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Safety and Effectiveness of Cochlear Implantation of Young Children, Including Those With Complicating Conditions

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Objective: Determine safety and effectiveness of cochlear implantation of children under age 37 months, including below age 12 months.
Study Design: Retrospective review.
Setting: Tertiary care children's medical center.
Patients: 219 children implanted before age 37 mos; 39 implanted below age 12 mos and 180 ages 12–36 mos. Mean age CI = 20.9 mos overall; 9.4 mos (5.9–11.8) and 23.4 mos (12.1–36.8) for the two age groups, respectively. All but two ≤12 mos (94.9%) received bilateral implants as did 70.5% of older group. Mean follow-up = 5.8 yrs; age last follow-up = 7.5 yrs, with no difference between groups.
Interventions: Cochlear implantation.
Main outcome measures: Surgical and anesthesia complications, measurable open-set speech discrimination, primary communication mode(s).
Results: Few surgical complications occurred, with no difference by age group. No major anesthetic morbidity occurred, with no critical events requiring intervention in the younger group while 4 older children experienced desaturations or bradycardia/hypotension. Children implanted under 12 mos developed open-set earlier (3.3 yrs vs 4.3 yrs, $p \leq 0.001$) and were more likely to develop oral-only communication (88.2% vs 48.8%, $p \leq 0.001$). A significant decline in rate of oral-only communication was present if implanted over 24 months, especially when comparing children with and without additional conditions associated with language delay (8.3% and 35%, respectively).
Conclusions: Implantation of children under 37 months of age can be done safely, including those below age 12 mos. Implantation below 12 mos is positively associated with earlier open-set ability and oral-only communication. Children implanted after age 24 months were much less likely to use oral communication exclusively, especially those with complex medical history or additional conditions associated with language delay. **Key Words:** American Society of Anesthesiology physical status—Children with additional conditions associated with language delay—Cochlear implant—Communication mode—Infants—Open-set speech perception—Oral communication—Pediatric anesthetic safety—Spoken language—Surgical and anesthetic complications.

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A recent long-term study of spoken language of implanted Australian children by Dettman et al. (1) demonstrated significant advantage of younger age at implantation, especially before age 12 months. The benefit of access to spoken language during infancy is not surprising in light of research on normal hearing infants demonstrating a relationship between perception at 6 months and language at 2 years (2). Despite the potential advantage of implantation during infancy, it is not common practice in the United States for reasons that include concerns regarding safety. Another group of patients who may not be receiving the benefits of early implantation are those with complex medical problems or conditions associated with language delay independent of hearing loss. Knowledge of the impact of early cochlear implantation (CI) on this population is not well understood in part due to the practice of excluding these children from studies of language outcome. Examples

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include the Dettman study which excluded children with even mild additional disability, as well as many other studies of early implanted children (1,3–8).

In addition to effectiveness, safety of an elective surgical procedure must be considered, especially for children whose age or medical conditions place them at increased anesthetic risk. From a pediatric anesthesia perspective, the risk of morbidity and mortality during elective CI surgery is highest for infants with severe comorbid conditions (9). These children have a higher American Society of Anesthesiology (ASA) Physical Status Classification (Table 1). However, the literature on safety of CI surgery in infants and children rarely has included ASA status or the occurrence of critical events requiring immediate intervention to avoid anesthetic complications.

This study explored the hypothesis that younger age of CI would be advantageous for all children, including those at risk for language delay for reasons other than hearing loss, and that implantation during infancy would provide further advantage. We also hypothesized that CI surgery may be done safely in infants and children, including those with elevated ASA status, using modern anesthetic techniques.

MATERIALS AND METHODS

Retrospective review of all children implanted at our institution in at least one ear under 37 months of age between 2007, when electronic medical records became available for ambulatory visits, and early 2015. Institutional Review Board approval was obtained (IRB protocol # 2017–722). Data included past medical history (e.g., neonatal intensive unit [NICU] stay, prematurity, elevated bilirubin, etiology of sensorineural hearing loss [SNHL], family history of SNHL and CI, cerebral palsy, autism spectrum disorder [ASD]); surgical information (use of the periosteal pocket technique to secure the receiver stimulator, degree of electrode insertion [determined by operative report], procedure duration and surgical complications within the first 30 days of surgery); intraoperative and post-anesthesia care unit (PACU) records for children implanted as of 2009 when this information became available in the electronic record (use of a cervical block, ASA status [Table 1], major anesthetic morbidity and mortality, events requiring additional management due to a difficult airway, desaturations, hypotension or bradycardia as well as more minor anesthetic issues such as nausea, vomiting, and pain requiring intervention in the PACU; and hospital stay following surgery); presurgical imaging of the cochlea and eighth nerve reviewed by an attending radiologist; and preoperative residual hearing (speech awareness threshold and Infant-Toddler Meaningful Auditory Integration Scale/Meaningful Auditory Integration Scale scores). Whether the child achieved any measurable open-set speech perception, as measured by a developmentally appropriate assessment such as the Multisyllabic Lexical Neighborhood Test, Lexical Neighborhood Test, Phonetically Balanced Kindergarten (PBK) word test, Pediatric AzBio Sentence, or Hearing in Noise Test for Children test and primary modes of receptive and expressive communication were routinely recorded in the chart by the audiologist at each evaluation; results from the last available follow up assessment served as the outcome measures. Actual open-set test scores, though recorded in the chart, were not used for this study because many children were tested with different measures or with monitored live voice rather than recorded test materials. The use of different measures and their administration was due to chronologic and developmental age and language level at time of last evaluation, as well as the addition of new test measures into the clinic protocol. Therefore, comparison of test scores between groups could not be validly performed. The present study is intended to evaluate outcomes related to age without the confounder of a degraded stimulus input. Therefore, children at significant risk to receive inadequate electrical stimulation due to less than full electrode insertion or cochlear nerve deficiency (CND) were excluded from analysis of performance outcome measures if they did not have a second ear with full insertion and normal nerve, for a total of 14 children excluded from performance analyses. Of those excluded, 11 had CND alone, two had CND and partial electrode insertion due to severe cochlear hypoplasia, and one had partial electrode insertion due to severe cochlear ossification. However, these children are included in all medical and surgical data analyses.

