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7 Language outcomes in deaf or hard of hearing teenagers who are spoken language users: Effects of

8 universal newborn hearing screening and early confirmation.

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Abstract

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2 Objectives: This study aimed to examine whether a) exposure to universal newborn hearing
3 screening (UNHS) and b) early confirmation of hearing loss were associated with benefits to
4 expressive and receptive language outcomes in the teenage years for a cohort of spoken language
5 users. It also aimed to determine whether either of these two variables were associated with
6 benefits to relative language gain from middle childhood to adolescence within this cohort.

7 Design: The participants were drawn from a prospective cohort study of a population sample of
8 children with bilateral permanent childhood hearing loss, who varied in their exposure to universal
9 newborn hearing screening, and who had previously had their language skills assessed at 6-10 years.
10 Sixty deaf or hard of hearing (D/HH) teenagers who were spoken language users and a comparison
11 group of 38 teenagers with normal hearing completed standardised measures of their receptive and
12 expressive language ability at 13-19 years.

13 Results: Teenagers exposed to UNHS did not show significantly better expressive (adjusted mean
14 difference = 0.40, 95% CI = -0.26 to 1.05, $d = 0.32$) or receptive (adjusted mean difference = 0.68,
15 95% CI = -0.56 to 1.93, $d = 0.28$) language skills than those who were not. Those who had their
16 hearing loss confirmed by 9 months of age did not show significantly better expressive (adjusted
17 mean difference = 0.43, 95% CI = -0.20 to 1.05, $d = 0.35$) or receptive (adjusted mean difference =
18 0.95, 95% CI = -0.22 to 2.11, $d = 0.42$) language skills than those who had it confirmed later. In all
19 cases effect sizes were of small size and in favour of those exposed to UNHS or confirmed by 9
20 months. Subgroup analysis indicated larger beneficial effects of early confirmation for those D/HH
21 teenagers without CIs ($N = 48$; 80% of the sample), and these benefits were significant in the case of
22 receptive language outcomes (adjusted mean difference = 1.55, 95% CI = 0.38 to 2.71, $d = 0.78$).

23 Exposure to UNHS did not account for significant unique variance in any of the three language scores
24 at 13-19 years beyond that accounted for by existing language scores at 6-10 years. Early

1 confirmation accounted for significant unique variance in the expressive language information score
2 at 13-19 years after adjusting for the corresponding score at 6-10 years (R^2 change = 0.08, $p=0.03$).

3 Conclusions:

4 This study found that while adolescent language scores were higher for D/HH teenagers exposed to
5 UNHS and those who had their hearing loss confirmed by 9 months, these group differences were
6 not significant within the whole sample. There was some evidence of a beneficial effect of early
7 confirmation of hearing loss on relative expressive language gain from childhood to adolescence.
8 Further examination of the impact of these variables on adolescent language outcomes in other
9 cohorts would be valuable.

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2 Approximately 1 in 1000 babies is born with bilateral permanent childhood hearing loss
3 (PCHL) of at least moderate severity (>40dB HL) (Davis et al. 1997). The impoverished access to
4 spoken language that is a consequence of childhood hearing loss places many deaf or hard of
5 hearing (D/HH) children at significant risk of delayed language development (Eisenberg 2007;
6 Luckner et al. 2010; Moeller et al. 2007; Moeller and Tomblin, 2015). Early identification of D/HH
7 children enables them to receive early intervention to improve the quality of their language input
8 during a ‘sensitive period’ for language development at the beginning of life (Lyness et al. 2013;
9 Thomas et al. 2008). However, historically, identification of children with congenital PCHL has been
10 delayed resulting in many months or years of restricted access to spoken language prior to
11 identification and intervention (Davis et al. 1997). The advent of universal newborn hearing
12 screening (UNHS) created the opportunity to identify children born with PCHL within the first few
13 days of life, including those children with no known risk factors for the condition. This in turn made it
14 possible for these children to be fitted with hearing devices that facilitate access to spoken language
15 (e.g. hearing aids or cochlear implants) very early in life, and for their families to enroll in early
16 intervention programs to support their child’s developing speech, language and communication skills
17 (Kasai et al. 2012; Meinzen-Derr et al. 2011; Moeller et al., 2013).

