

Outcomes of 3-Year-Old Children With Hearing Loss and Different Types of Additional Disabilities

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This research investigated the speech, language, and functional auditory outcomes of 119 3-year-old children with hearing loss and additional disabilities. Outcomes were evaluated using direct assessment and caregiver report. Multiple regressions revealed that type of additional disability and level of maternal education were significant predictors of language outcomes. Poorer outcomes were achieved in a combined group of children with autism, cerebral palsy, and/or developmental delay (DD) (Group A), compared with children with vision or speech output impairments, syndromes not entailing DD, or medical disorders (Group B). Better outcomes were associated with higher levels of maternal education. The association between better language outcomes and earlier cochlear implant switch-on approached significance. Further regression analyses were conducted separately for children with different types of additional disabilities. Level of maternal education was the only significant predictor of outcomes for Group A children, whereas degree of hearing loss was the strongest predictor for children in Group B. The findings highlight the variable impact that different types of additional disabilities can have on language development in children with hearing loss.

Introduction

Approximately 20–40% of children born with hearing loss also have significant additional disabilities that might prevent them from reaching their full potential

in regard to speech, language, cognitive, or social-communicative outcomes. Estimates vary according to the definition and participant samples used. [Kennedy et al. \(2006\)](#) reported that an additional disability was present in 19.2% of their sample of 120 British children with hearing loss. An almost identical figure (of 18.6%) was reported by [Berrettini et al. \(2008\)](#) for an Italian sample of similar size. A review by [Picard \(2004\)](#) suggested higher prevalence rates of about 30–40%, consistent with data collected through the Annual Survey of Deaf and Hard of Hearing Children and Youth in the United States, which indicated that 39% of the 31,784 school-aged children surveyed had an educationally relevant additional need ([Gallaudet Research Institute, 2008](#)).

Until recently, studies investigating the outcomes achieved by children with hearing loss and additional disabilities have been underrepresented in the literature ([Edwards, 2007](#)). Two factors have undoubtedly contributed to this situation. First, there are difficulties inherent in administration, scoring, and interpretation of formal assessments with such a heterogeneous subgroup of children; second, much of the research evaluating outcomes has focused on the benefits of cochlear implantation, a procedure for which these children were traditionally considered unsuitable ([Edwards, 2007](#)). Over the past 10 years or so there has been an

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increase in published research on this topic, much of which has been conducted with small groups of participants (e.g., Dammeyer, 2009; Dettman et al., 2004; Donaldson, Heavner, & Zwolan, 2004; Hamzavi et al., 2000; Pyman, Blamey, Lacy, Clark, & Dowell, 2000; Wiley, Meinzen-Derr, & Choo, 2008) or single cases (e.g., Fukuda et al., 2003; Malandraki & Okalidou, 2007).

Benefits of Audiological Intervention for Children With Additional Disabilities

One focus of recent investigations has been on the effectiveness of audiological intervention in the form of cochlear implantation. Performance on specific outcome measures has typically been compared pre-implant versus postimplant. Findings of these studies have generally been interpreted as showing that children with additional disabilities can benefit from cochlear implantation, albeit at a slower pace and/or to a lesser degree than children with no additional disabilities (e.g., Beer, Harris, Kronenberger, Holt, & Pisoni, 2012; Donaldson et al., 2004; Holt & Kirk, 2005; Pyman et al., 2000; Waltzman, Scalchunes, & Cohen, 2000; Yang, Lin, Chen, & Wu, 2004).

Waltzman et al. (2000) reported findings consistent with this view. Their participants were 29 children with profound hearing loss who received their cochlear implants (CIs) at ages ranging from 1.9 to 12 years. The children, who had been diagnosed with a diverse range of additional disabilities, were assessed on a variety of auditory-linguistic measures preoperatively and at yearly intervals (ranging from 1 to 8 years) following implantation. The results showed a gradual increase from year to year in both the number of children able to complete the assessments and the scores they obtained; however, these improvements were not as marked as those reported earlier for a group of children with hearing loss as their only disability (Waltzman et al., 1997). Moreover, there was substantial individual variation within the group of children with additional disabilities, many of whom were unable to recognize words or sentences at any postoperative assessment (Waltzman et al., 2000).

At around the same time, Hamzavi et al. (2000) reported on an investigation of 10 children with a diverse range of additional disabilities, who received

their CIs at ages ranging from 8 to 77 months. As in the study of Waltzman et al. (2000), substantial individual variability was evident in the postoperative outcomes achieved by the children after periods ranging from 12 to 55 months. Only two of the 10 children, both of whom were described as having “moderate learning difficulties” (Hamzavi et al., p. 172), developed the ability to recognize spoken words and sentences post implant. Of the remaining eight children, five did not develop any speech perception or production skills during the study period and three developed the ability to produce and/or understand just a few words. The highly variable nature of these results limits what we can draw from the data.

The primary focus of the research conducted by both Waltzman et al. (2000) and Hamzavi et al. (2000) was on evaluating improvements in children’s auditory-linguistic skills post implant. As noted by Edwards (2007), however, such an approach provides little information about whether children also developed more effective communication skills. A related concern was raised by Berrettini et al. (2008), who commented on the potential importance of evaluating “subjectively perceived benefits” (p. 200) for these children as well as objective improvements in auditory-linguistic skills. In line with this reasoning, Donaldson et al. (2004) used parent-report measures of communicative behavior, as well as formal measures of speech recognition ability and standardized tests of receptive and expressive vocabulary. Participants were seven children with autism spectrum disorder (ASD) who were fitted with CIs at ages ranging from 3 to 9 years. Although numerous missing scores made the results from formal assessments difficult to interpret, the two children who were able to complete most of the tasks showed some evidence of improved speech perception and vocabulary skills at 25 months postoperatively. More convincing results were obtained from parental ratings of children’s communicative skills, which showed consistent improvement for most children in aspects such as reacting to sound, vocalizing, making eye contact, responding to verbal requests, and attending to people.

Despite their use of a small and restricted sample of children, findings of Donaldson et al. (2004) illustrated that the benefits of cochlear implantation for some

children with ASD could be overlooked if only formal, objective assessments were included as outcome measures. A question that their study did not address, however, was whether improvement in children's perceptual skills post implant was associated with better communicative abilities. Berrettini et al. (2008) addressed this issue through their study of 23 children with profound hearing loss who had a diverse range of additional disabilities. All had 1–5 years of CI experience before their postoperative assessment, which was undertaken at ages ranging from 2.3 to 17 years. Assessments of auditory word recognition and communicative behavior (in the form of a parent questionnaire) were administered. The results showed an improvement in children's word recognition skills postoperatively, which was associated with an increase in their use of oral language and an improvement in their perceived communication skills (according to parental report).

Most studies examining outcomes for children with hearing loss and additional disabilities have evaluated the benefits of cochlear implantation. An exception is a study by Kaga, Shindo, Tamai, & Tanaka (2007), which focused on changes in children's auditory behaviors after the fitting of hearing aids (HAs). The participant sample included 28 children with hearing loss and additional disabilities who were fitted with HAs between 1 and 5 years of age. Of the 28 participants, six could not adapt to using HAs, and five showed no improvement in auditory behaviors over the course of the study. The remaining 17 children showed improved auditory behaviors in the months following HA fitting, although their rate of improvement was not as great as for a small control group of five children with no additional disabilities who were fitted at 1 year of age. Interpretation of this group difference is complicated however, by variation in the age at which HAs were fitted and children were assessed. In addition, no information was provided regarding children's degree of hearing loss.

The studies described above provide support for the effectiveness of audiological intervention, especially in the form of CIs, for children with hearing loss and additional disabilities considered as a group. They also reveal, however, marked individual variation between participants in their responses to intervention, which may in part reflect the influence of uncontrolled

demographic variables, including level of intellectual functioning.

Outcomes in Relation to Intellectual Disability

Previous research has provided evidence of a positive association between children's level of intellectual or cognitive functioning and their outcomes following cochlear implantation (e.g., Beer et al. 2012; Dettman et al., 2004; Edwards, Frost, & Witham, 2006; Lee, Kim, Jeong, Kim, & Chung, 2010; Meinzen-Derr, Wiley, Grether, & Choo, 2010; Wiley et al., 2008). It is unclear however, whether cognitive ability is the only factor, or even the most important factor, in determining outcomes for children with additional disabilities. Possible counterevidence came from Berrettini et al. (2008) who reported similar outcomes for a subgroup of children with intellectual disability as for their entire sample of children with additional disabilities. Furthermore, within the intellectually disabled subgroup, there was no significant association between degree of intellectual disability and any of the postimplant outcome measures. However, findings of Berrettini et al. need to be interpreted cautiously, because 8 of the 10 children with an intellectual disability were classified as mildly disabled. In a more recent study by Lee et al. (2010), a group of children with mild intellectual disability also performed similarly in some respects to children with no additional disabilities, although they achieved better postimplant outcomes than children with moderate intellectual disability on measures of speech perception, speech production, and receptive language.

