



Bilateral Cochlear Implants or Bimodal Hearing for Children with Bilateral Sensorineural Hearing Loss

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Abstract

Purpose of Review This review describes speech perception and language outcomes for children using bimodal hearing (cochlear implant (CI) plus contralateral hearing aid (HA)) as compared to children with bilateral CIs and contrasts said findings with the adult literature. There is a lack of clinical evidence driving recommendations for bimodal versus bilateral CI candidacy and as such, clinicians are often unsure about when to recommend a second CI for children with residual acoustic hearing. Thus, the goal of this review is to identify scientific information that may influence clinical decision making for pediatric CI candidates with residual acoustic hearing.

Recent Findings Bilateral CIs are considered standard of care for children with bilateral severe-to-profound sensorineural hearing loss. For children with aidable acoustic hearing—even in just the low frequencies—an early period of bimodal stimulation has been associated with significantly better speech perception, vocabulary, and language development. HA audibility, however, is generally poorer than that offered by a CI resulting in interaural asymmetry in auditory access, speech perception, head shadow, as well as brainstem and cortical activity and development. Thus, there is a need to optimize “two-eared” hearing while maximizing a child’s potential with respect to hearing, speech, and language while ensuring that we limit asymmetrically driven auditory neuroplasticity. A recent large study of bimodal and bilateral CI users suggested that a period of bimodal stimulation was only beneficial for children with a better-ear pure tone average (PTA) ≤ 73 dB HL. This 73-dB-HL cutoff applied even to children who ultimately received bilateral CIs.

Summary Though we do not yet have definitive guidelines for determining bimodal versus bilateral CI candidacy, there is increasing evidence that (1) bilateral CIs yield superior outcomes for children with bilateral severe-to-profound hearing loss and (2) an early period of bimodal stimulation is beneficial for speech perception and language development, but only for children with better-ear PTA ≤ 73 dB HL. For children with residual acoustic hearing, even in just the low-frequency range, rapid sequential bilateral cochlear implantation following a trial period with bimodal stimulation will yield best outcomes for auditory, language, and academic development. Of course, there is also an increasing prevalence of cochlear implantation with acoustic hearing preservation allowing for combined electric and acoustic stimulation even following bilateral implantation.

Keywords Cochlear implants · Bilateral · Bimodal hearing · Pediatric · Speech perception · Language

Introduction

In 1985, the Food and Drug Administration (FDA) approved multi-channel cochlear implants (CIs) for adults with bilateral

profound sensorineural hearing loss. Current adult indications for conventional CI systems specify bilateral moderate sloping to profound sensorineural hearing loss [1, 2]. Indications for hybrid or electric and acoustic stimulation (EAS) systems specify low-frequency thresholds ≤ 65 dB HL with precipitously sloping bilateral high frequency hearing loss [3, 4]. Approximately 75% of adult CI candidates receiving conventional CI systems have aidable low-frequency hearing in both ears prior to implantation [5], demonstrating the expanding auditory profile of adult CI recipients.

In 1990, the FDA approved cochlear implantation for children 2+ years of age with the minimum age lowered to

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18 months in 1998, 12 months in 2000, and most recently to 9 months in early 2020. Despite the most recent age change, few changes have been made to audiometric-based pediatric CI criteria over the past 30 years. The most recent amendment to audiometric criteria was made in 2000 when hearing loss severity was expanded to include children with bilateral severe-to-profound sensorineural hearing loss who are 2+ years [6], with criteria remaining most restrictive for children 9 months to 2 years (i.e., bilateral profound sensorineural hearing loss).

Though *audiometric criteria* for pediatric CI have not changed in over 2 decades, there are reports in the literature of expanded indications—albeit off-label—for children with less severe hearing losses. Carlson and colleagues [7] demonstrated that for 51 children implanted with better than severe-to-profound SNHL, CIs offered highly significant benefit for speech recognition in the implanted ear and in the bilateral, best-aided condition. Leigh and colleagues [8] reported speech perception outcomes for a group of 140 children—78 CI users and 62 bilateral hearing aid (HA) users—to determine a criterion pure tone average (PTA) above which cochlear implantation would be statistically more likely to yield significant benefit over bilateral HAs. They reported that children with PTAs above (i.e. poorer than) 60 dB HL, would have a 75% likelihood of achieving better outcomes with a CI as compared to bilateral HAs [8]. Given these reports in recent years, standard clinical practice has opened up cochlear implantation to children who have residual hearing, typically in the low-to-mid frequencies, in one or both ears.

