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Language Outcomes Improved Through Early Hearing Detection and Earlier Cochlear Implantation

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Hypothesis: Early identification and intervention, earlier cochlear implantation, and mother's level of education will directly and/or indirectly impact the language outcomes of children with cochlear implants (CIs).
Background: Identifying factors that contribute to the wide range of language outcomes in children who use CIs will assist healthcare and rehabilitation professionals in optimizing service delivery for this population. Universal newborn hearing screening provides an opportunity to examine the relationship between meeting the early hearing detection and intervention (EHDI) 1-3-6 guidelines and child language outcomes. These guidelines recommend screening by 1 month, confirmation of hearing loss by 3 months, and intervention by 6 months of age.
Methods: Participants were 125 children with CIs ranging from 13 to 39 months of age. Language ability was measured using the Child Development Inventory and MacArthur-Bates Communicative Development Inventories.
Results: Meeting EHDI 1-3-6, higher levels of maternal education and earlier cochlear implant activation had a direct, positive impact on language outcomes. Meeting the EHDI 1-3-6 guidelines also had an indirect positive effect on language outcomes via increasing the probability that the children's CIs would be activated earlier. Maternal education did not significantly predict age of cochlear implant activation nor whether a child met EHDI 1-3-6.
Conclusion: Ensuring families meet the EHDI 1-3-6 guidelines is an early step that can lead to higher language outcomes and also earlier cochlear implantation. **Key Words:** Cochlear implant—Early identification—Early intervention—Early hearing detection and intervention—Hearing loss—Language outcomes.

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Positive outcomes in children who use cochlear implants (CIs) have been found in a variety of domains (1,2) including, reading (3), speech (4,5), word-learning skills (6), and speech perception (7). However, not all children with CIs reach these high levels of performance. Cochlear implantation alone does not ensure that children will develop age-appropriate skills. To maximize the likelihood that children who receive CIs will achieve language skills on par with their hearing peers, it is critical to identify factors associated with higher language outcomes.

A variety of child and family characteristics have been found to contribute to the variability in outcomes of children with CIs, and a complex relationship between these variables has been described (8). Higher family income has been associated with better language performance before cochlear implantation as well as accelerated growth in language comprehension after cochlear implantation (9). Maternal responsiveness in early communication interactions has been associated with increased development in spoken language (9).

The United States Food and Drug Administration guidelines approve surgical placement of CIs in children starting at 12 months of age. In an effort to determine the importance of proceeding with cochlear implantation at this young age, a number of studies have looked at the impact of early cochlear implantation on speech and language outcomes. Numerous investigators have reported that earlier implantation is associated with better outcomes on spoken language measures and, in some cases, age-appropriate language scores (2,10–16). The rationale for earlier implantation is based on neural flexibility (17,18) and sensitive periods of learning (19–21).

The consistent support for earlier implantation can inform surgical guidelines and family decisions.

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However, there is an even earlier entry point into the intervention system for children with hearing loss. The early hearing detection and intervention (EHDI) guidelines state that children should be screened by 1 month of age, have a full hearing assessment by 3 months of age and be enrolled in early intervention by 6 months of age. This is commonly referred to as the EHDI 1-3-6 guidelines. Tomblin et al. (22) suggest that compliance with EHDI allows for several preliminary activities that are necessary for cochlear implantation to occur in the shortest time possible.

Considering the contribution of meeting the EHDI 1-3-6 guidelines to both age of implantation and later language ability may give practitioners an even earlier opportunity to positively impact the outcomes of children with CIs. This variable has not yet been considered in the CI literature. The purpose of the present article is to investigate the direct and indirect influence of meeting EHDI 1-3-6, age of CI activation, and level of maternal education on language outcomes of children with CIs.

MATERIALS AND METHODS

Participants

The participants were 125 children from 13 to 39 months of age (mean, 28.3; SD [standard deviation], 5.9) who used CIs. All met the following inclusion criteria: 1) congenital, bilateral hearing loss, 2) no apparent additional disabilities that would interfere with language development based on parent and interventionist report, and 3) use of one or two CIs for at least 3 months. See Table 1 for participant characteristics.

