World Journal of Otorhinolaryngology-Head and Neck Surgery (2017) **3**, 240–254

Available online at www.sciencedirect.com

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journal homepage: www.keaipublishing.com/WJOHNS; www.wjent.org

Review Article

Three challenges for future research on cochlear implants

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Received 1 November 2017; accepted 1 November 2017
Available online 2 January 2018

KEYWORDS
Hearing loss;
Cochlear implants;
Outcomes following implantation

Abstract Cochlear implants (CIs) often work very well for many children and adults with profound sensorineural (SNHL) hearing loss. Unfortunately, while many CI patients display substantial benefits in recognizing speech and understanding spoken language following cochlear implantation, a large number of patients achieve poor outcomes. Understanding and explaining the reasons for poor outcomes following implantation is a very challenging research problem that has received little attention despite the pressing clinical significance. In this paper, we discuss three challenges for future research on CIs. First, we consider the issue of individual differences and variability in outcomes following implantation. At the present time, we still do not have a complete and satisfactory account of the causal underlying factors that are responsible for the enormous individual differences and variability in outcomes. Second, we discuss issues related to the lack of preimplant predictors of outcomes. Very little prospective research has been carried out on the development of preimplant predictors that can be used to reliably identify CI candidates who may be at high risk for a poor outcome following implantation. Other than conventional demographics and hearing history, there are no prognostic tools available to predict speech recognition outcomes after implantation. Finally, we discuss the third challenge — what to do with a CI-user who has a poor outcome. We suggest that new research efforts need to be devoted to studying this neglected

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Peer review under responsibility of Chinese Medical Association.

Production and Hosting by Elsevier on behalf of KeAi

https://doi.org/10.1016/j.wjorl.2017.12.010

clinical population in greater depth to find out why they are doing poorly with their CI and what novel interventions and treatments can be developed to improve their speech recognition outcomes. Using these three challenges as objectives for future research on CIs, we suggest that the field needs to adopt a new narrative grounded in theory and methods from Cognitive Hearing Science and information processing theory. Without knowing which specific biological and neurocognitive factors are responsible for individual differences or understanding the underlying sensory and neurocognitive basis for variability in performance, it is impossible to select a specific approach to habilitation after a deaf adult or child receives a CI. Deaf adults and children who are performing poorly with their CIs are not a homogeneous group and may differ in many different ways from each other, reflecting the dysfunction of multiple brain systems associated with both congenital and acquired deafness. Hearing loss is not only an ear issue, it is also a brain issue too reflecting close links between perception and action and brain, body and world working together as a functionally integrated information processing system to support robust speech recognition and spoken language processing after implantation.

Introduction

Cochlear implants (CIs) are now universally considered to be the standard of care for the medical treatment of severe-to-profound sensory-neural hearing loss in adults and children. There is no longer any disagreement about the efficacy of CIs among specialists working in the medical community such as neurotologists, otolaryngologists and audiologists who diagnose and treat hearing loss. The anticipated benefits of implantation in restoring the sense of hearing to profoundly deaf adults and children is generally held to warrant the attendant risks of surgery and potential adverse side effects. For example, in the pediatric population, without a CI and access to the sounds of speech, prelingually-deaf infants and very young children with severe-to-profound hearing loss would be unable to acquire knowledge of the grammar of a natural language or develop the receptive and expressive spoken language skills needed to communicate effectively with family, friends and other people in their immediate environment using spoken language. Without cochlear implants deaf children may also have significant global developmental delays and functional limitations over their entire lifetime affecting their quality of life and social interactions with people they encounter in the real-world on a daily basis. Similar benefits are routinely observed in post-lingually deaf adults who have successfully acquired spoken language prior to the onset of their hearing loss. In the elderly adult population, a significant hearing loss acquired later in life has been found to be associated with cognitive declines and may underlie early onset dementia and Alzheimer's disease in some individuals.[1–4] Hearing loss may also be a significant, and potentially independent, risk factor for depression and other psychiatric disorders. Recently, Blake Wilson and his colleagues concluded that cochlear implants represent "one of the great success stories of modern medicine" and that "the cochlear implant is the most successful neural prosthesis developed to date" and "exceeds by orders of magnitude the number for all other types of neural prostheses".[5] The restoration of hearing with a cochlear implant and the stimulation of the auditory nerve with novel sensory input in both prelingually-deaf children and postlingually-deaf adults is now viewed as a significant landmark achievement in the fields of biomedical engineering, neurotology and speech and hearing science.[6]

In this paper, we discuss three major challenges for future research on cochlear implants in adults and children: (1) individual differences and variability in outcomes, (2) lack of preimplant predictors of outcomes and (3) the pressing need for novel interventions for patients who achieve poor outcomes after implantation. We believe these three particular issues are the most important and perhaps the most challenging research problems in the field that will need to be addressed in the future to insure that all patients who are candidates for CIs will be able to obtain maximum benefits from their CIs and reach optimal levels of speech recognition and spoken language performance. In some broader sense, we can think of these three issues as "grand challenges" for future research on CIs. It is our hope that discussing these issues here and making these challenges explicit will serve as a springboard for new research efforts on these three problems in the future.

This paper is organized into five main sections. We will not present any new research findings, but instead will focus our discussion on the existing research literature pertinent to the following three questions. First, why has so little progress been made in understanding and explaining the enormous individual differences and variability routinely observed in outcomes following implantation? Second, why are there no valid and reliable preimplant predictors of outcome? And, third, what does a hearing healthcare provider do for an adult or child who has a poor outcome after implantation? In the first introductory section, we review the efficacy versus effectiveness of CIs, process versus product measures, and the use of converging methods from the field of Cognitive Hearing Science. In the second section, we discuss why there has been so little substantive progress made in understanding the enormous individual differences and variability observed in speech and language outcomes in deaf children and adults who have received cochlear implants. In the third section, we discuss the pressing clinical need for developing novel preimplantation predictors of outcomes following implantation. At the present time, all that clinicians have available to assist with the prediction of outcomes is a set of variables including demographics, hearing history, and a small number of measures obtained from a battery of preimplant behavioral tests that are used to establish candidacy.[7] Currently, there are no reliable prospective behavioral or neural measures that can be applied preoperatively to predict post-implantation outcomes. The lack of valid and reliable preimplantation prognostic measures of outcomes represents a significant progress-hindering gap in our current knowledge and understanding of the efficacy and effectiveness of cochlear implants in both adults and children. This is a very important and neglected problem in the field of cochlear implantation that needs to be directly addressed in the future.

