

The NAL Study on Longitudinal Outcomes of Hearing-Impaired Children:
interim findings on language of early and later-identified children at 6
months after hearing aid fitting

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About one to two children in every thousand are fitted with hearing aids or cochlear implants by three years of age for a permanent hearing loss (Fortnum, Summerfield, Marshall, Davis, and Bamford, 2001; Prieve and Stevens, 2000; Ching, Oong and van Wanrooy, 2006). As congenital hearing loss impacts negatively on the communicative, educational and social developmental outcomes of children, the cost to society for providing health and educational care for these children is substantial (Downs, 1997; Davis, et al. 1997; Mohr, et al. 2000; Access Economic Report, 2006). With advances in electrophysiological testing that enable newborn hearing screening to be reliable and efficient, it has become possible to potentially alleviate the impact of hearing loss on children's development by early detection and intervention. A driving force for universal newborn hearing screening (UNHS) has been provided by studies that established an association between identification before six months of age and improved results in language at 3 years of age (e.g. Apuzzo and Yoshinaga-Itano, 1995; Yoshinaga-Itano, Sedey, Coulter and Mehr, 1998). Despite the frequent citing of these studies as evidence in support of the effectiveness of early identification in improving outcomes, a systematic literature review of the United States Preventive Services Task Force (USPSTF) in 2001 (USPSTF, 2001; Thompson, McPhillips, Davis, Lieu, Homer et al, 2001) concluded that "the evidence is insufficient to recommend for or against routine screening of newborns for hearing loss during the postpartum hospitalisation". They considered that the "evidence to determine whether earlier treatment resulting from screening leads to clinically important improvement in speech and language ... is inconclusive because of the design limitations of existing studies".

Since the review, several published studies have revealed information that diverged from the previous studies on the effectiveness of early identification in improving language outcomes. The dissimilarity in cohort compositions, evaluation age, evaluation instruments and factors that confounded the respective studies explains in part the differences in findings (see Table 1 for a summary).

Table 1 about here

Generally, program-based studies that assessed children at an early age indicated a statistically significant association between age of identification and language abilities. Seven of these studies included children enrolled in the Colorado Home Intervention Program (Apuzzo and Yoshinaga-Itano, 1995; Yoshinaga-Itano and Apuzzo, 1998a; Yoshinaga-Itano and Apuzzo, 1998b; Yoshinaga-Itano et al. 1998; Yoshinaga-Itano, Coulter and Thomson, 2000; Mayne, Yoshinaga-Itano, Sedey and Carey, 2000a; Mayne, Yoshinaga-Itano and Sedey, 2000b), one included children attending an early intervention program in Nebraska (Moeller, 2000), and another included children enrolled in a program in Washington (Calderon and Naidu, 2000). In these studies, children who remained in the program at the time of the study or for whom there were available data were included, and there was no information on attrition or follow-up rates. All these studies were based on unblinded assessments, with several relying predominately on parental reports of language abilities. Yoshinaga-Itano et al (1998) showed that children identified before age 6 months had higher language quotients at 3 years of age than children identified later, but found no significant difference in language performance for children identified between ages 6 to 34 months or interaction between severity of hearing loss and age of intervention.

Moeller (2000) reported that children enrolled prior to age 11 months had receptive vocabulary and reasoning scores within normal range at age 5 years, whereas those enrolled later had lower scores. Calderon and Naidu (2000) reported that children who enrolled before age 2 years had better language outcomes at age 3 years than those who enrolled later. All of these studies indicated, in principle, the benefits of early intervention. However, they also raise uncertainties. First, they suggest that there is no difference in language outcomes between children who first received their hearing aids at 7 months (Colorado Program studies) compared to those who first received their hearing aids when several years old. Second, there is no evidence as to whether outcomes are affected by when amplification is provided within the first 3, 6, or 9 months of life. Third, there is no evidence of an interaction between the degree of hearing loss and the timing of intervention. There is likely to be some interaction, as in the extreme case, no intervention is needed for children with normal hearing to obtain normal outcomes, on average.

Unlike the previous studies, several population-based cohort studies did not provide strong support for early intervention. A study in Australia (Wake, Hughes, Poulakis, Collins and Rickards, 2004) showed that severity of hearing loss, rather than age of diagnosis, correlated with language scores of children at 8 years of age. Although the study provided a comprehensive documentation of children's outcomes, the sample did not allow for an adequate evaluation of the effect of age of intervention because only 11 of the 89 children in the sample received their first hearing aids before age 6 months. These children did not demonstrate superior speech, language and reading outcomes to the remaining children who were first fitted between 7 months and

