Cochlear Implants Int. **8**(2), 92–116, 2007
Published online in Wiley InterScience
(www.interscience.wiley.com) **DOI**: 10.1002/cii.333

Childhood Development after Cochlear Implantation (CDaCI) study: Design and baseline characteristics

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ABSTRACT Children with severe to profound sensorineural hearing loss face communication challenges that influence language, psychosocial and scholastic performance. Clinical studies over the past 20 years have supported wider application of cochlear implants in children. The Childhood Development after Cochlear Implantation (CDaCI) study is the first longitudinal multicentre, national cohort study to evaluate systematically early cochlear implant (CI) outcomes in children. The objective of the study was to compare children who have undergone cochlear implantation, with similarly aged hearing peers across multiple domains, including oral language development, auditory performance, psychosocial and behavioural functioning, and quality of life. The CDaCI study is a multicentre national cohort study of CI children and normal hearing (NH) peers. Eligibility criteria include informed consent, age less than 5 years, pre- or post-lingually deaf, developmental criteria met, commitment to educate the child in English and bilateral cochlear implants. All children had a standardised baseline assessment that included

demographics, hearing and medical history, communication history, language measures, cognitive tests, speech recognition, an audiological exam, psychosocial assessment including parent-child videotapes and parent reported quality of life. Follow-up visits are scheduled at six-month intervals and include a standardised assessment of the full battery of measures. Quality assurance activities were incorporated into the design of the study.

A total of 188 CI children and 97 NH peers were enrolled between November 2002 and December 2004. The mean age, gender and race of the CI and NH children are comparable. With regard to parental demographics, the CI and NH children's families are statistically different. The parents of CI children are younger, and not as well educated, with 49% of CI parents reporting college graduation vs. 84% of the NH parents. The income of the CI parents is also lower than the NH parents. Assessments of cognition suggest that there may be baseline differences between the CI and NH children; however the scores were high enough to suggest language learning potential. The observed group differences identified these baseline characteristics as potential confounders which may require adjustment in analyses of outcomes.

This longitudinal cohort study addresses questions related to high variability in language outcomes. Identifying sources of that variance requires research designs that: characterise potential predictors with accuracy, use samples that adequately power a study, and employ controls and approaches to analysis that limit bias and error. The CDaCI study was designed to generate a more complete picture of the interactive processes of language learning after implantation. *Copyright © 2007 John Wiley & Sons, Ltd.*

Keywords: cochlear implants; children; severe to profound hearing loss; study design; baseline characteristics

Introduction

Children with severe to profound sensorineural hearing loss (SNHL) face communication challenges that influence language, psychosocial and scholastic performance. Clinical studies over the past 20 years have supported wider application of cochlear implants (CI) in children. However, the generalisability of these studies may be limited for single-centre, case-series designs that evaluated children using different implant technologies. Studies may have not included a control group nor measured separate variables that can modify outcome or act as confounds in assessing causality of that outcome. There may be greater explanatory strength in studies that examine the impact of cochlear implants from a longitudinal perspective that capture language, social and behaviour attributes of development at baseline, pre-intervention.

Although cultural resistance to early cochlear implantation has generally lessened, two areas of community equipoise regarding early cochlear implantation persist: 'Who should make the decision to implant?' and 'When should this decision be made?' Legal reviews sustain that parents are the reasonable representatives for children in CI candidacy (Brusky, 1995). Proponents of the right of a parent to make this choice argue that denial of the right to implantation of a young child

DOI: 10.1002/cii

'violates her right to an "open future"' (Davis, 1997), and the right to accept or reject the hearing world (Tucker, 1998). Opponents argue that implant technology underscores the observation that our majority culture fails to modify its institutions in order to accommodate the needs of the Deaf (Sparrow, 2005). Regardless of an observer's position along this ideological divide, parents are in the middle. Parental decisions are based on a presumption of 'best interests' that are often guided by considering the impact of an implant on a child's language learning (Yoshinaga-Itano et al., 1998; Nikolopolous et al., 2004) and the myriad implications for a life course that relate to language.

Thus an important measure of success with a (CI) is how a child's CI experience affects his or her ability to learn language. Language represents a complex behaviour that is shaped through auditory function, cognition, attention, communication-in-play and patterns of social interaction during childhood development. Fuller understanding of the effects of cochlear implantation on development calls for rigorous evaluation of these multidimensional domains, employing longitudinal assessment of patient groups beyond those that can be gathered by a single centre.

Summerfield and colleagues have employed multicentre designs to assess outcomes of early cochlear implantation (Summerfield et al., 2003; Barton et al., 2003, 2004, 2006). These investigators have noted that clinical approaches to early cochlear implantation are driven by the hypothesis that short-term gains in audition will translate into medium-term gains in social independence and quality of life, presumably through the communication competence achieved with implant experience. Summerfield and Marshall (1999) noted that prospective randomised controlled trials offer the greatest potential in confirming or refuting this basic hypothesis, but that it is implausible for investigators at this stage of the evolution of CI technology to comply with the tenets of treatment randomisation. Thus landmark studies by this group have utilised comprehensive, cross-sectional databases obtained from centres throughout the United Kingdom.

Longitudinal studies of outcome have shed light on patterns of developmental learning after cochlear implantation. For example, incremental growth in speech recognition ability in implanted children has been observed in noteworthy studies of implanted children followed for two years and longer (e.g. Carney et al., 1991; Fryauf-Bertschy et al., 1992, 1997; Gantz et al., 1994; Kirk et al., 1995; Osberger et al., 1991b; Miyamoto et al., 1996; Waltzman et al., 1995). Speech perception skills often flourish with increased CI experience, but age at implant is strongly and positively correlated with speech recognition ability (an observation noted early in the paediatric CI experience (Staller et al., 1991) and repeatedly confirmed (e.g. El-Hakim et al., 2002; Kirk et al., 1995)). There is also a rapidly growing body of research that charts the integration of auditory behaviours (e.g. McConkey Robbins et al., 2004) and oral language development (e.g. Robbins et al., 1995; Tomblin et al., 1999, 2005; Svirsky et al., 2000, 2004) over time. Such reports demonstrate the power of observing the sequence of developmental learning after implantation that is afforded by longitudinal analysis.