In the fourth section, we discuss the need for developing novel auditory, neurocognitive and linguistic interventions for CI with poor outcomes. Patients with poor outcomes following implantation have received insufficient attention in the overall research narrative on cochlear implants, and we believe that it is time to address this fundamental gap in our knowledge. At the present time, there is little consensus about precisely what to do with an adult or child who has a poor outcome — how to diagnose the patient's underlying problem and, importantly, how to improve poor performance with the patient's CI.[8]

Finally, in the last section, we provide a summary of our conclusions and then place our observations and recommendations about these three challenges within a broader context that emphasizes the need to critically rethink the current ongoing narrative about individual differences and variability following implantation, the clinical utility of preimplant predictors of outcomes, and the pressing need for the development of novel interventions for adults and children who display poor outcomes after implantation. While there have been significant accomplishments and truly enormous progress and discoveries made in the field of CIs over the last 25 years, there is still a great deal of important work to be done in the future to address these long-standing grand challenges which represent critical barriers to further progress and application of this technology to hearing impairment in adults and children.

Efficacy and effectiveness of cochlear implants

Much of the clinical research carried out on CIs over the past 25—30 years has been concerned with "efficacy" of CIs, that is, demonstrating and documenting that CIs work and provide significant benefits to profoundly deaf children and adults. It may be argued that this important initial phase of CI research, with respect to conventional CI candidates, has been concluded. In contrast, less research has been devoted to the "effectiveness" of CIs, that is, understanding and explaining the reasons for the enormous variability in outcome and benefit following implantation. When we consider the efficacy of a medical treatment or intervention, we are considering the ability or power of the intervention to produce the desired effect in an individual patient. That is, does a CI work, and does it provide benefit to a profoundly deaf person? In contrast, when we consider the effectiveness of a medical treatment or intervention, we are actually considering that treatment or intervention's ability to produce the expected effect in an individual patient in the real world. That is, does a CI work equally well and provide the desired benefit in everyday use in everyone who is a candidate and receives a CI? Most of the clinical research on CIs over the last 25—30 years has been concerned with device efficacy and has been designed to demonstrate that CIs work in an individual or samples of patients with severe-to-profound hearing loss; in contrast, very little sustained longitudinal research has focused on the effectiveness of CIs and why they often work very well in some patients but sometimes work more poorly or not at all in other patients of the same age, gender, demographics and medical hearing history.

Process vs. product measures

Much of the previous clinical research on CIs has focused on the effects of a small number of medical, device and demographic variables using traditional "product" or "endpoint" outcome measures based on assessment tools developed by clinical audiologists. These assessments have relied entirely on accuracy measures of performance (i.e., percent correct), with less understanding of process factors that explain the foundational processes driving accuracy outcomes. Although rarely discussed in the literature, all of the clinical outcome measures of performance used to assess the benefits of CIs are the final "product" or "endpoint" of a large number of complex sensory, perceptual, neurocognitive and linguistic processes that contribute to the observed variation among CI users. Until recently, no one obtained "process" measures of performance from patients with CIs to examine the underlying elementary information processing mechanisms used to perceive and produce spoken language in this clinical population.[9] Our recent findings summarized briefly in the sections below using a variety of process measures of performance have provided several new insights and important new knowledge about the underlying neurocognitive basis of individual differences and variability in profoundly deaf children with CIs.

Process measures of outcome and benefit have the potential to provide highly detailed knowledge and information about the underlying (latent) information processing mechanisms that conventional endpoint product measures are rooted in, but themselves cannot provide. All of the conventional endpoint product measures used routinely in the clinic were not designed to assess individual differences or variability in outcomes following implantation. These conventional measures were originally selected by the implant manufacturers to establish "efficacy" of cochlear implants for FDA approval. They were not developed to measure and assess "effectiveness" of CIs or the underlying foundational information processing operations that are used in speech perception and speech recognition, speech production and intelligibility, spoken word recognition and lexical access or retrieval processes underlying language comprehension. Moreover, almost all of these conventional product endpoint measures are based on accuracy and percent correct scores. None of the conventional outcome measures assess the speed of information processing or efficiency of the underlying neurocognitive processing operations. Moreover, conventional product or endpoint measures of outcome and benefit are, for the most part, just descriptive in nature — they are not explanatory, and, therefore, they are unable to provide detailed explanations of the elementary underlying information processing operations and mechanisms of action employed in a specific behavioral measure of performance.

In contrast, process measures of outcome rely on fundamentally different measures of performance such as processing speed, capacity of immediate short-term memory and working memory dynamics, learning, memory, inhibition and adaptation, and others. Process measures of performance are specifically designed to provide measures of the underlying mechanisms of action—neurocognitive operations such as: information processing speed, capacity, scanning and retrieval of information in active verbal short-term memory, retrieval of stored information from long-term memory, processing efficiency, flexibility, learning and memory, inhibitory control, perceptual normalization and adaptation — a small handful of elementary information processing components and subcomponents that reflect system integrity and functionality that can be used to assess how the whole information processing system works together in carrying out a specific behavioral task and what the core components are and how they contribute to the final product. The difference between product and process measures of performance is theoretically and clinically significant because these two different types of measures assess fundamentally different aspects of the perceptual process.

Although conventional endpoint product measures are important because they have clinical utility and strong ecological face validity, they are limited in their ability to identify and assess specific underlying subprocesses which process-based measures are specifically designed to measure. Sometimes process measures are called "latent variables" because they reflect an underlying neurocognitive construct such as registration, encoding, storage, rehearsal, retrieval, inhibition or capacity. All of these hypothesized latent variables are assumed to underlie the actual score of the "manifest variables," the actual dependent measures of performance typically obtained in any given behavioral study.

Process measures are extremely important measures to obtain from both normal hearing listeners as well as clinical populations with hearing loss because they assess core subprocesses that are assumed to be operative in all behavioral tests using complex speech signals as well as other non-speech signals like music and naturally-occurring environmental sounds. They are theoretically-motivated and grounded in a broader conceptual framework.

Cognitive hearing science and cognitive audiology

Over the last few years there has been an increased awareness and explicit recognition by a large number of hearing scientists and cognitive psychologists that the brain and central cognitive information processing operations and mechanisms play a critical role in supporting robust speech recognition and spoken language processing in a variety of clinical populations. The ear and brain are closely linked and coupled together by reciprocal neural connections and overlapping brain networks that support robust recognition under highly degraded listening conditions.[10—12] The awareness and acknowledgement of these links and connections and the complex interactions between ear and brain function have fostered and encouraged the development of two new closely-related emerging fields of study related to hearing, auditory cognition, and speech perception—Cognitive Hearing Science and Cognitive Audiology.[13]

In addition, because the field of clinical audiology is an applied science drawing knowledge and methods from several different disciplines, no common integrated theoretical framework motivates the choice of specific outcome measures, interprets the results and findings, provides explanations, or makes predictions. Without the benefit of a well-defined conceptual framework and additional theoretically motivated "process-based" measures of performance, it is very difficult to gain any new knowledge about the underlying neural and neurocognitive factors that are responsible for the observed variability in the traditional audiological outcome measures of performance. And, it is very difficult, if not impossible, to select a specific approach to habilitation and therapy after cochlear implantation without knowing precisely which underlying sensory and neurocognitive factors are responsible for the individual differences and variability in outcomes and the weaknesses in a specific patient. Moreover, all of the clinical research on CIs has been primarily descriptive and correlational in nature as opposed to experimental research designs and has not been motivated by hypothesis-testing or specific predictions from theories or models that would lead to understanding and explanation of process and mechanism. The bulk of CI research has focused on medical, demographic, hearing history and educational factors, not the underlying neurobiological and neurocognitive information processing operations and mechanisms that link brain and behavior.