The children in this investigation were enrolled in the National Early Childhood Assessment Project (NECAP), a multi-state study of early language outcomes of children with hearing loss supported by the Centers for Disease Control. This study was approved by the University of Colorado-Boulder Institutional Review Board. All NECAP participants who met the above selection criteria were included in the present study. The children lived in the following 12 states: Arizona, California, Florida, Idaho, Indiana, Maine, North Dakota, Oregon, Texas, Utah, Wisconsin, and Wyoming.

At the time of testing, 62 children used a CI in one ear and 63 wore bilateral CIs. Age of CI activation ranged from 5 to 34 months of age (mean, 16.1; SD, 4.9) with the implants of 63% of the participants activated at or before 15 months of age. On average, participants had worn their device for 12.7 months (range, 3–27 mo) before participation in the study. For additional information about CI use, see Table 2.

Age of identification of the children's hearing loss ranged from 1 week to 18 months (mean, 2.8 mo) with age of intervention ranging from 1 week to 23 months (mean, 5.0 mo). The EHDI guidelines of hearing screening by 1 month, confirmation of hearing loss by 3 months, and enrollment in early intervention by 6 months of age were met by 71% of the participants. The mothers' highest level of education ranged from less than a high school degree to a graduate degree, with 56% having obtained a degree higher than a high school diploma.

Measures

The participants' language was measured using the Child Development Inventory (23) and the MacArthur Communicative Development Inventories (24). A demographic form was completed by the family with assistance from their interventionist. Families signed a release of information, and audiologic/CI data were obtained directly from the participants' medical records.

TABLE 1. *Participant and family characteristics*

Characteristic	Percentage of Participants
Gender	
Male	56
Female	44
Ethnicity	
Non-Hispanic	69
Hispanic	31
Race	
White	91
African American/black	2
Asian	5
Native American	2
Communication mode used with the child	
Primarily spoken language	70
Spoken language only	25
Spoken with very occasional use of sign	45
Sign language + spoken language	30
Hearing status of the parent	
Both parents hearing	93.5
One or both parents deaf/hard of hearing[a]	6.5
Mother's highest educational degree	
High school diploma or less	44
Less than high school	13
High school	31
Beyond high school diploma	56
Vocational	7
Associates	14
Bachelor's	24
Graduate	10

[a]Of the parents who were deaf or hard of hearing, approximately one-third used sign language when communicating with their child.

TABLE 2. *Audiologic characteristics of the participants*

Characteristic	Percentage of Participants
Type of amplification	
Bilateral cochlear implants	50%
Unilateral cochlear implant	36%
Cochlear implant + hearing aid	13%
Cochlear implant + FM system	1%
Age at cochlear implant activation	
By 15 months	63%
After 15 months	37%
Average cochlear implant use per day	
Less than 3 hours	2%
3 to 5 hours	5%
6 to 10 hours	38%
11 or more hours	55%
Meets EHDI 1-3-6 guidelines	
Meets EHDI guidelines	71%
Does not meet EHDI guidelines	29%

EHDI indicates early hearing detection and intervention.

The first language measure, the Child Development Inventory (CDI), is a general development inventory assessing a young child's skills in a variety of domains. This study analyzed the Expressive Language and Language Comprehension subscales. The Expressive Language subscale examines expressive communication skills from pre-verbal behavior to complex grammar. The Language Comprehension subscale was named the Comprehension-Conceptual Language scale in a previous version of the CDI (25). Comprehension-Conceptual will be used throughout this article as it provides a better description of this subscale which assesses comprehension, production, and conceptual language skills. This instrument has been found to have strong reliability, concurrent validity, and a high level of sensitivity in identifying children with documented language delay (25,26). For this assessment, children received credit if a test item was understood/produced in spoken language or sign language.

The second language measure, the MacArthur-Bates Communicative Development Inventories (Mac-CDI) (24) evaluated the children's expressive vocabulary skills. Children were assessed with one of two inventories—the Mac-CDI: Words and Gestures or the Mac-CDI: Words and Sentences. In accordance with the instructions in the test manual when assessing children with potential language delay, the version of the inventory selected was based on an estimate of the children's vocabulary size rather than their chronological age. For this study, the Vocabulary Production subscale was considered because this is the one subscale that is common across the two inventories. This subscale presents a wide range of words organized in different semantic categories. Parents indicate words that their children produce spontaneously in either spoken language or sign language. The Mac-CDI is a widely-used assessment of early language skills, with numerous studies supporting its validity with children who are typically developing (27,28), those with various disabilities (29,30), and children with hearing loss (31,32).

