL]) model 4500; KayPENTAX, Lincoln Park, New Jersey). A standardized procedure was used for all recordings. Children were seated in a quiet environment. A microphone was held in front of them at an off-axis position of 45° and a constant mouth-to-microphone fixed distance of 10 cm. Children were instructed to produce the vowel /a/ for 3 seconds using a comfortable pitch and volume. Three repetitions were performed, and the mean of the 3 trials was used for analysis. The phoneme /a/ was chosen because it is a steady-state vowel and is easy for children to reproduce.

All acoustic voice samples were analyzed using Multi-Dimensional Voice Program software, which extracts up to 33 acoustic parameters from each sample. For the present study, the following 5 parameters were chosen for further analysis: fundamental frequency, short-term frequency perturbation (jitter), short-term amplitude perturbation (shimmer), vFo, and vAm. These parameters are commonly used to measure voice pathology. Moreover, children with unilateral cochlear implants have been shown previously to have poorer control over the pitch and loudness of their voice than their counterparts with normal hearing. Measures of short-term perturbations of frequency (jitter) and amplitude (shimmer) were normal, as would be expected in a population without primary laryngeal pathology.

To examine the influence of implant experience on voice outcomes, vFo and vAm were plotted against the total duration of cochlear implant use as shown in Figure 1, respectively. For vFo and vAm, a trend toward normal values was seen with increased duration of cochlear implant use. However, this relationship was significant only for vFo control (P = .03). When these same measures were compared with the duration of bilateral cochlear implant use, no significant relationship was found (P = .45 for vFo and P = .52 for vAM). Finally, when vFo and vAM were plotted against overall time in sound as shown in Figure 2, respectively, a similar trend emerged. The longer a child was exposed to sound, the better his or her control of pitch and loudness was. However, this relationship was statistically significant only for vFo (P = .02).

To investigate the contribution of all demographic variables on voice production parameters, a stepwise multiple linear regression analysis was performed. Predictor variables included in the model were sex, age at testing, age at first implant activation, age at second implant activation, and overall time in sound. Dependent vari-

### Table 2. Bilateral Cochlear Implant Users vs Pediatric Norms

<table>
<thead>
<tr>
<th>Variable</th>
<th>Bilateral Cochlear Implant Users, Mean (95% Confidence Interval)</th>
<th>Pediatric Norms, Mean</th>
<th>P Value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Fundamental frequency, Hz</td>
<td>268.19 (248.41-287.97)</td>
<td>279.05</td>
<td>.27</td>
</tr>
<tr>
<td>Jitter</td>
<td>1.36 (1.05-1.67)</td>
<td>1.24</td>
<td>.43</td>
</tr>
<tr>
<td>Shimmer</td>
<td>3.68 (3.25-4.10)</td>
<td>3.35</td>
<td>.13</td>
</tr>
<tr>
<td>Long-term frequency perturbation</td>
<td>3.31 (2.77-3.85)</td>
<td>1.75</td>
<td>&lt;.001</td>
</tr>
<tr>
<td>Long-term amplitude perturbation</td>
<td>25.3 (22.2-28.1)</td>
<td>15.1</td>
<td>&lt;.001</td>
</tr>
</tbody>
</table>

* Data are given as percentages unless otherwise indicated.
The results of the present study show a similar pattern in children who use bilateral cochlear implants. As summarized in Table 2, control of vFo and vAM was significantly worse in children with bilateral cochlear implants (compared with preexisting pediatric normative data). Therefore, despite the incredible opportunities that cochlear implants provide for auditory and linguistic development, abnormalities in acoustic voice outcomes persist.

Duration of cochlear implant use was found to significantly influence acoustic voice parameters. In a previous study at our center, Campisi et al.8 showed an improvement in control of vFo and vAm as the duration of unilateral cochlear implant use in children increased up to 6 months (the result was statistically significant only for vAm). As shown in Figure 1, a trend toward normal values for vFo and vAm was seen with increased total duration of cochlear implant use. This relationship was significant only for control of vFo. The duration of bilateral cochlear implant use did not seem to significantly influence voicing parameters in this group of children. Although our cohort of bilateral cochlear implant users was young, the most important factor influencing voice outcomes at this stage of bilateral implant use seemed to be the overall time a child was exposed to usable hearing and not the number of cochlear implants being used. However, specific effects of bilateral cochlear implantation on voice may emerge with longer duration of cochlear implant use that might be related to the period of interimplantation delay.

Although the longer children use a cochlear implant, the better their voice outcomes seem to be for control of vFo and vAm, it is unclear if this relationship is strictly because of implant use or some other confounding variable. Age at implantation and duration of cochlear implant use are related. Children who receive implants at younger ages have often had longer durations of cochlear implant experience by the time of testing. Because of shifting treatment paradigms and inclusion criteria, children who are older at the time of testing tend to have received implants at an older age. In addition, most children, especially those who received implants for the first time at an older age, are likely to have experienced some usable hearing before implantation using hearing aids.

In the present study, a stepwise multiple linear regression analysis was performed to investigate the contributions of age, sex, number of cochlear implants, duration of cochlear implant use, and overall time in sound on acoustic voice outcome parameters. As shown in Figure 2, overall time in sound was the only variable found to significantly influence control of vFo, even after accounting for all other potential confounders. This result underlines the importance of early recognition and treatment of children with hearing loss to provide auditory experience as soon as possible.
nations. These measures improve as the duration of exposure to usable hearing increases.

Targeted speech therapies that assist children using cochlear implants in monitoring and modifying the pitch and loudness of their voice would be useful in this setting. Future work is planned to develop and evaluate therapeutic tools that will specifically address these areas in implant recipients, with the objective of minimizing or eliminating these voice abnormalities.

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Author Contributions: Drs Holler, Campisi, Harrison, and Papsin had full access to all the data in the study and take responsibility for the integrity of the data and the accuracy of the data analysis. Study concept and design: Holler, Campisi, Harrison, Papsin, and Gordon. Acquisition of data: Holler, Allegro, Harrison, and Papsin. Analysis and interpretation of data: Holler, Campisi, Allegro, Chadha, and Gordon. Drafting of the manuscript: Holler, Campisi, Allegro, Chadha, and Gordon. Critical revision of the manuscript for important intellectual content: Holler, Campisi, Chadha, Harrison, and Papsin. Statistical analysis: Chadha and Gordon. Obtained funding: Campisi and Papsin. Administrative, technical, and material support: Holler, Campisi, Harrison, and Papsin. Study supervision: Campisi, Harrison, Papsin, and Gordon.

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