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Developmental trajectories of forward and backward digit spans in deaf children with cochlear implants

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Abstract

Background—Accounting for outcome variability among pediatric cochlear implant (CI) recipients is an enduring clinical challenge. Short-term memory and working memory (STM/WM), as indexed by digit span forward (DSF) and digit span backward (DSB), have been shown to be strongly correlated with speech and language (S/L) outcomes. The enormous variability observed in conventional outcome measures of S/L may reflect individual differences in STM/WM.

Methods—Repeated measure auditory digit spans were obtained from 110 children (age 3–15 years; mean 7.2 years) with at least 2 years of CI use. Growth curves were computed for each child, and linear functions were fit to both DSF and DSB. Slopes and intercepts were used as parameters in mixed-models to assess relations between STM/WM capacity change over time and S/L outcome measures including vocabulary (PPVT), open-set spoken word recognition (PBK), and sentence perception (HINT-C).

Results—For DSF, the percent of the sample more than 1 SD below the norm at each age ranged from 54 to 90% (mean = 66.5%). For DSB, the percent of the sample more than 1 SD below the norm at each age ranged from 23 to 42% (mean = 34.5%) at ages where there were at least five children. Four subgroups within our CI sample emerged: (Subgroup 1) children demonstrating age-appropriate growth in both DSF and DSB scores over time (49/110, 44.55%); (Subgroup 2) children demonstrating age appropriate growth in DSF over time but below average growth in DSB over time (23/110, 20.91%); (Subgroup 3) children demonstrating below average growth in DSF over time but age-appropriate growth in DSB over time (19/110, 17.27%); and (Subgroup 4) children demonstrating below average growth in both DSF and DSB over time (19/110, 17.27%). For all tests except CELF-3, Subgroup 4 demonstrated the poorest performance among the four DS slope subgroups. Significant differences were observed between Subgroup 1 and Subgroup 4 on last visit PBK-Word ($P = 0.029$), PPVT ($P = 0.018$), and HINT-C in Quiet ($P = 0.001$), but not CELF-3 ($P = 0.433$).

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Conclusion—The findings from this longitudinal study suggest that differences in the rate of development of STM/WM may influence S/L outcomes in children with CIs. The clinical implications of these findings are significant because they indicate that the rate of development of STM/WM, and not just the actual level of STM/WM at a single time point, predicts later S/L development in this clinical population. Targeted interventions to improve developmental rate of verbal STM/WM may hold promise for enhancing S/L skills in children with CIs.

Introduction

Why do some children with cochlear implants (CIs) demonstrate suboptimal speech and language (S/L) outcomes despite the presence of an apparently ideal set of known, established, conventional demographic, and medical indicators? The factors responsible for the large variability and individual differences in S/L performance following cochlear implantation remain a critical barrier to further progress in the field (NIH Consensus Statement, 1995).

Recent evidence suggests that individual differences following implantation are not anomalous, mysterious, or idiopathic, but represent systematic underlying differences in core elementary neurocognitive processes that influence performance on a wide range of traditional S/L outcome measures (Fagan *et al.*, 2007; Conway and Pisoni, 2008). Short-term memory (STM) and working memory (WM) processes, in particular, are critical for speech perception and language processing because they serve as temporary holding areas for incoming and outgoing verbal information, as well as storage spaces for linguistic information during immediate processing (Baddeley, 2007). Because development of verbal STM/WM is dependent, in part, on auditory perceptual experiences, children who experience a period of auditory sensory deprivation and/or degraded auditory input early in life are at high risk for disturbances and delays in verbal STM/WM processes (Bavelier *et al.*, 2008).

Identifying differences in STM/WM may provide better explanations of variability and individual differences in children who experience heretofore unexplained suboptimal S/L outcomes following cochlear implantation. Ultimately, improved prediction of who will do well with a CI and who is likely to struggle to obtain optimal benefit may lead to development and earlier initiation of novel targeted intervention strategies.

We conducted a prospective longitudinal cohort study of the development of verbal STM/WM using digit span forward (DSF) and digit span backward (DSB) raw scores in a large sample of deaf children with CIs. With repeated measures over time from each child, we used developmental growth curves to study the time course of learning and growth in STM/WM capacity. Using four different conventional S/L outcome measures, we sought to address how change in STM/WM over time is associated with performance on conventional clinical measures of S/L. The study was approved by our university Institutional Review Board.

Methods

Study participants

The sample consisted of 110 deaf children who received CIs at our medical center between the years of 1997 and 2007. Eligibility criteria included: (1) severe-to-profound hearing loss bilaterally, (2) a monolingual English home environment, and (3) use of a currently available, state-of-the-art CI system. Table 1 shows the study participants' characteristics.