To understand, explain and predict individual differences in outcome and benefit following cochlear implantation, it has become necessary to situate the problem of individual differences and variability in outcomes in this clinical population in a much larger global theoretical framework that fully recognizes and acknowledges that variability in brain-behavior relations is a natural consequence of biological development of all living systems.[14,15] The enormous variability observed in a wide range of speech and language outcome measures following implantation may not be unique to this particular clinical population but may reflect instead more general underlying sources of variability in behavior observed in speech and language processing in healthy typically-developing normal-hearing adults and children.[16] Moreover, it is very likely that the sources of the individual differences observed in speech and language outcomes in adults and deaf children with CIs also reflect variation in both domain-general and domain-specific neurocognitive processes.[17,18]

In order to investigate the sources of variability in performance and understand the neural and cognitive processes that underlie variation in outcome and benefits following implantation, new outcome measures are needed to assess a much wider range of behaviors and information processing skills beyond just the traditional clinical audiological, speech, and language endpoint measures that have been routinely used in the past. For example, the failure of an adult or child to obtain optimal benefits and achieve age-appropriate speech recognition outcomes from his/her CI may not be due directly to the functioning of the CI device itself but may reflect a combination of complex interactions among a number of contributing factors.[19] We have adopted the general working assumption in our research that many profoundly deaf children and adults who use CIs, especially patients with suboptimal outcomes, may have other neural, cognitive and affective sequelae resulting from a period of auditory deprivation combined with delays or disturbances in sensory coding and language processing before and after implantation. Thus, one of our core hypotheses is that the enormous variability observed in speech and language outcomes following implantation is not only due to hearing (i.e., detection and discrimination of auditory signals) and the early sensory encoding of speech but also reflects the contribution of other neurocognitive factors related to how sensory information is encoded, stored, and retrieved from memory (i.e., recognition, identification, categorization and classification of auditory signals), that is, how the sensory information delivered from a cochlear implant is "processed" by an adult or child with a significant hearing impairment. When we use the term "processed" in this paper, we mean that the information processing system carries out a series of operations on signals that lead to a new product or outcome via coding, recoding, organizing, binding, filtering or transforming of an input signal into another representation.

"Information processing" is a term that is routinely used to describe a broad-based approach to the study of complex high-level psychological processes such as perception, cognition and thought.[20,21] Information-processing theories are concerned with an analysis of "central processes" of large complex systems (such as human cognition) used in visual object recognition, perceptual learning and memory, speech perception, and various aspects of language processing such as comprehension or speech production. As Ulric Neisser put it many years ago in his seminal book Cognitive Psychology:

> "As used here, the term "cognition" refers to all the processes by which the sensory input is transformed, reduced, elaborated, stored, recovered, and used. It is concerned with these processes even when they operate in the absence of relevant stimulation, as in images and hallucinations. Such terms as sensation, perception, imagery, retention, recall, problem-solving, and thinking, among many others refer to hypothetical stages or aspects of cognition."(p4).[21]

A common goal of the information processing approach adopted by Cognitive Hearing Science and Cognitive Audiology is to examine the neurocognitive representations, elementary psychological processes and cognitive structures used in complex cognitive activities and to trace out the time-course of these processing operations.[20,22–24] Many sophisticated high-yield experimental methods have been developed by cognitive psychologists and cognitive scientists over the last 50 years to study cognitive processes such as perception, attention, learning and memory and inhibitory control within the framework of human information processing.[20–22] In addition, this approach has also provided a variety of novel theoretical and conceptual tools for modeling the mechanisms and processes involved in cognition and the underlying psychological phenomena.[25,26]

Brain-behavior relations

Our approach to the problems of individual differences and variability in outcome and benefit following cochlear implantation is motivated by recent findings and theoretical developments that suggest that deafness and hearing impairment cannot be viewed in isolation as a simple sensory impairment.[27] The enormous variability in outcome and benefit reflects numerous complex neural and cognitive processes that depend heavily on functional connectivity of multiple brain areas working together as a complex integrated system.[28] As W. Nauta[29] pointed out more than 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" (p125). Except for recent studies, measures of system-wide brain coordination and functional connectivity reflecting cognitive processes that are critical for robust highly adaptive speech and language behaviors have not been routinely obtained in deaf adults and children with CIs. We believe this is a promising new direction to pursue in clinical research on individual differences in deaf adults and children who have received CIs.

Challenge No. 1: Individual differences and variability in outcomes

The published research studies on speech and language outcomes following CI have documented that CIs work and often work very well in both the early implanted prelingually deaf pediatric population with congenital hearing loss and the post-lingually deaf adult population of patients with acquired hearing loss who meet the conventional criteria for CI candidacy.[7] Although CIs work and often work very well in most deaf children and adults who meet the candidacy criteria established by the FDA and adopted by most healthcare insurers, there are still many CI users who fail to display optimal levels of speech recognition and often perform poorly with their CIs even after several years of use and, when available, intensive aural rehabilitation (AR) carried out by experienced clinical audiologists and speech-language pathologists. The individual differences and variability in speech recognition outcomes following CI have been, and continued to be, one of the most important and most challenging research problems in the field of CIs today. Even after more than 25 years of clinical research on CIs, the enormous variability and individual differences in speech recognition outcomes still remains an enigma in the field of Otology and Audiology and represents a significant challenge for future research on CIs.[8] This is the first challenge we discuss below.

