The Effect of Inter-device Interval on Speech Perception Performance among Bilateral, Pediatric Cochlear Implant Recipients

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Abstract

Objective:
To determine if prolongation of the inter-device interval in children receiving bilateral cochlear implants adversely affects speech perception outcomes.

Study Design:
Retrospective chart review

Methods:
Retrospective review of our pediatric cochlear implant database was performed. Children who had undergone revision surgery or had less than 12 months listening experience with either the first or second implant were excluded. The inter-device interval, best PB-k-W score from each ear, and demographic data about each patient was collected. A ratio of PB-k-W was generated (PB-k-W 2nd side/PB-k-W 1st side) to minimize potential confounding from other individual patient factors that affect speech outcomes.

Results:
240 children met study criteria. Mean age at first CI was 3.2 years (0.6-17.9), second 6.6 years (0.8-22.4). Mean best PB-k score from the first CI side was 83.8% (0-100), the second was 67.5% (0-100) (p<0.001). When PB-k ratio was plotted against inter-device interval, R² was 0.47 (p<0.001). When analyzed for hearing stability, those with a progressive loss history demonstrated less influence of prolonged inter-device interval on performance. Multi-variate analysis did not identify other factors influencing the ratio. A line of best fit for those with stable hearing loss suggested best outcomes were with an inter-device interval less than 3-4 years. Beyond 7-8 years, very few achieved useful speech recognition from the second CI.
Conclusions:

Where possible, the second implant should be received within 3-4 years of the first to maximize outcome in those with stable, severe to profound sensorineural hearing loss.

Keywords: Cochlear implant, bilateral, speech perception outcomes, inter-device interval

Level of Evidence: 4
Introduction

During the last decade, an increasing number of children with severe to profound hearing loss have received bilateral cochlear implantation, either as a simultaneous or sequential procedure (1). The benefits of having access to information from both ears, an effect described as the binaural benefit, is seen in normal hearing individuals, individuals with hearing aids and has been reported in bilateral cochlear implant patients (2, 3). Utilizing both ears can provide better separation of the speech acoustic signals from noise sources within the environment. For children with congenital or a pre-lingual onset of deafness, unilateral cochlear implantation mitigates the effects of auditory deprivation (4). However, without access to binaural hearing, children with unilateral implantation are at risk of poorer educational performance compared to their binaural hearing peer (5). The now well-known benefits of bilateral cochlear implantation include better speech understanding in noise, sound localization, a reduced listening effort and improved quality of life (1, 4-13). Furthermore the second device provides a backup for device issues on the first side and potentially avoids periods of sensory deprivation while such issues are being resolved (14).

Though the benefits of early implantation on the first side have been well studied (15-19) the exact impact of a prolonged period between the first and second surgeries in bilateral recipients (the inter-device interval) is not well known. Research on the central auditory system would suggest that the period of highest neural plasticity is the first 3.5 years of life (15). Small studies of the effect of inter-device interval have shown trends towards worse outcome with with increased time between sequential surgeries (20, 21). The aim of this study is to determine if prolongation of the inter-device interval in children receiving bilateral cochlear implants
adversely affects speech perception outcomes in a large cohort of children in a single institution with a strong auditory-verbal rehabilitation program.
Material and Methods

Study Design and Subjects

The Biomedical Institutional Review Board approved the study. A retrospective review was performed of all pediatric patients (<18 years of age) who underwent bilateral cochlear implantation (CI) at our institution or received post-operative audiological and speech therapy rehabilitation in our service between 1992 and 2013. Those who later received revision surgery were excluded. Those who had less than 12 months listening experience with both the first and second implant or were unable to perform speech and language testing were also excluded.

Intellectual disability was defined as having significant developmental delays in areas other than speech and language. Hearing loss onset was determined by a combination of parental report and first available audiogram.

For all included children, all clinical and audiological assessments made since first diagnosis were reviewed to get a longitudinal assessment of each individual.

Cochlear implants were performed only once the child fit criteria of bilateral severe to profound hearing loss or hearing not sufficient for appropriate speech and language development despite adequate amplification.

Those with bilateral severe to profound hearing loss at first diagnoses, which did not change over time, were categorized as “stable”. Those with less severe hearing loss that later progressed to become severe to profound were categorized as “progressive”.

