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Profiles of Verbal Working Memory Growth Predict Speech and Language Development in Children with Cochlear Implants

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Abstract

Purpose—Verbal short-term (STM) and working (WM) memory skills predict speech and language outcomes in children with cochlear implants (CIs) even after conventional demographic, device, and medical factors are taken into account. However, prior research has focused on single endpoint outcomes as opposed to the longitudinal process of development of verbal STM/WM and speech-language skills. This study investigated relations between profiles of verbal STM/WM development and speech-language development over time.

Method—Profiles of verbal STM/WM development were identified using group-based trajectory analysis of repeated digit span measures over at least a two-year time period in a sample of 66 children (age 6-16 years) with CIs. Subjects also completed repeated assessments of speech and language skills during the same time period.

Results—Clusters representing different patterns of development of verbal STM (digit span forward scores) were related to the growth rate of vocabulary and language comprehension skills over time. Clusters representing different patterns of development of verbal WM (digit span backward scores) were related to the growth rate of vocabulary and spoken word recognition skills over time.

Conclusions—Different patterns of development of verbal STM/WM capacity predict the dynamic process of development of speech and language skills in this clinical population.

Cochlear implants (CIs) electronically process sound and transmit information to the auditory nerve, allowing dramatically improved perception of speech and environmental sounds for many individuals with severe to profound hearing loss. Approved by the United States Food and Drug Administration (FDA) in the 1980's, CIs became more widely used in

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pediatric populations in the 1990's, with 26,000 American children having a CI by 2010 (NIH, 2010). One major goal for children who have received CIs is development of speech perception, spoken word recognition, and language skills. As a result, much of the early research on CI benefits focused on endpoint product measures of speech and language outcomes, using cross-sectional or short-term longitudinal designs to provide a "snapshot" view of children's speech and language development following cochlear implantation. This research demonstrated improvement across a wide variety of spoken language tasks but also considerable variability in all speech and language outcome measures within the population of children with CIs (Geers, Brenner, & Davidson, 2003; Geers, Nicholas, & Sedey, 2003; Kirk, Hay-McCutcheon, Sehgal, & Miyamoto, 2000).

As the first cohorts of CI users implanted in early childhood reached late childhood and adolescent ages, it became possible to conduct increasingly more detailed and sophisticated longitudinal and long-term outcome studies of speech and language outcomes following cochlear implantation. These studies demonstrated, on average, clear and consistent patterns of improvement in speech and language skills for CI users over a period of years during early childhood (Niparko et al., 2010) and reaching into adolescence (Davidson, Geers, Blamey, Tobey, & Brenner, 2011; Geers & Sedey, 2011). Again, however, significant variability in developmental patterns (Niparko et al., 2010) and endpoint outcomes (Geers & Sedey, 2011) in speech-language functioning were consistently found.

Efforts to explain differences in developmental patterns and endpoints in speech-language functioning have identified several demographic, device, and medical factors that predict speech-language development following cochlear implantation. These factors include earlier age at implantation, greater experience with the CI, higher socioeconomic status, higher nonverbal IQ, and the use of auditory-oral communication strategies (as opposed to sign enhancement; Geers & Sedey, 2011; Niparko et al., 2010). Identification of these predictor variables has led to changes in clinical recommendations (e.g., earlier implantation, increased exposure to auditory-oral communication strategies) and has improved the identification of children who are at risk for suboptimal speech and language outcomes. However, even taking into account these demographic, device, and medical factors, a significant amount of the variance in speech-language outcomes for children with CIs remains unexplained (Pisoni, Conway, Kronenberger, Henning, and Anaya, 2010).

Because the enormous variability in speech-language outcomes observed in the child CI population is not sufficiently explained by conventional demographic, device, and medical factors, a number of researchers have continued to search for other influences on speech-language outcomes in children with CIs. One promising set of potential influences is domain-general neurocognitive functions that may be affected by a period of auditory deprivation and that, in turn, support the development of speech and language functioning. Auditory experience and speech-language functioning are part of a much broader interrelated set of neurocognitive abilities that develop dynamically through the lifespan, with especially rapid development during childhood (Luria, 1973). Hence, the impact of a period of deafness and sensory deprivation followed by exposure to highly degraded auditory signals has been shown to extend well beyond just speech perception and spoken language skills (Pisoni & Cleary, 2003).

One broad domain of neurocognitive functioning that may be affected in some children with deafness and degraded auditory experience following cochlear implantation is executive functioning, which includes processes such as controlled attention, active management of short-term memory, self-regulation, planning and sequencing (Pisoni et al., 2010). Figueras, Edwards, & Langdon (2008) found that children with CIs or hearing aids scored lower than normal-hearing controls in several areas of executive functioning, such as inhibition, planning, set-shifting, working memory, and some types of attention. In a more recent study, Beer, Pisoni, Kronenberger, & Geers (2010) found elevated scores (indicating more problems) on the BRIEF (Gioia, Isquith, Guy, & Kenworthy, 2000), a parent-report behavior checklist of executive functioning, in a sample of adolescents with CIs. A substantial amount of research also demonstrates that children with CIs score below age norms on measures of auditory-verbal short-term and working memory, which are considered to be core, foundational components of executive functioning (Kronenberger, Pisoni, Henning, Colson, & Hazzard, 2011; Pisoni et al., 2008; Pisoni, Kronenberger, Roman, & Geers, 2011).

In particular, verbal working memory (WM) has been shown to be affected by a period of deafness and degraded auditory experience following cochlear implantation (Pisoni et al., 2010). WM is the ability to encode, store, and manipulate information during a short period of time while simultaneously engaging in other cognitive activities (Baddeley, 2007). An extensive body of earlier research supports the existence of two domain-specific systems for storage of information in WM: a phonological loop specialized for verbal-phonological information and a visuospatial sketchpad for visual and spatial material. A third component, the central executive, functions to actively regulate the information in working memory by controlling attention and allocating cognitive resources and strategies (Gathercole & Baddeley, 1993), whereas a fourth component, the episodic buffer, acts to link information from the other three components and long-term memory (Baddeley, 2007). WM tasks can differ in the degree to which they engage each of these four components. Language-based WM tasks are processed largely through the domain-specific phonological loop, with involvement of the central executive to the extent that active regulation and controlled processing are required; similarly, visual and/or spatial WM tasks are processed through the visuospatial sketchpad and central executive (Baddeley, 2007; Gathercole & Baddeley, 1993). The role of the episodic buffer is dictated by the use of long-term memory information and skills in working memory processing.

The involvement of the central executive in working memory tasks varies based on the processing demands inherent in the task. Engle et al. (1999) demonstrated that tasks involving maintenance of memory information over short periods of time fall into two related categories: WM tasks, which require a high degree of active maintenance of information in memory in the face of competing information and mental activity, and short-term memory (STM) tasks, which involve simple rote memory span with less involvement of active controlled attention. Empirical results suggest that STM can be conceptualized as a subset of WM (Engle et al., 1999), with the degree of involvement of the central executive differentiating between WM (high degree of central executive involvement) and STM (lower degree of central executive involvement)(Cowan, 1995; Engle et al., 1999). Because it involves less intense activity of the central executive, STM relates more to the processing of the domain-specific subsystems (phonological loop and visuospatial sketchpad), although some active processing is also required for maintenance and retrieval of information in STM (Cowan, 1995; Engle et al., 1999).

Verbal STM/WM is used to maintain the phonological and lexical representations of words and sentences in immediate memory for verbal processing. As a result, 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). Development of the phonological loop and central executive supports the growth of skills in vocabulary, comprehension, reading, and speech production (Gathercole & Baddeley, 1993). Conversely, delayed or disturbed development of verbal STM/WM is related to disorders of language and learning (Alloway & Archibald, 2008; Gathercole, Alloway, Willis, & Adams, 2006). Children with CIs are at high risk for impaired verbal

STM/WM because of underspecified degraded phonological representations of verbal input, slower verbal rehearsal speed, increased demands on controlled attention during perception and recognition of spoken words, and reduced experience with real-time rapid phonological coding in speech perception and processing (Pisoni & Cleary, 2003; Pisoni et al., 2008, 2011).

Digit span tests (Wechsler, 1991) are widely used, well-validated measures of verbal STM/WM capacity. They consist of digits forward (DF) and digits backward (DB) components, which require subjects to repeat a series of spoken digits in either forward or reverse order, respectively. Items on digit span tests typically begin with a short sequence of digits (usually 2-3 digits) that increases incrementally in length after the subject successfully repeats a criterion number of sequences at each digit length. DF tasks require repetition of the digit sequences without any manipulation of the items in a sequence; hence, they emphasize verbal STM and the phonological loop much more than the central executive component of WM. DB tasks, on the other hand, require both verbal STM and the concurrent processing operations of actively reordering the sequence of digits before output. As a result, DB tasks require active controlled attention and allocation of conscious cognitive processing resources to divided tasks, requiring greater involvement of the central executive component of WM.

Using digit span tests, Pisoni, Geers, and colleagues investigated the development of verbal STM/WM in a sample of prelingually deaf children who received CIs by age 5 years (Pisoni & Cleary, 2003; Pisoni et al., 2011). Children with CIs were tested at ages 8-9 years (Pisoni & Cleary, 2003) and then again approximately 8 years later (Pisoni et al., 2011). At both age points, children with CIs scored lower on the digit span measures of verbal STM/WM skills than normal-hearing children. Relative to norms from the WISC-III, total digit span scores were more than 1 SD below the mean at both time points (Pisoni et al., 2011). DF tasks were more negatively affected than DB tasks, possibly because of the almost exclusive emphasis of DF tasks on known areas of severe risk for children with hearing impairment and CIs, including rapid phonological coding used for verbal rehearsal and rote-sequential processing. Unlike DF tasks, DB tasks also include a more significant component of controlled attention (for simultaneous management of memory and backward reordering of digits) which may be less severely affected albeit still below average relative to norms (Conway, Pisoni, & Kronenberger, 2009; Pisoni et al., 2008).

Studies with children who have milder forms of hearing impairment (e.g., mild to severe hearing impairment) and use hearing aids have shown smaller differences from normal hearing children in verbal STM/WM compared to studies of children with CIs. Stiles, McGregor, & Bentler (2012) did not find statistically significant differences on digits forward or digits backward tests between samples of hearing impaired children and normal hearing children, although there was a nonsignificant trend ($p=0.09$) for the hearing impaired group to score lower on digits forward, and the effect size comparing the groups on digits forward presented in quiet (the standard condition on the WISC-III) fell in the medium range ($d=0.74$). Hansson, Forsberg, Lofqvist, Maki-Torkko, & Sahlen (2004) also studied working memory skills in children with hearing impairment and found no differences compared to a group of normal hearing children, although their measure of working memory (a complex working memory task requiring subjects to remember the final word of each sentence in a series of sentences, after stating whether each sentence was factually accurate or inaccurate) differed markedly from the conventional digit span task. Similar to Stiles et al. (2012), they found no difference between the hearing impaired sample and a sample of normal hearing children that they obtained from a different study.