DOI: 10.1002/cii

While prior clinical studies of early cochlear implantation provide key insights into developmental learning, the ability to *explain* key outcomes remains elusive. Clearly, age at implant carries considerable impact. However, when examining predictors of communication outcome in the context of subgroups formed by factors that commonly associate with communicative competence, relatively little explanatory power is generated. For example, taken together, carefully performed studies by Miyamoto et al. (1997), Geers et al. (2003) and Nikolopoulos et al. (2004) found that between 35 and 62% of the variance in speech communication outcomes could be explained by conventional clinical predictors.

The Childhood Development after Cochlear Implantation Study (CDaCI) is the first longitudinal, multicentre, national cohort study to evaluate systematically early CI outcomes in children in the US (Niparko et al., 2005). The study compares children who have undergone cochlear implantation with similarly aged hearing peers across multiple domains, including oral language development, auditory performance, psychosocial and behavioural functioning, and quality of life. Videoanalytic measures are applied to all participants. The comprehensive, prospective approach of the CDaCI study is designed to collect early developmental characteristics of the child and his or her environment that may hold further explanatory power of the primary outcome of language learning. This article presents the CDaCI study protocol and baseline characteristics of the study population, highlighting characteristics of infants and toddlers who underwent cochlear implantation in the US in 2002–04 and features of the CDaCI study designed to fill existing gaps in our understanding of language acquisition after cochlear implantation.

Methods

The CDaCI is a multicentre, national cohort study of the effectiveness of paediatric cochlear implants. Table 1 presents the major study design features.

Study organisation

The CDaCI consists of six clinical implant centres, two preschools of normal hearing (NH) peers, a psychosocial measurement centre and a data coordinating centre. An external advisory board serves to monitor the progress of the study and to provide analytic guidance. The study protocol and informed consent were approved by the Institutional Review Boards at all participating centres.

Eligibility criteria

Children less than 5 years of age were eligible for enrolment. Those enrolled under the age of 2 had to have developmental scores on the Bayley Scales of Infant Development Mental Scale or Motor Scale (BSID II) of at least 70 (Bayley, 1993). Those enrolled over the age of 2 had to have a Leiter International Performance Scale-

DOI: 10.1002/cii

Table 1: Childhood Development after Cochlear Implantation (CDaCI) design summary

Purpose:	To identify factors influencing oral language-related outcomes that impact cognitive, psychosocial and scholastic performance in young children with cochlear implants (CI)
Type of study:	Concurrent prospective cohort study
Sample:	188 children with CI from 6 clinical implant centres 97 children with normal hearing (NH) from 2 preschools
Eligibility:	
Inclusion criteria:	Informed consent provided Less than age 5 years at baseline Pre- or post-lingually deaf Developmental criteria met – specified as BSID II mental or motor score better than 70 for children up to age 2 years **or** a Leiter-R IQ greater than 66 for children older than 2 years[a] Bilingual families who commit to educating child in English-speaking schools Unilateral or bilateral CI[b]
Exclusion criteria:	5 years or older at baseline assessment Patient family not able to participate Post-surgical CI complications Cannot be tested with RDLS in next 2 years No English in household
Enrolment:	1 November 2002 – 31 December 2004 for CI children and normal hearing peers
Follow-up:	Every 6 months for 3 years after enrolment
Outcomes:	
Primary:	Oral language skills (as measured on RDLS)
Secondary:	Speech recognition Attention and problem-solving skills (cognitive) Behavioural and social skills Social adjustment (parent/child) Health-related quality of life and cost-effectiveness

[a] A few children ($n = 39$) over the age of 2 years could not be tested with the Leiter-R and were therefore screened with the BSID II.
[b] CI children who receive a second implant can remain in the study and are followed from the date of their first implant activation.

Revised (Leiter-R) score of at least 66 (Roid and Miller, 2002). Bilingual families were included if they agreed to educate their child in English-speaking schools; and at least one parent could speak English. Children receiving either unilateral or bilateral cochlear implants were included. The audiological criteria for undergoing implantation were based on criteria established at each clinical centre. In general, this meant that the child had severe to profound hearing loss and was not demonstrating adequate benefit with hearing aids. Children were excluded if they had any condition that would preclude their ability to be tested with the Reynell Development Language Scales (RDLS) (Reynell and Gruber, 1990) after

DOI: 10.1002/cii

implantation, if their families could not commit to three years of follow-up and, for the implanted children, if they developed post-surgical CI complications.

Sample size and power analysis

The sample size was determined based on a projection of successful recruitment over a two-year period by six clinical centres, as well as the ability to provide sufficient statistical power to detect any sizable difference in oral language development between subgroups, such as grouping by age at implantation and length of hearing loss. Children in the CDaCI cohort are to be compared with NH age mates at annual timepoints over three years to assess their language learning trajectory. Power calculations were based on the following informed (Reynell, 1990; Svirsky et al., 2000) estimates derived from raw scores of Reynell Developmental Language Scale measures:

(1) NH children aged 1–7 years improve by an average of 20 points (standard deviation 7.5 points) over a three-year period;
(2) children with severe SNHL (Pure Tone Average (PTA) between 90 to 100 dB), aged 1–7 years, improve by an average of 7.5 points over a three-year interval;
(3) children with profound SNHL (PTA > 100 dB), aged 1–7 years, improve by an average of 6.5 points over a three-year interval.

These estimates suggest the difference in the annual rate of language growth between NH and the CI children could be as large as 4.5 points in RDLS measures, and provide estimates of standard deviation needed for power calculations.

The achievable statistical power based on an equal sample size of n for each subgroup is calculated as follows. We assume that raw scores of RDLS, denoted as Y, depend on an independent variable X, the follow-up time in years when Y is measured, as follows:

$$Y_{ijg} = \beta_{0g} + \beta_{1g} X_{ijg} + \varepsilon_{ijg}, i = 1,\ldots,n; j = 1,\ldots,m;$$

where i denotes the i^{th} child within the group, j denotes the j^{th} annual follow-up visit, and g is a label for the subgroup of children, β_{0g} is the intercept and β_{1g} is the rate of change in the RDLS (per year) over time for the g^{th} subgroup. Here n is the number of children in each subgroup and m is the number of repeated annual evaluations taken for each child over the follow-up ($m = 4$). We assume that $\text{var}(\varepsilon_{ijg}) = \sigma^2$. Furthermore, we assume that $\text{corr}(Y_{ijg}, Y_{ikg}) = \rho$ for all $j \neq k$ to account for the within-child correlation between the repeated measurements. Let $g = 0$ denote the control group of NH siblings, then the achievable statistical power for detecting the difference in learning rates (slopes) $\delta_s = \beta_{1s} - \beta_{10}$ between the control group and one of the implantation subgroup can be calculated as $\Phi(Z_Q)$, where