Why is there so much variability in outcomes and why are there such enormous individual differences in speech recognition performance following implantation? Some candidates who receive a CIs do extremely well with their devices, often approaching the levels of performance on open-set word recognition and sentence speech perception tests in the quiet that are, for all intents and purposes, comparable to scores obtained by age-matched normal-hearing peers without hearing loss. These exceptionally good CI users are frequently referred to as the "Stars" in the clinical research literature on CIs to distinguish them from the average, typical CI users, who derive some benefit from their CIs but fail to achieve such spectacularly good speech recognition scores that are often indistinguishable from normal hearing listeners on the same set of behavioral tests.[30] It is important to emphasize here that the "extraordinary" good performance displayed by the "Stars" on conventional clinical speech recognition tests serves as a very important benchmark about the "efficacy" and success of CIs and as such can be considered as a "proof of concept" or "proof of principle" that the current generation of signal processing algorithms and processing strategies developed for use in CIs provides a sufficient amount of spectral and temporal information about the information bearing elements of speech to support reliable speech recognition performance in some deaf individuals with CIs under quiet benign listening conditions in the clinic and research laboratory. Many of these exceptionally-good CI patients also do extremely well in real-world everyday listening conditions and speech communication tasks, and they routinely report substantial benefits from their CIs. Precisely why this small subset of CI users does so well with their CIs still remains unclear at this time, especially in light of other results with comparable deaf patients with the same medical and hearing histories who often display very poor performance on the same battery of standardized speech recognition and spoken word recognition tests.[31] At the present time, we do not have a well-articulated and fully-developed theoretical account of individual differences and variability in speech recognition following implantation, and we are therefore unable to provide any solid principled explanation of why some patients do so well with their CI and why others struggle to achieve even minimal benefits from their CIs.[8,9] Relying on conventional demographic and hearing history variables such as chronological age, age of implantation, or duration of deafness before implantation may be a useful general purpose heuristic for routine clinical purposes and counseling patients and families about medical decision-making. Unfortunately, demographic and hearing history measures, while clearly having some clinical face validity on the surface, are actually only "proxy measures" for the actual causal underlying processes, because like most of the behaviorally-based endpoint or product measures of outcomes, conventional demographic measures are not directly linked to any underlying causal mechanism of action or set of information processing operations carried out by the auditory system and the brain. The overwhelming bulk of what we currently know about outcomes following cochlear implantation is based on what is often called "weak science"— that is, descriptive research that relies almost entirely on observational or epidemiological studies based on correlational analyses. This approach may be contrasted with the "strong science" approach which relies on experimental studies of causation designed to test specific hypotheses and predictions revealed through manipulations of independent variables in true experimental designs with appropriate control conditions.

Although the problem of individual differences and variability in outcomes following implantation has been a very long-standing issue in the field going back to the very earliest days of basic research on the efficacy of CIs,[32] some progress has been made over the last few years by narrowing down the problem of individual differences to a more manageable size and rethinking the conventional clinical measures by adopting theory and experimental methodologies based on the information processing approach which has been embodied in the new emerging fields of Cognitive Hearing Science and Cognitive Audiology.[13] We have carried out several pioneering studies that measured the information capacity of immediate memory for sequences of highly familiar materials using measures of digit span.[33] In addition to measures of information capacity of verbal short-term memory as indexed by digit span length and total digits correctly recalled, we also obtained detailed speech timing measures of retrieval speed from verbal short-term memory based on scanning of items correctly recalled based on the vocal responses produced by subjects in carrying out the digit span task.[34] Other speech timing measures designed to assess verbal rehearsal speed were obtained from measuring the durations of spoken sentences elicited in different speech production task that was used to assess speech intelligibility.[33]

The first study of verbal short-term memory capacity using Wechsler Intelligence Scale for Children[35] digit span tests in deaf children with CIs was carried out by Pisoni and Geers in 1998.[36] They reported results obtained from a group of 43 prelingually deaf children who were between 8 and 9 years of age at test. Strong correlations were found between WISC digit spans and four different conventional outcome measures—speech perception using the closed-set WIPI test, speech production and intelligibility using playback methods and transcriptions of McGarr sentences, a global language functioning using the WISC similarities test and the TACL, and reading skills using the Woodcock Word Attack and PIAT vocabulary and comprehension subtests.[36] Pisoni and Geers's results showed that these four measures of spoken language processing and verbal short-term memory capacity indexed by digit spans were closely related and shared reciprocal links and processing resources that were used in speech perception, speech production, language comprehension and reading. Other follow up studies on verbal short-term memory with larger sample sizes using the same measures of digit span that were carried out by Pisoni and Cleary (2003)[33] and Burkholder and Pisoni (2005, 2006).[34,37] Their results established important links between conventional speech and language outcome measures, digit spans and verbal rehearsal speed as well as scanning and retrieval of digits from short-term memory.

In one set of analyses, Burkholder and Pisoni reported that deaf children with CIs were three times slower to retrieve verbal items (digits) from active verbal short-term memory even when all of the digits were correctly recalled compared to a control group of age-matched normal hearing typical-developing children.[34] Results from several other larger-scale longitudinal studies of digit span and verbal rehearsal speed in deaf children have been reported and showed: (1) that prelingually deaf children with CIs lag behind NH peers in verbal short-term memory (i.e., digit span) by about the same amount from ages 6—16 years and (2) that measures of early verbal short-term memory predict later language outcomes.[31,38—40] More recent research using visual and/or computer-based administration of the WISC digit span test demonstrated that these findings were not explained by audibility or speech production factors in the digit span test suggesting a central role for rapid verbal coding and verbal rehearsal processes.[41]

These initial studies of verbal short-term memory using information processing theory and methodologies demonstrated the clinical utility of examining the foundational elementary component operations that underlie several standard conventional end-point product measures of outcomes following implantation. In particular, the studies on digit span recall demonstrated that information processing capacity and processing speed, two elementary foundational components of cognition, underlie large amounts of variance observed in conventional clinical endpoint product measures in this population. Thus, individual differences and variability in conventional outcome measures can be traced back to more elementary component processes of neurocognition and information processing that reflect differences in speed of information processing operations such as verbal rehearsal, scanning and retrieval of items in verbal short-term memory and the rate of encoding phonological and lexical information in verbal working memory.[42]

These early studies on digit spans also demonstrated the central role of memory processes, specifically, short-term verbal memory capacity and working memory dynamics, as a common source of variance underlying different outcome measures. What a subject does with the initial sensory information they get from their CI is a very important source of variance in explaining individual differences and variability in performance on a range of outcome measures. We know from other research studies that CI users receive a compromised, highly-impoverished and spectrally-degraded transform of the acoustic signal from their CIs that they have to make efficient use of in information processing tasks that assess spoken word recognition, lexical access, speech production, language processing and reading. The signal processing algorithms and coding strategies in CIs significantly reduce the information content in the auditory nerve providing "sparsely-coded" and "underspecified" acoustic signals to the brain and higher speech and language centers. As a result, patients with CIs have very poor episodic encoding of contextual cues in memory resulting in a significant reduction in the registration and encoding of highly-detailed indexical attributes in the speech signal—the temporal fine structure of the signal that specifies the speaker's gender, regional dialect, emotional state.[43] Most CI users also have a great deal of difficulty encoding and recognizing individual voices because they are unable to encode and process detailed information about the idiosyncratic features of the talker's vocal tract transfer function and voicing source characteristics which are very poorly encoded and transmitted even by the current generation of multichannel cochlear implants in use today.[44]

Challenge No. 2: Preimplant predictors of outcomes after implantation

Until recently, clinicians and researchers working on CIs were unable to identify reliable preimplant predictors of outcome and benefit following implantation, above and beyond the conventional demographic and hearing history variables such as age at implantation and duration of deafness.[45—47] The absence of valid and reliable preimplant predictors is a theoretically significant finding, because it suggests that many complex interactions take place between the newly acquired sensory capabilities of a prelingually deaf child or a post-lingually deaf adult after a period of auditory deprivation, experience- and activity-dependent properties of the language-learning environment, and various interactions with family and caregivers after implantation. More importantly, however, the lack of reliable preimplant predictors of outcome and benefit makes it difficult for clinicians to identify adults and children who may be at high risk for poor outcomes at a time in development when changes and adjustments can be made to modify and improve their speech recognition skills.