4.5 years. The discrepant findings may be related to differences in intervention and some unknown differences in the samples studied, such as presumably higher incidences of risk factors and additional disabilities at birth for those identified early. In contrast, a controlled trial of UNHS in the United Kingdom of 120 children (Kennedy, et al. 2006) found that at 8 years of age, children who were screened and diagnosed prior to age 9 months had better receptive and expressive language skills than children who were diagnosed later. The speech production skills, however, did not differ significantly between the two groups. It was recognised that despite early diagnosis by 9 months of age, about half of the screened group did not receive intervention until after the age of 18 months (Kennedy, McCann, Campbell, Kimm, and Thornton, 2005). This trial of UNHS (Kennedy et al, 2006) was conducted before England commenced its Audiology Modernisation project (Department of Health Report, 2004). Vastly improved services and devices could alter outcomes differentially for those exposed and not exposed to UNHS, though it is hard to predict in which direction. More recently, an observational study that included 65 children from three auditory-verbal programs in Canada (Fitzpatrick, Durieux-Smith, Eriks-Brophy, Olds and Gaines, 2007) found that the oral communication development of 26 children who were screened and identified before 12 months of age was not superior to that of 39 unscreened and later-identified children. In the screened group, only 15 children were identified before 6 months of age. No significant relation between age of identification and any measure of oral language skills was found.

As outlined in Table 1 and published in extensive literature, factors including family involvement in intervention, maternal-infant interaction, severity of hearing

loss, non-verbal intelligence, maternal education level, socio-economic status, mode of communication, educational placement, presence of additional disabilities, device and etiology have been linked to children's outcomes. Examination of the effect of age of intervention in conjunction with multiple influences on outcomes and their interactions necessitates a large enough sample size to support the statistical analyses. Not only did most previous studies lack the sample size for sufficient power in analyses or used analysis methods that did not account for multiple confounders, they also neglected investigations of the amplification and intervention characteristics, both of which are expected to influence outcomes of children with hearing loss (Moeller, Tomblin, Yoshinaga-Itano, Connor and Jerger, 2007).

The real-ear amplification provided to children after diagnosis is an important but as yet unexplored factor. Despite consensus guidelines on fitting amplification to infants, no study has directly examined the requirements of infants and how to determine whether the amplification provided is optimal for speech and language development. While it is generally accepted that a prescriptive procedure should be used (Joint Committee on Infant Hearing, 2000), there is no evidence on which of the two widely used prescriptions, NAL-NL1 (Dillon 1999) or DSL[i/o] (Seewald et al. 1997; Scollie et al. 2005), is more effective. The DSL procedure provides more low-frequency gain for flat hearing loss and more high-frequency gain for sloping hearing loss, compared to the NAL procedure. The DSL procedure also prescribes much more overall gain than the NAL prescription. In 2001, the National Acoustic Laboratories collaborated with the University of Western Ontario (funded by the Oticon Foundation) to evaluate the two prescriptions for school-aged children with moderate hearing loss.

The study found that the choice of prescription has little consequence in speech perception or loudness rating by school-aged children, despite differences in overall gain (Seewald et al. 2002; Ching, 2006). Nevertheless, the gain difference may be important for development in speech and language skills in the short and longer term when amplification is provided during the first few months of life, especially when the hearing loss is severe. Early fitting has to be guided by evidence as to how hearing aids should be prescribed for infants and the adequacy of audibility provided by the respective prescriptions for speech and language development. Amplification characteristics, in addition to age of fitting and use of devices, would be expected to influence children's outcomes.

Intervention characteristics are also likely to affect outcomes. Some researchers suggested that mode of communication used in intervention did not impact on early language development (Yoshinaga-Itano et al. 1998; Mayne et al. 2000a, 2000b) whereas other researchers maintained that children from oral programs achieved higher expressive language scores and better spoken language than children from total communication programs (Musselman, Wilson and Lindsay, 1989; Moog and Geers, 2003). The latter is not surprising, as children who remained in oral programs probably did so because of their superior spoken language abilities. There is also some evidence that linked the quality of intervention to development of age-appropriate language performance (Nitttrouer and Burton, 2003). Clearly, prospective studies that are population-based rather than program-based are needed to identify the child and family characteristics associated with enrolment in particular intervention programs and with changes in intervention programs over time, and to better understand the role of

specific intervention features, in addition to other factors, that influence individual differences and long-term outcomes of early- and later-identified children.

Even though there is ample evidence on the negative impact of childhood hearing impairment on development of language and literacy (see Moeller, Tomblin, Yoshinaga-Itano, Connor and Jerger, 2007 for a summary), perceptual processing (see Jerger, 2007 for a summary), and psycho-social skills (see Moeller, 2007 for a summary) in children, little is known about its impact on children's use of spatial and interaural difference cues for speech perception in noise. This ability impinges on children's learning in noisy environments such as classrooms. Whereas normal-hearing children demonstrated the ability to use spatial cues in sound source segregation by the age of 3 years (Garadat and Litovsky, 2007), there is some evidence suggesting that many hearing-impaired children are not able to do so (Ching et al, in preparation). The extent to which this deficit may be lessened by early intervention remains to be investigated.

