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Verbal Short-Term Memory Development and Spoken Language Outcomes in Deaf Children with Cochlear Implants

Michael S. Harris, M.D.^a, William G. Kronenberger, Ph.D.^b, Sujuan Gao, Ph.D.^c, Helena M. Hoen, M.S.^c, Richard T. Miyamoto, M.D.^a, and David B. Pisoni, Ph.D.^{a,b,d}

^aDeVault Otologic Research Laboratory, Indiana University School of Medicine, Indianapolis, IN 46202

^bDepartment of Otolaryngology – Head & Neck Surgery Department of Psychiatry, Indiana University School of Medicine, Indianapolis, IN 46202

^cDivision of Biostatistics, Indiana University School of Medicine, Indianapolis, IN 46202

^dSpeech Research Laboratory, Department of Psychological and Brain Sciences, Indiana University, Bloomington, IN 47405

Abstract

Objectives—Cochlear implants (CIs) help many deaf children achieve near normal speech and language (S/L) milestones. Nevertheless, high levels of unexplained variability in S/L outcomes are limiting factors in improving the effectiveness of CIs in deaf children. The objective of this study was to longitudinally assess the role of verbal short-term memory (STM) and working memory (WM) capacity as a progress-limiting source of variability in S/L outcomes following CI in children.

Design—Longitudinal study of 66 children with CIs for pre-lingual severe-to-profound hearing loss. Outcome measures included performance on Digit Span Forward (DSF), Digit Span Backward (DSB), and four conventional S/L measures that examined spoken word recognition (PBK), receptive vocabulary (PPVT), sentence recognition skills (HINT), and receptive and expressive language functioning (CELF).

Results—Growth curves for DSF and DSB in the CI sample over time were comparable in slope, but consistently lagged in magnitude relative to norms for normal-hearing peers of the same age. For DSF and DSB, 50.5% and 44.0%, respectively, of the CI sample scored >1 SD below the normative mean for raw scores across all ages. The first (baseline) DSF score significantly predicted all endpoint scores for the four S/L measures, and DSF slope (growth) over time predicted CELF scores. DSF baseline and slope accounted for an additional 13%–31% of variance in S/L scores after controlling for conventional predictor variables such as: chronological age at time of testing, age at time of implantation, communication mode (AOC vs. TC), and maternal education. Only DSB baseline scores predicted endpoint language scores on PPVT and CELF. DSB slopes were not significantly related to any endpoint S/L measures. DSB baseline scores and slopes taken together accounted for an additional 4%–19% of variance in S/L endpoint measures after controlling for the conventional predictor variables.

Conclusions—Verbal STM/WM scores, process measures of information capacity, develop at an average rate in the years following cochlear implantation, but were found to consistently lag in

Corresponding Author: Michael S. Harris, M.D., DeVault Otologic Research Laboratory, Indiana University School of Medicine, Department of Otolaryngology – Head & Neck Surgery, Riley Research Wing 044, 699 Riley Hospital Drive, Indianapolis, IN 46202, Telephone: 317-274-1985.

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absolute magnitude behind those reported for normal hearing peers. Baseline verbal STM/WM predicted long-term endpoint S/L outcomes, but verbal STM slopes predicted only endpoint language outcomes. Verbal STM/WM processing skills reflect important underlying core elementary neurocognitive functions and represent potential intervention targets for improving endpoint S/L outcomes in pediatric CI users.

INTRODUCTION

With the widespread implementation of universal newborn hearing screening in the United States, approximately 12,000 children are identified as deaf or hearing-impaired each year (Shulman et al. 2010). The academic, social, and economic impact of deafness is profound and far-reaching on an individual and national basis (Mohr et al. 2000; Kritzer 2009). Cochlear implants (CIs) are now a well-established and widely available surgical intervention for adults and children with severe-to-profound (pure tone average thresholds of 70 dB HL) sensorineural hearing loss (SNHL). Cochlear implants have helped deaf children realize previously unattainable speech and language (S/L) developmental milestones.

Despite intervention with CIs, many deaf children fail to achieve typical S/L milestones even when tested under optimal listening conditions (Blake and Gordon, 2007; Peterson et al. 2010; Venail et al. 2010). Current candidacy criteria established by the Food and Drug Administration (FDA) include pre- or post-lingual deafness or severe-to-profound SNHL at mid-to-high frequencies (1000–8000 Hz) with limited benefit from a 6 to 12-month trial with well-fit bilateral hearing aids (NIH Consensus Panel 1995; Kral and O'Donoghue, 2010). Since CI technology was first extended to children as young as 2 years of age with FDA approval in 1990, indications for use of CIs have evolved to allow for implantation for children as young as 12 months (NIH Consensus Panel, 1995). The cost-utility of CIs compares favorably to many other implantable medical devices such as pacemakers and implantable defibrillators (Cheng et al. 2000; Bond et al. 2009).

In the years since CIs have become a routine clinical treatment for deafness and severe-to-profound hearing loss in children, several major findings about the effectiveness of CIs have been reported consistently in the literature. These findings have formed the basis for a small set of conventional predictors of S/L performance following cochlear implantation. First, age at implantation is crucially important for good outcomes. Earlier implanted children typically demonstrate better S/L performance relative to late implanted children (Niparko et al. 2010; Nikolopoulos et al. 2010; Peterson et al. 2010). Second, S/L abilities emerge gradually after implantation, reflecting the fact that children need to learn to use their devices over time to maximize both speech perception and speech intelligibility (Fagan and Pisoni, 2010; Peterson et al. 2010). Third, post-implantation linguistic and social experiences and activities are significant in facilitating optimal S/L outcomes. Children who are exposed to a language-learning environment that emphasizes an Auditory-Oral communication (AOC) approach perform consistently better on a wide range of speech perception and spoken language measures relative to children raised in Total Communication (TC) settings, which principally consist of manual communication (i.e., Signed Exact English or, less commonly, American Sign Language) along with speech (Wheeler et al. 2009). Fourth, there is an enormous degree of variability and individual differences in CI benefit in the acquisition of S/L, telephone proficiency, speech perception in noise, environmental sound awareness, and music appreciation (NIH Consensus Panel 1995).

Niparko and his colleagues recently reported a prospective, longitudinal study that assessed both receptive and expressive spoken language development following cochlear

implantation in a large cohort of deaf children and compared these findings to a group of age-matched normal-hearing, typically-developing children (Niparko et al. 2010). Replicating earlier findings, they reported that earlier age at implantation, shorter periods of hearing loss, greater residual hearing prior to surgery, higher ratings of parent-child interactions, and higher socioeconomic status were all associated with better performance on both expressive and receptive measures of spoken language development. Importantly, their data also demonstrated that even after accounting for the conventional predictors associated with demographic, medical, and device factors, a substantial and clinically-significant degree of unexplained variability and individual differences in S/L outcomes still remained in deaf children with CIs that was not observed in the normal-hearing, typically-developing peers, who showed close to ceiling levels of performance on these outcome measures. The factors responsible for the large variability and individual differences in S/L outcomes, however, are unclear at the present time and remain a critical barrier to further progress in the field as emphasized by the National Institutes of Health (NIH) Consensus Development Conference Statement on CIs in 1995 and the American Speech-Language-Hearing Association's Technical Report on CIs in 2004 (NIH Consensus Panel 1995; American Speech-Language-Hearing Association, 2004).

Why do some children with CIs demonstrate suboptimal S/L outcomes despite the presence of an apparently ideal set of conventional demographic and medical predictors? Current research efforts have begun to focus attention and resources on extending our understanding of the factors that underlie the wide range of variability in S/L performance following implantation beyond the small set of conventional indicators used in the past (Peterson et al. 2010). For both children and adults with deafness or hearing loss, *efficacy* – the power of a treatment or intervention to produce a desired effect – has now been well-established for CIs (Niparko et al. 2010; Nikolopoulos et al. 2010). There is still considerable room for progress, however, in improving the *effectiveness* of CIs – the ability to reliably and consistently produce optimum outcomes in all candidates in everyday, real-world settings (Pisoni et al. 2008).

