Speech and Language Outcomes in Adults and Children with Cochlear Implants

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Abstract

Cochlear implants (CIs) represent a significant engineering and medical milestone in the treatment of hearing loss for both adults and children. In this review, we provide a brief overview of CI technology, describe the benefits that CIs can provide to adults and children who receive them, and discuss the specific limitations and issues faced by CI users. We emphasize the relevance of CIs to the linguistics community by demonstrating how CIs successfully provide access to spoken language. Furthermore, CI research can inform our basic understanding of spoken word recognition in adults and spoken language development in children. Linguistics research can also help us address the major clinical issue of outcome variability and motivate the development of new clinical tools to assess the unique challenges of adults and children with CIs, as well as novel interventions for individuals with poor outcomes.

Keywords

cochlear implants, individual variability, speech perception, language development, adverse conditions
1. INTRODUCTION

Cochlear implants (CIs) are “one of the great success stories of modern medicine” (Wilson et al. 2011, p. 117), representing a significant engineering and medical milestone in the treatment of sensorineural hearing loss (SNHL) for both adults and children. CIs serve as the standard of care for adults and children with moderate to profound SNHL who do not receive sufficient benefit from hearing aids. CIs do not restore normal hearing to deaf individuals but rather provide a useful representation of sounds and, crucially, access to spoken language. The efficacy of CIs (i.e., that they work and provide significant benefits) is firmly established, especially when using basic clinical measures of speech recognition outcomes under idealized, highly controlled listening conditions in the audiology clinic or research laboratory. However, it is also clear that there is a tremendous degree of variability and individual differences in the effectiveness of CIs—the degree to which individual CI users experience useful benefits for real-world listening activities, such as listening to a talker in a crowded restaurant, communicating over the telephone, recognizing voices or emotions, enjoying music, or understanding speech produced by a nonnative speaker.

A major challenge in research on CIs is to understand the enormous variability and individual differences in speech and language outcomes in deaf adults and children who have received implants (Pisoni et al. 2018). Why do some deaf adults and children do quite well with their CIs, often achieving near-normal scores compared with typically developing, age-matched, normal-hearing (NH) peers, while other adults and children show significant weaknesses and delays in spoken word recognition and sentence processing? Furthermore, why do some adults and children with CIs do well in the research lab or clinic but display significant difficulties in their everyday lives? Answers to these questions have important clinical implications for improving diagnosis and treatment of both adults and children with hearing loss. Moreover, understanding individual differences in outcomes and specific challenges for adults and children with CIs is critical for developing new methods to identify individuals who may be at high risk for poor outcomes as early as possible, in order to use novel targeted interventions to help them achieve optimal levels of performance and benefits with their CIs.

This article provides a review of CI technology, a description of the adult and pediatric clinical populations who typically receive these devices, and an overview of what CIs can do for these individuals as well as the particular limitations and issues faced by CI users. Our overall goal in this review is to demonstrate the relevance of CIs specifically to the linguistics community. We suggest that CIs are an effective medical treatment for adults and children with moderate to profound SNHL, and that CIs not only give recipients access to sound but, more importantly, also provide access to spoken language. Furthermore, CI users can help us improve our basic understanding of spoken word recognition in adults and spoken language acquisition in children, by exploring impacts of hearing loss and restoration of audibility. Finally, we emphasize that linguistics research can help us address the major clinical issue of outcome variability and also motivate the development of new clinical tools to better assess the unique challenges of adults and children with CIs as well as novel interventions for individuals with poor outcomes.

1.1. What Is a Cochlear Implant, and What Does It Do?

A CI is a surgically implanted device that bypasses the sensory function of the inner ear to directly stimulate the cochlear nerve (Wilson & Dorman 2008). In most forms of SNHL, the main pathology is within the cochlea itself, but the cochlear nerve remains generally intact anatomically and functionally. Thus, the cochlear nerve does not receive adequate stimulation from the cochlea, and delivers an ineffective and impoverished signal to the central auditory system. By directly
stimulating the cochlear nerve, CIs deliver electrical signals to the functional cochlear neurons, providing synchronized stimulation across a broad frequency range.

Although there are several approved CI devices by various manufacturers, all CI hardware systems consist of an external sound processor worn by the listener and a surgically implanted internal device (for a detailed review and overview figure, see Carlson 2020). The external sound processor comprises one or more microphones, a digital signal processor, a battery to power the device, an external signal receiving/transmitting coil, and an external magnet. The microphone(s) captures the acoustic input and converts it into an electrical signal, which is delivered to a preamplifier that amplifies the signal. This signal is analyzed by the sound processor using digital filtering approaches, dividing the signal into different analysis bands (i.e., frequency bands or channels) to permit some degree of frequency-specific delivery of the signal. Additional signal processing may be applied to improve the signal-to-noise ratio for speech. The signal output then typically undergoes rectification and low-pass filtering to code the amplitude envelope for each frequency band. These amplitude envelopes are used to determine the magnitude of electrical stimulation that should be delivered by the implant. The signal processor then converts the processed signal into an electrical signal that is sent to the external coil and across the listener’s skin to the internal implant coil via electromagnetic induction/radio-frequency transmission.

