Age at Intervention for Permanent Hearing Loss and 5-year Language Outcomes

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Abbreviations: Permanent childhood hearing loss – PCHL; Universal newborn hearing screening - UNHS; 95% confidence interval – 95% CI; standard deviation – SD; Australian Hearing – AH; New South Wales – NSW; Victoria – VIC; Queensland – QLD; hearing aid – HA; cochlear implant – CI; Longitudinal Outcomes of Children with Hearing Impairment – LOCHI; better ear four-frequency average hearing level – BE4FA; kilo-Hertz – kHz; decibel hearing level – dB HL; socio-economic status – SES; Preschool Language Scale v.4 – PLS-4;

Peabody Picture Vocabulary Test v.4 – PPVT-4; Expressive Vocabulary Test v.2 – EVT-2; Parents' Evaluation of Aural/oral performance of Children - PEACH

Table of Contents Summary

The benefit of early age at intervention for language development of children with permanent childhood hearing loss (PCHL) increases with the degree of hearing loss.

What's Known on This Subject:
Universal newborn hearing screening is associated with early detection of permanent childhood hearing loss and superior language outcomes. However, the effect of variation of age at intervention and in severity of hearing loss on these associations is unknown.

What This Study Adds:
Compared with the same intervention at 24 months, 5-year language scores were 0.8 and 0.55 SD higher in children with 70 and 50 dB HL, respectively, with amplification at 3 months and 1.4 SD higher with implantation at 6 months.

Contributor's Statement

Dr. Ching conceptualized and designed the study. She took overall responsibility for all aspects of the study, drafted the initial manuscript and revised the manuscript for submission.

Dr. Dillon consulted in the design of the study, guided the analysis of data and interpretation of data, and reviewed the manuscript.

Ms. Button was responsible for acquisition of the evaluation data, provided guidance regarding data collection, and reviewed the manuscript.

Mr. Seeto was responsible for the analysis of data, in consultation with Dr. Dillon and Dr. Ching; and reviewed the manuscript.

Ms. Van Buynder was responsible for acquisition of the audiological data, provided guidance regarding data collection, and reviewed the manuscript.

Ms. Marnane assisted with management of the project, coordinated acquisition of demographic data and collation of data, and reviewed the manuscript.

Drs. Cupples and Leigh consulted in the design of the study, provided guidance regarding the data collection instruments, and reviewed and revised the manuscript.

All authors approved the final manuscript as submitted.

ABSTRACT

OBJECTIVE: Universal newborn hearing screening has been implemented to detect permanent childhood hearing loss (PCHL) early, with the ultimate goal of improving outcomes through early treatment. However, there is disagreement between studies on the size of this benefit, and in some cases whether it is significantly different from zero. There have been No studies of sufficient size in which researchers have determined reliably whether the effect varies with degree of PCHL. We aimed to explore how intervention timing influences 5-year language in children with PCHL.

METHODS: Via a prospective study of 350 children, we used standard multiple regression analyses to investigate the effect of age at intervention or hearing screening on language outcomes; after allowing for the effects of nonverbal IQ, degree of PCHL, sex, birth weight, maternal education, additional disabilities, and communication mode.

RESULTS: The benefit of early intervention for language development increased as hearing loss increased. Children whose amplification started at age 24 months had poorer language than those whose amplification started at 3 months. The difference was larger for 70 dB HL (-11.8 score points; 95% confidence interval [CI]: -18.7 to -4.8) than for 50 dB HL (-6.8; 95% CI: -10.8 to -2.8). Children who received cochlear implants at 24 months had poorer language than those implanted at 6 months (-21.4; 95% CI: -33.8 to -9.0). There was no significant effect of screening on outcomes.

CONCLUSIONS: Early intervention improves language outcomes, thereby lending support to streamlining clinical pathways to ensure early amplification and cochlear implantation after diagnosis.

INTRODUCTION

Bilateral permanent childhood hearing loss (PCHL), which occurs in 1 to 2 per 1000 newborns,[1,2] has major negative impacts on children's development[3-5] thereby incurring high societal costs.[6] Therefore, several program-based studies[7-9] that showed a retrospective association between earlier identification and better preschool language, despite methodological limitations,[10] have driven widespread implementation of universal newborn hearing screening (UNHS). Although the 2001 US Preventive Services Task Force (USPSTF) systematic review found good evidence that UNHS leads to earlier identification and treatment, it concluded that 'evidence to determine whether earlier treatment resulting from screening leads to clinically important improvement in speech and language is … inconclusive'.

Subsequent to the review, several population-based studies have examined the relationship of screening or early intervention with child outcomes. In the Wessex quasi-experimental study, Kennedy et al[11] compared language of 7-8 year-old English children with PCHL who were born in the mid-1990s in areas with and without UNHS. Exposure to UNHS had a significant positive effect on receptive language, but not expressive language or speech production. Confirmation of PCHL by age 9 months (comprising 67% of screened and 27% of unscreened children) was associated with higher scores for receptive and expressive language, but not for speech production. As the study was conducted before modernization of English audiological services, and confirmation by age 9 months would be considered 'late' relative to current recommendations,[12] the impact of early intervention remains to be further investigated.