Procedures

In all but one state, the assessment instruments were delivered to the family by their early intervention provider. The interventionist instructed the family on how to complete the forms and was available for questions as needed. At a subsequent intervention session, the early intervention provider collected the forms and reviewed them for completeness and accuracy. In the remaining state, instructions were provided to the family via telephone, and the assessment materials were distributed and collected via mail. Follow-up questions were asked by phone as needed.

For the two language assessments, age equivalent scores were obtained using the normative data of the respective instrument. Due to the varying chronological ages of the children at the time of testing, language quotient (LQ) scores were derived to make comparisons across the participants. Quotients were calculated by dividing the child's language age score by their chronological age and then multiplying by 100. Thus, a score of 100 indicated that the child's language age was equal to his/her chronological age. The test manual for the CDI states that LQs below 80 indicate significant language delay relative to age expectations. For the Mac-CDI, an LQ of 75 approximates the 10^{th} percentile; thus scores below this level indicate language skills are outside the normal range.

Analyses focused on the role and relationship of EHDI 1-3-6 goals and age of activation as predictors of language ability. Maternal education was included due to its well-documented relationship with child language development.

RESULTS

Primary statistical analyses used structural equation modeling (SEM). SEM is a flexible and powerful multivariate statistical approach that examines the complex causal relationships between multiple predictors and outcomes. It is a valuable tool for assessing mediational effects where one predictor has an effect on a second intermediate predictor (the mediator), which has an effect on the outcome of interest. For example, in this study, it allows us to examine how meeting the EHDI 1-3-6 goals may lead to earlier CI activation, which may lead to improved language outcomes. In addition, SEM provides fit indices, or statistical measures of how well the observed data actually fit the model being tested.

For this study, a common model was applied to each outcome: CDI Expressive Language, CDI Comprehension-Conceptual Language, and Mac-CDI Expressive Vocabulary. As illustrated in Figure 1, the model included paths from maternal education to EHDI 1-3-6 and language. Paths were also included from EHDI 1-3-6 to age of activation and language, and finally a path was included from age of activation to language. Maternal education was unrelated to age of activation, and so this path was excluded, allowing for the estimation of fit indices. Because EHDI 1-3-6 was dichotomous, the path coefficient between EHDI 1-3-6 and maternal education is a probit coefficient—not a traditional regression coefficient. For clarity, that coefficient is italicized in the figures. Paths leading to the language outcomes are unstandardized paths predicting the corresponding language quotient scores. One participant was missing data for the 1-3-6 variable. This small amount of missing data was addressed through the use of full information maximum likelihood estimation. All SEM analyses were conducted using M-Plus version 8 (Muthén & Muthén, Los Angeles, CA).

Expressive Language

Of the 125 participants, 94 completed the CDI. Two outliers were identified using the ''outlier labelling rule'' with a multiplier of 2.2 as recommended by Hoaglin and Iglewicz (33). Thus, the SEM analyses predicting CDI Expressive Language quotients (ELQs) were based on a total of 92 children. The mean ELQ for these children was 76.3 (SD = 18.9). Results for the ELQ SEM model are presented in Figure 1.

Fit indices suggested the model fit the data well. The χ^2 test was not significant (χ^2 (1, n = 92) = 0.59, $p = 0.442$), with a confirmatory fit index (CFI) = 1.000, and a root mean square error of approximation (RMSEA) < 0.001 (90% CI = 0.000, 0.251). The path from maternal education to age of CI activation was not significant and was excluded from the model so that fit indices could be estimated. Based on this model, the direct effect for maternal education was 1.870 ELQ points per year of education (standard error [SE] = 0.631, $p = 0.003$).