Children with syndromes and complex medical histories associated with language delays and at risk for cognitive deficits were not excluded. Children with preterm birth at less than 28 weeks (extreme prematurity), cerebral palsy, syndromes such as CHARGE, Goldenhar, Down syndrome or diagnosis of ASD, congenital cytomegalovirus, post-meningitic deafness, or hyperibilirubinemia requiring exchange transfusion were considered complex as such children are known to be at risk to have significant global developmental delays and poorer communication outcomes (10).

TABLE 1. *ASA physical status classification system*

ASA PS Classification	Definition of Patient Status
ASA I	Normal health
ASA II	Mild systemic disease
ASA III	Severe systemic disease
ASA IV	Severe systemic disease that is constant threat to life
ASA V	Moribund, not expected to survive without operation
ASA VI	Declared brain-dead

ASA indicates American Society of Anesthesiology.

Subject Characteristics

Subjects included 219 children with 383 implanted ears. All were implanted in at least one ear under age 37 months. Characteristics of the subjects are shown in Table 2 for all subjects and by age group at implantation of the first ear ($\leq$12 mo and $>$12 mo). Mean age at first CI was 20.9 months (5.9–36.8). In 24.2%, both ears were implanted simultaneously and another 50.7% were implanted in both ears by the time of last follow up. In the majority (63.5%), cause of hearing loss was unknown. A non-syndromic cause was identified in 23.7% and a syndrome in 7.3%. In 10.4%, a family member already had a cochlear implant. Preterm birth had occurred in 19.1% and history of a NICU stay greater than 5 days was present in 20.8%. Overall, a complex medical history was present in 15.5% (Table 3). Almost half (49.3%) were eligible for Illinois Medicaid when implanted. Mean time to last follow up was 5.8 years (1.1–10.4) for all children, with a mean age at follow up of 7.5 years (2.7–12.8).

The 39 children first implanted under age 12 months, had a mean age at implantation of 9.4 months (5.9–11.8), while the 180 children initially implanted between age 12 and 36 months had a mean age at implantation of 23.4 months (12.1–36.8).

TABLE 2. *Characteristics of 219 children implanted under the age of 37 months, for all subjects and by age at first CI group*

Characteristic	All[a] (n = 219)	First CI ≤12 months (n = 39)	First CI >12 months (n = 180)
Age first CI (mo): mean (SD, min–max)	20.9 (8.5, 5.9–36.8)	9.4 (1.6, 5.9–11.8)	23.4 (7.2, 12.1–36.8)
Sequence of implantation (n [%])[b]:			
Bilateral simultaneous	53 (24.2)	20 (51.3)	33 (18.3)
Bilateral sequential	111 (50.7)	17 (43.6)	94 (52.2)
Unilateral	55 (25.1)	2 (5.1)	53 (29.4)
Sex (%): M/F	53.4/46.6	59.0/41.0	53.4/46.6
Cause hearing loss (%):			
Unknown	63.5	46.2	63.5
Nonsyndromic	23.7	35.9	23.7
Syndrome	7.3	10.3	7.3
Bacterial meningitis	3.7	5.1	3.7
CMV	1.8	2.6	1.8
Family history hearing loss (%)	13.7	20.5	12.1
Family history CI (%)	10.4	15.4	9.2
Pre-term birth <37 weeks (%)	19.1	15.4	19.9
32 to <37 weeks	13.5	15.4	13.1
28 to <32 weeks	2.3	0.0	2.8
<28 weeks	2.8	0.0	3.4
Unknown degree	0.5	0.0	0.6
NICU stay (%): 0 to ≤5 days/>5 days	79.2/20.8	84.2/15.8	79.2/20.8
Complex medical history (%)	15.5	10.3	16.7
Illinois Medicaid eligible at time of first CI (%)[c]	49.3	28.2	53.9
Pre-op SAT for first CI (dB): mean (SD, min–max)	83.8 (20.2, 20-NR)	88.6 (14.7, 65-NR)	82.9 (21.0, 20-NR)
Pre-op MAIS for first CI (correct)[d]: Mean (SD, min–max)[d]	8.4 (7.6, 0–34)	4.8 (5.7, 0–26)	9.2 (7.8, 0–34)
Last follow-up (yr): mean (SD, min–max)	5.8 (2.3, 1.1–10.4)	6.0 (2.4, 2.1–10.4)	5.7 (2.3, 1.1–10.2)
Age at last follow-up (yr): mean (SD, min–max)	7.5 (2.4, 2.7–12.8)	6.7 (2.4, 2.7–11.0)	7.7 (2.4, 2.7–12.8)

[a]For some variables, particularly birth information, data were not available on all subjects so actual n may be slightly fewer.
[b]Statistically significant, $\chi^2 = 22.1$, $p \leq 0.001$: CI ≤12 months greater percentage bilateral simultaneous, fewer unilateral than CI >12 mo.
[c]Statistically significant, Fisher's exact = 0.005: CI >12 months more likely to be eligible for Illinois Medicaid than younger group.
[d]Statistically significant, Mann–Whitney *U* test, $p \leq 0.001$: CI ≤12 months lower mean score on MAIS.
CI indicates cochlear implant; CMV, cytomegalovirus; SD, standard deviation; SAT, speech awareness threshold; NR, no response.