In contrast, Wake et al[13] reported on a community-based cohort of children with PCHL who were exposed to risk-based newborn screening in Australia, and found no significant relationship between age at diagnosis and speech and language outcomes at 7-8 years old. The small number of children diagnosed before 6 months of age (n = 11) may provide inadequate power to evaluate an age effect. In a Canadian study[14] compared language abilities between screened and unscreened children aged 2 to 5 years, and did not find significant differences between groups. The finding may have been limited by the small sample size (n = 26 screened, 39 not screened).

In the Netherlands, Korver et al[15] compared language scores of Dutch children aged 3 to -5 years born in UNHS and non-UNHS regions; and found no significant difference between groups. The study did not adjust for hearing loss severity, which has been negatively associated with language outcomes in previous studies;[13,16] and relied on parent reports without direct assessments of children's abilities. The effect of early confirmation of PCHL on language outcomes was not examined.

The 2008 update of the USPSTF review[17] and a recent review by Pimperton and Kennedy[18] confirmed that there was good evidence that UNHS leads to earlier intervention, defined by cut-off points at 6 months[7] and 9 months,[11] and improves language outcomes. The benefit of UNHS was smaller than that of early identification, presumably because of the failure of screening programs to screen the entire target population or to proceed promptly to identification of true cases among children screening positive for PCHL.[18]

In A recent prospective study, the Longitudinal Outcomes of Children with Hearing Impairment (LOCHI) study, evaluated the effect of age at intervention on outcomes of a population-based cohort born between 2002 and 2007 in the states of New South Wales (NSW), Victoria (VIC) and Queensland (QLD) in Australia. All children with PCHL receive uniform post-diagnostic services administered by a national government-funded organization (Australian Hearing, AH) at no cost to families, but access to UNHS depended on whether state-funded UNHS was operating.[19] Through the service network, the LOCHI study enrolled children whose PCHL was identified through either UNHS or standard care. On average, better language at 3 years of age was significantly associated with earlier fitting of a cochlear implant (CI), but age at fitting of a hearing aid (HA) had a weak effect on outcomes.[16] Possibly, assessments at this young age might not provide sufficient scope for manifestation of the benefits of early intervention on language development.[14,20]

Although researchers who in previous studies have included sufficient observations have reported some benefits of early intervention for later language skills, their studies have not been powered to determine if the extent of the benefit varies with severity of PCHL. Here, we addressed this question by drawing on the 5-year language outcomes of the cohort in the LOCHI study.

We aimed to explore the following:

1. Effects of age at HA fitting by degree of hearing loss for children using HAs; and
2. Effects of age at CI activation for children using CIs.

We hypothesized that children who started amplification earlier would have better language, and the benefit would increase with hearing loss. Also, we hypothesized that children who received a CI earlier would have better language.

Our secondary aim was to examine the impact of UNHS on outcomes, separately for children using HAs or CIs. We hypothesized that, because access to UNHS decreases age at intervention (fitting of HA or CI), access to UNHS would improve outcomes.

METHODS

Study population

The participants were 350 children with PCHL and a comparison group of 120 children with normal hearing that was very similar to the children with PCHL with respect to age, sex, and socio-economic status (SES). In This study, we focused on the impact of intervention within the age range for which there is uncertainty about the effect of age of intervention, and we therefore excluded children whose device was fitted later than 3 years of age, and those whose long-term audiograms revealed a progressive hearing loss. The participants included children from the population-based cohort from three states in Australia who participated in the LOCHI study (see Introduction and Figure 1) and who had received by 3 years of age, a device of the same type (i.e. HA or CI) that they were using at age 5 years. This study was approved by the AH Human Research Ethics Committee.

Sampling

The AH national service network served as the sampling frame. All families of children who met inclusion criteria were presented with an information statement and consent form at a time as close to initial fitting of HAs as possible, in person or by post. Once written informed consent was obtained from parents, their child was assessed at several intervals.

Post-diagnostic follow-up for all children with PCHL, including this cohort, is performed by AH audiologists until the children reach 26 years of age. The service includes audiological assessment and fitting of HAs, and referrals for CI, medical and communication intervention, in accordance with the AH national protocol.[21]

Measures

Main outcomes measures[22-32] are summarized in Table 1. Standardized measures of language, vocabulary, letter knowledge, speech production and oral reading were directly administered to children at age 5-6 years, by speech pathologists who were blinded, as much as possible, to age at intervention and hearing loss severity. Parents reported on their child's functional auditory performance in real life, language, and social skills. All measures are widely used for assessing development of children with normal hearing or PCHL, with known validity and reliability. The same test instruments and protocols were administered across all sites.