$$Z_Q = (nms_X{}^2 \delta_s{}^2 / 2\sigma^2 (1-\rho))^{1/2} - Z_a,$$

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and s_X^2 is a measure of the variability of the independent variable X (Diggle and Kenward, 1994). Here $X_{ijg} = X_j = j - 1$, for all i and g, indicating the time from implantation to each annual follow-up evaluation, will be roughly identical for every child in all subgroups of this study. Specifically, we used $X_1 = 0$, $X_2 = 1$, $X_3 = 2$ and $X_4 = 3$, indicating the time (in years) of baseline (pre-implantation), first, second and third annual post-implantation follow-up visits, in our calculation and arrived with $s_X^2 = \sum_j (X_j - X)^2/m = 1.25$. The variance of the difference between two evaluation scores three years apart approximates $(7.5)^2$, which is an estimate of $2\sigma^2(1 - \rho)$ if ρ is the correlation between two measurements three years apart. We assumed $\text{corr}(Y_{ijg}, Y_{ikg}) = \rho$ for all $j \neq k$ regardless of when Y_{ijg} and Y_{ikg} are observed and using 56.25 to estimate $2\sigma^2(1 - \rho)$ in the calculation of statistical power. With two-sided type I error = 0.05, and appropriately Bonferroni-adjusted α, we have more than 90% power to detect a 1.3 points/year between group difference in RDLS growth with subgroup sizes of 90 (i.e. two equally sized implanted subgroups plus the NH control group). These calculations yielded a needed sample size of 180 implanted children with 90 NH controls to allow sufficient statistical power to detect a clinically significant effect in subgroup analyses.

Data collection

The CDaCI consists of six clinical implant centres at House Ear Institute, Los Angeles, CA; Johns Hopkins University Listening Center, Baltimore, MD; the University of Miami, Miami FL; the University of Michigan, Ann Arbor, MI; the University of North Carolina, Chapel Hill, NC; and the University of Texas (UT) Callier Research Center, Dallas, TX.

Screening and enrolment

After initial site visits to each clinic, test material development and investigator training, screening of potential participants with SNHL began in October 2002, and the first children were enrolled in November 2002. Recruitment of children continued at all six clinical implant sites until 31 December 2004.

Normal hearing controls were recruited from two preschools affiliated with UT-Dallas and the Johns Hopkins University Listening Center between February 2003 and December 2004. These schools were chosen because they have children below the age of 5 and staff trained in the administration of language and speech recognition tests.

Baseline assessment

All families of children enrolled provided written informed consent and had a standardised baseline assessment. Table 2 presents the CDaCI data collection schedule and the testing protocol. The baseline assessment was typically conducted during two half-day appointments so as not to exhaust the child or the family. The

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Table 2: Data collection schedule and procedures by construct measured

	Visit/target date Forms and/or procedures	BL (Pre-op)[b]	Surg[c]	Post-op[d] (activ.)	6[a]	12[a]	18[a]	24[a]	30[a]	36[a]
	Registration/eligibility review	x								
	Consent	x								
	Communication/educational history	x				x		x		x
	Hearing and medical history	x								
	Audiological test results	x								
	Surgical report form		x							
	Post-op activation report form			x						
LANGUAGE	RDLS (*1–6 years 11 mo*)	x			x	x		x		x
	MacArthur	x			x	x		x		x
	BSID II (*up to 2 years*)	x								
	Leiter-R (*2 years+*)	x								
	OWLS[e] (*3 years+*)	x			x	x		x		x
SPEECH	Infant-toddler meaningful auditory integration scale (IT-MAIS) (*1–3 years*)	x			x	x	x	x	x	x
	Early speech perception (low verbal) (ESP) (*2 years+*)	x			x	x	x	x	x	x
	PSI[f]-Format II (*age 3–7*) (standard)	x			x	x	x	x	x	x
	Early speech perception (*age 3–7*)	x			x	x	x	x	x	x
	MLNT (*age 3+*)	x			x	x	x	x	x	x
	LNT (*age 4+*)	x			x	x	x	x	x	x
	MAIS (*age 4+*)	x			x	x	x	x	x	x
	PBK (*age 5+*)	x			x	x	x	x	x	x
	HINT-C (*age 5+*)	x			x	x	x	x	x	x
PSYCHOSOCIAL	Videotasks: – Free play – Puzzles – Art gallery	x			x	x	x (*free play only*)	x	x (*free play only*)	x

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	– Symbolic play – Noun learning (50+ words) – Speech production (Brown) – Puppet video (*under age 3*)					not at F36
	Continuous performance task (*3 yrs+*)	x	x	x	x	
	Child behaviour checklist	x	x	x	x	
	ECBI (*2–5 years*)	x	x	x	x	
	SSRS[g] (*6–12 years*)					
	SSRS for child (8–12 years)					
	Family Stress Scale – all ages	x	x	x	x	
	Parental Stress Index (1 month–12 years)	x	x	x	x	
	Parental involvement (SPISE)	x	x	x	x	
	Impact of hearing loss – all ages	x	x	x	x	
	From teacher/therapist:	x	x	x	x	
	– SCBE (*2–5 yrs*)					
	– SSRS[g] (*6–12 yrs*)					
	– SEAI < (*2–7 yrs*) / (*7–13 yrs*)					
	– Parental Involvement Scalec (SPISE-T)[h]					
QOL/UTILITY	Ontario Health Utility Index	x		x	x	x
	Visual analogue and time trade-off	x		x	x	x

[a] Six, 12, 18, 24, 30 and 36 months from the post-op CI activation visit.
[b] BL = Baseline.
[c] Surg = Surgery.
[d] Post-op (activ.) = Post-operative (activation).
[e] OWLS = Oral and Written Language Scales.
[f] PSI = Pediatric Speech Intelligibility.
[g] SSRS = Social Skills Rating System.
[h] SPISE-T = Scale of Parental Involvement and Self-Efficacy – Teacher Rating.