Although the reasons for discrepant findings between verbal STM/WM studies of children with CIs and similar studies of children with hearing aids are unclear, it is possible that greater severity of hearing loss and more degraded auditory input may put children with CIs at higher risk for delayed development of verbal STM/WM. Alternatively, the more recent studies of children with hearing aids used relatively small samples of children with hearing impairment (e.g., N=18 in the Stiles et al., 2012 study; N=18 in the Hansson et al., 2004 study), so the power to detect a smaller effect size was limited. Similar to children with CIs (Pisoni et al., 2011), children who use hearing aids did show slower articulation rates than normal hearing children (Stiles et al., 2012), and articulation rate is an important component of verbal STM/WM (Pisoni et al., 2011). Hence, additional investigation is needed to understand and explain the relationship between mild to moderate hearing impairment and verbal STM/WM.

Because verbal STM/WM serves as the core obligatory interface or gateway used to map speech signals onto phonological and lexical representations in long-term memory, any deficits or disturbances in verbal STM/WM may also cascade and contribute to delays in language development (Pisoni & Cleary, 2003). This hypothesis has been supported in several earlier studies of children with CIs, which demonstrate that digit span measures of verbal STM/WM are strongly correlated with a range of spoken language outcomes in samples of children with CIs (Pisoni & Cleary, 2003; Pisoni et al., 2011). Similarly, support for a strong relationship between verbal STM/WM and language development has been found in samples of children with mild to moderate hearing impairment (Hansson et al., 2004; Stiles et al., 2012).

Although earlier studies have documented the relations between verbal STM/WM and endpoint product speech-language outcome measures in children with CIs, there has been little investigation of the development of verbal STM/WM over time following CI or assessment of how developmental patterns of verbal STM/WM are related to the process of development over time of speech and language skills. Pisoni et al. (2011) found improvement in children with CIs relative to normal-hearing peers in DF scores between (approximately) ages 8 and 16 years, but over half of the CI sample continued to lag 1 SD or more behind normal-hearing peers at the later age. DB scores in the CI sample differed less than the DF scores from norms at ages 8-9 years, but the percentage of the total sample of 112 children differing by more than 1 SD from norms increased for DB scores over the subsequent 8 year period. Moreover, early DF and DB scores predicted speech-language skills 8 years later, although DF was found to be a stronger predictor, and DB predicted only higher-order spoken language skills such as vocabulary, reading, and comprehension (Pisoni et al., 2011).

Taken together, the extant research demonstrates that verbal STM/WM, as assessed with digit span, is atypical and significantly delayed in children with CIs relative to normal-hearing peers and that measures of verbal STM/WM capacity predict long-term speech-language outcomes. However, little is known about the dynamic process of development of verbal STM/WM over time following cochlear implantation or how developmental patterns of STM/WM relate to the trajectory of speech-language growth during childhood (as opposed to single endpoint measures). A significant advantage of research investigating the development of verbal STM/WM capacity over time is the ability of longitudinal data to provide new insights concerning plasticity and change in verbal STM/WM skills. By analyzing growth trajectories over time, researchers can better understand the types of changes that can occur in verbal STM/WM, as well as the relations between those types of changes and development of speech-language outcomes. For example, some verbal STM/WM growth trajectories may be associated with children who catch up to normal hearing peers in speech-language outcomes, whereas others may be associated with continued risk of decline and delayed speech and language skills.

In this paper, we sought to address three central questions resulting from the gap in research on growth trajectories of verbal STM/WM in children with CIs: (1) What patterns of developmental growth over time in verbal STM/WM skills (as measured by digit span) are shown by children with cochlear implants? (2) What proportion of children demonstrate each pattern of growth (e.g., which growth patterns are most and least common)? (3) How is the development of verbal STM/WM related to the growth in speech and language skills during that same period of time?

These three questions address fundamentally different issues than have been considered in past research. First, none of the earlier studies have attempted to identify profiles of growth in verbal STM/WM over *multiple time points* during child development following cochlear implantation. In order to address this question, at least three measurements of digit span over a period of two years or greater were obtained from all subjects in the current study. Earlier studies have obtained only one or two time-point measurements (Pisoni et al., 2011), which are less than ideal for identifying clusters of developmental trajectories. Second, this study sought to identify *empirically-derived clusters* of digit span growth within a CI sample, in order to better characterize the enormous variability in verbal STM/WM growth over time that has been found in the CI population (Pisoni et al., 2011). Similar empirical clustering techniques have contributed to the development of highly valuable and widely used classification systems in the behavioral sciences, such as categorization of children's behavior problems (e.g., Thompson, Kronenberger, & Curry, 1989). Finally, this study specifically investigated relations between developmental patterns (clusters) of verbal STM/WM change and growth in speech-language skills, as opposed to predicting only single end-point speech-language outcomes. In other words, relations between the growth patterns of both verbal STM/WM and speech-language skills were investigated, as opposed to examination of the endpoints alone.

Method

Participants

Participants in the study were 66 children who were 6.0 to 11.5 years of age (mean=7.6 years, SD=1.4) at the time of initiation of data collection for this study and 8.1 to 16.1 years of age (mean=11.4 years, SD=2.2) at the completion of data collection for this study. Participants were required to have prelingual severe-to-profound bilateral hearing loss, cochlear implantation at age 8 years or younger, use of a currently available, state-of-the-art CI system, and a monolingual English home environment. 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), and two or more speech-language assessments during that same time period. Table 1 provides a description of other characteristics of the sample.

Digit span measurements for a specific time point were included only if a subject achieved a raw score above zero on either DF or DB, 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). Three different subjects each provided one digit span measurement that was dropped from further analyses because both DF and DB raw scores were 0. 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).

Procedure

Subjects were ongoing participants in a long-running, longitudinal study of development of spoken language and related cognitive skills in children with cochlear implants. Families of subjects were approached about participation in the study as close to the time of the implant surgery as possible, although some subjects entered the study later because they were implanted at other sites and/or chose to start participation in the study at a later date. All subjects were administered a battery of tests to assess speech perception, spoken word recognition, vocabulary knowledge, language comprehension/understanding, and other cognitive skills (e.g., digit span). Visits were scheduled at 6 month intervals from the time of implantation until age 6 years, and at 1-year intervals thereafter. Although every attempt was made to complete all return visits and to obtain all data at each visit, some missing data occurred because of missed appointments, inability to complete some tests in the battery because they were too advanced/difficult for the child, and/or difficulty completing the entire battery during an appointment. As a result, subjects completed different numbers of tests at different ages within the study age range of 6-16 years. A total of 299 visits were obtained for the 66 study subjects.

All speech-language and neurocognitive tests were administered by licensed speech-language pathologists who had extensive experience testing children with CIs. The study was approved by the university institutional review board, and parental consent (with child assent when appropriate) was obtained prior to initiation of any study procedures. Subjects were paid $20/hour to offset expenses related to time and travel.

Measures

Verbal short-term/working memory capacity—The Digit Span subtest of the Wechsler Intelligence Scale for Children, Third Edition (WISC-III; Wechsler, 1991), which consists of Digits Forward (DF) and Digits Backward (DB) tasks, was administered using standard live-voice format. The DF task requires subjects to repeat a sequence of random spoken digits between 1 and 9 (inclusive) in forward order, beginning with a 2-digit sequence. Two items are presented at each sequence length, and if subjects answer at least one item correctly, the sequence length is increased by one until subjects answer both items incorrectly at the same sequence length. The DB task is identical to the DF task except that subjects must repeat the sequences in reverse order. DF and DB raw scores (the total number of items correctly reproduced) were used as measures of verbal STM and verbal WM capacity, respectively, for this study. Norm-based scores (scaled scores with a mean of 10 and standard deviation of 3 based on a nationally-representative normal hearing normative sample; Kaplan, Fein, Kramer, Delis, & Morris, 1999) were also calculated for DF and DB in order to provide a comparison to a representative sample of normal-hearing children. Consistent with prior studies (e.g., Pisoni & Cleary, 2003; Pisoni et al., 2011), DF and DB scaled scores for the first ($t(65)=11.8$ and 9.5, respectively, $p<0.001$) and final ($t(65)=9.8$ and 5.4, respectively, $p<0.001$) visits fell well below scale norm means (Table 1).

Speech and language—Four measures were used in this study to assess a range of speech and language skills: the Phonetically Balanced Kindergarten test (spoken word recognition), Hearing in Noise Test for Children (sentence repetition), Peabody Picture Vocabulary Test (one-word receptive vocabulary), and Clinical Evaluation of Language Fundamentals (complex language and comprehension skills).

***Phonetically Balanced Kindergarten* (PBK; Haskins, 1949):** The PBK test is an open-set measure of spoken word recognition that uses 50 spoken (live-voice) words. Scores are provided for the number of correctly identified phonemes and words, expressed as percentages correct. Raw percentage correct scores for the PBK words were used in the present study to measure open-set speech perception skills. A total of 52 subjects completed the PBK two or more times during the study period (mean number of assessments=4.1, SD=1.3), for a total of 211 PBK assessments (71% of the 299 total study visits).

***Hearing in Noise Test for Children* (HINT-C; Nilsson, Soli, & Sulllivan, 1994):** The HINT-C is used to measure children's ability to perceive and repeat recorded sentences presented in the quiet and in masking noise (+5dB SNR). Scores represent the percentage of words repeated correctly. For the present study, only HINT-C scores in quiet were used to measure sentence perception and repetition skills because this task yielded more valid results at young ages (i.e., some children at the low end of the age range who could complete HINT-C in quiet were unable to provide a valid score for HINT-C in noise), and therefore the HINT-C in quiet could be used with greater validity across the entire age range of the study. The correlation between HINT-C in quiet and HINT-C in noise was 0.81 for the first measurement of HINT-C completed by subjects in the study (N=37 because not all subjects were able to complete the HINT-C in noise), indicating a strong relationship between performance on the HINT-C in quiet and HINT-C in noise. A total of 40 subjects provided valid HINT-C scores two or more times during the study period (mean number of assessments=3.3, SD=1.3), for a total of 131 HINT-C measurements across the 299 visits (44% of study visits).

***Peabody Picture Vocabulary Test – Third Edition* (PPVT-III; Dunn & Dunn, 1997):** The PPVT-III is a measure of single-word receptive vocabulary that requires participants to select a picture that matches a word that is spoken and/or signed (depending on the communication preference of the child) by the examiner. Norm-referenced criteria based on a nationally representative sample of normal-hearing children are used to produce a total standard score. For the present study, PPVT-III raw scores were used in order to model growth in vocabulary-related language skills across age. A total of 65 subjects completed the PPVT-III two or more times during the study period (mean number of assessments=4.3, SD=1.3), for a total of 282 PPVT-III assessments across the 299 study visits (94% of visits).