In the past, a small number of prognostic factors have emerged as potentially useful measures in predicting outcomes and individual differences in success with a cochlear implant. Although the impact for prelingually deaf, early-implanted pediatric cochlear implant users is still unclear, in postlingually deafened adults, the positioning and depth of insertion of the electrode array inside the cochlea has been shown to reliably predict some portion of the variability among adult patients in word recognition scores attained postimplantation.[48,49]

Very few reliable behavioral preimplant predictors of success with a cochlear implant are available for young children. The insufficient number of reliable preimplant predictors of benefit and success suggests that basic underlying neurocognitive factors such as learning, memory and, attention and inhibitory control may be the important targets for preimplant and post-implant evaluations of both adults and children with cochlear implants.

Moreover, the lack of reliable preimplant predictors in children is also important because it suggests the presence of complex interactions among the newly acquired sensory and perceptual capabilities of an adult or child after a period of sensory deprivation, the properties of the language-learning environment, and the various interactions with parents and caregivers that the patient is exposed to early on after receiving a cochlear implant. The absence of reliable preimplant predictors of outcome also makes it difficult to identify in a timely manner low-functioning adults and children who may benefit from early initiation of intervention and aural rehabilitation.

Complex interactions exist in the language-learning environment that affect the way early sensory information is perceived, encoded, stored and interpreted by adults and children with CIs. Investigation of these intermediate processes may provide valuable new insights into the wide variation observed in outcome performance. While the lack of preimplant predictors based on traditional outcome measures may be somewhat troubling for clinicians and researchers who would like to maximize the benefits of cochlear implants by modifying or adjusting intervention strategies soon after implantation, other more basic measures of performance that are related to the information processing operations such as verbal short-term and working memory capacity and coding and rehearsal strategies may be worth exploring in greater detail in addition to the traditional audiological outcome measures that have been used over the years to assess performance.[50,51]

Two studies carried out in the past have looked at this problem in greater depth and reported encouraging findings suggesting that it may be possible to develop reliable preimplant predictors of outcome performance. The first study was carried out with post-lingual adults; the second investigated prelingually deaf children. In a study of postlingually deafened adults, Knutson et al (1991)[52] found that preimplant performance on a visual monitoring task (VMT) predicted audiological outcome after 18 months of implant use. Strong and highly significant correlations were found between VMT performance in a signal detection task and scores on four sound-only audiological measures, sentence perception, consonant and vowel perception and phoneme recognition in words. These results obtained with adult patients demonstrated that the cognitive processing operations and skills needed to rapidly extract information from sequentially arrayed visual stimuli may also be used in processing auditory signals and may be an important processing factor that underlies the successful use of a cochlear implant (see also Gantz et al, 1993).[53] The findings obtained in Knutson et al's study support the hypothesis that higher-level cognitive factors related to perception, attention and working memory capacity play an important role in predicting outcome with an implant. More importantly, these results show that preimplant measures of information processing in the visual modality can be used to predict speech perception performance in the auditory modality.

Studying the pediatric population, Tait, Lutman and Robinson (2000)[54] reported moderate correlations between pre-verbal communication measures extracted from an analysis of videotapes and several outcome measures of speech perception obtained from prelingually deaf children three years after implantation. Video recordings of 33 children were transcribed and scored for various turn-taking and autonomy behaviors before implantation. Outcome measures of sentence perception, discourse tracking and telephone use were obtained without the use of visual cues and correlations were computed with the behaviors obtained from the coded videotapes. Although positive correlations were found between each of the outcome measures and the preimplant behaviors coded from the videotape analysis, none of the correlations with the turn-taking behaviors reached significance. However, the correlations with the autonomy behaviors were significant suggesting that some pre-verbal communicative behaviors that are present before implantation are associated with audiological outcome measures of speech perception and language processing obtained three years later.[52] The findings reported by Tait et al (2000)[54] suggest that several important aspects of the development of spoken language are already present in infancy in these deaf children. These underlying pre-verbal communication skills may function as the "prerequisites" and serve as a type of "scaffolding" for speech and language development in very young prelingually deaf children and, therefore, may be quite general in nature reflecting multi-modal interactions between perception and action that are not tied to a specific sensory modality. The Tait et al findings are, of course, correlational in nature and it is necessary not only to replicate these findings but also try to specify more precisely the underlying neural and perceptual mechanisms that are responsible for these differences. It is possible that differences in imitation behaviors, gestures and perceptuomotor links related to joint attention between mother and child are the fundamental processes that actually underlie the observations obtained from the analysis of the videotapes.[55–57]

Measures of verbal learning and memory as predictors of outcomes

Two recent studies have reported findings obtained from the California Verbal Learning Test (CVLT) that may be useful as a prognostic index of speech recognition outcomes following implantation in postlingually deaf adults.[49,58] The CVLT is routinely used to assess verbal learning and memory processes in a wide range of different clinical populations. Both studies used a non-standard version of the CVLT that combined simultaneous visual (print) and live-voice auditory presentation of the test items on the study lists. The standard clinical format of the CVLT uses "live-voice" presentation of the stimulus materials. In the first study, Heydebrand and colleagues (2007)[58] reported that a composite free recall score based on several subcomponent measures of free recall performance accounted for 42% of the variance in CNC monosyllabic word recognition scores six months post-implantation. The authors suggested that verbal learning and memory tasks like the CVLT could be used to predict speech recognition outcomes in postlingually deaf CI users, but they did not provide any additional details about precisely what kinds of verbal learning and memory measures would be clinically useful and which specific domains of verbal learning and memory should be investigated further.

In a more recent study with a larger sample size encompassing a broader age range, Holden and colleagues (2013)[49] also reported a significant relation between a composite free recall score on the CVLT and speech recognition outcomes, but this correlation was eliminated when they controlled for chronological age. However, despite use of combined visual-auditory presentation of stimuli, both of these studies were also limited in their conclusions because the CVLT measures they collected may have relied heavily on auditory abilities of the participants. Moreover, detailed analysis of the critical "process" and "contrast" measures of performance from the CVLT, which was designed to measure specific capacities of verbal encoding, storage, retrieval, and self-generated organizational strategies used in retrieval, were not reported in either of these two earlier studies. The process measures of verbal learning and memory obtained from the CVLT have been shown in numerous studies with other clinical populations to be highly informative about underlying cognitive information processing strategies, because they provide detailed information and quantitative measures about what participants are doing with the verbal information they encode, store, and retrieve from memory in this task. Without examining the process measures provided by the CVLT, such as learning rates, proactive interference (PI) and retroactive interference (RI), retrieval inhibition, release from PI, and organizational strategies such as semantic, serial and subjective clustering of output responses, as well as repetitions and intrusions in free recall, detailed insights cannot be gained into the possible differences in underlying information processing operations and neurocognitive mechanisms used by participants in carrying out the CVLT task protocol. Focusing on only the "first-order" primary free recall measures obtained from the immediate and delayed free recall trials provides only a global overall assessment of the foundational information processing operations underlying verbal learning and memory for lists of categorized words in this clinical population. Without detailed information about the process measures on the CVLT, only an incomplete picture of the strengths, weaknesses, and milestones in these patients was obtained in both of these earlier studies.