There were few statistically significant differences between the two age groups in their preimplant characteristics. Although the younger group was a little more likely to have a family history of hearing loss or a CI in the family, these differences did not achieve statistical significance. Funding for implantation did differ between the groups, with most children in the younger group funded by commercial insurance as fewer were eligible for Medicaid than in the older group (28.2% versus 53.9%, $p = 0.005$). Time to last follow up and age at last follow up were not statistically different by age group. The older group did have a higher mean preoperative MAIS score, something that might be expected just based on age, but mean speech awareness threshold did not differ between groups.

TABLE 3. *Complex medical conditions (n [% of cases])*

Medical Condition	All (n = 219)	First CI ≤12 months (n = 39)	First CI >12 months (n = 180)
CMV	4 (1.8)	1 (2.6)	3 (1.7)
Bacterial meningitis	8 (3.7)	2 (5.1)	6 (3.3)
Down syndrome	1 (0.5)	0	1 (0.6)
CHARGE	5 (2.3)	0	5 (2.8)
Goldenhar	1 (0.5)	1 (2.6)	0
Tracheotomy	1 (0.5)	0	1 (0.6)
Preterm <28 weeks	41 (19.1)	6 (15.4)	35 (19.9)
Autism spectrum disorder	12 (5.5)	0	12 (6.7)
Cerebral palsy	6 (2.7)	0	6 (3.3)

CHARGE indicates coloboma, heart defect, atresia choanae, restricted growth and development, genital abnormality, and ear abnormality; CI, cochlear implant; CMV, cytomegalovirus.

The greatest difference between age groups is that significantly more patients implanted at less than or equal to 12 months underwent bilateral simultaneous implantation (51.3% versus 18.3%). A similar percentage of children in both age groups received sequential implantation of a second ear, but only 5.1% of the younger group received just one implant compared with 29.4% of the older group.

The majority of implanted ears had normal preoperative cochlear anatomy (Table 4). The most common abnormal finding on magnetic resonance imaging was enlarged vestibular aqueduct (4.8% of 376 ears).

Data Analysis

Data from a variety of sources (audiology, medical/surgical, radiology, anesthesiology) were entered into Excel files with

TABLE 4. *Preoperative imaging findings for all implanted ears*

Image	CT (n = 244)	MRI (n = 376)
Cochlea (%):		
Normal	84.0	91.8
Hypoplastic, full basal	6.6	2.4
Hypoplastic, <full basal	1.2	0.8
Common cavity	0.4	0.3
Dysplastic, full basal	4.9	1.9
Labyrinthitis ossificans	0.8	2.4
Other	2.0	3.8
Enlarged vestibular aqueduct (%)	4.1	4.8
Vestibule/Semicircular canal (%):		
Normal	88.5	88.2
Dysplastic	10.2	8.0
Ossified/fibrotic	0.8	3.7
Other	0.4	0.0
Cochlear nerve deficiency (%)	–	4.8 (n = 18)

CT indicates computer tomography; MRI, magnetic resonance imaging.

unique subject identifiers and later combined into a common statistical file. Data related to medical history and outcome measures were analyzed by subject while surgical complications and radiology findings were analyzed by ears implanted. Anesthesia outcomes were analyzed by the number of anesthesia CI episodes, with anesthesia variables only counted once for simultaneous implantation of both ears. Descriptive and inferential statistical analyses were performed using SPSS/PC (SPSS Inc., now IBM SPSS Statistics, Armonk, NY). Comparisons between categorical variables used χ^2 or Fisher's exact test. Because of unequal sample sizes and lack of homogeneity of variance, comparisons of interval-level data were performed using the nonparametric Mann–Whitney *U* test. Criterion for statistical significance was set at $p \leq 0.05$, two-tailed.

RESULTS

Distribution of age at first and second CI is presented in Table 5. Twenty children in the younger group had bilateral simultaneous implantations and an additional two received their second sequential implant below age 12 months, accounting for the 22 second ears implanted under 12 months of age. About 38% of second ears were implanted at greater than 3 years of age.

TABLE 5. *Distribution of age at first and second cochlear implant (n [%])*

Age range	First CI (n = 219)	Second CI[a] (n = 164)
≤12 months	39 (17.8)	22 (13.4)
12–18 months	49 (22.4)	19 (11.6)
18–24 months	52 (23.7)	20 (12.2)
24–30 months	41 (18.7)	18 (11.0)
30–37 months	37 (17.4)	22 (13.4)
>3–5 years	–	40 (24.4)
>5–7 years	–	14 (8.5)
>7 years	–	9 (5.5)

[a]Includes bilateral simultaneous implantation second ears.
CI indicates cochlear implant.

Family history of hearing loss was not related to age at implantation. However, having a family member (immediate or extended) with a CI was significantly associated with younger mean age at first implantation (15.6 mo versus 21.2 mo, $p \leq 0.008$).