***Clinical Evaluation of Language Fundamentals* (CELF-3 or CELF-4, depending on the time period of the study during which subjects were assessed; Semel, Wiig, & Secord, 1995, 2003):** The CELF provides a global measure of complex receptive and expressive language skills such as comprehension of linguistic concepts and ability to formulate or recall sentences. A total of 39 subjects completed a version of the CELF during two visits of the study period (mean number of CELF assessments=3.3, SD=1.4), for a total of 130 CELF assessments during the 299 study visits (43% of study visits). Thirty-three subjects completed the CELF-3, and 28 subjects completed the CELF-4 (22 subjects completed the CELF-3 at earlier visits and CELF-4 at later visits, because the CELF was revised during the time period of this study). For the CELF-3, the Total Language Score was used for this study, whereas for the CELF-4, the Core Language Score was used. These two scores (both of which are norm-based Standard Scores, allowing for the combination of corresponding scores across CELF versions) are the total summary scores for the respective CELF versions and share content (including several overlapping subscales) about a child's understanding of words and concepts, recalling sentences, and formulating sentences. For the 22 subjects who completed both CELF-3 and CELF-4 scales during the study, the correlation between the CELF-4 Core Language Score and the immediately preceding CELF-3 Total Language Score was 0.87, which is approximately the test-retest validity of the CELF-3 Total Language and CELF-4 Core Language Scores reported in the test manuals (Semel et al., 1995, 2003); this analysis provides support for the decision to combine CELF-3 Total Language and CELF-4 Core Language Scores.

Statistical Analysis

Statistical analyses were conducted in two stages. In the first stage, DF and DB growth clusters were determined. In the second stage, eight repeated measures models were fit for the four speech-language measures and two sets of cluster groups (DF and DB). In the first stage of analysis, Nagin's group-based trajectory analysis was used to derive empirically-based DF and DB developmental trajectories over time (Nagin 1999; Jones and Nagin, 2007). Different from the traditional cluster analysis such as K-means clustering, the group-based trajectory analysis is a model-based approach that provides probabilistic classification through the posterior probability of cluster membership. The shape of the cluster trajectories and the size of each cluster are jointly estimated using a maximum likelihood approach. The trajectory groups represent subpopulations of subjects that follow similar developmental trajectories. Since the DF and DB scores are nonnegative, censored normal distribution was used when modeling the cluster trajectories. Group-based trajectory analysis was run specifiying 1, 2, 3, 4, and 5 clusters. Then the optimal number of clusters was determined by comparing Bayes Information Criteria (BIC) between models and choosing the model with the largest BIC and significant improvement from the previous model ($2 \times (\mathrm{BIC}_{\text{k-1 clusters}} - \mathrm{BIC}_{\text{k clusters}}) > 2$) (Jones et al., 2001).

In addition, we required that each cluster contain at least 10% of the sample to ensure that the size of all clusters was adequate for further analysis. Linear trajectories were considered for clusters based on evidence that DF and DB scores in normative samples grow in a linear fashion throughout the age range used in this study (Kaplan et al., 1999). After the shape of the cluster trajectories and size of the clusters were determined, the posterior probability that a subject belonged to each of the clusters, given the observed DF or DB scores over time, was computed. The subject was then assigned to his/her most likely cluster. The accuracy of the classification was examined using classification tables as explained in Nagin (1999).

The linear trajectories of each cluster are reported in this paper as a baseline score at age 6 years (the projected value for the cluster at age 6 years, which is the youngest age eligible for the study) and a slope (increase in digit span raw score per age-year) over the duration of the study. The baseline score represents a projected “starting point” for the digit span raw score (as a measure of verbal STM/WM), whereas the slope represents growth in that score over time (age). Sample characteristics for each cluster are also reported, in order to identify factors associated with each of the DF and DB developmental clusters.

In the second stage of our analysis, the association between DF and DB clusters and speech and language growth was analyzed using eight separate repeated measures mixed effects models (separate models for DF and DB clusters for each of the four speech-language measures). Chronological age was used as the time effect. All visits for which speech and language measures were obtained were utilized in the models. One of the advantages of mixed models is that a missed visit or missing score does not eliminate a subject from inclusion (DeLucia & Pitts, 2006). However, sample size varied for each speech and language measure depending on the number of subjects that were able to complete the measure two or more times during the time period of the study (in order to provide a growth estimate).

In these mixed models, the DF or DB clusters, age, and DF/DB cluster × age factors were used as predictors of the repeated measures speech-language scores, while controlling for major demographic and device variables that have been identified in past research to be related to language outcomes: age at implant, best pre-implant pure tone average, communication mode, and maternal education as a proxy for SES. Because chronological age was a main effect in these models, the main effect of DF and DB clusters was evaluated independently of a main effect of age. Time since implantation was not controlled because it is derived directly from two factors already in the model (age at implant and chronological age) and is therefore redundant with those factors. Participant was modeled as a random effect to account for the correlation of within-subject language scores, with a compound symmetry variance-covariance structure.

In these analyses, the DF/DB cluster × age interaction represents the relationship between DF or DB cluster and growth (e.g., change with age) in the criterion speech-language skill. In other words, this interaction measures the association between DF or DB growth (as represented by the clusters) and language growth over time. In cases where this interaction was not significant, main effects of cluster and age were tested, in order to evaluate the relationship between DF/DB cluster and speech-language scores independent of growth in speech-language scores over time, and the relationship between age and speech-language scores independent of DF/DB cluster membership, respectively. Analyses were conducted using SAS 9.3.

Results

Verbal STM/WM Developmental Clusters

Results of the trajectory analysis for DF supported 3 or 4 cluster solutions, but a 3 cluster solution was ultimately adopted because the 4 cluster solution resulted in a small subgroup with an unacceptably low N (N=5). Classification tables reflecting the probabilities of subjects belonging to their assigned cluster produced probability scores greater than 0.90 for the 3 DF cluster solution (these tables are available from the authors on request). The linear trajectories for the three DF clusters, shown in the left-hand panels of Figure 1, revealed notable differences in baseline scores at age 6 years and the overall elevation (height) of the growth curve between ages 6 and 16 years (Table 2). The slopes of the growth curves over time showed much more modest differences.

DF Cluster 1 reflected the lowest baseline score, lowest overall elevation of the DF scores across the study age range, lowest slope of the growth curve over time, and lowest DF scaled (i.e., norm-based) scores (Figure 1, Panel A; Table 2); as a result, Cluster 1 was labeled “Low Baseline, Low Growth” (DF-LL). DF Cluster 2 had a baseline score, overall elevation, and scaled scores in a mid-range between Clusters 1 and 3, with a slope that was slightly greater than Cluster 1 (Figure 1, Panel B; Table 2). Therefore, Cluster 2 was labeled as “Mid Baseline, High Growth” (DF-MH). DF Cluster 3 had the highest baseline score, elevation, and scaled scores across the study age range, with a slope comparable to that of Cluster 2 (Figure 1, Panel C; Table 2). Hence, Cluster 3 was characterized as “High Baseline, High Growth” (DF-HH).

Results of the trajectory analysis for DB also supported a 3 cluster solution. Classification tables reflecting the probabilities of subjects belonging to their assigned cluster produced probability scores greater than 0.86 for the three DB cluster solution. The DB clusters shown in the right-hand panels of Figure 1 displayed greater differences in slope values than the DF clusters (Table 2). DB Cluster 1 had a low baseline and elevation, as well as a low slope and low scaled scores relative to norms (Figure 1, Panel D; Table 2). This cluster was labeled “Low Baseline, Low Growth” (DB-LL). DB Cluster 2 had a baseline falling between Clusters 1 and 3 and a slope comparable to Cluster 1 (Figure 1, Panel E). This cluster was therefore labeled “Mid Baseline, Low Growth” (DB-ML). DB Cluster 3 had a baseline score slightly higher than Cluster 2 and a slope that was higher than Clusters 1 or 2 (Figure 1, Panel F), resulting in norm-based scaled scores in the average range (Table 2). This cluster was labeled “High Baseline, High Growth” (DB-HH).

Membership and background characteristics of the DF and DB clusters are summarized in Tables 2 and 3. For DF, almost half of subjects fell in the DF-MH cluster (Cluster 2), and most of the remaining subjects were in the DF-LL cluster (Cluster 1). The DF-HH cluster was smaller, encompassing only about 18% of the sample. The DF clusters did not differ significantly in age at implantation, calendar year at implant, best PTA, gender, or communication mode (Table 3). However, DF clusters did differ significantly in age at first testing, with subjects in the DF-LL cluster tested initially at older ages on average than subjects in the other two clusters (Table 3). For the DB clusters, approximately 2/3 of the sample fell in the DB-ML group (Cluster 2), with only 23% and 12% falling in Clusters 1 (DB-LL) and 3 (DB-HH), respectively. The DB clusters did not differ significantly in age at implantation, calendar year of implant, age at first testing, best PTA, gender, or communication mode (Table 3).

Membership in the low, moderate, or high growth clusters for DF was associated with membership in similar growth clusters for DB (Table 4). Half or more of subjects in the low and moderate growth clusters of DF overlapped in the corresponding cluster for DB, while over half of subjects in all three clusters of DB overlapped in the corresponding cluster of DF (Mantel-Haenszel $X^2(1)=20.1$, $p<0.0001$).

Verbal STM/WM Developmental Clusters and Language Growth Over Time

For PBK, the cluster × age effect was not significant for DF (F(2,155=1.95, p>0.14) but was significant for DB (F(2,155)=3.94, p=0.0214), indicating that language growth over time differed for the three different DB clusters. Main effects for DF cluster (F(2,46)=5.24, p=0.0089) and age (F(1,155)=44.79, p<0.0001) on PBK scores were found, indicating that PBK scores improved with age and that DF Cluster 3 (DF-HH) had higher PBK scores at all ages than DF Clusters 1 (DF-LL) and 2 (DF-MH)(Figure 2). For DB, Cluster 2 (DB-ML) had the highest growth rate of PBK scores, followed by Cluster 1 (DB-LL) and Cluster 3 (DB-HH)(Table 5; Figure 2). It should be noted that DB Cluster 3 results were constrained by a ceiling effect, because almost all subjects in that cluster entered the study with high PBK scores (60-80%), and the maximum PBK score is 100% (Figure 2). In contrast, the majority of subjects in DB Clusters 1 and 2 had PBK baseline scores of 40-50% or less. Hence, the lower growth rate in Cluster 3 can be explained by the high baseline scores and ceiling of 100%, as opposed to slow development of spoken word recognition skills.