Insert Table 1 about here

Information about age at HA fitting, age at CI activation, audiometric thresholds and hearing device were obtained from clinical records. The degree of PCHL at the time of evaluation was characterized in terms of better-ear four-frequency-average hearing loss (BE4FA, average of hearing thresholds at 0.5, 1, 2, and 4 kilo-Hertz [kHz]) in decibels hearing level (dB HL). Nonverbal IQ was assessed by a psychologist. Parents completed custom-designed questionnaires on demographic characteristics.

Potential *a priori* confounders were determined on the basis of current literature, including earlier findings at 3 years of age.[16] The variables included birth weight, sex, degree of hearing loss, nonverbal IQ, additional disabilities, maternal education (university degree, vocational training certificate, ≤12 years of formal schooling), SES (census-based Index of Relative Socio-Economic Advantage and Disadvantage for home postcode;[33] higher scores reflect greater advantage), and communication mode during educational intervention (oral vs combined [speech and sign]). These were used as predictors in regression analyses to maximally account for variance in scores so as to increase the sensitivity with which we can assess the impact of intervention timing or access to UNHS.

Statistical Analysis

The statistical analysis was done using R, version 3.0.2,[34] and Statistica, version 10.[35] All analyses used two-tailed tests, except where otherwise stated, with statistical significance set at $p < 0.05$.

Children unable to be directly assessed because of parent-reported or apparent disabilities or not using spoken English were assigned the poorest score observed for that test (receptive/expressive language: n = 33, 9.4%; vocabulary: n = 62, 17.7%; speech production: n = 47, 13.4%; reading: n = 66, 18.9%; letter name: n = 63, 18.0%; maths: n = 69, 19.7%). A multiple imputation method[36] was applied to handle missing values in variables, assuming that data were missing at random. The analysis was averaged over ten imputations.

Scores from 20 measures were aggregated using factor analysis to form a global language score. This approach was justified by an exploratory factor analysis showing that such measures all loaded onto a single underlying factor. The global score was scaled so that a normal population would have a mean of 100 and a standard deviation of 15. The use of a global score eliminates the disadvantages associated with performing multiple hypothesis testing on correlated data and increases reliability by reducing random measurement error through averaging across measures.

Outcomes by age at HA fitting and hearing loss severity for the HA group (Aim 1): We performed standard multiple linear regression analysis with the global score as the dependent variable. Predictors included age at initial HA fitting (log-transformed), degree of PCHL, nonverbal IQ, birthweight, and an interaction between age at HA fitting and degree of PCHL as continuous variables; and sex, additional disabilities, maternal education, SES, and communication mode as categorical variables.

Outcomes by age at cochlear implantation for the CI group (Aim 2): We performed multiple linear regression analysis, using the global score as dependent variable, with predictor variables as for the HA group, but we performed it with age at HA fitting replaced by age at activation of the first CI (log-transformed), and without the interaction between age at HA fitting and degree of PCHL.

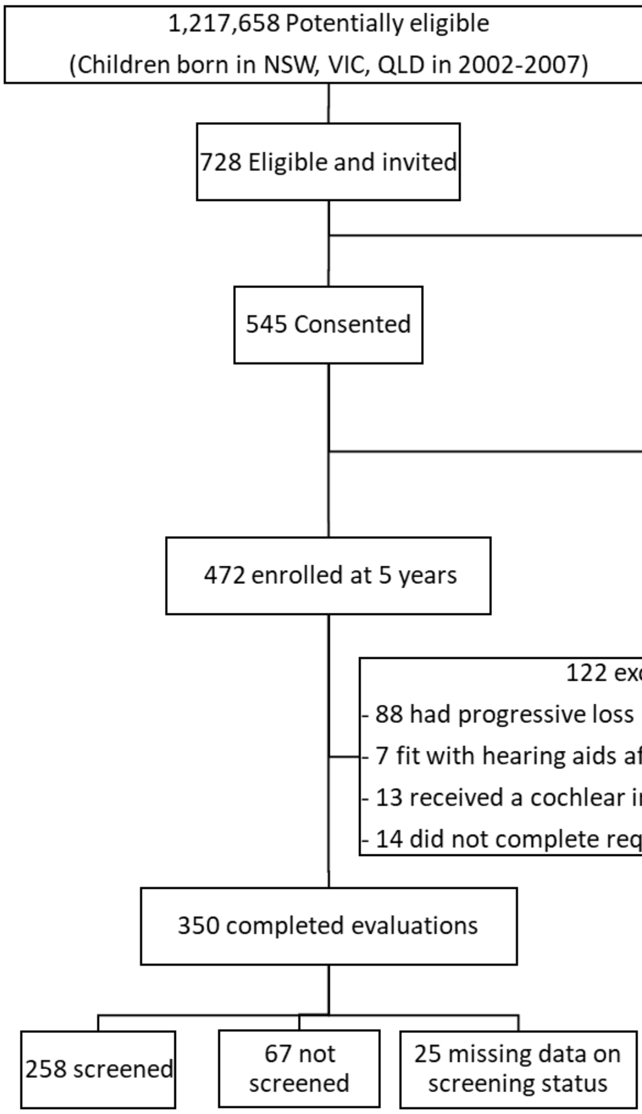
Outcomes by screening status for the HA and CI groups (Aim 3): Analyses for subgroups of users of HAs and CIs were as described above, but with age at intervention replaced by screening status (screened vs non-screened).