A query of our pediatric CI REDCap [9] database revealed that for the 379 children (577 ears) implanted over a 6-year range from January 2013 through December 2018, 26% had 250-Hz thresholds \leq 80 dB HL in the ear to be implanted and 40% of 181 unilateral CI recipients had 250-Hz thresholds \leq 80 dB HL in the non-implanted ears. While this proportion of pediatric CI users with potentially aidable low-frequency hearing is substantially lower than our adult population, the audiometric severity and configurations for our pediatric CI candidates is consistent with two primary treatment options, namely bimodal hearing (CI + contralateral HA) or bilateral CI. It is important to note that pediatric cochlear implantation with acoustic hearing preservation in the implanted ear(s) remains a viable option [10–19]. Specifically, there is no clinical or scientific reason that hearing preservation cochlear implantation should not be attempted for both pediatric and adult patients who have acoustic hearing to preserve. There is even evidence suggesting that in the absence of hearing to preserve, minimally traumatic surgical techniques hold potential for higher CI outcomes presumably due to preservation of intracochlear structures [20–22]. Despite the possibility and benefits of hearing preservation cochlear implantation for pediatric CI users, for the purposes of this manuscript, we will be focusing on auditory and language outcomes for children

utilizing traditional bimodal hearing and bilateral CIs who have relatively symmetric degrees and configurations of hearing loss. That is, we will not be covering CIs as related to EAS systems, cases of single-sided deafness (SSD), or highly asymmetric hearing.

Benefits Obtained from Bimodal Hearing and Bilateral CIs

Bimodal stimulation yields benefit as compared to unilateral CI stimulation for speech recognition in quiet, noise, and music perception for both adults [23–29] and children [30, 31–34]. Bimodal benefits are more variable for horizontal plane localization with some studies showing significant bimodal benefit [31, 32, 35] and others showing little-to-no benefit as compared to the CI-alone condition [35–37]. Additionally, there are reports that bimodal hearing offers substantial qualitative benefits resulting in a more natural sound quality for speech and music stimuli as compared to CI-alone listening [28, 38].

On the other hand, bilateral cochlear implantation is the standard of care intervention for adults and children with bilateral severe-to-profound SNHL. Bilateral CI benefit is noted as compared to unilateral CI for both adult and pediatric CI recipients on measures of speech recognition in quiet and noise [39–42] as well as for horizontal-plane localization [36, 42]. Horizontal-plane localization is generally superior for bilateral CI users as compared to bimodal listeners for both between-group analyses [36] and within-subject analyses for bimodal listeners who later received a second CI [35].

Bimodal Vs. Bilateral CI Candidacy: Current Practice

There are no data-driven guidelines determining when the expected benefit to be gained from a second CI would exceed that of a HA used in a bimodal configuration. This issue is most relevant for young children due to the critical, time-sensitive periods of auditory, speech, and language development. Further complicating this matter is that there is increasing evidence that binaural effects, such as summation, as well as speech and language outcomes decrease with delay between surgeries, suggesting a sensitive period for bilateral cochlear implantation [43–49, 50••].

Decisions regarding bilateral CI candidacy are often made for our youngest bimodal listeners on the basis of audiometric thresholds in the non-CI ear. However, based on data from adult CI users, the degree of bimodal benefit provided by contralateral acoustic hearing varies dramatically across listeners and is not *reliably related* to the unaided audiogram. Though there is a correlation ($r = 0.25$ to 0.38) between bimodal benefit and unaided audiometric thresholds in the non-CI ear for adult listeners [25, 29], the magnitude of bimodal benefit for individuals with unaided thresholds ranging from

40 to 100 dB HL extends from negative benefit (bimodal interference) to over 60-percentage points for speech recognition in noise [24, 25, 27, 32]—rendering this relationship not clinically useful for guiding recommendations at the individual patient level. Comprehensive datasets highlighting the relationship between unaided audiometric thresholds and bimodal benefit for pediatric CI users have not been published, and as such, there are no data supporting clinical guidance regarding expected benefit from bimodal hearing on the basis of the unaided audiogram.