For HINT-C, neither the age × cluster interaction effects (for DF, F(2,87)=0.30, p>0.74; for DB, F(2,87)=0.02, p>0.98) nor the cluster main effects (for DF, F(2,34)=2.36, p>0.10; for DB, F(2,34)=0.65, p>0.52) were significant. However, chronological age was related to HINT-C scores in both the DF (F(1,87)=11.41, p=0.0011) and DB (F(1,87)=11.29, p=0.0012) analyses, indicating that HINT-C improved significantly over time, regardless of DF or DB cluster membership. Inspection of plots for HINT-C scores by chronological age within each DF and DB cluster (Figure 3) revealed considerable variability in HINT-C baseline and slope in DF and DB Clusters 1 and 2, with a ceiling effect (all scores 80-100%) for a majority of subjects in DF and DB Cluster 3.

For PPVT-III scores, the cluster × age effect was significant for both DF (F(2,213)=5.54, p=0.0045) and DB (F(2,213=7.20, p=0.0009), indicating that vocabulary growth over time with CI use differed for the different DF and DB clusters. For DF, Cluster 1 had the lowest growth rate of PPVT-III scores with age, whereas Clusters 2 and 3 had higher and similar growth rates (Table 5). For DB, Clusters 1, 2, and 3 had incrementally increasing growth rates of PPVT-III scores with age (Table 5). Inspection of plots for PPVT-III scores by chronological age within each cluster (Figure 4) showed a great deal of variability in PPVT-III score baseline values and slopes (growth across age) in Cluster 1 of both DF and DB, with the majority of subjects showing very low baseline scores compared to the other clusters. Additionally, several subjects in Cluster 1 showed little or no growth in PPVT-III raw scores. Clusters 2 and 3 of both DF and DB, on the other hand, showed more uniform and positive patterns of growth in PPVT-III scores, with some variability in baseline values within each cluster (Figure 4).

For CELF scores, the cluster × age effect was significant for DF (F(2,87)=4.23, p=0.0176). Cluster 1 had a much lower growth rate than Clusters 2 or 3 (Table 5), with most subjects showing no change or modest declines in CELF standard scores over time, despite having very low baseline CELF scores (Figure 5). Most subjects in DF Clusters 2 and 3, on the other hand, showed a positive growth rate in CELF scores, although the baseline and elevation of their CELF growth lines was highly variable within clusters (Figure 5). The DB cluster × age effect was nonsignificant for CELF (F(2,87=1.42, p>0.24), as was the main effect for DB cluster (F(2,33)=0.77, p>0.47), likely as a result of the high variability in baseline and slope for CELF scores within each cluster (Figure 5). However, a significant relationship was found between age and CELF scores, indicating a significant growth rate with age for all children (F(1,87)=20.96, p<0.0001).

Discussion

The results of this study provide several new insights about the growth patterns of verbal STM/WM skills that occur in children with CIs and how these growth patterns are related to the development of speech and language skills. Unlike prior research, the use of multiple time point evaluations of verbal STM/WM skills in the present study allowed for investigation of types and frequencies of growth trajectories of verbal STM/WM skills. Furthermore, the longitudinal data obtained for this study revealed relationships between the pattern of growth of verbal STM/WM skills and the development of speech-language skills. Some growth trajectories of verbal STM/WM skills were related to more positive language skill development, whereas other trajectories were associated with slower rates of language skill development.

For DF, a measure of verbal STM capacity, the most common cluster trajectory (N=32/66 subjects) was a mid-range baseline score followed by a high slope. Relative to norms, this cluster fell well below the average for normal hearing children at both the first and final visits, with mean scaled scores of 5.5 and 6.7, respectively (Table 2). On the other hand, a small proportion (12/66) of the sample showed a high DF baseline score and high slope, with mean scaled scores that were approximately average for age at the first and final visits. However, one-third of the sample (22/66 subjects) displayed a low baseline with a low slope of growth of DF scores as well as mean scaled scores that were 2 standard deviations or more below those for the typically developing, normal hearing normative WISC-III sample, suggesting the presence of significant vulnerability to delayed development of verbal STM. It is notable that this latter group was older than the other DF clusters at the first digit span testing visit (Table 3), although all of the DF clusters were assessed across a wide range of ages (Figure 1).

For DB, a measure of verbal WM capacity, the most common cluster trajectory (43/66 subjects) also fell into the mid-range, with a mid-range baseline score followed by a low slope. Again, however, mean scaled scores for that cluster were below age norms for normal hearing children (Table 2). Similar to DF, a small (8/66) cluster of subjects demonstrated high baseline, high growth, and mean DB scaled scores in the average range for their chronological age. Membership in the low-slope, low-baseline group (Cluster 1) was less common for DB than for DF, with 23% of subjects falling in that group, which fell well below scale norms.

The DF and DB clusters generally did not differ in conventional demographic, device, and medical factors, such as age when first tested, age at implantation, chronological year of implantation, best pre-implant PTA, or communication mode. Only the low-performing DF cluster was found to have been initially tested at a later chronological age than the other DF clusters. Given the existing research demonstrating relations between demographic, device, and medical factors and speech-language outcomes in children with CIs (Geers & Sedey, 2011; Niparko et al., 2010), it may be surprising that so few relationships were found between demographic, device, and medical factors and verbal STM/WM development in this longitudinal dataset. However, this finding is generally consistent with cross-sectional research investigating relationships between digit span and demographic, device, and medical factors. Prior cross-sectional research has found a positive relationship between DF scores and use of auditory-oral communication strategies (vs. simultaneous communication strategies), but DB was not related to any of the conventional demographic, device, or medical factors, replicating earlier findings reported by Pisoni & Cleary (2003). Furthermore, the relationship between digit span scores and speech-language scores was attenuated minimally (and remained significant) after partialing out demographic, device, and medical factors (Pisoni et al., 2011). Thus, demographic, device, and medical factors may play a smaller role in verbal short-term/working memory development than they do in speech-language development. Furthermore, the relationship between verbal STM/WM development and speech-language development appears to be only minimally affected by conventional demographic, device, and medical factors.

Cluster membership for DF was strongly predictive of the growth of higher-order language scores such as receptive vocabulary (PPVT-III) and complex receptive/expressive language (CELF), whereas cluster membership for DB was predictive of the growth of vocabulary (PPVT-III) and word recognition (PBK) scores, even after controlling for conventional demographic, device, and medical factors. This is the first study to empirically derive clusters of verbal STM/WM growth as a function of age in a longitudinal sample of children with CIs and to demonstrate that these clusters are related to the *growth* of language skills, as opposed to endpoint language outcome measures obtained at only one point in time. Viewed in this way, the present findings are relevant not for single endpoint outcomes of STM/WM and speech-language skills but for understanding the dynamic process of development of these core underlying foundational skills.

The DF clusters identified in this study differed primarily in baseline score and much less in growth with increasing age (i.e., slope). This finding suggests that types of verbal STM growth in children with CIs may be related more to the level of those skills at early ages (e.g., baseline) as opposed to differences in the way that those skills change and grow over time with CI use. A high initial baseline score tended to be associated with high elevation throughout development, whereas a low initial baseline score tended to be associated with low elevation throughout development. Furthermore, more children fell in a low baseline-low growth cluster (Cluster 1; DF-LL) than in a high baseline-high growth cluster (Cluster 3; DF-HH), consistent with prior longitudinal research demonstrating that most deaf children with CIs lag significantly behind their normal-hearing peers in verbal STM capacity (as measured by DF) skills, with little “catch up” over time (Pisoni et al., 2011).

The DB clusters, on the other hand, differed not only in initial baseline but also in slope (growth over time), with two subgroups showing lower growth in verbal WM capacity (as measured by DB) skills than the third subgroup that had much higher growth in verbal WM skills. In the case of DB, the majority of the sample had a mid-range baseline score but lower growth, consistent with Pisoni et al.'s (2011) earlier finding that children with CIs scored close to DB age norms at age 8-9 years, but that this difference widened with increases in age when tested about 8 years later.

DF and DB scores in the sample differed from each other in growth over time (e.g., cluster slopes) and in scaled scores compared to norms. Compared to DF, the DB clusters showed a larger slope (Table 2), indicating that DB raw scores grew at a faster pace than DF raw scores. This finding likely is a result of lower DB baseline raw scores allowing more room for growth in raw scores over time (see Figure 1). Additionally, DB scaled scores at the first and the last visit were higher than DF scaled scores (Table 1), showing that the digit span performance of the CI sample was less discrepant than that of age peers for DB than for DF (although the CI sample scored well below norms for both DB and DF). This may reflect the fact that DF places almost sole emphasis on rapid phonological coding and short-term verbal memory, which are severely affected in children with CIs, whereas DB also includes a component of controlled attention during reordering of digits, which may be less impaired (although still below average) in children with CIs relative to norms. As a result, relative to norms, it is possible to obtain a higher scaled score with a shorter sequence of digits on the DB task than on the DF task because the demands of controlled attention during reordering of digits in DB reduce the number of digits that can be recalled by the average individual. To the extent that verbal STM (sequence length) is a greater challenge than controlled attention (retaining memory during backward reordering) for children with CIs, they will score lower relative to norms on the task that exclusively focuses on verbal STM (DF) compared to the task that includes both verbal STM and a reordering component (DB).

Strong and significant relations between verbal STM capacity as measured by DF scores and conventional endpoint product measures of speech-language performance have been reported in earlier studies of children with CIs. Pisoni & Cleary (2003) were the first researchers to report strong associations between DF scores and spoken word recognition scores in a cross-sectional study of 8-9 year old children with CIs (see also Pisoni & Geers, 1998). Using longitudinal follow-up data, Pisoni et al. (2011) found significant relations between DF scores at age 8-9 years of age and speech perception, sentence recognition, vocabulary, and complex language skills eight years later at 16-17 years of age. DF scores at age 16-17 years were also significantly correlated with speech and language functioning measured at that same time point. Furthermore, the change in DF scores between ages 8-9 years and 16-17 years predicted speech perception, vocabulary, and complex language skills at age 16-17 years (Pisoni et al., 2011). The current findings with a different sample of children are also consistent with findings from studies of normal hearing children, children with language/learning disorders (Gathercole & Baddeley, 1993), and children with mild to moderate hearing impairment (Hansson et al., 2004), which all show that verbal STM/WM is related to vocabulary and language development.

The present study differs from earlier research by demonstrating that the *pattern of development* of verbal STM/WM over time is related to the *growth trajectory* of speech-language skills, as opposed to just a single endpoint measurement. This is a theoretically important distinction, because the same endpoint can be reached with different types of growth and development over time. By demonstrating that growth patterns of development of verbal STM/WM capacity are related to growth in speech-language skills, the present findings reveal a relationship between the dynamic, transactional processes of development of verbal STM/WM and speech-language skills over time, as opposed to a single point predictive or concurrent relationship as shown in earlier studies (e.g., Pisoni et al., 2011).

Although the DF clusters were primarily differentiated by the baseline DF score as opposed to the rate of growth of DF scores over time, the growth (trajectory) of vocabulary (PPVT-III) and complex language development was different for the different DF clusters. Children with higher baseline verbal STM achieved faster growth in those language skills, compared to children with lower baseline verbal STM. This pattern suggests that children with more robust early verbal STM skills may have better endpoint speech-language outcomes due to faster growth in speech-language skills, as opposed to simply having stronger speech-language skills at baseline (Pisoni et al., 2011).

