Cochlear Implantation for Unilateral Hearing Loss

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KEYWORDS

• Pediatric • Unilateral hearing loss • Cochlear implantation • Single sided deafness

KEY POINTS

- Cochlear implantation in the pediatric severe to profound unilateral hearing loss patient allows for significant improvement in speech recognition in quiet and noise as well as sound localization.
- Outcomes in cochlear implantation in pediatric patients with severe to profound unilateral hearing loss may be impacted by etiology, duration of deafness, and family/patient motivation.
- Additional research is required to determine optimal timing and device choice to allow for maximal outcomes.

INTRODUCTION

Treatment of pediatric unilateral hearing loss (UHL) has changed radically over the past few decades. Before the implementation of newborn hearing screening the average age of diagnosis for UHL was more than 8 years old.[1] As late as the 1970s, the predominant management strategy was reassurance—informing parents UHL has no negative consequences.[2] Research in the 1980s began to shed light on the significance of UHL. Thirty-five percent of children with UHL failed 1 or more grades and up to 60% required special educational services.[3–6] Children with UHL were also noted to have more behavioral issues than their peers with normal hearing, including social withdrawal, aggression, and difficulties with interpersonal and social adjustment.[3,7,8] More recent research has suggested that children with UHL may lag behind in terms of speech and language and cognition as well.[8–10]

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As part of the Americans with Disabilities Act, the 504 plan ensures that children with UHL receive accommodations to improve access to school curriculums and improve their academic success. Educational support for children with UHL typically involves preferential seating as well as a remote microphone system to minimize the impact of background noise. A hearing aid is another essential tool for the child with UHL who has hearing loss within the range to benefit from traditional amplification. Amplification is usually beneficial for UHL of mild to moderate degree affecting the speech frequencies (500 Hz to 4 kHz).

Children with at least a severe UHL in the speech frequencies may not benefit from traditional amplification. Amplification of greater degrees of hearing loss may not make speech audible, or the perception of speech may be very poor (limited recognition of word and sentences with hearing alone). Children initially benefitting from unilateral amplification may experience progression of hearing loss and no longer benefit.[11] Children with UHL who are unable to benefit from traditional amplification typically have a moderate-to profound loss in the affected ear impacting the speech frequencies (500 Hz to 4 kHz). When hearing loss exceeds these levels, acoustic amplification results in distortion of the speech signal and poor overall speech perception. Children with this degree of UHL and normal hearing in the opposite ear are often referred to as having single-sided deafness (SSD). Treatment options for this population have historically been limited to contralateral routing of signal or bone conduction hearing devices. However, these interventions do not allow for binaural hearing, which is important for auditory development and hearing in dynamic environments.[3,12,13]

There are a number of advantages of hearing from 2 ears, especially in complex listening environments. Stimulation of both auditory pathways allows the brain to take advantage of head shadow, binaural squelch, and binaural summation.[14] These phenomena are best observed when the speech signal and noise are spatially separated, as illustrated in **Fig. 1**. When speech and noise are collocated in front of the child, binaural hearing allows for binaural summation. This improves the determination of loudness and allows for an increase of up to 3 dB in signal-to-noise ratio and up to a 28% improvement in speech perception.[15] When the masker is moved to the better hearing ear, or the left ear in the **Fig. 1** example, the head creates a barrier that results in a decrease in the noise signal on the side of the poorer hearing ear. This is known as the "head shadow effect" and allows for an improved signal-to-noise ratio at the ear contralateral to the noise source. In those with normal hearing bilaterally, the head shadow effect results in improved speech perception as compared with the collocated condition. In the case of SSD, the child would be unable to access the improved signal-to-noise ratio on the poorer hearing side without access to bilateral hearing and the signal is dominated by the noise. Binaural squelch is a more complicated process that allows central auditory pathways to use timing and phase differences in the signal from each ear to improve spatial hearing.

Binaural hearing also allows for sound source localization. Sound arrives with greater amplitude and earlier timing at the ear closest to the signal source. The brain uses these cues for comparison between ears to localize sound, using low frequencies to compare the timing differences and the higher frequencies to compare level differences.[14] These mechanisms are completely dependent on binaural information.

