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Electric-Acoustic Stimulation Outcomes in Children

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Objectives: This study investigates outcomes in children fit with electric-acoustic stimulation (EAS) and addresses three main questions: (1) Are outcomes with EAS superior to outcomes with conventional electric-only stimulation in children? (2) Do children with residual hearing benefit from EAS and conventional electric-only stimulation when compared with the preoperative hearing aid (HA) condition? (3) Can children with residual hearing derive benefit from EAS after several years of listening with conventional electric-only stimulation?

Design: Sixteen pediatric cochlear implant (CI) recipients between 4 and 16 years of age with an unaided low-frequency pure tone average of 75 dB HL in the implanted ear were included in two study arms. Arm 1 included new recipients, and Arm 2 included children with at least 1 year of CI experience. Using a within-subject design, participants were evaluated unilaterally with the Consonant-Nucleus-Consonant (CNC) word list in quiet and the Baby Bio at a +5 dB SNR using an EAS program and a conventional full electric (FE) program. Arm 1 participants' scores were also compared with preoperative scores.

Results: Speech perception outcomes were statistically higher with the EAS program than the FE program. For new recipients, scores were significantly higher with EAS than preoperative HA scores for both the CNC and Baby Bio in noise; however, after 6 months of device use, results in the FE condition were not significantly better than preoperative scores. Long-term FE users benefited from EAS over their FE programs based on CNC word scores.

Conclusions: Whether newly implanted or long-term CI users, children with residual hearing after CI surgery can benefit from EAS. Cochlear implantation with EAS fitting is a viable option for children with HAs who have residual hearing but have insufficient access to high-frequency sounds and poor speech perception.

Key words: cochlear implant, pediatrics, eas, electric-acoustic.

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INTRODUCTION

As cochlear implant (CI) candidacy continues to expand to recipients with more residual hearing, modified electrode arrays and surgical techniques are resulting in postoperative hearing preservation (Gantz et al. 2005; Yao et al. 2006; Gstoettner et al. 2008; Adunka et al. 2010b; Brown et al. 2010; Carlson et al. 2011; Erixon et al. 2012; Tamir et al. 2012; Havenith et al. 2013; Santa Maria et al. 2013, 2014; Adunka et al. 2014; Mahmoud et al. 2014; Skarzynski et al. 2014; Anagiotos et al. 2015; Kisser et al. 2016; Sweeney et al. 2016; Pillsbury et al. 2018). When low-frequency hearing is preserved after implantation, there is an opportunity to provide low-frequency cues through acoustic hearing and high-frequency hearing through electric stimulation within the same ear via electric-acoustic stimulation (EAS).

CI users have poor spectral resolution due to limited neural survival and limitations of electrical stimulation. This can lead to difficulty with speech understanding and hearing in noise. Low-frequency acoustic hearing affords access to better temporal and spectral cues than low-frequency electrical stimulation in bimodal listeners (Zhang et al. 2013) which could translate to EAS users. Research with adult CI recipients indicates that unilateral EAS use results in significantly improved listening skills in quiet (Gantz et al. 2005; Gstoettner et al. 2008; Adunka et al. 2010a; Adunka et al. 2013; Mahmoud et al. 2014; Pillsbury et al. 2018) and noise (Gantz et al. 2005; Gstoettner et al. 2008; Adunka et al. 2013; Sheffield et al. 2015; Pillsbury et al. 2018) over fully acoustic or electric stimulation.

Audibility is of utmost importance when it comes to the hearing needs of children. Children with normal hearing require even greater audibility of speech than adults to acquire spoken language and process conversational speech (Stelmachowicz et al. 2000). Adult CI recipients, who typically have later onset of deafness, have preoperative linguistic skills that provide a lexical and acoustic reference to derive information for speech understanding when presented with the novel electrical signal. In contrast, children with CIs are building an auditory and lexical base through the use of technology. Early implantation, which exploits neural plasticity, has resulted in outcomes for children with CIs that parallel auditory, speech, and language outcomes of their hearing peers (Geers et al. 2003; Davidson et al. 2011; Tobey et al. 2013).

Best practices for validation of acoustic output to ensure audibility and prevent overamplification are well established (Seewald et al. 2005; Bagatto et al. 2010). Because children have smaller ear canals than adults, measurements of real ear-to-coupler difference values and the use of real ear measurements are critical for an optimized fitting (Seewald et al. 1985; Ross & Seewald 1988; Seewald & Ross 1988). Children require audibility of soft, medium, and loud speech inputs to derive necessary information for the development of spoken language (Strauss & van Dijk 2008; Sininger et al. 2010; Davidson et al. 2011, 2014; Stiles et al. 2012; McCreery et al. 2017). Given the known differences between children and adults when it comes to auditory needs and the development of language, there is potential for significant error in assuming EAS fitting and outcomes in adults could be extrapolated to children. When providing fundamental frequency and often first formant information through acoustic stimulation, it is important that audibility is ensured and outcomes are validated.

As of yet, there are few peer-reviewed studies that investigate EAS in the pediatric population, and existing reports are on relatively small groups of children. Wolfe et al. (2017) report on a cohort of 7 children aged 6 to 16 who were fit with EAS speech processors and evaluated in quiet and in noise. They demonstrated statistical benefit from EAS in noise and in quiet when compared acoustic-only conditions and a trend toward benefit between EAS and electric programs that did not reach significance. The children in this study had been using electric

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stimulation devices for at least 6 months, before the EAS speech processor was fit, and were given ≥4 weeks to acclimate to the device. The researchers tested the electric condition by disabling acoustic input and relying on a truncated electric map. Quality of life measures comparing listening situations before and after EAS fit did indicate significant benefit. In a second study, Scholz et al. (2017) also report on a group of six children (nine ears) aged 6 to 10 years who had worn electric programs for an average of 5.9 years before being fit with EAS processors. Contrary to Wolfe et al., there was no difference between the EAS and electric conditions. Skarzynski et al. (2007) reported on six children using EAS, some of whom had CI-alone experience and some who did not, and demonstrated an improvement in speech perception over preoperative scores. Skarzynski et al. reported on six children using EAS, some of whom had CI-alone experience and some who did not, and demonstrated an improvement in speech perception over preoperative scores. Skarzynski and Lorens (2010) described benefit of EAS for children over conventional hearing aids (HAs), but neither of these articles compares EAS outcomes to conventional electric listening. There are no peer-reviewed studies that focus on the use of EAS in children who are fit with the device from initial stimulation or differentiate this cohort from children who have been wearing full electric (FE) maps and are converting to EAS.