Recent evidence suggests that individual differences following cochlear implantation are not anomalous, mysterious, or idiopathic, but represent systematic underlying differences in several core elementary neurocognitive processes that influence performance in a wide range of traditional S/L outcome measures (Fagan et al. 2007; Conway & Pisoni, 2008). Short-term memory (STM) and working memory (WM), for example, are core foundational constructs developed in the field of cognitive psychology that have been found to be central to speech perception and spoken language functioning (Baddeley, 2007; Cowan, 2005; Dempster, 1981; Gathercole & Baddeley, 1993). Together STM/WM serve as the temporary holding area for incoming and outgoing verbal information, as well as the storage space for linguistic information during real-time immediate processing of spoken language (Alloway et al. 2009). Short-term memory involves the storage and retrieval of information over short periods of time; WM requires additional manipulations and/or transformations of information in STM as well as retention and retrieval of information over short periods of time (Alloway et al. 2009).

Cognitive psychologists study perception, attention, learning and memory within a theoretical framework known as human information processing (Haber, 1969). One of the fundamental principles of information processing theory is that neurocognitive processes such as sensation, perception, memory, thought, and other complex processing activities should be viewed as representing a continuum of cognitive processing (Haber, 1969). These processing activities are assumed to be mutually interdependent and cannot be easily divided up into separate subsystems. Furthermore, an analysis of one subsystem, such as perception, cannot be carried out successfully without incorporating the contributions of the other major

subsystems such as memory, attention, learning and cognitive control (Lachman, Lachman, & Butterfield, 1979). This approach to studying human cognition has also provided a variety of novel conceptual tools for theorizing about the structures and processes involved in complex cognitive activity and the underlying psychological phenomena. Information processing theories are concerned with an analysis of “central processes” of large complex systems used in visual object recognition, perceptual learning and memory, speech perception and spoken language processing (Haber, 1969). A common goal of this theoretical approach is to investigate and understand the neural and cognitive representations and elementary neurocognitive processes and cognitive structures that are used in these kinds of complex cognitive activities and to trace out and describe the time-course of these processing operations (Pisoni, 2000).

Within this broad theoretical framework, STM/WM is viewed as a highly flexible multi-component information processing system that is used to temporarily store and process verbal and visual-spatial memory codes for short periods of time (Baddeley, 2007; Cowan, 2005). In particular, WM is used to support a wide range of everyday cognitive activities such as listening and speaking, reading and writing, mathematical cognition, problem solving, decision-making and complex thought processes (Baddeley, 2007). STM and WM are often considered as the mental workbench or workspace of the mind because they play mandatory roles in language processing, thought and action. Large individual differences are routinely found in measures of the capacity and efficiency of STM/WM, reflecting underlying neurobiological sources of variability in more basic elementary processes involved in attention, learning, memory, and cognitive control that are routinely used in encoding, storing and processing information for on-going real-time neurocognitive operations (Baddeley, 2007; Cowan, 2005; Gathercole & Baddeley, 1993).

Over the last thirty years, a substantial body of research has documented the close links and central role of STM/WM as the interface between memory and language processing in a wide variety of behavioral tasks such as word recognition, vocabulary development, sentence comprehension and language production (Gathercole & Baddeley, 1993). Many of these findings have been incorporated in a well-known and highly influential model of WM developed by Baddeley and his colleagues (Baddeley, 2007; Gathercole & Baddeley, 1993). The current multicomponent model of WM consists of four subcomponents: (1) a domain-general central executive that controls attention and processing activities and regulates the flow of information in the processing system, (2) the phonological loop that is used for the temporary storage of verbal phonological memory codes, (3) the visual-spatial sketchpad that maintains and processes visual and spatial representations, and (4) an episodic buffer that is used to integrate and bind memory codes from different processing domains into larger chunks of information.

Evidence for the critical role of verbal phonological short-term memory in a wide range of speech and language processing tasks has been accumulating steadily over the years since the seminal publication of Baddeley, Gathercole and Papagno (1995). In this paper, Baddeley et al. proposed that language learning, specifically, learning of new spoken words, requires the use of a temporary specialized verbal short-term memory system, the “phonological loop,” which encodes, stores and processes phonological representations of speech. The phonological loop in Baddeley’s WM model serves as the gateway or interface between the early sensory registration for speech-- the initial encoding of the speech waveform by the auditory system, and more stable and permanent auditory-verbal-linguistic phonological memory codes for speech sounds and spoken words stored in permanent long-term memory. As a result, components of verbal STM and WM, specifically the phonological loop and the active control processes of the central executive, are core factors underlying the efficiency of language processing and, over a long period of time, verbal

development and language learning (Cowan, 2005; Gathercole & Baddeley, 1993). Disturbances and delays in these core elementary components of verbal STM/WM have been demonstrated to adversely affect language development in a variety of domains, including receptive and expressive vocabulary, reading, speech production, and phonological processing (Gathercole & Baddeley, 1993). Verbal STM/WM has been shown to be particularly important for spoken language development in children who are already vulnerable to speech-language delays as a result of degraded auditory input and hearing impairment (Pisoni, Kronenberger, Roman, & Geers, 2011).

The development of verbal STM/WM during childhood represents a significant cognitive achievement that facilitates the development of a broad set of related language skills (Gathercole & Baddeley, 1993). During the grade school years, children show dramatic increases in their ability to remember verbal information, as a result of a complex interplay of cognitive processes ranging from phonological loop capacity to controlled attention to coding and chunking in STM/WM (Cowan, 2005; Dempster, 1978, 1981). Increases in verbal STM/WM capacity and processing efficiency are accompanied by similarly dramatic improvements in speech-language skills (Gathercole & Baddeley, 1993). In these studies, age is likely a proxy for the developing brain and the accumulation of experience that promotes the growth of robust verbal memory skills. Thus, factors that reduce access to auditory-phonological experiences and activities are likely to impair and delay the growth of verbal STM/WM skills, which is then likely to produce a downstream impact on the development of speech and language skills which depend on these basic foundational elementary information processing skills (Pisoni, Kronenberger, Roman et al., 2011).

In one of our earlier studies, we measured the capacity of STM/WM in deaf children with cochlear implants and found large differences in verbal STM/WM capacity in a group of 176 early implanted 8- and 9-year old children who had used their CIs for more than five years (Pisoni & Cleary, 2003). Both forward and backward auditory digit spans were atypical and significantly shorter than the memory spans obtained from a group of age-matched typically-developing normal-hearing children. Moreover, measures of verbal STM/WM in these deaf children were found to be strongly correlated with several different conventional speech and language outcome measures obtained concurrently with the digit span scores (Pisoni & Cleary, 2003). Pisoni & Cleary (2003) argued that the atypical forward and backward digit spans which are assumed to measure the information processing capacity of verbal STM/WM reflected more basic differences in processes used for rapid phonological encoding and storage of phonological memory codes in verbal STM/WM.