Since the audiogram provides little clinical utility for determining bilateral CI candidacy for individuals with bilateral moderate-severe or poorer sensorineural hearing loss, the next logical measure is aided speech recognition performance. However, determining bilateral CI candidacy is not likely to be sensitive when using measures of speech recognition in the bilateral, best-aided condition. This is potentially problematic as most audiology clinics use a single loudspeaker delivering co-located speech and noise—a test environment for which there is no evidence that a second CI would yield greater benefit over bimodal hearing [23, 24, 51–56]. That is, even for bilateral CI users who exhibit significant benefits beyond that offered by bimodal hearing, they may not demonstrate said benefits in typical clinical testing environments. In fact, such testing environments may actually favor bimodal hearing as this condition affords different, yet complementary information across ears [57]. In fact, a recent cross-sectional study of 85 adult bimodal and bilateral CI users demonstrated that while clinical assessments of speech recognition were not sensitive to distinguish between bimodal and bilateral CI performance, the use of complex listening scenarios with multiple speakers and diffuse noise did demonstrate performance differences across groups [24]. To date, no such studies have been published for pediatric bimodal and bilateral CI populations.

Purpose of Review

Because we do not currently have clinical evidence driving recommendations for bimodal and bilateral CI candidacy for either adults or children, this poses a clinical quandary for audiologists and otologists who may be unsure when to recommend a second CI for children with residual acoustic hearing in the non-CI ear—particularly given the potential for lost acoustic hearing. Thus, the purpose of this review is to describe speech perception and language outcomes for children using bimodal hearing as compared to children with bilateral CIs and to contrast those findings with the adult literature. The goal is to highlight scientific information that may guide clinical decision making for pediatric CI candidates with residual acoustic hearing as well as to highlight gaps in our current knowledge base so as to motivate future investigation.

Recent Findings

Speech Recognition in Quiet and Noise: Bimodal and Bilateral CI Performance

Binaural summation exhibited for children using bimodal stimulation or bilateral CIs is similar in magnitude to that described for adult bimodal and bilateral CI users [24, 39, 58] in the range of 1- to 5-percentage points in quiet [32, 41, 59], approximately 2- to 10-percentage points in noise for fixed SNR tasks [32, 41, 59], and 2 to 4 dB for adaptive speech receptive thresholds [32, 60, 61]. Deep and colleagues [59] reported significantly greater binaural summation for pediatric bimodal listeners (13-percentage points) as compared to a group of sequential bilateral CI users (2-percentage points) for sentence recognition in quiet; this finding is not unexpected given the complementary information offered by acoustic lower frequency hearing (F0 and temporal fine structure) combined with broadband audibility from the CI ear.

Despite similar binaural summation for bimodal and bilateral CI listeners, pediatric bimodal listeners have greater interaural asymmetry for speech recognition in quiet and in noise [50••, 62]. This trend for greater interaural asymmetry holds negative consequences for speech perception and has been similarly reported for adult bimodal listeners [23, 24, 58]. Degree of interaural asymmetry is similar across age groups with approximately 68% of bimodal children [50••] and 67 to 94% of bimodal adults [24, 58] exhibiting significant interaural asymmetry in speech perception. In contrast, just 14% of bilaterally implanted children [41, 50••] and 16–25% of bilaterally implanted adults [24, 58] demonstrate significant interaural asymmetry in speech perception.