The lack of strong evidence to support the effectiveness of UNHS in improving long-term outcomes puts even well-accepted programs at risk of their funding providers (Bailey, Bower, Krishnaswamy and Coates, 2002), and leaves the discrepant findings in previous studies unresolved. As each of the previous studies provided a snapshot of children's outcomes at a point in time, whether an advantage of early intervention observed at an early age may be dissipated in later years of life is not clear. Without longitudinal follow-through, the apparently contrastive findings cannot be resolved, the relation between early performance and later outcomes cannot be quantified, and the longer term efficacy of UNHS remains unproven. As summarised by the USPSTF in

2001, “there are no prospective controlled studies that directly examine whether newborn hearing screening and earlier intervention result in improved speech, language or educational development.” Because UNHS coverage is already well over 90% in the US (Marge and Marge, 2005) and the UK (Uus and Bamford, 2006), it is no longer possible to recruit an adequate sample for a comparative study of early- and later-identified children in those countries. In Australia, the National Acoustic Laboratories took advantage of a narrow time window during which UNHS coverage was markedly different between states (ranging from 30% to well over 90% across states, Leigh, 2006) to conduct a prospective population-based study to examine whether early intervention leads to improved long-term outcomes; to quantify the influence of multiple factors, including age of intervention, on a range of outcomes; and to describe the etiologic bases of hearing loss in children (www.outcomes.nal.gov.au). Because all children with hearing loss in Australia are referred to Australian Hearing (AH), a government-funded organization that provides standardized pediatric services with quality procedures for free to all children under the age of 21 years, the same approaches to assessment and amplification are followed from the time of diagnosis for all children in all states. The national client database maintained by AH also enables the sampling frame of the present study to be consistent across states.

This study is important for four reasons:

1. Improvement in long-term outcomes is the underlying reason for newborn hearing screening programs. There are as yet no prospective studies that examine the long-term speech, language, functional and psychosocial outcomes of large numbers of early and late-identified children.

2. Outcomes studies are essential for estimating the negative impact of hearing loss, and defining how much of the negative impact may be alleviated by advances in device and non-device intervention. There is a clear need for an evidence base to guide professionals in fitting amplification and in their counsel to parents regarding choice/change of device and non-device intervention.

3. Investigations of the etiology of early and later-identified children will increase knowledge on the epidemiology of hearing loss. An understanding of the causes of early and late-onset pre-lingual deafness will contribute to better strategies for screening and monitoring hearing of newborns.

4. Longitudinal studies help to understand why children do well or poorly over time. In trying to understand better the variability in short and longer-term outcomes of early and later-identified children, we incorporate novel factors including the etiology of hearing loss, accuracy of early amplification, real-ear aided gain characteristics for children with hearing aids, the presence/absence of auditory neuropathy and cortical responses with amplification or implantation.

We are now faced with a new generation of children many of whom have access to early identification, early intervention and advanced hearing technologies. For practical purposes of optimising audiological and intervention strategies to best support these children, we need to strengthen the evidence base to better understand the effect of multiple factors and their interaction on individual children's outcomes. Without doubt, an increased understanding of the influence of early auditory perception and auditory experience on various aspects of a child's development is of theoretical interest. Not only will the evidence complement current knowledge on the role of early

sensory experience on the organisation of sensory information for language (Sharma, Gilley, Dorman and Baldwin, 2007; Ponton and Eggermont, 2001; Shepherd and Hardie, 2001), it will also have practical implications on whether additional factors need to be modified to improve long-term outcomes of children. In an environment where health resources are highly competitive, empirical support for the efficacy of universal newborn hearing screening in improving long-term outcomes of children with hearing impairment is crucial in indicating the need for UNHS to be universal and to be maintained in the long term.

Our study aimed to 1) describe the levels of performance on speech, language, functional and psychosocial skills and educational attainment of early- and late-identified children; 2) identify the factors including age of intervention, etiology, nature and amount of intervention, device type and characteristics, and other family- and child-related factors that influence outcomes; 3) describe the etiology of hearing loss by determining the presence of congenital CMV infections, the presence of *GJB2* and *SLC26A4* mutations and the mtDNA A1555G mutation; and 4) relate early performance to later outcomes.

The study is currently in its second year. In this paper, we report auditory comprehension and expressive communication skills measured by using the PLS-4 (Zimmerman, Steiner and Pond, 2002) of 123 children in an interim investigation of the effect of age of fitting, severity of hearing loss and hearing aid prescription on language development at 6 months after initial fitting.