All children were fit with binaural amplification prior to their first cochlear implant surgery. When significant residual hearing existed in the contralateral hear, continued hearing aid use was encouraged following implantation. Many rejected the second side hearing aid once the first cochlear implant was placed.
Speech Perception Testing

The open set measure used for this study was the Phonetically-Balanced Kindergarten word lists (PB-k words). The PB-k test consists of phonetically balanced lists of 50 monosyllabic words that the child is required to repeat (6). The PB-k lists were administered when a child had achieved a score significantly above chance on closed-set tests. Tests are administered in a sound controlled environment using live-voice or recorded stimuli presented at 60 decibel SPL to each ear separately. Best PB-k word scores from each ear were identified from the patient’s charts and the pediatric database once the child had at least 12 months of listening experience with each device. The best score achieved on the side of the second implanted ear was divided by the best score achieved on the side of the first implanted ear to create a ratio. This ratio was used to try to control for individual factors that affect speech perception outcomes, such as labyrinthine malformation, intellectual disability, medical co-morbidities and age at first implantation.

Statistical Analysis

Statistical analysis was performed using SPSS software version 20.0 (SPSS Inc., Chicago IL). Means were compared using t-tests. The ratio of second to first side PB-ks were plotted against the time between the first and second implant operations (inter-device interval) using scatter plots. Regression analysis was performed using both linear and non-linear regression models. Lines of best fit were plotted using a LOESS (Local Polynomial Regression) method with 99% of data-points used. Multi-variate analysis was performed to look for the potential influence of gender, age at first CI, side of first operation, stability of hearing loss, intellectual disability, presence of labyrinthine malformation, auditory neuropathy spectrum disorder (ANSD), medical
co-morbidities, hearing loss onset and known causes for hearing loss. Subset regression analysis was then repeated with data divided into those with progressive versus non-progressive (stable) hearing loss.
Results

There were 349 children with bilateral CIs identified in our database. Once those who underwent revision surgeries were excluded 290 remained. Only 14 patients received their CIs in simultaneous procedures; the rest of them underwent surgery for the second ear within a minimum of two-months and a maximum of 16.1 years 145 (50%) patients were male, and 203 (70%) of them received an implant on the right side first (Table 1). Mean age at first CI was 3.2 years (range 0.6-17.9 years) and at the second CI was 6.6 years (range 0.8-22.4 years). 214 (74%) children received a Cochlear Corp (Sydney, AUS) device, 25 (8.6%) a MedEl Corp. (Innsbruck, Austria), and 51 (17.6%) received an Advanced Bionics device (Valencia, California) (Table 2). The same manufacturer was always used on both sides. The cause and onset of hearing loss for the 290 subjects are shown in Figure 1 and 2.

PB-k word scores were available for both ears with a minimum of 12 months of listening experience for 240 children. The mean best PB-k score from the first implanted ear was 83.8 (range 0-100%) and from the second implanted ear 67.5 (range 0-100%) (p<0.001). The mean inter-device interval was 3.4 years (range 0-16.1 years) (Table 3).

Inter-device interval

The ratio of best PB-k second side over first side was plotted against inter-device interval (Figure 3). When a linear regression model was used the R^2 was 0.47 (p<0.001). This did not change significantly when a non-linear regression model was used. When multivariate regression was then performed with consideration of the following variables: gender, age at first implantation, side of first operation, intellectual disability, presence of a labyrinthine malformation, ANSD, medical comorbidities, hearing loss onset, known cause for hearing loss and stability of hearing...
loss, there was little change in the $R^2$ value ($R^2 = 0.48$). The only variable other than inter-device (p<0.001) interval to reach significance (p<0.05) was stability of hearing loss (p=0.041), suggesting the ratio format was sufficient to control for many of these individual patient factors (Table 3). A line of best fit calculated using the LOESS method indicated a trend towards worse speech perception score with the second implant when compared to the first implant as a function of more prolonged inter-device intervals. Very few individuals achieving 80% of his/her first side PB-k score (ratio 0.8) with an inter-device interval over 8 years.

Children with progressive hearing loss were separated from stable hearing loss and their second versus first side PB-k ratio plotted against inter-device interval (Figure 4). This analysis suggested that many of the outliers to trends seen in Figure 3 appeared to be related to those children with progressive hearing loss. When separate lines of best fit were calculated for each group it is clear that there is a more precipitous decline in outcome with prolonger inter-device interval in those with stable hearing compared to progressive hearing loss (Figure 4). When linear regression was performed for these two groups, there was greater correlation between inter-device interval and speech perception outcomes on the second side in those with stable hearing loss ($R^2 = 0.57, p <0.001$) compared to progressive hearing loss ($R^2 = 0.23, p<0.001$), suggesting the length of the inter-device interval has greater impact in this group. Again non-linear regression analysis and multi-variate analysis looking at gender, age at first CI, side of first operation, intellectual disability, presence of cochlear malformation, ANSD, medical comorbidities, hearing loss onset and known cause for hearing loss changed the $R^2$ values very little and no other factor reached statistical significance (Table 3). The line of best fit for those with stable hearing loss would suggest that the best chance of achieving 80% or more of the speech perception performance attained with the first implant is when the second side
implantation occurs within 3-4 years of the first operation. Furthermore, when the second device is received beyond 7-8 years after the first operation, very few achieved good speech perception from the second side.
Discussion

The results of this study demonstrate the negative consequences of prolonging the inter-device interval in children who receive sequentially placed, bilateral cochlear implants. The effect was more pronounced in those with stable severe to profound sensorineural hearing loss compared to those with a progressive decline in hearing, supporting the notion of a critical period of intervention to maximize the response of the auditory system to electrical stimulation following auditory deprivation. This is in accord with the current understanding of neural plasticity of the auditory system (15).