18 A significant body of evidence from around the world has demonstrated the efficacy of
19 UNHS in increasing rates of early identification of babies born with PCHL (see Nelson et al. 2008;
20 Thompson et al. 2001 for reviews). This includes evidence from a controlled trial undertaken in the
21 Wessex region of England (Kennedy et al. 1998; Kennedy et al. 2005). The Wessex trial was unique in
22 that UNHS was given/not given according to a controlled experimental regimen, a situation made
23 ethically possible because the screening tests involved were at the time novel and unproven. This
24 created two cohorts of babies that were very similar in all respects other than their exposure to
25 UNHS. In the cohort of babies who were exposed to UNHS, 74% of all cases of true PCHL were

1 referred to audiological services before they were 6 months old, more than double the 31% referred
2 before 6 months in the cohort who had not been exposed to UNHS (Kennedy et al. 2005). A recent
3 population-based study in Australia also reported that UNHS was associated with a reduction in the
4 mean age at which infants with PCHL were identified from 16.2 months to 8.1 months when
5 compared with the contemporary birth cohort in another Australian state that adopted a policy of
6 screening only infants known to be at increased risk of PCHL (Wake et al., 2016).

7 To determine whether UNHS, and the resulting early identification of PCHL, is associated
8 with the predicted benefits to language outcomes, studies have compared these outcomes between
9 groups of D/HH children who were exposed and not exposed to a programme of UNHS, and
10 between groups of early- and late-identified children (see Nelson et al. 2008; Pimperton and
11 Kennedy 2012; Thompson et al. 2001 for reviews). Of these studies, three major population-based
12 studies have prospectively examined the effect of exposure (or not) to a UNHS programme at birth
13 on subsequent language outcomes (Kennedy et al., 2006; Korver et al., 2010; Wake et al., 2016).
14 D/HH children who were involved in the controlled Wessex trial of UNHS participated in a follow-up
15 study at the age of 6-10 years alongside an additional cohort of D/HH children from Greater London
16 who also varied in their exposure to UNHS (Kennedy et al., 2006). Compared to those not exposed,
17 children in populations exposed to a programme of UNHS at birth showed significantly superior
18 receptive language skills but no significant advantages for their expressive language or speech skills.
19 Within the same cohort, confirmation of PCHL at ≤ 9 months was associated with significant
20 benefits to both receptive and expressive language but not to speech skills. Furthermore, the effect
21 sizes for the early vs. late confirmation expressive and receptive language comparisons were larger
22 than those for the UNHS vs. no UNHS comparisons. This pattern of findings may be explained by the
23 fact that some babies born in periods with UNHS were not confirmed early, and some born in period
24 without UNHS were confirmed early, both of which could account for greater benefits of early
25 confirmation (Kennedy et al., 2006; Pimperton and Kennedy 2012). It is important to note that the
26 benefits associated with early confirmation did not bring the average performance level of these

1 early-confirmed children to the same level as their peers with normal hearing: the D/HH children
2 who were early-confirmed still showed significant deficits in both their receptive (1.76 SD below the
3 hearing mean) and expressive (0.59 SD below the hearing mean) language skills. Even with early
4 confirmation and intervention (e.g. provision of hearing aids or cochlear implants) it is likely that
5 D/HH children continue to experience a greater degree of inconsistent access to linguistic input, and
6 hence accrue reduced cumulative linguistic experience relative to their hearing peers (Moeller and
7 Tomblin, 2015) and it is likely that this contributes to their persistent language delays.

8 Korver et al. (2010) compared language outcomes for 3-5 year old D/HH children who were
9 born in regions of the Netherlands where UNHS was in place with those of D/HH children who were
10 born in regions where there was no UNHS programme. They found that the children born in regions
11 where there was no UNHS produced significantly more signed words than the children born in
12 regions with UNHS. The number of signed words used was inversely related to the number of spoken
13 words meaning that the children exposed to UNHS showed an advantage in terms of number of
14 spoken words used. This advantage was not statistically significant but the authors argued it was
15 clinically important. There were no differences between the UNHS and no UNHS groups in terms of
16 their mean length of utterance or the complexity of the sentences they produced.