Method

A longitudinal prospective study involving multiple assessment intervals over 5 years is used to examine a range of child characteristics, audiological and device related factors, family characteristics and intervention-related factors on child outcomes, and to track changes over time. A randomised controlled trial is used to examine the effect of hearing aid prescription.

Participants

All children (and their families) who present at Australian Hearing for first fitting under the age of 3 years in paediatric hearing centres in New South Wales, Victoria and Queensland are invited to participate in this longitudinal study. Our target sample size is 400, determined from considerations to support statistical analyses. Firstly, to ensure an adequate ratio of data points to unknown coefficients in multiple regression analyses, 400 data points at a measurement interval and 30 raw predictors give a very favorable ratio of 13:1 for the initial analysis. This ratio will reduce by an amount determined by the number of predictors for which we find it necessary to non-linearly transform the predictor variable, but will increase as predictors found to be unrelated to the outcomes are deleted. We will not allow the ratio to drop below 7:1, and we anticipate it being considerably higher. Secondly, we anticipate performing analyses of variance on outcomes for early- versus late-identified children. We can detect an effect size of 0.25 within-group standard deviations (generally considered to be a small effect) with a power of 80%, for an alpha level of 0.05, if we have 200 children in each group. It is worth noting that as the outcomes data will have been corrected for the effects of all other predictor variables, the within-group standard deviation that applies for this power analysis is likely to be considerably smaller than

the raw standard deviation that would otherwise apply. A total of 400 children therefore give us a design that is very sensitive to the effect of early intervention.

Key outcome areas

The key outcome areas include communicative function, educational attainment, and social competence. Within the area of communicative function, measures include binaural speech perception, auditory comprehension, expressive communication skills, articulation and phonological development, speech intelligibility and a global measure of functional performance in real life. Within the educational area, reading, writing and numeracy are the major components. Mediating variables for measurement at an early age include letter-naming, phonological awareness, phonological memory, vocabulary size and non-word repetition skills. Reading words, reading comprehension, vocabulary size, writing and numeracy are assessed when children reach an age appropriate for these tests. The grade level of the children and their results from state-wide assessments are recorded at appropriate ages. Within the social competence domain, we assess development of motor skills, social-emotional skills, self-help ability, general development, strength and difficulties and executive functioning ability. In all domains, a combination of child-administered tests and subjective reports from parents and teachers are used, and interactions between outcomes in different domains are examined.

Key predictors

For hearing-loss related factors, objective measures of hearing acuity using electrophysiological and behavioural methods, acoustics of the real ear, electro-acoustic measurements of hearing aids, and assessments of aided cortical responses are used.

Children are randomly assigned to either the NAL or the DSL prescription at first fitting and the quality of audiological intervention is controlled by adherence to consistent protocols and procedures across all hearing service centres. Strict criteria for matching hearing aids to prescriptive targets are observed in all fittings. Changes in hearing sensitivity and real-ear amplification characteristics are recorded at all evaluation intervals. For children who subsequently receive cochlear implants, the age at implantation and implant parameters are recorded. Also, information about whether the hearing loss is progressive, fluctuating, and whether auditory neuropathy is present are recorded.

Other child-related factors including ethnicity, gender, presence or absence of additional disabilities and family-related factors including socio-economic status, maternal education and language used at home are recorded. In addition, information about the amount and type of intervention as well as family involvement in intervention are recorded. The interplay of these characteristics, their changes over time and their effects on outcomes will be examined.

Procedure overview

The children's outcomes are assessed by using standardized tests and validated measures that combine child-administered tests with report-type questionnaires. Over a period of 5 years, children are assessed at 6 and 12-months post-fitting/post-implantation, and at chronological ages of 3 and 5 years (we expect that a second wave of the study will examine the children's outcomes at 8 and 11 years of age). Depending on the age at first fitting and age at study enrolment, children are assessed for a minimum of 3 intervals and a maximum of 4 intervals. All measures that are

appropriate to the age of the child at each interval are administered. Information about demographic characteristics, family characteristics, and intervention-related characteristics are collected via questionnaires, interviews or retrieved from service providers' database at each evaluation interval.

All evaluations are blinded to the age of intervention and hearing aid characteristics of children. Each child is assessed by a qualified speech pathologist and/or audiologist not directly involved in providing habilitation service to the child, to the extent possible.

Results

Table 2 shows the age of fitting for 123 children and Table 3 shows the auditory comprehension and expressive communication standard scores of the children.

Tables 2 and 3 about here

Because hearing threshold information was so far retrieved from audiological files for only 95 children, the following analyses were based on data from these children.