The total effect of meeting EHDI 1-3-6 was 11.395 ELQ points (SE = 1.837, $p < 0.001$), a relatively large

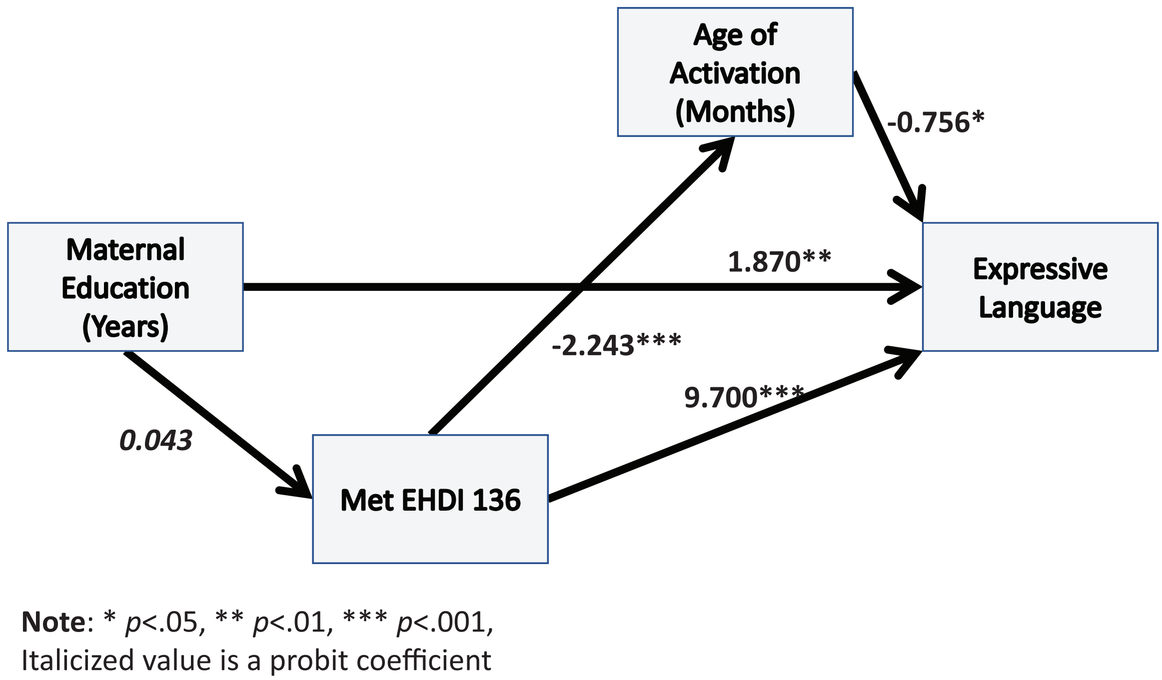

FIG. 1. SEM model predicting expressive language quotients. SEM indicates structural equation modeling.

and meaningful effect on language development. The direct effect of meeting EHDI 1-3-6 was equal to a gain of 9.700 points (SE = 1.998, $p < 0.001$), which is its unique effect after controlling for maternal education and is also unrelated to age of activation. The indirect effect of EHDI 1-3-6 through age of activation was equal to a gain of 1.695 points (SE = 0.687, $p = 0.014$), which is the effect EHDI 1-3-6 has on Expressive Language by leading to earlier cochlear implant activation. In contrast, the direct effect of age of activation was a reduction of 0.756 ELQ points per month (SE = 0.314, $p = 0.016$).

For context, the effect of EHDI 1-3-6 is equivalent to a 15.1 month difference in age of activation. In other words, the effect of meeting EHDI 1-3-6 is equal to the difference one would expect between two otherwise equivalent children, where one received CI activation 15.1 months earlier than the other. The EHDI 1-3-6 effect is similarly equal to 6.1 years of additional maternal education.

Comprehension-Conceptual Language

Of the 125 participants, 94 completed the CDI and so SEM analyses predicting CDI Comprehension-Conceptual Language quotients (CCQs) were based on these 94 children. The mean CCQ was 70.4 (SD = 17.7). Results for the CCQ SEM model are presented in Figure 2.

As with Expressive Language, fit indices suggested the model fit the data well. The χ^2 test was not significant (χ^2 (1, n = 94) = 0.002, $p = 0.962$), with a CFI = 1.000, and a RMSEA < 0.001 (90% CI = 0.000, 0.001). Based on this model, the direct effect for Maternal Education was 2.459 (SE = 0.558, $p < 0.001$).

