Normal-like Motor Speech Parameters Measured in Children With Long-term Cochlear Implant Experience Using a Novel Objective Analytic Technique

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IMPORTANCE Although voice has been studied extensively in children who use cochlear implants (CIs), speech production has not been studied in this population using the Motor Speech Profile. Whether children who receive CIs gain normal speech production abilities is unknown.

OBJECTIVE To assess speech and articulation in deaf, long-term CI users who had undergone early unilateral cochlear implantation, compared with their normal-hearing peers.

DESIGN, SETTING, AND PARTICIPANTS Cross-sectional study at a tertiary pediatric hospital of 16 children aged 8 to 17 years who had undergone early implantation, are longstanding users, and had excellent audiogram and speech perception scores. Results were compared with a historical pediatric normal-hearing group.

INTERVENTION Unilateral cochlear implantation.


RESULTS The CI users had normal articulation and timing but poorer than normal intonation stimulability, particularly frequency variability. Diadochokinesis rates were within the 95% confidence interval of age-matched pediatric norms for 11 of 16 (69%) and 11 of 15 (73%) children with CI when they were performing /pa/ and /pataka/ tasks, respectively. The magnitude and rate of the second formant transitions were within normal limits for 9 of 16 (56%) and 10 of 12 (83%) children, respectively. The variability in frequency and amplitude of intonation stimulability domains were within normal limits for 7 of 16 (44%) and 16 of 16 (100%) children, respectively. The syllabic rate and duration were both within normal limits for 14 of 16 children (88%).

CONCLUSIONS AND RELEVANCE Despite significant improvements in speech after cochlear implantation, abnormalities remain, particularly in frequency variability. Such deviations can present as a decreased expression of emotion in speech and likely reflects decreased auditory frequency resolution provided by the CI. These deficits have been the focus of ongoing work to advance CI technologies and speech-processing strategies.
It is reasonable to suspect abnormalities in speech production in children who are deaf. The auditory system is necessary for normal voice, speech, and language development in children. Through an auditory feedback mechanism, speech sound is acquired prelinguistically.1 Later in life, this same feedback modulates voice and speech production via continuous fine adjustments to voice and speech.2,3

By contrast, children who are deaf demonstrate abnormal voice and speech production; specifically, they have less pitch variation,4 less variation in fundamental frequency,5 more period-to-period variation in fundamental frequency,6,7 and abnormalities with intonation6-7 as compared with normal-hearing peers. A study using a standard method of voice assessment (multidimensional voice program [MDVP]) found that children with profound deafness awaiting cochlear implantation had poor frequency and amplitude control during sustained vowel phonation as compared with normal-hearing peers.8

Voice has been studied extensively in this population,9-11 as has acquisition of speech and language.12-18 However, the objective techniques used to study motor speech parameters have not been used in the population of pediatric cochlear implant (CI) users beyond validating pediatric norms.9,19 In the present study, we used these tools to examine whether children who receive CIs gain normal speech production abilities. A systematic review of the literature revealed that this question has not yet been answered.9

Because CIs provide children who are deaf with access to sound, it is hoped that they will also achieve more normal speech production skills. Cochlear implant electrodes directly stimulate nerve fibers in the cochlea, providing some auditory input, albeit limited and unlike that of normal-hearing cochleae. Given the diminished representation of voice frequency provided by CIs,8 we hypothesize that long-term CI users will not achieve completely normal intonation as measured by objective motor speech parameters.

The primary objective of this study was to assess motor speech characteristics in children with early-onset deafness who were provided with unilateral CIs early in life and who had considerable experience with these devices. A standardized and validated hardware-software package with known pediatric normative values19 was used to collect objective measures of their speech production and to determine whether these skills differ from those of a group of normal-hearing peers. A secondary objective was to assess whether voice quality differs between longstanding CI users and normal-hearing children.20 Collection and dissemination of such data may become important in understanding the Motor Speech Profile (MSP) for clinical routine use, particularly in tertiary or quaternary voice clinics.