For children receiving sequential bilateral implants, degree of interaural asymmetry approximated that of pediatric bimodal listeners in quiet and was much less prevalent for speech recognition in noise [50••, 59]. That is, children with sequential bilateral implants exhibit more similar scores for speech recognition in quiet and noise when comparing performance for each ear individually. Less interaural asymmetry in performance across the two ears leads to greater binaural summation and greater speech recognition benefits with roving speech signals (e.g., conversations between multiple talkers and group gatherings) [24, 58]. The timing between sequential bilateral CI surgeries is a critical variable, particularly when interaural symmetry in speech recognition is important, such as in communicative environments where speech and noise are spatially separated and in conditions for which multiple talkers are present and may be roving, such as in a school cafeteria or on the playground. Degree of interaural asymmetry is an important clinical consideration as it impacts benefit afforded by binaural summation. Multiple studies have shown that bimodal and bilateral CI users exhibiting the greatest interaural asymmetry derive little-to-no binaural

summation—a finding that has been reported both for adults [24, 63, 64] and children [41, 50••].

Interaural symmetry in audibility and speech recognition is also critical for head shadow—a key benefit of *two-eared hearing*. Though head shadow is not a true binaural benefit as even monaural listeners can benefit provided that noise is directed toward the poorer ear, individuals exhibiting positive and symmetric head shadow across ears are most likely to derive benefit from spatial release from masking and are less influenced by moving talkers and/or noise location [58, 65]. Adult bimodal listeners have demonstrated little-to-no benefit from head shadow for the poorer hearing ear, typically the non-CI ear [23, 37, 58, 66–69]. In contrast, adult bilateral CI users demonstrate symmetrical head shadow across ears due to greater symmetry in speech recognition across ears as well as more similar interaural audibility bandwidth [23, 39, 40, 58, 69, 70]. In a study of 14 bilaterally implanted children, Sheffield et al. [41] showed symmetric head shadow across ears in the range of 20- to 30-percentage points, on average. Further, the head shadow exhibited by this group of bilaterally implanted children was equivalent to the magnitude of head shadow exhibited by bilaterally implanted adults [23, 40]. In contrast, pediatric bimodal listeners have been shown to demonstrate variable head shadow in the range of 0- to 17-percentage points [71, 72] for fixed SNR studies, and approximately 3 dB for adaptive speech receptive thresholds [61] with head shadow present only for conditions with noise directed to the poorer ear. Consequently, preferential seating is more critical for bimodal listeners as compared to bilateral CI recipients as bimodal listeners are more likely to have significant asymmetry in functional speech recognition across ears. In summary, bimodal listeners exhibiting significant interaural asymmetry in speech recognition would likely be better served with a second CI to achieve greatest binaural summation and equivalent head shadow across ears—the latter of which contributes to greater benefit from spatial release from masking which will be less dependent upon speech and noise locations [58].

Speech Recognition in Quiet and Noise: Cross-Sectional Studies of Bimodal and Bilateral CI Users

Deep et al. [59] reported speech recognition outcomes for 88 pediatric CI recipients (57 bimodal and 31 sequential bilateral CI). Their between-subjects analyses revealed no difference between bimodal and bilateral CI listeners on tasks of word recognition or sentence recognition in quiet or co-located noise. Choi et al. [31] described word recognition in quiet and in multi-talker babble for 32 pediatric CI recipients (19 bimodal and 13 bilateral CI users). Similarly, they found no between-group differences in word recognition in quiet or noise for most listening conditions including quiet, co-located speech and noise, and conditions for which noise

was directed to either the poorer ear or both ears simultaneously; however, bilateral CI recipients exhibited significantly better word recognition in noise for conditions in which babble was directed toward the better hearing ear with the bilateral CI group outscoring bimodal listeners by nearly 30-percentage points, on average. The trends reported by Choi and colleagues (2018) as well as Ching et al. [73] are similar to cross-sectional studies with adult bimodal and bilateral CI users such that these two groups achieve similar outcomes for most listening conditions, except for configurations in which noise is directed to the better hearing ear [23] or when the target speech is directed to the poorer hearing ear [58].