RESULTS

Figure 1 summarizes participant enrollment. Of the 728 infants who met inclusion criteria for the LOCHI study, 545 (74.8%) families gave consent for participation. Participants were similar to non-participants with respect to age, sex, degree of PCHL, and SES. By 5 years of age, 472 were enrolled in the study, of whom 384 children had non-progressive hearing loss. Of these, 350 completed the required evaluations. All except 3 completed at least one directly-administered test. Demographic characteristics of the cohort are shown in Table 2.

Insert Figure 1 about here

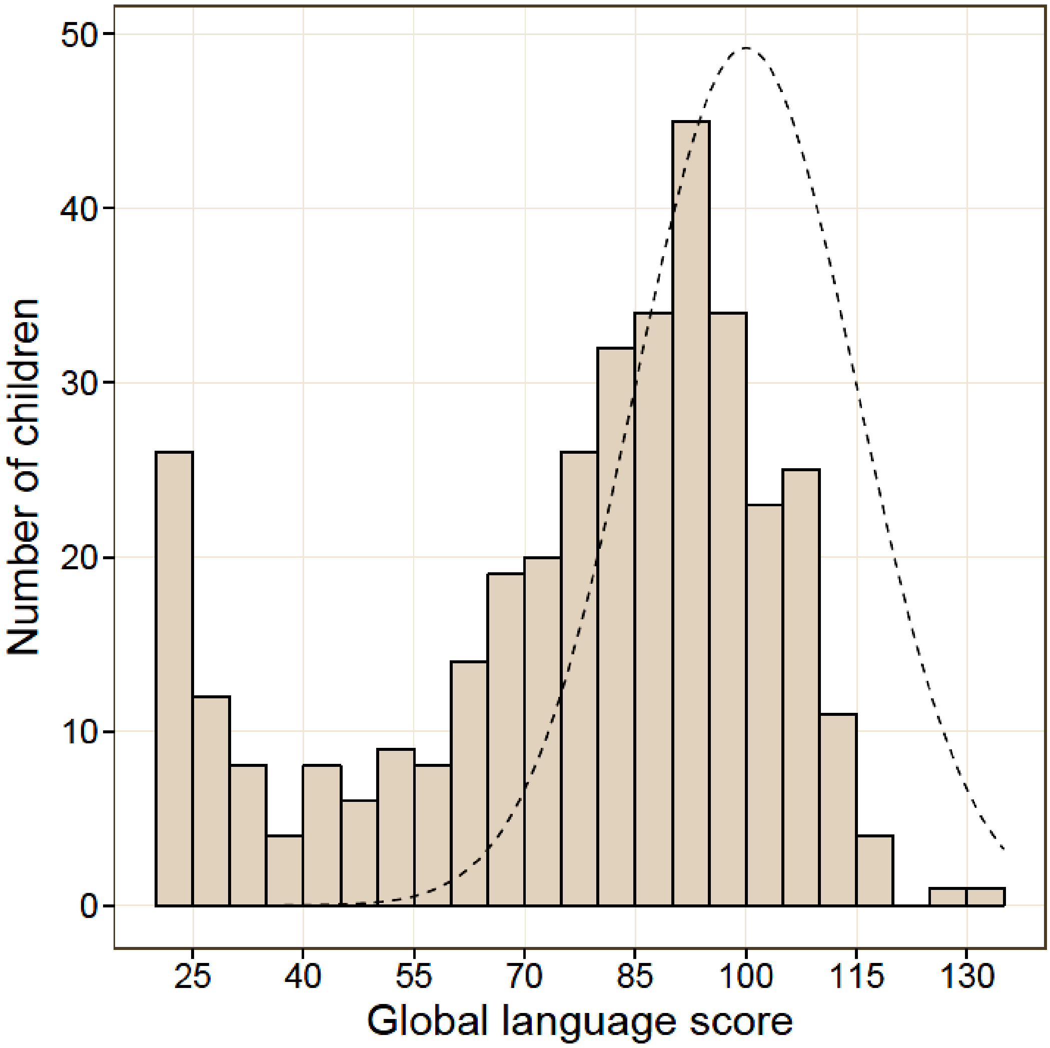
Figure 1. Study recruitment.



Insert Table 2 about here

Figure 2 shows the distribution of the global language scores of the cohort.