Verbal working memory is related, in part, to factors such as encoding efficiency, memory scanning speed, and subvocal rehearsal speed, which allow the individual to rapidly find information in memory and to rehearse more information in the same duration of time, respectively (Cowan et al., 1994, 1998). Children with CIs have much slower subvocal verbal rehearsal speed and longer verbal memory scanning times than normal hearing children, likely as a result of degraded or poorly specified phonological representations of spoken language (Burkholder & Pisoni, 2003; Pisoni et al., 2011). Furthermore, verbal rehearsal speed was found to be strongly related to both DS performance and to speech and language outcomes in children with CIs (Pisoni et al., 2011). Additionally, improvement in verbal working memory is associated with improvement in memory scanning and articulation rate (Kronenberger et al., 2010). These findings suggest that working memory may be related to speech-language skills by reflecting information processing capacity, or “the degree to which the individual is able to rapidly and fluently (efficiently) encode, store, maintain, and retrieve phonological and lexical representations from short-term working memory (Pisoni et al., 2011, p. 71S).” Because this capacity provides for access to speech and language learning experience as well as retention of speech and language information

during verbal processing, it is likely to relate to speech and language outcomes (Pisoni et al., 2011).

STM and WM have been found to be critical for the development of speech perception and spoken language (Gathercole & Baddeley, 1993; Pisoni & Geers, 2000; Pisoni & Cleary, 2003). The strong relations between STM/WM and the development of S/L skills has been demonstrated in numerous studies with normal-hearing children (Gathercole et al. 2006; Alloway et al. 2009). Because the development of verbal STM/WM is strongly dependent on early auditory experiences and activities, children who experience a period of auditory deprivation along with degraded auditory input following cochlear implantation are at high risk for disturbances and delays in verbal STM/WM processes (Bavelier et al. 2008). Deaf children with CIs also have been shown to exhibit significant delays and disturbances in several other closely related neurocognitive processes such as executive functioning, theory of mind, mathematics, reading, and behavioral and emotional regulation (Pisoni & Geers, 2000; Pisoni & Cleary, 2003; Cleary et al. 2005; Fagan et al 2007; Watson et al. 2007; Bavelier et al. 2008). As a result, delays and deficits in verbal STM/WM may be one critical link or interface between degraded auditory input and S/L development.

Identifying differences in basic underlying neurocognitive processes such as verbal STM/WM may provide a better understanding and more precise explanations of variability and individual differences in deaf children who experience heretofore unexplained suboptimal S/L outcomes following cochlear implantation. The clinical implications of these findings would be significant, allowing for improved evaluation, treatment, and ongoing management of S/L development following cochlear implantation by integrating neurocognitive factors into conventional clinical assessments and treatment models. Ultimately, improved prediction of who will do well with a CI and who may be likely to struggle to obtain optimal benefits will lead to development and earlier initiation of novel targeted intervention strategies that are focused on the underlying causal factors responsible for delays in S/L (Kronenberger et al. 2010).

Recent work by our research team examined verbal digit span (DS), a well-established neurocognitive measure of information processing capacity that directly reflects verbal STM/WM processes (Cowan, 2005; Dempster, 1978), and verbal rehearsal speed in deaf children after more than 10 years of CI use (Pisoni et al. 2011). The DS test consists of two parts, a Digits Forward (DSF) subtest which requires subjects to repeat spoken sequences of digits in forward order, and a Digits Backward (DSB) subtest which requires subjects to repeat spoken sequences of digits in reverse order. Digit Span was chosen as a measure of verbal STM/WM capacity because of its brevity and sensitivity to short-term/working memory deficits in children (Kaufman, 1990; Sattler, 1992; Wechsler et al., 2004). There are several limitations of the DS test (see, for example, Reynolds, 1997): DSF and DSB, while correlated, are affected by somewhat different factors. DSF measures rote-sequential verbal short-term memory capacity with minimal interference or competition, whereas DSB includes an additional concurrent processing component (reversing digits) that introduces interference and competition, and requires more active involvement of executive control processes (Kaufman, 1990; Wechsler et al., 2004). Furthermore, DSB places limited and specific demands (reverse sequencing) on executive functioning, and a broad assessment of verbal working memory may include other types of demands on executive functioning and methods of blocking rehearsal (e.g., Wechsler et al., 2004). DS performance is not based on a single neurocognitive ability, but rather can be affected by multiple factors, including anxiety, familiarity with numbers, memory, attention, and sequencing ability (Sattler, 1992). Nevertheless, DS scores have been consistently validated as measures of verbal STM/WM capacity and are a mainstay of neurocognitive and memory testing in adults and children (Kaufman, 1990). Consistent with the empirical research on DS subtests and

recommendations of numerous authors (e.g., Reynolds, 1997), we analyzed results for DSF and DSB separately in this report.

Using DS as a measure of verbal STM/WM capacity in children with CIs, we found that longest DS forward scores at ages 8 to 9 years were significantly correlated with all S/L outcomes in adolescence, but backward digit spans correlated significantly only with measures of higher-order language functioning over that time period (Pisoni & Cleary, 2003; Pisoni, Kronenberger, Roman, et al., 2011). The current study substantially extends the earlier descriptive work by our group and others by examining the longitudinal time course of learning and growth in verbal STM/WM capacity in a different sample of children with CIs, using multiple DS data points over time, in order to model the contribution of both baseline and growth in verbal STM/WM in predicting later speech and language outcomes.

Development and growth (as opposed to static, single time-point measurement) of verbal STM/WM capacity is an important and unexplored area in deaf children with CIs. Whereas the first, or baseline, measure of DS in a longitudinal series of measurements reflects the initial starting point of verbal STM/WM capacity, such a single measure does not capture the contribution of growth and development of that core capacity over a longitudinal time period of child development. It is possible, for example, to have high initial STM/WM capacity but to show little subsequent growth of that cognitive ability, or conversely, to have low initial STM/WM capacity but rapid developmental growth. Therefore, with the exception of ceiling effects (which have minimal effects on measures of DS until mid to late adolescence), the baseline level and growth rate of DS are not necessarily related in any specific individual. Furthermore, both baseline (as a proxy for basic, absolute immediate memory capacity) and growth (as a proxy for trajectory of development of that capacity) of verbal STM/WM may underlie the development of endpoint S/L outcomes in children with CIs. Studies with normal hearing children, for example, show that STM/WM performance develops rapidly throughout childhood (Dempster, 1978) and that this developmental process is important for the growth of later speech and language skills (Gathercole & Baddeley, 1993).

Using four different conventional endpoint S/L outcome measures, we sought to address two important unanswered questions about variability and individual differences in the effectiveness of CIs: First, how does verbal STM/WM capacity change over time in a large group of deaf children with CIs, and how does this developmental trajectory compare to normative benchmarks based on the performance of typically-developing peers with normal hearing? Based on prior findings demonstrating significant lags in DS scores in both children and teenagers with CIs (e.g., Pisoni et al., 2011), we hypothesized that verbal STM/WM capacity in deaf children with CIs would lag consistently behind norms throughout childhood and early adolescence. Second, what components of STM/WM growth predict endpoint future performance on S/L measures, and what degree of previously unexplained variance does this account for beyond the small set of conventional demographic, medical and device predictors? We hypothesized that baseline (e.g., first) measures of DS would predict endpoint future performance on S/L measures, consistent with earlier findings (e.g., Pisoni et al., 2011). However, there have been no studies of growth rate of DS (or any verbal STM/WM measure) and endpoint S/L outcomes. Recognizing that there is an absence of guiding prior research, we expected that children with higher growth rates of DS (as a measure of verbal STM/WM) would also have better endpoint S/L outcomes.

We chose endpoint (i.e., final visit) S/L performance as the criterion for this study in order to test a predictive model of S/L outcomes, as opposed to a growth model of S/L development. In other words, we sought to evaluate the relations between verbal STM/WM development and the final endpoint of S/L skills in our sample, as opposed to where the S/L

skills started or how the S/L skills reached those endpoints. Endpoint S/L measures are important because they show the final level of speech and language functioning for the time period studied. They also allow us to investigate of how well verbal STM/WM predicts future speech and language functioning, as opposed to concurrent speech and language functioning.