Davidson et al. [30•] completed a cross-sectional investigation of word recognition in quiet and noise and suprasegmental perception (e.g., emotion identification, talker and stress discrimination) for a group of 117 pediatric CI users (29 bimodal, 65 sequential bilateral, and 23 simultaneous bilateral). Though they did not perform between-subjects analyses, hierarchical regression was completed to investigate the relationship between various listener variables on speech perception. Children with longer HA use and lower (i.e., better) unaided audiometric thresholds (≤ 73 dB HL) achieved better word (segmental) recognition and suprasegmental perception than children with higher (i.e., poorer) unaided thresholds. This finding cannot necessarily be interpreted as pediatric bimodal listeners exhibiting better speech perception for segmental and suprasegmental features. The reason is that the regression did not include participant group as an independent variable; rather the regression included best ear pure tone average (PTA for 2nd CI ear for bilateral recipients and non-CI ear PTA for bimodal listeners) representing a continuum for all 117 participants. In fact, 14% of the 65 sequential bilateral recipients and 38% of the 29 bimodal listeners had unaided PTA ≤ 73 dB HL. The effect of HA use was only significant for children with the best unaided audiograms—specifically with unaided PTA ≤ 73 dB HL. Thus, this finding can only be interpreted that some period of bimodal stimulation was beneficial for segmental and suprasegmental speech perception; however, this bimodal benefit was only noted for children with the best unaided audiometric thresholds.

Speech Recognition in Quiet and Noise: Within-Subjects Comparison of Bimodal and Bilateral CI Users

Up to this point, all descriptions of pediatric bimodal versus bilateral CI outcomes have been focused on between-subjects analyses in cross-sectional studies. Between-subjects comparisons can be problematic due to differences in patient selection and degree of residual hearing across groups. For example, children receiving bilateral CIs generally have poorer unaided audiometric thresholds [30•, 74], which potentially confounds data analysis, interpretation, and clinical application of findings for between-group comparisons. There is, however, a

recent study that reported bimodal and bilateral CI outcomes using between-subjects analyses, as well as a repeated-measures, within-subjects cohort. Deep et al. [59] completed a retrospective review of 579 pediatric CI recipients and identified 88 pediatric bimodal listeners, for which 31 (57%) pursued a second CI. Their between-subjects results are described in previous paragraphs and consistent with other studies showing no significant difference between bimodal and bilateral CI conditions for clinical measures of speech recognition and significant interaural asymmetry in speech recognition for bimodal as compared to bilaterally implanted children. Their within-subjects analyses on clinical measures of speech perception revealed no differences between bimodal and bilateral CI performance for the 31 sequentially implanted children, suggesting that the children showed minimal bilateral CI benefit on clinical measures of speech perception as compared to their previous bimodal listening configuration—an effect which is consistent with the adult literature examining within-subjects effects for clinical measures [35, 51, 55, 75]. However, Deep and colleagues [59] also showed that the 31 sequential bilateral CI recipients closed the gap in interaural asymmetry that they had exhibited in their bimodal hearing configuration—a finding that holds potential for significantly greater and symmetric head shadow across ears and potentially greater spatial release from masking (irrespective of signal and noise source location). Additional investigation is warranted to prospectively and longitudinally investigate the benefits afforded by bilateral cochlear implantation as compared to bimodal hearing for children in various auditory scenarios.

Neurophysiologic, Perceptual, and Subjective Consequences of Interaural Asymmetry

The general consensus across studies is that on most measures of speech recognition for bimodal or bilateral CI listeners in their best “two-eared condition,” both adults and children achieve roughly equivalent speech recognition outcomes for words, sentences in quiet, and sentences in co-located noise. A consistent trend is also that bimodal listeners tend to exhibit significant interaural asymmetry in speech recognition, an effect that deleteriously impacts head shadow with noise to the better ear as well as spatial release from masking and roving speech perception. Because CIs provide consistent audibility from approximately 200 through 8000 Hz resulting in greater symmetry in interaural audibility bandwidth, speech recognition performance across ears, and better localization abilities, many children utilizing a bimodal hearing configuration are simply not optimized for audibility and auditory resolution across ears. Consistent interaural asymmetry resulting from bimodal listening or sequential implantation with long inter-implant delay have been shown to result in aural preference syndrome [44, 45, 50••]. Aural preference syndrome describes asymmetrically driven auditory neuroplasticity for which