For DB, past research has generally found somewhat weaker relations with endpoint speech and language functioning (albeit significant in several areas), and DB scores tend to be more highly correlated with complex language functioning measures than with elementary speech perception and sentence recognition skills (Pisoni et al., 2011). The results of the present study are partly consistent with those earlier findings using a different sample: DB cluster was related to growth in vocabulary (PPVT-III) but not complex language (CELF) scores. DB clusters with higher baseline and/or higher slope (growth) of DB scores had greater vocabulary growth. Interestingly, DB cluster membership was also associated with growth in spoken word recognition (PBK) scores, although this finding should be interpreted with caution because one of the clusters (Cluster 3; DB-HH) had an artificially low PBK growth rate because of a ceiling effect for the high-performing children in this subgroup. It is not clear why DB cluster membership was related to growth of PPVT-III scores but not to growth of CELF scores. This finding contrasts with findings for DF and CELF in the present study and with findings for single time-point measurement of DB and CELF scores from other studies (e.g., Pisoni et al., 2011). It is possible that smaller CELF sample size may have reduced statistical power for some analyses.

Unlike the other speech-language measures, growth over time in sentence repetition (HINT-C) scores was unrelated to either the DF or DB clusters. This finding was unexpected, given that sentence repetition skills in children with CIs require the use of verbal STM/WM (Kronenberger et al., 2011). Furthermore, sentence recognition skills have been found to be related to verbal STM scores as measured by digit span (Pisoni & Cleary, 2003; Pisoni et al., 2011). It may be that the relationship between verbal STM/WM skills and sentence repetition skills is driven much more by the initial baseline abilities in these areas than by differences in the way that these skills develop over time. If this is the case, then a relationship between verbal STM/WM and sentence repetition skills would reflect a consistent association of those skill areas over time (as opposed to differences in how the skills grow) and would result in a significant correlation of the endpoint measures, as has been reported in prior research. Alternatively, HINT-C baseline and slope values in the present sample showed wide variability within DF and DB Clusters 1 and 2 as well as a ceiling effect in Cluster 3, which may have reduced the statistical power of the analyses.

Although significant relations were found between verbal STM/WM development and speech-language development in this sample, the variability of speech-language development within each DF and DB cluster is also quite striking (Figures 2-5). The enormous range of variability within subgroups indicates the need for further exploration of additional factors that may explain different baselines and trajectories of development of speech-language skills in children with CIs. However, some patterns can be observed within the variability of speech-language outcomes. For PBK and HINT-C, the greatest variability occurred in Clusters 1 and 2 for DF and DB, whereas Cluster 3 tended to have more uniformly higher scores (to the point of approaching ceiling effects, as shown in Figures 2 and 3). This may indicate that the strongest verbal STM/WM development is related to more uniform positive development of speech perception and sentence repetition scores, while other factors are more influential for mid-range and poor verbal STM/WM development. For PPVT-III, we found considerably less variability in scores within each DF and DB cluster (Figure 4), and the age × cluster effect was strongest for PPVT-III for both DF and DB. This suggests robust relations between verbal STM/WM development and vocabulary development in children with CIs. For CELF, the most striking observation was the uniformly very low performance of children in Cluster 1 (Low-Low) groups, with almost all scores below a standard score of 70 and a decline in most scores with time. Because these are standard scores, a decline in scores reflects poorer performance relative to normal-hearing peers, as opposed to a true decline in absolute language skills. In contrast, almost all subjects in DF and DB Cluster 3 (High-High) scored above 80 on the CELF and showed positive trajectories in their scores, indicating improvement in language skills relative to peers over time (Figure 5). These findings should be interpreted cautiously because of missing data for some speech-language measures and because of limited size for some clusters, but they provide important directions for future investigations.

The results of this study have several important clinical implications. First, early baseline skills in verbal STM (DF) may be very important for anticipating and characterizing verbal STM growth over time and in predicting later vocabulary and language growth. For verbal WM (DB), on the other hand, differences in slope (growth) of skills over time were at least as influential in cluster membership as baseline scores, with one cluster showing marked improvement over time relative to the other two clusters. This high-growth DB cluster also had much higher growth in vocabulary (PPVT-III) skills than the other DB clusters, suggesting that the pace of verbal WM growth coincides with the pace of vocabulary growth, a finding that is consistent with earlier research in normal-hearing children (Pickering & Gathercole, 2004). Hence, for children with CIs, low baseline and/or slow growth of verbal STM/WM as assessed by digit span may indicate elevated risk for slow development of speech-language skills and need for early intervention. As research on the important role of working memory in language processing and the value of novel interventions to improve working memory becomes available, this information may be discussed with parents at the time of evaluation for candidacy to support a plan for future evaluation of working memory and future interventions as needed based on the results of working memory evaluations. Second, only a small minority (12-18%) of the sample fell into the high performing (HH; Cluster 3) DF and DB clusters. For both DF and DB, more of the sample fell into the lowest performing than the highest performing cluster, an effect that was particularly strong for DF, with 1/3 of the sample falling in the DF-LL cluster. Hence, verbal STM appears to be at high risk not only for a low baseline but also for low growth in many deaf children with CIs (particularly those in DF Cluster 1). Similar results have been found in other studies (Pisoni et al., 2011), but this is the first study to demonstrate this finding using empirically-based clustering techniques with a longitudinal design employing multiple observations of digit span and speech-language measures over time for the same children. Third, this study demonstrated that verbal STM/WM may be important not only for predicting endpoint speech-language functioning in children with CIs but also for characterizing how speech-language performance grows throughout development during this age range.

Given these clinical implications of relations between verbal STM/WM development and speech-language growth in children with CIs, it is possible that interventions to improve verbal STM/WM may positively influence the dynamic process of development of speech and language in children with CIs. Kronenberger et al. (2011), for example, found improvement in verbal working memory capacity, sentence repetition, and sentence memory skills in a small sample of deaf children with CIs who completed short-term computer-based working memory training. These improvements may be explained by improvements in memory scanning speed and verbal rehearsal speed, which were also found to increase after working memory training (Kronenberger et al., 2010). The findings from the current study suggest that improvements in working memory capacity may help not only the endpoint speech-language achievement of children with CIs but also the dynamic process of development of these speech-language skills. Alloway (2011) provides several suggestions for improving working memory skills in academic environments; these interventions may offer the potential to enhance didactic learning and explicit training of speech and language skills, especially in this clinical population. More research is needed to investigate these possibilities.

The results of the present study must be interpreted in the context of several methodological characteristics and limitations. First, the number of subjects in the study, while large for a longitudinal study of children with CIs, is a limiting factor that constrains the number of clusters that can be empirically identified. Clusters that are more unusual would be more difficult to identify in a study with a modest sample size. Hence, additional clusters may be identified in future research with larger sample sizes. Second, the sizes of the strongest performing DF and DB clusters (Cluster 3; High-High) were much smaller than the other two clusters, which may have limited the statistical power of analyses involving these clusters. Third, because the sample was already participating in an ongoing longitudinal study, subjects completed the digit span test at different ages. Although chronological age factors were statistically controlled in all analyses, it is possible that age differences had some influence on results. Fourth, because digit span tests were completed multiple times, practice effects may have influenced results to some extent. However, the role of practice effects was likely to be minimal because subjects were tested no more frequently than at 6 month intervals (and typically at longer intervals), and retest effects on digit span scores are minimal, even for intervals as short as 1 month (Wechsler et al., 2004). Fifth, subjects sometimes were unable to complete scheduled evaluations as a result of personal or family factors, failing to show up at scheduled times, and medical issues, resulting in missing data at specific time points. Sixth, subjects were sometimes unable to provide valid digit span or other test scores because of fatigue, lack of sufficient effort, or difficulty understanding the tasks. In such cases, their data could not be obtained by the examiner or (in the case of DF and DB scores both equal to 0) were dropped from analysis because validity was suspect. This effect was largest for the CELF (N=39) and HINT-C (N=40) scores, which were completed by less than 2/3 of the total sample. Because we did not systematically code for the reason that data were not obtained, we cannot describe the cause for each instance of missing data. Seventh, we did not systematically measure the functioning of the CI at each visit, so variability in the functioning of the CI could have impacted scores at some visits. However, this is not likely to have been a significant factor because all subjects were regularly followed by audiology (with adjustments as needed), and the speech-language pathologists conducting the testing for the study administered tests only if the functioning of the CI was sufficient to obtain valid results. Finally, we did not include a normal hearing control group because the emphasis in this study was on patterns of verbal STM/WM development and growth over time in children with CIs. However, it may be important in future studies to compare these growth patterns to the types and frequencies of growth patterns of verbal STM/WM development obtained from normal hearing samples. While we expect that a CI group would show disproportionately large membership in the lower performing digit span clusters (based on research showing verbal STM/WM delays in CI samples), inclusion of a normal hearing sample would allow a direct empirical test of this.

In summary, this study obtained empirically derived clusters of verbal STM/WM growth for 66 children with CIs based on digit span scores. These DF and DB clusters were found to be related to the growth of several core speech and language skills. The present findings suggest that development of verbal STM/WM over time is important not only for endpoint speech and language outcomes but also for understanding the dynamic process of growth in core underlying foundational speech and language skills in children with CIs as they develop through childhood and early adolescence.