17 Most recently, Wake et al. (2016) looked at outcomes for three populations of children with
18 congenital PCHL; one exposed to UNHS, one contemporary birth cohort exposed to risk factor
19 screening, and one earlier birth cohort exposed to opportunistic detection (i.e. no systematic UNHS
20 or risk factor screening programmes). They found that in children without intellectual disability
21 exposure to UNHS was associated with significant population-level benefits to expressive and
22 receptive language skills compared to exposure to risk factor screening and that population language
23 scores improved incrementally from opportunistic detection to risk factor screening to UNHS.

24 Other studies have also been conducted to look at the effects of UNHS exposure on
25 language outcomes. Yoshinaga-Itano et al. (2000) found significantly higher receptive and expressive

1 language outcomes for D/HH children aged between 9 months and 6 years who had been born in
2 hospitals offering UNHS compared with those born in hospitals that did not. By contrast, Fitzpatrick
3 et al. (2007) did not find any significant advantages in terms of expressive or receptive language
4 outcomes for children who had been screened as newborns compared with those that had not. They
5 suggested that one of the reasons they may have been unable to detect benefits of early
6 confirmation in their study was the inclusion of a relatively high proportion of children with cochlear
7 implants (CIs). They argued that for these children, early confirmation was likely to have less of an
8 impact on language outcomes than the age at which they received their implant, i.e. when they
9 achieved 'access to effective intervention', and that age at implantation was similar across the
10 screened and unscreened groups. A recent large scale study in Australia also found that age at
11 implantation was a significant predictor of language outcomes in children with cochlear implants
12 (Ching et al., 2013).

13 From 2003 onwards, UNHS has been implemented as national or regional policy in
14 numerous countries around the world, including the USA, where in 2009 an estimated 5073 cases of
15 PCHL were detected by UNHS (Howell et al. 2012). This figure accounted for 43.3% of all detected
16 cases of any of the 29 medical conditions for which newborn screening is recommended in the USA.
17 To date, however, no study has followed up infants involved in trials of UNHS through to the teenage
18 years. As a result the longer-term effects of UNHS and early identification of children who are born
19 D/HH on language outcomes are as yet unknown. Following up the cohort of teenagers from the
20 Wessex and Greater London birth cohorts described above whose language skills at primary school
21 age have been reported previously (Kennedy et al., 2006) provided us with a unique opportunity to
22 test whether exposure to UNHS and early identification of PCHL brings significant benefit to
23 outcomes in the teenage years. We have reported elsewhere on the literacy outcomes for this
24 cohort (Pimperton et al., 2016), including significant benefits of early confirmation of PCHL, but not
25 exposure to UNHS, on reading comprehension, the primary outcome in the teenage phase of this
26 cohort study.

1 In the present paper we focus on a subset of the cohort who were spoken language users
2 and therefore able to provide data to address the question of whether exposure to UNHS and
3 confirmation of PCHL by \leq 9 months brings benefits to spoken receptive and expressive language
4 outcomes in the teenage years. Consistent with the argument of Fitzpatrick et al. (2007) regarding
5 early confirmation having less of an impact for children with CIs, the benefits of early confirmation
6 to reading comprehension reported in Pimperton et al. (2016) were larger in those D/HH teenagers
7 without cochlear implants. We therefore examined whether age at confirmation also has differential
8 importance for the spoken language skills of the D/HH teenagers with and without CIs.

9 Following this sample of D/HH participants from middle childhood through to adolescence
10 also provided us with a unique opportunity to address the question of whether exposure to UNHS
11 and early confirmation were associated with superior spoken language development during this
12 period. To address this we examined whether UNHS or early confirmation were associated with
13 variation in language outcomes at 13-19 years (i.e. in the current phase of the study) while adjusting
14 for the level of pre-existing language skills, as assessed at 6-10 years.