Other evidence has suggested that a moderate intellectual disability may not always prevent a child with hearing loss from achieving postoperative speech, language, and communication outcomes that resemble those achieved by children with no additional disability. Fukuda et al. (2003) described a single case study of a young male with moderate intellectual disability, who was fitted with a CI at the age of 4 years 8 months. He made good progress in the perception and production of spoken language in the months and years that followed implantation, which, according to the authors, was similar to that observed in previous Japanese studies of children with hearing loss and no additional

disabilities. It may be that the presence of a severe intellectual disability would, however, be sufficient to preclude typical communicative development, regardless of a hearing loss.

Findings reported by [Holt and Kirk \(2005\)](#) provided additional support for the view that degree of intellectual disability alone cannot account for variation in the postimplant outcomes of children with hearing loss and additional disabilities. Holt and Kirk selected participants with the aim of reducing heterogeneity and investigated the impact of a mild intellectual disability per se. Nevertheless, a large amount of (unexplained) intersubject variability was evident in their results, leading them to conclude that there was “a need to determine the impact of other disabilities, such as autism, low vision, physical impairments, and combinations of disabilities on speech and language development in deaf children with cochlear implants” (p. 147). This aspect was addressed in the research described here although the focus was not restricted to children with CIs but also extended to children with HAs.

Outcomes in Relation to Demographic Variables

Individual variation in the outcomes achieved by participants with hearing loss and additional disabilities might also reflect differences in audiological and family-related demographic variables. Although previous systematic research on this topic is limited, [Meinzen-Derr et al. \(2010\)](#) found that neither parental education nor income was associated with post-CI language outcomes in a small, heterogeneous sample of twenty 5-year-old children with hearing loss and additional disabilities. It is worth noting, however, that levels of maternal education were relatively restricted in their study, with 79% of mothers educated beyond a high school level. A different pattern of results might emerge if greater variability in parental education levels were present.

In the same study, [Meinzen-Derr et al. \(2010\)](#) used multiple regression techniques to identify possible associations between specific audiological variables and children’s language outcomes. The results showed that both age at diagnosis of hearing loss

and duration of CI use accounted for significant unique variance in outcomes after controlling for variation in nonverbal cognitive ability. More specifically, earlier age at diagnosis and “shorter” duration of implant use were both associated with better receptive and expressive language quotients on the Preschool Language Scale Fourth Edition (PLS-4; [Zimmerman, Steiner, & Pond, 2002](#)). It is noteworthy that the authors downplayed the significance of their finding in regard to duration of use. They noted its counterintuitive nature (“longer” term usage might have been expected to accompany better language outcomes) and suggested that it might have reflected the inclusion in their participant sample of several long-term implant users whose language skills were poor. Fifteen of the participants from this original study also formed the basis for a subsequent report by the same authors ([Meinzen-Derr, Wiley, Grether, & Choo, 2011](#)). In this more recent investigation, age at cochlear implantation was not correlated with children’s receptive or expressive language outcomes as measured by the PLS-4. Furthermore, subsequent multiple regressions revealed that no audiological variables (including average four-frequency thresholds) accounted for significant variance in PLS-4 outcomes once nonverbal cognitive ability was controlled.

By contrast with [Meinzen-Derr et al. \(2010, 2011\)](#) who used the directly administered PLS-4 to investigate children’s outcomes, [Beer et al. \(2012\)](#) used a measure of auditory functioning based on parental report, the Infant-Toddler Meaningful Auditory Integration Scale (IT-MAIS; [Zimmerman-Phillips, Robbins, & Osberger, 2000](#)). Participants included 46 children with CIs: 23 children who had been diagnosed with a disability in addition to hearing loss and a matched group of 23 children with hearing loss and no additional disabilities. Correlational analyses involving a set of audiological variables and a measure of children’s post-CI improvements in auditory functioning revealed no significant associations within the group of children with additional disabilities, despite the fact that better auditory outcomes were associated with earlier age at implantation in the matched group of children with no additional disabilities. On the basis of these differential findings, Beer et al. concluded that “established early predictors

of functional auditory benefit may not be as critical or meaningful in predicting benefit in deaf children with ADs” (p. 497). One aim of the current study was to explore this suggestion through investigation of a large participant sample using a wide range of outcome measures.

To summarize, previous research examining outcomes achieved by children with hearing loss and additional disabilities is limited. There have been few studies reported in the literature, and those that have been published typically contained data from fewer than 30 participants with additional disabilities, who often varied widely in regard to the nature of their disabilities, age at implantation, and duration of device use. The use of small, heterogeneous samples creates difficulties for generalization, and for examining the impact of different types of disability in isolation from other, potentially confounding, audiological and family-related variables. As a result, researchers have recently called for “further studies including multiple centres and wider, more homogeneous samples ... in order to develop standardized measures to evaluate the overall outcomes and to better define the prognostic factors and expected benefits in this population” (Berrettini et al., 2008 p. 207).

Current Study

In order to meet this demand and address the shortcomings of previous investigations, the present research measured speech, language, and functional auditory outcomes in a large sample of 3-year-old children with hearing loss and additional disabilities. These children were drawn from a population-based cohort who participated in the 3-year-old assessment phase of a longitudinal study investigating outcomes of children with hearing loss (the “Longitudinal Outcomes of Children with Hearing Impairment” or “LOCHI” study, as described by Ching et al., 2013). In the wider LOCHI study, children’s outcomes were quantified in terms of global factor scores, which were computed for each child using a combination of individual test scores encompassing measures of receptive and expressive language, speech production, auditory functional performance, and psychosocial functioning. Multiple regression analysis revealed that the presence of an additional disability

was associated with a significant reduction of 10.4 global factor score points, thus making it one of the strongest predictors of children’s speech, language, and functional outcomes (Ching et al., 2013).

The aim of this investigation was to provide detailed information about outcomes achieved by children with hearing loss and additional disabilities who took part in the wider LOCHI study. Because the sample was large and heterogeneous, it was possible to identify subgroups of children with different types of additional disabilities, thus reducing within-group variability and enabling direct examination of the impact of type of additional disability on children’s outcomes. All of the children were fitted with CIs or HAs. The inclusion of a sample of children with HAs distinguished this research from the majority of previous studies in the area, which have focused predominantly on outcomes for children with CIs (although see Kaga et al., 2007, for an exception). In line with recommendations from previous research, outcome measures included both formal assessments of speech and language development as well as more subjective measures of functional auditory behavior based on parent/clinician report.

Research Questions and Hypotheses

Two research questions were addressed.

1. Which demographic variables are associated with subjective and formal assessments of speech, language, and functional auditory outcomes in young children with hearing loss and additional disabilities? Do different outcome measures reveal similar patterns of association? The specific demographic variables under consideration were derived from previous research and included both audiological variables (degree of hearing loss, type of sensory device, age at fitting of sensory devices) and child- and family-related variables (gender, type of additional disability, maternal education, communication mode at home).
2. Are similar demographic variables important in predicting outcomes for children with hearing loss who have different types of additional disabilities?

Given the paucity of published literature relating directly to our research questions, predicted outcomes were tentative. Nevertheless, in the absence of strong evidence to the contrary, we hypothesized that: (a) demographic variables, including age at fitting of sensory devices and the nature of a child's additional disability, would be associated with variation in speech, language, or functional auditory outcomes in our sample of children with hearing loss and additional disabilities and (b) similar demographic variables would be important in predicting outcomes for children across a range of additional disabilities.

Method

Participants

A sample of 119 children with hearing loss and additional disabilities took part in this investigation. They were drawn from a population-based cohort participating in the LOCHI study referred to earlier. An invitation to participate in a prospective study on outcomes was issued to all families of children who were born between 2002 and 2007 and who presented for hearing services below 3 years of age at pediatric centers administered by Australian Hearing (the government-funded hearing service provider for all children in Australia) in New South Wales, Victoria, and Southern Queensland. As part of the LOCHI data collection procedure,

demographic information describing the children, their families, and their environment was elicited from caregivers using custom-designed questionnaires. Among other things, caregivers were asked to indicate whether or not their child had been diagnosed with a disability in addition to hearing loss by a qualified professional. All children who had been diagnosed with an additional disability by the age of 3 years were included in this study (approximately 26.4% of the total LOCHI population). A wide range of additional disabilities was reported. For descriptive purposes these additional disabilities were grouped into eight nonoverlapping categories, as shown in Table 1.