METHODS

Study Participants

The sample consisted of 66 deaf children who received CIs at a large university-based hospital, using FDA criteria in place at the time of their pre-operative evaluation. All subjects completed the Wechsler Intelligence Scale for Children, Third Edition (WISC-III) Digit Span subtest (see below) following implantation. In order to be considered eligible for this study, participants had to have: (1) a pre-lingual onset (under age 3 years) severe-to-profound hearing loss bilaterally, (2) age at time of implantation ≥ 8 years (in order to select children who received a CI early in schooling), (3) a monolingual English home environment, (4) use of a currently available, state-of-the-art CI system in the judgment of study personnel (Otolaryngologists, Speech-Language Pathologists, and/or Audiologists), and (5) participation in a rehabilitative program (speech-language therapy and educational setting) that encouraged the development of spoken language and listening skills. Additionally, in order to be eligible for the current study, participants had to provide at least 3 digit span assessments (mean=4.5, SD=1.3) over a 2 year period or more (mean=3.9 years, SD=1.7); only digit span assessments conducted at least 12 months post-implant were used, in order to allow time for subjects to accommodate to their CI device. Digit span measurements for a specific time point were included only if a subject achieved a raw score above zero on either DSF or DSB, in order to ensure that subjects were capable of performing the digit span task at that time point (e.g., that subjects were oriented and fully understood the task). Only visits after the age of 6 years were considered for study inclusion because the digit span task used in this study was designed for the 6–16 year age range (Wechsler, 1991). Potential participants were excluded from the study if they had any additional pediatric or neurological conditions (including intellectual delays and/or developmental disabilities) that impacted quality of life or day-to-day functioning.

Study Design & Procedure

Data for the present study were obtained from a longitudinal research project investigating speech and language development in children with CIs. As a part of this study, repeated measures of Digit Span Forward (DSF) and Digit Span Backward (DSB) as well as four conventional clinical S/L outcome measures were obtained (along with other speech and language data collection not used in the present study) at research appointments scheduled at 6-month intervals for post-implant years 0–7 and once a year thereafter until 12 years of age. An attempt was made to obtain complete data for all participants, who were called for scheduling at regular time intervals per the research protocol. However, some participants missed (or elected to skip) visits; some participants did not enroll in the study until their children were older; and some participants were unable to provide full data at the time of their testing because of fatigue, time constraints, difficulty understanding directions, etc. This resulted in variability in age at first testing, number of times evaluated, and ages at time of evaluation. In order to address possible effects of this variability, we used statistical analyses that are not dependent on evaluation at fixed intervals, and we controlled for age variables in regression analyses. Additionally, not all subjects completed S/L outcome measures at the final visit, for reasons stated above.

The mean number of test sessions was 4.53 (SD = 1.27; Range = 3.0–9.0). Data from evaluations prior to cochlear implantation were not considered in the present set of analyses because we were primarily interested in predicting endpoint S/L performance based on growth (slope) of DS over time following implantation. All tests were administered by licensed speech-language pathologists with expertise in hearing impairment in children and CIs. The study was approved by our university Institutional Review Board (IRB), and written informed consent was obtained from parents prior to initiation of any study procedures including data collection.

Measures

Verbal STM & WM—The WISC-III Digit Span (DS) subtest requires the child to reproduce progressively longer lists of digits that are spoken live-voice (with the child able to view the examiner's face) by the test administrator at a rate of approximately one digit per second (Wechsler, 1991). The task consists of two recall conditions: Digits Forward (DSF) and Digits Backward (DSB). The DSF task requires subjects to repeat a sequence of random digits between 1 and 9 (inclusive) in forward order, beginning with a 2-digit sequence. Two items are presented for each sequence length, and if subjects reproduce at least one item correctly, the sequence length is increased by one until subjects repeat both items incorrectly at the same sequence length. The DSB task is identical to the DSF task except that subjects must reproduce the sequences in reverse order.

Digit span is one of the most widely used cognitive psychological tests (Kaufman, 1990), and its psychometrics in normal hearing samples have been well studied, demonstrating good to excellent reliability and validity (Sattler, 1992; Wechsler, 1991; Wechsler et al., 2004). It is administered and scored following highly standardized instructions, which promote a high degree of reliability in administration (Wechsler, 1991; Wechsler et al., 2004). In the present study sample, the test-retest reliability of DSF was ($r=$) 0.69 for the first to second assessment visits (average of 0.92 years between assessments, SD=0.32) and 0.69 for the second to third assessment visits (average of 1.05 years between assessments, SD=0.28); these values compare favorably with DSF test-retest values reported for the WISC-IV normative sample over a much shorter (32 day) time period ($r=$ 0.72; Wechsler et al., 2004). The test-retest reliability of DSB in the present study sample was 0.60 for the first to second assessment visits and 0.59 for the second to third assessment visits; these values also are comparable to DSB test-retest values reported for the WISC-IV normative sample over a shorter (32 day) time period ($r=$ 0.67; Wechsler et al., 2004).

In samples of children with CIs, digit span is the most widely studied measure of verbal short-term/working memory (Kronenberger, Pisoni, Henning, Colson, & Hazzard, 2011; Pisoni & Cleary, 2003; Pisoni, Kronenberger, Roman, et al, 2011). In a cross-sectional study, Pisoni & Cleary (2003) demonstrated that digit span scores are approximately normally distributed in samples of children with CIs and that mean DSF and DSB scores are stable across same-age subsamples of children with CIs. Digit span scores are strongly related to measures of speech-language outcomes, verbal rehearsal speed, and articulation speed, demonstrating construct validity (Pisoni, Kronenberger, Roman, et al., 2011; Pisoni & Cleary, 2003). DSF scores are improved following targeted working memory training in children with CIs, indicating that DS is sensitive to working memory processes in the CI population (Kronenberger et al., 2011). In the current study, DSF and DSB raw scores (the total number of items correctly reproduced) were used as measures of verbal STM and verbal WM capacity (St. Clair-Thompson, 2010), respectively

Speech Perception & Language Outcomes—Four conventional, standardized, behaviorally-based S/L outcome measures commonly used in clinical assessment of

performance following cochlear implantation were obtained from each child in order to assess a broad range of speech perception and language skills considered to be “gold-standard” speech-language outcomes in children following cochlear implantation (Davidson et al., 2011; Geers & Sedey, 2011): spoken word recognition, sentence recognition, receptive vocabulary, and broad/complex language processing. The four S/L measures selected for this study have been very widely used and have well-documented psychometric properties including reliability and validity (Kirk & Choi, 2009). For the present set of analyses, only the final test session’s performance was considered in this report.

Stimuli for the spoken word recognition test were presented live-voice, auditory only (lips hidden) consistent with standardized instructions. Stimuli for the sentence recognition test were presented digitally over a loudspeaker in soundfield at 65 dB at a distance of about 3 feet. Stimuli for the vocabulary and language tests were presented in the child’s preferred communication mode (either live voice or live voice + signed exact English). Participants were tested with CI alone (no other assistive devices).

1. Spoken Word Recognition

The Phonetically Balanced Kindergarten (PBK) Word test (N=45) was used to measure open-set recognition of 50 monosyllabic isolated words presented live-voice (Haskins, 1949). Performance is scored in terms of either the percentage of phonemes (speech sounds) or the percentage of words identified correctly. For this study, the PBK Word score (percentage of words identified correctly out of the total words administered) was used. As with the Hearing in Noise Test for Children (HINT-C) discussed below, there are no published normative values for the PBK, although it is a mainstay of pre-implantation assessment (e.g. current US FDA CI candidacy criteria for children require <30% correct on PBK, HINT-C, or a similar “age appropriate” word list) as well as post-implantation follow-up assessments.