Measures

Measures of STM and WM—The Wechsler Intelligence Scale for Children, 3rd edition (WISC-III) Digit Span (DS) subtest requires the child to reproduce a progressively longer list of digits spoken live-voice by a test administrator at a rate of approximately one digit per second with the administrator's face visible to the child. The task consists of two recall conditions that tap STM/WM, respectively: DSF, which requires the child to repeat the list of digits in the serial order originally presented (passive storage and retrieval); and DSB, which requires the child to repeat the list of digits in backward order from the original presentation (storage, active processing, and retrieval) (St Claire-Thompson, 2010).

Speech and language outcome measures—Four conventional clinical S/L outcome measures were obtained for each child: (1) the phonetically balanced kindergarten (PBK) word test as a measure of open-set spoken word recognition, (2) the peabody picture vocabulary test (PPVT) as a measure of receptive vocabulary, (3) the hearing-in-noise test for children (HINT-C) as a measure of sentence perception, and (4) the clinical evaluation of language fundamentals-3 (CELF-3) as a measure of receptive and expressive language. Only the final test session's performance was considered in the present set of analyses.

Statistical analysis

Normative comparisons—To compare our subjects' performance on DSF and DSB to a benchmark normative growth curve, we used normative means obtained from the Children's Memory Scale (CMS) as a proxy for the WISC-III because the WISC-III does not report normative data separately for DSF and DSB raw scores and the CMS numbers subtest is identical in length and instructions. Scores based on samples of normal-hearing, typically developing children were obtained for the PPVT and CELF-3 from published manuals. No norms exist for the PBK test or the HINT-C.

Modeling developmental growth—Growth curves of DSF and DSB raw score across chronological ages were modeled using mixed-effects repeated measures from multiple assessments, specifying random slopes and intercepts to account for individual variability in the within-person development of DSF and DSB over time.

Results

Developmental growth

Fig. 1 shows individual DSF and DSB scores of all 110 subjects over time fit with a linear growth model. In order to evaluate development of DSF and DSB in our CI sample relative to norms, we compared the CI sample's slope and intercepts to normative values obtained from the CMS. Estimated norm slopes for DSF and DSB were 0.36 and 0.43, respectively. For DSF, the percent of the sample more than 1 SD below the norm at each age ranged from 54 to 90% (mean = 66.5%). For DSB, the percent of the sample more than 1 SD below the norm at each age ranged from 23 to 42% (mean = 34.5%) at ages where there were at least five children.

Digit span profile analysis

Fig. 2 shows a scatterplot for DSF and DSB slopes across all 110 study participants. Applying norm scores from the CMS, four subgroups within our CI sample emerged: (Subgroup 1) children demonstrating age-appropriate growth in both DSF and DSB scores over time (i.e., slope equal to or greater than the slope of the CMS normative sample) (49/110, 44.55%); (Subgroup 2) children demonstrating age-appropriate growth in DSF over time but below average growth (i.e., slope less than that of the CMS norm sample) in DSB

over time (23/110, 20.91%); (Subgroup 3) children demonstrating below average growth in DSF over time but age-appropriate growth in DSB over time (19/110, 17.27%); and (Subgroup 4) children demonstrating below average growth in both DSF and DSB over time (19/110, 17.27%).

An analysis of the final test session's S/L outcomes was performed across the DS profile subgroups. For all tests except CELF-3, Subgroup 4 (–) demonstrated the poorest performance among the four DS slope subgroups. Analysis of variance demonstrated significant differences between Subgroup 1 (++) and Subgroup 4 (–) on last visit PBK-Word ($P = 0.029$), PPVT ($P = 0.018$), and HINT-C in Quiet ($P = 0.001$), but not CELF-3 ($P = 0.433$). Panels A–D of Fig. 3 show performance on the S/L outcomes across the four DS subgroups. Interestingly, Subgroup 3 (–+) consistently performed at or above the mean level of performance demonstrated by Subgroup 1 (++) —children who had DS slopes that were most like the normal hearing, typically developing normative benchmark.

Discussion

Accumulating evidence suggests that individual differences in S/L outcome measures are not anomalous, mysterious, or idiopathic, but rather represent sources of variability in core elementary neurocognitive processes that underlie all conventional S/L outcome measures (Fagan *et al.*, 2007). The current study found that development of STM/WM as indexed by DSF/DSB is delayed or disturbed in deaf children using CIs, and those children demonstrating disturbances in both verbal STM and verbal WM (Subgroup 4) have the poorest performance on S/L outcome measures.