children exhibit subjective, perceptual, and neurophysiological “preference” for the better hearing ear—generally the CI ear for bimodal listeners and the first CI ear for sequentially implanted bilateral CI users. This is a well-known phenomenon first described in the auditory physiology literature demonstrating developmental changes in auditory cortical overrepresentation of the better hearing ear following unilateral auditory deprivation [76, 77] or asymmetric auditory stimulation [78–80]. Research has shown lasting effects in brainstem and cortical activity (neural activation area, response amplitude, and processing latency) for children who retain bimodal hearing with interaural asymmetry in performance and audible bandwidth [44, 46, 50••, 81] as well as for children receiving sequentially placed bilateral CIs with interaural surgical delays exceeding approximately 12–18 months [43, 44, 47].

It is possible for sequential bilateral CI recipients to largely overcome the effects of aural preference and interaural asymmetry with inter-implant delays up to 3–4 years [50••, 81–83]; however, the timing of the first CI is also critical with < 4 years for the first CI coupled with < 4 years inter-implant delay resulting in the least interaural asymmetry in speech recognition [83]. This is not to say that children receiving sequential bilateral CIs with larger inter-implant delays—including those who may have received their first CI after 4 years of age—do not benefit from bilateral implantation; rather, greater delays are simply more likely to result in asymmetries in interaural performance [43, 50••, 82–86], auditory neural processing [44, 81], and subjective aural preference [45, 83].

Auditory-Based Language Development: Bimodal Hearing Vs. Bilateral CI

It is not known whether aural preference syndrome results in protracted language development compared to children with bilateral CI with symmetrical interaural function. This point holds high potential for clinical and educational impact as many pediatric CI users continue to display persistent speech, language, and reading difficulties despite early implantation and early intervention [87–89]. In this last section, we will review the current literature with respect to auditory-based language development for pediatric bimodal and bilateral CI recipients.

There are a number of studies demonstrating significant benefit of bilateral over unilateral CI, without use of a contralateral HA, for language development [90, 91]; however, Nittrouer and Chapman [92] were the first to describe language outcomes for 58 children with various CI configurations including 15 unilateral CI users (without contralateral HA), 17 bimodal listeners, and 26 sequential bilateral CI users. The three groups were well controlled for age at first CI, socioeconomic status, and early intervention. Nittrouer and Chapman [92] reported on norm-referenced assessments of receptive language and expressive vocabulary as well as

measures of generative language (mean length of utterance (MLU) and number of pronouns used) obtained from video recorded naturalistic language samples. Comparisons of language outcomes at 42 months of age revealed no differences in receptive language, expressive vocabulary, or generative language across the three CI groups. However, when the 58 children were grouped by whether or not they bimodal hearing experience—which included all bimodal and many sequential bilateral CI users—the children with some bimodal experience significantly outperformed those who only had electric hearing experience on both measures of generative language. The authors theorized that acoustic stimulation, even for an ear with severe-to-profound hearing loss, offered better spectral resolution as compared to electric hearing, including F0 and temporal fine structure allowing for development and resolution of suprasegmental speech features, such as prosody, as well as better spectral representation of voiced formants. All children in this study had better ear PTA ≥ 70 dB HL; however, it is not clear whether previous bimodal experience was associated with lower (i.e., better) PTA in the better ear.

Ching et al. [73] investigated language, articulation, and speech recognition outcomes for a large cohort of 61 bimodal and 61 bilateral CI listeners who had been enrolled in the Longitudinal Outcomes of Children with Hearing Impairment (LOCHI) study. Norm-referenced assessments of receptive and expressive language and speech production (articulation) were completed at 3 years of age. They found no significant differences between bimodal and bilateral CI users for receptive or expressive language, receptive vocabulary, and speech production (articulation). However, there was no description of audiometric thresholds in the non-CI ear for the bimodal listeners and thus it is unclear how much access acoustic amplification provided for the bimodal group.