A recent survey sent electronically to 35 well-known cochlear implant clinics located in Europe, Australia, the United States, and Canada revealed that children who underwent bilateral cochlear implant (BCI) in sequential procedures make up 70% of all BCI children and 49% of the entire BCI user group (pediatric and adult) worldwide (22). This would suggest that, though simultaneous implantation is desirable, it is often not practically achieved. Knowledge of the impact of inter-device interval is important for maximizing outcomes for children.

The available studies on the impact of the inter-device implantation are conflicting. Previous studies published to date relating to the inter-device interval suggest that a longer interval has either a negative effect (4, 21, 23) or no impact (4, 24-26) on postoperative speech perception outcomes in quiet. Zeitler et al. reported that speech perception benefits continue to increase over time, despite the length of deafness in either ear, inter-device interval, or age at implantation (24). Two studies showed statistically significant poorer speech perception in quiet for longer inter-implant intervals or an older age at the second implantation (4, 27).

Sparreboom et al. and Grieco-Calub et al. report no such impact of older age at the second implantation on postoperative outcomes (28-31).
These studies have all been limited by small numbers (<100) of subjects or short follow-up periods (3 months of implant use). The current study achieves strong statistical evidence of the impact of inter-device interval through a large cohort and a minimum of 12-months of implant experience with each ear.

Furthermore, the analysis of the effect of inter-device interval on speech perception outcomes can be complicated by several factors that are known to influence speech perception scores such as; age at the first implantation, intellectual disability, presence of cochlear malformation, ANSD, medical comorbidities, hearing loss onset and known cause for hearing loss (1, 4-13).

The use of a ratio of second to first side outcomes in this study allowed isolation of the inter-device interval from other individual patient factors which may affect the raw speech perception outcome score achieved on the second side. In essence each individual child’s first side PB-k score was used as a benchmark for what that child is capable of achieving with all these factors present. By comparing the second side directly to this value, many of these factors stay the same and so should not influence the ratio generated. The multi-variate regression analysis, showing no significant impact of the aforementioned characteristics on the outcomes seen, validates this measure as a way to specifically control bias from these factors. Though likely to affect the magnitude of the PB-k score achieved by each ear, the factors that showed no affect on the ratio with multivariate analysis appear to have remained static between the first and second implant.

In contrast, the stability of hearing loss in our total cohort appeared to be a significant confounder, necessitating subset analysis. This was confirmed when a difference in outcome trend was seen in those with progressive hearing loss when separated from those with stable severe to profound sensorineural hearing loss.. This difference was further highlighted by a large shift in correlation values when the groups were separated (Figure 4).
There are many possible contributors to outcome in those with progressive hearing loss that change over time and may influence the speech perception achievable on the second side. In contrast to those children with stable, bilateral severe to profound sensorineural hearing loss, those with progressive losses have variable amounts of residual hearing in the second ear over different durations of time. Hearing aid use may therefore also be a significant factor in this group. A major restriction to the author’s ability to draw meaningful conclusions about those with progressive hearing loss is that significant longitudinal information about hearing aid use was not available, as data logging in many instances has only recently become available. Furthermore, this was considered an extremely complex task in those with progressive hearing loss, as it would require knowledge about the rate of decline in hearing, hours of hearing aid use and the frequency of programming the hearing aid to ensure the patient was maximally amplified in the setting of declining hearing. This was considered beyond the ability of a retrospective review to reliably do. In contrast, in those with stable severe to profound hearing loss, hearing aid use between the first and second implants is less likely to have affected speech perception outcomes. Conclusions of the impact of inter-device interval of speech perception outcomes were therefore confined solely to those children with stable (non progressive) severe to profound hearing loss.

The authors considered a good outcome to be that each child achieved at least 80% or more of the speech perception score that he/she achieved with their first side (ratio of 0.8). This was achieved by most children with stable hearing loss when the second implant surgery occurred within 3-4 years of the first side. After 7-8 years no child with stable hearing loss achieved this benchmark from the second side. In contrast, those with progressive hearing loss achieved good
outcomes with much longer intervals, with many achieving a ratio 80% or more, 9-10 years after their first implant.