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first day included parent-reported questionnaires of the family's demographics, the child's hearing and medical history and the communication and educational history of the child. The child was then assessed with the language measures appropriate for his age, the cognitive tests, the speech recognition hierarchy and an audiological exam. The second day of the baseline assessment was devoted to the psychosocial questionnaires, the videotaped tasks and the parental report of quality of life. If the child was in a preschool or auditory verbal therapy, the teacher was asked to complete standardised questionnaires about the child's behaviour, social skills and parental involvement.

Children who were candidates for a CI were typically scheduled for surgery two to four weeks after their baseline assessment. At the time of surgery, the surgeon completed a surgical report form detailing the type of device and ear implanted and a report of any physiologic findings or complications. After surgery, the child returned in four to six weeks for a post-operative visit and implant activation. The study activation form documented the date of activation and this date was used to set the follow-up schedule at six-month intervals. For the NH children, their first six-month follow-up visit was lagged by four weeks in order to approximate the interval between the baseline assessment, surgery, implant activation and the first six-month visit for CI children.

Follow-up visits

The six- and 12- month follow-up visits included the full battery of language, speech recognition, psychosocial and quality of life measures. Questionnaires assessing child behaviour, social skills, parenting stress and involvement were completed by parents and the child's teacher or therapist. Because rapid change in language and speech recognition was expected post-implant, we wanted to have complete assessments using the full battery at two time points (six and 12 months) in the first year in order to capture these changes longitudinally. After the first year, the annual visits at two and three years included all age-appropriate measures while the intervening mid-year assessments at 18 and 30 months included only the speech recognition hierarchy and a videotape of the free play activities between the parent and child.

Families who enrolled in the study were given honoraria for each year of their participation and gift cards after each completed visit. CI families were provided with a two-year extension of their child's CI processor warranty after completing three years of follow-up. The families of the CI and NH children were given a DVD compiling the videotaped activities over three years.

Quality assurance

Quality assurance activities established by the Data Coordinating Center at Johns Hopkins University and the Psychosocial Measurement Center at the University of Miami were incorporated into the design of the study. Monthly conference calls

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of the investigative team addressed protocol administration, recruitment and retention of participants, and standardisation of all measurements. Site visits to each clinic occurred prior to the start of the study to evaluate the facility for the videotaping component as well as to review the protocol and standardise the clinic team on data collection methods. Each site prepared and submitted a 'pilot' videotape that was reviewed by the Psychosocial Measurement Center to standardise their videotaping technique. Site visits with written feedback occurred yearly since the start of data collection. A *Manual of Procedures* was created and has been updated with changes to the protocol and the addition of new forms or procedures. Data entry was performed at the Data Coordinating Center independent of personnel who provide clinical care to participants. Review, coding and entry of video-based data tapes are similarly performed by study personnel who are masked to performance variables. The study biostatistician also independently evaluates data quality and completeness, conducts data analysis and confirms any statistics provided by the clinical centres to further ensure data quality. Data query reports that summarise the data received at the Data Coordinating Center are sent to the clinical sites quarterly for resolution of outstanding or missing forms and data inconsistencies.

Statistical analysis

Baseline demographic, socioeconomic and medical history factors are described as means (SD) for continuous variables and as frequency distributions for categorical variables. Comparisons between the CI children and the NH peers were tested using two-sample *t*-tests or analysis of variance (ANOVA) for continuous variables and chi-square tests for categorical variables. Distribution of continuous variables such as those from Leiter-R, Brief Form (Roid and Miller, 2002) assessments were compared using standard cutoffs from the instrument. Bayley Scales of Infant Development-Second Edition (BSID-II) (Bayley, 1993) mental and motor developmental scores were converted to respective developmental age and related to the child's chronological age first using scatter plots and non-parametric regression. The differences in mental and motor developmental age between CI and NH children at baseline were then compared using general linear regression models consistent with the exploratory analysis. Pearson correlation between the mental and motor developmental scores was also calculated for the CI and the NH group.

Results

A total of 188 CI children and 97 NH peers were enrolled between November 2002 and December 2004. Table 3 shows that 425 CI children were screened in order to enrol 188 CI children. In applying the findings of cohort studies, it is important to establish whether there are any systematic differences between the characteristics of study participants and eligible non-participants which might

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Table 3: Screening of eligible cochlear implant (CI) children and reasons for exclusion

	N	%
Total screened (11/2002–12/2004)	425	100%
No. enrolled	188	44%
No. excluded	237	56%
Reasons for exclusions		
Non English household	70	30%
No consent given	80	34%
Cognitive impairment	43	18%
Could not test with RDLS	15	6%
Not a CI candidate	29	12%

affect the generalisability of the study results. A log of the characteristics of enrolled, as well as eligible but non-enrolled subjects was kept for evaluation of the representativeness of the study population. Of 425 CI candidates screened, 268 children were eligible for the study (188 consented to participate in the study and 80 declined consent to participate). This 70% participation rate is substantially higher than participation rates that are generally quoted in the 15 to 30% range (Starfield, 1998). For the 80 children who met criteria for the study but did not consent to enrol, there was no difference in the average age of the CI candidate. Comparing participating and non-participating families indicates a higher percentage of African-American families in the non-participating group (19% in the non-participating group vs. 9% in the participating group). Parental age was willingly provided by 41% and socioeconomic status (SES) by 15% of the non-participating families. For those that provided SES data, parental age and SES were similar to participating families.

General baseline characteristics are presented in Table 4. While the mean age of the two groups is comparable (2.2 years for CI vs. 2.3 years for NH), the actual age distributions of the groups were statistically significantly different; with 18% of the CI children being under the age of 1 compared to 5% of the NH children. The difference in age distribution is the result of the NH children being selected from two preschools with very few children under the age of 1. There are sufficient numbers of children in the other age categories for comparison. With regard to gender and race, the two groups of children do not differ. There are more females than males in both groups. Approximately three quarters of the study population describe themselves as white, 9% of the CI and 13% of the NH children are African-American and 5% of the CI and 2% of the NH children are Asian. Twenty per cent of the CI children are of Hispanic origin compared to 9% of the NH children. The larger proportion of Hispanic families among the CI children can be attributed to the location of three of the six clinical sites (Los Angeles, Miami and Dallas) in Hispanic communities. With regard to parental demographics, the CI and the NH children's families are statistically different. The parents of the CI

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Table 4: Baseline characteristics of the Childhood Development after Cochlear Implantation (CDaCI) cochlear implant (CI) children and normal hearing (NH) control children