Nittrouer and colleagues have since published a number of studies describing longitudinal language, phonological awareness, and literacy for children using bimodal stimulation and bilateral CIs. In a group of 35 children with hearing loss (27 with at least one CI and 8 bilateral HA) and 17 children with normal hearing in Kindergarten (mean age 6.6 years), Nittrouer et al. [93] demonstrated that children with hearing loss achieved significantly poorer outcomes on measures of phonological awareness and literacy (word reading and reading comprehension) than children with NH. Children with CI tended to exhibit poorer phonological processing and literacy outcomes than children with bilateral HA; however, this did not reach statistical significance. They also completed a separate analysis grouping CI recipients by whether or not they had a period of bimodal stimulation revealing that children with previous bimodal experience significantly outperformed children with CI-only hearing for all measures of phonological processing and receptive language, but not for expressive vocabulary. As explained in their 2009 study, the authors theorized children with previous bimodal experience had better

spectral resolution from the acoustic hearing ear allowing better lexical and phonotactic representation of speech thereby affording higher phonological awareness, receptive language skills, and reading abilities. Though all children had better ear PTA ≥ 70 dB HL, as stated previously, it is unclear whether PTA viewed as a continuous variable may have at least partially explained this effect.

Moberly et al. [74] prospectively investigated language, phonological awareness, and literacy in a group of 48 children with CIs upon completing 2nd and 4th grade. Of the 48 children with CI, 32 had received bilateral CIs prior to entering 4th grade (29 sequential bilateral). For the 29 sequentially implanted bilateral CI users, 24 had continued use of a contralateral HA prior to receipt of the 2nd CI. Of the remaining 24 children in the 48-participant sample, none had previous bimodal experience meaning that they either had a single CI, simultaneous bilateral CI, or discontinued HA use following the 1st CI. This study did not involve a controlled assignment approach to bimodal or bilateral CI, as there are ethical concerns surrounding such an investigation. Rather, this rigorously controlled study of children receiving 1 or 2 CI who may or may not have had a period of bimodal stimulation, allowed for an investigation of natural factor variation in auditory and language outcomes resulting from different clinical recommendations and audiologic intervention. Thus in this study, the experimenters mainly investigated between-group effects for which groups were stratified by previous bimodal experience versus those who never had bimodal experience—the latter of which included some unilaterally implanted children. They found that the group with previous bimodal experience significantly outperformed the group with electric-only hearing on measures of expressive language, phonological awareness, and word reading abilities at both time points. Post hoc analyses revealed that for children completing 2nd grade, the bilateral CI recipients with bimodal experience outperformed the unilateral CI users with bimodal experience on measures of phonological awareness; however, by the 4th grade, this gap between the two groups had been closed. To further investigate this effect noted at the 2nd grade, the authors characterized phonemic awareness standard scores in the 2nd grade as a function of better ear PTA. At the group level, they found no significant correlation between phonemic awareness and better ear PTA for the group of children with or without bimodal hearing experience; however, excluding the 3 children with the best PTA (≤ 70 dB HL) who scored more than 2 standard deviations *below* age-normative performance, there was a clear trend between better ear PTA and phonemic awareness warranting further investigation with larger samples.

Nittrouer and colleagues [94•] later reported auditory, language, and literacy outcomes in this same cohort in 6th grade (mean age = 12.4 years). Their previous findings held consistent such that (1) children with hearing loss demonstrated

significantly poorer outcomes on all measures of language, phonological awareness, and literacy as compared to children with NH, (2) there were no differences in outcomes between bimodal and bilateral CI users and (3) irrespective of CI status, prior bimodal experience was associated with higher outcomes on measures of phonological awareness and language (receptive language and expressive vocabulary). Without additional information regarding the potential effects of unaided PTA as a continuous variable and a clear distinction between unilateral CI users without HA use, bilateral CI users, and bimodal listeners, it is unclear whether the benefits afforded by early bimodal experience is limited to children with the lowest (i.e., best) better ear PTA.