With recent changes in electrode design and minimally traumatic surgery techniques, evidence of postoperative hearing preservation in pediatric CI recipients during routine clinical care has been documented. Hearing preservation after CI in children is possible (Skarzynski et al. 2007; Brown et al. 2010; Bruce et al. 2014; Carlson et al. 2017) and perhaps even better than in adults (Anagiotos et al. 2015). As with any new fitting paradigm, is important to validate fitting methods with EAS in the pediatric population and to document outcomes. This study seeks to investigate outcomes with currently available EAS technology and current practices, which will allow for further work on fitting methods. The primary aim of this study is to investigate speech perception performance with unilateral EAS use in two cohorts of pediatric CI recipients with residual hearing: experienced conventional CI users and new recipients. Three questions are addressed: (1) Are outcomes with EAS superior to outcomes with conventional electric-only stimulation in children? (2) Do children with residual hearing benefit from EAS and conventional electric-only stimulation when compared with the preoperative HA condition? (3) Can children with residual hearing derive benefit from EAS after several years of listening with conventional electric-only stimulation?

METHODS

Study Design

This study was approved by the Institutional Review Board at the University of North Carolina at Chapel Hill. The within-subjects design allowed each subject to serve as his or her own control, accommodating for some of the variability among children with CIs. Only one ear was included and tested for each subject. If the contralateral ear had acoustic hearing, it was occluded with a foam plug.

Subjects

Inclusion criteria included use of a CI system produced by Cochlear Limited, an unaided threshold of 80 dB HL or better at 500 Hz and low-frequency pure tone average of 75 dB HL or better in the ear to be studied. For the purposes of this study, the low-frequency pure tone average is defined as the average at 125, 250, and 500 Hz. Twenty-one subjects were enrolled and assigned to two different Arms. Arm 1 consisted of ears fit with a CI speech processor for the first time, and Arm 2 subjects had worn conventional fully electric speech processors for at least 1 year before EAS fitting. Three subjects in Arm 1 lost hearing before the first study-test interval and were withdrawn. One additional subject was lost to follow-up, leaving nine subjects in Arm 1 and seven subjects in Arm 2. Average age of enrollment was 9.94 years for Arm 1 (range, 4.88–15.60; SD = 3.60) and 11.38 years for Arm 2 (range, 8.22–16.27; SD = 2.53).

The mean age of implant for the test ear was 9.89 years for Arm 1 (range, 4.83–15.55; SD = 3.61) and 7.50 years for Arm 2 (range, 2.42–13.36; SD = 3.93). The mean time of use before EAS fitting for Arm 2 ranged from 2.14 to 7.57 years with a mean of 4.04 years (SD = 2.06). Subject demographics can be found in Tables 1 and 2.

At the time of enrollment, eight subjects were bilateral CI recipients, one being a bilateral EAS user. This Arm 1 participant received simultaneous CIs. Preoperative thresholds were within 5 dB at all frequencies, and aided speech perception scores differed by 6% points between ears. The right ear was chosen arbitrarily as the study ear, and the left ear was occluded with a foam plug for all test measures. The remaining subjects used a hearing aid on the contralateral ear, which was occluded with a foam plug during

TABLE 1. Demographics of Subjects in Arm 1

Subject ID	Etiology	Contra Ear	Age at Implant (yrs)	LFPTA (dB HL)	Highest Frequency Target Met (Hz)	Overlap (Hz)
02	Unknown	HA	13.77	27	750	104
03	Unknown	HA	15.55	58	500	93
04	Claudin 14	HA	4.83	45	500	195
06	Unknown	CI	12.32	51	750	347
07	Ototoxicity	HA	7.93	33	750	189
08	Kernicterus	CI	11.90	53	1000	577
13	Suspected hereditary	EAS	6.68	58	2000	629
19	Unknown	CI	8.33	52	500	387
20	Prematurity complications	CI	7.69	45	750	412

Contralateral ear lists the device used on the opposite ear at the time of testing. Age at implant lists the age of implantation in years for the test ear.
CI, cochlear implant; EAS, Electric Acoustic stimulation; HA, hearing aid; LFPTA, low-frequency pure tone average (125, 250, and 500 Hz).

TABLE 2. Demographics of Subjects in Arm 2

Subject ID	Etiology	Contra Ear	Age at Implant (yrs)	Length of CI Use (Yrs)	LFPTA (dB HL)	Highest Frequency Target Met (Hz)	Overlap (Hz)
01	Prematurity complications	CI	7.46	2.91	65	750	201
05	Suspected hereditary	CI	10.46	2.24	55	500	130
09	Malformation: EVA	HA	8.99	2.14	57	750	270
10	Ototoxicity	HA	13.36	3.14	33	500	163
14	Connexin 26	HA	6.94	4.26	50	500	287
17	Suspected hereditary	CI	2.88	7.57	55	500	112
18	Unknown	HA	2.42	6.01	38	1500	1592

Contralateral ear lists the device used on the opposite ear at the time of testing. Age at implant lists the age of implantation for the test ear. Length of CI use lists the time of full electric use in years before enrollment.
CI, cochlear implant; EVA, enlarged vestibular aqueduct; HA, hearing aid; LFPTA, low-frequency pure tone average (125, 250, and 500 Hz).

testing. Only the implanted ear with residual hearing was isolated and measured for purposes of this study.

Device Fitting

Subjects were fit with a Nucleus CP910 Sound Processor and programmed using Custom Sound software. Each participant ultimately received two programs: a FE program and an EAS program. For both programs, electrical threshold and comfort levels were established using behavioral methods. The FE program utilized a default frequency allocation table where the frequency of the most apical channel began at 188 Hz. It did not include any acoustic amplification. For the EAS program, the device was programmed in Hybrid mode with an acoustic component. The acoustic component was fit with either a dome or personal earmold depending on the child's individual fit and venting needs. Real ear-to-coupler difference values were obtained using the Verifit2 system, and real ear measures were used to verify the acoustic fitting. Using a 65 dB SPL speech input, Desired Sensation Level (DSL) V5.0 targets were met for frequencies at which audiometric thresholds were ≤75 dB HL. Within the recipient's map, the frequency boundary of the most apical channel was programmed so that the electric output starting (EOS) frequency was a frequency slightly lower than the highest frequency where target was successfully met, providing some overlap between acoustic and electric stimulation. Acoustic output was reduced via a steep roll-off at the lowest frequency where audiometric thresholds were >75 dB HL, and the acoustic output endpoint (AOE) was defined as the frequency where the maximum possible output crossed audiometric threshold as visualized on the speechmap. For all but one subject, the hearing loss sloped in a manner that limited the acoustic bandwidth to 1500 Hz or lower. For one subject, acoustic targets were met through 2000 Hz and real ear measures verified an AOE of 2640 Hz. Individual audiograms for the test ear are presented in Figure 1.

Overlap was calculated as AOE − EOS. For some subjects, the EOS frequency was adjusted based on subjective feedback. These adjustments occurred mostly in Arm 2 subjects who would have otherwise had an extreme shift in their frequency allocation table and were not acutely comfortable with the sound quality. For these subjects, the EOS was moved to a lower frequency until the subjects reported an improvement in sound quality. This resulted in greater overlap as the acoustic settings were not changed. No subjects required a higher frequency EOS than the original fit.