In 2019, the US Food and Drug Administration approved the MED-EL cochlear implant (CI) system for adults and children age 5 years and older with SSD. This is the first CI system approved for this indication in the United States. Cochlear implantation is the first and only treatment option to provide binaural hearing to children with SSD. Positive results are emerging from ongoing clinical trials and retrospective

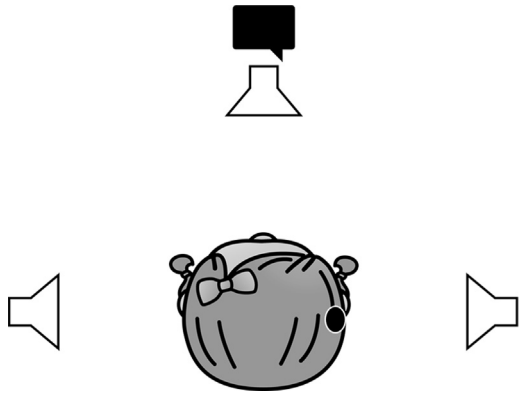

Fig. 1. Spatial hearing in noise. Speech is presented in front (black speech bubble) and the noise signal (white speaker) can be collocated with the speech in front of the listener, directed to the normal hearing (*left*) ear, or directed to the poorer hearing (*right*/implanted) ear. The head shadow effect can be observed here by noting that the implanted ear (*right*) benefits from the physical presence of the head creating an advantageous signal-to-noise ratio (SNR) when noise is directed to the better hearing ear when a CI is in place but not when a CI is off. The head decreases the amount of noise reaching the right ear leading to an improved signal to noise ratio for that ear.

studies have indicated that cochlear implantation in children with SSD is an effective treatment option.[16]

CRITERIA FOR CANDIDACY

Etiology of Hearing Loss

The etiology of the hearing loss has an impact on candidacy. Etiologies of pediatric UHL are more often unknown because there is no comprehensive genetic test battery for UHL, as for bilateral sensorineural hearing loss (SNHL). However, unlike bilateral SNHL, cochlear nerve deficiency (CND) is common in congenital SSD. Therefore imaging, in particular MRI, to directly visualize the eighth nerves, is essential to determine implant candidacy in this population. Other etiologies of significance include congenital cytomegalovirus (CMV) and cochlear malformations. The potential impacts of these conditions are outlined elsewhere in this article.

Cochlear nerve deficiency

CND refers to hypoplasia or aplasia of the cochlear nerve branch of the eighth nerve. CND is the most common etiology for pediatric congenital profound UHL with a reported incidence of 26% to 58%.[17–19] Pediatric CI recipients with bilateral CND have poorer CI outcomes compared with other etiologies. Development of open-set speech perception is not predictable and is often very slow to emerge in this population.[20] The use of CI despite limited benefit in bilateral CND may be warranted because these children otherwise have no access to sound. However, providing children with 1 normal hearing ear with a severely impoverished and distorted auditory signal in the opposite ear is likely to provide a poorer perception of speech. In this situation, the advantages of binaural hearing are not expected, and nonuse is likely. Thus, cochlear implantation is not recommended for individuals with UHL secondary to CND.[17] Ruling

out CND with high resolution 3-dimensional MRI is an essential part of CI candidacy evaluation for children with SSD.

Congenital cytomegalovirus infection

Another common cause of pediatric UHL is congenital CMV infection.[17,19] UHL owing to congenital CMV can be progressive and eventually become bilateral, impacting both treatment of the deaf ear and follow-up of the better hearing ear. With the significant risk of progression, children with SSD owing to congenital CMV should be considered for timely CI.[21]

Additional etiologies

Other potential etiologies for congenital or progressive UHL include inner ear malformations, trauma, meningitis, auditory neuropathy spectrum disorder, sudden idiopathic hearing loss, labyrinthitis, and ototoxic medications, as well as neurologic, syndromic, and other unknown causes. In regard to inner ear malformations, the most common is an incomplete partition type 2 followed by a narrowed internal auditory canal and enlarged vestibular aqueduct.[22] Although children with incomplete partition type 2 and/or an enlarged vestibular aqueduct can perform as well as children with normal cochleae, severely malformed cochlea may also limit functional outcomes and therefore are not suited for implantation in children with SSD.[23]

Duration of Deafness

Early implantation, thus minimizing the period of auditory deprivation, is associated with better speech perception of children with bilateral SNHL who receive CIs. Evidence in the SSD population also supports an optimal window for intervention. Those with SSD have the unique issue of aural preference syndrome, a reorganization of the central auditory pathways for the dominant hearing ear.[24] To prevent this central preference and to allow for a good binaural hearing, outcome data suggest that implantation should be within the first 5 years of life. Evidence supporting this window includes histopathology and MRI studies examining central auditory pathway myelination and plasticity.[25–27] Case studies of cortical evoked potential changes after CI in children with SSD have also supported these findings by noting improvements in cortical reorganization in children with less than 5 years of deafness.[28,29]

Some retrospective case series of children with congenital SSD have demonstrated poorer speech perception outcomes when the duration of deafness is prolonged.[20,30,31] The authors of these works recommend implantation at less than 3 or 4 years of age to encourage maximal CI outcomes in children with SSD.[30] However, there are case reports indicating positive CI outcomes in cases of prolonged congenital SSD.[32] Given the small number of published reports involving CI in children with SSD, and variables such as programming techniques, electrode length, consistency of device use, and postimplant habilitation, which may impact outcomes, the age range within which significant benefit may still occur remains undefined.