The internal CI hardware consists of the receiving/transmitting coil, an internal magnet, a second digital signal processor, a stimulator to generate electrical pulses, and an electrode array (i.e., multiple electrode contacts for current multichannel devices). Once the internal coil receives the signal from the external processor, it is again analyzed by the internal digital signal processor. The stimulator produces biphasic electrical pulses, typically at a fixed moderate to fast rate, and the amplitude of these pulses is modulated on the basis of the amplitude of the signal envelope at each frequency band. Amplitude-modulated electrical pulses are then delivered to individual electrode contacts on the electrode array that have been surgically positioned within the cochlea. Multichannel CIs attempt to capitalize on the tonotopic frequency-specific organization of the cochlea, delivering high-frequency signals to the electrodes positioned near the basal end of the cochlea and low-frequency signals to more apical electrodes.

1.2. Who Receives a Cochlear Implant?

The vast majority of adults who receive CIs are postlingually deafened; their SNHL progresses to the point of meeting CI indications after having previously developed normal spoken language skills. The greatest growth in application of cochlear implantation is in adults aged 60 years and over (Fakurnejad et al. 2020). The increase in CI patients is thought to reflect the growth of the aging population as well as the increasing prevalence of SNHL with advancing age (i.e., presbycusis). Other postlingual adult CI candidates may demonstrate bilateral moderate to profound SNHL as a result of noise exposure, Ménière’s disease, ototoxic medications, autoimmune inner ear disease, meningitis, or trauma (Blamey et al. 2013). CIs are also gaining use in individuals with pathology of the cochlear nerve, such as auditory neuropathy spectrum disorder and tumors affecting the cochlear nerve (e.g., acoustic neuroma/vestibular schwannoma) (Bartindale et al. 2019, Shearer & Hansen 2019). A smaller proportion of adult CI candidates can be considered pre- or perilingual deaf, meaning that their hearing loss began before development of spoken language skills was complete. Although outcomes in these adults are often poorer and less consistent, some of them demonstrate improved open-set speech perception, audiovisual speech perception, and hearing-related quality of life after implantation (Debruyne et al. 2020).

Although early CIs were single-channel devices implanted as aids to support lip-reading abilities in profoundly deaf adults in the 1970s, advances in device technology that support greater
benefits to spoken language communication have led to a gradual broadening of indications (Sladen et al. 2017). Since 1985, the US Food and Drug Administration (FDA) indications for adult cochlear implantation have evolved beyond bilateral profound SNHL with 0% open-set sentence recognition. Official indications from 2005 (Cent. Medicare Medicaid Serv. 2005) are bilateral, moderate to profound SNHL with limited benefit from amplification, meaning scores of 50% (or fewer) words correct on open-set sentence recognition in the ear to be implanted (and worse than 60% in the best-aided condition). The Minimum Speech Test Battery (MSTB) for adults serves as a best-practice recommendation for evaluating CI candidacy and outcomes (Luxford 2001). Initially, two measures of speech recognition were recommended for clinical use, specifically the Consonant-Nucleus-Consonant (CNC) isolated word recognition test (Peterson & Lehiste 1962) and the Hearing in Noise Test (HINT) (Nilsson et al. 1994). The HINT is an open-set sentence recognition test that includes lists of meaningful, simple English sentences spoken by a single male talker in a clear speaking style. However, as CI candidacy criteria broadened and speech recognition abilities improved, many CI users reached ceiling levels of performance on the HINT sentences in quiet (Gifford et al. 2008). As a result, the MSTB (Audit. Potential 2011) was revised to include the AzBio sentence test (Spahr et al. 2012). The AzBio test includes lists of meaningful sentences spoken by four talkers, two male and two female, in a conversational speaking style. However, speech recognition abilities continue to improve in adult CI users with expanding candidacy criteria, and some CI users reach ceiling levels of performance on AzBio after only 3 months of device use (e.g., Sladen et al. 2017).

As studies continue to demonstrate substantial benefits for adults with milder degrees of hearing loss or asymmetric SNHL, off-label cochlear implantation is often done, meaning that implants are offered to individuals who do not meet FDA-approved indications (Carlson 2020). More recently, one CI manufacturer obtained FDA approval for cochlear implantation for single-sided deafness (SSD) or asymmetric hearing loss, which is a profound SNHL in the ear to be implanted and NH or mild to moderate hearing loss in the other ear for listeners who do not receive sufficient benefit from more traditional devices, like hearing aids (Kornak 2019).

The FDA first approved multichannel CIs in 1990 for children between 2 and 17 years of age with bilateral profound (worse than 90 dB HL) SNHL and no open-set speech recognition. By the late 1990s, studies had demonstrated that pediatric CI users with bilateral severe (worse than 70 dB HL) SNHL also improved in their word recognition ability. Current FDA guidelines differ among CI manufacturers and depend on patient age. All children receiving CIs must demonstrate little progress in age-appropriate spoken language and auditory milestones when fitted with hearing aids for 3 months (for children age 12–23 months) or 6 months (for children age 2–17 years). Children between 12 and 23 months of age must have bilateral profound SNHL, and those over age 2 years generally must demonstrate severe to profound SNHL with poor best-aided word recognition when fitted with hearing aids. Since the development of these pediatric guidelines, however, clinicians have advocated for continued expansion of pediatric CI criteria to patients with milder degrees of hearing loss, auditory neuropathy spectrum disorder, and cochlear nerve deficiency with some functional nerve; children younger than 12 months of age; and even some cases of SSD (Teagle et al. 2019).