Characteristic	CI (n = 188) N (%)	NH (n = 97) N (%)	p-value
Age at enrolment (in years)			
<1	34 (18%)	5 (5%)	0.006
1–2	59 (31%)	39 (40%)	
2–3	43 (23%)	32 (33%)	
3–4	34 (18%)	10 (10%)	
4–5	18 (10%)	11 (11%)	
Mean age (yr (SD))	2.2 (1.2)	2.3 (1.1)	
Gender			
Male	90 (48%)	37 (38%)	0.117
Female	98 (52%)	60 (62%)	
Race			
White	134 (71%)	76 (78%)	0.305
African-American	17 (9%)	13 (13%)	
Asian	10 (5%)	2 (2%)	
Hawaiian	0 (0%)	0 (0%)	
American Indian	0 (0%)	0 (0%)	
Other	20 (11%)	5 (5%)	
No answer	7 (4%)	1 (1%)	
Ethnicity			
Hispanic	37 (20%)	9 (9%)	0.059
Not Hispanic	145 (77%)	86 (89%)	
No response	6 (3%)	2 (2%)	
Parents age (years)			
<19	5 (3%)	0 (0%)	0.0001
20–29	72 (38%)	17 (18%)	
30–39	96 (51%)	59 (61%)	
40–49	12 (6%)	19 (20%)	
50–59	0 (0%)	0 (0%)	
60+	0 (0%)	0 (0%)	
Decline/no response	3 (2%)	2 (2%)	
Parent's education			
<High school	14 (7%)	5 (5%)	<0.0001
High school graduate	26 (14%)	2 (2%)	
Some college	55 (29%)	8 (8%)	
College graduate	92 (49%)	81 (84%)	
No response	1 (1%)	1 (1%)	
Parents income			
<$15,000	15 (8%)	5 (5%)	<0.0001
$15–29,999	22 (12%)	4 (4%)	
$30–49,999	42 (22%)	6 (6%)	
$50–74,999	31 (16%)	14 (14%)	
$75–100,000	26 (14%)	13 (13%)	
$100,000+	31 (16%)	49 (51%)	
Declined/don't know	21 (11%)	6 (6%)	

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Table 4: *Continued*

Characteristic	CI (n = 188) N (%)	NH (n = 97) N (%)	p-value
Full term pregnancy			
Yes	161 (86%)	76 (78%)	0.045
No	21 (11%)	11 (11%)	
Don't know/missing	6 (3%)	10 (10%)	
Biological child			
Yes	175 (93%)	85 (88%)	0.019
No	11 (6%)	5 (5%)	
Don't know/missing	2 (1%)	7 (5%)	
No. of siblings			
0	61 (32%)	32 (33%)	0.344
1	75 (40%)	46 (47%)	
2	36 (19%)	15 (15%)	
3	12 (6%)	2 (2%)	
4 or more	4 (2%)	2 (2%)	
Medical conditions present[a]			
Cerebral palsy	1 (1%)	0 (0%)	0.020
Visual impairment	4 (2%)	1 (1%)	
No medical conditions	154 (82%)	67 (69%)	
Don't know/missing	29 (15%)	29 (30%)	

[a]From medical records.

children are younger, and not as well educated, with 49% of CI parents reporting college graduation vs. 84% of the NH parents. The income of the CI parents is also lower than the NH parents with only 16% of CI families reporting a family income of $100,000 or more vs. 51% of the NH children. These socioeconomic differences between the two study groups can be attributed to the homogeneous nature and smaller catchment areas of the two preschools. The observed group differences identified these baseline characteristics as potential confounders which may require adjustment in analyses of outcomes. The two groups did not differ with regard to the enrolled child being born after a full-term pregnancy. Eleven per cent of children in each group were not full term. A total of 93% of the CI and 88% of the NH children were the biologic offspring of the parents. The family size of the two groups is similar; for one-third of the families in each group, the enrolled child is their only child. Forty per cent of the CI and 47% of the NH children have one other sibling.

The medical history was obtained from clinic records at the time of enrolment into the CDaCI. The CI children enrolled had few co-morbidities with 82% having no medical condition other than being deaf. Among the NH children, only one had reported visual impairment at baseline. CI children were enrolled at the time of their first implant. As of August 2006, 21 CI children have received bilateral

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implants. It is expected that this number of bilateral implants will increase over the course of follow-up and the study will document the date, surgical outcomes and activation of the second implant. Four of the 16 CI children had both ears implanted on the same day. The remainder received their second implant at a later date. These children are followed from the date of activation of their first implant. Subgroup analysis of the language outcomes of bilateral vs. unilateral implanted children will be conducted.

Table 5 presents the cause of hearing loss and the communication methods and therapy programmes for the CI children at baseline. The cause of hearing loss was abstracted from the medical record at the time of enrolment. Uncertainty regarding the precise aetiology of hearing loss predominated (29%). Of identifiable aetiologies, a non-syndromic genetic cause was noted in 28% and meningitis in 4%.

Table 5: Cause of hearing loss and communication history of the cochlear implant (CI) children at baseline

Characteristic	CI (n = 188) N (%)
Cause of hearing loss[a]	
Cytomegalovirus (CMV)	3 (2)
Genetic	52 (28)
Head injury	0 (0)
Viral infection	0 (0)
Maternal rubella	0 (0)
Meningitis	7 (4)
Fever	1 (1)
Waardenburg's syndrome	4 (2)
Prematurity	6 (3)
Rhesus (rh) incompatibility	1 (1)
Other cause	59 (31)
Cause unknown	55 (29)
Current communication methods used[b]	
American Sign Language (ASL)	90 (32)
Auditory verbal/oral/aural	98 (52)
Total communication	64 (23)
Other	2 (1)
Don't know/missing	23 (8)
Type of (pre)school or therapy programme	
Parent-infant programme	52 (28)
Other auditory-verbal (A-V) therapy	79 (42)
None	50 (27)
Don't know/missing	7 (4)

[a] From medical records.
[b] From parent report validated by medical records, total N = 277 reflecting multiple communication methods used by 188 children.

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With regard to communication history, parents reported their children were using multiple methods at baseline with 52% of the CI children using oral communication. Thirty-two per cent were using sign and 23% were using total communication in combination with other methods. Twenty-eight per cent were enrolled in parent-infant programmes, 42% were receiving auditory-verbal therapy and 27% were not in any preschool or therapy programme.