2. Receptive Vocabulary

The Peabody Picture Vocabulary Test (most current revision used, depending on date of testing: 57 PPVT-III, 3 PPVT-IV) was used to evaluate each child’s single-word receptive vocabulary knowledge (Dunn and Dunn, 1997). The child was required to select one drawing from a series of four alternatives, identifying the picture that corresponds to the word spoken or signed and spoken (based on the child’s preferred communication modality), by the test administrator. PPVT standard scores (population mean = 100, population SD = 15) were used for outcomes analyses.

3. Sentence Recognition

The HINT-C (N=37) was used to measure children’s ability to perceive and immediately reproduce meaningful recorded English sentences presented in quiet (Nilsson & Sullivan, 1994). Children who concurrently used a hearing aid and a CI in the contralateral ear were administered sentence lists under 3 listening conditions: hearing aid alone, CI-alone, and combined use of both hearing aid and CI. For the purposes of these analyses, only the data from the CI-alone condition was considered. As with the PBK test described above, performance on the HINT-C was reported in terms of the percentage of words recognized correctly; there are no published normative values for this test.

4. Receptive and Expressive Complex Language Processing

The Clinical Evaluation of Language Fundamentals-3 (CELF-3; N=38) was used to evaluate a child’s linguistic knowledge and use of the basic foundations of language (Semel et al.

1995). The CELF-3 consists of several subtests that yield subdomain scores for receptive language (i.e., the ability to demonstrate comprehension of sentence structure, concepts and directions, and word classes) and expressive language (i.e., the ability to recall spoken sentences, use appropriate word structure, and formulate grammatical sentences). An overall CELF-3 global score (Core Language Score, expressed as a standard score based on norms) is derived from these subtests, which was used for all analyses reported here.

Reasons for missing data points across test intervals typically fell into one of three categories: (1) participant's disposition at the time of testing precluded completion of the test measure, (2) time restrictions prevented completion of the measure at a given interval, or (3) the child and parent were not able to attend the scheduled testing session.

Statistical Analysis

Modeling Developmental Growth of DS Scores—Overall growth curves of DSF and DSB raw scores across chronological ages (see Figure 1) and across duration of CI use (i.e., “hearing age;” see Figure 2) were modeled using mixed-effects models from multiple assessments, specifying random slopes and intercepts to account for within individual variability in development of DSF and DSB over time. Models of linear and quadratic age effects both fit the data almost identically, differing only at the tails of the distribution where fewer data points were available. Therefore, the linear model was used in the present analyses because it was the most parsimonious.

To compare our study participants' performance on DSF and DSB to a benchmark normative growth curve to assess developmental milestones, we used cross-sectional normative values obtained from the WISC-III standardization sample (Kaplan, Fein, Kramer, Delis, & Morris, 1999). Mean raw scores for each age range of the WISC-III norms were derived from regression equations, based on norm tables that give Scaled Scores (population mean = 10, population SD = 3) for raw score values at the different ages (Kaplan et al., 1999). Scores based on samples of normal-hearing, typically-developing children were also obtained for the PPVT and CELF-3 from published manuals (Dunn and Dunn, 1997; Semel 1995). No norms exist for the PBK test or the HINT-C; results on these measures are clinically reported in terms of percentage correct.

Predicting Speech & Language Outcomes—A two stage analysis approach was used to predict S/L outcomes obtained at the last visit based on characteristics of DS growth from repeated DS scores obtained prior to the last visit. In stage one, individual slopes were computed for each child using linear regression analysis on DS measures prior to the last visit when the S/L outcomes were obtained. In addition, a baseline DS scaled score was derived using age-specific WISC-III norms for the first DS measure completed by each child. The slope parameter provides an estimate of change in DS performance over time, whereas the baseline DS scaled score reflects a child's STM/WM capacity relative to normal-hearing children at baseline (first DS assessment at or after age 6 years). In stage two of the analysis, regression models were used to model each of the four S/L outcome measures from the last test session. In addition to DSF and DSB baseline and slope estimates, predictor variables in the regression models included: age at the time S/L measures were obtained, age at implant, communication mode (AOC vs. TC), and maternal education. In order to keep the number of predictor variables to a minimum, age of onset of deafness and best pre-operative Pure Tone Average (i.e., average hearing level at 500 Hz, 1000 Hz, and 2000 Hz) were not entered as predictor variables in the regressions because of nonsignificant correlations ($p > 0.15$ for all correlations) with all four of the S/L outcome measures.

R^2 values were calculated for the “full” model, representing the contributions of the conventional CI outcome predictors together with the two novel predictors—DS baselines and DS slopes. Values for the increase in R^2 from the addition of DSF or DSB baseline and slope scores were also determined in order to provide an estimate of the unique variance in S/L outcomes accounted for by the STM/WM measures above and beyond the variance predicted by the conventional CI outcome predictors. Statistical analyses were performed using SAS software, Version 9.2.

RESULTS

Sample Characteristics

Study participants' characteristics are summarized in Table 1. Most of the sample ($N=54$; 82%) were deaf at birth; five children (8% of the sample) had onset of deafness between ages 2 and 3 years. Approximately 15% of the sample was implanted between ages 1 and 2 years ($N=10$); 24% ($N=16$) were implanted between ages 2 and 3 years; 20% ($N=13$) were implanted between ages 3 and 4 years; 27% ($N=18$) were implanted between ages 4 and 6 years; and 14% ($N=9$) were implanted between ages 6 and 8 years.

Examination of the bivariate correlations among participant characteristics showed expected relations between time, age, and duration variables. Specifically, participants who were older at their first visit were more likely to have received their CIs at older ages ($r=0.62$, $p<0.001$), have a longer duration of deafness prior to implant ($r=0.60$, $p<0.001$), have had their implant for longer period of time at the time of the first visit ($r=0.26$, $p<0.05$), and be older at the final study visit ($r=0.64$, $p<0.001$). Participants who were older at the final study visit were more likely to have received their CIs at older ages ($r=0.42$, $p<0.001$), have a longer duration of deafness prior to implantation ($r=0.32$, $p<0.01$), have had their implant for longer period of time at the time of the final visit ($r=0.69$, $p<0.001$), have been followed in the study for a longer period of time ($r=0.76$, $p<0.001$), and have been followed for more visits ($r=0.54$, $p<0.001$). Participants who were older at the time of implantation had a longer period of deafness prior to implant ($r=0.92$, $p<0.001$), better PTA thresholds ($r=-0.50$, $p<0.001$), and a shorter period of implant use prior to the first ($r=-0.59$, $p<0.001$) and last ($r=-0.38$, $p<0.01$) study visits. Participants with later age of onset of deafness had a shorter period of deafness prior to implant ($r=-0.33$, $p<0.01$) and a longer period of time during which they were followed for the study ($r=0.28$, $p<0.05$). Participants with a longer period of time from deafness to implant had better PTA thresholds ($r=-0.48$, $p<0.001$) and shorter durations of use of CIs at the first ($r=0.52$, $p<0.001$) and last ($r=0.41$, $p<0.001$) study visits. Participants with longer duration of CI use at the first study visit had higher (worse) PTA thresholds ($r=0.64$, $p<0.001$) and fewer study visits ($r=-0.32$, $p<0.01$), whereas participants with longer duration of CI use at the final study visit had higher (worse) PTA thresholds ($r=0.49$, $p<0.001$) and were followed in the study for a longer period of time ($r=0.76$, $p<0.001$) and for more visits ($r=0.41$, $p<0.001$). Finally, participants who were followed in the study for a longer period of time had more study visits ($r=0.79$, $p<0.001$); a table of all correlations between participant characteristics is available from the authors).

Modeling Developmental Growth of DS Scores—Figure 1 shows individual slopes based on DSF and DSB raw scores by chronological age for all 66 subjects over time fit with linear regression models. Each line (with the exception of the solid/bold line) represents fitted digit span raw scores of a child over time based on linear regression. The solid/bold line represents the average slope of the entire CI sample from a mixed effects repeated measures model. The DSF scores of the CI sample consistently lagged approximately one standard deviation behind norms at all ages (Figure 1, top panel),

whereas the DSB scores ranged from approximately one-half to one standard deviation behind norms (Figure 1, bottom panel).

