Transcutaneous Osseointegrated Implants for Pediatric Patients With Aural Atresia

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**IMPORTANCE** Patients with aural atresia typically have maximal conductive hearing loss, which can have negative academic and social consequences. Transcutaneous osseointegrated implants (TOIs) can potentially restore hearing on the affected side.

**OBJECTIVES** To review the demographic, audiological, and surgical outcomes of TOI placement in pediatric patients with aural atresia and to describe a modification in incision technique in anticipation of later auricular reconstruction.

**DESIGN, SETTING, AND PARTICIPANTS** This retrospective case series reviewed 41 cases of TOI placement in pediatric patients between January 1, 2014, and September 30, 2016, at Lurie Children’s Microtia and Aural Atresia Clinic. Patients, all younger than 18 years and with atresia or microtia, received at least 6 months of follow-up and underwent testing before and after surgery.

**MAIN OUTCOMES AND MEASURES** Patient age, indication for procedure, ear sidedness, case length, incision type, complications, and other postoperative events. Audiological outcomes before and after implantation were measured using pure-tone averages and the Hearing In Noise Test for Children, presented in variable signal to noise ratios.

**RESULTS** In total, 46 TOIs were performed in 38 pediatric patients, but only 41 implantations in 34 patients were included in this study. Of the 34 patients, 13 (38%) were males and 21 (62%) were females, with a mean age of 8.9 (range, 5-17) years at the time of TOI placement. Microtia on the implanted side was present in 39 cases (95%). A modified posterior-superior scalp incision technique was used in 30 (73%) of 41 ears, all in cases of microtia. One perioperative surgical complication occurred: a seroma requiring drainage. Two patients developed minor skin irritation and erythema at the magnet site related to the overnight use of the processor, which resolved when removed while sleeping. The mean (SD; range) score for the Speech In Noise test at 5 dB signal to noise ratio improved from 75.3% (14.4%; range, 50%-92%) correct in unaided/preoperative condition to 93.6% (6.95%; range, 80%-100%) correct in the aided/postoperative condition. The mean improvement in score was 18.3% (95% CI, 10.8%-25.9%), with an effect size of 1.62 (95% CI, 0.95-2.29). The mean pure-tone averages (SD; range) similarly improved from 63.7 (13.2; range, 25-11) dB to 9.6 (4.9; range, 5-15) dB.

**CONCLUSIONS AND RELEVANCE** Transcutaneous osseointegrated implantation has a low complication rate among pediatric patients with atresia or microtia and can provide excellent audiological results. It should be included as a treatment option for this population of patients who meet audiological criteria.
Osseointegrated implants are well established for unilateral conductive, mixed, or sensorineural hearing loss as well as bilateral mixed or conductive loss. These implants include percutaneous implants with an abutment (Baha, Cochlear Ltd; Ponto, Oticon Medical) and, more recently, transcutaneous implants, which use an implanted magnet coupled with a magnet on the processor (Sophono Alpha 2, Medtronic; Baha Attract, Cochlear Ltd). Both pediatric patients (<18 years) and their parents often find the transcutaneous implant appealing because it is hidden under the skin, allowing for a near-normal–appearing implant site when the processor is removed. In addition to favorable aesthetics, the transcutaneous magnetic-based implants have also shown comparable hearing results in early trials, albeit with some concern that they may provide less sound transduction than do percutaneous implants.

Patients with aural atresia, often with concurrent microtia, are excellent candidates for osseointegrated devices. These patients have maximal conductive hearing loss (CHL) but usually normal bone conduction hearing based on audiometry. Therefore, the osseointegrated implant can “bypass” the missing ear canal. A recent systematic review of atresioplasty and osseointegrated device placement showed better hearing outcomes and a lower surgical complication rate with the osseointegrated devices. As opposed to atresioplasty, implantation is reasonably quick, is safe with relatively low morbidity, and typically restores hearing on the atretic side to normal or near-normal levels.