Analysis of variance with the PLS-4 subscale standard scores as dependent variables, with age of fitting (< 6 months vs ≥ 6 months) and prescription (NAL vs DSL) as categorical within-group factors, and averaged three-frequency hearing loss as a continuous covariate indicated that the main effect of age of fitting was highly significant ($p = 0.0004$), hearing loss was significant ($p = 0.006$), but prescription was not significant ($p = 0.1$). The same results apply to both auditory comprehension and expressive communication. Figure 1 shows the mean scores for children who first received amplification before 6 months and those at or after 6 months of age.

Figure 1 about here

Inspection of individual data also suggested that earlier fitting was associated with higher scores, and the effect was nonlinear with larger differences occurring during the first 10 months than between 11 and 36 months. A logarithmic transform of the age at fitting was significantly correlated with auditory comprehension ($r = -0.25$, $p < 0.05$) and expressive communication ($r = -0.31$, $p < 0.05$). Further examination of the language scores in relation to hearing loss suggested that a quadratic equation characterised the relation between hearing loss and auditory comprehension ($r = -0.33$, $p < 0.05$). No significant correlation was found between hearing loss and expressive communication ($r = -0.13$, $p > 0.05$).

To examine the effect of age of fitting as a continuous variable together with co-varying factors, we used multi-nonlinear stepwise regression analyses with auditory comprehension scores as a dependent variable. After accounting for hearing loss (Beta = -0.39, $p = 0.00006$), age of fitting was significant (Beta = -0.29, $p = 0.003$). The analysis was repeated for expressive communication scores, with results indicating that the choice of prescription affected the scores ($p = 0.03$) and the severity of hearing loss almost reached significance ($p = 0.05$). After accounting for the effect of prescription, age of fitting was significant (Beta = -0.33, $p = 0.001$) suggesting that the significant effect of age of fitting was not due to the co-varying prescription. Figure 2 presents the adjusted scores in relation to age at fitting.

Figure 2 about here

Although the regression line of best fit suggests that children who received hearing aids prior to 8 months of age were associated with auditory comprehension scores within 1 SD of the normative mean (100 ± 15), this finding is only limited to the

investigation of the effect of age of fitting on language development as measured by the PLS-4, after adjusting for severity of hearing loss and prescription for children at 6 months after initial fitting. When all data become available, it will be possible to consider other factors that may affect outcomes, both in the short term and in the longer term.

For 78 children, we have also obtained language data at two assessment intervals. A preliminary estimate of rate of growth was carried out by dividing the increase in language-equivalent age by the increase in chronological age. A growth rate of one denotes development at the normal rate, irrespective of the actual ability at the start of the period. For children who received first fitting before 8 months of age, the mean rate was 0.8 (SD = 0.5, range = 0 to 2.2, n = 56) for development of auditory comprehension, and 1.1 (SD = 0.6, Range = 0.1 to 2.8, n = 57) for development of expressive communication. For children who received later fitting, the mean rates were 1.1 (SD = 0.8, range = 0 to 3.2, n = 21) and 0.9 (SD = 0.6, range = 0 to 2.0, n = 21) for development of auditory comprehension and expressive communication respectively. The inter-subject variability in growth rate is large possibly because of measurement errors in having only two data points over a relatively short time interval. When all longitudinal data become available, we will be able to estimate the growth rate more accurately.

Discussion

The preliminary analyses of expressive and receptive language ability at 6 months after fitting suggest that children who received intervention before 8 months of age developed auditory comprehension at age-appropriate levels whereas children who

received later intervention revealed deficits in these skills compared to their normal-hearing peers. The benefit due to early intervention is consistent with findings from 39 children who received a cochlear implant before or after 12 months of age and who were assessed at 6 months after implantation (Ching et al, accepted). Even though the present data strongly suggest that more normal language acquisition is achieved by children who were identified earlier, it must be emphasised that the effect of age of intervention on long-term outcomes is still open to question. As multiple factors are potentially important in affecting performance, these will have to be examined at a later stage of the study when data from all subjects are available. Also, it is not clear whether near-normal performance measured early in life may be maintained through to school-age children. However, from 6 months post-fitting to 12 months post-fitting, the mean rate of language acquisition is the same for early- and late-identified children, suggesting that early differences in performance are being maintained. Finally, the impact of age of intervention on long-term speech production, speech perception, language, educational and social development remains to be examined when longitudinal data and data from other measures of the same children become available.

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Figure legend.

Figure 1. Mean Auditory Comprehension (AC, filled circles) and Expressive Communication (EC, open squares) subscale scores for children who received hearing aid fitting before 6 months of age ($< 6m$) and those at or after 6 months of age ($\geq 6m$). The left panel shows data of children whose hearing aids were fitted according to the NAL prescription (Pres: NAL) and the right panel shows data of children whose hearing aids were fitted using the DSL prescription (Pres: DSL).