Surgical and Anesthesia Characteristics

A periosteal pocket, without a recessed bed, was used in the majority regardless of age group (Table 6). There were few surgical complications, with no difference by age group. Cerebrospinal fluid (CSF) leak was the most common complication (1.8%), with four of seven occurrences in children under age 12 months, including one who underwent repair of a mastoid tegmen dural tear from below. The remaining seven ears had intra-cochlear CSF. Of these seven, two had hypoplasia (basal turn only, hypo/dysplastic vestibular system); two had severe hypoplasia (limited portion of basal turn, hypo/dysplastic vestibular system); one had severe hypoplasia (limited portion basal turn, absent vestibular system); two, in the same child, had normal findings. No CSF leaks reoccurred after sealing at time of implantation. One wound infection in the older group was managed by explantation with interval re-implantation. No facial nerve or other infectious complications occurred.

ASA status class in children who underwent anesthesia for single ear implantation was similar in the younger and older groups. For children undergoing bilateral simultaneous CI, fewer children in the younger group had ASA status II or III ($p \leq 0.012$). Most in both age groups received a cervical block (81.5 and 85.6%) to aid in pain control and reduce nausea and vomiting by minimizing narcotic use. No major anesthetic morbidity (cardiac arrest, neurologic injury, or death) occurred. None of the children had difficult airways requiring more than three attempts at intubation or advanced airway management techniques, and no episodes of laryngospasm requiring neuromuscular blockade or reintubation occurred. In the younger group, there were no episodes of desaturation, bradycardia/hypotension or airway events requiring intervention. In the older group, four children undergoing unilateral CI (two ASA II and two ASA III), had anesthesia-related events requiring intervention. There was no relationship between duration of the surgical procedure and occurrence of anesthetic events.

More than 90% of patients undergoing unilateral and 85% undergoing bilateral simultaneous implantations were discharged on the day of surgery, with no significant difference by age group. Duration of surgery was significantly longer in those kept for overnight observation than those discharged the same day (179.4 min versus 130.2 min, $p \leq 0.007$). Results were similar if excluding simultaneous bilateral surgeries (141.1 min versus 112.4 min, $p \leq 0.006$). For single ear surgeries, children in ASA III were less likely to be discharged on the day of surgery than those with ASA I or II status (62.5% versus 95.2% and 89.7%, respectively, $p \leq 0.001$)

TABLE 6. *Surgical and anesthesia characteristics for all implanted ears, by age at surgery*

Surgery	All (n = 383)	CI ≤12 months (n = 61)	CI >12 months (n = 322)
Periosteal pocket technique (%)	95.0	93.4	95.3
Surgical complications (%)			
None	97.9	93.4	98.8
CSF leak	1.8 (n = 7)	6.6 (n = 4)	0.9 (n = 3)
Wound infection	0.3 (n = 1)	0.0	0.3 (n = 1)
Anesthesia[a]	**ALL (n = 236)**	**CI ≤12 months (n = 27)**	**CI >12 months (n = 209)**
ASA status (% status I/II/III)			
Single CI surgery[b]	40.9/52.9/6.2	43.8/50.0/6.3	40.7/53.1/6.2
Two simultaneous CIs[c]	55.6/38.9/5.6	71.4/28.6/0.0	45.5/45.5/9.1
Use of cervical block (%)	85.2	81.5	85.6
Anesthesia event (n [%])			
None	225 (95.3)	25 (92.6)	200 (95.7)
Desaturation	2 (0.8)	0 (00)	2 (1.0)
Bradycardia/hypotension	2 (0.8)	0 (00)	2 (1.0)
Difficult airway	0 (0.0)	0 (0.0)	0 (0.0)
Vomiting	1 (0.4)	0 (0.0)	1 (0.4)
Pain control	2 (0.8)	1 (3.7)	1 (0.5)
Postop nausea	4 (1.7)	1 (3.7)	3 (1.4)
Hospital stay (%):			
Single CI surgery[b]			
Discharged	90.8	84.2	91.3
Overnight	7.0	5.3	7.1
Admitted	2.2	10.5	1.6
Bilateral simultaneous			
Discharged	85.5	90.5	82.4
Overnight	14.5	9.5	17.6
Admitted	0.0	0.0	0.0

[a]Simultaneous bilateral implants are only counted once.
[b]Initial or sequential implantation of one ear at a time.
[c]Of simultaneous implantations, younger group has higher rate of ASA 1, $\chi^2 = 8.8$, $p \leq 0.012$.
ASA indicates American Society of Anesthesiology; CI, cochlear implant.

while for simultaneous bilateral surgeries, both ASA II and III were less likely to be discharged than ASA I (76.2 and 66.7%, respectively, versus 93.3%, $p \leq 0.024$) (Table 7). Of the 34 patients not discharged on the day of surgery, 29 were observed overnight in the PACU and five were admitted to the hospital and discharged within 48 hours (Table 8). Two of the five admissions to the pediatric intensive care unit (PICU) were children of ASA status III. One PICU admission was planned because of a sleep study documenting apnea and the other was arranged to monitor the child after desaturations occurred immediately post-extubation. These two children were discharged the following day. Three children who underwent sealing of CSF leak were admitted to the floor for additional observation. Eight children who underwent bilateral simultaneous CI (two ASA I and six ASA II) were observed overnight. Four of these children had a history of recent meningitis with fibrosis/ossification of the cochleae.