The first two categories were ASD and cerebral palsy (CP), respectively, both of which occurred either alone or in combination with other disabilities. The most frequent combinations were with DD, which was reported in 3 out of 9 participants with ASD and 16 out of 24 participants with CP. The third and fourth disability categories were DD groupings, in which DD was reported either in combination with other disabilities or syndromes (excluding ASD and CP), or as a child's only disability. The remaining four categories involved no reported DD. They encompassed disorders of vision, speech output, various syndromes that could not be assumed to entail DD, and a diverse set of medical disorders, often affecting major body organs or motor skills (see Table 1 for further detail).

Table 1 Numbers (and percentages) of children with different types of disabilities ($N = 119$)

Type of disability	No. of children (%)	
1. Autism spectrum disorder (ASD)	9	(7.6)
2. Cerebral palsy (CP)	24	(20.2)
3. Developmental delay (DD) with a syndrome/condition other than ASD or CP ^a	27	(22.7)
4. DD only	14	(11.8)
5. Vision ^b	9	(7.6)
6. Speech output only ^c	4	(3.4)
7. Various syndromes (not entailing DD) ^d	19	(16.0)
8. Medical ^e	13	(10.9)
Total	119	

^aThis category included five participants whose caregivers did not specify DD but whose syndromes entailed DD; namely, Down syndrome ($n = 1$), CHARGE syndrome ($n = 1$), and Cornelia de Lange ($n = 3$).

^bThis category did not include 26 children with a visual disability who also had CP ($n = 10$), DD ($n = 14$), or another syndrome (not entailing DD; $n = 2$). Those children were included in categories 2, 3, and 7 as appropriate.

^cThis category included Oromotor/oral dyspraxia ($n = 2$), speech disorder ($n = 1$), and stuttering ($n = 1$).

^dThis category included Treacher-Collins syndrome ($n = 4$), Waardenburg syndrome ($n = 3$), bronchio-oto-renal ($n = 2$) syndrome, Otspondylomegaepiphyseal dysplasia, Pendred's syndrome, Goldenhar's syndrome, Stickler syndrome, proximal symphalangism, glycogen storage disorder, sensory overload, long QT syndrome, translocation of chromosomes 2 and 6, and an unspecified genetic disorder ($n = 1$ each).

^eThis category included disorders of the brain (microcephaly), heart, kidneys, thyroid, bones, muscles, and nervous system.

Table 2 presents relevant background data on the cohort of 119 participants, more than half of whom were boys. Audiological information was collected from the databases of Australian Hearing and relevant intervention agencies. Hearing loss is represented as a four-frequency average in the better ear (4FA HL; see Table 2). Across the cohort, hearing loss at 3 years of age ranged from mild to profound ($M = 66.4$, $SD = 31.3$, range = 20.0–123.8). The majority of children were HA users, with just over one quarter using unilateral or bilateral CIs. Device use was associated with degree of hearing loss. All children with mild or moderate losses used HAs, as did 79% (19 out of 24) of children with a severe loss. By contrast, the majority of children with a profound loss (29 out of 32 or 90.6%) used a CI. On average, children had been diagnosed with a hearing loss at 6.1 months of age ($SD = 7.7$, range = 0.07–34.4) and first fitted with HAs approximately three months later ($M = 9.2$, $SD = 9.2$, range = 0.9–34.8). For children using CIs, devices were first switched-on between 5.4 and 35.6 months of age ($M = 17.5$, $SD = 7.5$). Given that age at CI switch-on

and duration of CI use provided essentially redundant information in the current study, duration of use is not reported in detail. It is noteworthy, nevertheless, that 32 of the 34 children with CIs (94.1%) had more than 10 months of experience with their device.

Children's socioeconomic status was measured using the Index of Relative Socioeconomic Advantage and Disadvantage (IRSAD) from the Socio-Economic Index for Areas (Australian Bureau of Statistics, 2006). Lower IRSAD scores indicate geographic areas with relatively fewer resources, whereas higher scores indicate geographic areas with relatively more resources. Scores are expressed as deciles. The majority of children in the current cohort lived in more advantaged areas, with 66.9% scoring 7 or above on the IRSAD (Median = 7.0, mode = 9.0, range = 1–10). Parental education was measured using a three-point scale. Both female and male caregivers were fairly evenly divided between those who had a university qualification, those with a diploma or certificate, and those with 12 years or less of school attendance (see Table 2).

Table 2 Participants' background information ($N = 119$)

Variable	No. of participants (%) ^a	
Gender (male)	74	(62.2%)
Degree of hearing loss (4FA HL)		
Mild (20–40 dB)	25	(21.0%)
Moderate (41–60 dB)	38	(31.9%)
Severe (61–80 dB)	24	(20.2%)
Profound (>80 dB)	32	(26.9%)
Device use		
Hearing aid		
Bilateral	72	(60.5%)
Unilateral	13	(10.9%)
Cochlear implant (CI)		
Bilateral	14	(11.8%)
Unilateral	6	(5.0%)
CI plus hearing aid	14	(11.8%)
Maternal education ($n = 117$)		
1. University qualification	45	(38.5%)
2. Diploma or certificate	34	(29.1%)
3. 12 years or less of schooling	38	(32.5%)
Paternal education ($n = 100$)		
1. University qualification	37	(37.0%)
2. Diploma or certificate	32	(32.0%)
3. 12 years or less of schooling	31	(31.0%)
Communication mode at home		
Oral only	68	(57.1%)
Mixed (sign and speech)	51	(42.9%)

Note. 4FA HL = the average of hearing threshold levels at 0.5, 1, 2, and 4 KHz, represented nonlinearly.

^aOwing to missing data for some variables, scores are based on different numbers of participants as specified.

Caregivers were asked to describe their children's method of communication at home as being oral only, manual/sign only, or mixed (i.e., sign and speech). Most children in this cohort used oral communication only at home, although a significant minority used mixed communication (see Table 2). No children were reported to communicate using sign only. More specifically in regard to spoken language, all of the children used English at home, with a small number of these ($n = 10$, 8.4%) using another spoken language as well.

Evaluation Tools

Evaluation tools included both formal assessments of children's speech and language development as well as more subjective measures of functional auditory behavior based on parent/clinician report.

Formal assessments. Formal assessments included the PLS-4 (Zimmerman et al., 2002), Peabody Picture Vocabulary Test Fourth Edition (PPVT-4; Dunn & Dunn, 2007), and Diagnostic Evaluation of Articulation and Phonology (DEAP; Dodd, Hua, Crosbie, Holm, & Ozanne, 2002).

The PLS-4 was used to provide a formal assessment of children's overall receptive and expressive language abilities. At 3 years of age, this test incorporates interactive play, picture pointing, and verbal elicitation activities targeting children's knowledge of English semantics, morphology, and syntax. In recent studies, the PLS-4 has been used with children who have developmental disability in the form of ASD (e.g., Volden et al., 2011) and CP (e.g., Hustad, Gorton, & Lee, 2010).

The PPVT-4 was used to provide a second formal measure of receptive language, in particular, vocabulary. This widely used test is based on a four-alternative, forced-choice, picture-selection format and has been used successfully to assess children from a range of special populations, results of which are presented in the test manual.

The phonology subtest of the DEAP was included to provide a quantitative measure of children's speech production ability. Single-word utterances are elicited using pictures, verbal cues, and/or imitation. For present purposes, children's speech output was scored in

terms of percent consonants correct (PCC) and percent vowels correct (PVC).

Report-based evaluations. Report-based evaluations included the Child Development Inventory (CDI; Ireton, 2005), the Parent Evaluation of Aural/Oral Performance of Children (PEACH; Ching & Hill, 2007), and a Speech Intelligibility Rating (SIR; Yoshinaga-Itano, 1998).

The CDI provides a caregiver report of participants' receptive and expressive language abilities. This tool takes the form of a questionnaire, which comprises 300 statements to which caregivers are asked to respond "yes" or "no" based on observations of their child's behaviors. Items are divided into eight scales, results for only two of which, language comprehension (50 items) and expressive language (50 items), are reported here. It should be noted that in two instances, children's early intervention specialists completed the CDI because caregivers were unable to do so.

The PEACH was included to provide a measure of participants' functional auditory performance in everyday situations as judged by their caregivers. Caregivers were asked to answer 13 questions based on observations of their child over a period of at least 1 week. Two questions addressed the child's use of sensory devices and occurrence of discomfort in response to loud sounds. The remaining 11 questions solicited information about the child's ability to listen and to communicate in quiet and in noise, use of the telephone, and the child's response to environmental sounds in everyday situations. An overall score of functional performance was calculated on the basis of the summed ratings provided in response to the 11 questions.

The final report-based evaluation was a rating of speech intelligibility (SIR) completed by a research speech pathologist. For this purpose, a 6-point scale described by Yoshinaga-Itano (1998) was used: (1) I always or almost always understand with little or no effort; (2) I always or almost always understand; however, I need to listen carefully; (3) I typically understand about half of the child's speech; (4) I typically understand about 25% of the child's speech; (5) I understand only occasional words; and (6) I never or almost never understand the child's speech. For children who had

few or no word approximations, a response of “no rating” was recorded.