Davidson et al. [30•] investigated speech perception as well as norm-referenced assessments of receptive vocabulary and language for a group of 117 pediatric CI users (29 bimodal, 65 sequential bilateral, and 23 simultaneous bilateral). They completed hierarchical regression to determine the relationship between various listener variables, including segmental and suprasegmental speech perception, on receptive vocabulary and language. In addition to nonverbal IQ and maternal education, they found that suprasegmental speech perception (e.g., emotion identification, talker and stress discrimination) was significantly related to receptive language skills. For receptive vocabulary, they found that in addition to nonverbal IQ and maternal education, both segmental and suprasegmental speech perception were significantly related. As mentioned previously, children with longer HA use and lower (i.e., better) unaided audiometric thresholds (≤ 73 dB HL) achieved higher word (segmental) recognition and suprasegmental perception [30•]; however, this cannot necessarily be interpreted as bimodal listeners achieving higher speech perception and hence superior receptive vocabulary and language outcomes. The reason is that the regression did not include *participant group* as an independent variable. Rather the regression included best ear pure tone average (PTA for the 2nd CI ear for bilateral recipients and non-CI ear PTA for bimodal listeners) representing a continuum for all 117 participants. Pooling their speech perception and language outcomes together, Davidson et al. [30•] reported the benefits of having *some experience with bimodal stimulation*, even if only a few months prior to receiving a 2nd CI, for speech perception (both segmental and suprasegmental), receptive vocabulary, and receptive language—a finding that was consistent with several other studies [74, 92, 93, 94•]. However, Davidson et al. [30•] also showed bimodal hearing benefit was only effective for children with the *best unaided audiometric thresholds* and the benefits derived seemed to plateau after 3–4 years of bimodal experience. What we can interpret from these findings is that (1) children with better-ear thresholds ≥ 73 dB HL are more likely to achieve better auditory and language outcomes with *early bilateral cochlear implantation*, and (2) children with better-ear thresholds ≤ 73 dB HL could

still obtain benefit from sequential bilateral cochlear implantation, but these children will derive significant auditory and language benefit from early bimodal experience. The most important finding from this paper was that for the vast majority of children receiving cochlear implants (i.e., better-ear thresholds ≥ 73 dB HL), early bilateral cochlear implantation would result in best auditory and language outcomes.

Summary and Conclusions

We have much to learn about what constitutes the best “two-eared” listening configuration for children with bilateral moderate-to-profound SNHL as well as the optimum timing of intervention, particularly as related to sequential bilateral implantation. As described here, it is important to consider individual ear performance as interaural asymmetry in performance and audibility bandwidth is associated with aural preference syndrome. Aural preference syndrome includes asymmetric auditory development, little-to-no binaural summation, minimal spatial release from masking, greater reliance on preferential seating, and poorer localization. The current literature supports the recommendation of sequential bilateral cochlear implantation for children with better-ear thresholds consistent with a severe or poorer hearing loss—following a period of bimodal stimulation—to achieve highest outcomes for speech recognition (segmental and suprasegmental) as well as receptive vocabulary, receptive language, and expressive/generative language. Indeed, it is quite possible that bilateral cochlear implantation is the best intervention option even for children with less severe bilateral hearing losses [7, 8]. Further research is warranted including large multi-center prospective studies allowing for within-subjects analyses of sequential bilateral CI recipients as well as between-subjects comparisons to children retaining bimodal hearing. Furthermore, we have a great need for the identification of clinically feasible, evidence-based measures that provide clinical guidance for the determination of bimodal versus bilateral CI candidacy. Researchers are investigating the possibility of using objective measures as well as the development of audiometric criteria regarding interaural symmetry in audibility for our youngest patients and including speech recognition and subjective reports for older children. Though outside the scope of the current review, we must also consider the possibility for *bilateral cochlear implantation with acoustic hearing preservation*. There is a growing population of adults and pediatric CI recipients with acoustic hearing preservation in the implanted ear(s) who are successfully combining electric and acoustic stimulation. This intervention offers the best option for the developing auditory system providing F0 and temporal fine structure via acoustic hearing in one or both ears and greater interaural symmetry in audibility bandwidth and speech perception with bilateral implants. We will likely see an

increasing number of peer-reviewed reports for this growing population in the years to come.

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Compliance with Ethical Standards

Conflict of Interest The author is a consultant for Advanced Bionics, Cochlear, and a member of the clinical advisory board for Frequency Therapeutics.

Human and Animal Rights and Informed Consent This article does not contain any studies with human or animal subjects performed by any of the authors.

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- Of importance
- Of major importance

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