15 To summarise, this paper addressed two key questions:

- 16 1. Are a) UNHS and b) early confirmation of PCHL associated with benefits to adolescent
17 language outcomes in spoken language users?
- 18 2. Are a) UNHS and b) early confirmation of PCHL associated with benefits to relative language
19 gain from middle childhood to adolescence in spoken language users?

20

21 Methods

22 *Participants*

23 The eligible sample for this follow-up study (T2) comprised 120 D/HH teenagers and a
24 comparison group of 63 teenagers with normal hearing (NH) who had taken part in the previous

1 phase of this research at primary school at the age of 6-10 years (T1; see Figure 1). As detailed in
2 Kennedy et al. (2006), those 120 teenagers were drawn from all 160 contactable children with
3 bilateral PCHL of at least 40 dB HL in the better ear identified from a birth cohort of 157,000 children
4 in eight districts of southern England. Children with a known postnatal cause of their hearing loss
5 (e.g., bacterial meningitis) were not included. The children in the sample were born over a three year
6 period (1993-1996 inclusive) in four districts in the Wessex region or over a five year period (1992-
7 1997 inclusive) in two pairs of adjacent districts in the Greater London region. The four districts in
8 the Wessex subgroup had provided the birth cohort for the Wessex trial, in which a program of
9 universal newborn screening was or was not in place in each pair of districts for birth cohorts born in
10 alternate four- or six-month periods. The Greater London subgroup consisted of children born in the
11 only two districts in the United Kingdom that provided UNHS for PCHL in the early 1990s and in two
12 other adjacent districts. Protocols for the identification, confirmation and management of PCHL
13 were similar at all sites apart from variation in the details of UNHS provision (Kennedy et al., 2005;
14 Tucker & Bhattacharya, 1992; Watkin & Baldwin, 1999). The children exposed to UNHS and those
15 who were not were, in all but a small number of cases, treated by the same audiological service
16 providers.

17 Seventy six of the 120 D/HH teenagers and their families who had been assessed at primary
18 school age agreed to participate in this follow-up phase of the research. Of these 76 D/HH
19 teenagers, 60 and 59 completed the receptive and expressive language assessments respectively
20 and were therefore included in the present study on spoken language outcomes (see Figure 1).
21 Those who did not complete the assessments either used British Sign Language (BSL) as their
22 preferred language, rendering these spoken English assessments inappropriate, or had severe
23 additional disabilities that precluded the development of sufficient language to attempt the tests.

24 The eligible comparison group of 63 teenagers with NH who participated at T1 were drawn
25 from the same birth cohorts as the group of 120 D/HH children. Thirty eight of the 63 (60%)

1 teenagers with NH who had participated at T1 took part in the present study (see Figure 1). All 38
2 hearing teenagers completed both the receptive and expressive language assessments.

3

4 *Procedure*

5 This study was approved by the Southampton and South West Hampshire Research Ethics
6 Committee. Written informed consent for participation in the study was obtained from principal
7 caregivers and from the teenage participants. Each teenage participant was assessed by a trained
8 researcher who was unaware of their audiological history. Testing was undertaken in a quiet room at
9 the teenager's home or at their school according to their expressed preference.

10 Spoken language skills were assessed with the following measures:

11 *Receptive language.* The Test for Reception of Grammar Version 2 (TROG-2; Bishop 2003),
12 standardised on the age range 3 to 16 years 11 months as well as with adults, was used to assess
13 participants' receptive skills for spoken English grammar. Items in the task assess understanding of
14 increasingly complex grammatical contrasts, including plurals, passives, negatives, and relative
15 clauses. Participants must point to a picture from a choice of four alternatives that corresponds to a
16 spoken sentence.

17 The British Picture Vocabulary Scale Third Edition (BPVS-3; L. M. Dunn et al. 2009), standardised on
18 the age range 3 years to 16 years 11 months, provided a measure of the participants' receptive skills
19 for spoken English vocabulary. Participants must point to a picture from a choice of four alternatives
20 that corresponds to a spoken word. Earlier editions of both the BPVS and the TROG were used to
21 measure the participants' receptive language skills at primary school age (T1).