Despite concerns and reservations about these earlier studies, the free recall findings reported by Heydebrand et al (2007)[58] and Holden et al (2013)[49] suggest that measures of verbal memory and learning processes could serve as useful predictors of speech recognition outcomes in adult CI users and, therefore, might provide new process-based behavioral measures that could help explain the underlying basis of the enormous individual differences and variability observed in outcomes following implantation.

Recently, we completed a new study on verbal learning and memory in post-lingually-deaf adults with CIs using an updated and revised version of the California Verbal Learning Test (CVLT-II).[51] The standard clinical version of the CVLT-II uses live-voice presentation of the stimulus materials that could be a potential problem for elderly patients who have significant hearing loss. To address issues related to audibility and early sensory encoding of auditory input, we developed a version of the CVLT-II that used visual presentation of the stimulus items on a computer display screen with no accompanying spoken words. Thus, differences observed among CI users or between CI users and normal-hearing control participants cannot be due to modality-specific sensory effects related to audibility or sensory processing and encoding of the test materials. Rather, any differences observed must reflect a level or levels of information processing that involve modality-independent differences associated with verbal coding, phonological and lexical processing, storage, retrieval, and information processing operations that are not compromised by prior hearing loss or differences in audibility or early sensory registration and processing of auditory signals by their CIs.

This new study was designed to compare performance of experienced elderly adult CI users (ECIs) and age-matched older normal-hearing (ONH) control participants using a visually presented CVLT-II and to investigate the relations between verbal learning and memory and speech recognition outcomes in the ECI users.[51] In addition to the CVLT-II, we also collected several non-auditory visually-based neurocognitive measures to investigate the relations between measures of verbal learning and memory obtained from the CVLT and neurocognitive scores from tests of non-verbal fluid reasoning (IQ), reading fluency, immediate memory span, and vocabulary knowledge.

While the two groups of elderly participants did not differ on most of the measures of verbal learning and memory obtained with the visual CVLT-II, several significant differences related to the build-up of proactive interference (PI; earlier learned information interfering with memory for information presented later) and retrieval induced forgetting (RIF) were found. RIF is a form of forgetting that is due to retrieving specific items from long-term memory that subsequently impairs recall of other semantically-related items. Within the ECI group, nonverbal fluid IQ scores from the Ravens Progressive Matrices, reading fluency indexed by the Towre-2 test, and resistance to the build-up of PI from the CVLT-II consistently predicted better speech recognition outcomes. While still preliminary in nature and based on a relatively small sample size ($n = 25$), the results of this study suggest that several underlying neurocognitive abilities are related to speech recognition outcomes following implantation in older adults and may serve as prognostic measures for predicting outcomes after implantation. A true prospective study of verbal learning and memory processes in cochlear implant candidates using the visual CVLT-II format in a pre-implant vs. post-implant design is currently underway to determine the prognostic utility of measures of verbal learning and memory in predicting speech recognition outcomes after implantation.

Challenge No. 3: Developing novel interventions for poor outcomes

One of the most important challenges for future research on CIs is the adult or child who displays a poor outcome after several years of CI use. This is a very important issue that has received little attention in the fields of otology and audiology despite its direct clinical relevance to improving outcomes following implantation. Almost all of the research in the field of CIs has been focused on studying patients who achieve successful outcomes after implantation. There is no question that a significant selection bias is present in the published research studies on CI outcomes. CI efficacy studies have focused much less on poor-outcomes and have concentrated more on the average and exceptionally good CI users—the "Stars" who often display extraordinary good performance on a wide range of outcome measures that is comparable to results routinely observed with age-matched NH controls." In fact, the inclusionary and exclusionary criteria in many published research studies make this observation explicit by specifying a lower limit on the performance of participants to avoid floor effects in their statistical analyses. One consequence of this approach of "weeding out" and excluding poor performers from research studies is that patients with suboptimal outcomes fall through the cracks and are underrepresented in large-scale studies of outcomes following implantation. With the exception of recent work by Moberly et al (2016),[8] there have been very few studies that focus specifically on patients with poor outcomes. This is very unfortunate because there are a large number of patients with poor outcomes at every CI center around the world and their failure to display benefits with a CI is a legitimate and important research problem that needs serious attention by clinicians and researchers working on CIs, particularly the small number of researchers who study of individual differences and variability in outcomes following implantation.

Why have children and adults with poor outcomes after cochlear implantation received insufficient attention in earlier research studies? We believe there are two major reasons for this situation: First, if a child or adult fails to display rapid adaptation to the novel electrical stimulation provided by a CI, they may often fail to surface to the top of the priority list because there are few interventions or assessment protocols available to work with beyond basic general medical and audiological guidelines, such as a device integrity check of the CI to make sure it is functioning according to the manufacturers specifications, a follow-up neuroimaging study to verify that the electrode array has been properly inserted in the cochlear, unaided pure-tone audiograms in each ear, unaided word recognition scores in each ear and sentence scores in quiet and noise. In typical clinical care, no other diagnostic tests (especially neurocognitive or behavioral tests) are routinely carried out to identify the locus of the specific weakness or problem that is responsible for the poor outcomes.[8] It is often a difficult and challenging problem to find out precisely why an individual patient is doing poorly with his/her CI. What is wrong with the patient's device? Where is the problem located? Is there a device failure or something related to the surgical procedure and placement of the electrode array? More importantly, what kinds of interventions, treatments and aural rehabilitation protocols are available to improve outcomes for low-functioning CI patients who experience poor outcomes? These are a few of the first-line questions that neurotologists, audiologists and other health care providers are confronted with when a patient fails to make adequate progress after implantation.

The second reason that patients with suboptimal outcomes may receive insufficient attention is because doing this kind of clinical diagnostic research takes additional time and money. Beyond the initial expense of the implantation surgery, early mapping and routine follow-up. Insurance carriers frequently do not cover the costs involved in tracking down the cause of a poor outcome following implantation, and guidelines for such evaluation and intervention are limited by the dearth of explanatory and evidence-based treatment research.

Given our current understanding of the sensory, neurocognitive and linguistic factors that are responsible for the enormous individual differences and variability in outcomes after implantation discussed earlier in this paper under Challenge No. 1, it is very clear that we also need substantial new research efforts on assessment, diagnosis and treatment of the underlying cause of poor outcomes following implantation. Only after detailed audiological and neurocognitive assessments and careful diagnosis is it appropriate to consider options for novel targeted, individualized interventions and treatments of the patient with a suboptimal CI outcome. Every patient with hearing loss is different and every patient with a CI is likely to display a different profile of strengths, weaknesses, limitations and milestones in their peripheral and central auditory functioning as well as their neurocognitive and linguistic information processing skills. Other family and psycho-social factors may also contribute to poor outcomes too.[57,59–61] These domains need to be assessed and carefully evaluated as well along with quantitative measures obtained from behavioral tests.