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Appendix

Appendix

Device and Strategy for Subjects in Sample

Subject	Device	Strategy at Visit 1	Strategy at Final Visit
1	CC-Nucleus 22	MPEAK	SPEAK
2	CC-Nucleus 22	MPEAK	SPEAK
3	CC-Nucleus 22	SPEAK	SPEAK
4	CC-Nucleus 22	SPEAK	SPEAK
5	CC-Nucleus 22	SPEAK	SPEAK
6	CC-Nucleus 22	SPEAK	SPEAK
7	CC-Nucleus 22	SPEAK	SPEAK
8	CC-Nucleus 24	ACE	ACE
9	CC-Nucleus 24	ACE	ACE
10	CC-Nucleus 24	ACE	ACE
11	CC-Nucleus 24	ACE	ACE
12	CC-Nucleus 24	ACE	ACE
13	CC-Nucleus 22/24*	SPEAK	ACE
14	CC-Nucleus 22	SPEAK	SPEAK
15	CC-Nucleus 22	SPEAK	SPEAK
16	CC-Nucleus 22	SPEAK	SPEAK
17	CC-Nucleus 22	SPEAK	SPEAK
18	CC-Nucleus 22	SPEAK	SPEAK
19	CC-Nucleus 22	SPEAK	SPEAK
20	CC-Nucleus 22	SPEAK	SPEAK
21	CC-Nucleus 22	SPEAK	SPEAK
22	CC-Nucleus 22	SPEAK	SPEAK
23	ABC-CL	CIS	CIS
24	CC-Nucleus 22	SPEAK	SPEAK
25	CC-Nucleus 22	SPEAK	SPEAK
26	CC-Nucleus 22	SPEAK	SPEAK
27	ABC-CL	CIS	CIS
28	ABC-CL	CIS	CIS
29	ABC-CL	CIS	CIS
30	ABC-CL	CIS	MPS
31	ABC-CL	CIS	HiRes
32	CC-Nucleus 22	SPEAK	SPEAK
33	CC-Nucleus 22	SPEAK	SPEAK
34	CC-Nucleus 22	SPEAK	SPEAK
35	CC-Nucleus 22	SPEAK	SPEAK
36	CC-Nucleus 22	SPEAK	SPEAK
37	CC-Nucleus 22/24*	SPEAK	ACE

Subject	Device	Strategy at Visit 1	Strategy at Final Visit
38	CC-Nucleus 24	ACE	ACE
39	ME-Combi 40+	CIS	CIS
40	CC-Nucleus 24	SPEAK	ACE
41	CC-Nucleus 24	ACE	ACE
42	ABC-CL-HF	SAS	SAS
43	CC-Nucleus 24	ACE	ACE
44	ABC-CL	MPS	MPS
45	ABC-CL	SAS	HiRes
46	CC-Nucleus 24	ACE	ACE
47	ABC-CL-HF	SAS	SAS
48	ABC-CL-HF	SAS	SAS
49	CC-Nucleus 24	ACE	ACE
50	CC-Nucleus 24	ACE	ACE
51	CC-Nucleus 24	ACE	ACE
52	CC-Nucleus 24	ACE	ACE
53	CC-Nucleus 22	MPEAK	SPEAK
54	CC-Nucleus 24	SPEAK	ACE
55	CC-Nucleus 22	SPEAK	SPEAK
56	CC-Nucleus 22	SPEAK	SPEAK
57	CC-Nucleus 22	SPEAK	SPEAK
58	ABC-CL	CIS	CIS
59	CC-Nucleus 24	SPEAK	SPEAK
60	CC-Nucleus 22	SPEAK	SPEAK
61	CC-Nucleus 22	SPEAK	SPEAK
62	ME-Combi 40+	CIS	CIS
63	CC-Nucleus 22	SPEAK	ACE
64	CC-Nucleus 22	SPEAK	SPEAK
65	CC-Nucleus 24	ACE	ACE
66	ABC-CL	MPS	MPS

Note: CC=Cochlear Limited; ABC=Advanced Bionics Corporation; ME=MED-EL

[*] Device changed to CC Nucleus 24 prior to final visit

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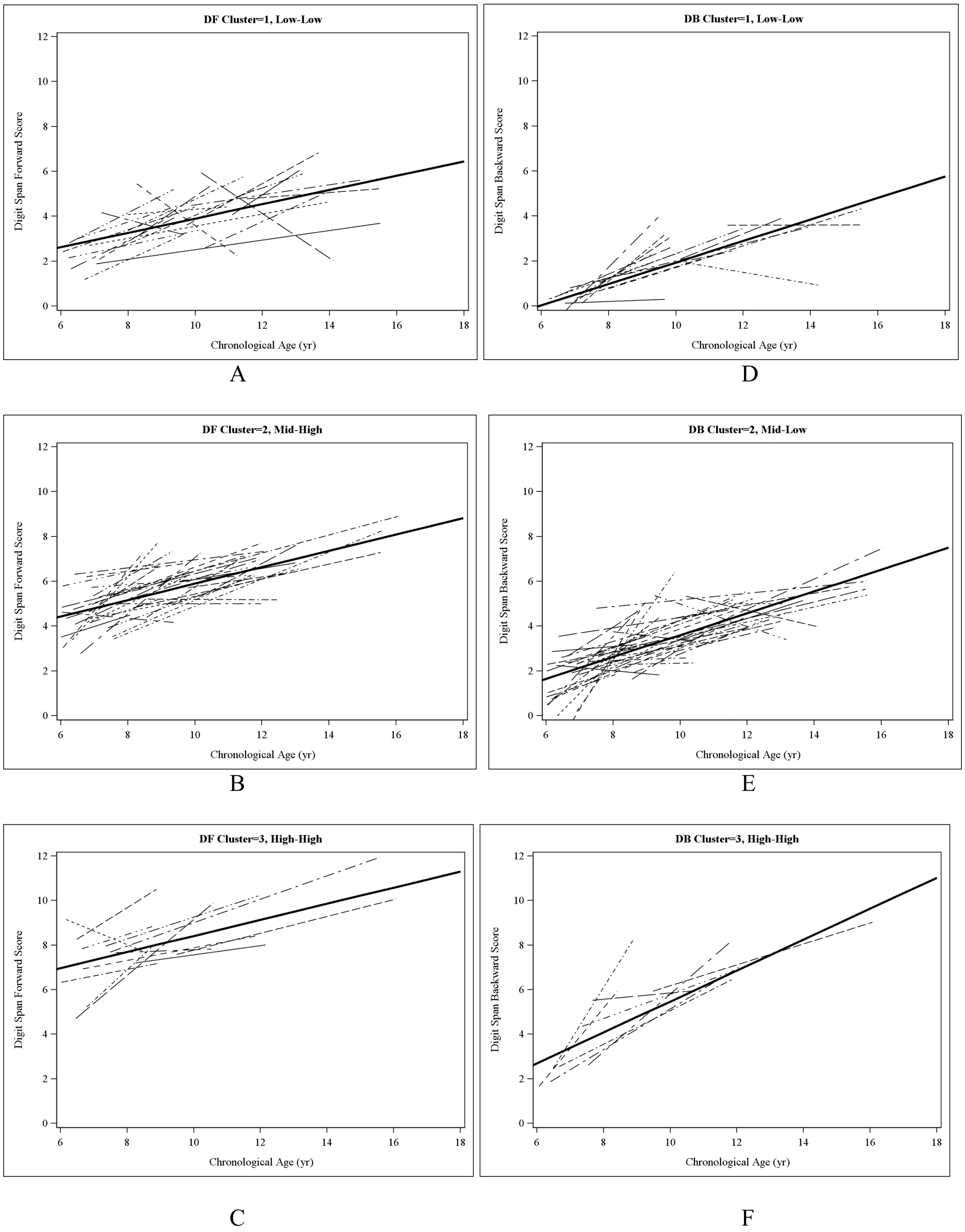
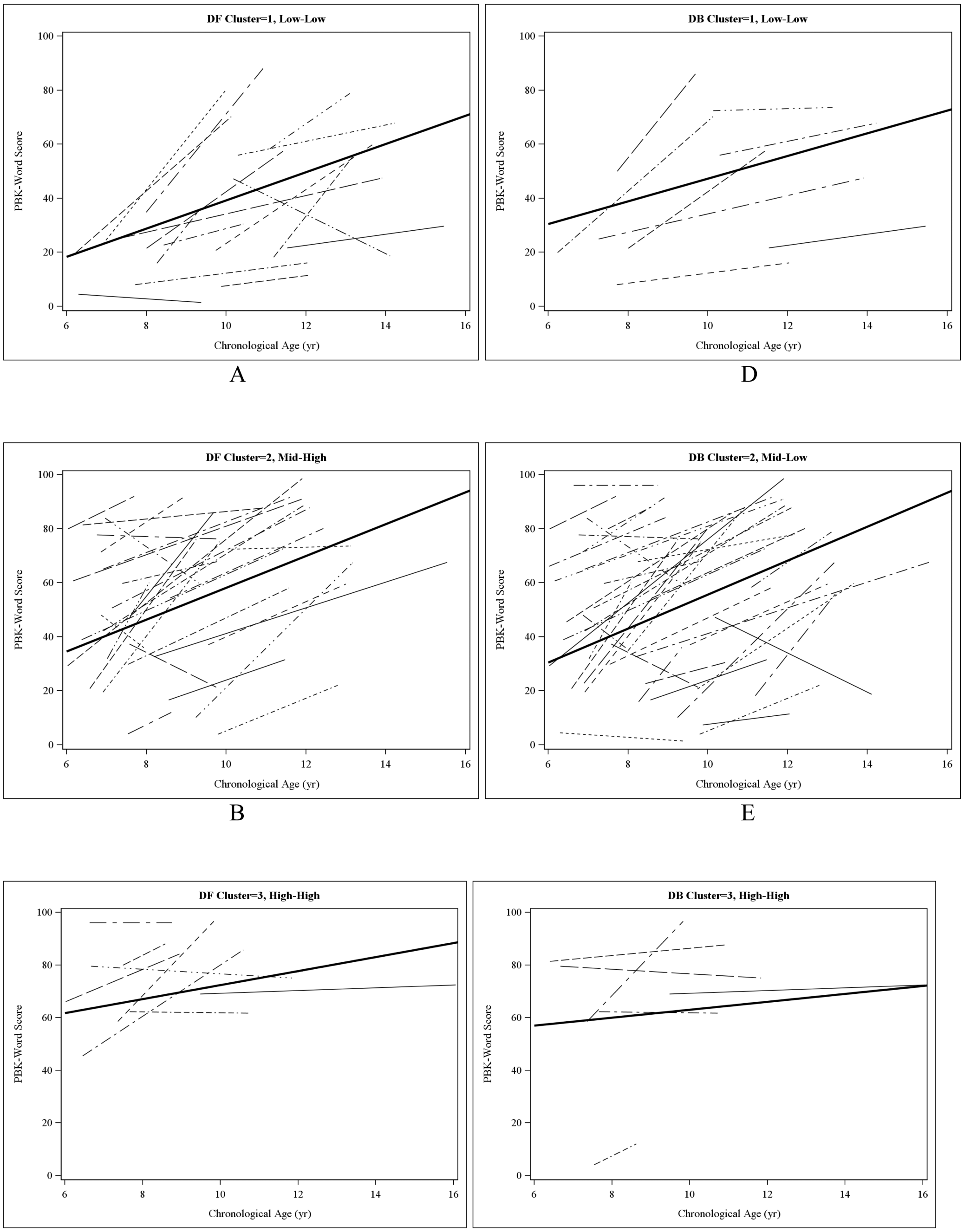


Figure 1.

Developmental Clusters for Digit Span Forward (DF) and Backward (DB). Lines represent fitted scores (based on linear regression) of WISC-III Digit Span raw scores by age for individual subjects for DF Clusters 1 (Panel A), 2 (Panel B), and 3 (Panel C) and for DB Clusters 1 (Panel D), 2 (Panel E), and 3 (Panel F). The bold solid line in each plot represents the linear developmental trajectory for the cluster group depicted.