The total effect of EHDI 1-3-6 was 7.413 (SE = 2.205, $p = 0.001$). Both the direct effect of EHDI 1-3-6 (4.820, SE = 2.377, $p = 0.043$) and its indirect effect through age of activation (2.593, SE = 0.828, $p = 0.002$) were statistically significant. The unique effect of age of activation on language scores that is unrelated to EHDI 1-3-6 or maternal education was −1.191 (SE = 0.315, $p < 0.001$). For context, the EHDI 1-3-6 effect suggests that the effect of meeting the EHDI goals on comprehension-conceptual language is equivalent to a 6.2 month difference in age of activation or 3.0 years of maternal education.

Expressive Vocabulary

Of the 125 participants, 120 completed the Mac-CDI and so SEM analyses predicting Mac-CDI Expressive Vocabulary quotients (EVQs) were based on these 120 children. The mean EVQ for these children was 66.7 (SD = 16.7). Results for the EVQ SEM model are presented in Figure 3.

Fit indices again suggested the model fit the data well. The χ^2 test was not significant (χ^2 (1, n = 120) = 0.254, $p = 0.614$), with a CFI = 1.000, and a RMSEA < 0.001 (90% CI = 0.000, 0.193). Based on this model, the direct effect for Maternal Education was 1.151 (SE = 0.542, $p < 0.034$).

The total effect of EHDI 1-3-6 was 7.853 (SE = 1.356, $p < 0.001$), with a direct effect of 6.122 (SE = 1.426, $p < 0.001$) and indirect effect through age of activation of 1.731 (SE = 0.675, $p = 0.010$), both of which were statistically significant. A significant indirect effect of EHDI 1-3-6 leading to earlier activation, which leads

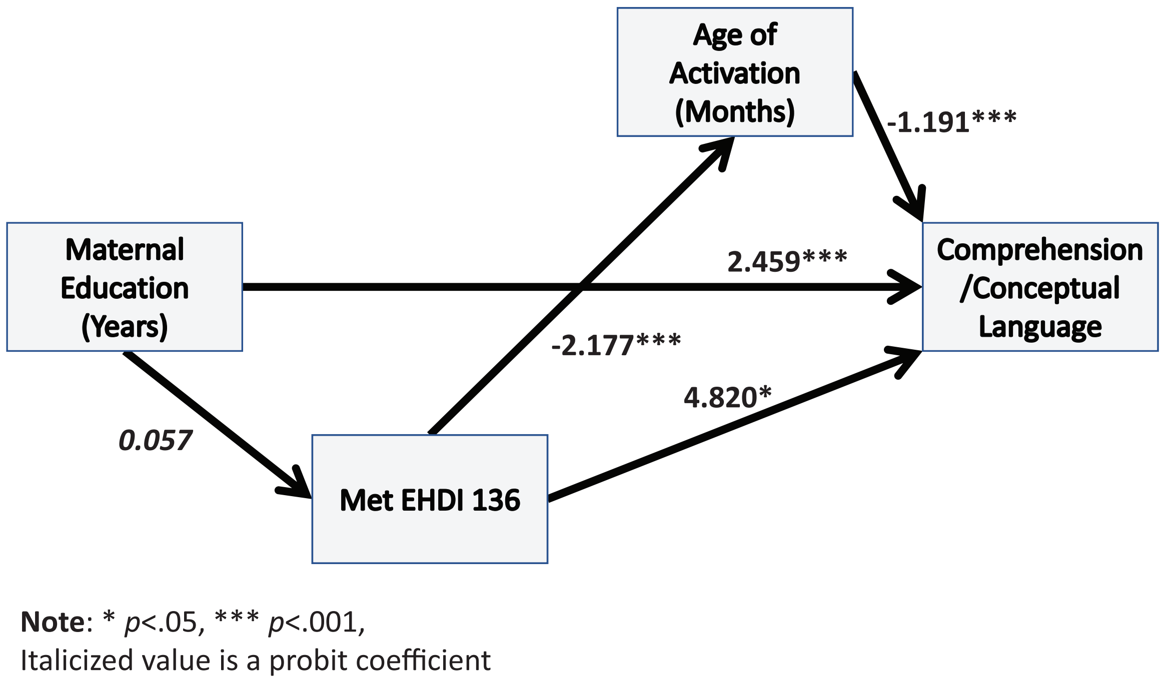

FIG. 2. SEM model predicting comprehension-conceptual language quotients. SEM indicates structural equation modeling.

to higher language outcomes was therefore consistent across all three language measures. Finally, the unique effect of age of activation after controlling for maternal education and EHDI 1-3-6 was −0.900 (SE = 0.294, $p = 0.002$). For context, this suggests that the effect of meeting EHDI 1-3-6 on expressive vocabulary is equivalent to an 8.7 month difference in age of activation or 6.8 years of maternal education.