Improving performance beyond the effects of practice alone (IPBEPA Effects)

Everyone who works in the field of CIs—the neurotologists, otologists, audiologists and neuroscientists believes that it should be possible, at least in principle, to improve speech recognition outcomes in patients who are doing poorly with their CIs. The key question and grand challenge is "how do you accomplish this goal?". What techniques, methodologies, protocols, training and test materials for aural rehabilitation of CI patients are available for use in children and adults with poor CI outcomes? What is currently available for immediate use in the CI clinic and research laboratory and what new methods and training materials need to be developed in the future to address these issues? These are pressing and important clinical issues in the field today that are not routinely discussed at professional conferences, workshops and meetings.

To take one example, the current generation of computer-based "auditory training" programs available through some cochlear implant manufacturers are basically all generic – "one-size-fits-all" in nature. These auditory training programs often provide very narrowly defined benefits to a small subgroup of some low-functioning patients. While some patients may show benefits from these general-purpose auditory training programs, many patients do not derive any benefits at all other than "practice effects" showing some improvement in performance on the specific tasks and specific stimulus materials that they were trained on but little, if any, evidence for generalization to novel stimuli or robust transfer-of-training (i.e., near or far transfer) effects to new stimulus materials and novel processing tasks that were not part of the original training protocol.[62,63]

Other potential interventions may focus on underlying neurocognitive abilities that support auditory processing and spoken language skills, such as executive functioning and working memory.[64] In a recent study, Kronenberger et al (2011),[65] for example, enrolled 9 prelingually deaf, early implanted children with CIs in the Cogmed Working Memory Training Program, a 5 week computer-based training program of working memory exercises.[66] Of note, the Cogmed program trains working memory adaptively, tailoring the difficulty of training to the maximum level of the subject's abilities as improvement occurs. During a 5-

week pretraining period (during which time children did not complete any additional training or exercises), the sample showed no improvement in verbal working memory or executive functioning skills. However, after 5 weeks of Cogmed training, the sample improved in verbal short-term memory (digit span forward), visuospatial short-term memory (spatial span forward and backward), parent-reported executive functioning/working memory (on a behavior checklist), and sentence repetition skills. However, only improvements in sentence repetition skills were retained at 6-month follow-up, and measures of generalization and transfer of the effect were limited. Furthermore, other studies of working memory and cognitive training in older adult CI users have produced less robust or contradictory results.[67] Thus, cognitive training interventions for CI users with suboptimal outcomes are in need of further refinement and investigation, particularly as they are related to generalization and transfer-of-training to real-world outcomes.

The long-term objective of any intervention program based on auditory, neurocognitive or linguistic training with hearing impaired listeners is improving performance beyond the effects of practice alone (i.e., "IPBEPA Effects"). Why is there no improvement beyond simple practice effects? The core design features of general-purpose auditory training programs like "LACE," "Angel-Sounds" and "Sound and Way Beyond" involve repeated practice and repetition of isolated words taken out of meaningful linguistic context. Training human listeners, and for that matter, even training speaker-independent speech recognition systems on isolated words taken out of context, is not going to generalize robustly to recognition of words in meaningful sentences or longer passages of connected fluent speech.[68–71] Furthermore, repetition practice just recognizing and identifying isolated words produced by only one high-intelligibility talker, even with immediate feedback in effect after each training trial, is unlikely to produce generalization and robust near- and far-transfer-of-training effects to novel materials produced by new talkers, unfamiliar talkers with marked regional dialects or non-native speakers who produce English with a foreign accent.[72]

What interventions can be recommended for adults and children who have poor outcomes following implantation? While we do not have complete answers to this very important central question at this point in time, we do know that the auditory brain is highly plastic, especially in infants, toddlers and young children and it should, in principle, be possible to develop an individualized targeted intervention protocol that "matches" the specific weaknesses of an individual patient who has a poor outcome after implantation.[5,6] However, before we can recommend any intervention protocol for treatment, we need to know what the patient's strengths, weaknesses, and milestones are across multiple information processing domains, some of which are directly dependent on audibility and sensory processing of the signal and others that are domain-general in nature such as verbal learning and memory processes, controlled attention and inhibitory control. Only after a complete comprehensive sensory and neurocognitive assessment and individual profile has been compiled on the patient will we be able to identify the underlying problems and then be in a position to recommend an evidence-based intervention protocol that targets the specific sensory and information processing domain and weaknesses. This is one of the most important and pressing challenges for future research in the field of CIs. Whether we can achieve this grand challenge and improve outcomes for patients with suboptimal outcomes remains to be seen in the future. For now, simply making this goal explicit here may encourage new research efforts on a pressing clinical problem that has been ignored in the past.

Summary and concluding remarks

A CI is not a passive sensory aid or sensory substitution device that simply replaces a damaged or defective cochlea to restore normal hearing. To achieve a successful outcome following implantation, all patients who receive a CI require a prolonged period of aural rehabilitation (AR) that involves perceptual learning, adaptation and readjustment of their attentional networks during which the brain and central nervous system undergo substantial reorganization and realignment to adapt to the highly-degraded, compromised, incomplete and sparsely-coded novel electrical input signal that is transmitted to the auditory nerve by the CI.[14] As Carol Flexer[73] observed a few years ago, hearing loss is not just an ear issue—it is a brain and information processing issue too. The emphasis and narrow focus on early registration and sensory encoding of acoustic signals by a CI has relegated other domains of perception and cognition, learning, memory and attention and inhibition to the sidelines.

It is well-known in the field of human speech perception and spoken language understanding that the "heavy lifting" in speech perception and spoken language processing and the seemingly robust performance routinely observed in normal-hearing listeners under a wide range of challenging conditions is carried out very rapidly and almost effortlessly by highly automatic down-stream predictive coding strategies that draw on the enormous reservoir of knowledge and prior experience and activities that have been pre-compiled and stored in the listener's long-term phonological and lexical memory.[71,74–76] When deaf children and adults receive a CI as a treatment for severe-to-profound hearing loss, they do not simply have their hearing restored at the auditory periphery. After implantation, they receive novel electrical stimulation to the auditory nerve and degraded and highly compromised neural representations of speech to specialized cortical areas of their brain that are critical for the development and processing of spoken language, specifically, automatized phonological processing and lexical retrieval skills that are used to rapidly encode, process, and reproduce speech signals linking up sensory and motor systems in new ways. Moreover, many different neural circuits in other areas of the brain also begin to receive inputs from the auditory cortex and brainstem, and these contribute to the overall global connectivity patterns and integrative functions linking multiple brain regions in regulating speech and language processes in a highly coordinated manner.