Family and Patient Factors

Parents of young children with SSD are less likely to pursue CI than those with children with bilateral SNHL.[33] The decision-making process is fundamentally different because children with SSD develop spoken language and learning delays may not be apparent until they reach elementary school. Therefore, the benefits of binaural hearing, and not mode of communication, are weighed against the risk of surgery, the use of an external device, and the need for lifelong audiologic care.

For families that pursue CI, their commitment to ensure their child consistently use the device is critical to performance.[34,35] Unlike children with bilateral CIs, those with

SSD may struggle, but can still communicate with spoken language without their device. Therefore, the need for consistent device use may not be as clear or compelling to the family, and the risk of device nonuse is increased.

Older children with SSD have been reported to be at increased risk to become nonusers.[31,36] Therefore, it is important that older children be involved in the decision-making process and be willing to commit to device use.

OBSERVATIONAL OUTCOMES OF COCHLEAR IMPLANTATION FOR SINGLE-SIDED DEAFNESS

Word Recognition in Quiet

Unlike other hearing interventions, cochlear implantation allows for speech perception in the ear with SSD. Overall, the literature demonstrates the potential of a CI to improve word recognition. Implanted children with SSD, particularly those with a shorter duration of unilateral deafness, demonstrate a significant improvement in their word recognition scores in the CI-only condition, with scores as high as 90%.[17,31,35–38] Children with longer durations of deafness, ranging from 4 to 13 years often have poorer word recognition outcomes.[31,35,36] However, isolated word recognition is not a measure of the real-world impact of CI on the daily lives of children with SSD. These impacts would include listening effort, localization of sound, hearing in noise, and academic and social skills.

Sound Source Localization

One of the principal benefits of binaural hearing is sound source localization. Studies have demonstrated a significant improvement in sound localization in the "CI on" condition in the majority of CI recipients with SSD.[17,32,39] Localization abilities tend to correlate with speech perception outcomes, although there have been some patients who demonstrate improvement in sound localization with limited or no speech perception in quiet in the implanted ear.[17,35] Improved sound localization has been seen as early as 1 month after CI activation with benefit maintained long term.[35] Parental surveys of auditory behavior also have demonstrated subjective improvement in localization and spatial hearing.[17,32]

Speech Perception in Noise

In children, the ability to hear in dynamic listening environments, which include varying degrees of background noise, is paramount to learning. Speech perception testing in noise is important because it permits the measurement of a binaural advantage, particularly when speech and noise are separated in their presentation (**Fig. 1**). Separating the signals allows for the evaluation of binaural summation, binaural squelch, and the head shadow advantage. Binaural summation can be measured by comparing speech perception scores with unilateral versus bilateral hearing when speech and noise are collocated in front of the listener. Studies have demonstrated summation benefits in the collocated condition among children with UHL and CI.[30,35,40,41] Similarly, head shadow benefit can be calculated by comparison of unilateral versus bilateral conditions (CI on vs CI off) when the speech signal is in front of the listener and the noise is directed to the better hearing ear. Children with SSD who use a CI have shown evidence of benefit from head shadow.[17,32,37,40,41] Benefit from the squelch effect can be measured with by comparing unilateral and bilateral speech perception outcomes obtained with speech in front of the listener and noise at the poorer hearing ear. Evidence of squelch is mixed in adult studies, with some noting no benefit in CI users with SSD[42,43] and others noting benefit after 1 or more years of CI use.[42–45] Small amounts

of binaural squelch benefit may be measurable in some children with SSD who use CI for more than 1 year.[41]

Some studies of speech perception in noise support duration of deafness and etiology as influencing outcome, whereas others do not.[17,30,32,40] Interestingly, similar to localization abilities, some children with SSD and CI who do not have measurable open set word recognition still report subjective improvement in hearing in noise.[35] In general, a comparison of speech perception in spatially separated speech and noise with CI on versus CI off has demonstrated that a CI does not interfere with overall speech understanding, regardless of performance with the CI alone, alleviating concerns regarding binaural interference.[36]

Device Use

Device use is critical to improvement and benefit from a CI. This point has been demonstrated clearly in individuals with bilateral SNHL. Most pediatric SSD CI studies report full time use in more than 75% of children with a minority of recipients as nonusers.[9,17,30,31,36,40] Reasons cited for device nonuse include a subjective lack of improvement with device use, nonauditory stimulation, poor family support, stigmatization and negative attention as a result of device use, and older implantation age.[17,36] Counseling is an important tool to establish realistic expectations and encourage full-time use.