DISCUSSION

This article investigated the interactive role of three predictor variables (meeting EHDI 1-3-6 guidelines, age

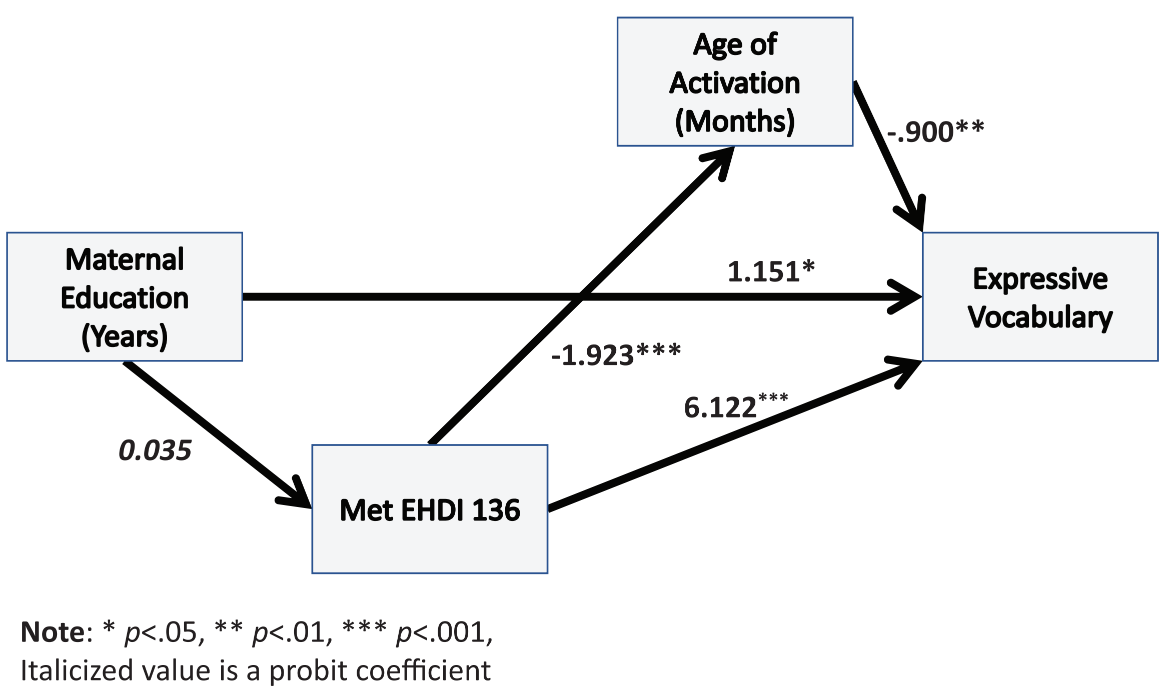

FIG. 3. SEM model predicting MacArthur vocabulary quotients. SEM indicates structural equation modeling.

of implant activation, and maternal level of education) in terms of their effect on the scores of young children with CIs on three different language measures. Meeting EHDI 1-3-6, maternal level of education, and CI activation age had direct positive effects on all three language outcomes. Meeting EHDI 1-3-6 had both a direct effect and a significant indirect effect via age of activation on all three language measures.

The positive impact of maternal level of education on language outcomes is consistent with a number of previous studies. Higher levels of maternal education and a closely-related variable, socio-economic status (SES), have been associated with a language advantage for typically developing children (34–36) as well as children who use CIs (9,37). Although this characteristic cannot be directly changed by healthcare providers and interventionists, awareness of a family's level of education may inform the quantity and nature of the intervention a family receives. For example, research on providing parents from lower educational and SES backgrounds with information and feedback about their use of language with their child has been found to improve parent knowledge of language development. Additionally, it has been shown to increase the number of adult words and conversational turns occurring in the home (38).