After a stable soundfield audiogram of 20 to 30 dB HL across octave frequencies from 250 Hz to 6 kHz was verified in the EAS condition, typically by 5 to 9 weeks after the initial stimulation, subjects were given access to both EAS and FE programs to use at home.

Audiometry and Speech Perception

Testing was performed in a single-walled sound-attenuating booth, utilizing a Grason-Stadler GSI-61 audiometer. Soundfield testing was completed at 0° azimuth, and unaided testing

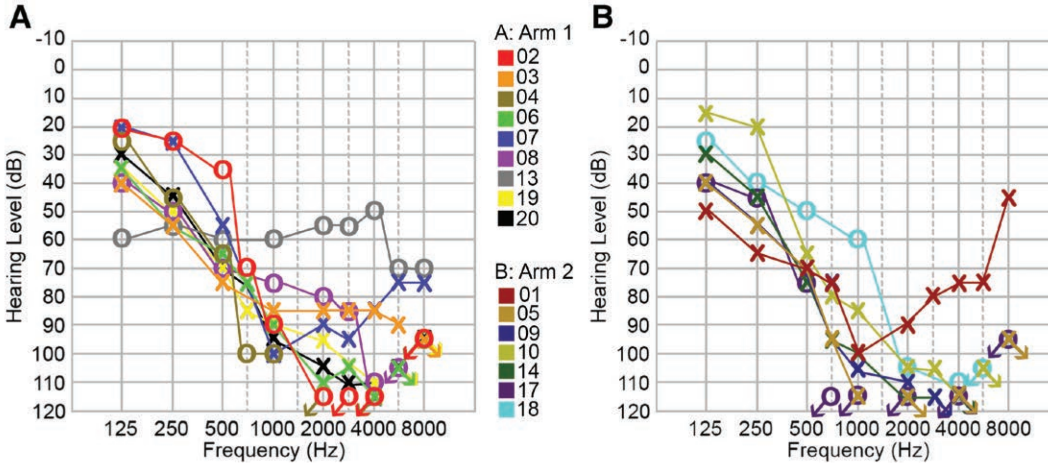

Fig. 1. Unaided air conduction thresholds for the test ears. Audiograms of subjects enrolled in Arm 1 (A) and audiograms of subjects enrolled in Arm 2 (B).

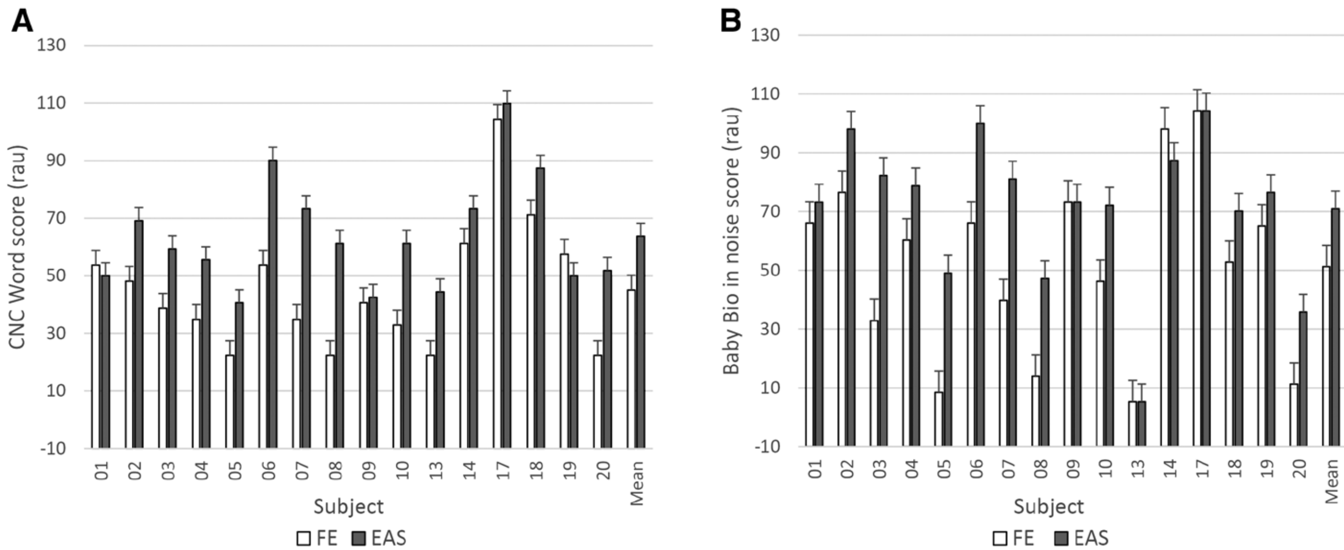

Fig. 2. Electric acoustic stimulation (EAS) vs full electric (FE) program outcomes. Individual mean and scores for all participants. CNC word scores (A) and Baby Bio in noise scores (B) are in rationalized arcsine transform units (rau). FE scores are represented in white and EAS scores are in gray. There was no significant interaction between Arms and programs for either test. For main effects, EAS scores were significantly higher than FE scores for the CNC and Baby Bio in noise ($p < 0.001$ for each test). There was no main effect of Arm for either measure.

was completed using insert earphones. Speech perception testing included the Consonant-Nucleus-Consonant (CNC) word list (Peterson & Lehiste 1962) and the pediatric AZBio sentences (known as the Baby Bio sentences) (Spahr et al. 2014). Both lists were presented at 60 dBA. The CNC was presented in quiet, and the Baby Bio was presented at a +5 dB SNR.

Preoperatively, subjects in Arm 1 were tested using the above measures with their HA in the ear to be implanted only. The HA had been fit to DSL targets using real ear-to-coupler difference. Preoperative data were not collected on Arm 2 subjects as their preoperative test measures differed greatly and could not be compared. Postoperatively, all subjects were tested at the study test point with the FE and EAS programs in the test ear alone. For all test points, the contralateral ear was occluded with a foam plug when there was acoustic hearing, and the test ear was occluded for the FE test condition. Children in Arm 1 were tested as near to the 6 month postactivation mark as possible. Children in Arm 2 returned to the clinic for testing after an acclimatization period of 1 to 3 months. Program use and processor settings were verified in Custom Sound. A CR230 Remote Assistant was used to change programs and verify settings during testing.

Statistical Analysis

Before analysis, CNC Word scores and Baby Bio scores in noise were converted to rationalized arcsine transform units (rau) following the rationalized arcsine method outlined by Studebaker (1985). Data were assessed for normal distribution using Shapiro-Wilk test. Mean outcomes were compared using paired-samples *t* tests, one-way repeated measures analyses of variance (ANOVAs), and two-way repeated measures mixed ANOVAs as described in the Results. Bonferroni adjustments were incorporated for multiple comparisons. Secondary factors were investigated with the Spearman rank-order correlation, comparing changes in scores to various demographic factors.