2. WHAT CAN COCHLEAR IMPLANTS DO FOR ADULTS?

Because postlingually deafened adults with bilateral SNHL are the most common recipients of CIs, we focus our discussion of CI outcomes on this population first. Clinically, adult CI outcomes are traditionally assessed using the same open-set word and sentence recognition materials that are used to establish CI candidacy. In the United States, the most frequently used tests for evaluating
Figure 1

Trajectories of word and sentence recognition improvement over the first 6 months of cochlear implant use for adults. The thin, solid black lines indicate the individual trajectories for percent words correct on AzBio sentences, and the thin, dashed red lines indicate the trajectories on Consonant-Nucleus-Consonant (CNC) words. Mean trajectories for AzBio sentences and CNC words are indicated by the thick black and red lines, respectively.

Outcomes are the CNC word recognition test (Peterson & Lehiste 1962) and the AzBio sentence recognition test (Spahr et al. 2012), described above. The average long-term (i.e., beyond 2 years after implantation) CNC word recognition score for postlingual adults in quiet is approximately 60% words correct (Holden et al. 2013), while the average AzBio sentence score in quiet is around 70% words correct (Gifford et al. 2008). However, there is enormous variability in clinical speech recognition outcomes across individual CI users. Moreover, the time course by which listeners learn to adapt to the novel input provided by the CI is highly variable. Figure 1 demonstrates word (CNC) and word-in-sentence (AzBio) recognition accuracy for CI users prior to implantation and after 1, 3, and 6 months of CI use. As shown in the figure, some individuals display some open-set speech recognition on the day of device activation, while others require 6 months to fully adapt to the device. Still other CI users may demonstrate improvements beyond 2 years after device activation (Dillon et al. 2013).

2.1. How Do We Predict and Explain the Variability in Adult Outcomes?

Individual differences and variability in speech and language outcomes are not new problems in the field of cochlear implantation. In fact, several important issues surrounding the study of variability in outcomes in both children and adults were explicitly addressed as high-priority areas of research in two early National Institutes of Health Consensus Development Conferences on Cochlear Implants, held in 1988 and 1995. Clinical research studies designed to predict (before surgery) or explain (after surgery) this variability have focused primarily on demographic, audiologic, and surgical factors. Older age has a weak negative association with adult CI speech recognition outcomes (Beyea et al. 2016, Williamson et al. 2009). Longer duration of hearing loss and worse preoperative severity of hearing loss have also been associated with poorer outcomes (Blamey et al. 2013, Holden et al. 2013, UK Cochlear Implant Study Group 2004). Surgical
factors, including the distance between the electrode contacts and the cochlear nerve as well as surgery-related trauma to the cochlea from device insertion, also contribute to outcomes (Dalbert et al. 2020, Holden et al. 2013). The health and functional status of the cochlear nerve that receives the electrical stimulation are also associated with speech recognition outcomes (Fitzpatrick et al. 2014, Schwartz-Leyzac & Pfingst 2018).

More recently, rather than focusing solely on the predictive value of demographic, audiologic, and surgical factors, several researchers have begun to investigate why these traditional clinical factors contribute to outcomes. By understanding the underlying sensory, cognitive, and linguistic mechanisms by which these factors contribute, we can better target these mechanisms in our assessments, resulting in a better method to predict or explain outcomes. Moreover, this approach allows us to gain a better understanding of the processes involved in spoken word recognition as well as perceptual learning and adaptation to the highly degraded signals transmitted by the CI. Exploring individual differences in speech recognition in adult CI users can also provide a useful way to investigate how sensory and cognitive processes support spoken word recognition more generally, since the contributions of these factors may not be observed across typically developed NH listeners in the favorable listening conditions of the research laboratory (Matty et al. 2012).

One such demographic factor that has traditionally been found to contribute to outcomes in adult CI users is duration of hearing loss before implantation (e.g., Beyea et al. 2016), which is typically evaluated by self-reports, subjective estimates of changes in hearing. Although difficult to assess accurately in adult CI users, duration of hearing loss appears to have a negative effect on the ability to recognize words in sentences. Yet, this relation does not give us any insight into the effects of long periods of deafness and sensory deprivation on the underlying processes involved in speech perception that relate to outcome variability following implantation. Specifically, previous research suggests that lack of audible speech input has a lasting impact on phonological processing. Adults with severe to profound SNHL demonstrate poor phonological processing, which is at least partially attributable to the deterioration of phonological representations experienced during a long period of SNHL (Lyxell et al. 1998). Interestingly, a decline in phonological processing is not observed uniformly across adults with SNHL. Some individuals with SNHL may be able to maintain stronger and more robust phonological representations through visually based phonological processing (i.e., lip-reading) during everyday speech communication activities (e.g., Lazard et al. 2012). Thus, there is variability in the quality of linguistic representations in individuals with SNHL.