22 *Expressive language.* The Expression, Reception and Recall of Narrative Instrument (ERRNI; Bishop
23 2004), standardised on the age range 4 years to adults, provided a measure of participants'
24 expressive spoken language skills. Participants were required to produce a narrative based on a

1 series of picture cues, and subsequently to reproduce that narrative without the support of the
2 pictures. Their narrative productions were audio-recorded, subsequently transcribed and scored
3 according to the ERRNI manual to produce three scores: an Initial score for the quality of their initial
4 narrative, a Recall score for the quality of their recalled narrative, and a Mean Length of Utterance
5 (MLU) score which reflected the average length of their utterances in words across both the initial
6 and recall narrative narratives. An inter-rater reliability exercise, following Whitehouse et al. (2009),
7 was carried out to check the reliability of the scoring: 12 randomly selected narratives (12% of the
8 total) were transcribed and scored by a second rater. There was good agreement (intraclass
9 correlations, r_{ic}) between the two ratings for all three scores (Initial, $r_{ic} = .82$; Recall, $r_{ic} = .90$; MLU, r_{ic}
10 $= .95$).

11 The measure used to assess expressive language skills at primary school age (T1), the
12 Renfrew Bus Story Test (Renfrew 1995), was designed for use with 3-8 year olds. This test involved
13 children listening to a story told by the experimenter while viewing a series of pictures that
14 corresponded to the story. They then had to tell the story in their own words (i.e. produce their own
15 narrative), using the pictures as prompts. Two scores were derived from this measure, reflecting
16 both the inclusion of relevant information in the narrative and the length of utterances produced,
17 and were combined into an expressive language composite score (Kennedy et al., 2006). The ERRNI
18 was selected for this current phase of the study because it was similar in design to the Bus Story
19 Test, enabling the derivation of both a score for the information content of the narrative produced
20 as well as a measure of utterance length, and had been designed for an age range within which our
21 participants fell.

22 *Non-verbal ability.* A twenty minute timed version (Hamel et al. 2006) of Raven's Standard
23 Progressive Matrices Plus (Raven's SPM+; Styles et al. 1998) was used to assess non-verbal ability.
24 Participants were given twenty minutes to work their way through a series of progressively more

1 difficult matrix reasoning puzzles. Raw scores reflecting the total number of correct items out of a
2 possible 60 were calculated.

3 *Demographic and audiological characteristics.* Other characteristics of the teenager and their family,
4 including maternal education level and languages used in the home, were also documented. The
5 most recently available audiological data were documented from audiology and cochlear implant
6 centre records. Severity of hearing loss was categorized from the most recent audiological records as
7 moderate (40 to 69 dB HL), severe (70 to 94 dB HL), or profound (≥ 95 dB HL) according to four-
8 frequency averaging of the pure-tone thresholds from 500 to 4000 Hz for the better ear. For
9 participants with cochlear implants, we collected unaided pure-tone thresholds obtained during
10 assessment for implantation.

11

12 *Analysis strategies*

13 *Effects of UNHS and early confirmation*

14 For the purpose of comparisons within the group of D/HH teenagers, we used norms
15 obtained from the participating children with NH (Kennedy et al. 2006). The group mean score and
16 standard-deviation score for a particular measure in teenagers with NH were used to derive age-
17 adjusted z scores for the D/HH teenagers on that measure. When language outcomes were
18 examined in this cohort at 6-10 years, the BPVS and TROG z scores were averaged into a receptive
19 language composite and the information and sentence length scores from the ERRNI were averaged
20 into an expressive language composite (Kennedy et al., 2006). To check the validity of using the
21 same composite structure at the current time point, correlations between the measures were
22 examined and a Principal Component Analysis (PCA) was conducted. The two receptive language
23 measures (BPVS and TROG) showed strong positive correlations ($n = 98$, $r = .71$, $p < .001$) and in the
24 PCA both loaded highly (.92) on the first component. They were therefore combined into a single
25 receptive language composite for analysis purposes. Of the three expressive language scores, the

1 two information scores (initial and recall) showed strong positive correlations with each other (n =
2 97, r = .71, p < .001) but weaker relationships with the MLU score (n = 97, r = .20, p = .048, and n =
3 97, r = .26, p = .009 respectively). A PCA on these expressive score identified a single component
4 with an eigen value greater than 1. On this component the two information scores had loadings
5 greater than .5 (initial .88 and recall .90). The MLU had a loading of .49. The two information scores
6 were therefore combined into an Expressive Information composite, and the MLU score was
7 reported separately for analysis purposes. Thus the three language outcomes examined in this study
8 were a Receptive language composite (BPVS and TROG scores), an Expressive Information composite
9 (ERRNI initial and recall information scores), and Expressive MLU score.