The functional results prove even more important as a growing body of research indicates that children with aural atresia or microtia are at higher risk for developmental and academic delays. For example, Jensen et al performed a retrospective medical record review of 74 patients with aural atresia to investigate whether this population is at an increased risk for speech and learning problems. They found that 86% of patients with bilateral aural atresia and 43% of patients with unilateral aural atresia required speech therapy and that both school problems and educational interventions were common in this population. This improvement in hearing status, along with the aesthetic advantage of transcutaneous devices, further contributes to the growing interest in implantation for these individuals.

At Lurie Children’s Microtia and Aural Atresia Clinic (Chicago, Illinois), we follow more than 200 children with microtia, most of whom have atresia with associated maximal CHL. These patients have varying degrees of disability related to their hearing loss. We have established a standard protocol for audiological testing that includes a trial with a bone-conduction headband using speech perception tests in varying noise levels. We recommend implantation for those children (and parents) who show that the headband is useful and who are interested in pursuing surgery.

Bilateral implantation has historically been somewhat controversial given that a single implant provides a signal to both cochleae. However, evidence, including a systematic review, shows that bilateral implantation provides advantages in sound detection and localization as well as improved speech perception in both quiet and noise. Pediatric patients report an increased quality of life and high satisfaction scores with bilateral implants. Bilateral implants also ensure that patients have access to at least 1 functioning implant should the other processor malfunction. For these reasons, we offer concurrent bilateral implants to patients with bilateral atresia.

In addition, we have developed a modified incision for patients with microtia who are candidates for auricular reconstruction (Figure 1 and Figure 2). The standard incision is performed near the postauricular sulcus, directly in the site of the skin pocket used during auricular reconstruction surgery.
which may compromise the skin flap. To avoid this possibility, we place the incision posterosuperiorly in the scalp. The incision is designed to allow the implant to sit 5.5 to 6.5 cm from the presumed site of the external auditory canal, using the contralateral side as a guide if the microtia is unilateral. To do so, we trace the circle of the magnet around the implant site using the template and then mark the incision as an arc 15 mm from the internal magnet site.

In this study, we sought to examine our experience with transcutaneous osseointegrated implants (TOIs) in pediatric patients with aural atresia. In addition to discussing audiometric results before and after implantation, we review patient demographics and surgical outcomes.

Methods

We performed a retrospective review of pediatric patients who underwent TOI placement between January 1, 2014, and September 30, 2016, at Lurie Children’s Microtia and Aural Atresia Clinic (Chicago, Illinois). At the beginning of the study period, 2 types of TOI were available for use: Sophono Alpha 1 or 2 and Baha Attract. However, the audiological protocol was applied specifically to patients using Baha Attract, which is the only device that we have used in the past 3 years. For clarity and because the surgical technique and magnets differ, we included only patients using Baha Attract in this study. This study received approval from the institutional review board of the Ann & Robert H. Lurie Children’s Hospital of Chicago. Written patient informed consent was obtained from all participants.

Patient demographics, including age at implantation and the presence of microtia or aural atresia, were identified. Surgical demographics included length of surgery, scalp thickness, unilateral vs bilateral implantation, and implant size. Postoperative data included loading time, initial magnet strength, and postsurgical complications. All patients who underwent TOI placement were seen or contacted 1 week after surgery and evaluated 1 month after surgery, which is typically when the device is activated. Patients were then evaluated at 3, 6, and 12 months after surgery and then yearly thereafter. Speech perception testing on patients was completed before (unaided) and after (aided) surgery if time permitted. The Hearing In Noise Test for Children (HINT-C) at 50 dB in aided and unaided conditions was presented in quiet and with variable signal to noise ratios of 10, 5, and 0 dB. Testing was completed in noise because this type of testing better reflects the patient’s ability to understand spoken language during daily-life listening situations in which competing background noise is present. The most reflective of classroom conditions is 5 dB, and therefore the 5-dB HINT-C was used after surgery. Preoperative and postoperative pure-tone averages (PTAs) were calculated using 0.5, 1, 2, and 3 kHz frequencies. Effect size (Cohen’s $d$) and number needed to treat were calculated to assess the clinical significance for the HINT-C before and after implantation.