Procedure

The data reported in this paper were collected when children reached a chronological age of approximately three years. Although there was some variation in age at testing across individual children and tasks (range: 34 to 42 months for CDI, PLS-4, PPVT-4, and PEACH; and 34 to 44 months for DEAP), the majority of assessments across all tasks (88.8%) were conducted between 36 and 40 months of age.

A team of research speech pathologists directly assessed children in their homes or early intervention centers. During evaluations, children wore HAs and/or CIs at their personal settings. For children who used speech and sign to communicate, tests were administered in speech and sign by a qualified speech pathologist/sign language interpreter. As far as possible, research speech pathologists were blinded to children’s severity of hearing loss and hearing device settings.

Reliability. Inter-rater reliability was computed for the group of participants in the larger LOCHI study. Formal assessments were video/audio recorded, and randomly selected samples were subjected to a second, independent scoring by a member of the research

speech pathologist team who was not involved in the initial test administration or scoring. A total of 34 PLS-4 assessments (10.9%) and 13 PPVT-4 assessments (5.7%) were double-scored. Agreement was high on test items administered for both PLS-4 receptive (98.6%) and PLS-4 expressive (98.6%), and for the PPVT-4 (98.1%). A total of 21 DEAP assessments (10%) were transcribed a second time from the audio and/or video recording by a different speech pathologist. Point-to-point agreement was 88% for consonants and 94% for vowels (90% for all phonemes).

Task administration. Standardized measures of language (PLS-4, PPVT-4, and DEAP) were directly administered where each child’s abilities allowed. Administration of standardized assessments was slightly modified in some cases to cater for children’s abilities. Children with poor hand control, for example, completed the PLS-4 using larger blocks; children with vision impairment were allowed additional time to look at pictures. Despite these accommodations, some children remained unable to cope with the demands of formal, standardized testing as shown in Table 3. In particular, around 40% of children were unable to cope with the demands of the PPVT-4, and nearly 48% were unable to complete the DEAP. Furthermore, the rates of noncompletion of these two formal assessments varied as a function of disability type, such that children

Table 3 Number and percentage of participants unable to cope with demands of directly administered tasks as a function of disability type ($N = 119$)

	Assessment task		
	PLS-4	PPVT-4	DEAP
Disability type			
1. Autism spectrum disorder, ASD ($n = 9$)	0 (0.0%)	7 (77.8%)	9 (100.0%)
2. Cerebral palsy (CP, $n = 24$)	4 (16.7%)	17 (70.8%)	15 (62.5%)
3. Developmental delay (DD) plus other ($n = 27$)	2 (7.1%)	14 (50.0%)	17 (60.7%)
4. DD only ($n = 14$)	1 (6.7%)	6 (46.7%)	7 (53.3%)
5. Vision ($n = 9$)	0 (0.0%)	0 (0.0%)	2 (22.2%)
6. Speech output ($n = 4$)	0 (0.0%)	0 (0.0%)	1 (25.0%)
7. Various syndromes ($n = 19$)	0 (0.0%)	3 (15.0%)	4 (20.0%)
8. Medical ($n = 13$)	0 (0.0%)	1 (7.7%)	2 (15.4%)
Total ($N = 119$)	7 (5.7%)	48 (40.2%)	57 (47.5%)
Disability group			
Group A (ASD, CP, DD; $n = 74$)	7 (9.5%)	44 (59.5%)	48 (64.9%)
Group B (Other; $n = 45$)	0 (0.0%)	4 (8.9%)	9 (20.0%)

Note. PLS-4, Preschool Language Scale, Fourth Edition; PPVT-4, Peabody Picture Vocabulary Test, Fourth Edition; DEAP, Diagnostic Evaluation of Articulation and Phonology.

with ASD, CP, and/or DD were more often than not unable to cope with the task demands, whereas children with vision or speech output impairments, various syndromes not entailing DD, or medical disorders achieved consistently high rates of completion (75% or better). On the basis of this differential performance, two disability groups were formed. Group A included children with ASD, CP, and/or DD; Group B included children with other disabilities (see Table 3 for average rates of noncompletion in these two participant groups).

Statistical Considerations and Preliminary Data Analysis

A range of individual test scores was used to investigate outcomes relating to receptive language (PLS-4, CDI, and PPVT-4), expressive language (PLS-4 and CDI), speech output (percent consonants and vowels correct on the DEAP, and SIR), and functional auditory performance (PEACH). Statistical analysis was conducted using raw scores rather than standard scores for several reasons. First, and most important, use of standard scores sometimes resulted in a marked loss of sensitivity and variability; for example, of 85 children who completed the PLS-4, 35 (29.4%) achieved the lowest possible standard score of 50 on receptive language. The corresponding raw scores achieved by this subgroup of children varied considerably, however, from a minimum of 4 to a maximum of 23, with an approximately normal distribution ($M = 17.9$, $SD = 4.3$). Second, because there was little variation in assessment age across the participant sample (as outlined above), the use of raw scores was not considered problematic. Even if scores increased with age, as might be expected, its effects as a confounding factor should be negligible. Finally, the primary purpose of this study was not to compare the performance of this cohort of children directly with published assessment “norms,” but rather to examine their speech, language, and functional auditory outcomes in regard to demographic and disability-related variables as measured within the sample.

In line with this primary research purpose, our first aim in analyzing the data was to examine the extent to which children’s individual speech, language, and functional auditory outcomes were associated with each of a group of audiological variables (4FA HL,

sensory device [HA or CI], and age at fitting of sensory device), and child and family-related variables (gender, disability group, maternal education, communication mode at home). This initial statistical analysis was conducted using the Pearson’s product-moment correlational procedure. Not all of the variables described in Table 2 were included in the analysis, primarily because they measured related characteristics. Level of maternal education was significantly correlated with both level of paternal education (Pearson’s $r [N = 100] = .44$, $p < .001$) and socioeconomic status (Pearson’s $r [N = 111] = -.25$, $p = .009$); that is, children with more highly educated mothers tended to live in geographic areas with relatively more resources). Maternal education was selected for inclusion because it was more evenly distributed across the participant sample than was socioeconomic status, and there were fewer missing data points than for paternal education (see Table 2). Age at fitting of HAs was included in preference to age at diagnosis of hearing loss, because the two variables were highly correlated (Pearson’s $r [N = 119] = .87$, $p < .001$), and the former variable was a specific focus of the research. Because all children in the sample, even those who eventually received a CI, were fitted with HAs initially, correlational analyses involving age at fitting were based on data for the entire sample of 119 children. Importantly, however, when these correlations were recomputed using data from the smaller set of 85 participants who were still using HAs at 3 years of age, the pattern of significant findings was identical, except for the correlation between age at fitting and 4FA HL, which was no longer significant (Pearson’s $r [N = 85] = -.16$). This difference reflected the decrease in variability in hearing loss within the more restricted participant sample.

Subsequent to the overall correlational analysis, multiple regression techniques were employed to identify demographic variables accounting for significant unique variance in receptive or expressive language outcomes assessed using the PLS-4 and CDI. Use of multiple regressions was restricted to outcome data from the PLS-4 and CDI for two main reasons. First, it ensured sufficient numbers of participants per predictor variable. Second, it minimized the number of missing data points resulting from participants’ inability to cope with the demands of directly administered tasks,

which were not evenly distributed across disability groups (see Table 3).

For each of the four dependent variables (PLS-4 receptive, PLS-4 expressive, CDI receptive, and CDI expressive), three regression models were fitted. The first model included four categorical variables and one continuous variable. The categorical variables were gender, device type (HA or CI), communication mode at home (oral or mixed), and maternal education, which was recoded as two binary variables using university-level education as the reference category. The continuous variable was 4FA HL. In the second model, two more continuous variables, age at HA fitting and age at CI switch-on, were added to the set of predictors. Because age at switch-on was available only for participants with CIs, there were numerous nonrandom missing data points, which were replaced with the average value for this variable¹. In the third and final model the categorical variable of disability group was added. In the event that disability group was a significant predictor of language outcomes, further multiple regressions were planned to investigate the association between demographic variables and outcomes within each disability group (research question 2).

In line with standard practice, a Type I error rate of $\alpha = .05$ (two-tailed) was adopted for all statistical analyses, which were performed using SPSS and R (R Development Core Team, 2011) with the additional R package, rms (Harrell, 2011).

Results

Some data were missing as a result of participants being unable to cope with the demands of formal testing (see Table 3). On other occasions, participants were unavailable for testing or were not using their HAs, assessments were attempted but not completed, or questionnaires were not returned. Remaining participant numbers ranged from a low of 33 (for the DEAP) to highs of 83 (for the CDI) and 85 (for the PLS-4).