TABLE 7. *Hospital stay by ASA status for one ear and bilateral simultaneous procedures (%)*

Hospital Stay	ASA I	ASA II	ASA III
Procedure - one ear[a]	(n = 105)	(n = 136)	(n = 16)
Discharged	95.2	89.7	62.5
Overnight observation	2.9	9.6	18.8
Admitted	1.9	0.7	18.8
Procedure – bilateral[b]	(n = 30)	(n = 21)	(n = 3)
Discharged	93.3	76.2	66.7
Overnight observation	6.7	23.8	33.3
Admitted	0	0	0

[a]Statistically significant, $\chi^2 = 17.1$, $p \leq 0.001$, ASA III lower rate of discharged than ASA I and II, with higher rates of overnight and admitted.
[b]Statistically significant, $\chi^2 = 7.5$, $p \leq 0.024$, ASA II and III lower rates of discharged and higher rates of overnight observation than ASA I.
ASA indicates American Society of Anesthesiology.

Open-Set and Communication Mode at Most Recent Follow Up

The ability to score on a developmentally appropriate open-set speech perception measure and communication mode at last follow-up were evaluated in 204 children, 34 first implanted below 12 months who had a minimum follow up of 2 years and 170 first implanted between 12 and 37 months of age (Table 9). The majority of both age groups were able to show at least some degree of

TABLE 8. *Characteristics of children requiring overnight observation or admission after cochlear implant procedure*

Admit or Overnight Observation	Age (mo)	No. of Ear (s) Implanted at Procedure	ASA status	Surgical Cx	Syndrome	Cochlear Malformation (Y/N) or Ossification
Admission	8	One	1	CSF leak[c]	–	N
Admission	10	One	2	CSF leak	Goldenhar	Y
PICU admission[a]	21	One	3	–	CHARGE	Y
Admission	28	One	1	CSF leak	–	Y
PICU admission[b]	36	One	3	–	–	N
Overnight observation	7	Bilateral	2	–	–	N
‘‘	7	Bilateral	1	–	–	N
‘‘	10	One	2	CSF leak	Goldenhar	Y
‘‘	10	One	3	CSF leak	–	Y
‘‘	12	One	2	–	Pendred	Y
‘‘	12	Bilateral	2	–	=	Ossification
‘‘	12	One	2	–	–	Y
‘‘	15	Bilateral	2	–	–	Ossification
‘‘	16	One	1	–	–	Y
‘‘	16	Bilateral	2	–	–	Ossification
‘‘	17	Bilateral	2	–	–	N
‘‘	18	Bilateral	1	–	–	Ossification
‘‘	19	One	2	–	–	N
‘‘	21	One	2	–	–	N
‘‘	22	One	2	–	–	Y
‘‘	23	One	2	–	–	N
‘‘	25	One	2	–	–	N
‘‘	25	One	3	CSF leak	CHARGE	Y
‘‘	27	One	2	–	–	N
‘‘	27	One	3	–	–	N
‘‘	28	Bilateral	3	–	–	N
‘‘	29	One	1	CSF leak	–	N
‘‘	29	One	2	–	–	Y
‘‘	32	One	2	–	Pendred	Y
‘‘	33	One	3	–	–	N
‘‘	40	One	1	–	–	Y
‘‘	74	One	2	–	Pendred	Y
‘‘	92	One	2	–	–	N
‘‘	122	One	2	–	Pendred	Y

[a]Planned PICU admission due to apnea recorded on presurgical sleep study.
[b]Transferred to PICU for observation after post-extubation bradycardia/hypotension.
[c]CSF leak secondary to tegmen dural tear.
CHARGE indicates coloboma, heart defect, atresia choanae, restricted growth and development, genital abnormality, and ear abnormality.

TABLE 9. *Open-set speech discrimination and mode of communication outcomes at last follow-up by age group at first implantation*

Outcome	First CI ≤12 months (n = 34)	First CI >12 months (n = 170)	Statistical Significance
Attained open-set (%)	94.1	82.7	NS
Age at open-set (yr): mean (SD)	3.3 (0.6)	4.3 (1.5)	$p \leq 0.001$
Receptive communication (%)			$p \leq 0.001$[a]
Oral	88.2	52.4	
Oral and sign	11.8	43.5	
Sign	0.0	1.2	
Other	0.0	2.9	
Expressive communication (%)			$p \leq 0.001$[a]
Oral	94.1	55.9	
Oral and sign	5.9	34.7	
Sign	0.0	2.4	
Other	0.0	7.1	
Oral communication exclusively	88.2	48.8	$p \leq 0.001$

[a]Comparison of oral versus all others grouped.
NS = not significant.

TABLE 10. *Factors affecting attainment of open-set speech discrimination and oral communication*

	Open-set		
Factor	Yes	No	Statistical Significance
Age first CI (mo): mean (SD)	20.0 (8.2)	24.6 (8.7)	$p \leq 0.009$
Age second CI (mo): mean (SD)	36.2 (24.3)	38.7 (20.5)	NS
Last follow-up (yr): mean (SD)	6.1 (2.3)	4.5 (2.3)	$p \leq 0.001$
Age last follow-up (yr): mean (SD)	7.8 (2.3)	6.5 (2.7)	$p \leq 0.007$
Complex medical history (%):	9.6	20.0	NS
	Oral-Only Communication		
	Yes	No	
Age First CI (mo): mean (SD)	17.5 (7.7)	25.2 (7.4)	$p \leq 0.001$
Age second CI (mo): mean (SD)	31.8 (24.6)	43.1 (20.6)	$p \leq 0.001$
Last follow-up (yr): mean (SD)	5.9 (2.4)	5.7 (2.3)	NS
Age last follow-up (yr): mean (SD)	7.3 (2.3)	7.8 (2.5)	NS
Complex medical history (%):	8.8	20.9	$p \leq 0.016$

NS = not significant.

measurable open-set ability, including 94.1% of the younger group and 82.7% of the older group. Although the proportion of the two age groups achieving open-set was not statistically different, open-set was achieved at a younger mean age in the group implanted younger (3.3 yr versus 4.3 yr, $p \leq 0.001$). All children who attained open-set in the first ear implanted also attained open-set in the second ear implanted. As noted earlier, because of the wide variety of open-set measures and the need to use monitored live voice as well as recorded test material due to the differences in childrens' chronologic and developmental age and language ability, valid comparison of actual test scores between groups was not possible. Therefore, these findings indicate only progress to open-set and not the level of open-set speech perception ability.