Results

Patient Demographics

In total, 46 Baha Attract TOIs were placed in 38 pediatric patients from January 1, 2014, to September 30, 2016. Forty-one implantations were performed for 34 patients with aural atresia and were included in our results. All patients had a follow-up of more than 6 months. Of the 34 patients, 13 (38%) were males and 21 (62%) were females, with a mean age of 8.9 (range, 5-17) years at the time of TOI placement. “Older” patients (>10 years) in this study fell into 2 main categories: (1) they had not been seen at our clinic and were not aware of the option for an osseointegrated implant or (2) they were not having academic difficulties previously but, as school became more difficult and social situations more complex, began to struggle and sought improved hearing. All (100%) of the TOIs were done for maximal CHL. Microtia was present in 39 cases (95%). One patient had hemifacial microsomia, and another patient had Treacher Collins syndrome.

Surgical Results

For the 34 unilateral TOIs, 20 (59%) were right-sided implants and 7 (21%) were left-sided (Table). Seven patients (21%) had concurrent bilateral implantation for bilateral aural atresia. Thirty-nine (95%) of the 41 cases were done in a single stage, and of the remaining 2 patients, 1 had a previous percutaneous implant with chronic skin overgrowth that necessitated the removal of the abutment. This patient was the only one in the study who had a prior implant. After 3 months of skin healing, a Baha Attract magnet was fitted into his existing implant. The other patient had a 2-staged percutaneous implant planned, but she opted for the transcutaneous magnet at the second stage when it became available between her stages.

The modified posterior-superior incision was used in 30 of 41 ears (73%), all in cases of microtia. The 4-mm implant was used in 33 of 41 ears (80%), and the 3-mm implant was used for the remaining 8 ears (20%). The mean scalp thickness mea-
Transcutaneous Osseointegrated Implants for Pediatric Patients With Atresia

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Objective: To evaluate the outcomes of children and adolescents with unilateral atresia who received transcutaneous osseointegrated implants (TOIs) and to correlate scalp thickness and magnet strength.

Methods: This study describes the results of TOI placements for 41 pediatric patients. Preoperative and postoperative PTA data were available for 32 patients. The mean (SD; range) preoperative PTA was 63.7 (25.4; range, 15-11) dB. The mean (SD; range) postoperative PTA was 9.6 (6.1; range, 2-15) dB with the use of the processor. Preoperative and postoperative HINT-C results were available for 12 patients. The mean (SD) speech in noise test at 5 dB SNR signal to noise ratio improved from 75.3% (14.4%; range, 50%-92%) correct in unaided/preoperative condition to 93.6% (6.95%; range, 80%-100%) correct in the aided/postoperative condition. The mean improvement in score was 83.8% (95% CI, 10.8%-25.9%), with an effect size of 1.62 (95% CI, 0.95-2.29). Therefore, based on this effect size, the number needed to treat to achieve a favorable outcome is 1.7.

Discussion

This study describes the results of TOI placements for 41 pediatric patients with aural atresia, making our pediatric series with TOI the largest to date, to our knowledge. Carr et al, reporting on 10 patients with a mean age of 43.8 years who received TOI, found an improvement in word discrimination scores when implantation indications included single-sided deafness, otosclerosis, meatal stenosis, and hearing loss associated with chronic mastoid drainage or chronic suppurrative otitis media. Iseri et al presented a series comparing TOI outcomes with percutaneous osseointegrated implant outcomes and included 7 (in a total of 16) pediatric patients receiving the Baha Attract device. They reported mean speech reception threshold gains of 24 dB in the transcutaneous group; implanted indications included hearing loss with chronic suppurative otitis media or atresia. Baker et al compared Baha Attract with Sophono and reported on 17 children (mean age, 10.7 years) with TOIs. Six children underwent implantation for single-sided deafness and conductive loss secondary to osicular chain abnormalities, and they experienced a mean speech reception threshold improvement of 56 dB HL (hearing level) and PTA improvement of 41 dB HL.