Figure 2. Adjusted Auditory Comprehension (AC, filled circles) and Expressive Communication (EC, open squares) subscale scores for children as a function of age at fitting. The solid line shows the regression line for AC, and the broken line shows the regression line for EC scores.

Figure 1.

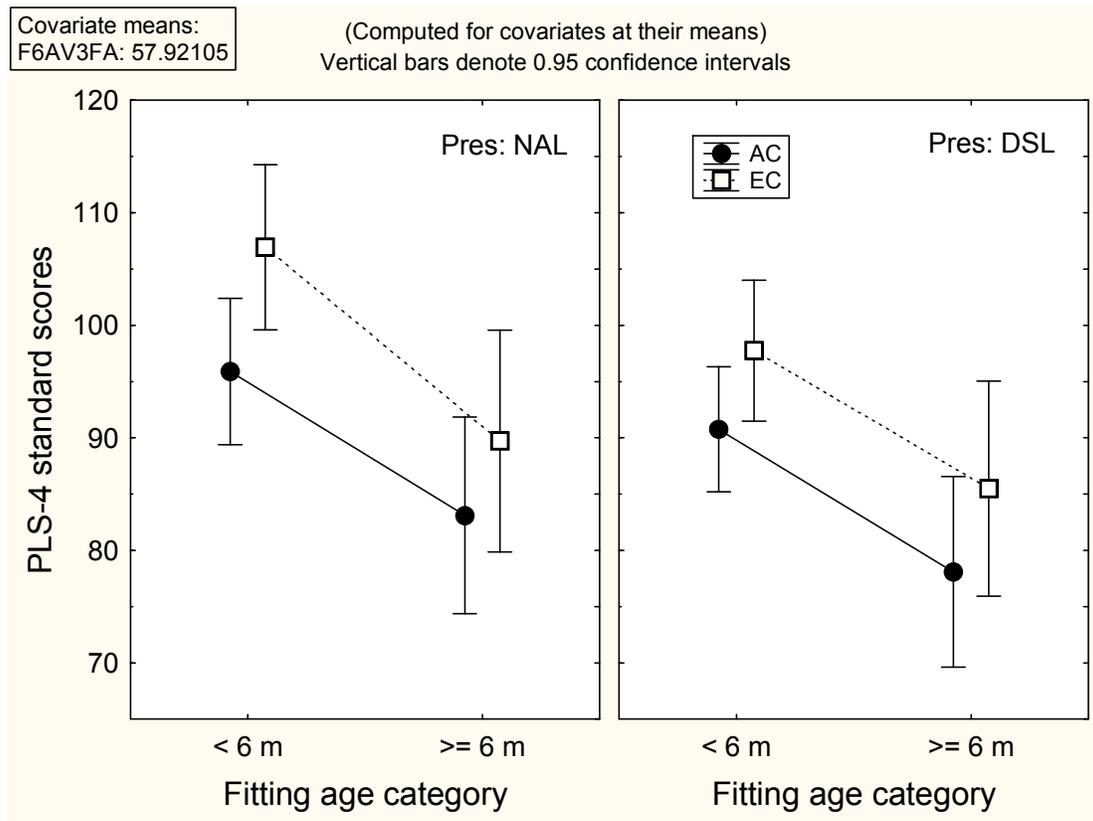


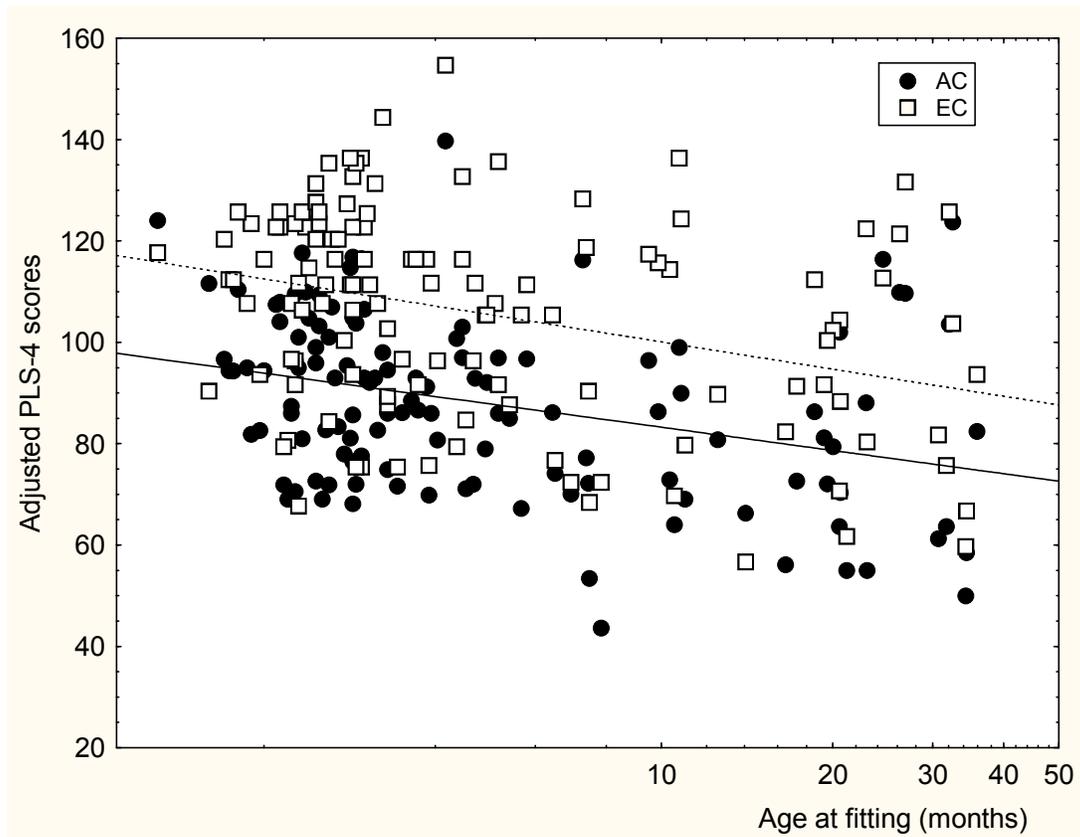
Figure 2.

Table 2. Age at first fitting for 123 children.

State	n	Age at first fitting (months)			
		Median	Mean	25 th percentile	75 th percentile
New South Wales	40	2.8	4.5	2.3	4.2
Queensland	45	4.0	10.0	2.9	17.3
Victoria	38	5.2	8.6	2.9	10.5
Total	123	3.5	7.8	2.5	9.5

Table 3. Mean standard scores for the Auditory Comprehension and Expressive Communication subscales score of the PLS-4 at 6 months after fitting.

		Auditory comprehension	Expressive Communication
Fitted before 6 months of age (n = 84)	Mean	92.2	100.7
	SD	16.2	18.5
	Range	61.0 to 142.0	62.0 to 150.0
Fitted at or after 6 months of age (n = 39)	Mean	79.2	86.2
	SD	20.4	21.7
	Range	50.0 to 120.0	52.0 to 127.0

Table 1. Summary of studies reporting language outcomes of children.

STUDY	SUBJECT GROUPS	INCLUSION	MEASURE	AGE OF ASSESSMENT	ANALYSIS	RESULTS
Apuzzo & Yoshinaga-Itano, 1995	69 high-risk infants diagnosed at 2 to 25 months. Children enrolled in the Colorado Department of Health Home Intervention Program (CHIP).	No Severe cognitive delay (development quotient >60). Infants with data available on age of identification, age at testing, test scores and hearing loss category.	Parent report	Mean age of 40 months (range between 25 and 60 months)	Analyses of variance, adjusted for covariance of degree of hearing loss and cognitive ability. No multiple regression analysis to adjust for potential confounders.	Infants identified before 2 months had higher expressive language scores than those identified later.