The fidelity and specificity of these phonological representations, negatively affected by SNHL, are crucial for understanding speech after audibility is restored by a CI. Spoken word recognition relies on mapping the acoustic input onto long-term memory representations of spoken words (Rönnberg et al. 2019). A mismatch between the speech signal and long-term representations, arising from degraded speech input, triggers explicit information processing that relies heavily on working memory and controlled attention (Rönnberg et al. 2013, Rudner et al. 2008), resulting in challenging and effortful speech processing. This mismatch may also occur as a result of degraded phonological representations in long-term memory (Classon et al. 2013, Rönnberg et al. 2011). Previous research using auditory (e.g., Moberly et al. 2016, 2017) and nonauditory tasks of phonological processing (Tamati et al. 2021) has demonstrated that phonological processing skills are related to speech recognition outcomes in experienced CI users. Moreover, individual differences in the efficiency or speed of lexical access are also related to speech recognition in adult CI users (Kaandorp et al. 2016).

Our research has found that phonological processing skills are important for predicting and explaining speech recognition outcomes in adult CI users, particularly when nonauditory tasks are
used. Assessment of phonological processing using auditory stimuli is confounded by the presence of poor audibility preimplantation and the fact that the spectro-temporal properties of the auditory input delivered by the CI post implantation is highly degraded. Therefore, efforts have been made to use information-processing tasks that rely on nonauditory stimuli. In a recent study (Tamati et al. 2021), we examined the relevance of reading efficiency (e.g., timed reading of individual words and nonwords, without the context of a meaningful passage) for explaining and predicting outcome variability. To accomplish this, we used the Test of Word Reading Efficiency, Second Edition (TOWRE-2) (Torgesen et al. 2012), which measures single-word, context-free reading accuracy and fluency for word and nonword subtests and provides word, nonword, and overall (word + nonword) reading efficiency scores. Reading efficiency relies both on rapid phonological decoding processes for unfamiliar or nonwords and on direct access to lexical representations for real words (e.g., Baddeley et al. 1985, Jackson & McClelland 1979), reflecting a set of underlying cognitive-linguistic skills relevant to processing both orthographic and spoken language.

Our study found that word reading efficiency, reflecting the speed of lexical access, helps explain variability in speech recognition outcomes in experienced CI users with more than 1 year of CI use (Figure 2a). In contrast, nonword reading efficiency, reflecting the efficiency of phonological decoding, helps predict variability in speech recognition outcomes in new CI users (Figure 2b). Here, sentence recognition scores were determined using semantically anomalous sentences (Herman & Pisoni 2000), which are challenging due to their lack of meaningful semantic content. Interestingly, these findings suggest that the factors that contribute to spoken word recognition under an adverse condition (specifically, speed of lexical access) may be different from the factors that predict the ability to adapt to and make optimal use of a new CI (phonological decoding). Thus, core linguistic skills of the listener may at least partially account for the observed relations between typical demographic factors and speech recognition outcomes, and may provide better insights into the underlying linguistic processes involved in speech recognition in adults with CIs.

Figure 2

Scatterplots demonstrating the relation (a) between postoperative word reading efficiency, based on the Test of Word Reading Efficiency, Second Edition (TOWRE-2), and sentence recognition (percent key words correct) for anomalous sentences in experienced cochlear implant (CI) users ($r = 0.48; p < 0.001$) and (b) between preoperative nonword reading efficiency, based on TOWRE-2, and sentence recognition (percent key words correct) for anomalous sentences in new CI users at 6 months of CI use ($r = 0.36; p = 0.056$).
2.2. What Are the Specific Limitations and Issues for Adults?

Open-set word and sentence recognition have been considered the gold standard in the clinical measurement of performance in adult CI users and in the evaluation of stimulus parameters or different speech coding strategies (e.g., Zeng et al. 2008). Many adult CI users are able to achieve reasonably good open-set speech recognition on conventional clinical speech recognition tests, which largely contain idealized speech materials, that is, carefully articulated, clear speech produced by a single talker or a small number of talkers, such as the AzBio sentences. However, these conventional outcome measures may not reflect the everyday challenges of adult CI users. For many CI users, speech communication remains challenging and effortful, particularly in everyday, real-world listening conditions (e.g., Pals et al. 2013, 2020; Hughes et al. 2018).

Recently, multidisciplinary approaches, based largely on prior research findings in cognitive psychology and linguistics, have sought to better understand the neurocognitive resources that CI users utilize to adapt to and understand the degraded speech delivered by the implant. CI users rely on input signals that are spectro-temporally degraded, due to the limitations of the electrode–nerve interface and the relatively broad electrical stimulation of the auditory nerve (for a review, see Ba¸skent et al. 2016). Accurate recognition of degraded speech can often be achieved, but at the cost of increased cognitive effort and mental workload (Pichora-Fuller 2016). Speech recognition generally remains effortful for adult CI users (e.g., Pals et al. 2013, Winn et al. 2015), who rely heavily on predictive coding and downstream cognitive resources to process the degraded speech input.