10 We pre-specified the definition of 'early' confirmation of PCHL as confirmation at ≤ 9
11 months of age consistent with the definition used in our previous trial of UNHS (Kennedy et al. 1998)
12 and our evaluation of language at primary school age (Kennedy et al. 2006). We separately assessed
13 the associations between a) exposure to UNHS (i.e., birth during periods when UNHS was in place),
14 and b) confirmation of PCHL at ≤ 9 months of age, and each of the three receptive and expressive
15 language scores (Receptive, Expressive Information, Expressive MLU) before and after adjustment in
16 a multiple linear regression for severity of hearing loss, maternal education, and non-verbal ability
17 which were pre-specified as potential confounders of the study outcomes (Kennedy et al., 2006) and
18 English as an additional language in the home which was identified as a potential confounder of the
19 outcomes at the current time point due to unequal distribution between the early and late
20 confirmed groups (Pimperton et al., 2016). We tested for an interaction between the effects of a)
21 UNHS vs. no UNHS and b) early vs. late confirmation of PCHL and cochlear implantation (CI vs. no CI)
22 by entering an additional term reflecting this interaction into regression analyses predicting the
23 combined receptive and expressive language scores.

24 Normality and homogeneity of the residual variance were examined for all measures to
25 ensure that the regression models were appropriate. All reported *p* values are two-sided and 95%
26 confidence intervals (95% CI) are given.

1 *Language development from childhood to adolescence*

2 In order to examine whether a) exposure to UNHS and b) early age at confirmation were
3 associated with variability in relative language gains from middle childhood to adolescence, a
4 hierarchical multiple regression analysis was conducted which assessed whether UNHS exposure or
5 age at confirmation predicted significant unique variance in T2 language (assessed at the current
6 assessment time point, aged 13-19 years) when adjusted for T1 language (assessed at the previous
7 assessment time point, aged 6-10 years), as well as the confounding variables (severity of hearing
8 loss, maternal education level, non-verbal ability, and English as a main language at home). The
9 analysis was run separately for receptive and the two expressive language measures. Normality and
10 homogeneity of the residual variance were examined for all measures to ensure that the regression
11 models were appropriate. This analysis necessarily included only D/HH participants who had
12 provided spoken language data at both time points (Receptive, N = 59; Expressive, N = 54). For this
13 longitudinal element of the analysis it was important to directly compare the same D/HH
14 participants relative to the same NH control group at both time points (T1 and T2). We therefore
15 recalculated the T1 language z scores for the D/HH participants using the norms only from those
16 participants with NH who provided the norms for the T2 phase of the research (N = 38) and used
17 these in the analysis.

18
19 Results

20 *Participants*

21 Table 1 presents descriptive statistics on key Time 1 demographic variables for the teenagers
22 who did and did not provide spoken language data for the present study. The D/HH teenagers who
23 provided spoken language data in the present study did not differ significantly from those who did
24 not provide spoken language data in terms of age, sex, severity of hearing loss, use of a cochlear
25 implant, birth in periods with UNHS, confirmation at ≤ 9 months, or maternal education level (all

1 $ps > .10$) but there was a non-significant tendency for those who did provide spoken language data
2 to be more likely to have English as the main language at home ($\chi^2 (2, N = 98) = 5.22, p = .07$). The
3 teenagers with NH who provided data in the present study did not differ significantly from those
4 who were lost to follow up in terms of age, sex and use of English as a first language at home (all
5 $ps > .10$) but, compared to those who were lost, those who were retained showed higher maternal
6 education levels ($\chi^2 (2, N = 98) = 6.13, p = .05$). Both the D/HH and NH groups who provided T2
7 spoken language data showed higher receptive language z scores at T1 than their counterparts who
8 did not (D/HH: $t (99) = 1.98, p = .05$; NH: $t (61) = 2.22, p = .03$).