Methods

This study was approved by the Research Ethics Board of the Hospital for Sick Children, Toronto, Ontario, Canada. Informed written consent was obtained for each child. The study sample consisted of 16 children who received unilateral CIs (14 right, 2 left) during the period 1994 through 2003. Children with voice disorders or motor speech disorders and those unable to repeat a short passage in English were excluded. Motor speech parameters of participants with CIs were compared with those of a cohort of 112 normal-hearing children.19

Outcome Variables

The MSP, in conjunction with the Computerized Speech Laboratory (KayPENTAX), is an integrated software-hardware system for voice and speech analysis. The MSP was used to provide a noninvasive, reproducible, and objective measure of motor speech in the present study and in the previously published normative study.19

Children were asked to complete a set of program-prescribed tasks that were then used to assess different voice and speech parameters (Table 1): diadochokinetic (DDK) rate, second formant transitions, intonation patterns, and syllabic rates. The DDK rate was measured during repetition of single-syllable and multisyllabic stimuli (eg, /pa/ and /pataka/). The MSP also assessed second formant transition during vowel repetition /i-u/, intonation stimulability during production of sentence stimuli, and syllabic rate during production of sentence stimuli and conversational speech (Table 1). Details of materials, equipment, and methodology used to acquire the data have been previously reported.19 The normative values from this earlier study constitute our historical control group.

Covariates: Other Potential Predictors of Speech and Voicing

In the CI cohort, information on potential predictors of speech and voicing was collected. Demographic data including children’s age at time of assessment, first language, communication mode, history of developmental delay, parent and sibling hearing status, and education mode were recorded.

| Table 1. Motor Speech Profile (MSP) Tasks and Output* |
|---------------------------------|-----------|-----------------|--------|--------|
| Motor Speech Domain       | MSP Task | MSP Parameter   | Unit   |
| DDK                     | /pa/     | paDDK rate      | /s     |
|                         | /pataka/ | patakaDDK rate  | /s     |
| F2                      | /i-u/    | Rate of F2 variation | /s |
|                          |          | Magnitude of F2 variation | Hz |
| Intonation stimulability | “Are you leaving today or tomorrow?” | rvF0     | %     |
|                          |          | rvAM             | %     |
| Syllabic rate and duration | “We knew you were away all year.” | SSrate  | /s     |
|                          |          | SSDur            | ms    |

Abbreviations: DDK, diadochokinesis; F2, second formant transition; rvAM, amplitude variability; rvF0, frequency variability; SSDur, syllabic duration; SSrate, syllabic rate.

* Modified from Wong et al.19
Voice Disorders
The absence of voice abnormalities was confirmed using both perceptual and acoustic voice analysis. The MDVP was used to assess acoustic parameters including jitter, shimmer, fundamental frequency variation ($vF_0$), and peak amplitude variation ($vAM$). Jitter and shimmer percentage are measures of the relative period-to-period variability of pitch (frequency) and amplitude, respectively. As compared with these short-term measures, $vF_0$ reflects the variation of the fundamental frequency ($vAM$) reflects the peak-to-peak amplitude variations, both over the long term. A sustained vowel was used to analyze the MDVP as required by the software.

The Consensus Auditory Perceptual Evaluation of Voice (CAPE-V) was used to perceptually assess voice production. All CAPE-V parameters were reported: overall severity, roughness, breathiness, strain, pitch, loudness, and a summative total score. The maximum score for each parameter is 100, providing a maximum total score of 600, with a higher score indicating more deviant or abnormal voice quality. This was measured by a single expert rater (L.R.), the speech language pathologist for the Centre for Paediatric Voice and Laryngeal Function at the Hospital for Sick Children.

Auditory Experience
To assess the effect of past auditory experience on speech, “time in sound” was calculated by subtracting sound deprivation time from the child’s age at time of assessment. Duration of auditory deprivation was calculated as the time from identification of deafness to hearing aid fitting plus the time from identification of inadequate hearing aid use, defined as hearing thresholds of greater than 40 dB hearing level (with or without hearing aids), to cochlear implantation. We also reviewed audiograms and a speech perception battery to the nearest date within the 3-month period following hearing aid activation and CI activation to confirm expected thresholds (20-40 dB hearing level) after activation of these devices.

Speech Perception
The speech perception battery included closed and open set tests based on the child’s age and abilities to provide the required response. The results of these tests can be combined using the Pediatric Ranked Order Speech Perception (PROSPER) score.21 The PROSPER score, which ranges from 0 (poor) to 34 (excellent), was created to integrate all available speech perception outcome results into 1 score that could be followed over time because of the typical change in speech perception tests used as children age and acquire oral speech and language.21 For this study, we report PROSPER scores within 6 months of the speech production test date to confirm the degree of speech perception.

Statistical Analyses
Three tokens were analyzed and averaged for each MSP task. We a priori selected 8 MSP parameters to be studied on the basis of clinical experience and our hypothesis that there would be differences between the control and experimental groups.