Limitations in CI hearing may result in even greater difficulties understanding speech in everyday, real-world challenging conditions. NH listeners are able to recognize and successfully understand speech under an enormously wide range of adverse and challenging conditions, such as noise or other sources of signal degradation, or in the presence of competing talkers. Real-world listening conditions are also characterized by a vast amount of acoustic–phonetic variability originating from multiple talkers with different voices and diverse language and developmental histories (e.g., Gilbert et al. 2013, Mattys et al. 2012, Pisoni 1997). Speech recognition in NH listeners is largely successful despite this acoustic–phonetic variability. NH listeners adjust quickly and almost effortlessly to acoustic differences in speech, facilitated by their ability to perceive, encode, and retain in memory detailed talker information (e.g., Johnsrude et al. 2013, Nygaard & Pisoni 1998, Nygaard et al. 1994, Souza et al. 2011). However, recognizing speech from multiple talkers or coping with different regional or foreign accents may also be more demanding and effortful than idealized lab speech (e.g., Nusbaum & Magnuson 1997). Furthermore, even in NH listeners, studies have revealed large individual differences in performance and suggest that several core neurocognitive skills are factors underlying multitalker sentence recognition (e.g., Tamati & Pisoni 2014, Tamati et al. 2013).

Currently, we know very little about how CI users process compromised signals in real-world conditions, or about the nature of their rapid perceptual adaptation and normalization. Compared with NH listeners, CI users appear to be less sensitive to fine-grained talker-specific acoustic information, and they demonstrate relatively poor or atypical talker, gender, and accent discrimination (e.g., Clopper & Pisoni 2004, Fuller et al. 2014, Hay-McCutcheon et al. 2018, McDonald et al. 2003, Tamati et al. 2014). Similarly, studies in CI users (or in NH listeners using vocoding to simulate CI hearing) demonstrate that the observed limitations in talker perception likely impede adaptation to an unfamiliar talker’s voice (Kapolowicz et al. 2018, 2020; Tamati et al. 2020). Furthermore, adult CI users have also been found to display relatively less accurate recognition of conversationally reduced speech (Liu et al. 2004), highly variable speech produced by multiple talkers (Faulkner et al. 2015, Sommers et al. 1997, Tamati et al. 2020), and foreign-accented speech (Ji et al. 2014, Kapolowicz et al. 2020) in comparison to idealized clear speech. Together,
these studies suggest that talker adaptation processes may be compromised in CI users as a result of limitations imposed by CI hearing.

Conventional assessments, involving idealized speech materials in well-controlled laboratory environments, may overestimate the abilities of CI users and fail to capture meaningful individual variability in performance. Speech materials and listening tasks that better reflect real-world challenges may result in reduced word or sentence recognition performance for CI users and might reveal a wider range of meaningful individual differences in performance. Furthermore, we note that, even with research expanding to study more real-world listening challenges, few studies have examined linguistic processing beyond speech perception and spoken word and sentence recognition in CI users, nor have these tasks become routine assessments in clinical settings. For example, the ability to comprehend spoken discourse and answer questions regarding linguistic content (e.g., Sommers et al. 2011, Tye-Murray et al. 2008) has received little attention from clinical researchers. Similarly, although CIs were originally developed as a supplement to assist listeners with speech-reading in the 1970s, auditory-only, open-set speech recognition abilities have become the gold standard research and clinical assessment tools. Multimodal audiovisual processing abilities, despite their relevance for typical everyday listening conditions for CI users and other listeners with hearing loss (Dorman et al. 2016), are not routinely assessed by audiologists working with CI patients. Finally, to better understand the specific challenges that CI users face in everyday life, researchers have also focused on examining outcomes from the patient’s perspective using qualitative approaches (Finlay & Molano-Fisher 2008, Hallberg & Ringdahl 2004, Hughes et al. 2018, Mäki-Torkko et al. 2015). Relatedly, patient-report outcome measures represent an ongoing advance in the study of outcomes and outcome variability in this patient population (Hinderink et al. 2000, Hughes et al. 2018, McRackan et al. 2018). Therefore, future basic and clinical research studies should consider broadening the approaches for assessing speech recognition, comprehension, and multimodal listening abilities, as well as specific challenges from the perspectives of CI listeners (see the sidebar titled Related Resources). By broadening outcome measures, we may be able to better capture real-world speech recognition abilities in the research lab or clinic.

3. WHAT CAN COCHLEAR IMPLANTS DO FOR CHILDREN?

Cochlear implants are the standard of care for infants and children with severe to profound hearing loss. With the widespread implementation of universal newborn hearing screening, children who are born with severe to profound bilateral hearing loss are now identified at very early ages,
often only a few days after birth. Medical interventions using hearing aids and CIs are routinely recommended to mitigate the effects of spoken language deprivation on speech perception, language development, and intellectual functioning. Approximately 95% of deaf children are born to hearing parents who have no knowledge of sign language (Mitchell & Karchmer 2004). The vast majority of these deaf-of-hearing children receive a CI with the goal of achieving functional spoken language and literacy. Almost all hearing parents of deaf infants want their children to learn to listen and speak and to be mainstreamed in local schools (Geers et al. 2017).

Lack of access to a language model during early sensitive periods for language development in childhood can result in the child failing to achieve native-speaker competence in any language. Given the close interdependencies of language and cognition during early stages of development, a period of auditory and linguistic deprivation increases the risk of cognitive deficits in addition to excluding the child from social communication. Thus, parents and professionals must provide deaf children with a path by which they can acquire full competence in a language. Whether children who receive CIs should be taught sign language is a matter of ongoing debate (see Geers et al. 2017, Napoli et al. 2015). Deaf children may be well served by early exposure to sign language before they receive an implant, but currently there is a lack of clear evidence regarding the development of spoken language with or without the addition of sign language (although see Geers et al. 2017 for results suggesting reservations about this approach).