RESULTS

Outcomes With EAS Versus FE Programs

All subjects were included in the analysis to determine whether children with residual hearing perform better with EAS than FE programs. A two-way repeated measures mixed ANOVA was run to determine the effect of listening programs (EAS and FE) and Arms (1 and 2) on CNC words scores in rau. There was no significant interaction between Arm and program, $F(1,14) = 4.112$; $p = 0.062$; partial $\eta^2 = 0.227$. The main effect of program showed that CNC rau scores were significantly higher using the EAS program than scores using the FE program $F(1,14) = 29.867$; $p < 0.001$; partial $\eta^2 = 0.681$. There was no significant main effect of Arm, $F(1,14) = 1.387$; $p = 0.259$; partial η2 = 0.09. A second two-way repeated measures mixed ANOVA comparing the impact of Arms and programs on converted rau scores obtained on the Baby Bio in noise also indicated no significant interaction between Arm and program, $F(1,14) = 3.105$; $p = 0.100$; partial $\eta^2 = 0.182$. The main effect of program showed that speech perception scores in noise with EAS were significantly higher than those obtained with the FE programs, $F(1,14) = 20.695$; $p < 0.001$; partial $\eta^2 = 0.596$. Again, there was no significant main effect of Arm, $F(1,14) = 1.387$; $p = 0.263$; partial $\eta^2 = 0.089$. Individual and mean scores can be found in Figure 2.

CNC rau scores obtained in the FE program were subtracted from scores obtained with EAS for a difference score. The same was calculated for Baby Bio rau scores. Secondary factors of age at implantation and length of EAS use were included in a Spearman rank-order correlation to investigate their impact on the difference score. There was a strong positive correlation between the difference in scores obtained with the Baby Bio at a +5 dB SNR and the age at implantation, $r_s = 0.631$; $p = 0.009$ (Fig. 3). The correlation between the noise score difference and the length of EAS use was not significant, and there were no significant correlations for the difference in CNC rau scores. Correlation and significance values can be found in Table 3.

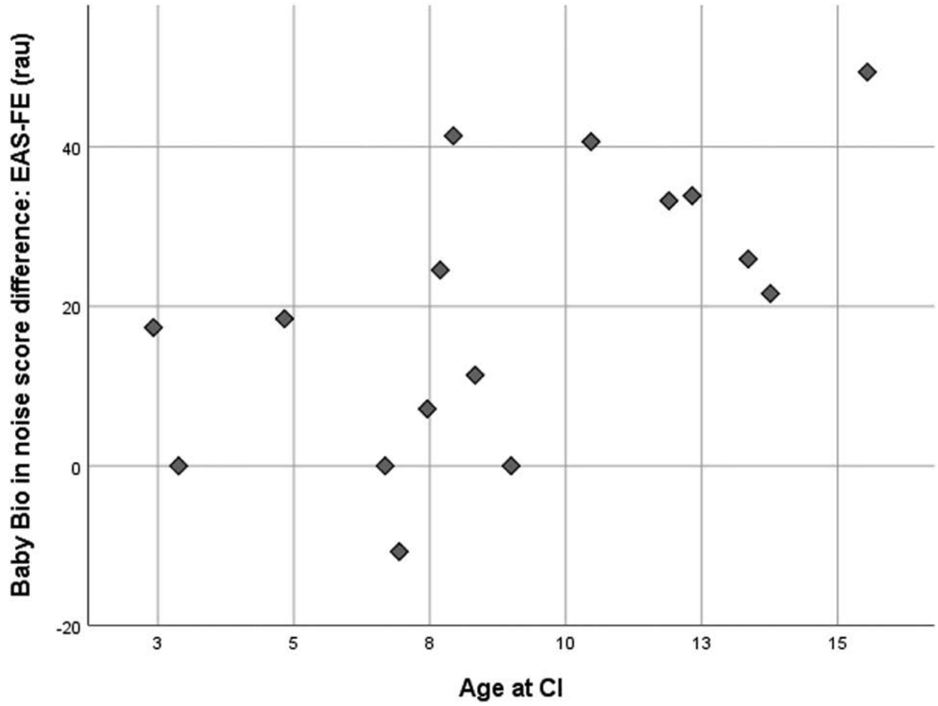

Fig. 3. Correlation between age at implantation and the difference between electric acoustic stimulation (EAS) and full electric (FE) scores in noise for all participants. A strong positive correlation exists between the difference in FE and EAS scores obtained in noise and the age at implantation ($r_s = 0.631$; $p = 0.009$). CI indicates cochlear implant; rau, rationalized arcsine transform units.

Outcomes When Compared With Preoperative Scores

Results from Arm 1 participants were included in a one-way repeated measures ANOVA to determine whether there was a statistically significant change in CNC word scores or Baby Bio scores at +5 dB SNR from pre- to postoperative listening programs. For the first analysis, CNC word scores in rau were compared for each program (preoperative HA, postoperative EAS, and FE). Mean scores in rau were 24.89 (SD = 14.31) when measured preoperatively with an HA, 37.21 (SD = 13.58) with the FE program, and 61.67 (SD = 14.02) with the EAS program. There was a significant effect of time point, $F(2,16) = 18.606$; $p < 0.001$; partial $\eta^2 = 0.699$. Post hoc analysis revealed that CNC rau scores did increase significantly from the preoperative HA condition when measured with EAS (M = 36.77; 95% confidence interval, 13.52, 60.03; $p = 0.004$), but not when measured with FE programs (M = 12.32; 95% confidence interval, −4.30, 28.94; $p = 0.167$). A second analysis compared scores obtained in each program on the Baby Bio in noise test. Mean rau scores were 19.39 (SD = 44.92) when measured preoperatively with an HA, 41.25 (SD = 26.93) with FE, and 67.23 (SD = 31.39) with EAS. Taking a Greenhouse-Geisser correction into account, there was a significant effect of time point, $F(1.187,9.497) = 8.589$; $p = 0.013$; partial $\eta^2 = 0.518$. Post hoc analysis indicated that the increase in scores from listening with an HA to an EAS program was statistically significant (M = 47.84; 95% confidence interval, 9.028, 86.658; $p = 0.018$). Despite more a more than two-fold increase in scores, there was no significant difference between the HA and FE programs (M = 21.86; 95% confidence interval, −21.758, 65.472; $p = 0.507$). Results are presented in Figure 4.

A Spearman rank-order correlation was again used to analyze secondary factors of age at implantation and length of device use between difference scores. For these analyses, scores obtained preoperatively with an HA were subtracted from those obtained with the EAS program. There was no correlation between the difference in scores for CNC words or the Baby Bio at a +5 dB SNR. Statistical data are listed in Table 3.