Table 9 also shows the percentage of children in each age group at first CI using the different forms of communication at last follow-up. The percentage of those using oral-only communication was significantly higher in the younger age group for receptive communication, expressive communication, and combined for exclusively oral communication. The differences were quite large, with 88.2% of the children implanted below 12 months using exclusively oral communication compared with just 48.8% of those implanted between 12 and 37 months ($p \leq 0.001$).

Table 10 shows that those who developed open-set had a younger mean age at first implantation compared with those who did not (20.0 mo versus 24.6 mo, $p \leq 0.009$), as well as longer follow-up time and older age at last follow up. Preoperative hearing did not differ significantly between those who achieved measurable open-set ability and those who did not. Although those who did not achieve open-set had a higher rate of complex medical history, this difference did not achieve statistical significance, and 72.7% of complex cases did obtain open-set speech perception. Mean ages at both first and second CIs, were significantly younger in those using oral-only communication, and preoperative hearing was also slightly better for this group. Those who did not use oral-only communication had a higher rate of complex medical history than those who do use oral-only communication (20.9% versus 8.8%, $p \leq 0.016$), but 34.5% of complex cases did achieve oral-only communication. Table 11 summarizes the effect of age at implantation across all children and by presence of complex medical history for both open-set discrimination and oral communication. Those implanted after age 24 months show a considerable decline in the attainment of oral-only

TABLE 11. *Rate of positive outcomes by different age at first implantation groupings and presence of complex medical history (%)*

Outcome	≤12 months	12–24 months	>24 months	Statistical Significance
Open-set	94.1	84.2	80.6	NS
Non-complex	93.5	84.5	84.7	NS
Complex	100.0	81.8	50.0	–[a]
Oral communication exclusively	88.2	62.2	30.6	$p \leq 0.001$
Non-complex	90.3	64.3	35.0	$p \leq 0.001$
Complex	66.7	50.0	8.3	–[a]
Oral receptive	88.2	64.3	36.1	$p \leq 0.001$
Oral expressive	94.1	68.4	38.9	$p \leq 0.001$

[a]Cell sizes too small for statistical validity.
NS = not significant.

communication compared with those implanted younger. The age at implantation effect is especially pronounced in medically complex children, both for attainment of open-set speech perception and oral-only communication. There was no significant difference in the percent of children categorized as complex across the three age groups. However, the rate of oral-only communication in complex kids was 66.7% for those implanted by 12 months of age, decreasing to 50.0% for those first implanted from 12 to 24 months and to only 8.3% for those implanted after 24 months. Due to the small number of medically complex cases who were tested when divided into three age groups, statistical tests were not valid. However, two-thirds of complex children were able to achieve oral-only communication if implanted at or below age 12 months. There was no significant difference in mean age at first CI between complex and non-complex children (22.2 mo and 20.7 mo, respectively), although age at CI 2 did differ, with complex children implanted younger (21.5 mo versus 38.1 mo, $p \leq 0.004$) as they more often had simultaneous bilateral implantation.

DISCUSSION

The ability to hear spoken language early in life is critical to development of spoken language. A number of studies have provided evidence that implantation of children younger than age 12 months may improve spoken language (1,4,8,11–13). The largest series of implanted infants was published in 2016 by Dettman et al. (1). They reported the long-term outcomes of congenitally deaf children including 151 implanted by age 12 months and younger. The 12 months and younger group had significantly better language and speech production outcomes than those implanted at 13 to 18 months and 19 to 24 months, when evaluated at school entry and late primary school.

Unlike many studies, this series does not exclude children with additional disabilities and conditions associated with language delay. These children are included because they are a growing proportion of CI candidates (10). Taking advantage of neuroplasticity by early implantation and providing effective auditory habilitation may be more important for maximizing the outcomes of this population than those having only an auditory deficit. Our results demonstrate that a significant number of complex children achieve measurable open-set skills and oral communication as their primary mode of communication, especially when implanted at a younger age.

The most striking finding in our series is the relationship between oral-only communication mode and younger age of first CI. The decline in rate of oral-only communication was particularly striking for all children implanted over age 24 months. Because communication mode in this study was determined at last follow up when the average age was 7.5 years, this reflects communication mode as many of the children entered grade school. In our locale, the vast majority of children who use sign in addition to oral communication are placed in total communication classrooms. Placement in total communication rather than oral-only or mainstream classrooms for most children requiring sign is, in our experience, influenced by these children often having lower oral language ability.

Although children implanted in the younger group (<12 mo) and the older group (12–36 mo) had similar preoperative hearing thresholds, the cause of hearing loss was variable and often unknown. It is likely that some children had hearing at birth and, therefore, more access to sound before CI. Earlier residual hearing would more likely advantage those in the older group. However, the children implanted below 12 months developed auditory skills more rapidly and were more likely to develop oral-only communication.