Interestingly, Subgroups 2 (+–) and 3 (–+) – children who demonstrated below average growth in one area of DS (DSB or DSF, respectively) but average or better improvement in the other area of DS — consistently performed at the same level as Subgroup 1 (++) on the S/L measures. We suspect that this is a result of the different distributions of DSF and DSB in the sample: The subgroups with below average DSB slopes (2 and 4) show a similar range of low DSB slope values (Fig. 2) ranging from nearly average to well below average. However, for the subgroups with below average DSF slopes (3 and 4), the range of DSF slope scores differs markedly between subgroups. Specifically, DSF slopes for Subgroup 3 are close to the normative DSF slope mean (as shown by the green triangles falling very close to the vertical line in Fig. 2), whereas most DSF slopes for Subgroup 4 are well below the normative mean (as shown by the blue squares falling more to the left of the vertical line in Fig. 2). Thus, while both Subgroups 3 and 4 have DSF slopes that are below average, the actual DSF slope values for Subgroup 4 are clearly lower than those for Subgroup 3. This suggests that developmental delays in DSF growth (which are clearly most profound in Subgroup 4) are driving differences found between Subgroup 4 and the other three subgroups.

Conclusions

The findings from this longitudinal study suggest that differences in the rate of development of STM/WM may influence S/L outcomes in children with CIs. The clinical implications of these findings are significant because they indicate that the rate of development of STM/WM, and not just the actual level of STM/WM at a single time point, predicts later S/L development in this clinical population. Targeted interventions to improve developmental rate of verbal STM/WM may hold promise for enhancing S/L skills in children with CIs.

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Figure 1.

Red lines are from linear regression of each child's digit span scores over time; black lines represent the average growth curve of the sample. Panel A shows digit recall forward across chronological age. Panel B shows digit recall backward across chronological age. Means and standard deviations of norm data from a mixed effects model specifying random slopes and intercepts (from CMS) are shown at each age by black dots and bars.

Figure 2.

Scatterplot showing digit span slopes over time for all subjects. Black cross-hatch lines represent the normative slopes over time for normal hearing peers for digits forward (vertical line) and digits backward (horizontal line) obtained from the CMS. The right upper quadrant shows Subgroup 1 (++), children who demonstrated age-appropriate slopes for both DSF and DSB. The right lower quadrant shows Subgroup 2 (+-), children who demonstrated age-appropriate DSF slope, but delayed DSB slope. The left upper quadrant shows Subgroup 3 (-+), children who demonstrated delayed DSF slope but age-appropriate DSB. The lower left quadrant shows Subgroup 4 (--), children who demonstrated both delayed DSF slope and DSB slope.

Figure 3.

Mean scores with 95% confidence interval bars for the final test session's performance on the four S/L outcome measures (A: PBK-Word; B: CELF; C: PPVT; D: HINT-C in Quiet) across the four digit span slope subgroups. rs = raw score; ss = standard score.

Table 1Characteristics of study participants, $n = 110$

Variable	Mean (SD; range)
Age at CI implant (years)	3.98 (2.16; 1.06–13.14)
Chronological age at first visit (years)	7.17 (2.22; 3.26–15.42)
Duration of implant use at first visit (years)	3.19 (1.79; 0.01–9.05)
Time followed (years)	3.32 (1.88; 0.43–8.91)
Number of test sessions	4.5 (1.6; 2–9)
Best pre-operative PTA *	104.47 (13.51; 58.33–120.07)
Mother's education †	4.75 (1.92; 1–9)
	n (%)
Male	58 (52.7%)
Race/ethnicity ‡	
White	104 (94.6%)
African American	7 (6.4%)
Asian	2 (1.8%)
Hispanic	3 (2.7%)
Etiology of hearing loss	
Unknown	72 (65.4%)
Meningitis	11 (10%)
CMV §	4 (3.6%)
Genetic aberration	13 (11.8%)
Anatomic anomalies ¶	7 (6.4%)
Ototoxicity	1 (.9%)
Auditory neuropathy	2 (1.8%)
Communication mode first visit **	
Aural-oral (AO)	74 (67.2%)
Total communication (TC)	36 (32.7%)
Device ††	
Unilateral	99 (84%)
Bilateral	4 (3%)
CI + HA ‡‡	15 (13%)

* PTA is the pure tone average (500, 1000, and 2000 Hz).

† Education of mother or female caregiver; education of father in one case; education is an ordinal scale of 1–9, based on years of education ranging from 'some high school' to 'doctorate'.

‡ More than one race/ethnicity was recorded for some children.

§ Cytomegalovirus.

¶ For example, Enlarged Vestibular Aqueduct, Mondini Malformation.

** Three children switched from TC to AO over the course of testing.

^{††} Sum is greater than 110 because some children switched from unilateral to bilateral over the course of testing.

^{‡‡} Concurrently used a CI in one ear and a hearing aid in the contralateral ear.