Figure 2. Distribution of global language scores. The dashed line depicts the theoretical distribution of scores in the 'normal' population (normally distributed with mean=100, SD=15).



Outcomes by age at HA fitting and hearing loss severity for the HA group (Aim 1): Children who received HAs earlier had better language outcomes. In addition, the regression model indicates that the effect of age at intervention increased as hearing loss increased (*p* = 0.035; 1-tailed). Table 3 gives the effect sizes associated with changes in age at amplification for different degrees of PCHL. Compared with amplification at 3 months, those who started amplification at 24 months had poorer language: -11.8 score points (95% CI: -18.7 to -4.8) for 70 dB HL, and -6.8 score points (95% CI: -10.8 to -2.8) for 50 dB HL (see Figure 3). These effects were observed after adjustments for nonverbal IQ, birth weight, additional disabilities, maternal education, sex, SES, and communication mode in a model that accounted for 74% of total variance in global score.

Insert Figure 3 about here

Figure 3. Adjusted global language scores by age at hearing aid (HA) fitting (log-transformed) in children using HAs at age 5 years. For display, children are grouped according to their BE4FA hearing loss in terms of dB HL. Data points are included for children who had 20 dB HL < BE4FA hearing loss < 40 dB HL in the first panel, 40 dB HL < BE4FA hearing loss < 60 dB HL in the second panel, and 60 dB HL < BE4FA hearing loss < 80 dB HL in the third panel. The points are adjusted with nonverbal IQ, birth weight, and SES set to the mean values of the entire HA group, no additional disabilities, and with maternal education, sex and communication mode set to university degree, female and oral respectively. In each panel, the regression line shows predicted mean score, and the shaded band depicts the 95% CI.

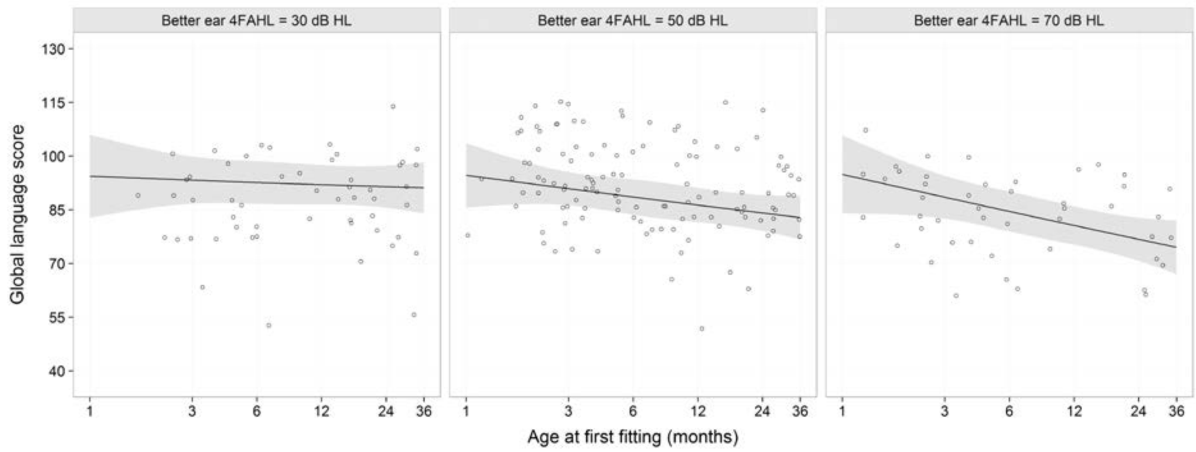
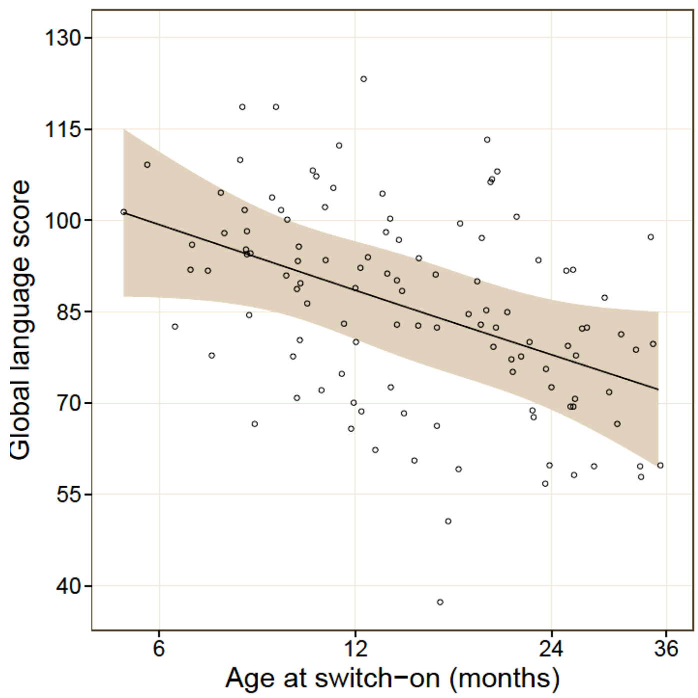