Yoshinaga-Itano & Apuzzo, 1998a	40 high-risk infants: 15 identified before age 6 months, 25 after age 18 months). Children enrolled in the	No Severe cognitive delay. Infants with data available on age of identification, age of testing test scores and	Parent report	Mean age of 40 months (range between 25 and 60 months)	Adjusted for covariance of degree of hearing loss and cognitive ability. No multiple regression analysis	Children identified before 6 months had higher expressive and receptive language than those identified after 18 months.

	CHIP.	hearing loss category.			to adjust for potential confounders.	
Yoshinaga-Itano & Apuzzo, 1998b	82 infants: 34 identified before age 6 months via high-risk registry, 48 identified 7-18 months by usual care. Children enrolled in the CHIP.	No Severe cognitive delay. Infants with data available on age of identification, age of testing, test scores, gender, and hearing loss category,	Parent report	Mean age of 26 months, range between 19 and 36 months	Adjusted for covariance of cognitive ability and chronological age at testing. No multiple regression analyses to adjust for potential confounders.	Children identified before 6 months had better expressive and receptive language than those identified later. The earlier identified group also had better vocabulary than the later identified group.
Yoshinaga-Itano et al, 1998	150 children: 72 identified before 6 months, 78 identified after 6 months. Children enrolled in the CHIP.	Infants with data available.	Parent report	13 to 36 months	Adjusted for covariance of cognitive ability. Examined gender, maternal education level, mode of communication, additional disabilities singly in analyses of variance. No multiple regression analyses	Children identified before 6 months had higher receptive and expressive language quotients than later-identified children. No difference among four age-of-identification levels for children identified between 7 and 34 months. For children with low cognitive ability (cognitive quotient below 80), differences