As the outcome measures with the largest number of observations, PLS-4 and CDI results are reported in detail below. PPVT-4 scores were available for 40 children, who knew an average of 27.8 words. There was marked variability however, with individual scores ranging from 1 to 84 ($SD = 19.2$). For the 33 children

who completed the DEAP, speech output accuracy was better for vowels ($M = 83.3\%$, $SD = 12.4$) than for consonants ($M = 48.6\%$, $SD = 20.7$). Speech intelligibility ratings, which were available for 49 children, revealed relatively poor outcomes, with a mean rating of 4.2 ($SD = 1.8$) on a scale from 1 to 6, where 1 is 100% intelligible.

Associations Between Demographic Variables and Outcome Measures

Table 4 presents the results of a bivariate correlational analysis conducted to address the first research question. As shown, four demographic variables were significantly correlated with children's speech, language, or functional auditory outcomes.

1. Use of oral communication rather than a mixture of oral and sign language was associated with better receptive and expressive language scores on both the PLS-4 and the CDI, and better auditory functional scores on the PEACH.
2. Higher levels of maternal education were associated with better receptive and expressive language scores on both the PLS-4 and the CDI, and better auditory functional scores on the PEACH.
3. More severe levels of hearing loss were associated with poorer receptive vocabulary scores.
4. Children in Disability Group A (ASD, CP, DD) achieved lower receptive and expressive language outcomes than children in Disability Group B (other disabilities) on both the PLS-4 and CDI. In regard to these associations with Disability Group, Table 5 shows the mean scores achieved on the PLS-4 and CDI as a function of disability group and the more specific, disability type. These mean data illustrate the consistency with which children allocated to Group B outperformed children allocated to Group A; that is, the overall group effect was not due to the performance of children with just one or two disability types. Finally, Group A children also received poorer caregiver ratings of their functional auditory performance on the PEACH, and poorer clinician ratings of speech intelligibility (SIR; see Table 4).

Table 4 Correlations (Pearson's r) between demographic variables and outcome measures with number of paired observations in parentheses

	Mode	MatEd	4FA HL	Device	AgeHA	AgeSO	DisabGrp	CDIRec	PLSRec	PPVT-4	CDIExp	PLSExp	SIR	PCC	PVC	PEACH
Gender	.10 (119)	.10 (117)	.15 (119)	.08 (119)	.09 (119)	.13 (34)	.14 (119)	-.03 (83)	.01 (85)	-.10 (40)	.00 (83)	-.05 (85)	-.04 (49)	.02 (33)	.04 (33)	-.06 (66)
Mode	1.00 (119)	.19* (117)	.15 (119)	.09 (119)	-.23* (119)	-.09 (34)	-.26** (119)	-.34** (83)	-.29** (85)	-.05 (40)	-.38*** (83)	-.38*** (85)	.02 (49)	.09 (33)	.01 (33)	-.38*** (66)
MatEd	1.00 (117)	1.00 (117)	-.05 (117)	-.18* (117)	-.07 (117)	.37* (33)	-.11 (117)	-.32** (83)	-.31** (85)	-.18 (40)	-.30** (83)	-.33** (85)	.27 (49)	-.09 (32)	.00 (32)	-.31* (66)
4FA HL	1.00 (119)	1.00 (119)	1.00 (119)	.85*** (119)	-.28** (119)	-.07 (34)	.11 (119)	-.16 (83)	-.20 (85)	-.34* (40)	-.12 (83)	-.16 (85)	-.07 (49)	-.10 (33)	-.24 (33)	-.06 (66)
Device	1.00 (119)	1.00 (119)	1.00 (119)	1.00 (119)	-.25** (119)	— ^a	.16 (119)	-.05 (83)	-.12 (85)	-.25 (40)	-.03 (83)	-.10 (85)	.05 (49)	-.02 (33)	-.15 (33)	.08 (66)
AgeHA	1.00 (119)	1.00 (119)	1.00 (119)	1.00 (119)	1.00 (119)	.31 (34)	.02 (119)	.09 (83)	.18 (85)	.05 (40)	.06 (83)	.12 (85)	.27 (49)	-.04 (33)	-.25 (33)	-.02 (66)
AgeSO	1.00 (119)	1.00 (119)	1.00 (119)	1.00 (119)	1.00 (119)	1.00 (34)	.23 (119)	-.20 (23)	-.33 (24)	.12 (13)	-.17 (23)	-.38 (24)	.19 (17)	.08 (12)	.38 (12)	-.41 (20)
DisabGrp	1.00 (119)	1.00 (119)	1.00 (119)	1.00 (119)	1.00 (119)	1.00 (119)	1.00 (119)	.58*** (83)	.49*** (85)	-.03 (40)	.58*** (83)	.54*** (85)	-.33* (49)	-.14 (33)	-.04 (33)	.36*** (66)

Note. Gender (1 = male; 2 = female); mode = mode of communication at home (1 = oral; 2 = mixed); MatEd = maternal education (1 = university; 2 = diploma/certificate; 3 = 12 years or less formal schooling); 4FA HL = four-frequency average hearing loss in the better ear; Device (1 = hearing aid [HA]; 2 = cochlear implant [CI]); AgeHA = age at hearing aid fitting; AgeSO = age at CI switch-on; DisabGrp = disability group (1 = autism spectrum disorder, cerebral palsy, or developmental delay; 2 = vision, speech output, various syndromes, medical); CDIRec = Child Development Inventory receptive language (language comprehension) raw score; PLSRec = Preschool Language Scale Fourth Edition receptive language (auditory comprehension) raw score; PPVT-4 = Peabody Picture Vocabulary Test Fourth Edition receptive vocabulary raw score; CDIExp = Child Development Inventory expressive language raw score; PLSExp = Preschool Language Scale Fourth Edition expressive communication raw score; SIR = speech intelligibility ratings; PCC = Diagnostic Evaluation of Articulation and Phonology (DEAP) percent consonants correct; PVC = DEAP percent vowels correct; PEACH = Parent Evaluation of Aural/Oral Performance of Children grand total.

^aAge at switch-on applies only to children with CIs.

* $p < .05$; ** $p < .01$; *** $p \leq .001$.

Table 5 Receptive and expressive language outcomes as a function of assessment task and disability

Disability type	Preschool Language Scale Fourth Edition (PLS-4)						Child Development Inventory (CDI)							
	Receptive			Expressive			Receptive			Expressive				
	N	Mean	(SD)	Range	Mean	(SD)	Range	N	Mean	(SD)	Range	Mean	(SD)	Range
1. Autism spectrum disorder (ASD)	8	19.5	(2.7)	17-24	23.6	(4.0)	18-28	9	9.7	(5.0)	6-19	6.9	(4.3)	1-16
2. Cerebral palsy (CP)	17	23.7	(11.6)	4-44	26.1	(12.7)	7-46	18	13.3	(13.6)	0-37	13.9	(15.1)	0-42
3. Developmental delay (DD) plus other	18	22.2	(6.5)	8-35	24.6	(5.7)	10-35	17	12.2	(10.4)	0-35	10.5	(11.3)	0-40
4. DD only	9	26.2	(6.8)	18-37	29.0	(5.7)	22-39	11	14.3	(8.0)	0-24	11.4	(8.4)	0-25
5. Vision	7	32.1	(8.0)	24-45	36.0	(6.7)	26-47	6	32.8	(12.0)	12-46	34.3	(12.9)	12-48
6. Speech output	4	38.5	(11.8)	27-54	40.5	(11.1)	27-53	4	30.3	(11.5)	16-44	27.0	(12.2)	9-36
7. Various syndromes	13	33.3	(9.8)	19-51	36.5	(7.2)	25-49	9	27.3	(10.7)	10-44	27.8	(11.4)	15-45
8. Medical	9	31.8	(12.1)	15-48	36.0	(11.8)	18-54	9	25.2	(11.8)	5-45	26.8	(13.6)	5-47
TOTAL	85 ^a	27.0	(10.3)	4-54	30.0	(10.1)	7-54	83 ^a	17.9	(13.0)	0-46	17.1	(14.3)	0-48
Disability group														
Group A (ASD, CP, DD)	52	23.0	(8.3)	4-44	25.7	(8.5)	7-46	55	12.6	(10.4)	0-37	11.2	(11.5)	0-42
Group B (Other) ^b	33	33.3	(10.1)	15-54	36.7	(8.8)	18-54	28	28.3	(11.2)	5-46	28.8	(12.2)	5-48

^aThere was not total overlap between the samples of participants with PLS-4 and CDI scores: 74 children had scores on both measures, whereas an additional 11 children had PLS-4 scores but no score for the CDI, and 9 children had CDI scores but no score on the PLS-4.

^bGroup B (Other) includes children with disorders of vision or speech output, various syndromes not entailing DD, and medical disabilities.

None of the four remaining demographic variables (gender, device, age at HA fitting, or age at CI switch-on) was significantly associated with children's outcomes.