However, the scientific question of the best approach to promote spoken language development in deaf children is often clouded by broader issues surrounding the potential implications of CI use in deaf children on the Deaf community (e.g., Byrd et al. 2011, Crouch 1997, Tucker 1998). Although this topic is largely outside the scope of this review, we note that CIs are viewed by some members of the Deaf community as threats to Deaf culture. The increased use and availability of CIs have the potential to reduce the number of sign language users, and consequently members of the Deaf community, limiting the transmission of Deaf culture (Sparrow 2005). More compatible with this viewpoint, some research promoting sign language emphasizes the limitations of CI hearing, including the high degree of variability in outcomes, and stresses that reliance solely on spoken language may result in linguistic deprivation with detrimental impacts on language and cognitive development (Humphries et al. 2012; Hall et al. 2018, 2019). However, whether limited familial and clinical resources will be more helpful to the child’s linguistic development when devoted to learning and teaching only spoken language, or spoken and sign language in parallel, remains an individual parental decision that depends on the available resources and abilities of the child, family, and community. Although debates about whether deaf children of deaf parents should receive CIs (Hall et al. 2018, 2019) are ongoing, our focus here is on spoken language development in deaf-of-hearing children who have received CIs.

Over the last 15–20 years, several large-scale longitudinal projects have been carried out by research groups around the world to study outcomes in prelingually deaf children with multi-channel CIs (Ching et al. 2013, 2018; Dettman et al. 2016; Geers et al. 2011; Leigh et al. 2016; Niparko et al. 2010). Other studies are still in progress (Ching et al. 2018, Dettman et al. 2016, Leigh et al. 2016). Typically, a sample of prelingually deaf children is recruited for testing, and a battery of conventional audiometric and language tests is administered at several intervals after implantation, usually every year over some period of time, to study the development of speech and language skills.

The current conventional outcome tests, specified in the Pediatric Minimum Speech Test Battery (Uhler et al. 2017), consist of measures designed to evaluate speech discrimination for infants and word and sentence recognition for children before entering school, using developmentally appropriate behavioral measures. Assessments include closed- and open-set measures of speech recognition, with recommended age ranges for application based on typical development. In
addition to conventional measures, parent-report questionnaires are used to provide a glimpse into a child’s language and linguistic skills in a real-world environment and to supply information for children who cannot complete behavioral assessments due to chronological age or developmental level.

Until the pioneering research program by Ann Geers and collaborators at Washington University in St. Louis, almost all of the previous studies on pediatric outcomes reported findings on very small sample sizes, typically with only a few dozen children. Geers et al. (2003) recruited 181 deaf children with CIs from the United States and Canada and tested 45 of them in St. Louis each summer from 1997 to 2000. All of the children in the initial phase of the project were between 8 and 9 years old, were prelingually deaf, received their CIs before they were 2–5 years of age, and had used their CIs for at least 5.5 years. Eight years after the initial testing, Geers et al. (2011) recruited 112 of the original sample, who were then in high school, to complete follow-up tests after 13.3 years of CI use. The tests also included a large battery of age-appropriate speech and language tests and several novel measures of information-processing capacity and speed that were originally developed by cognitive and developmental psychologists (Pisoni & Cleary 2003, Pisoni et al. 2011).

The overall results of this project were very encouraging, demonstrating the benefits of CIs in prelingually deaf children. Measures of speech perception, speech intelligibility, language, reading, and psychosocial adjustment obtained in high school exceeded scores typically obtained for profoundly deaf children who used conventional hearing aids. Furthermore, Geers & Nicolas (2013) found that more than half of the high school students achieved test scores on language, reading, and psychosocial measures that were within 1 standard deviation (SD) of scores obtained from NH peers. High school students with CIs showed increased participation in mainstream academic settings; greater reliance on spoken language for everyday communications; and a preference for age-appropriate language, academic, and social interactions with their NH peers.

Measures of speech recognition were found to be strongly correlated with measures of speech production, suggesting that robust speech perception skills may be a prerequisite of and underlie improvements in speech motor control, speech intelligibility, and expressive language. Language and reading outcomes were also strongly correlated (Geers & Hayes 2011). Deaf children with CIs who displayed excellent vocabulary and syntax in elementary school achieved better literacy outcomes in high school. One of the most important findings from the tests carried out in elementary school was that verbal rehearsal speed, a measure of information-processing speed and efficiency, was a strong and independent predictor of all speech and language outcomes, above and beyond conventional demographic, hearing, family, and socioeconomic factors. Finally, the children in elementary school who relied exclusively on spoken language showed faster verbal rehearsal speed and achieved higher scores in high school on tests of speech recognition, speech intelligibility, language, and literacy compared with children who used a combination of speech supplemented by signed exact English, suggesting significant benefits in development for experience-dependent learning based on modality-specific auditory information in speech.