All data were tested for normality, and when it was not found, nonparametric descriptors and tests were used. We used a 2-sided $t$ test to compare means for the MDVP parameters between our group and a group of normal-hearing children and deaf children.8,20 We present our main findings in 8 graphs, 1 for each MSP parameter. A previous study from our group demonstrated that speech, as measured through MSP parameters, develops with age.19 Linear regression analysis of the normative data was performed for each of the 8 speech parameters and time in sound, which for the control group is simply age whereas for the experimental group was calculated as described. Regression lines, as well as the 95% confidence interval lines of the data, rather than the 95% confidence interval of the regression line, were plotted. The CI group data were then plotted on each of these 8 graphs to assess how many children fell outside the 95% confidence interval of the normative data. The aforementioned covariates between CI users who fell within and outside the age-specific norms were then compared.

Results

Control Group: Normal-Hearing Children
Our historical normative control group was based on 112 children (54 girls, 58 boys) between the ages of 4 and 18 years.19 Because sex was not a significant predictor for any of the 8 MSP parameters,19 we combined the data across sexes for regression analyses.

Cochlear Implant Cohort: Deaf Children With CIs
The demographic and audiologic history of our experimental group is described in Table 2. These were 16 children, all of whom used English as their first language, used oral communication as their primary communication mode at home and at school, and wore bilateral hearing aids after identification of their deafness. One child had mild developmental delay, and another child had a parent and a sibling with substantial hearing loss necessitating amplification. All children previously benefitted to a limited degree from hearing aid use; most had undergone cochlear implantation at a young age and were long-standing users at voice assessment. After cochlear implantation, their pure-tone average and speech perception scores were normal; maximum PROSPER scores were reached by all children.

Objective and Subjective Voice Quality Measures
The MDVP (objective) parameters of voice are summarized in Table 3. Mean measures of jitter and shimmer are within established pediatric norms.20 Although the difference in shimmer is statistically significant compared with normative data, the difference between these 2 groups is not clinically significant because all of the children presented with normal or clear voice quality. It is not surprising that these short-term measures are normal in this group of children without suspected vocal disease. The mean $vF_0$ (3.54%) and $vAM$ (23.73%) were elevated from established pediatric norms (Table 3)20 and comparable to those of a group of children awaiting cochlear im-
plantation with nonsignificant t test P values (not shown in Table 3). In other words, these experienced unilateral CI users had considerable difficulty with long-term control of frequency and amplitude during sustained vowel production, as has been previously observed in a group of children awaiting cochlear implantation.8

The CAPE-V (subjective) parameters of voice are summarized in Table 4. Median CAPE-V parameter and total scores were well within normal limits, reflective of perceptually normal or clear voice quality, as would be expected for a group of children without vocal disease.

MSP Analysis

As in other published pediatric voice studies, not all children were able to perform certain tasks of a required token, and in some cases, not all samples recorded were of acceptable quality for analysis. This is reflected in varying sample size for various MSP parameters as seen in Figures 1, 2, 3 and 4. Specifically, /pataka/ DDK rate was missing for 1 child and second formant transition rate could not be assessed in 4 children. The remainder of the MSP data are complete.

Figures 1 through 4 display the linear regression line with the 95% confidence interval for the normative data. Children with results outside the 95% confidence interval were considered statistically different (α < .05) from their age-matched normal-hearing peers. Most children in the CI group performed the speech tests within normal limits. Figure 1 shows that DDK rates were within the 95% confidence interval of age-matched pediatric norms for 11 of 16 (69%) and 11 of 15 (73%) children with CI when they were performing /pa/ and /pataka/ tasks, respectively. As shown in Figure 2, the magnitude and rate of the second formant transitions were within normal limits for 9 of 16 (56%) and 10 of 12 (83%) children, respectively. The variability in frequency and amplitude of intonation stimulability domains were within normal limits for 7 of 16 (44%) and 16 of 16 (100%) children, respectively, as shown in Figure 3. In Figure 4, the syllabic rate and duration were both within normal limits for 14 of 16 children (88%).

Figure 5 demonstrates a summary of data presented in Figures 1 through 4. The MSP parameters that had the lowest proportion of children falling within the normative range were (1) intonation stimulability (frequency variability), which appeared flat regardless of time in sound as shown in Figure 3A; and (2) the magnitude of the second formant transition plotted in Figure 2A.

Covariate Analysis

Our covariate analysis was limited by an overall small sample size and made more difficult by our exclusionary criteria. For instance, all children used English as their first language, were primarily oral communicators, and participated in mainstream education programs. Only 1 child had a developmental delay whereas another had a parent and sibling with hearing loss. The MDVP and CAPE-V results were largely normal and had narrow ranges, preventing us from performing meaningful subgroup analyses. Children with higher vF0 and vAM values were not more likely to have poorer MSP results. This group consisted of competent CI users. Post-CI audiograms and speech perception (PROSPER) scores were excellent. Even the PBK, an open set test of perception of monosyllabic words, had scores of greater than 80% for all children. The homogeneity in our group meant that we could not meaningfully identify covariates that might explain our MSP results.