Predicting Outcomes as a Function of Demographic Variables

A series of four multiple regressions was conducted to investigate in more detail the associations described above. The most salient finding was that disability group accounted for significant unique variance in receptive and expressive language outcomes on both the PLS-4 and the CDI, after controlling for the variance associated with all other demographic variables (see Table 6). Disability group (Group A or B) accounted for between 15% and 21% of variance across the four language outcomes and was the only variable to account for significant unique variance in CDI outcomes (see Table 6 for regression coefficients for all variables included in the final multiple regression model). For PLS-4 outcomes, level of maternal

education was also a significant predictor ($p < .05$) and age at CI switch-on approached significance ($p < .06$). These findings reflect the fact that better receptive and expressive language scores were associated with higher levels of maternal education and earlier CI switch-on.

Given the significant differentiation between children with different types of additional disabilities (Groups A and B), two further sets of multiple regressions were conducted to identify the demographic variables predicting language outcomes within each group. To compensate for the reduced number of participants in each analysis, gender, age at HA fitting, and age at CI switch-on were omitted from the set of predictor variables due to their nonsignificant zero-order correlations with the four outcome measures. The remaining variables were 4FA HL, maternal education, sensory device (HA or CI), and communication mode (oral or mixed).

For Group A (ASD, CP, DD), the four variables combined did not account for significant variance in receptive or expressive language outcomes (see Table 7). Level of maternal education did, however,

Table 6 Multiple regression summary table for receptive and expressive language outcomes

Predictors	Dependent variable			
	Preschool Language Scale Fourth Edition ($N = 85$)		Child Development Inventory ($N = 83$)	
	Receptive	Expressive	Receptive	Expressive
	<i>R</i> ² change			
Gender, 4FA HL, mode, maternal education (MatEd), device	.20**	.25***	.20**	.21**
Age at hearing aid (HA) fit, age at cochlear implant (CI) on	.03	.02	.01	.01
Disability Group	.15***	.16***	.21***	.20***
Total <i>R</i> ²	.38***	.43***	.42***	.42***
	Regression coefficients			
Gender (reference male)	0.351	-0.997	-0.769	0.251
4FA HL	-0.064	-0.019	-0.094	-0.080
Device (reference HA)	-0.957	-3.137	0.008	0.008
Mode (reference oral)	-0.964	-2.809	-2.144	-4.754
MatEd (reference University level)				
Certificate/Diploma	-2.164	-2.497	-2.559	-1.947
12 years or less	-5.494*	-5.694*	-4.795	-3.686
Age at HA fit	0.150	0.095	-0.045	-0.105
Age at CI on	-0.507	-0.474	-0.410	-0.387
Disability group (reference Group A)	8.917***	9.235***	14.293***	15.522***

Note. Regression coefficients are for the final model containing all predictor variables; Mode = communication mode at home; Device = HA versus CI; Disability Group A = autism spectrum disorder, cerebral palsy, and developmental delay; Disability Group B = vision, speech, various syndromes, & medical; 4FA HL = four-frequency average hearing loss in the better ear; MatEd = maternal education (1 = university; 2 = certificate/diploma, 3 = 12 years or less) was coded as two binary variables in regression analyses using university education as the reference category.

* $p < .05$, ** $p < .01$, *** $p \leq .001$.

account for significant unique variance in three of the four language measures when all other variables were controlled (11.2% for CDI receptive language, $p < .05$; 14.1% for PLS-4 receptive language, $p < .05$; and 15.8% for PLS-4 expressive language, $p < .05$). A different pattern of results emerged for Group B (other disabilities). For three of the four outcome measures, the set of four predictor variables combined accounted for significant variance of between 41% and 48% (see Table 7). In all cases, 4FA HL accounted for significant unique variance (from 9.6% to 17.6%) once all other variables were controlled. There was only one other significant unique predictor, communication mode, which accounted for 9.4% of the variance in PLS-4 expressive language scores ($p < .05$).

Discussion

The primary aim of this investigation was to evaluate speech, language, and functional auditory outcomes in

a large sample of 3-year-old children with hearing loss and additional disabilities. In line with recommendations in the literature, a range of outcome measures was used, involving both direct assessment and caregiver report (e.g., Berrettini et al., 2008; Edwards, 2007). A total of 119 children took part in the study, which was aimed at exploring: (a) the demographic variables that were associated with speech, language, and functional auditory outcomes in this cohort of children and (b) whether similar demographic variables were important in predicting outcomes for children with different types of additional disabilities.

With regard to the first of these research aims, results from correlational analyses showed that higher levels of maternal education and the use of oral communication at home were both associated with improved outcomes in receptive and expressive language (on the PLS-4 and CDI) and functional auditory skills (on the PEACH). This analysis also revealed that milder levels of hearing loss were associated with better outcomes

Table 7 Multiple regression summary table for receptive and expressive language outcomes as a function of disability group

Predictors	Disability group							
	Group A (ASD, CP, DD)				Group B (other disabilities)			
	Dependent variable							
	PLS-4		CDI		PLS-4		CDI	
	Receptive	Expressive	Receptive	Expressive	Receptive	Expressive	Receptive	Expressive
	<i>R</i> ² change							
4FAHL, MatEd, device, mode	.15	.19	.16	.13	.44**	.48**	.34	.41*
<i>N</i>	52	52	55	55	33	33	28	28
	Regression Coefficients							
4FA HL	0.005	0.045	0.000	0.035	-0.204**	-0.170*	-0.315**	-0.333**
MatEd	(reference university)							
Certificate/diploma	-5.129	-6.597*	-7.982*	-8.148	1.616	2.561	4.052	6.141
≤ 12 years	-7.820**	-8.151**	-8.222*	-6.678	-2.442	-2.968	-4.618	-7.768
Device	-0.932	-4.015	-4.038	-5.651	1.142	1.466	13.107	15.752
	(reference hearing aid)							
Mode	-0.668	-2.644	-2.689	-4.142	-5.405	-6.430*	-2.504	-7.088
	(reference oral)							

Note. 4FA HL = four-frequency average hearing loss in the better ear; ASD = autism spectrum disorder; CDI = Child Development Inventory; CP = cerebral palsy; DD = developmental delay; PLS-4 = Preschool Language Scale Fourth Edition. Regression coefficients are for the final model containing all predictor variables; MatEd = maternal education (1 = university; 2 = certificate/diploma, 3 = 12 years or less) was coded as two binary variables in regression analyses using university education as the reference category.

* $p < .05$; ** $p < .01$.

in receptive vocabulary (on the PPVT-4) but not with receptive or expressive language (on the PLS-4 or CDI). There was, however, no significant association between age at fitting of sensory devices (HAs or CIs) and children's performance on any of the outcome measures in the current participant sample.

The nature of children's additional disabilities was significantly correlated with language outcomes on the PLS-4 and CDI, as well as with functional auditory performance (on PEACH) and speech intelligibility (on SIR). Children with ASD, CP, and/or DD (Group A) achieved consistently poorer outcomes than children with vision or speech output impairments, various syndromes not entailing DD, or medical disorders (Group B). Multiple regressions were conducted using PLS-4 and CDI receptive and expressive language scores as dependent variables. The results revealed a consistent and significant effect of disability group, which accounted for between 15% and 21% of the variance in language outcomes after controlling for gender, hearing loss, device type (HA or CI), communication mode at home (oral vs. mixed), level of maternal education, age at fitting of HAs, and age at CI switch-on (see [Table 6](#)). In fact, disability group was the only variable to account for significant unique variance in all four language outcomes, with children in Group A (ASD, CP, DD) performing more poorly than children in Group B (other disabilities).

The only other significant predictor of language outcomes in these overall multiple regressions was level of maternal education, which accounted for unique variance in PLS-4 receptive and expressive language scores only. In particular, children whose mothers had completed postsecondary education performed better than children whose mothers had 12 years or less formal schooling. This association might in part reflect the fact that children whose mothers had higher levels of education tended to live in geographic areas with more resources, as reflected in a significant correlation between maternal education and socioeconomic status.

Finally, some weak evidence emerged for the potential benefits of earlier cochlear implantation, with the predictor variable of age at CI switch-on almost reaching significance ($p < .06$) in analyses of PLS-4 receptive and expressive language outcomes. It is interesting that children's mode of communication at home, which had

a significant zero-order correlation with language outcomes on the PLS-4 and CDI, did not emerge as a significant unique predictor in these multiple regressions. Inspection of the data suggest that this finding was due to the overlap between disability group and communication mode, whereby the majority (73.3%) of children in Group B (other disabilities) used oral communication only, whereas most children (52.7%) in Group A (ASD, CP, DD) used a mix of sign and speech.