In order to further evaluate DSF and DSB in our CI sample relative to norms, we compared the CI sample's DS scores at each visit to cross-sectional normative values at each age derived from the norms tables of the WISC-III, as described in statistical methods above. For DSF, the percent of the CI sample that scored more than 1 SD below the normative mean at each age range (annually until age 13, then aggregated from age 13–16 because of small N at those ages) varied from 33.3% at age 6 years to 80.0% at age 12 years (50.5% across all ages). For DSB, the percent of the sample more than 1 SD below the normative mean at each age range varied from 25.0% at age 7 years to 64.5% at age 9 years (44.0% across all ages). By comparison, in any normally distributed sample, 16% would be expected to be more than one SD below the normative mean. Thus, although DSF and DSB measures for the CI sample appeared to have approximately the same slope (growth over time) as the normal-hearing WISC-III normative sample (Figure 1), larger than expected percentages of the CI sample fell below average (more than 1 SD below the normative mean) relative to the normative sample for DSF and DSB at each age (Figure 1).

Figure 2 shows individual slopes based on DSF and DSB by hearing age, defined as duration of CI use in the study sample, compared to norms by hearing age (defined as chronological age) for the normal-hearing WISC-III normative sample. Only DSF and DSB scores corresponding to 6 years or more of CI use were included in this figure in order to map appropriately onto the WISC-III normative sample ages of 6 years and older. Eighteen of the total study sample N of 66 provided 3 or more Digit Span scores for 6 years of CI use or more and are included in the figure. As expected, because hearing age (i.e., duration of CI use in the study sample) is by definition less than chronological age for individuals with CIs, adjusting the curves for duration of CI use resulted in trajectories closer to the normative data than trajectories based on chronological age (Figure 1). However, scores for DSF continued to consistently lag one-half to one standard deviations below norms. Scores for DSB, after initially falling at about norm values (possibly as a result of floor effects) at ages 6 and 7 years, lagged by about one-quarter to one-third SD below norms for the later ages. As with chronological age, regression lines based on duration of CI use appeared to have approximately the same slope (growth over time) as the WISC-III normative sample. Thus, analyses based on duration of CI use as compared to chronological age produced trajectories closer to (but still consistently lagging below) the norm values.

Predicting Speech & Language Outcomes—The mean age at the visit during which the endpoint S/L measures were obtained ranged from 11.08 years (SD=2.07, range=8.06 to 15.56) for the HINT to 12.01 years (SD=2.25, range=8.06 to 16.06) for the PPVT. Overall, the percentages of CI users who scored more than one SD below the normative mean on PPVT and CELF-3 were 58.3% and 63.2%, respectively. Correlations between the DSF and DSB baseline and slope scores and the S/L variables are displayed in Table 2. DSF and DSB baseline scores predicted all S/L endpoint outcome scores with the exception of a nonsignificant trend for the correlation between DSB and PBK Word scores. DSF and DSB slope scores were not significantly correlated with any of the S/L endpoint outcome scores.

After controlling for the conventional predictor variables (chronological age at time of testing, age at time of implantation, communication mode (AOC vs. TC), and maternal education), DSF baseline scores significantly predicted final PPVT Standard Score, and DSF slope scores showed a nonsignificant trend ($p<0.10$) toward predicting PPVT Standard Score. DSF baseline and slope scores taken together accounted for an additional 28% of variance above and beyond the conventional predictor variables in predicting PPVT scores (Table 3). For CELF, both DSF baseline and slope scores significantly predicted endpoint

scores, accounting for an additional 31% of variance above and beyond the conventional predictors. The speech perception (PBK and HINT-C) scores were significantly predicted only by the DSF baseline score above and beyond the conventional predictors; DSF slope was not significantly related to the speech perception scores in the regression equations. The additional percent of variance accounted for by DSF baseline and slope scores was more modest for PBK (14%) and HINT (13%) than for the language (PPVT and CELF) scores.

For DSB, only the two language measures (CELF and PPVT) were significantly predicted by DSB baseline score but not slope, with 9–19% of variance accounted for after controlling for the conventional predictor variables (Table 4). DSB baseline and slope scores taken together were not related to the speech perception variables and accounted for an additional 6% to 14% of the variance in PBK and HINT scores after accounting for the conventional predictor variables.

DISCUSSION

This longitudinal study examined the development of verbal STM/WM capacity over time in a cohort of 66 deaf children following cochlear implantation. Slopes of DSF and DSB performance representing growth and development of verbal STM/WM capacity in the CI sample were similar in magnitude to values obtained from a normal-hearing sample using WISC-III cross-sectional norms. However, consistent with our hypotheses and broadly consistent with prior research, the mean DSF and DSB scores for each age of the CI sample fell consistently below the mean DSF and DSB scores for the normative sample. Moreover, at each age a higher than expected percentage of children in the CI sample scored more than 1 SD below the normative mean scores for the DSF and DSB. These findings reveal that while a normative rate of growth in verbal STM/WM is observed in deaf children with CIs following implantation, deaf children failed to catch up to their normal-hearing peers for either DSF or DSB in the age ranges investigated in this study. Furthermore, correcting for age at implant and duration of CI use (by calculating hearing age) did not fully account for these delays. Because STM/WM as indexed by DSF/DSB is known to be a fairly robust measure of information processing capacity with relatively little improvement in DSF/DSB scores in normal-hearing samples after the late teen years (Wechsler 2008), the gap demonstrated in this sample of CI users is of particular interest both clinically and theoretically in terms of understanding the foundational sources of individual differences in S/L outcomes in this clinical population.

The importance of the finding of parallel but delayed development of verbal STM/WM capacity in pediatric CI users is underscored by the fact that a combination of initial performance (“baseline”) and developmental growth (“slope”) in verbal STM/WM, as assessed by DSF and DSB, predicted additional clinically and statistically significant variance in future S/L performance even after accounting for conventional CI outcome predictors. This finding was particularly robust for DSF scores and for the language (PPVT and CELF) measures. These findings represent a significant advancement in our understanding of the underlying sources of variability and individual differences universally reported in S/L performance following implantation. Our results demonstrate that the basic elementary neurocognitive processes of verbal STM/WM account for an additional meaningful portion of heretofore unexplained suboptimal S/L outcomes observed in deaf children following cochlear implantation (NIH Consensus Panel, 1995; Peterson et al. 2010).

Baseline DSF and DSB scores were better predictors of S/L outcomes than were DS slopes. This is contrary to our hypotheses that DS slopes would consistently predict S/L outcomes, and it suggests that the absolute level or capacity of verbal STM/WM has a much greater

impact on S/L outcomes later in time than the pace of growth in STM/WM (although pace of growth of DSF was predictive of language outcomes independent of baseline score in the regression equations). This pattern may occur for several reasons. First, the DS baseline score reflects the observed level of STM/WM skills, whereas the slope over time reflects growth independent of the actual level of skills. For example, a high baseline score indicates superior STM/WM capacity that will be maintained with even a developmentally average slope (growth curve), whereas a high slope (above average developmental growth) may occur for individuals with poor initial STM/WM skills that do not fully recover. Second, DS slope may be a better predictor of rate of development of S/L skills than of endpoint S/L skills. If this is the case, then DSF/DSB slopes should be related more strongly to the slopes of development of S/L skills over time than with endpoint S/L scores. For this reason, DS slope may have better prognostic utility for children with initial low functioning STM/WM than those with better STM/WM skills. We are currently studying this possibility. Finally, it is important to note here that none of the subjects in this sample received any specific interventions designed to improve STM/WM capacity because such interventions are not routinely available (other than in one small pilot study conducted at our site, which did not include any of the children studied here) for children with CIs. The relationship between DS slope and S/L outcome observed in this study might be changed if children received highly focused interventions to markedly alter the development of STM/WM capacity (Kronenberger et al., 2011).