Speech and Language Development

Although children with SSD develop spoken language, there is evidence of impact upon language and academics in these children in comparison to children with normal hearing.[9] One longitudinal study compared 6 children with congenital SSD, implanted between ages 8 and 26 months, with normal hearing peers and with unimplanted children with congenital SSD.[9] Language comprehension, expressive vocabulary, morphosyntactic knowledge, and cognitive skills were examined. The SSD CI group performed similarly to the normal hearing group, whereas approximately 50% of the nonimplanted children with SSD performed lower than the normal hearing group in each category.[9] This study supports the concept that CI may enable development of language skills and cognitive milestones in children with SSD equivalent to normal hearing children.

SUBJECTIVE OUTCOMES OF COCHLEAR IMPLANTATION FOR SINGLE-SIDED DEAFNESS

Speech, Spatial, and Qualities of Hearing Scale

The Speech, Spatial, and Qualities of Hearing Scale is a self-reported measure with a modified version for parental report to evaluate subjective hearing abilities in 3 separate areas sensitive to binaural hearing.[46] Studies of the impact of CI on children with SSD have demonstrated consistent improvement in all 3 areas evaluated by the Speech, Spatial, and Qualities of Hearing Scale survey, with the greatest improvement in spatial hearing.[17,31,39,40,47] High performers tend to show correlation between Speech, Spatial, and Qualities of Hearing Scale scores and behavioral CI measures including word recognition scores, sound localization, and daily CI use.[17]

Tinnitus

Tinnitus, one of the original reasons for cochlear implantation in adults with SSD, has also been examined in the pediatric population. The incidence of tinnitus in children with a CI is estimated to be 38%.[48] Zeitler and colleagues[38] reported on the impact

of CI on tinnitus in children with SSD. The authors found that 50% experienced partial suppression and 50% complete resolution while using a CI.[38]

Academic and Personal Performance

SSD also impacts children's academic and personal well-being. In 1 study, a parental survey on a nonvalidated questionnaire demonstrated positive behavior change attributed to CI was noted by parents of one-half of the implanted children, as well as improvement in mental serenity and tranquility in 28%.[40] Improvement in peer interactions after implantation has also been reported on parental survey.[32] Children with SSD with little to no speech perception in quiet with the CI side only have also reported improvement in quality of life as well as academic performance both on parental and child report, likely related to the improvements still noted in sound localization as well as speech perception in noise.[35]

Child and Family Satisfaction

Parental and child satisfaction after implantation has been reported to be favorable in several studies. One study reported that 84.2% of parents would select cochlear implantation again if given the choice.[40] Another study reported that all children in the cohort were very satisfied with their decision to undergo implantation.[32]

DEVICE CONSIDERATIONS

There are theoretic reasons why implantation of longer electrodes may be beneficial for implanted patients, particularly those with SSD. Longer electrode arrays, which extend into the apex of the cochlea, allow for improved frequency–place matching when programming a CI by representing a larger range of frequencies in the cochlea.[49,50] Frequency–place matching refers to stimulation of the cochlea at sites that are appropriate for the desired frequency based on the normal tonotopic organization of the cochlea. The more accurate frequency–place matching, the more closely electric hearing may approximate hearing in the normal ear, thereby hastening the integration of binaural hearing of 2 different types of auditory stimulation.[51] This factor may decrease the brain remapping necessary for the integration of electric and acoustic hearing. Rapid integration soon after device activation may aid in achieving device acceptance and consistent use, which may be especially advantageous in children. Additionally, longer electrode arrays have also been shown to improve spatial hearing.[52] Studies of the impact of frequency–place mapping specifically on SSD outcomes are needed. However, in light of current knowledge, longer electrode length should be a consideration when implanting children with SSD.

FUTURE DIRECTIONS

The current pediatric SSD and CI literature primarily consists of retrospective reviews of small series of CI recipients. Given the significant heterogeneity of the population in terms of length of deafness, etiology, cognition, and CI device/electrode, well-designed prospective clinical trials are needed to understand best practices to optimize outcome, potential benefits and range of outcome.

SUMMARY

Cochlear implantation in the pediatric patient with SSD can result in significant objective improvements in speech perception in the deafened ear, speech recognition in noise, and localization, as well as subjective improvements in tinnitus, speech and

language development and academic performance. Eligible patients should have a normal cochlear nerve. Candidacy considerations should include etiology of deafness including risk of future loss in the normal hearing ear, duration of deafness, and the child and family's commitment to cochlear implantation.

CLINICS CARE POINTS

- Improvements that can be expected from cochlear implantation in the child with SSD include better speech perception in quiet and noise, sound localization, speech and language development, and quality of life.
- Children with SSD owing to CND are not candidates for CI. Nerve deficiency should be evaluated by high resolution 3-dimensional MRI of the internal auditory canals.
- CI for SSD is of special consideration in children with history of congenital CMV or cochlear malformation that places the child's one normal hearing ear at risk for future hearing loss.
- The duration of deafness, with shorter being more favorable, and patient and family motivation should be considered in the decision for implantation.

DISCLOSURE

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