DF Cluster=1, Low-Low

DB Cluster=1, Low-Low

A

D

DF Cluster=2, Mid-High

DB Cluster=2, Mid-Low

B

E

DF Cluster=3, High-High

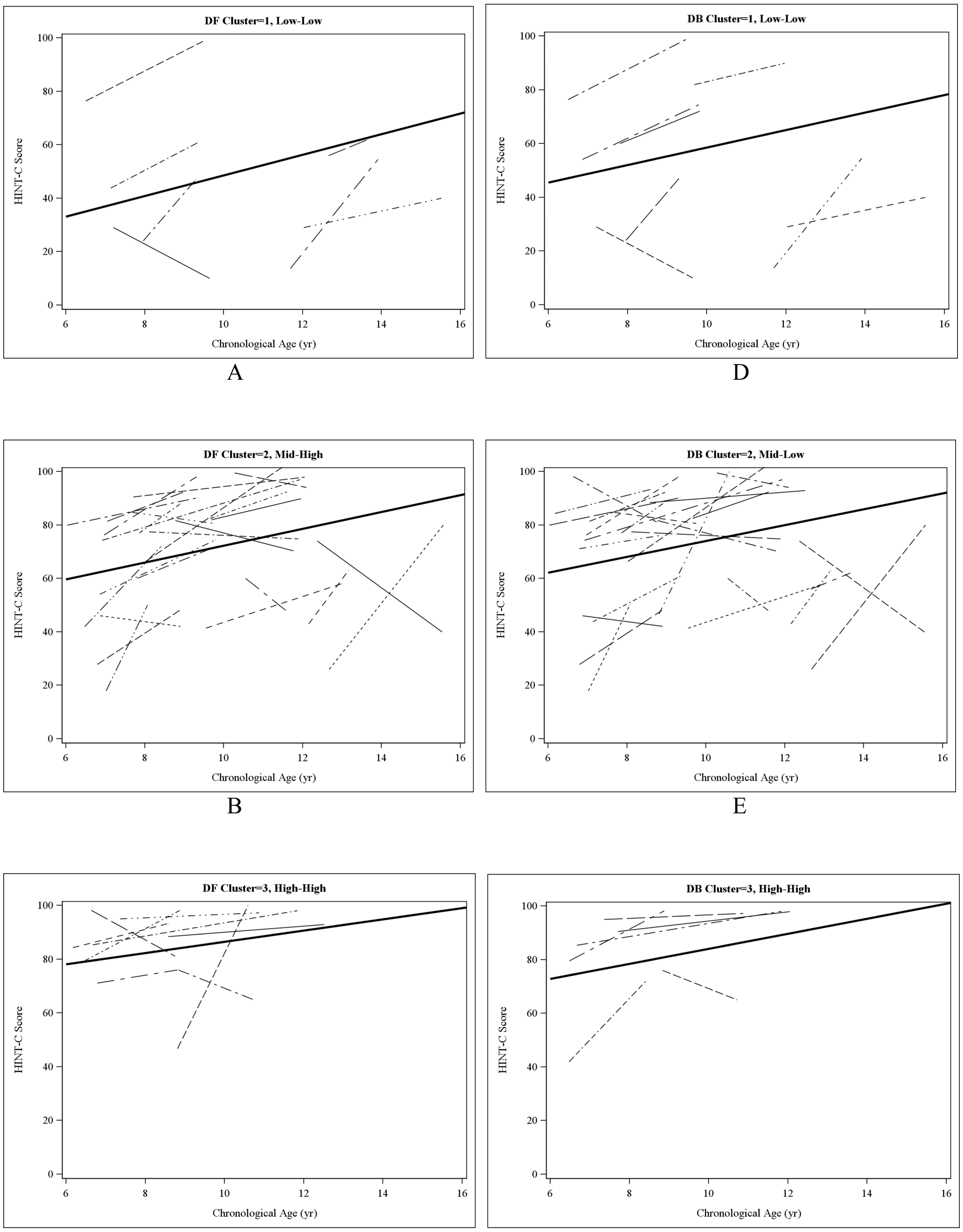
DB Cluster=3, High-High

C

F

Figure 2.

Growth in PBK Scores for DF and DB Clusters. Lines represent fitted scores (based on linear regression) of PBK Word % Correct scores by age for individual subjects for DF Clusters 1 (Panel A), 2 (Panel B), and 3 (Panel C) and for DB Clusters 1 (Panel D), 2 (Panel E), and 3 (Panel F). The bold solid line in each plot represents the predicted regression line for PBK from a mixed effects repeated measures model for each DF and DB cluster across age at the average value of baseline covariates (age at implantation, best pre-implant PTA, maternal education, and communication mode).



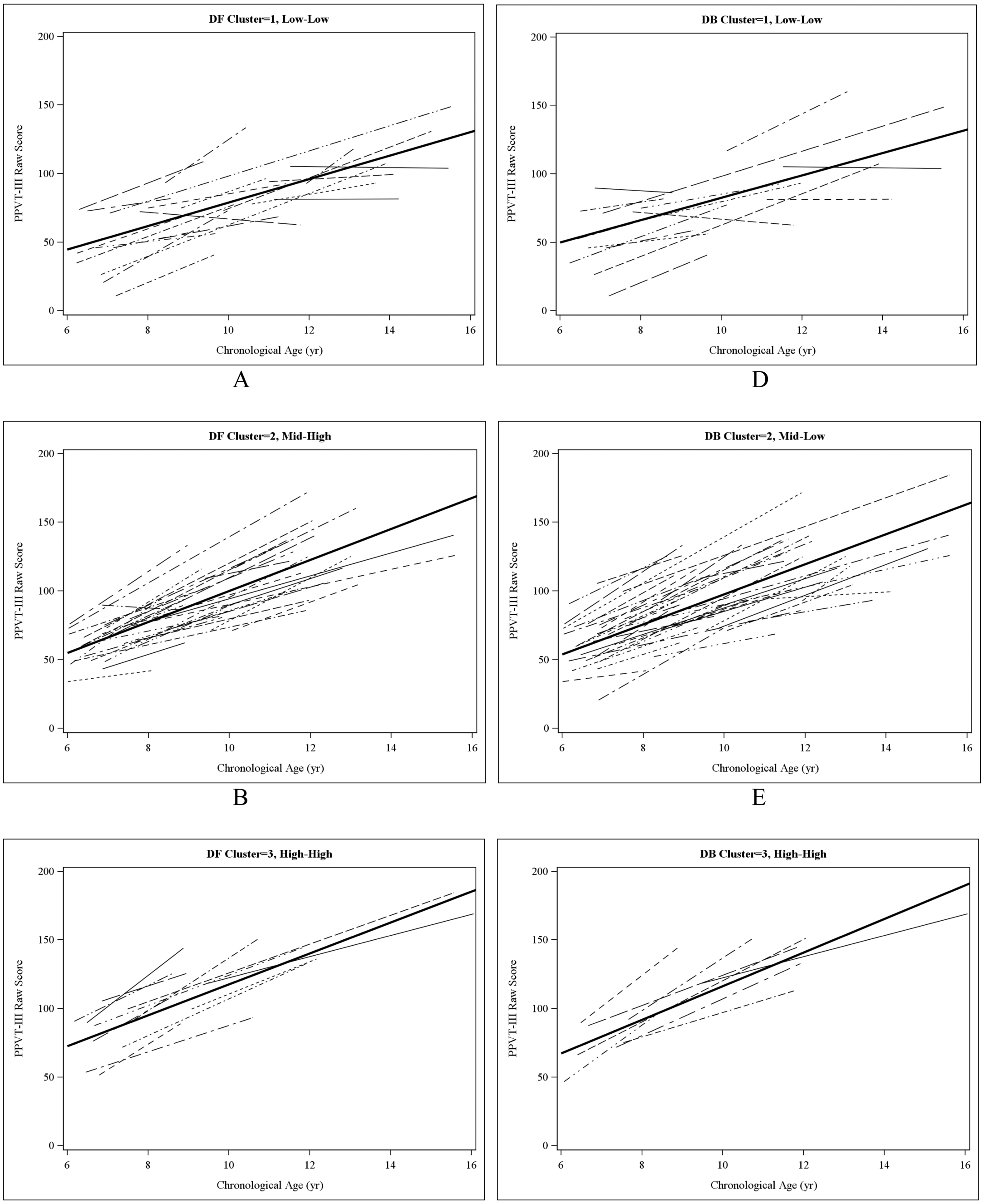
A D

B E

C F

Figure 3.

Growth in HINT-C Scores for DF and DB Clusters. Lines represent fitted scores (based on linear regression) of HINT-C scores by age for individual subjects for DF Clusters 1 (Panel A), 2 (Panel B), and 3 (Panel C) and for DB Clusters 1 (Panel D), 2 (Panel E), and 3 (Panel F). The bold solid line in each plot represents the predicted regression line for HINT-C from a mixed effects repeated measures model for each DF and DB cluster across age at the average value of baseline covariates (age at implantation, best pre-implant PTA, maternal education, and communication mode).