Table 3 about here

Outcomes by age at CI (Aim 2): Earlier age at CI activation was associated with better language at 5 years of age. Table 4 gives the effect sizes for variations in age at implantation. Compared to those who first received CIs at age 6 months, children who received a CI at 24 months had lower scores by 1.4SD (-21.4 score points, 95% CI: -33.8 to -9.0). These effects were observed after adjustments for nonverbal IQ, additional disabilities, maternal education, and communication mode in a multilinear regression model that accounted for 70% of total variance in global score. Figure 4 shows the relationship between age at implantation and the adjusted global scores.

Figure 4. Adjusted global language scores by age at cochlear implant (CI) activation (log-transformed). The regression line shows predicted mean score, and the shaded band depicts the 95% confidence interval.



Outcomes by screening status (Aim 3): The effect size of hearing screening (screened vs non-screened) was 0.3 score points (95% CI: -4.2 to 4.9; p = 0.88) for children using HAs; and 6.4 score points (95% CI: -2.8, 16.2, p = 0.19) for children using CIs.

DISCUSSION

Principal findings: In This prospective study, we demonstrated a strong positive effect of early intervention on 5-year language in children with PCHL. The younger a child received intervention, the better the language outcomes. In addition, more substantial benefits of earlier access to useful hearing via both HAs and CIs were obtained by those with worse hearing. Earlier intervention, rather than access to UNHS, improved outcomes.

Strengths of the study:

This study has a sample size that was powered for investigating the effect of intervention timing on outcomes, separately for children using HAs or CIs; after allowing for the effects of multiple demographic characteristics. In this relatively large study, we include language data from 350 children fitted with HAs before 3 years of age, comprising 189 (54%) before age 6 months (at 5 years: 111 use HAs, 78 use CIs) and 161 between 6 months and 3 years (at 5 years: 125 use HAs, 36 use CIs). These data therefore constitute the best opportunity to examine how severity of PCHL influences the association between age at intervention and language outcomes. Because all children received uniform hearing services and high-level technology from a single government-funded agency, we controlled for variations in post-diagnostic services. We evaluated outcomes using direct assessments of children by researchers who were blinded to age at diagnosis and parent reports.

The median age at HA fitting for the entire screened group (3.9 months [interquartile range or IQR: 2.3 to 10.1]) is earlier than that of the cohorts in the Wessex study (15 months; interquartile range: 10 - 40);[11] the Netherlands study (mean 15.7 months [SD, 14.0]);[37] and an earlier Australian study[38]. Unlike recent population-based studies in which researchers included few children with CIs (Wessex study: 16; Netherlands study: 32),[11,37] the present sample comprised 114 children who received CIs before 3 years of age, 42 (36.5%) of whom received them before 12 months of age. All children were assessed at 5 years of age, unlike previous studies that evaluated children at an older (Wessex study: 6-10 years) or a younger age (Netherlands study: 3-5 years).

Limitations:

While the sample size was large, the 48.1% (350/728) inclusion rate was lower than that in the Wessex study,[11] but it was comparable to the proportion with measured outcomes in the Netherlands study,[37] or a recent Australian study.[39] As age, sex, hearing loss and SES were similar in participants and non-participants, response bias seems unlikely. The participation rate may limit inferences on the merits of UNHS, but it does not affect conclusions on the impact of intervention timing on outcomes.

Although our regression models have accounted for a major proportion of variance in global scores, there remains about 30% of unexplained variance; so some combination of other factors and measurement error has also influenced outcomes. Future investigations of the effect of intervention timing on scores for specific assessments would also be worthwhile as age at intervention may affect some abilities more than others.[18]

Finally, we excluded children with progressive hearing loss (n = 88), so the results will likely not generalize to that population.

Interpretation in light of other studies: Our findings broadly agree with, but extends findings in recent reports. Earlier intervention improved language outcomes of children with PCHL.[18] Unlike previous studies that used cut-off points for early identification of 6 months[7] or 9 months,[11] suggesting a time window for language development to maximally benefit from intervention or a 'sensitive period' that ends before 1 year,[18] our data do not support this concept. Rather, the earlier intervention is provided (at least down to 3 months for HA and 6 months for CI) over the first 3 years of life, the better. Furthermore, the benefit increases as hearing loss increases (see Tables 3 and 4). The positive strong effect of early CI is consistent with results at age 3 years [16] and previous literature.[40,41]

Although access to UNHS led to earlier intervention (72% of the screened group commenced amplification before 6 months, compared to 32% in the unscreened group), and earlier intervention led to better outcomes, there is insufficient evidence to conclude that UNHS is beneficial. The diminished effect size of UNHS compared to that of age at intervention is likely because even though UNHS maximizes the opportunity of early intervention, not all screened children commenced amplification early, and not all unscreened children commenced amplification late (see Table 2).