TABLE 3. Spearman ρ Correlation Coefficients and Two-Tailed Significance Values for Secondary Correlations

		Time Since EAS Fitting	Length of Device Use	Age at Surgery
Q1: All				
CNC difference	r_s	—	−0.41	0.33
(EAS-CI)	p	—	0.12	0.21
BabyBio difference	r_s	—	−0.48	0.63*
(EAS-CI)	p	—	0.06	0.01*
Q2: Arm 1				
CNC difference	r_s	—	−0.05	0.45
(EAS-HA)	p	—	0.90	0.22
BabyBio difference	r_s	—	−0.25	0.58
(EAS-HA)	p	—	0.52	0.10
Q3: Arm 2				
CNC difference	r_s	−0.18	0.14	0.36
(EAS-CI)	p	0.70	0.76	0.43
BabyBio difference	r_s	0.05	−0.25	0.49
(EAS-CI)	p	0.92	0.59	0.27

Q1 relates to question 1 and includes all participants. Q2 relates to question two and only includes Arm 1 participants. Q3 relates to question three and includes only Arm 2 participants.
**p < 0.05.*
CI, cochlear implant; CNC, consonant nucleus consonant test; EAS, electric-acoustic stimulation; HA, hearing aid.

Outcomes After Long-Term Electric-Only Stimulation

A paired samples *t* test was used to investigate benefits of EAS in children with residual hearing who have been wearing

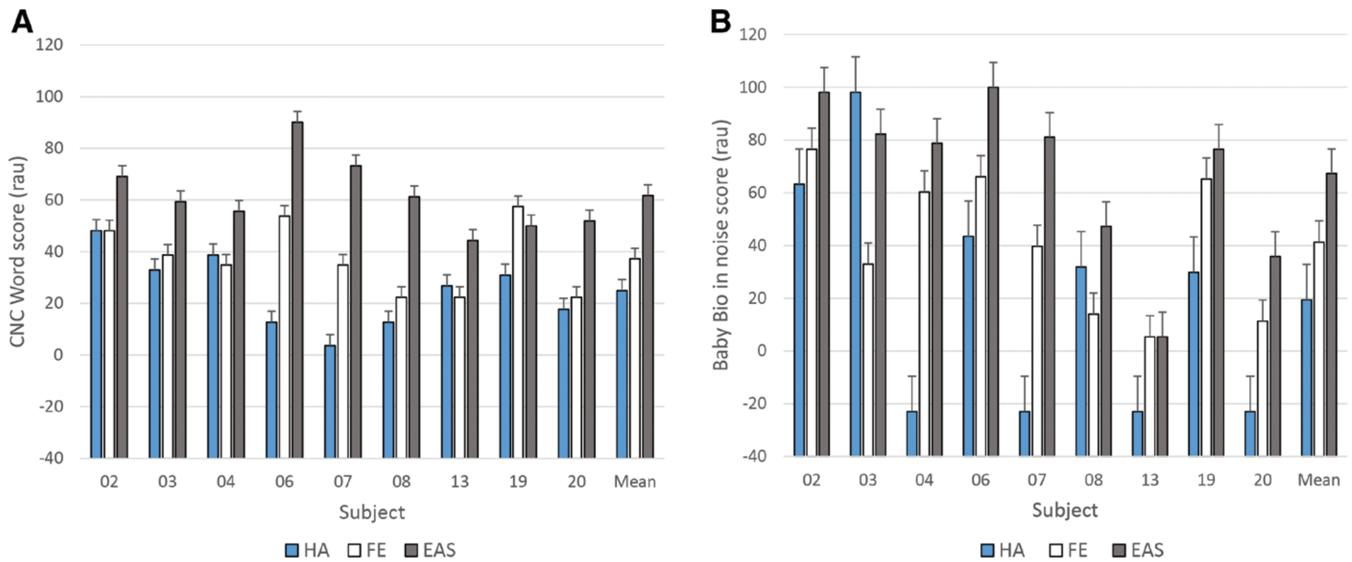

Fig. 4. Pre- and postoperative listening outcomes. Individual mean and scores for Arm 1 participants. CNC word scores (A) and Baby Bio in noise scores (B) are in rationalized arcsine transform units (rau). Preoperative results measured with a hearing aid (HA) are represented by the blue bars, full electric (FE) scores in white, and electric acoustic stimulation (EAS) scores in gray. CNC word scores increased significantly from the preoperative HA condition when measured with EAS ($p = 0.004$), but not when measured in the FE condition ($p = 0.167$). Baby Bio in noise scores followed a similar pattern, increasing significantly from the preoperative HA condition when measured with EAS ($p = 0.018$), but not when measured in the FE condition ($p = 0.507$).

FE programs for over a year (Arm 2). Speech perception scores in both EAS and FE programs were compared. Mean CNC rau scores were 27.36 (SD = 27.36) for the FE programs and 66.41 (SD = 25.49) for the EAS programs. These scores were significantly different, $t(6) = 2.73$; $p = 0.034$; $d = 1.03$. For the Baby Bio in noise, mean rau scores were 64.18 (SD = 36.67) with FE and 75.66 (SD = 16.92) with EAS. These scores were not significantly different, $t(6) = 1.719$; $p = 0.136$; $d = 0.65$. Results are shown in Figure 5.

The scores obtained with the FE program were subtracted from the scores obtained with the EAS program for a difference score. Secondary factors of age at implantation, time since EAS fitting, and time since CI fitting were included in a Spearman rank-order correlation. There was no significant correlation to the difference score for any of these factors for either measure. Correlation and significance values can be found in Table 3.

DISCUSSION

Question 1

Are Outcomes With EAS Superior to Outcomes With Conventional Electric-Only Stimulation in Children? • In this study, children with residual hearing performed better on both single words in quiet and sentences in noise with EAS when compared with conventional FE programs. These findings are similar to those reported by Wolfe et al. (2017) who also found that children with residual hearing benefit from the use of EAS in noise; however, findings of the current study did reach statistical significance. In addition, the current study used FE programs rather than truncated electric maps, allowing access to a wider frequency spectrum. These results echo outcomes of many adult studies investigating unilateral EAS use as well (Gantz et al. 2005; Gstoettner et al. 2008; Adunka et al. 2010a; Adunka et al. 2013; Mahmoud et al. 2014; Sheffield et al. 2015; Pillsbury et al. 2018).

The positive correlation between EAS benefit in noise and age at CI was an interesting finding. Many of the children in this study had progressive hearing loss that resulted in a later age of implantation. Others had significant levels of measurable residual hearing but waited to receive a CI until single word speech perception scores decreased or there was a plateau in language development. It is possible that longer exposure to acoustic hearing leads to a greater ability to integrate acoustic and electric cues. Or perhaps there was better hair cell survival in this population that lead to a longer period of perceived HA benefit and delayed implantation. This finding warrants future investigation with a larger cohort of participants.