					to adjust for potential confounders.	between early and later identified groups were not significantly different when either receptive or expressive language quotient was considered.
Yoshinaga-Itano et al, 2000	50 children: 25 born in a hospital with UNHS, 25 in a hospital without UNHS. Children enrolled in the CHIP.		Parent report	9 to 61 months	Pairs matched on age of testing, degree of hearing loss, cognitive ability.	Language development was within normal range for 56% of screened group, but only 24% of unscreened group
Mayne et al, 2000a	113 children, 54 identified by age 6 months, 59 after age 6 months. Children enrolled in the CHIP.		Parent report	Mean age of 31 months, ranged between 24 and 37 months	Regression analyses with child's age, age of identification, cognitive ability, additional disability force-entered in three blocks.	Child's age and age of identification accounted for 23%, cognitive ability accounted for 30% and the presence of additional disability accounted for 3% of variance in expressive vocabulary. Expressive vocabulary scores were higher with increased age, increased cognitive ability, identification by age 6

						months, and absence of additional disabilities.
Mayne et al, 2000b	168 children, 73% identified with hearing loss by age 6 months. Children enrolled in the CHIP.		Parent report	8 to 22 months	No multiple regression analyses to adjust for potential confounders.	Age of identification was not significantly correlated with receptive vocabulary ability.
Calderon & Naidu, 2000	80 children who graduated from the Early Childhood Home Instruction (ECHI) Program: 9 enrolled before 12 months of age, 39 enrolled between 12 to 24 months, 32 enrolled after 24 months.	Enrolled in the program between 1989 and 1994. No severe developmental delay or disabling condition as diagnosed medically.	Parent report	3 years, at program exit.	Adjusted for degree of hearing loss and baseline test levels obtained within 2 weeks of program entry.	Age at program entry explained 43.5% of variance in receptive language and 49% of variance in expressive language. Children who enrolled before age 2 years had better outcomes in receptive and expressive language than those enrolled after age 2 years.
Moeller 2000	112 children aged 5 years who graduated from the Diagnostic Early	Nonverbal IQ ≥ 70 , no evidence of major secondary	Receptive vocabulary, nonverbal reasoning	5 years	Multiple regression analyses adjusted for family involvement, degree of hearing	Age of enrolment accounted for 11.4% and family involvement accounted for 35% of variance in vocabulary

	Intervention Program (DIEP). Only 24 children were identified before age 11 months.	disabilities, Participated in the DIEP between 1981 and 1994.			loss, nonverbal IQ.	scores. Adjusted mean receptive vocabulary and reasoning scores were within normal range for children enrolled prior to 11 months, but were lower for later-identified children
Wake et al, 2004	89 children: 11 fitted before 6 months; 17 fitted between 6 and 12 months; 55 fitted between 12 months and 4.5 years.	Children born between 1991 and 1993 with congenital hearing loss, fitted with hearing aids by 4.5 years. No intellectual disability. English-speaking background. No serious medical condition	Child administered tests: CELF, PPVT	7 to 8 years	Adjusted for IQ.	Language and receptive vocabulary decreased with increased severity. No significant correlation between language outcomes and age at diagnosis.
Kennedy et al, 2006	120 children born between 1992 and 1997 :	No known postnatal cause of bilateral permanent	Parent report, test for reception of grammar,	Mean age of 7.9 years, ranged	Adjusted for degree of hearing loss, Maternal education	Children diagnosed by 9 months had better language skills than those diagnosed

	57 diagnosed by 9 months, 63 diagnosed later (up to 6years).	hearing impairment (e.g. bacterial meningitis).	receptive vocabulary, narrative speech.	between 5.4 and 11.7 years		after 9 months. Speech production was not significantly different between the early and later identified groups.
Fitzpatrick et al, 2007	65 children born between 1998 and 2003 enrolled in 3 auditory-verbal programs: 26 screened and identified before 12 months of age (15 before 6 months), 39 identified later.	- Age < 5 years; - Bilateral hearing loss > 20 dB HL - Congenital or onset < 6 months. - Consistent use of hearing device and enrolled in auditory-verbal program - Intervention in English - No complex medical and developmental disabilities.	Child administered measures included PLS-4, PPVT, and GFTA-2; Parent report based on CDI.	Ranged between 43 and 57 months	Multiple linear regression using age at diagnosis, degree of hearing loss, family education and self-help quotient as predictor variables and child-administered measures as dependent variables (based on data from 43 children)	Age of diagnosis was not associated with improved outcomes for any measure. No significant difference in oral communication development between children identified before 12 months of age and those identified later. No significant difference between screened and unscreened groups.