The present findings, consistent with a recent review of evidence,[18] have important implications. First, timely device-fitting and educational support needs to follow UNHS to ensure earlier access to hearing, communication and language development.[42] Second, research needs to focus on post-diagnostic intervention to capitalize on early detection. Third, future follow-up of the existing cohort could evaluate whether the long-term language outcomes improve as the age at intervention decreases. confirm, or otherwise, the individual and societal benefits of early intervention made possible by UNHS.[43]

CONCLUSION

In This study, we show that it is vital to implement a seamless clinical pathway from screening to diagnosis to intervention (fitting HA or CI) so that the opportunity offered by UNHS to improve language development can be captured. Further, the significance of early implantation highlights the importance of vigilant monitoring of early outcomes with amplification so that children at risk for language delay can receive CIs at the earliest possible age.

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Table 1. Measures.

	Administration	Measure	Information about measure and scoring
Receptive language	Direct assessment	Preschool Language Scale v.4 (PLS-4)[22]	Auditory Comprehension subscale. Standardized: mean 100, SD 15, range 50-140.
	Parent report	Child Development Inventory[23]	Language comprehension subscale, based on 50 items. Quotients calculated.
Expressive language	Direct assessment	Preschool Language Scale v.4 (PLS-4)[22]	Expressive communication subscale. Standardized: mean 100, SD 15, range 50-140.
	Parent report	Child Development Inventory[23]	Expressive language subscale, based on 50 items. Quotients.
Receptive vocabulary	Direct assessment	Peabody Picture Vocabulary Test v.4 (PPVT-4)[24]	228 items. Standardized: mean 100, SD 15, range 20-160.
Expressive vocabulary	Direct assessment	Expressive Vocabulary Test v.2 (EVT-2)[25]	190 items. Standardized: mean 100, SD 15, range 20-160.
Reading	Direct assessment	Woodcock Diagnostic Reading Battery[26]	3 subscale scores: Word Attack, Word Identification, and Passage Comprehension. Standardized: mean 100, SD 15.
Maths	Direct assessment	Wechsler Individual Achievement Test[27]	Maths reasoning subtest. Standardized: mean 100, SD 15.
Number	Parent report	Child Development Inventory[23]	Number subscale, based on rating of 15 items. Quotients.
Letter knowledge	Direct assessment	Phonological Abilities Test[28]	Letter knowledge subtest, measures letter name. Raw score range 0-26.
	Parent report	Child Development Inventory[23]	Letter subscale, based on rating of 15 items. Quotients.
Speech	Direct	Diagnostic Evaluation of Articulation and	2 subscale scores: vowel and consonant; each based on 50

production	assessment	Phonology[29]	target words elicited by pictures. Standardized: mean 100, SD 15.
Functional auditory behaviour	Parent report	Parents' Evaluation of Aural/oral performance of Children (PEACH)[30]	11 items. Standardized: mean 100, SD 15.
Social skills	Parent report	Child Development Inventory[23]	Social subscale, based on ratings of 40 items. Quotients.
Behavior problems	Parent report	Strengths and Difficulties Questionnaire[31]	25 items. Standardized: mean 100, SD 15.
	Teacher report	Strengths and Difficulties Questionnaire[31]	25 items. Standardized: mean 100, SD 15.
Health-related quality of life	Parent report	Pediatric Quality of Life Inventory[32]	23-items. Standardized: mean 100, SD 15.

Table 2. Characteristics of the sample.

			HA at 5yrs: #Screened N=174	HA at 5yrs: Non-screened N=47	Difference *p* value*	CI at 5 yrs: #Screened N=84	CI at 5 yrs: Non-screened N=20	Difference *p* value
Child	Age at hearing aid (HA) fitting in months, mean (SD)		9.1 (8.9)	19.1 (10.7)	<0.001	4.0 (4.2)	12.9 (6.3)	<0.001
	Age at HA fitting in months, median (interquartile range, IQR)~		5.1 (2.6 to 12.2)	21.3 (9.2 to 28.6)		2.5 (1.8 to 4.1)	12.8 (7.4 to 18.2)	
	Fitted with HAs before 6 months of age, n (%)^		97 (55.7%)	9 (19.1%)	<0.001	70 (83.3%)	3 (15.0%)	<0.001
	Age at activation of first cochlear implant (CI) in months, median (IQR)		-	-		13.04 (9.7 to 19.2)	20.9 (16.3 to 26.1)	
	Fitted with CI before 12 months of age, n (%)		-	-		37 (44.0%)	2 (10.0%)	0.01
	Male gender, n (%)		97 (55.7%)	30 (63.8%)	0.41	41 (46.5%)	5 (25.0%)	0.09