<table>
<thead>
<tr>
<th>Table 2. Demographic Characteristics</th>
<th>Value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age at time of voice assessment, median (range), y (N = 16)</td>
<td>13.5 (8-17)</td>
</tr>
<tr>
<td>Duration of CI use prior to voice assessment, median (range), y (N = 16)</td>
<td>7 (1.5-12)</td>
</tr>
<tr>
<td>Female sex, No. (%) (N = 16)</td>
<td>7 (44)</td>
</tr>
<tr>
<td>Prelingual onset of hearing loss, No. (%) (N = 16)</td>
<td>16 (100)</td>
</tr>
<tr>
<td>Time between identification of deafness and HA fitting, median (range), mo (N = 16)</td>
<td>1 (0-4)</td>
</tr>
<tr>
<td>Audiology: initial pure-tone average with HA, median (range), dB (n = 12)</td>
<td>36 (22-58)</td>
</tr>
<tr>
<td>Age at initial CI, median (range), mo (N = 16)</td>
<td>33 (10-193)</td>
</tr>
<tr>
<td>Right-sided CI, No. (%) (N = 16)</td>
<td>14 (88)</td>
</tr>
<tr>
<td>Audiology: initial pure-tone average with CI, median (range) (n = 15)</td>
<td>17 (7-28)</td>
</tr>
<tr>
<td>PROSPER Score with CI, median (range) (n = 11)</td>
<td>34 (34)</td>
</tr>
</tbody>
</table>

Abbreviations: CI, cochlear implant; HA, hearing aid.

<table>
<thead>
<tr>
<th>Table 3. Multidimensional Voice Program (MDVP) Voice Quality Measures</th>
<th>Mean (SEM), %</th>
<th>t Test P Value</th>
</tr>
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<tbody>
<tr>
<td>Jitter</td>
<td>1.24 (0.07)</td>
<td>1.38 (0.18)</td>
</tr>
<tr>
<td>Shimmer</td>
<td>3.35 (0.12)</td>
<td>5.10 (0.63)</td>
</tr>
<tr>
<td>vF0</td>
<td>1.75 (0.08)</td>
<td>3.54 (0.67)</td>
</tr>
<tr>
<td>vAM</td>
<td>15.1 (0.8)</td>
<td>23.73 (2.42)</td>
</tr>
</tbody>
</table>

Abbreviations: CI, cochlear implant; SEM, standard error of the mean; vF0, fundamental frequency variation; vAM, peak amplitude variation.
Discussion

Without access to sound, children who are deaf develop abnormalities in voice production. Specifically, such children demonstrate less pitch variation, more period-to-period variation in fundamental frequency, and abnormal intonation when compared with normal-hearing peers. Using a standard method of voice assessment (MDVP), we found that children who are profoundly deaf awaiting cochlear implantation had abnormally poor frequency and amplitude control with sustained phonation compared with children with normal hearing.

In the present study, differences in voice quality between experienced CI users and deaf patients awaiting cochlear implantation were observed. Jitter and shimmer were within established pediatric norms, and the differences observed in our data set were not clinically relevant. This is not surprising because our cohort did not have vocal disease. Long-term CI users did however demonstrate substantial difficulty with long-term control of frequency and amplitude with sustained phonation, similar to a group of children awaiting cochlear implantation. This could be because the

<table>
<thead>
<tr>
<th>CAPE-V Parameter</th>
<th>Median (Range)</th>
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<tr>
<td>Overall severity</td>
<td>10 (0-20)</td>
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<tr>
<td>Roughness</td>
<td>9 (0-28)</td>
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<tr>
<td>Breathiness</td>
<td>2.5 (0-21)</td>
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<tr>
<td>Strain</td>
<td>0 (0-26)</td>
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<tr>
<td>Pitch</td>
<td>6.5 (0-23)</td>
</tr>
<tr>
<td>Loudness</td>
<td>0 (0-20)</td>
</tr>
<tr>
<td>Total</td>
<td>42.5 (12-89)</td>
</tr>
</tbody>
</table>

Figure 1. Diadochokinesis (DDK)

Figure 2. Second Formant Transition (F2)

The dotted line represents the linear regression line with the 95% confidence interval for the normative data (solid lines) for /pa/ DDK rate (A) and /pataka/ DDK rate (B). Each data point represents data from 1 child. Children with results outside the 95% confidence interval line were considered statistically different (α < .05) from their age-matched normal-hearing peers.
Figure 3. Intonation Stimulability

The dotted line represents the linear regression line with the 95% confidence interval for the normative data (solid lines) for intonation stimulability (frequency variability) (A) and intonation stimulability (amplitude variability) (B). Each data point represents data from 1 child. Children with results outside the 95% confidence interval line were considered statistically different (α < .05) from their age-matched normal-hearing peers.