Comparisons using level of speech perception ability were not possible in this study due to the young ages and range of developmental status which required clinical use of different test measures and procedures. This is a significant limitation of the study as achieving a low but greater than zero level on a test can represent a considerable difference in ability from achieving a score near the high end of a test. In addition, only information regarding communication mode(s) rather than comprehensive information about receptive and expressive language at last follow up was available. We also note that there may be unknown differences such as parenting and socioeconomic status between children who receive an implant under age 12 months and those who do not, that might influence performance outcome. For example, the finding of a lower rate of Medicaid funding in the younger age group suggests the possibility that those who received CIs at this younger age came from families likely to have more financial resources and higher education level which may have benefited the child's learning and development. These are all areas for future study.

Regarding outcomes of the complex children, it was not possible to grade the relative severity of complicating conditions. Therefore, complex children in the older group may have been more severely affected and for reasons beyond age of first CI, less likely to develop open-set ability and oral-only communication outcomes in comparison to those in the younger group.

Growing evidence of the advantages of implantation below age 12 months must be balanced against potential increased anesthetic and surgical risk (14). Previous series describing surgical complications in children under 12 months reported complication rates similar to those found in older children and adults (13,15–18). An additional publication focused solely on soft tissue complications in 94 ears of 66 children implanted below 12 months reported only one minor wound infection that was successfully treated without surgery (19). Our series is similar, with a low rate of major surgical complications even in the children under 12 months of age. Most were related to intra-cochlear CSF and were anticipated.

Yeh et al. (20) previously published the most comprehensive study of anesthesia outcomes in 123 children implanted under age 18 years, of which 12 were below age 12 months. Few events requiring intervention occurred

and all but one were respiratory. Other authors have concluded anesthesia risk of implanting infants and young children is acceptable (13,15–18). However, ASA status or occurrence of respiratory critical events was often not reported (15–18).

From a pediatric anesthesia perspective, risk of morbidity and mortality is highest for infants with severe comorbid conditions and for children undergoing emergent surgery (9). Study of major morbidity requires thousands of patients because these complications are exceedingly rare. One of the first reliable studies of cardiac arrest in the perioperative setting was published in 2000 and evaluated 289 cardiac arrests in over one million episodes of pediatric anesthesia (21). More than half occurred in infants of who two-thirds had significant comorbidities placing them in ASA status III–V (Table 1). Halothane played a causal role in two-thirds of cardiac arrests. Cardiovascular events caused the majority of anesthesia-related arrests in patients without underlying heart disease and were often related to blood loss and/or inappropriate fluid therapy. The next most common cause of anesthesia-related arrests was respiratory arrests, most commonly caused by laryngospasm and problems managing a difficult airway. By 2000 in the United States, Halothane was replaced by safer agents. Modern monitoring equipment and new approaches to proactively minimize risk have dramatically improved safety. The unique anesthetic needs of children, especially those who are very young and/or have comorbid conditions, has become widely recognized and resulted in pediatric anesthesia becoming a board certified subspecialty in 2013. For these reasons, major morbidity for children, including those less than 12 months, has declined.

Today, for children less than 36 months of age undergoing CI surgery, anesthetic management and risk does not differ from other elective procedures. Although the incidence of cardiovascular and respiratory critical events is significantly lower compared with previous decades, infants under 12 months, especially those with underlying systemic comorbidities or difficult airways, remain at heighten risk (22,23). These risks, however, are minimal in the otherwise healthy ASA I or II child undergoing CI.

Studies have demonstrated more favorable outcomes of elective surgery in children at highest anesthetic risk, namely those with congenital heart disease, craniofacial anomalies and syndromes such as CHARGE (commonly ASA status II or III), who are managed by a specialized pediatric anesthesiologist in a well-resourced setting, rather than the occasional pediatric anesthesiologist or trainee (21,22).

In our series, 236 episodes of anesthesia for CI surgery were reviewed for laryngospasm, desaturation, bradycardia and hypotension requiring intervention, in addition to major morbidity. There were no major morbidities despite implantation of children with complex medical histories and comorbid conditions, an expected finding given the rarity of these types of complications. However, lesser anesthetic events, such as laryngospasm or cardiovascular events requiring management were also uncommon in our series. The paucity of even minor anesthetic events in our series may be due to the nature of our practice within a tertiary care children's hospital staffed by board certified pediatric anesthesiologists.

The two PICU admissions were for monitoring due to concerns about increased risk for complications. The vast majority of children were discharged on the day of surgery, including most that underwent bilateral simultaneous implantations under 12 months of age. Most of the 28 children who were observed overnight in the hospital (Table 8) were ASA status II or above, while only five were ASA status I. A difference in anesthetic-related problems, including pain control and nausea and vomiting, did not account for overnight versus same day discharge. Longer surgery duration, which may be associated with more complex surgery, was related to overnight observation. Although reasons for overnight stay may reflect parental anxiety and geographic distance from the hospital, overnight stay of many of these children may have been influenced by higher ASA status, and longer surgery, especially for children with longer bilateral procedures such as those with ossified cochleae.

Despite decades of research demonstrating the benefits of early implantation, many children are not implanted below age 2 years. There are many barriers to implantation, often based on socioeconomic status, including lack of referral for evaluation and insurance benefits, especially for children under age 12 months. Commercial insurance and Medicaid plans often deny coverage based upon outdated Food and Drug Administration audiological and age guidelines created when commercial approval for use in children 12 months and older was granted almost two decades ago. Despite significant evidence of benefit, CI before age 2 years is not a goal of federal early intervention legislation nor does the Joint Committee on Infant Hearing, an influential multidisciplinary group, promote early implantation (24).