Question 2

Do Children With Residual Hearing Benefit From EAS and Conventional Electric-Only Stimulation When Compared With the Preoperative HA Condition? • Children with residual hearing in this study showed significant benefit with EAS for both single words and sentences in noise when compared with the preoperative HA condition. Although all of the Arm 1 participants had been fit with EAS from initial stimulation, these findings are similar to Skarzynski and Lorens (2010) and Skarzynski et al. (2007). The lack of significant difference between the preoperative and FE conditions is surprising. A trend toward improvement was evident in noise, however, as scores in the FE condition were more than double those obtained preoperatively. Children with progressive postlingual high-frequency hearing loss have been shown to do well with conventional CI programming (Meredith et al. 2017; Ahmad et al. 2012), as have teens and adults with significant residual hearing (Hughes et al. 2014). In the current study, the small sample size may be a factor. The majority of the participants were using the EAS program on a daily basis and were tested acutely with the electric-only programs which could have influenced outcomes. Only one subject, subject 3, showed a decrement with an FE

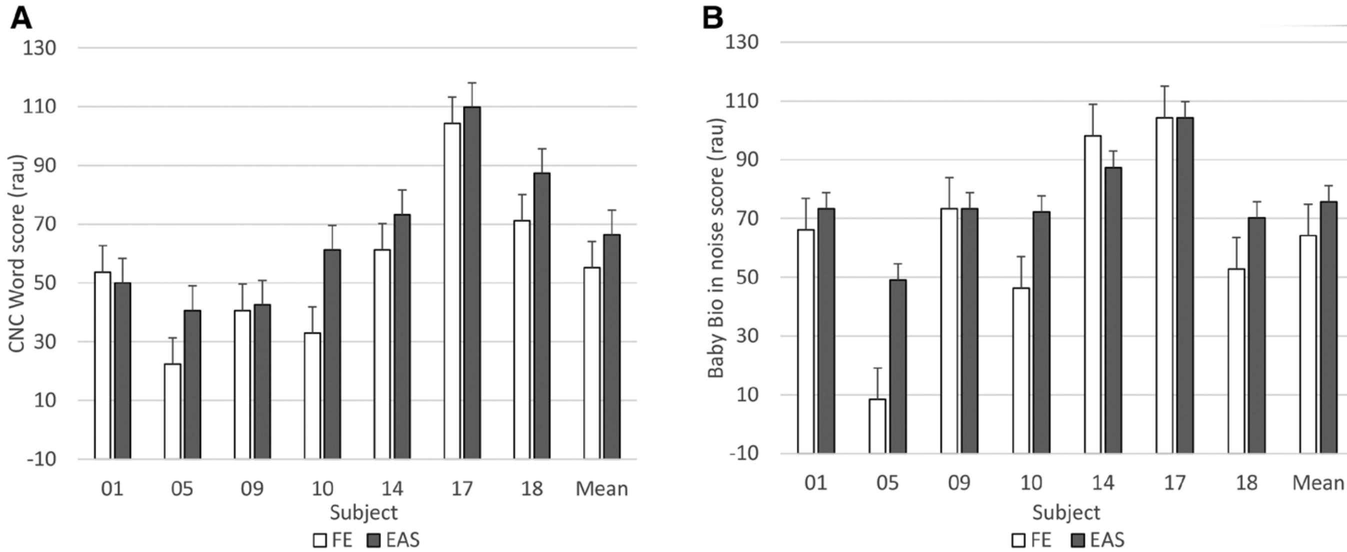

Fig. 5. Electric acoustic stimulation (EAS) outcomes after long-term full electric (FE) stimulation. Individual and mean scores for Arm 2 participants. CNC word scores (A) and Baby Bio in noise scores (B) are in rationalized arcsine transform units (rau). FE scores are represented in white and EAS scores are in gray. Scores in the EAS condition were significantly higher than those obtained in the FE condition for CNC words ($p = 0.034$), but not for the Baby Bio in noise ($p = 0.136$).

program, and extraneous patient factors may have affected performance on the day of data collection. In addition, these scores were obtained at just 6 months poststimulation. It is likely that with time, scores would improve. Conversely, results when comparing scores obtained with EAS and FE programs when including all subjects suggest that Arm, or previous experience with electric-only programs, is not a factor. Children who had been listening to an FE program for an average of 4 years while retaining residual hearing did not perform differently than the newly implanted group. Preoperative scores on CNC words and Baby Bio sentences at +5 dB SNR are not available for Arm 2 participants because the children were either too young to participate or the test was not part of our standard battery at that time. While EAS was the superior intervention for this group and clearly provided better outcomes than an HA, all but one participant also did as well or better in the CI condition than the preoperative HA condition.

In general, however, the CNC word scores in the FE programs are rather low. As all but one participant in this group was a full-time user based on datalogging, the mean age of implant (9.89 years) is a concern. All of the children were born with some degree of hearing loss and over half of them were progressive. Often children with significant levels of residual hearing are not referred for implantation until professionals and caregivers feel that there is a plateau in language development or difficulty in school. It may be that these children were referred after an extended period of time listening to degraded input and would have had better outcomes if they had been implanted sooner. Unfortunately, the preoperative histories of this cohort are not enough to make a conclusion.

Question 3

Can Children With Residual Hearing Derive Benefit From EAS After Several Years of Listening With Conventional Electric-Only Stimulation? • All subjects did as well or better with EAS than FE stimulation after several years of FE program use; however, only results for CNC words reached statistical significance. Of the subjects who did not show significant improvement in sentence in noise performance, ceiling effects may be at play. Some of the individual outcomes for participants were of interest. For example, subject 1 experienced an acute increase of 37% points in Baby Bio in noise scores and 22% points CNC word scores after adjustment to his electric frequency boundary several months after this study was completed. After 5 additional months of listening, subject 14 had an increase in CNC word scores with the EAS device, resulting in a 38% point difference between conditions. Subjects 14 and 17 were at or near ceiling on the Baby Bio in noise, and subject 9 was lost to follow-up. In the study by Wolfe et al. (2017), all of the subjects were experienced FE listeners and had about 4 weeks of acclimatization to the EAS program. There was a trend in improvement with EAS that did not reach significance. These findings are similar to the current study, although electric programs differed. Wolfe et al. disabled the acoustic input for the electric condition without changing electric frequency boundaries, so the subjects were listening to truncated electric maps. It is possible that both the EAS and electric conditions were still novel in that study and that the EAS condition was still novel in the current study. Children who start with FE programs may need more than 1 to 3 months to adjust to EAS and to find the appropriate frequency boundaries.

Low CNC word scores in the FE program is of concern for the participants in this Arm as well. Again, the mean age of implant (7.5 years) may be a factor. Many of these children were implanted when centers were just beginning to expand indications to include children with greater levels of residual hearing. They may have had poor word recognition and an extended period of time listening to degraded input before referral.