	Birthweight in gms, mean (SD)		2967.7 (1022.2)	3144.2 (843.4)	0.67	3184.8 (883.5)	2923.4 (941.1)	0.18
	Degree of hearing loss depicted by better ear four frequency average hearing thresholds (BE4FA) in dB HL, mean (SD)		49.1 (14.8)	47.0 (15.9)	0.42	101.9 (14.5)	97.0 (16.9)	0.29
		Mild (BE4FA ≤40 dB HL), n (%)	47 (27.0%)	13 (27.7%)		-	-	
		Moderate (BE4FA: 41-60 dBHL), n (%)	88 (50.6%)	26 (55.3%)		-	-	
		Severe (BE4FA: 61-80 dB HL), n (%)	36 (20.7%)	7 (14.9%)		7 (8.6%)	3 (20%)	
		Profound (BE4FA: >80 dB HL), n (%)	3 (1.7%)	1 (2.1%)		74 (91.4%)	12 (80%)	
	Presence of any additional		49 (31.3%)	21 (47.7%)	0.06	23 (31.9%)	2 (11.1%)	0.14

	disabilities, n (%)							
		Cerebral palsy	10 (6.3%)	3 (6.7%)		5 (6.7%)	2 (11.1%)	
		Autism spectrum disorder	6 (2.8%)	1 (2.3%)		4 (5.4%)	0 (0.0%)	
		Vision impairment	30 (13.9%)	8 (17.8%)		4 (5.4%)	0 (0.0%)	
		Cranio-facial abnormalities	9 (5.7%)	0 (0.0%)		1 (1.3%)	0 (0.0%)	
		Genetic	17 (10.6%)	4 (8.9%)		5 (6.8%)	0 (0.0%)	
		Other	20 (12.6%)	12 (26.7%)		10 (13.5%)	0 (0.0%)	
	Nonverbal cognitive ability, mean (SD)		95.7 (25.4)	96.3 (24.5)	0.81	92.5 (26.4)	92.1 (24.0)	0.74
	Oral communication during educational intervention, n (%)		79 (83.2%)	19 (76.0%)	0.59	44 (83.0%)	5 (62.5%)	0.38
Family	Language other than English household,		33 (21.4%)	6 (14.6%)	0.46	14 (20.6%)	3 (20.0%)	1.00

	n (%)							
	SES[@], mean (SD)		1018.8 (74.5)	1023.9 (74.8)	0.68	1015.6 (73.5)	1010.5 (78.0)	0.83
	Maternal education: ≤ 12 years schooling, n (%)		59 (34.7%)	19 (43.2%)	0.39	24 (29.6%)	8 (42.1%)	0.44
	Maternal education: Vocational training Certificate, n (%)		43 (25.3%)	10 (22.7%)	0.88	18 (22.2%)	5 (26.3%)	0.94
	Maternal education: University degree, n (%)		68 (40.0%)	15 (34.1%)	0.59	39 (48.1%)	6 (31.6%)	0.29

#Accessed universal newborn hearing screening (UNHS). Information about screening status was missing for 25 of the 350 children. **p*-value for the test of whether the distributions in the screened and non-screened groups are the same using Mann-Whitney tests for the continuous variables and tests of proportions for the discrete variables. ~ Median age at HA fitting of the entire screened group comprising users of HA and CI was 3.9 months (interquartile range or IQR: 2.3 to 10.1); and the median age at fitting of the entire non-screened group was 17.3 months (IQR: 7.5 to 25.9). ^All percentages are relative to the number of non-missing values. [@]Socio-economic status (SES) represented by the Index of Relative Social Advantage and Disadvantage, which has a mean of 1000 and a standard deviation of 100. The study cohort has a range of 300 to 1250.

Table 3. Estimated effect size and 95% confidence interval in the mean global language score associated with the stated change in the age at intervention (e.g. 'compared to fitting at 3 months, fitting at 6 months' is expressed as '3 → 6'), if the other predictor variables are constant. The degree of hearing loss is expressed in terms of BE4FA in dB HL.

a. **Group with hearing aids**

	Estimate and 95% Confidence Interval		
Age at amplification	4FA = 30 dB HL	4FA = 50 dB HL	4FA = 70 dB HL
3 → 6	-0.7 (-4.3, 2.9)	-2.3 (-5.4, 0.8)	-4.0 (-7.6, -0.4)
6 → 12	-0.6 (-2.8, 1.6)	-2.3 (-3.6, -0.9)	-3.9 (-6.3, -1.6)
12 → 18	-0.3 (-2.3, 1.6)	-1.3 (-3.0, 0.4)	-2.3 (-4.3, -0.2)
18 → 24	-0.2 (-1.8, 1.4)	-0.9 (-2.4, 0.6)	-1.6 (-3.3, 0.1)

b. **Group with cochlear implants**

Age at implantation	Estimate and 95% Confidence Interval
6 → 12	-10.8 (-22.3, 0.8)
12 → 18	-6.2 (-9.3, -3.1)
18 → 24	-4.4 (-8.6, -0.1)

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