CONCLUSION

There is a significant body of literature indicating that young age of CI is advantageous. Despite this knowledge, implantation of infants remains relatively uncommon in the US. Our study adds to the growing literature that implantation below age 12 months results in more rapid auditory skill development and exclusively oral communication. In addition, the outcomes of this study provide evidence that CI is also safe and effective for infants and children with complex medical problems, including conditions known to cause language delay. It is likely that significant improvement in language would result if changes in public policy reduced barriers to early implantation for all children.

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REFERENCES

1. Dettman SJ, Dowell RC, Choo D, et al. Long-term communication outcomes for children receiving cochlear implants younger than 12 months: a multicenter study. *Otol Neurotol* 2016;37:e82–95.

2. Tsao F, Liu H, Kuhl PK. Speech perception in infancy predicts language development in the second year of life: a longitudinal study. *Child Dev* 2004;75:1067–84.
3. Fagan MK, Pisoni DB. Hearing experience and receptive vocabularly development in deaf children with cochlear implants. *J Deaf Stud Deaf Educ* 2010;15:149–61.
4. Nicholas JG, Geers AE. Will they catch up? The role of age at cochlear implantation in spoken language development of children with severe to profound hearing loss. *J Speech Lang Hear Res* 2007;50:1048–62.
5. Niparko JK, Tobey EA, Than DJ, et al. Spoken language development in children following cochlear implantation. *JAMA* 2010;303: 1498–506.
6. Richter B, Eissele S, Laszig R, et al. Receptive and expressive language skills of 106 children with a minimum of 2 years' experience in hearing with a cochlear implant. *Int J Pediatr Otorhinolaryngol* 2002;64:111–25.
7. Tobey EA, Thal D, Niparko JK, et al. Influence of implantation age on school-age language performance in pediatric cochlear implant users. *Int J Audiol* 2013;52:219–29.
8. Svirsky M, Teoh S, Neuburger H. Development of language and speech perception in congenitally, profoundly deaf children as a function of age at cochlear implantation. *Audiol Neurootol* 2004;9:224–33.
9. Paterson N, Waterhouse P. Risk in pediatric anesthesia. *Paediatr Anaesth* 2011;21:848–57.
10. Young NM, Weil C, Tournis E. Redefining cochlear implant benefits to appropriately include children with additional disabilities. In: Young NM, Kirk KI, editors. *Pediatric Cochlear Implantation - Learning and the Brain*. New York: Springer-Verlag; 2016. pp. 213–65.
11. Colletti L, Mandala M, Zoccante L, et al. Infants versus older children fitted with cochlear implants: performance over 10 years. *Int J Pediatr Otorhinolaryngol* 2011;75:504–9.
12. Leigh J, Dettman S, Dowell R, et al. Communication development in children who receive a cochlear implant by 12 months of age. *Otol Neurotol* 2013;34:443–50.
13. Holman MA, Carlson ML, Driscoll CLW. Cochlear implantation in children 12 months of age and younger. *Otol Neurotol* 2013;34:251–8.
14. Young NM. Infant cochlear implantation and anesthetic risk. *Ann Otol Rhinol Laryngol Suppl* 2002;189:49–51.
15. Dettman SJ, Pinder D, Briggs RJ, et al. Communication development in children who receive the cochlear implant younger than 12 months: risks versus benefits. *Ear Hear* 2007;28:11S–8S.
16. O'Connell BP, Holcomb MA, Morrison D, et al. Safety of cochlear implantation before 12 months of age: Medical University of South Carolina and Pediatric American College of Surgeons–National Surgical Quality improvement program outcomes. *Laryngoscope* 2016;126:707–12.
17. Roland JT Jr, Cosetti M, Wang KH, et al. Cochlear implantation in the very young child: long-term safety and efficacy. *Laryngoscope* 2009;119:2205–10.
18. Colletti L, Mandala M, Colletti V. Cochlear implants in children younger than 6 months. *Otolaryngol Head Neck Surg* 2012;147: 139–46.
19. Das Purkayastha PK, Jewell S, James AL, et al. Soft tissue complications after pediatric cochlear implantation in children younger than 12 months. *Otol Neurotol* 2011;32:780–3.
20. Yeh JS, Mooney KL, Gingrich K, et al. Anesthetic complications in pediatric patients undergoing cochlear implantation. *Laryngoscope* 2011;121:2240–4.
21. Morray J, Geiduschek J, Ramamoorthy C, et al. Anesthesia related cardiac arrest in children. Initial findings of the pediatric perioperative cardiac arrest (POCA) registry. *Anesthesiology* 2000;93:6–14.
22. Habre W, Disma D, Virag K, et al. Incidence of severe critical events in paediatric anaesthesia (APRICOT): a prospective multicenter observational study in 261 hospitals in Europe. *Lancet* 2017;5:412–25.
23. Fiadjoe JE, NIshisaki A, Jaqannathan N, et al. Airway management complications in children with difficult tracheal intubation from the pediatric difficult intubation (PeDI) registry: a prospective cohort analysis. *Lancet Respir Med* 2016;4:37–48.
24. Joint Committee on Infant Hearing. Year 2007 position statement: principles and guidelines for early hearing detection and intervention programs. *Pediatrics* 2007;120:898–921.