Although DS baseline scores were better predictors of endpoint S/L scores, DSF slopes were also important additional independent predictors of endpoint language measures (CELF and, in a nonsignificant trend, PPVT) in regression equations (Table 3). Interestingly, however, DSF slopes were not significantly correlated with endpoint language measures in the zero-order Pearson correlation analyses (Table 2). The discrepancy between the correlation and regression findings for DSF slope indicates that DSF slopes were predictive only of the residual of the language scores after the shared variance of DSF baseline with those language scores was removed. In other words, DSF slope predicted a small and nonsignificant portion of the total variance in language (PPVT and CELF) scores but a (relatively) large portion of the variance in language scores that was not associated with DSF baseline. This indicates that DSF growth contributed a small but significant and independent (of DSF baseline effects) portion of the variance to the language outcomes.

In the regression equations, DSF slope scores significantly predicted endpoint performance on a global language measure involving use of linguistic knowledge and comprehension (CELF), and a nonsignificant trend was found between DSF slope scores and receptive vocabulary (PPVT). This pattern suggests that growth in verbal STM, while not predictive of endpoint speech perception skills (assessed by the PBK and HINT), predicts endpoint higher-order language processing skills after baseline verbal STM is accounted for. This finding may occur because the development of language skills continues to be significant throughout childhood and adolescence (Semel et al., 1995), whereas speech perception skills show more rapid development early in childhood and then typically level off after that time. Hence, verbal STM growth during childhood and adolescence may be more valuable for language outcomes because it is occurring concurrently with dramatically increasing demands on language learning skills.

Recently, Niparko and his colleagues reported the results of a 3-year prospective, longitudinal study of a large cohort of deaf children following cochlear implantation and a control group of normal-hearing, typically-developing children using the Reynell Developmental Language Scales (RDLS), a measure of spoken language development that yields both receptive and expressive language scores (Niparko et al. 2010). Niparko et al. found that earlier age at implantation, shorter periods of hearing loss, greater residual

hearing prior to surgery, higher ratings of parent-child interaction, and higher socioeconomic status were all factors associated with better performance on measures of spoken language development assessed by the RDLs. Importantly, Niparko and his colleagues also presented results showing that, in contrast to a group of normal-hearing, typically-developing peers, the CI users' receptive and expressive scores spanned the entire range of performance. Our findings suggest that an additional component of the variance underlying receptive and expressive language may be explained by the contribution of verbal STM/WM capacity studied here, which assesses the basic underlying elementary information processing skills of these children.

Many deaf children with CIs have a disturbance, a delay, or a deficit in rapid phonological coding of speech into lexical and sublexical representations in verbal STM/WM (Pisoni & Geers, 2000; Pisoni & Cleary, 2003; Fagan et al. 2007). The "phonological deficit hypothesis" suggests that deaf children have degraded, incomplete, or otherwise "underspecified" phonological representations for spoken words in STM/WM. All of the conventional behavioral tests currently used to assess S/L outcomes following cochlear implantation rely very heavily on fundamental elementary neurocognitive processes of sensory registration, encoding, storage, retrieval, response organization and execution – all of which are mediated by verbal STM/WM. Variation in verbal STM/WM capacity and processing speed may therefore ultimately be one of the core contributing factors that underlies the large individual differences observed in a wide range of conventional endpoint S/L outcome measures that constitute the standard of care for post-CI follow-up (Pisoni & Geers, 2000; Pisoni et al. 2003; Fagan et al. 2007).

The approach taken in the current study is novel because it relies on a fundamentally different type of performance measure than is typically used to assess S/L outcomes following cochlear implantation. Historically, research on CI outcomes has used conventional clinical *endpoint product measures* of speech and language performance, which reflect a composite of more basic underlying elementary processes that contribute to S/L functioning without offering direct insight into these processes themselves. Although the conventional batteries of S/L outcome measures have good face validity, they were not developed to measure real-world, ecologically valid effectiveness of CIs or to uncover the underlying sources of variability responsible for the large individual differences in speech and language outcomes in this clinical population. Digit span, a neurocognitive *process measure* of the information capacity of verbal STM and verbal WM, provides an assessment measure of an underlying elementary process that forms one of the foundations of S/L performance and which is known to be at-risk in children with CIs (Pisoni & Geers, 2000). The use of a robust set of S/L outcome measures and a predictive model provides further novel converging evidence that verbal STM/WM capacity is predictive of a broad set of future S/L skills in deaf children with CIs. With this set of data, we are able to move beyond just an associative model establishing correlation between verbal STM/WM and begin to document the predictive value of process measures of verbal STM/WM performance and S/L outcomes several years following cochlear implantation.

The results of this study have several limitations, which should be considered in interpreting study results. First, in order to compare the performance of our sample of CI users to a normative standard, we used values from the WISC-III normative sample. This sample was not matched to our sample on variables such as geographical region or SES. An alternative approach would be to use a normal-hearing, age-matched control group, as Niparko and colleagues have done (Niparko et al. 2010). Second, our sample of pediatric CI users was heterogeneous and consisted of children with different lengths of CI use, different ages of testing, different durations between baseline DS and S/L testing, and different numbers of DS data points separated by different amounts of time. For these very reasons, however, our

sample is also highly reflective of the typical clinical population of deaf children at large, and we believe our findings are directly applicable to issues associated with clinical management. Furthermore, we statistically controlled for some of the main components of this heterogeneity in the regression analyses. Third, we did not measure or control for IQ or nonverbal ability in analyses investigating the relationship between DS scores and S/L outcomes. It is therefore possible that the relationship between DS and S/L outcomes could be mediated by nonverbal intellectual ability, although one prior study of CI users found that nonverbal IQ did not fully attenuate the relationship between DS score and language scores measured at high school ages (Geers & Sedey, 2011). Additionally, Pisoni, Kronenberger, Roman, et al. (2011) reported significant cross-sectional correlations between DS scores and S/L outcomes but no significant correlations were found between nonverbal IQ and S/L outcomes in children and adolescents with CIs. Finally, we did not investigate interaction effects between the conventional predictors of S/L performance (i.e., Block 1 of the regressions in Tables 3 and 4) and DS scores in predicting outcomes. It is possible that DS scores are more or less predictive of S/L performance in subgroups of children who differ in one or more of those conventional predictors. Future research with larger sample sizes will be necessary to fully investigate this possibility.

It should also be noted that the DSB task used here can function both as a measure of verbal STM or verbal WM, depending upon strategic factors that, while varying with chronological age at the time of evaluation, are also likely to differ on an individual basis (St. Claire-Thompson, 2010). Indeed, a number of alternative process measures of working memory capacity, namely, complex sentence span, exist that are also widely used as measures of verbal STM/WM. Digit span was used here, however, because at this early stage of the research, it was important to use a brief, easy to administer measure that can be employed efficiently in CI evaluations and is, therefore, more clinically applicable. However, it is important to integrate the findings of this study, which are based on Digit Span as the sole measure of verbal STM/WM, with future studies that include additional measures of verbal STM/WM (Alloway, 2007). Future research should also investigate a more diverse set of verbal and visuospatial STM/WM measures to obtain a more robust profile of each child's information processing skills.