Our finding that age of activation has a direct impact on language outcomes is in keeping with existing literature. Multiple studies using different assessment tools have found that earlier placement of CIs improves language outcomes (1,9,12,22,39–43) including vocabulary (44,45). Most studies, including the current investigation, report on short-term benefits of earlier cochlear implantation. While there is variability in the reports of the impact on long-term outcomes, some researchers have documented language benefits after 10 years of device use (14,46). These long-term benefits highlight the importance of expediting a child through the EHDI system so that implantation can be achieved as early as is appropriate.

An encouraging finding from this study was that neither meeting EHDI 1-3-6 nor age of CI activation were significantly impacted by the mother's level of education. This indicates that our national EHDI systems are providing equal opportunities for families with lower and higher levels of education in terms of access to early identification, intervention, and cochlear implantation. Thus, although maternal level of education has a direct impact on language outcomes, the benefits of early identification and intervention and earlier cochlear implantation appear to be equally available to families across a wide range of educational backgrounds.

Previous studies have examined the relationship of a single component of EHDI to the timing of cochlear implantation. For example, Dettman et al. (47) found that an earlier age of hearing aid fitting led to earlier CI surgery. Niparko et al. (9) noted that children implanted earlier typically were diagnosed with hearing loss earlier. The current investigation is consistent with these findings, however, it is the first to examine the impact of meeting all three components of the EHDI guidelines on both age of CI activation and language outcomes. In addition to EHDI making a positive indirect impact on all three language measures via earlier cochlear implantation, there was also a significant direct effect of EHDI on all three language measures. This indicates that EHDI is having additional positive impacts on language outcomes unrelated to early CI activation. The higher language outcomes associated with meeting EHDI guidelines are likely due to a variety of benefits that are possible as a result of prompt identification of hearing loss and early access to intervention. This includes timely emotional support to families which can lead to reductions in parental stress and improved parent–child bonding. In addition, early education on communication strategies assists parents in providing an optimal language learning environment for their child.

Taken as a group, both previous research and the current study point to the importance of increasing the number of children who meet the EHDI 1-3-6 guidelines as this is likely to lead to earlier implantation and to better language outcomes (48). The results from this study found that the effect of meeting the EHDI 1-3-6 guidelines, both directly on language and indirectly through earlier CI activation, had a large effect—even relative to the well-documented effects of maternal education and age of CI activation. Typically, studies on children with CIs do not include any information about meeting 1-3-6, but this study raises the possibility that meeting 1-3-6 could offset the negative impact of lower maternal level of education. The positive impact of meeting 1-3-6 could be greatest for those families who have the greatest educational and social risks. With a larger sample size, we hope to be able to investigate this more thoroughly. Otolaryngologists, pediatricians, and other healthcare professionals can play a role in increasing compliance to the EHDI guidelines by impressing upon families whose children have referred on their newborn hearing screen, the importance of prompt hearing assessment and timely enrollment into early intervention.

Similar to the findings of Niparko et al. (9), on average, the language abilities of the children in this study were considerably below those of the hearing children on whom the language tests were normed. Specifically, the mean language quotients were ELQ = 76.3, CCQ = 70.4, and EVQ = 66.7. In contrast, children who met EHDI 1-3-6, had their CIs activated at 11 months of age, and had mothers with a college degree had the following predicted mean scores: ELQ = 87.4, CCQ = 82.5, EVQ = 76.7. Although these scores are 10 to 12 quotient points better than the overall group averages, they are still not at the target quotient of 100. Fortunately, there is some evidence that over time, with continued intervention and CI use, language quotients increase. For example, Yoshinaga-Itano et al. (49), found that from 4 to 7 years of age, children with CIs made greater than 1 year of language gain within a single year.

One of the positive features of the current study is the generalizability of the results due to the diversity of the participating families. In contrast to many previous studies on language and CIs, the following groups were well represented: Hispanic ethnicity, parents who are deaf/hard of hearing, parents with lower levels of education, and children who were predominantly receiving services through publicly-funded local educational agencies or state-wide systems.

Limitations of the current study include the use of a restricted range of independent variables to predict language outcomes. Although this was unavoidable due to power limitations and the nature of the data collected, it would be valuable to learn more about the contribution to children's language outcomes of important variables such as amount of daily implant use and quantity and quality of intervention services. In addition, longitudinal studies examining language growth over time after more extended implant use would allow us to determine characteristics of children who are able to maintain typical language growth trajectories or demonstrate accelerated growth curves that allow them to close early language gaps.

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