Aspects of the hearing history have been given careful consideration. Figures 1 and 2 reveal the pattern and type of hearing loss as well as time spent with hearing, deprivation (known severe SNHL without amplification), and amplification. Over 56% of the children enrolled in the CDaCI had congenital hearing loss.

As an eligibility screen for cognition that would allow for language acquisition, children under age 2 years were tested with the BSID-II, while children 2 years and older were assessed with the Leiter-R. A few children in the CI group (n = 39) over the age of 2 years could not be tested with the Leiter-R and were screened with the BSID-II. The distributions of Leiter-R Brief IQ Composite (BIQ) scores for CI and NH children were assessed and respective histograms using standard cut-points for the population distribution (i.e. standard distribution) provided in the test manual were plotted side-by-side against the frequencies of the standard distribution (Figure 3a). Clearly, both CI and NH groups assessed with Leiter-R at baseline had slightly higher BIQ score distributions compared to the standard distribution.

For those assessed with BSID-II at baseline, both mental and motor developmental scores were converted to respective developmental age related to the child's chronological age for comparison using scatter plots and non-parametric regression. As shown in Figure 3b, the motor developmental age for both CI and NH groups

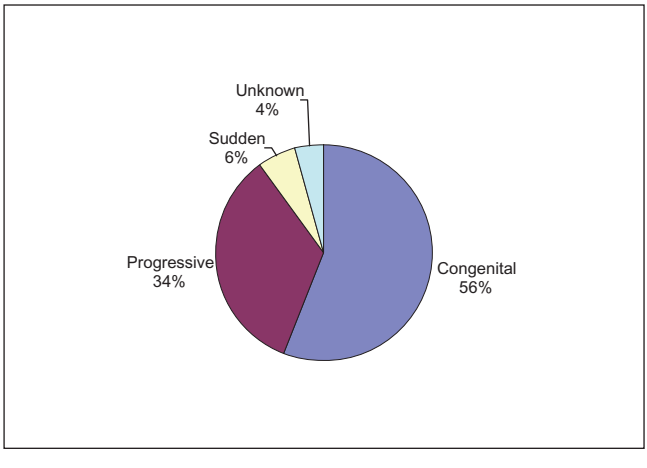

Figure 1: Distribution of hearing loss by type of onset (n = 188). The onset was characterised as congenital (n = 105), progressive (64) and sudden (11). The pattern of onset was unknown in eight adopted children. This figure is available in colour online at www.interscience.wiley.com/journals.cii.

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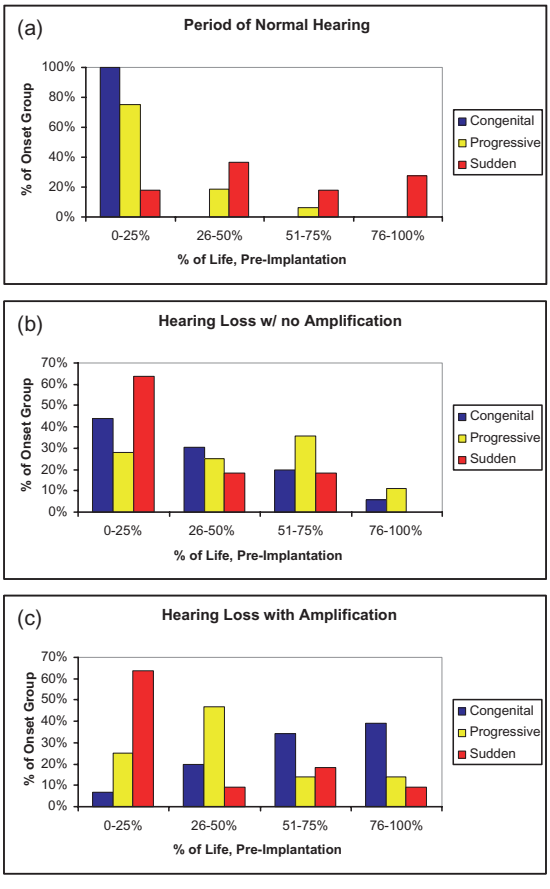

Figure 2: Distributions of periods of (a) hearing, (b) deprivation and (c) amplification prior to cochlear implantation expressed as a percentage of life span at implantation according to type of hearing loss. This figure is available in colour online at www.interscience.wiley.com/journals.cii.

was along the mean developmental trajectory and the slopes of developmental age on chronological age were not significantly different from 1 or from each other ($p = 0.60$). However, the BSID-II mental developmental age at baseline for the CI group was below the normal mean trajectory and the deficit was more pronounced for CI children who were older at baseline (Figure 3c). The slope of mental developmental age on chronological age for the NH group was 1.06 (95% confidence interval, 0.96 to 1.16), which was not significantly different from the standard slope of 1. The slope was 0.69 (95% confidence interval, 0.64 to 0.74) for the CI group, which was significantly different from the standard slope of 1 and the slope of the NH group ($p < 0.0001$). The correlations between the BSID-II mental and motor scale were 0.94 for the NH group, and 0.89 for the CI group.

Assessments of cognition suggest that there may be baseline differences between the CI and NH children. We are currently investigating the potential impact of language level at baseline on the assessment of BSID-II in hearing-impaired