One of the most important foundational properties of human speech perception and spoken language processing is its robustness in the face of diverse acoustic stimulation over a wide range of listening conditions that produce significant degradations in the first-order sensory acoustic—phonetic and phonological properties of speech signals. In comparison to other information processing domains, speech perception is probably one of the most flexible and highly adaptive information processing skills that humans have developed over the course of evolution. At first glance, human listeners appear to adapt and compensate very quickly and effortlessly to large acoustic changes in the vocal sound source—the talker's gender, age, regional dialect, speaking rate and speaking style. Human listeners also rapidly adjust to numerous sources of acoustic degradation in the speech signal such as noise, filtering, reverberation and temporal and spectral degradation in their immediate listening environment without any significant loss of speech intelligibility. The talker's intended linguistic message is extremely robust and is highly resistant to many different sources of signal degradation. Moreover, normal hearing listeners are able to recognize and successfully understand speech from an enormous range of unfamiliar novel talkers, non-native speakers, as well as computer-generated synthesis-by-rule systems and they can do this under an enormously wide range of adverse and challenging listening conditions such as multi-talker babble or talking over the telephone with significantly reduced bandwidth and limited dynamic range. Human listeners can also successfully recognize and understand significantly degraded vocoded spectrally-transformed and spectrally-rotated speech as well as highly impoverished sine-wave replicas of natural speech, time-compressed speech and temporally-interrupted speech signals maintaining very high levels of intelligibility.[77—80]

These are several of the traditional benchmarks used for evaluating the robust speech perception skills of normal-hearing listeners and they serve as the gold-standard and foundation for evaluating the performance of sensory aids for the hearing impaired listeners as well as automatic speech recognition and spoken language understanding systems for machines.

The three challenges that we have considered in this paper demonstrate the pressing need to reassess and reevaluate the current narrative about CIs — where the field is right now and where basic and clinical research needs to go in the future. While enormous advances have been made over the last 25 years in the medical and surgical management of profound SNHL in both adults and children using CIs, it is now becoming necessary to rethink and reconsider the research agenda and narrative for the future and move beyond the narrow focus on hearing, audibility and the early registration and encoding of sensory input provided by a CI. Some of these changes in emphasis are already occurring in several research groups around the world who are now actively carrying out new research on the "auditory brain" and fully acknowledging the important contribution of cognition and brain function in speech recognition and spoken language understanding.[5,6]

To properly address the three challenges discussed here, we believe it is necessary to reevaluate several long-standing tacit assumptions that have guided research on cochlear implants. First, we need to rethink the clinical utility of conventional end-point product-based outcome measures that have been used universally to assess benefit following implantation. While there is a clear need to retain the endpoint product measures of outcome and benefit because they serve as useful clinical benchmarks of performance in the clinic and laboratory, new sustained and expanded research efforts also need to be redirected and focused on explaining variability in outcomes and on developing measures of early preimplant predictors of speech recognition outcomes that can be used to identify CI candidates who may be at high risk for a poor outcome following implantation. At the present time, other than conventional demographics and hearing history measures, we do not have any valid and reliable prognostic methods and tools available to us for predicting speech recognition and spoken language processing outcomes after implantation. This is a serious gap in our knowledge and understanding about the efficacy and effectiveness of CIs.

Second, we believe that the current battery of outcome measures should be broadened substantially to encompass other information processing domains related to cognition and cognitive hearing science, specifically, process measures of verbal learning, memory, attention, inhibitory control and executive functioning. New global measures of systems integrity and functional assessments need to be developed as well in order to measure how all the individual components of the language processing systems work together, especially in more complex linguistic tasks like language comprehension and learning from listening. Moreover, these new preimplant measures need to be validated in studies using large sample sizes so that normative data can be obtained to establish benchmarks and milestones for both children and adults that can be used to evaluate their strengths, weaknesses and limitations. Novel efficacious interventions cannot be recommended for patients with poor outcomes following implantation without knowing what is wrong and what systems should be targeted.

Finally, we believe that new intensive broad-based basic and clinical research programs need to be developed to study the poor CI performers in order to find out why they are doing poorly with their CIs and what novel interventions can be developed to improve their outcomes. Multidisciplinary research programs encompassing all of the relevant stakeholders in the new fields of Cognitive Hearing Science and Cognitive Audiology need to be created to focus new concentrated research efforts on both the adults and children who are doing poorly with their CIs in order to begin understanding why these particular patients fail to achieve adequate outcomes. Additionally, it is important for these research programs to determine how we can improve speech recognition and spoken language processing skills, as well as other information processing domains that are dependent on speech perception and spoken language comprehension such as executive function, controlled attention, inhibitory control, self-regulation and psycho-social functioning, core information processing domains that have received little attention in the past by researchers working in the mainstream of clinical research on CIs.

Researchers and clinicians working in the field of CIs also need to adopt a unifying vision and theoretically-sound conceptual framework for basic and clinical research on CIs in the future. A new narrative needs to be developed that is motivated by a well-defined theoretical and conceptual framework.[81] The enormous individual differences and variability routinely observed in outcomes following implantation are not mysterious, anomalous or idiopathic in nature. When framed within the context of cognitive information processing theory and the new emerging fields of Cognitive Hearing Sciences and Cognitive Audiology, it becomes possible to begin studying and understanding individual differences and variability in outcomes. Recent developments in cognitive science and cognitive neuroscience have also established the utility of viewing the development of speech and language as embodied processes linking brain, body, and world together as a functionally integrated system of perception and action. [82] There is every reason to believe that these new theoretical ideas will also provide fundamental new insights into the enormous variability and individual differences in outcome and benefit following cochlear implantation in deaf children and adults. Without knowing which specific biological and neurocognitive factors are responsible for the enormous individual differences in CI outcomes or understanding the underlying neurocognitive basis for variation and individual differences in performance, it is impossible to select a specific approach to habilitation and treatment after a deaf adult or child receives a CI. Deaf adults and children who are performing poorly with their CIs are not a homogeneous group and may differ in numerous ways from each other, reflecting the dysfunction of multiple brain systems associated with both congenital and acquired deafness and profound hearing loss. Moreover, it seems very unlikely that an individual patient will be able to achieve optimal benefits from his/her CI without researchers and clinicians knowing why a specific patient is having problems and what particular neurocognitive domains and information processing subsystems underlie these problems.

Acknowledgements

Preparation of this paper was supported by NIH NIDCD Research Grant R01 DC-000111-38 and the IU Grant Linking University-wide Expertise award to DBP and NIDCD R01 Research grant DC-015257-01 to DBP and WGK. Additional support was also provided NIH NIDCD Career Development Award 5K23DC015539-02 and the American Otological Society Clinician-Scientist Award to ACM and the 2017 American Neurotologic Society Research Grant to MSH. The authors would like to express their special thanks to Dr. Richard Miyamoto for his continued help and support for the long-standing program of research on cochlear implants at the IU School of Medicine in Indianapolis. Each of us has benefitted from the intellectual environment in the DeVault Lab since its founding in the mid 1980’s by Dr. Miyamoto and his research team.

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Edited by Xin Jin