In light of the finding that disability group accounted for a significant percentage of the variance in children's receptive and expressive language outcomes, further multiple regressions were conducted to address our second research question: whether similar demographic variables were important in predicting outcomes for children with different types of additional disabilities. Level of maternal education was the only demographic variable to predict significant unique variance in language outcomes for children in Group A (ASD, CP, DD); whereas for children in Group B (other disabilities), 4FA HL was the strongest predictor, accounting for significant unique variance in all four language outcomes.

Results of the current study supported the larger LOCHI study in revealing that higher levels of maternal education, in particular postsecondary education, were associated with better outcomes. Although in this study, that association appears to be due primarily to the combined group of children with ASD, CP, and/or DD, this apparent group difference might instead reflect an underrepresentation of mothers with less than 12 years education in the "other" disability group. Just 6 out of 33 children (18.2%) with other types of disabilities had mothers with less than 12 years education, compared with 22 out of 52 children (42.3%) of the group with ASD, CP, and/or DD. As noted earlier, a similar argument could be made in regard to the failure of [Meinzen-Derr et al. \(2010\)](#) to find an association between parental education level and post-CI language outcomes on the PLS-4. In their sample of 20 children with hearing loss and additional disabilities, 79% of mothers were educated beyond a high school level.

Another similarity between the current findings and those reported in the larger LOCHI study lies in the observed association between children's degree of hearing loss and their language outcomes. In the current

study, children with less severe hearing losses achieved better receptive vocabulary scores (on the PPVT-4), as reflected in a significant negative correlation between the variables. It is important to remember, however, that most children who were able to complete the PPVT-4 belonged to Group B (with disabilities other than ASD, CP, or DD). This group of children was also the one for whom 4FA HL was the major predictor of PLS-4 and CDI language outcomes, as reflected in the results from multiple regression analyses (see Table 7). In the current study, however, degree of hearing loss was not an important correlate of language outcomes for the combined group of children with ASD, CP, and/or DD, even though the distribution of 4FA HLs was similar in this group of participants as it was in the group of participants with other disabilities. It is possible that for children with ASD, CP, and/or DD, the additional disability itself was of paramount importance in determining their capacity to acquire language skills.

Finally, it is noteworthy that in the current study the effect of age at CI switch-on was in the expected direction (better language outcomes on the PLS-4 were associated with earlier switch-on), even though the required significance level of $p < .05$ was not met. It is possible that a stronger effect of early auditory stimulation on language development may emerge at an age older than 3 years. By design, the children in this study will be evaluated again at 5 years of age, when effects of the predictor variables on outcomes will be investigated further. Pending collection of additional data, any conclusions drawn from the current findings must remain tentative, given the lack of statistical significance. Nevertheless, the findings provide limited support for recommendations of early cochlear implantation in children with hearing loss and additional disabilities (e.g., Lee et al., 2010).

This research has made an important contribution to the literature in showing that children's receptive and expressive language outcomes differed according to the nature of their additional disabilities. The limited number of participants included in most previous studies made it difficult to test this proposal in an objective manner. There has also been a tendency to downplay the predictive role of disability type and focus instead on the degree of cognitive impairment when

determining outcomes (e.g., Beer et al., 2012; Edwards, 2007; Pyman et al., 2000; Wiley et al., 2008). Beer et al. (2012) stated that "The presence of any ADs, regardless of type, could place a strain on the child, family, and habilitation team that might affect long-term CI performance" (p. 492). Nevertheless, in the present study, a classification procedure was established using the nature of children's additional disabilities as its basis, and group membership proved to be highly effective, not only in predicting children's language outcomes but in identifying a differential influence of important demographic variables on performance. In particular, although degree of hearing loss did not account for significant unique variance in language outcomes for the combined group of children with ASD, CP, and/or DD, it was a significant predictor of language outcomes for the group of children with other types of additional disabilities.

A possible weakness of the current classification system is the lack of independent confirmation of children's status within the subgroup classified as having DD alone. It is possible that children in this category might have received their diagnosis at least partly as a result of perceived communication difficulties, which for our purposes were also the primary outcome measures. Although one might argue against this possibility on the grounds that children in this subgroup performed similarly in all observed respects to children with a diagnosis of DD plus another syndrome/condition, a stronger argument could be made on the basis of independent assessment of children's nonverbal cognitive abilities. The 5-year-old assessments conducted as part of the wider LOCHI study incorporate a measure of nonverbal cognitive ability, which should address this shortcoming.

A second potential weakness lies in the fact that our participant sample was not large enough to enable comparison of children grouped according to more specific disability types. On the basis of data reported here, an interesting contrast would involve children with ASD versus those with CP and/or DD. Children with ASD were less likely to cope with the demands of formal testing than children in any other disability category and attained the lowest average scores on both receptive and expressive language scales of the PLS-4 and CDI, but their limited number (only nine children

were diagnosed with ASD) prevented formation of a separate disability group.

Regardless of these potential weaknesses, the current study has a number of advantages over previous investigations of outcomes for children with hearing loss and additional disabilities. In contrast with previous published literature in the area, which has been dominated by small-sample or individual case studies (e.g., [Dammeyer, 2009](#); [Donaldson et al., 2004](#); [Fukuda et al., 2003](#); [Hamzavi et al., 2000](#); [Malandraki & Okalidou, 2007](#); [Meinzen-Derr et al., 2010, 2011](#); [Wiley et al., 2008](#)), our participant sample was large and heterogeneous, comprising 119 children with a diverse range of additional disabilities. Use of a large, heterogeneous sample meant that it was feasible to evaluate the influence of a range of demographic variables that have not been examined in previous studies and also compare statistically the outcomes achieved by groups of children with different types of additional disabilities.

The inclusion of a range of outcome measures, some directly assessed and others based on caregiver report, is another strength of the current research, especially because the pattern of results obtained in the multiple regression analyses differed for the PLS-4 (directly administered) compared with the CDI (a parent-report measure). In fact, the PLS-4 was a particularly effective assessment in the context of the current study, because the majority of children were able to cope with the demands of testing, even though a large number of them achieved receptive language raw scores that mapped onto the lowest possible standard score. (See [Volden et al., 2011](#), for a similar pattern of results based on data from 294 preschool children with ASD but no severe hearing impairment). Not all outcome measures involving direct assessment were as appropriate for this sample of children with additional disabilities at 3 years of age. In particular, a substantial number of children, especially those with ASD, CP, or DD (Group A), were unable to cope with the task requirements of the DEAP and the PPVT. At the 5-year-old assessments, we would expect this pattern of results to change, as more children become able to cope with the more difficult tasks.

Finally, in order to generalize the findings reported here to the population of children with hearing loss

and additional disabilities, it is important to consider the extent to which our sample is representative of the wider group. In this regard, it is noteworthy that the current sample was drawn from an Australian, population-based cohort who participated in the 3-year-old assessment phase of the LOCHI study ([Ching et al., 2013](#)). The only additional criterion for inclusion in the current investigation was the presence of a diagnosed disability in addition to hearing loss by the age of 3 years. Also encouraging are the nature and distribution of disability types within our sample. Thus, although the total percentage of children with reported additional disabilities was somewhat lower in the current study (26.4%) than in the 2007–2008 Annual Survey of Deaf and Hard of Hearing Children in the United States (in which 39.3% of children had “educationally relevant conditions”), the range of reported disability types was similar. Their relative frequencies, although computed differently, both revealed a large percentage of children with DD (DD, mental retardation, or specific learning disability) and smaller percentages with other disabilities including visual impairment (or deafblindness), orthopedic impairment (including CP), and ASD.

In conclusion, this investigation of outcomes for children with hearing loss and additional disabilities has shown that at least some of the same factors that are associated with outcomes in the wider population of children with hearing loss were also associated within this cohort, although the strongest associations varied according to the nature of children’s additional disabilities. More specifically, level of maternal education was a significant predictor of children’s language outcomes in general, and degree of hearing loss was important, especially for the combined group of children with visual or speech output impairments, syndromes not entailing DD, and medical disorders. In regard to age at fitting of sensory devices, the results provided weak evidence to suggest that age at CI switch-on might be associated with better outcomes in receptive and expressive language, although the results fell just short of statistical significance. Finally, the results of this investigation have made an important contribution to our understanding of the variable impact that additional disabilities of different types can have on language development in children with hearing loss.

Notes

1. Given the large number of nonrandom missing values for age at CI switch-on (i.e., for children who do not have CIs), two sets of additional multiple regressions were conducted for each of the four dependent variables. In the first set, we omitted age at CI switch-on as a predictor. The same patterns of significance and nonsignificance were obtained for the remaining predictors. In the second set, we created a new continuous variable, age at fitting of primary device, which was either the age at HA fit or the age at CI switch-on. When this new variable was substituted for the two component variables, the pattern of results did not change.

Conflicts of Interest

No conflicts of interest were reported.