In summary, variability in STM/WM, one of the basic underlying core neurocognitive factors that are common information processing components of all behaviorally-based conventional measures of S/L performance, predicted an additional significant portion of the heretofore unexplained variability and individual differences in S/L outcomes following cochlear implantation. The longitudinal findings on the development of DS presented here suggest that process measures of verbal STM and verbal WM capacity predict long-term S/L outcomes and that the rate of growth of verbal STM/WM is delayed relative to normative data obtained from typically-developing normal-hearing age-matched peers. Future research should aim to better understand the patterns of verbal STM/WM development in children with CIs, including the identification of possible subgroups of children with different STM/WM developmental profiles. Novel interventions targeting the core underlying elementary information processing mechanisms may offer the potential for helping more pediatric CI users develop better and more robust S/L skills and, therefore, narrow the developmental gap between poor CI performers and exceptionally good CI users who are performing within the range observed for their normal-hearing age-matched peers.

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Figure 1.

Digit Span Scores by Chronological Age. The top panel shows the individual slopes of digit span forward (DSF) raw subscale scores of deaf children with cochlear implants ($n=66$) by chronological age. The bottom panel shows the digit span backward (DSB) raw subscale slopes of deaf children with cochlear implants by chronological age. Each line represents fitted digit span raw subscale scores of a child over time based on linear regression. The heavy solid black line represents the average slope of the CI sample obtained from a mixed effects repeated measures model. Means and standard deviations of the normative data are shown at each age by black dots and error bars. Norms in both panels were obtained from the WISC-III (Kaplan et al., 1999).

Figure 2.

Digit Span Scores by Hearing Age. Hearing Age is defined as chronological age for the normal-hearing WISC-III normative sample or years of CI use for the study sample of children with CIs. Only data for hearing age of 6 years or older is displayed in order to correspond to WISC-III norms. The top panel shows the individual slopes of digit span forward (DSF) raw subscale scores of deaf children with cochlear implants by years of CI use. The bottom panel shows the digit span backward (DSB) raw subscale slopes of deaf children with cochlear implants by years of CI use. Each line represents fitted digit span raw subscale scores of a child over time based on linear regression. The heavy solid black line represents the average slope of the CI sample obtained from a mixed effects repeated measures model. Means and standard deviations of the normative data are shown at each age by black dots and error bars. Norms in both panels were obtained from the WISC-III (Kaplan et al., 1999).

Table 1

Characteristics of Study Participants, N = 66

Variable	Mean (SD; Range)
Age at CI implant (yrs)	3.81 (1.73; 1.40–8.01)
Age at Onset of Deafness (yrs)	0.30 (0.71; 0.00–3.00)
Duration of Deafness (yrs)	3.51 (1.84; 0.49–8.01)
Chronological Age at 1st Visit (yrs)	7.55 (1.44; 6.01–11.53)
Duration of implant use at 1 st Visit (yrs)	3.74 (1.41; 1.02–8.00)
Time Followed (yrs)	3.88 (1.69; 2.00–8.91)
Number of Test Sessions	4.53 (1.27; 3.00–9.00)
Best pre-operative PTA [†]	108.36 (9.90; 83.33–120.07)
Mother/Female caregiver education [*]	4.65 (1.77; 2.00–9.00)
	<u>N (%)</u>
Male	34 (51.52%)
Race/Ethnicity ^{**}	
White	63 (95.45%)
African American	4 (6.06%)
Asian	0 (0%)
Hispanic	1 (1.52%)
Etiology of Hearing Loss	
Unknown	43 (65.15%)
Meningitis	7 (10.61%)
CMV [‡]	2 (3.03%)
Genetic	9 (13.64%)
Anatomic Anomalies [£]	5 (7.58%)
Communication Mode at 1 st visit	
Auditory-Oral Communication (A-OC)	43 (65.15%)
Total Communication (TC)	23 (34.85%)
Device	
Unilateral CI	64 (96.97%)
Bilateral CIs	0
CI + HA [§]	2 (3.03%)

[†]PTA = Pure Tone Average (500, 1000, 5000, 2000 Hz)^{*} Ordinal scale of 1–9; includes education of father in one case^{**} More than one race/ethnicity was recorded for some children[‡] Cytomegalovirus[£] e.g., Enlarged Vestibular Aqueduct, Mondini Malformation[§] Concurrently used a CI in one ear and a hearing aid in the contralateral ear

CI = cochlear implant; HA = hearing aid

Table 2

Correlations between digit span and speech-language outcome measures.

	PBK Word	HINT-C	PPVT	CELF
<u>Digit Span Forward</u>				
Digit Span Forward – Baseline	0.51 ***	0.42 *	0.58 ***	0.72 ***
Digit Span Forward – Slope	–0.14	–0.05	–0.15	–0.06
<u>Digit Span Backward</u>				
Digit Span Backward – Baseline	0.26 ^a	0.40 *	0.40 **	0.51 ***
Digit Span Backward – Slope	–0.21	–0.16	–0.09	0.14

Note: Values are zero-order Pearson correlation coefficients. PBK=Phonetically Balanced Kindergarten test; HINT-C=Hearing in Noise Test for Children in Quiet; PPVT=Peabody Picture Vocabulary Test; CELF=Clinical Evaluation of Language Fundamentals Fourth Edition Core Language Score.

^a
p<0.10

*
p<0.05

**
p<0.01

p<0.001

Table 3

Digit span forward growth and speech-language outcomes in deaf children with cochlear implants.

	PBK Word	HINT-C	PPVT	CELF
<u>Block 1: Conventional Predictors</u>				
Age at Last Visit	-1.07	-1.18	0.68	-0.63
Age at Implantation	3.15	0.91	-1.97	-4.57 [*]
Mother's Education	1.93	2.12	3.66 [*]	3.14
Communication Mode	29.22 ^{***}	29.27 [*]	-2.34	17.6 ^a
R ² for Block 1	0.33 ^{**}	0.21 ^a	0.13	0.32 [*]
<u>Block 2: Digit Span Forward</u>				
Digit Span Forward – Baseline	4.29 ^{**}	3.42 [*]	4.30 ^{***}	5.62 ^{***}
Digit Span Forward – Slope	-1.14	3.85	5.29 ^a	8.90 [*]
R ² for Block 2	0.47 ^{***}	0.34 [*]	0.41 ^{***}	0.63 ^{***}

Note: Values are unstandardized regression coefficients. PBK=Phonetically Balanced Kindergarten test; HINT-C=Hearing in Noise Test for Children in Quiet; PPVT=Peabody Picture Vocabulary Test; CELF=Clinical Evaluation of Language Fundamentals Fourth Edition Core Language Score.

^a
p<0.10

^{*}
p<0.05

^{**}
p<0.01

^{***}
p<0.001

Table 4

Digit span backward growth and speech-language outcomes in deaf children with cochlear implants.

	PBK Word	HINT-C	PPVT	CELF
<u>Block 1: Conventional Predictors</u>				
Age at Last Visit	-1.07	-1.18	0.68	-0.63
Age at Implantation	-3.15	0.91	-1.97	-4.57 [*]
Mother's Education	1.93	2.12	3.66 [*]	3.14
Communication Mode	29.22 ^{***}	29.27	-2.34	17.60 ^a
R ² for Block 1	0.33 ^{**}	0.21 ^a	0.13	0.32 [*]
<u>Block 2: Digit Span Backward</u>				
Digit Span Backward – Baseline	1.66	3.33 ^a	2.37 [*]	4.91 ^{**}
Digit Span Backward – Slope	-4.69	-2.20	-0.31	6.38
R ² for Block 2	0.39 ^{**}	0.35 [*]	0.22 [*]	0.51 ^{***}

Note: Values are unstandardized regression coefficients. PBK=Phonetically Balanced Kindergarten test; HINT-C=Hearing in Noise Test for Children in Quiet; PPVT=Peabody Picture Vocabulary Test; CELF=Clinical Evaluation of Language Fundamentals Fourth Edition Core Language Score.

^a
p<0.10

^{*}
p<0.05

^{**}
p<0.01

^{***}
p<0.001