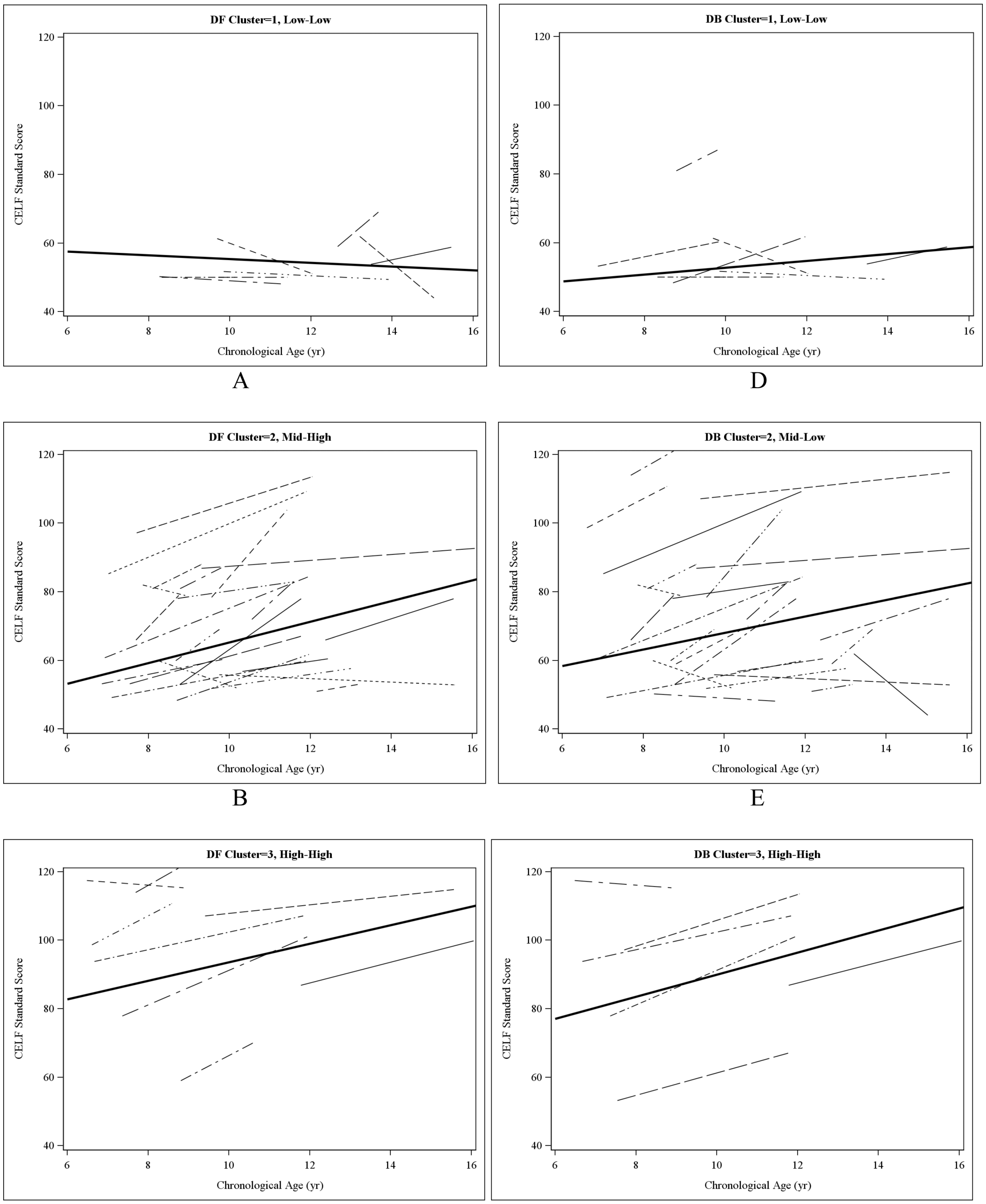
A D

B E

C F

Figure 4.

Growth in PPVT-III Scores for DF and DB Clusters. Lines represent fitted scores (based on linear regression) of PPVT-III raw scores by age for individual subjects for DF Clusters 1 (Panel A), 2 (Panel B), and 3 (Panel C) and for DB Clusters 1 (Panel D), 2 (Panel E), and 3 (Panel F). The bold solid line in each plot represents the predicted regression line for PPVT-III from a mixed effects repeated measures model for each DF and DB cluster across age at the average value of baseline covariates (age at implantation, best pre-implant PTA, maternal education, and communication mode).



A D

B E

C F

Figure 5.

Growth in CELF Scores for DF and DB Clusters. Lines represent fitted scores (based on linear regression) of CELF Total (CELF-3) or Core Language (CELF-4) standard scores by age for individual subjects for DF Clusters 1 (Panel A), 2 (Panel B), and 3 (Panel C) and for DB Clusters 1 (Panel D), 2 (Panel E), and 3 (Panel F). The bold solid line in each plot represents the predicted regression line for CELF from a mixed effects repeated measures model for each DF and DB cluster across age at the average value of baseline covariates (age at implantation, best pre-implant PTA, maternal education, and communication mode).

Table 1

Sample Characteristics

	N	Mean	SD	Range
Age (Years) at First Visit	66	7.6	1.4	6.0-11.5
Age (Years) at Final Visit	66	11.4	2.2	8.1-16.1
Maternal Education[a]	66	4.7	1.8	2-9
Pre-Implant Mean PTA[b]	66	108.4	9.9	83.3-120.1
Age (Years) at Implantation	66	3.8	1.7	1.4-8.0
Duration of CI Use (Years) – First Visit	66	3.7	1.4	1.0-8.0
Digits Forward Scaled Score – First Visit	66	5.8	2.9	2.0-16.0
Digits Forward Scaled Score – Final Visit	66	6.5	2.9	1.0-13.0
Digits Backward Scaled Score – First Visit	66	7.0	2.6	2.0-13.0
Digits Backward Scaled Score – Final Visit	66	7.9	3.2	2.0-15.0
Number of Digit Span Assessments	66	4.5	1.3	3.0-9.0
Time Period of Digit Span Assessments (First Visit to Final Visit, Years)	66	3.9	1.7	2.0-8.9
Number of PBK Assessments	52	4.1	1.3	2.0-7.0
Number of HINT-C Assessments	40	3.3	1.3	2.0-7.0
Number of PPVT-III Assessments	65	4.3	1.3	2.0-9.0
Number of CELF Assessments	39	3.3	1.4	2.0-7.0
Communication Mode – Oral	43			
– Simultaneous	23			
Gender – Male	34			
– Female	32			
Race/Ethnicity[c] – White	63			
– Black	4			
– Hispanic	1			

Note: Digits Forward and Digits Backward scores are scaled scores based on comparison to age-based norms (normative mean of 10, SD of 3; Kaplan et al., 1999). Communication mode was coded as oral or simultaneous based on the child's preference for communication during the testing visit.

[a] Coded on a 1 (no GED or high school diploma) to 5 (2 years college) to 9 (doctoral graduate degree) scale.

[b] Mean unaided pure-tone average in dB HL.

[c] Race adds to more than total N because some subjects reported more than one race.

Table 2

Baseline and Slope Values for Digits Forward (DF) and Digits Backward (DB) Clusters

			Raw Scores		Mean Scaled Score	
	Cluster	N	Baseline Score	Slope Value	First Visit	Final Visit
DF	1 (LL)	22	2.63 (0.31)	0.32 (0.06)	3.9 (0.2)	4.0 (0.3)
	2 (MH)	32	4.44 (0.27)	0.37 (0.06)	5.5 (0.3)	6.7 (0.3)
	3 (HH)	12	6.97 (0.36)	0.36 (0.08)	10.3 (0.7)	10.8 (0.6)
DB	1 (LL)	15	0.03 (0.38)	0.48 (0.07)	6.0 (0.5)	5.5 (0.4)
	2 (ML)	43	1.65 (0.38)	0.49 (0.06)	6.8 (0.4)	8.4 (0.5)
	3 (HH)	8	2.69 (0.63)	0.69 (0.24)	9.6 (0.7)	11.1 (0.6)

Note: Digit Span raw scores are depicted for baseline score and slope value, whereas scaled scores (norm-based scores) are depicted for first and final visit. Baseline score and slope values are derived from Digits Forward or Digits Backward raw scores, based on linear estimate for each cluster. Baseline score is value of linear estimate for each cluster at age 6 years, 0 months. Slope value is slope of linear estimate for each cluster (growth over time [age years]). Mean scaled scores are average of DF or DB scaled scores (relative to WISC-III norms) for each cluster for the first visit and final visit of each subject. Values in parentheses are standard errors.

Table 3

Demographic and Device Variables Differences by Digit Span Cluster

	CY Of Implant		Age at Implant		Age at First Testing		Best PTA		Comm Mode
Digits Forward	**Mean**	**SE**	**Mean**	**SE**	**Mean**	**SE**	**Mean**	**SE**	**% OC**
Cluster 1 (LL)	1995.55	(0.59)	4.19	(0.73)	8.21	(0.30)	110.34	(2.12)	59.1
Cluster 2 (MH)	1996.72	(0.49)	3.65	(0.31)	7.27	(0.25)	107.21	(1.76)	62.5
Cluster 3 (HH)	1997.25	(0.80)	3.54	(0.50)	7.12	(0.40)	107.80	(2.87)	83.3
F (2,63)	1.81		0.81		3.74[*]		0.67		---
p-value	0.18		0.45		0.03		0.52		0.36

Digits Backward	**Mean**	**SE**	**Mean**	**SE**	**Mean**	**SE**	**Mean**	**SE**	**% OC**
Cluster 1 (LL)	1996.87	(0.71)	3.83	(0.45)	7.76	(0.38)	108.37	(2.59)	53.3
Cluster 2 (ML)	1995.95	(0.42)	3.78	(0.27)	7.55	(0.22)	108.67	(1.53)	67.4
Cluster 3 (HH)	1998.13	(0.97)	3.98	(0.62)	7.21	(0.52)	106.68	(3.55)	75.0
F (2,63)	2.34		0.04		0.38		0.13		---
p-value	0.11		0.96		0.69		0.88		0 .55

Note: DF=Digits Forward; DB=Digits Backward; CY=chronological year; PTA=pure tone average in better hearing ear at evaluation prior to CI surgery (in dB HL); Comm Mode=Communication Mode (% oral communication [OC] with remainder being % simultaneous communication). F-values are for one-way ANOVA comparing DF or DB clusters. For Communication Mode, only the p-value for Fishers' Exact Test is reported. Values in parentheses are standard errors (SE).

[*] $p<0.05$

Table 4

Number of Subjects in Digits Forward and Digits Backward Clusters

	Digits Backward Cluster		
	1-LL	**2-ML**	**3-HH**
Digits Forward Cluster			
1 – LL	11	11	0
2 – MH	4	25	3
3 – HH	0	7	5

Note: Values are number of subjects. Mantel-Haenszel $X^2(1)=20.1$, $p<0.001$.

Table 5

Mixed Effects Model Results of Speech-Language Growth Across Digit Span Clusters

Digits Forward	**PBK**	**HINT-C**	**PPVT-III**	**CELF**
Cluster 1 (LL)	5.22 (1.14) [16]	3.86 (1.93) [7]	8.58 (0.68) [21]	-0.55 (1.13) [8]
Cluster 2 (MH)	5.90 (0.85) [27]	3.15 (0.97) [24]	11.30 (0.56) [32]	3.01 (0.50) [23]
Cluster 3 (HH)	2.65 (1.42) [9]	2.11 (1.48) [9]	11.29 (0.85) [12]	2.71 (0.69) [8]
Age × Cluster F (df)	1.95 (2,155)	0.30 (2,87)	5.54 (2,213)	4.23 (2,87)
p-value for Age × Cluster	0.15	0.75	0.005**	0.02*
Digits Backward				
Cluster 1 (LL)	4.20 (1.39) [8]	3.25 (1.83) [8]	8.16 (0.73) [15]	0.99 (1.10) [8]
Cluster 2 (ML)	6.29 (0.78) [38]	2.96 (0.99) [26]	10.93 (0.52) [42]	2.41 (0.52) [25]
Cluster 3 (HH)	1.51 (1.61) [6]	2.82 (1.62) [6]	12.29 (0.97) [8]	3.22 (0.75) [6]
Age × Cluster F (df)	3.94 (2,155)	0.02 (2,87)	7.20 (2,213)	1.42 (2,87)
p-value for Age × Cluster	0.03*	0.99	0.0009***	0.25

Note: Values are slopes for lines of language scores across chronological age from mixed effects repeated measures of language scores, controlling for age at implantation, best pre-implantation pure tone average, communication mode, and maternal education. Values in parentheses are standard errors; values in brackets are N. DF=Digits Forward; DB=Digits Backward; PBK=Phonetically Balanced Kindergarten test; HINT-C=Hearing In Noise Test for Children; PPVT-III=Peabody Picture Vocabulary Test, Third Edition; CELF=Clinical Evaluation of Language Fundamentals.

* $p<0.05$

** $p<0.01$

*** $p<0.001$.