This study does not aim to address the full impact of inter-device interval on speech perception outcomes with bilateral cochlear implantation. Previous studies on the impact of inter-device interval on speech understanding in noise have shown no effect (4, 25, 26) but have been limited by small numbers of participants, short follow up periods and been confined to older children (most over 7 years old). Further prospective studies on the effect of inter-device interval in bilateral CI experience could look at the impact on function in background noise and sound localization.
Conclusion

When considering bilateral, sequential cochlear implantation for children with stable severe to profound hearing loss, the present study suggests that results are better when the second device is implanted within 3-4 years of the first device placement. Moreover, inter-device intervals beyond 7-8 years result in much poorer performance when compared to first device performance. Further larger studies on the effect of inter-device interval in bilateral CI experience could look at function in background noise and sound localization for a longer period of time.
References


Figure legends

Figure 1. Cause of hearing loss

Figure 2. Onset of hearing loss

Figure 3. The ratio of the best PB-k achieved on the second side over the best PB-k achieved on the first side, plotted against inter-device interval (time between surgeries in years).

Figure 4. The ratio of the best PB-k achieved on the second side over the best PB-k achieved on the first side, plotted against inter-device in years (time between surgeries in years), broken down by progressive versus stable (non progressive) hearing loss.

Table 1. Demographics

Table 2. Overall results

Table 3. Multivariate analysis of regression analysis
Table 1. Demographics

<table>
<thead>
<tr>
<th>Variable</th>
<th>Percent of Study Group</th>
</tr>
</thead>
<tbody>
<tr>
<td>Male</td>
<td>50%</td>
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<tr>
<td>First Side - Right</td>
<td>69.7%</td>
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<tr>
<td>Intellectual Disability*</td>
<td>7.2%</td>
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<tr>
<td>Cochlear Malformation</td>
<td>11.4%</td>
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<tr>
<td>ANSD</td>
<td>14.1%</td>
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<td>Medical Co-morbidities</td>
<td>19.7%</td>
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<tr>
<td>Progressive Hearing Loss</td>
<td>30.7%</td>
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</table>

* Significant developmental delays other than speech and language
Table 2. Overall Results

<table>
<thead>
<tr>
<th>Variable</th>
<th>Mean</th>
<th>Std deviation</th>
<th>Range</th>
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<tbody>
<tr>
<td>Age first Implant</td>
<td>3.2 years</td>
<td>2.6 yrs</td>
<td>0.6-17.9 yrs</td>
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<tr>
<td>Age Second Implant</td>
<td>6.6 years</td>
<td>4.5 yrs</td>
<td>0.8-22.4 yrs</td>
</tr>
<tr>
<td>Inter-device Interval</td>
<td>1235.6 days</td>
<td>3.4 years</td>
<td>0-5869 days</td>
</tr>
<tr>
<td>First Side Best PB-k</td>
<td>83.8</td>
<td>20.0</td>
<td>0-100</td>
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<tr>
<td>Second Side Best PB-k</td>
<td>67.5</td>
<td>30.0</td>
<td>0-100</td>
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</table>

Mean PBK First side versus Second side p <0.001
Table 3: Multiple Regression Analysis of Ratio of Second side CI/First side CI speech perception score

<table>
<thead>
<tr>
<th>Factor</th>
<th>P value on Multivariate Analysis – Total Group</th>
<th>P value on Multivariate Analysis - Progressive Only Group</th>
<th>P value on Multivariate Analysis - Stable Hearing Loss Only Group</th>
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<td>&lt;0.001</td>
<td>&lt;0.001</td>
<td>&lt;0.001</td>
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<tr>
<td>Age at First Operation</td>
<td>0.374</td>
<td>0.677</td>
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<td>0.414</td>
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<td>0.135</td>
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<td>Cause for Hearing Loss</td>
<td>0.359</td>
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<tr>
<td>Gender</td>
<td>0.118</td>
<td>0.069</td>
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<td>Side of First Operation</td>
<td>0.239</td>
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<td>Progressive versus Stable</td>
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<td>n/a</td>
</tr>
</tbody>
</table>

* Significant developmental delays other than speech and language
Figure 1. Cause of hearing loss
89x69mm (300 x 300 DPI)
Figure 2. Onset of hearing loss
89x71mm (300 x 300 DPI)
Figure 3. The ratio of the best PB-k achieved on the second side over the best PB-k achieved on the first side, plotted against inter-device interval (time between surgeries in years).

R² = 0.47
p < 0.001
Figure 4. The ratio of the best PB-k achieved on the second side over the best PB-k achieved on the first side, plotted against inter-device in years (time between surgeries in years), broken down by progressive versus stable (non progressive) hearing loss.

Stable (non progressive) $R^2 = 0.57$
Progressive $R^2 = 0.23$
Both $p < 0.001$
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