Table. Demographics, Operative Findings, and Perioperative and Postoperative Complications for All Cases

<table>
<thead>
<tr>
<th>Variable</th>
<th>Cases, No. (%) (N = 41)</th>
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<tbody>
<tr>
<td>Demographics</td>
<td></td>
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<tr>
<td>Ears with aural atresia</td>
<td>41 (100)</td>
</tr>
<tr>
<td>Conductive hearing loss</td>
<td>41 (100)</td>
</tr>
<tr>
<td>Ears with microtia</td>
<td>39 (95)</td>
</tr>
<tr>
<td>Bilateral</td>
<td>7 (21)</td>
</tr>
<tr>
<td>Right-sided</td>
<td>20 (59)</td>
</tr>
<tr>
<td>Left-sided</td>
<td>7 (21)</td>
</tr>
<tr>
<td>Syndromic patients</td>
<td>3 (7)</td>
</tr>
<tr>
<td>Female patients</td>
<td>25 (61)</td>
</tr>
<tr>
<td>Male patients</td>
<td>16 (39)</td>
</tr>
<tr>
<td>Operative findings</td>
<td></td>
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<tr>
<td>Use of modified posterior-superior incision</td>
<td>30 (73)</td>
</tr>
<tr>
<td>Requiring scalp thinning</td>
<td>3 (7)</td>
</tr>
<tr>
<td>Use of the 4-mm implant</td>
<td>33 (81)</td>
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<tr>
<td>Use of the 3-mm implant</td>
<td>8 (20)</td>
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<tr>
<td>Single-stage surgery</td>
<td>39 (95)</td>
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<tr>
<td>Perioperative and postoperative</td>
<td></td>
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<tr>
<td>complications</td>
<td></td>
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<tr>
<td>Seroma</td>
<td>1 (2)</td>
</tr>
<tr>
<td>Skin site pain and erythema at magnet site</td>
<td>2 (5)</td>
</tr>
</tbody>
</table>

* Two patients, 1 case bilateral.
* Twenty-one patients, 4 bilateral.
* Thirteen patients, 3 bilateral.
* Required incision and drainage for treatment.
* Resolved after discontinuing nighttime external processor use.

Magnet strength plotted as a function of scalp thickness for each implanted device. Some data points are superimposed because they represent the same values. Pearson correlation testing reveals a modest positive correlation (Pearson r = 0.36; n = 38).
Our study did not directly compare the results of different osseointegrated devices. However, the positive results in these previous studies correlate well with our findings of significant improvement on multiple audiometric variables in the HINT-C before and after implantation, particularly in the Speech In Noise Test. Although the HINT-C data were available for only 12 patients, the improvement in scores was clinically significant (number needed to treat, 1.7), and these 12 patients all had unilateral aural atresia. Therefore, our results indicate that a TOI can improve hearing in noise even when the child has a normal hearing ear.

Clinical evidence for academic deficiencies and decreased hearing-related quality of life in children with unilateral sensorineural hearing loss has been well established, and evidence of similar rates of academic and social challenges for patients with CHL due to aural atresia is growing.10,11 In addition, laboratory results have shown that there is a critical window for the development of the auditory cortex and auditory processing, and unilateral hearing loss can lead to permanent changes to auditory perception.12-16 This process is also seen with unilateral CHL, with the development of amblyaudio and persistent distortions in the neural tonotopic maps when CHL was induced during the critical window.17

We believe it is important to follow up with patients with aural atresia and associated hearing loss, and we advocate for hearing resources as needed. This effort includes receiving regular audiograms and intervention within the school system for preferential seating and frequency modulation systems. Patients are encouraged to undergo a trial of a bone-conduction aid with the HINT-C at different signal to noise ratios to characterize their speech perception abilities with or without the aid. Often, patients are not aware of how much they are missing. Implantation is beneficial even for older children who already have fully developed language skills. For example, the oldest patient in our study who underwent TOI placement improved his scores from 71% unaided to 100% aided on the 5-dB HINT-C, which translates to a large clinical advantage. For those who show that such testing is helpful and are interested in implantation, we offer a TOI placement.