A large-scale, multisite longitudinal project was carried out by John Niparko and colleagues at Johns Hopkins University. The children recruited for this study were implanted at different ages and tested annually with a traditional battery of clinical tests of speech and language. One subgroup of the 188 prelingually deaf children recruited for this study was implanted at what was considered a very early age at the time, under 18 months (N = 28). Two other subgroups of children in the project were implanted at 18–36 months (N = 48) and above 36 months (N = 21). Figure 3 depicts some of the major findings on receptive and expressive language outcomes obtained by Niparko et al. (2010), based on the Reynell Developmental Language Scales (RDLS) (Reynell & Gruber 1990, Reynell & Huntley 1985). The figure shows the results for the children who were
implanted before 18 months of age as well as for their NH peers. The individual developmental trajectories show enormous variability in both receptive and expressive language RDLS scores for the children with CIs. However, Niparko et al. (2010) did not address individual differences in their report.

3.1. How Do We Predict and Explain the Variability in Pediatric Outcomes?

Similar to research in adults with CIs, most of the past clinical research on pediatric CI outcomes focused on demographic, medical, device, and hearing history factors. In pediatric CI users, age at implantation is one of the primary demographic factors that has been found to influence almost all outcome measures. Children who receive a CI at a young age generally perform better on a wide range of speech and language outcome measures than children who receive a CI at older ages (e.g., Kirk et al. 2000, Niparko et al. 2010). Length of auditory deprivation or duration of deafness before implantation is also related to outcomes, with children who have been deaf for shorter periods of time before implantation demonstrating better outcomes. Both findings on age of implantation and duration of deafness before implantation demonstrate the important contribution of neural plasticity and the role of early sensitive periods in sensory, perceptual, and spoken language development (Konishi 1985, Marler & Peters 1987).

As for adults, however, these demographic measures are not strong and reliable predictors of CI benefit in the pediatric population. Niparko et al. (2010) observed an enormous variability in outcomes even in the earliest-implanted group of children, who had received their CIs under 18 months of age, suggesting that other factors had influenced outcomes (for suggestions, see Geers et al. 2011). As discussed above for adults, demographic factors like age of implantation or duration...
of deafness represent proxies for a number of more basic underlying elementary information-processing operations that have yet to be identified precisely. Moreover, as the age of implantation continues to steadily decrease, variation in age of implantation will be substantially reduced by early implantation below 12 months of age. This will motivate a new active search for other reliable predictors of speech and language outcomes beyond age at implantation that are more closely linked to the underlying cognitive and linguistic mechanisms of action.

Along these lines, a long-term outcome study of neurocognition and executive functioning in deaf children with CIs, carried out by David Pisoni and William Kronenberger at Indiana University School of Medicine, sought to investigate the cognitive functions of early-implanted deaf children with CIs and to understand the factors contributing to receptive and expressive speech and language skills (Montag et al. 2014, Ruffin et al. 2013). The study focused on outcomes after 15 years of CI use in older children, adolescents, and young adults who received their CIs at a young age and were followed over time. The study design included traditional clinical audiometric measures of speech recognition and several clinical normed and standardized measures of language; a broad range of neurocognitive measures of working memory, attention, inhibition, executive functioning, and verbal learning and memory; and several psychosocial measures. As mentioned above for adult CI users, special effort was devoted to developing visually based neurocognitive measures that did not place demands on audibility of the test materials.

Overall, the average speech perception scores reported by Ruffin et al. (2013) and Montag et al. (2014) were consistent with those from earlier outcome studies (e.g., Davidson et al. 2011, Geers et al. 2003, Spencer et al. 2004). Although their mean language test scores were lower than those of typically developing NH children, a majority of the CI users scored within 1 SD of the normative mean or higher in both receptive and expressive speech and language skills. Speech recognition scores were negatively correlated with hearing loss due to meningitis, older age at implantation, poorer preimplant unaided pure tone average thresholds, lower family income, simultaneous use of speech and signed exact English (i.e., Total Communication), and lower likelihood of bilateral CI use. Age of implantation and amount of spoken language exposure assessed by measures of communication mode and family income were also related to speech intelligibility outcomes. Children in the oldest group who had used their CIs for 15 years or more were more likely to have these characteristics and were also more likely to score lower on measures of speech recognition compared with children who had used their CIs for 14 years or less. The aggregation of these risk factors in the older group accounts for their lower speech recognition scores and may reflect the more conservative CI candidacy criteria in use in the early to mid-1990s in terms of unaided residual hearing and age of implantation. These children, who were among the very first prelingually deaf children to receive multichannel CIs, provide benchmark findings on long-term speech and language outcomes from early childhood into adulthood.

In addition to demographic measures, several other post-CI factors explain outcome variability. Early post-CI speech and language skills are strongly associated with speech and language skills with long-term CI use. Several previous studies found that pediatric CI users’ speech and language skills in elementary school and high school were strongly associated (Geers & Sedey 2011, Hay-McCutcheon et al. 2008). Castellanos et al. (2014, 2016) found that preschool measures of vocabulary size, speech intelligibility, and parent ratings of communicative development predict long-term language and cognitive outcomes. In a prospective, longitudinal study, Nittrouer et al. (2014, 2018) demonstrated that children with CIs show phonological deficits relative to NH peers starting in kindergarten that persist through at least sixth grade, and that individual differences in these phonological abilities were related to lexical skill development such as expressive vocabulary. Moreover, a recent study has shown that speech language skills measured as early as 6 months after implantation predict language and cognitive outcomes after more than 10 years of CI use (Hunter
Taken together, these findings demonstrate that early speech and language skills following implantation facilitate speech, language, and cognitive development in pediatric CI users.