Figure 4. Syllabic Rate and Duration

The dotted line represents the linear regression line with the 95% confidence interval for the normative data (solid lines) for syllabic rate (A) and syllabic duration (B). Each data point represents data from 1 child. Children with results outside the 95% confidence interval line were considered statistically different (α < .05) from their age-matched normal-hearing peers.

Figure 5. Proportion With Normal Speech by Motor Speech Profile (MSP) Parameter

Summary of data presented in Figures 1 through 4. Specifically, the proportion of children who fell within the normative range, from lowest to highest (ascending order), is plotted for each MSP parameter. DDK indicates diadochokinesis.
implant is not providing the necessary auditory input for sufficient auditory feedback to enable fine tuning of these voice features. Alternatively, the implant may have been provided too late in development to have had an effect.

The main objective of the present study was to examine speech production using the MSP in a cohort of children who are longstanding users of CIs. The cohort of children had excellent auditory and speech perception scores as measured by the PROSPER score. Most of the children (>70%) performed within normal limits for all of the MSP parameters measured except for the magnitude of the second formant transition. Although the attributes of speech are mostly normal after cochlear implantation, specific abnormalities persist compared with normal-hearing peers, particularly as related to frequency variability. Abnormal measurements of frequency variability may affect prosody, which is used to reflect emotions or emotional states during speech as has been previously demonstrated. We could not find demographic, family, or clinical predictors for outliers due to the relative homogeneity of our study sample.

These difficulties in emotional speech reception and expression are related to the inability of the implant to properly code pitch. The abnormalities that are not corrected with cochlear implantation require advances in implant technologies that would provide the CI recipient with more diverse auditory input. The future of implant technology lies in better neural survival, better electrodes to reach the auditory nerves, and better sound-coding strategies. At present, some have argued that it is best to preserve low-frequency acoustic hearing for access to pitch cues in combination with the electrical input in the cochlear basal segment.22,23

The measures obtained from the MSP are consistent with and extend those from prior reports. As far as we know, this is the first objective analytic report of speech abnormalities in experienced CI users who had undergone early implantation. An important strength of this study is the use of the MSP, which is objective and has well-developed pediatric normative values.19

These data must be interpreted in the context of the study design. This study involves narrow inclusion criteria: experienced CI users who had undergone early implantation with excellent speech perception and without any vocal disease. As a result, our results are likely not generalizable to all children using CIs. Nonetheless, many children using CIs would fall in this group and the narrow inclusion criteria helped us arrive at our conclusions.

Conclusions

Cochlear implantation is associated with significant improvements in motor speech production for children who are deaf; however, objective differences exist compared with normal-hearing peers, particularly as it relates to frequency variability. This can affect prosody and may result in blunted speech emotion expression, although verifying this would require additional study. Long-term CI users did not have demonstrable voice quality improvement after implantation, as they continue to have significant difficulty with long-term control of frequency and amplitude with sustained phonation. These findings may be attributable to limitations in the auditory feedback provided by a CI.

ARTICLE INFORMATION


Author Contributions: Drs Eskander and Gordon had full access to all of 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: Gordon, Tirado, Campisi.

Acquisition, analysis, or interpretation of data: All authors.

Drafting of the manuscript: Eskander, Tirado, Russell, Allegro, Campisi.

Critical revision of the manuscript for important intellectual content: Eskander, Gordon, Hopyan, Papsin, Campisi.

Statistical analysis: Eskander, Gordon.

Obtained funding: Papsin.

Administrative, technical, or material support: Eskander, Russell, Allegro, Papsin, Campisi.

Study supervision: Gordon, Papsin, Campisi.

Conflict of Interest Disclosures:

Previous Presentation: This study was presented in the form of a poster at the 2013 American Academy of Otolaryngology–Head and Neck Surgery Annual Meeting: September 23, 2013; Vancouver, British Columbia, Canada.

Additional Contributions: The authors thank the entire team of the Cochlear Implant Program at the Hospital for Sick Children, Archie’s Cochlear Implant Laboratory, and the Centre for Paediatric Voice and Laryngeal Function. Stephanie Jewell, HBSc, and Selena Verney, Cochlear Implant Laboratory, are owed thanks for assisting with the acquisition of missing data points. Ms Jewell was paid for her contribution to the study.

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