Surgery was well tolerated, with minimal recovery and low complication rate. We believe that the seroma is associated with the patient having a thick scalp that required skin thinning, with subsequent tissue weeping into the dead space of the developed pocket. This situation might be potentially prevented by tacking stitch to avoid pincushioning or by applying a pressure dressing for a longer period. Surgeons should keep this in mind in older pediatric patients and when skin thinning is needed. In addition, we encountered no issues with single-stage surgery and subsequent activation, even in younger patients.

Longer-term skin issues with TOI and the magnet site were limited to pain and redness from device overuse, which resolved at subsequent follow-up visits after giving the site breaks and counseling parents and patients on the optimal use of the device. This low rate of skin problems is noteworthy when comparing percutaneous with transcutaneous implants. Although we did not directly compare TOI results with percutaneous implant outcomes, Chan et al18 recently published a study of 45 pediatric patients who underwent placement of percutaneous osseointegrated implants, which found a high complication rate of 58 complications in 29 patients. The most common of these complications stemmed from skin infection or overgrowth, with 17 of 45 patients (38%) requiring revision surgery for skin overgrowth. Our study was of a similar sample size, but no skin infections were noted among our participants. In addition, using the transcutaneous model negates skin overgrowth as a complication. Despite the relatively high complication rate in the study by Chan et al,18 their survey data revealed that parents still found the percutaneous osseointegrated implant device satisfactory. Considering both our TOI results and those of a similarly structured study using percutaneous osseointegrated implants, we strongly recommend the use of TOIs in this population because these devices allow for excellent hearing results without the higher risk of surgical site infections or the need for surgical site revision, both of which can be costly and emotionally taxing.

Skin breakdown has been raised as a concern for TOIs, but in our series, this was not an issue, even during the later follow-up dates of more than 6 months after surgery. Cutaneous complications associated with transcutaneous devices have been highlighted in a study by O’Niel et al.19 In their series, a skin complication rate of 37.5% is reported, nearly a third of which resulted in actual skin breakdown. As a consequence, their recommendations included a weaker magnet strength at initial fitting, a graduated wearing schedule, and increased parental education about appropriate use. Carr et al10 reported that 3 of their 10 patients experienced pain, necessitating a reduction in magnet strength, and 2 required upgrading magnet strength. None experienced skin breakdown. Baker et al9 reported no skin breakdown problems, but 1 patient experienced insufficient magnet strength. Iseri et al17 reported 1 patient with temporary erythema and 3 with pain, all of whom improved with the reduction in magnet strength, which led the investigators to conclude that opting for a lower-strength magnet initially is a preferred strategy.17

Also of importance in our case series, the posterior incision with anterior-based skin flap worked well and did not add difficulty to the case or affect healing. Existing published series either use the anterior-based incision or do not comment.1,2,8,20 However, this modification allowed for an incision that was well away from the potential auricular reconstruction site with no risk to the skin pocket or blood supply, including the superficial temporal artery or postauricular arteries. Thus far, 2 patients have undergone concurrent TOI placement with the first-stage auricular reconstruction, and 1 patient had subsequent auricular reconstruction. With this incision technique, we have not encountered any issues associated with incision placement or vascular supply to the reconstruction site. We recommend this incision when there is consideration for later or concurrent auricular reconstruction.

Limitations
The limitations of this study include its retrospective nature and the exclusive attention given to transcutaneous implantation. Accordingly, a comparison of the audiological outcomes with the outcomes of those opting for percutaneous implantation or...
Transcutaneous Osseointegrated Implants for Pediatric Patients With Atresia

Conclusions

Transcutaneous implants present a valid alternative to percutaneous implants or atresioplasty for hearing habilitation. For pediatric patients with aural atresia or microtia, surgical and audiological outcomes are positive. This procedure should be a part of the options discussed for patients with aural atresia.

REFERENCES