Clinicians and researchers have historically been unable to identify reliable preimplantation predictors of speech and language development after implantation (see, however, Bergeson & Pisoni 2004, Horn et al. 2005). The lack of preimplantation predictors of outcomes presents a challenge for clinicians to identify those children who may be at high risk for poor outcomes at a time when interventions could be made to improve their language skills. Recently, Derek Houston and his research group at The Ohio State University discovered differences in habituation to visual images of novel nonsense objects between deaf infants and age-matched typically developing NH infants (Monroy et al. 2019). Deaf infants required more trials to habituate to a novel visual object, displayed slower rates of habituation, and demonstrated lower look-away rates during habituation than NH infants, suggesting slower and less efficient processing of visual stimuli. Moreover, for the group of deaf infants, visual habituation measures were found to be related to early language skills. These novel findings suggest that early measures of visual information processing obtained preoperatively from deaf infants using visual habituation techniques could be used to predict speech and language outcomes after implantation.

3.2. What Are the Specific Limitations and Issues for Children?

The conventional clinical outcome measures discussed above are the final endpoint or product of a large number of more basic elementary sensory, perceptual, cognitive, and linguistic processes that contribute to the observed variability among CI users. Many deaf children with CIs have co-morbid disturbances and delays in several basic elementary cognitive processes that are integral components of the core information-processing system used in speech perception and spoken language processing, and these disturbances are secondary to their hearing loss and delay in language. A period of auditory and language deprivation during early critical developmental periods before implantation affects cognitive and linguistic development in a variety of ways. Above and beyond the pressing immediate clinical issues of helping deaf children learn to speak and listen like typically developing NH children and reach their full intellectual potential, deaf children with CIs also serve as a unique “model system” to investigate the effects of sensory and language deprivation as well as the restoration of hearing following implantation on both receptive and expressive language (Kral & Sharma 2012, Kral et al. 2016).

Although the spoken language outcomes following CI in profoundly deaf children and adolescents are quite good overall, some deaf children with CIs display specific weaknesses and delays in core areas of speech perception and neurocognitive information processing. Numerous studies have indicated weaknesses in immediate short-term memory, sequence memory and learning, implicit learning and memory, executive function, and rapid phonological coding (Deocampo et al. 2018, Kronenberger et al. 2020). Depending on the specific information-processing domain investigated, some of the differences observed are quite substantial in nature when compared with typically developing NH children, while others are somewhat more subtle. Regarding speech and language outcomes specifically, Kronenberger & Pisoni (2019) observed significant weaknesses in higher-level language processing on the language-processing subtests of the Clinical Evaluation of Language Fundamentals (Semel et al. 2003), a standardized normative test of language-processing skills. In particular, their findings identified delays in processing higher-order complex linguistic information in sentences in prelingually deaf CI users, which are not typically assessed clinically. These recent findings again highlight the critical need for the development and use of a broad-based, comprehensive assessment of speech and language skills that relies on a multidisciplinary team approach.
4. CONCLUSIONS

Fortunately, moderately to profoundly deaf adults and children are generally able to achieve very good speech and language outcomes with CIs, demonstrating the efficacy of cochlear implantation as a medical intervention for the treatment of profound SNHL. However, there remain several challenges in the field that deal with the effectiveness of CIs in individual patients. These challenges represent significant research gaps in our understanding of the underlying foundational mechanisms of action responsible for the enormous individual differences and variability that are routinely observed in speech and language outcomes following cochlear implantation, as well as specific limitations unique to adult and pediatric CI users.

It is becoming clear in the field of CIs that the individual differences and variability in speech and language outcomes following implantation are not merely an ear issue that deals with the sensory coding and processing operations reflecting the upstream contributions from the peripheral auditory pathways. CI outcomes in both adults and children are also brain and language issues, reflecting the downstream contributions of the whole information-processing system, working in an integrated fashion to successfully solve the problem of spoken language understanding with degraded and underspecified representations of speech provided by the CI. Successful outcomes with a CI are very likely due to basic information-processing, learning, and memory processes. Moreover, the enormous variability in outcomes reflects numerous complex neural and cognitive processes that depend heavily on functional connectivity of multiple brain areas working together as an integrated system (Luria 1973). As Nauta (1964, p. 125) pointed out almost 50 years ago, “no part of the brain functions on its own, but only through the other parts of the brain with which it is connected.”

Thus, taken together, the findings reviewed here suggest that the challenges of explaining and predicting individual differences and accounting for poor outcomes after implantation are not just problems for the medical fields of otology and audiology. Rather, this work encompasses several closely related fields of study, including cognitive psychology, cognitive science, developmental sciences, and linguistics. To make further progress, the field of CIs would benefit greatly from contributions from multiple disciplines to generate new knowledge on speech and language outcomes in adults and children with CIs, as well as on the information-processing mechanisms by which underlying factors contribute to outcome variability. Such knowledge will be crucial in providing a scientific basis for the development of novel clinical assessments and rehabilitation protocols to optimize outcomes in adult and pediatric CI patients.

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