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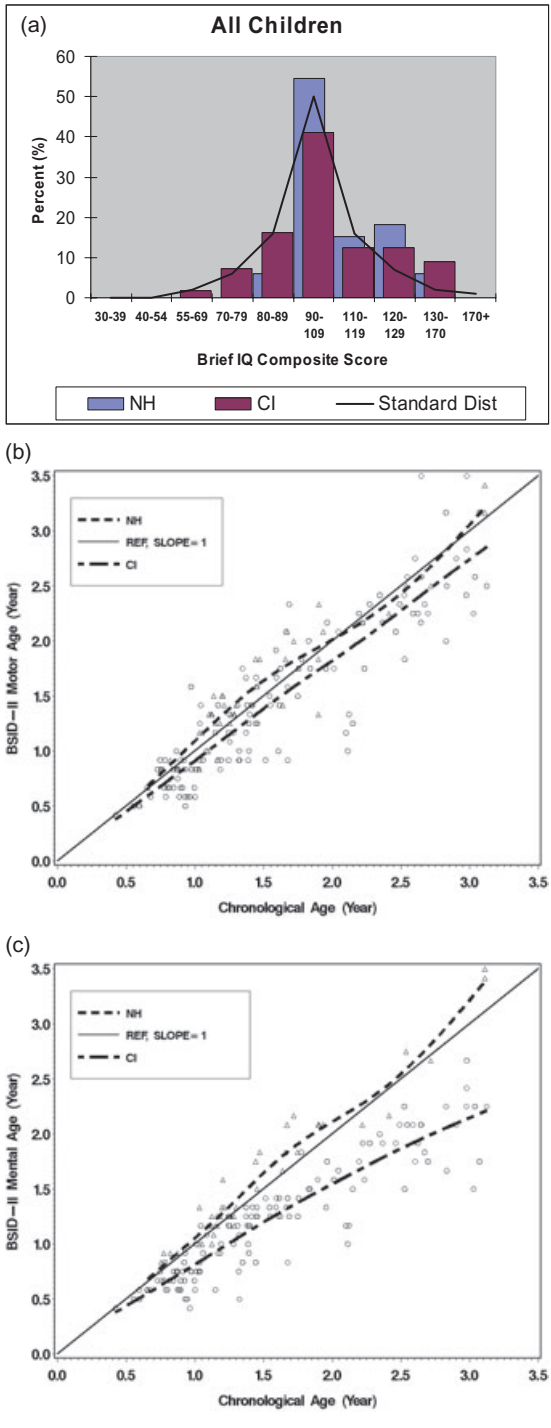

Figure 3: (a) Histograms of Leiter-R International Performance Scale-Revised Brief IQ Composite scores for the cochlear implanted (CI) children and the normal hearing (NH) children in the Childhood Development after Cochlear Implant (CDaCI) study. This figure is available in colour online at www.interscience.wiley.com/journals.cii. (b) BSID-II motor development age versus chronological age for the cochlear implanted (CI) children and the normal hearing (NH) children in the CDaCI Study. (c) BSID-II mental developmental age versus chronological age for the cochlear implanted (CI) children and the normal hearing (NH) children in the CDaCI Study.

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children to gain better insight into the actual cognitive developmental level of the CI group in the CDaCI cohort. The reliability of these measures in very young children will also be assessed with follow-up testing with the Wechsler Intelligence Scale for Children-Fourth Edition (WISC-IV) at age 7 years in both groups. While the mean scores in BSID-II mental scale at all ages were lower for the CI children than the NH children, the scores were high enough to suggest language learning potential.

The CDaCI baseline assessment included an assessment of oral language with the age-appropriate MacArthur Communicative Developmental Inventories (CDI) (Fenson et al., 1993) and with the RDLS (Reynell and Greuber, 1990). Two versions of the CDI were used: the Words and Gestures form (CDI-WG) and the Words and Sentences form (CDI-WS).

Audiologic testing at baseline assessed the ability of the child to identify words and sentences in quiet and in competition using hearing aids, if appropriate (Eisenberg et al., 2006). Testing was structured according to the child's age and functional hearing ability. Typically most hearing-impaired children were not able to perform these tests at baseline.

The psychosocial aspects of childhood development were measured through videotaped assessments of the child with their primary caregiver in five standardised interaction tasks: free play, problem solving, art gallery, symbolic play and noun classification.

Quality of life was assessed by parent-proxy through questionnaires: The Ontario Health Utility Index, two visual analogue scales and a time trade-off survey to derive health utility were administered.

Discussion

We believe the study design employed for the CDaCI study will enable us to address questions related to high variability in language outcome. As in every other aspect of human development, the development of language is characterised by variation (Bates et al., 1995). Identifying sources of that variance requires research designs that:

– characterise potential predictors with accuracy
– use samples that adequately power a study (to support conclusions of differences between groups (or lack thereof)),
– employ controls and approaches to analysis that limit bias and error.

The postulated advantages in generating a more complete picture of the interactive processes of language learning after implantation that were used to design the CDaCI Study are summarised in Table 6.

A comprehensive picture of the effects of auditory deprivation followed by modern CI intervention, provides an opportunity to examine the influence of auditory input provided by a CI on the development of the whole child. For example, variability in language learning likely represents the effects of multiple

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Table 6: Key features of the Childhood Development after Cochlear Implantation (CDaCI) study design

KEY FEATURE:

1. **Prospective design**: Elicitation of biological, environmental and intervention variables in temporal proximity to their occurrence. Averts need for reconstructing past events and the consequences of imprecision of memory. Avoids tendency to externalise responsible factors in order to dissociate outcome and precursors (Gordis, 1996). Recall bias poses a particular challenge in cases of suboptimal outcome whereby measurements are biased by knowledge of the outcome.
2. **Adequate sample size**: As small samples are prone to risks of confounding and bias, we used methodologically rigorous procedures for calculating the needed sample size to adequately power the study in assessing language-related outcomes. The large sample size also allows consideration of sub-groups of interest, such as early-implanted vs. late implanted CI children, bilateral and bimodal CI implanted children vs. unilateral implanted children. While findings within the smaller sub-groups may be less generalisable than the global study results, they should prove valuable guides for further research.
3. **Data processing and statistical analysis**: Performed independent of site personnel who provide care to participants. Review, coding and entry of video-based data tapes are similarly performed by study personnel who are blind to performance variables.
4. **Multicentre participation**: Six clinical sites recruited a demographically diverse population of CI subjects, in numbers mandated by sample size calculations. In addition to the diversity of subjects achieved, the CDaCI subject accrual maintained similarity in the generation of CI provided. Calculations indicate that an individual participating centre would be incapable of generating the needed subjects to detect the size of language learning effect of interest with adequate power.
5. **Multidimensional testing**: Given the multiple factors that underlie language learning, our approach establishes variance of this primary outcome and identifies factors that are most closely related. Prospective assessment of an array of domains is more likely to capture and clarify the role of intermediary and latent variables in influencing an outcome as multidimensional as language. We will develop models of development that unify measures within hierarchical testing of language and speech recognition in CI participants.
6. **Study controls**: CI and NH children accrued from multiple sites have enabled us to begin to address questions of how well children with implants are developing the linguistic skills needed for mainstream participation from a perspective of representative, real-world effectiveness. We will also access the NICHD Early Childcare Database that includes language, behavioural, academic and parent-child interactional data on 1347 children from birth to age 10.