Overall, the current study provides evidence that children with residual hearing who have been wearing FE programs can adapt to EAS and benefit. Scholz et al. (2017) also fit EAS on children with long-term use of FE programs, but did not find statistical significance. The authors note the variability in outcomes within their group and the difficulty their subjects had with acceptance after 4 weeks of acclimatization time. Two of the Arm 2 subjects have opted to discontinue use of the acoustic component. Neither liked using the receiver in canal (RIC) device. One recognized the benefit but was not comfortable with dome or personal earmold fitting. The other was an exceptional performer with his FE program, and while he liked the sound quality, he and his parents did not feel the benefits outweighed the additional troubleshooting and care of the RIC. Other subjects have continued to use their acoustic component, and three of the four bimodal listeners in Arm 2 decided to become bilateral EAS users rather than bimodal listeners. All four bimodal users enrolled in Arm 1 have since received a second CI and are currently bilateral EAS users. Acceptance in this study seems to be greater than what was found by Scholz et al. (2017) and more closely aligned with findings by Wolfe et al. (2017).

Limitations

This study did not concentrate on optimizing settings such as acoustic bandwidth, electric start frequencies, or acoustic fitting formula. Future studies should be designed to establish best practices for programming EAS in this special population. Optimal fittings of the RIC devices for smaller ears and ideal acoustic targets should be a focus. In the current study, DSL targets were used in order to ensure audibility soft and loud speech for children still developing language. In truth, DSL was designed for a full bandwidth of acoustic hearing, and children who are utilizing EAS are using acoustic information in a completely different way. They may require a specific pediatric EAS fitting formula that has yet to be developed.

CONCLUSIONS

Whether new implant recipients or long-term CI users, children with residual hearing should be afforded the opportunity to use EAS. In most cases, children with functional low-frequency hearing who may be able to benefit from a CI do not meet candidacy criteria under manufacturer labeling; however, this study supports consideration for implantation. Children with HAs who have residual hearing, but insufficient access to high-frequency sounds, should be considered for CI surgery and fit with EAS when hearing is preserved. The potential for improved speech understanding in quiet and in noise, as documented here, could increase the accessibility of spoken language. Higher speech perception scores in general are related to higher language function (Blamey et al. 2001; Desjardin et al. 2009; Davidson et al. 2011; Geers et al. 2013). Furthermore, children with CIs are known to have greater difficulty understanding in noise than children with HAs who have moderate to severe losses (Eisenberg et al. 2004). Combining acoustic and electric information through the use of EAS provides an improvement over FE stimulation, and better hearing in noise presents more opportunities for exposure to spoken language and incidental learning.

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REFERENCES

Adunka, O. F., Dillon, M. T., Adunka, M. C., et al. (2013). Hearing preservation and speech perception outcomes with electric-acoustic stimulation after 12 months of listening experience. *Laryngoscope*, *123*, 2509–2515.

Adunka, O. F., Dillon, M. T., Adunka, M. C., et al. (2014). Cochleostomy versus round window insertions: Influence on functional outcomes in electric-acoustic stimulation of the auditory system. *Otol Neurotol*, *35*, 613–618.

Adunka, O. F., Pillsbury, H. C., Adunka, M. C., et al. (2010a). Is electric acoustic stimulation better than conventional cochlear implantation for speech perception in quiet? *Otol Neurotol*, *31*, 1049–1054.

Adunka, O. F., Pillsbury, H. C., Buchman, C. A. (2010b). Minimizing intracochlear trauma during cochlear implantation. *Adv Otorhinolaryngol*, *67*, 96–107.

Ahmad, F. I., Demason, C. E., Teagle, H. F., et al. (2012). Cochlear implantation in children with postlingual hearing loss. *Laryngoscope*, *122*, 1852–1857.

Anagiotos, A., Hamdan, N., Lang-Roth, R., et al. (2015). Young age is a positive prognostic factor for residual hearing preservation in conventional cochlear implantation. *Otol Neurotol*, *36*, 28–33.

Bagatto, M., Scollie, S. D., Hyde, M., et al. (2010). Protocol for the provision of amplification within the Ontario infant hearing program. *Int J Audiol*, *49*(Suppl 1), S70–S79.

Blamey, P. J., Sarant, J. Z., Paatsch, L. E., et al. (2001). Relationships among speech perception, production, language, hearing loss, and age in children with impaired hearing. *J Speech Lang Hear Res*, *44*, 264–285.

Brown, R. F., Hullar, T. E., Cadieux, J. H., et al. (2010). Residual hearing preservation after pediatric cochlear implantation. *Otol Neurotol*, *31*, 1221–1226.

Bruce, I. A., Felton, M., Lockley, M., et al. (2014). Hearing preservation cochlear implantation in adolescents. *Otol Neurotol*, *35*, 1552–1559.

Carlson, M. L., Driscoll, C. L., Gifford, R. H., et al. (2011). Implications of minimizing trauma during conventional cochlear implantation. *Otol Neurotol*, *32*, 962–968.

Carlson, M. L., Patel, N. S., Tombers, N. M., et al. (2017). Hearing preservation in pediatric cochlear implantation. *Otol Neurotol*, *38*, e128–e133.

Davidson, L. S., Geers, A. E., Blamey, P. J., et al. (2011). Factors contributing to speech perception scores in long-term pediatric cochlear implant users. *Ear Hear*, *32*(Suppl 1), 19S–26S.

Davidson, L. S., Geers, A. E., Nicholas, J. G. (2014). The effects of audibility and novel word learning ability on vocabulary level in children with cochlear implants. *Cochlear Implants Int*, *15*, 211–221.

Desjardin, J. L., Ambrose, S. E., Martinez, A. S., et al. (2009). Relationships between speech perception abilities and spoken language skills in young children with hearing loss. *Int J Audiol*, *48*, 248–259.

Eisenberg, L. S., Kirk, K. I., Martinez, A. S., et al. (2004). Communication abilities of children with aided residual hearing: Comparison with cochlear implant users. *Arch Otolaryngol Head Neck Surg*, *130*, 563–569.

Erixon, E., Köbler, S., Rask-Andersen, H. (2012). Cochlear implantation and hearing preservation: Results in 21 consecutively operated patients using the round window approach. *Acta Otolaryngol*, *132*, 923–931.

Gantz, B. J., Turner, C., Gfeller, K. E., et al. (2005). Preservation of hearing in cochlear implant surgery: Advantages of combined electrical and acoustical speech processing. *Laryngoscope*, *115*, 796–802.

Geers, A. E., Davidson, L. S., Uchanski, R. M., et al. (2013). Interdependence of linguistic and indexical speech perception skills in school-age children with early cochlear implantation. *Ear Hear*, *34*, 562–574.