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References

- Australian Bureau of Statistics (2006). Information paper: An introduction to Socio-Economic Indexes for Areas (SEIFA) (ABS cat. no. 2039.0). Retrieved from [http://www.ausstats.](http://www.ausstats.abs.gov.au/ausstats/subscriber.nsf/0/D729075E079F9FD_ECA2574170011B088/$File/20390_2006.pdf)
- Beer, J., Harris, M. S., Kronenberger, W. G., Holt, R. F., & Pisoni, D. B. (2012). Auditory skills, language development, and adaptive behaviour of children with cochlear implants and additional disabilities. *International Journal of Audiology, 51*, 491–498. doi:10.3109/14992027.2012.664291
- Berrettini, S., Forli, F., Genovese, E., Santarelli, R., Arslan, E., Chilosi, A. M., & Cipriani, P. (2008). Cochlear implantation in deaf children with associated disabilities: Challenges and outcomes. *International Journal of Audiology, 47*, 199–208. doi:10.1080/14992020701870197
- Ching, T. Y. C., Dillon, H., Marnane, V., Hou, S., Day, J., Seeto, M., ... Yeh, A. (2013). Outcomes of early- and late-identified children at 3 years of age: Findings from a prospective population-based study. *Ear and Hearing, 34*, 535–552. doi:10.1097/AUD.0b013e3182857718
- Ching, T. Y. C., & Hill, M. (2007). The Parent's Evaluation of Aural/Oral Performance of Children (PEACH) Scale: Normative data. *Journal of the American Academy of Audiology, 18*, 220–235. doi:10.3766/jaaa.18.3.4
- Dammeyer, J. (2009). Congenitally deafblind children and cochlear implants: Effects on communication. *Journal of Deaf Studies and Deaf Education, 14*, 278–288. doi:10.1093/deafed/enn042
- Dettman, S. J., Fiket, H., Dowell, R. C., Charlton, M., Williams, S. S., Tomov, A. M., & Barker, E. J. (2004). Speech perception results for children using cochlear implants who have additional special needs. *The Volta Review, 104* (monograph), 361–392.
- Dodd, B., Hua, Z., Crosbie, S., Holm, A., & Ozanne, A. (2002). *Diagnostic evaluation of articulation and phonology (DEAP)*. London, UK: Harcourt.
- Donaldson, A. I., Heavner, K. S., & Zwolan, T. A. (2004). Measuring progress in children with Autism Spectrum Disorder who have cochlear implants. *Archives of Otolaryngology—Head and Neck Surgery, 130*, 666–671. doi:10.1001/archotol.130.5.666
- Dunn, L. M., & Dunn, D. M. (2007). *Peabody Picture Vocabulary Test* (4th Ed.). Minneapolis, MN: Pearson Assessments.
- Edwards, L. C. (2007). Children with cochlear implants and complex needs: A review of outcome research and psychological practice. *Journal of Deaf Studies and Deaf Education, 12*, 258–268. doi:10.1093/deafed/enm007
- Edwards, L. C., Frost, R., & Witham, F. (2006). Developmental delay and outcomes in paediatric cochlear implantation: Implications for candidacy. *International Journal of Pediatric Otorhinolaryngology, 70*, 1593–1600. doi:10.1016/j.ijporl.2006.04.008
- Fukuda, S., Fukushima, K., Maeda, Y., Tsukamura, K., Nagayasu, R., Toida, N., ... Nishizaki, K. (2003). Language development of a multiply handicapped child after cochlear implantation. *International Journal of Pediatric Otorhinolaryngology, 67*, 627–633. doi:10.1016/S0165-5876(03)00016-8
- Gallaudet Research Institute (2008). Regional and national summary report of data from the 2007–08 annual survey of deaf and hard of hearing children and youth. Retrieved from: http://research.gallaudet.edu/Demographics/2008_National_Summary.pdf

- Hamzavi, J., Baumgartner, W. D., Egelierler, B., Franz, P., Schenk B., & Gstoettner, W. (2000). Follow up of cochlear implanted handicapped children. *International Journal of Pediatric Otorhinolaryngology*, *56*, 169–174. doi:10.1016/S0165-5876(00)00420-1
- Harrell, F. E. (2011). rms: Regression modelling strategies (Version 3.3-1). Retrieved from <http://cran.r-project.org/package=rms>
- Holt, R., & Kirk, K. (2005). Speech and language development in cognitively delayed children with cochlear implants. *Ear and Hearing*, *26*, 132–148. doi:10.1097/00003446-200504000-00003
- Hustad, K. C., Gorton, K., & Lee, J. (2010). Classification of speech and language profiles in 4-year-old children with cerebral palsy: A prospective preliminary study. *Journal of Speech Language and Hearing Research*, *53*, 1496–1513. doi:10.1044/1092-4388(2010/09-0176)
- Ireton, H. (2005). *Child development inventory*. Minneapolis, MN: Child Development Review.
- Kaga, K., Shindo, M., Tamai, F., & Tanaka, Y. (2007). Changes in auditory behaviours of multiply handicapped children with deafness after hearing aid fitting. *Acta Oto Laryngologica Supplement*, *559*, 9–12. doi:10.1080/03655230701596368
- Kennedy, C. R., McCann, D. C., Campbell, M. J., Law, C. M., Mullee, M., Petrou, S., ... Stevenson, J. (2006). Language ability after early detection of permanent childhood hearing impairment. *New England Journal of Medicine*, *354*, 2131–2141. doi:10.1056/NEJMoa054915
- Lee, Y-M., Kim, L-S., Jeong, S-W., Kim, J-S., & Chung, S-H. (2010). Performance of children with mental retardation after cochlear implantation: Speech perception, speech intelligibility, and language development. *Acta Otolaryngologica*, *130*, 924–934. doi:10.3109/00016480903518026
- Malandraki, G. A., & Okalidou, A. (2007). The application of PECS in a deaf child with autism: A case study. *Focus on Autism and Other Developmental Disabilities*, *22*, 23–32. doi:10.1177/10883576070220010301
- Meinzen-Derr, J., Wiley, S., Grether, S., & Choo, D. I. (2010). Language performance in children with cochlear implants and additional disabilities. *The Laryngoscope*, *120*, 405–413. doi:10.1002/lary.20728
- Meinzen-Derr, J., Wiley, S., Grether, S., & Choo, D. I. (2011). Children with cochlear implants and developmental disabilities: A language skills study with developmentally matched hearing peers. *Research in Developmental Disabilities*, *32*, 757–767. doi:10.1016/j.ridd.2010.11.004
- Picard, M. (2004). Children with permanent hearing loss and associated disabilities: Revisiting current epidemiological data and causes of deafness. *The Volta Review*, *104*, 221–236.
- Pyman, B., Blamey, P., Lacy, P., Clark, G., & Dowell, R. (2000). The development of speech perception in children using cochlear implants: Effects of etiologic factors and delayed milestones. *The American Journal of Otolaryngology*, *21*, 57–61.
- R Development Core Team (2011). R: A language and environment for statistical computing (Version 2.13.1). Retrieved from <http://www.r-project.org/>
- Volden, J., Smith, I. M., Szatmari, P., Bryson, S., Fombonne, E., Miranda, P., ... Thompson, A. (2011). Using the Preschool Language Scale, Fourth Edition to characterize language in preschoolers with Autism Spectrum Disorders. *American Journal of Speech Language Pathology*, *20*, 200–208. doi:10.1044/1058-0360(2011/10-0035)
- Waltzman, S. B., Cohen, N. L., Gomolin, R. H., Green, J. E., Shapiro, W. H., Hoffman, R. A., & Roland, J. T. (1997). Open-set speech perception in congenitally deaf children using cochlear implants. *The American Journal of Otolaryngology*, *18*, 342–349.
- Waltzman, S. B., Scalchunes, V., & Cohen, N. L. (2000). Performance of multiply handicapped children using cochlear implants. *The American Journal of Otolaryngology*, *21*, 329–335.
- Wiley, S., Meinzen-Derr, J., & Choo, D. (2008). Auditory skills development among children with developmental delays and cochlear implants. *Annals of Otolaryngology, Rhinology & Laryngology*, *117*, 711–718.
- Yang, H-M., Lin, C-Y., Chen, Y-J., & Wu, J-L. (2004). The auditory performance in children using cochlear implants: Effects of mental function. *International Journal of Pediatric Otorhinolaryngology*, *68*, 1185–1188. doi:10.1016/j.ijporl.2004.04.011
- Yoshinaga-Itano, C. (1998). Early speech development in children who are deaf or hard of hearing: Interrelationships with language and hearing. *The Volta Review*, *100*, 181–211.
- Zimmerman, I. L., Steiner, V. G., & Pond, R. E. (2002). *Preschool Language Scale* (4th Ed.). San Antonio, TX: The Psychological Corporation.
- Zimmerman-Phillips, S., Robbins, A. M., & Osberger, M. J. (2000). Assessing cochlear implant benefit in very young children. *Annals of Otolaryngology, Rhinology & Laryngology*, *109*, 42–43.