ADVANTAGE:

1. Prospective data accrual heightens accuracy by avoiding recall and informational biases, thus averting threats to validity in estimating effects of independent variables. A *priori* identification of antecedents enhances the ability to assess a factor's explanatory power of variability in outcome. True associations and potential confounds can be discerned. We will be able to correlate patterns of emerging language with accurately recorded antecedents.
2. Results of clinical studies may have limited explanatory power. An incorrect inference that means between the CI and NH groups are not statistically different (i.e. Type 2 error: reaching a false conclusion of no difference when one exists) is a commonly cited concern. Adequate statistical power to reduce the probability of Type 2 error requires that the study has sufficient sample size. CDaCI has calculated sample size under a rigorous study design to ensure proper inferences.

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Table 6: *Continued*

3. Dissociates analysis from the patient-provider bond and awareness of language outcomes. Prevents bias in coding the parent-child interactions based on demographic, site or language characteristics. Similarly, data entry is blinded to these variables.
4. Variance related to the generation of technology employed is avoided. Single-centre, case-series designs have often evaluated children using several different generations of implant technologies. Geers (2004), for example, suggested that this factor may have produced difficult to measure effects on outcome. In addition to controlling a potential source of variance, CDaCI's multisite design provides demographic and geographic diversity and enhanced generalisability of observations.
5. Prospective multidimensional study strategies accurately record early characteristics of subject and environment in a comprehensive manner, allowing for analysis of characteristics that may carry strong associations. Because interrelated factors are measured over time, prospective cohort studies establish antecedents and sequencing that strengthens causal inferences. Repeated testing through a child's development enables models of growth across age-appropriate measures.
6. Measures across domains that are simultaneously performed under stringent protocols are instrumental in characterising childhood participants at baseline and longitudinally, particularly when performed concurrently in CI and NH controls across multiple sites. Concurrent cohort study further establishes temporal relationships, strengthening inferences of causality. Affords meaningful comparison of CI and NH groups, as both have been assessed in the same way contemporaneously.

domains. Though expectations of implantable technologies in promoting language learning have grown, there are limitations in auditory transmission via a CI that persist (likely owing to altered neuronal substrate, channel interaction, compression characteristics and a constrained soundfield). A firm understanding of the impact of CI technology on child development will direct decisions about who should receive the technology, when they should receive it, what additional interventions are needed and how payers should consider the financial implications of this technology. Based on this knowledge, we can also ask whether more technology (bilateral cochlear implants) yields a worthy cost-benefit ratio. Strong study designs offer the best prospects for quality data on which parental decisions should be based.

Control group considerations

Selection of an appropriate control group for CI children has been controversial. Prior studies have compared implanted children's performance pre- to post-implantation (Tobey et al., 1991), and to children using tactile or conventional hearing aids (Boothroyd et al., 1991; Osberger et al., 1991a; Tobey and Geers, 1995; Geers, 1997) or to NH children (Svirsky et al., 2000). The stability of a control group using hearing aids is reduced due to cross-over to the treatment group (e.g. Tomblin et al., 1999), and there is evidence that (1) families who choose early cochlear implantation may differ from those who do not with respect to

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communication methodology (Osberger, 1991b) and (2) that selection criteria may be further relaxed in view of superior performance after implantation for children with residual hearing (Eisenberg et al., 2000; Gantz et al., 2000). The selection of NH children as controls extends the quest for a control group that provides a reference to which to compare the stability of testing procedures as well as between-group performance. Normal hearing controls will allow us to address the challenge of directly assessing the potential for an implanted child to overcome the effects of auditory deprivation to close identified gaps (e.g. Robbins et al., 1995; Tomblin et al., 1999, 2005; Svirsky et al., 2000, 2004) in language learning as well as cognitive, social and behavioural development (Quittner et al., 2004). With NH controls evaluated under the same study protocol as the CI children, the CDaCI design allows for statistical modelling adjustment to account for the level of potential confounding factors such as family income and parental education which are differentially observed between the implanted and control groups.

Multicentre design

Multicentre studies carry both advantages and disadvantages (Spilker, 1991). Disadvantages stem from more complex administrative arrangements, higher costs, a wider range of independent variables and potential disagreement between ethics committees at participating institutions. Advantages lie in rapid subject recruitment, broader research protocols by virtue of dedicated resources, reduced likelihood of investigator bias in the design, conduct or analysis of the data, and most importantly for this population, larger samples with greater heterogeneity.

The CDaCI multicentre design offers several strengths. As indicated by our calculations of sample size, an individual participating centre would be incapable of generating the number of subjects needed in a reasonable time frame to detect the size of language learning effect of interest with adequate power. The short time frame of accruing subjects affords better control of variables related to device design by assessing children who are fitted with same-generation multichannel implants. Evolving criteria, as well as evolving technology, have bedevilled the analysis of outcomes in prior studies (Rubinstein et al., 1999; Cheng et al., 1999). As a completely novel approach, we intend to perform a multidimensional assessment of children enrolled in the study simultaneously from multiple institutions. Thus the design insures that changes in performance over time or across subjects are not related to effects from different generations of technology. Another advantage to this design is the opportunity to evaluate children from geographically diverse sites wherein selection criteria, practice patterns and habilitation/community resources may vary. Data from this longitudinal design may thus enhance prediction of the size and rate of gains in verbal language achieved with cochlear implantation as stratified by other modifying variables. Moreover, this study will provide data that are amenable to multivariate analysis for assessing the independent contribution of predictors on how well young children with cochlear implants develop communicative competence.

DOI: 10.1002/cii

Overall, the CDaCI project is unified by a focus on early language learning. We attempt to address the complexity of language development under conditions of profound hearing loss, especially in the very young child when a variety of operational skills develop rapidly. The study of communication development in hearing-impaired children with cochlear implants requires an examination of cognitive, behavioural and social resources that contribute to the development of linguistic behaviours. Thus the study recognises the multiple parameters of early developmental learning, particularly as it relates to spoken language in children with advanced hearing loss, in order to more clearly determine prognosis for childhood cochlear implantation. Strong study design, as employed in the CDaCI, offers the best prospect for quality outcomes data on which parental decisions should be based.

Acknowledgement

The research was supported by NIDCD National Institute on Deafness and other Communication Disorders-R01DC004797 (JK Niparko, PI Principal Investigator).

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DOI: 10.1002/cii