Geers, A. E., Nicholas, J. G., Sedey, A. L. (2003). Language skills of children with early cochlear implantation. *Ear Hear*, *24*, 46S–58S.

Gstoettner, W. K., van de Heyning, P., O'Connor, A. F., et al. (2008). Electric acoustic stimulation of the auditory system: Results of a multi-centre investigation. *Acta Otolaryngol*, *128*, 968–975.

Havenith, S., Lammers, M. J., Tange, R. A., et al. (2013). Hearing preservation surgery: Cochleostomy or round window approach? A systematic review. *Otol Neurotol*, *34*, 667–674.
Hughes, M. L., Neff, D. L., Simmons, J. L., et al. (2014). Performance outcomes for borderline cochlear implant recipients with substantial preoperative residual hearing. *Otol Neurotol*, *35*, 1373–1384.
Kisser, U., Wünsch, J., Hempel, J. M., et al. (2016). Residual hearing outcomes after cochlear implant surgery using ultra-flexible 28-mm electrodes. *Otol Neurotol*, *37*, 878–881.
Mahmoud, A. F., Massa, S. T., Douberly, S. L., et al. (2014). Safety, efficacy, and hearing preservation using an integrated electro-acoustic stimulation hearing system. *Otol Neurotol*, *35*, 1421–1425.
McCreery, R. W., Brennan, M., Walker, E. A., et al. (2017). Perceptual implications of level- and frequency-specific deviations from hearing aid prescription in children. *J Am Acad Audiol*, *28*, 861–875.
Meredith, M. A., Rubinstein, J. T., Sie, K. C. Y., et al. (2017). Cochlear implantation in children with postlingual progressive steeply sloping high-frequency hearing loss. *J Am Acad Audiol*, *28*, 913–919.
Peterson, G. E., & Lehiste, I. (1962). Revised CNC lists for auditory tests. *J Speech Hear Disord*, *27*, 62–70.
Pillsbury, H. C., III, Dillon, M. T., Buchman, C. A., et al. (2018). Multicenter US clinical trial with an electric-acoustic stimulation (EAS) system in adults: Final outcomes. *Otol Neurotol*, *39*, 299–305.
Ross, M., & Seewald R. C. (1988). Hearing aid selection and evaluation with young children. In F. H. Bess (Ed.), *Hearing Impairment in Children* (pp. 190–213). Parkton, MD: York Press.
Santa Maria, P. L., Domville-Lewis, C., Sucher, C. M., et al. (2013). Hearing preservation surgery for cochlear implantation: Hearing and quality of life after 2 years. *Otol Neurotol*, *34*, 526–531.
Santa Maria, P. L., Gluth, M. B., Yuan, Y., et al. (2014). Hearing preservation surgery for cochlear implantation: A meta-analysis. *Otol Neurotol*, *35*, e256–e269.
Scholz, S., Todt, I., Olze, H., et al. (2017). Benefit of a hybrid speech processor in implanted young children with residual hearing. *JSM Health Educ Prim Health Care*, *2*, 1028.
Seewald, R., Moodie, S., Scollie, S., et al. (2005). The DSL method for pediatric hearing instrument fitting: Historical perspective and current issues. *Trends Amplif*, *9*, 145–157.
Seewald, R. C., & Ross, M. (1988). Amplification for young hearing-impaired children. In M. Pollack (Ed.), *Amplification for the Hearing-Impaired* (3rd ed.) (pp. 213–271). New York, NY: Grune & Stratton.
Seewald, R. C., Ross, M., Spiro, M. K. (1985). Selecting amplification characteristics for young hearing-impaired children. *Ear Hear*, *6*, 48–53.
Sheffield, S. W., Jahn, K., Gifford, R. H. (2015). Preserved acoustic hearing in cochlear implantation improves speech perception. *J Am Acad Audiol*, *26*, 145–154.
Sininger, Y. S., Grimes, A., Christensen, E. (2010). Auditory development in early amplified children: Factors influencing auditory-based communication outcomes in children with hearing loss. *Ear Hear*, *31*, 166–185.
Skarzynski, H., & Lorens, A. (2010). Electric acoustic stimulation in children. *Adv Otorhinolaryngol*, *67*, 135–143.
Skarzynski, H., Lorens, A., Matusiak, M., et al. (2014). Cochlear implantation with the nucleus slim straight electrode in subjects with residual low-frequency hearing. *Ear Hear*, *35*, e33–e43.
Skarzynski, H., Lorens, A., Piotrowska, A., et al. (2007). Partial deafness cochlear implantation in children. *Int J Pediatr Otorhinolaryngol*, *71*, 1407–1413.
Spahr, A. J., Dorman, M. F., Litvak, L. M., et al. (2014). Development and validation of the pediatric AzBio sentence lists. *Ear Hear*, *35*, 418–422.
Stelmachowicz, P. G., Hoover, B. M., Lewis, D. E., et al. (2000). The relation between stimulus context, speech audibility, and perception for normal-hearing and hearing-impaired children. *J Speech Lang Hear Res*, *43*, 902–914.
Stiles, D. J., Bentler, R. A., McGregor, K. K. (2012). The Speech Intelligibility Index and the pure-tone average as predictors of lexical ability in children fit with hearing AIDS. *J Speech Lang Hear Res*, *55*, 764–778.
Strauss, S., & van Dijk, C. (2008). Hearing instrument fittings of pre-school children: Do we meet the prescription goals? *Int J Audiol*, *47*(Suppl 1), S62–S71.
Studebaker, G. A. (1985). A "rationalized" arcsine transform. *J Speech Hear Res*, *28*, 455–462.
Sweeney, A. D., Hunter, J. B., Carlson, M. L., et al. (2016). Durability of hearing preservation after cochlear implantation with conventional-length electrodes and scala tympani insertion. *Otolaryngol Head Neck Surg*, *154*, 907–913.
Tamir, S., Ferrary, E., Borel, S., et al. (2012). Hearing preservation after cochlear implantation using deeply inserted flex atraumatic electrode arrays. *Audiol Neurootol*, *17*, 331–337.
Tobey, E. A., Thal, D., Niparko, J. K., et al.; CDaCI Investigative Team. (2013). Influence of implantation age on school-age language performance in pediatric cochlear implant users. *Int J Audiol*, *52*, 219–229.
Wolfe, J., Neumann, S., Schafer, E., et al. (2017). Potential benefits of an integrated electric-acoustic sound processor with children: A preliminary report. *J Am Acad Audiol*, *28*, 127–140.
Yao, W. N., Turner, C. W., Gantz, B. J. (2006). Stability of low-frequency residual hearing in patients who are candidates for combined acoustic plus electric hearing. *J Speech Lang Hear Res*, *49*, 1085–1090.
Zhang, T., Spahr, A. J., Dorman, M. F., et al. (2013). Relationship between auditory function of nonimplanted ears and bimodal benefit. *Ear Hear*, *34*, 133–141.