9 Table 2 presents descriptive statistics on key Time 2 demographic variables for the teenagers
10 who provided language data for this study. The D/HH teenagers did not differ significantly from the
11 teenagers with NH with respect to gender, non-verbal ability, or use of English as the main language
12 at home (all $ps > .10$). The teenagers with NH were significantly younger than the D/HH teenagers (t
13 $(96) = 2.65, p = .01$) and there was a non-significant tendency for them to have higher maternal
14 education levels ($\chi^2 (2, N = 98) = 5.22, p = .07$). Scores were age-adjusted prior to analysis, and
15 maternal education was adjusted for in the group comparisons.

16 The D/HH participants confirmed at ≤ 9 months did not differ significantly from those who
17 were confirmed > 9 months with respect to age, gender, severity of hearing loss, use of a cochlear
18 implant, non-verbal ability, aetiology, and maternal education level (all $ps > .10$) (Table 2). There
19 was a non-significant tendency for more teenagers confirmed at ≤ 9 months to have English as the
20 main language at home ($\chi^2 (1, N = 60) = 3.51, p = .06$). This variable was adjusted for in the group
21 comparisons. Those exposed to UNHS did not differ significantly from those not exposed to UNHS
22 with respect to age, gender, severity of hearing loss, use of a cochlear implant, non-verbal ability,
23 aetiology (all $ps > .10$). There was a non-significant tendency for lower maternal education in the
24 group exposed to UNHS ($\chi^2 (2, N = 60) = 5.34, p = .07$). This variable was adjusted for in the group
25 comparisons.

1 *Language outcomes NH vs D/HH*

2 The teenagers with NH showed significantly higher adjusted mean receptive language z
3 scores than the D/HH teenagers, but no significant advantage in terms of expressive language z
4 scores (Table 3). This contrasts with the findings in this cohort aged 6-10 years, when the D/HH
5 participants showed significant deficits in both receptive *and* expressive language relative to the
6 comparison group with NH (Kennedy et al. 2006). When we looked only at those participants who
7 provided receptive and expressive language data at both time points to make a direct comparison,
8 the same pattern of results was evident in that while the magnitude of the receptive language deficit
9 shown by the D/HH participants (N = 54) relative to the participants with NH (N = 38) remained
10 similar from T1 to T2 (T1 *M* Difference = -2.01, 95% CI = -2.50 to -1.51; T2 *M* Difference = -1.78, 95%
11 CI = -2.45 to -1.10), the expressive deficits for both MLU (T1 *M* Difference = -0.96, 95% CI = -1.52 to -
12 0.39; T2 *M* Difference = -0.15, 95% CI = -0.55 to 0.25) and Information score (T1 *M* Difference = -
13 1.28, 95% CI = -1.85 to -0.71; T2 *M* Difference = -0.20, 95% CI = -0.64 to 0.24) were much reduced.

14 To examine whether the patterns of language deficits shown by the D/HH group in this study
15 were a function of the NH reference group used, we also examined standard scores on the receptive
16 and expressive language measures to assess their performance relative to the larger hearing samples
17 on which the tests were standardised. Standard scores (M = 100, SD = 15) were available for all
18 participants who completed the ERRNI. The D/HH group did not show evidence of substantial
19 deficits on this task when examining their scores relative to the standardisation sample; their
20 standard scores were close to or above the mean of the standardisation sample and were similar to
21 those obtained by the NH reference group who participated in this study (Table 3).

22 Standard scores (M = 100, SD = 15) were available for all participants on the TROG. For the
23 BPVS, standard scores (M = 100, SD = 15) were available for participants up to 16;11 years. For all
24 participants in our sample over this age, we allocated them the standard score for the highest
25 available age bracket (16;09-16;11). For participants whose raw score placed them below the basal

1 standard score of 70 (N = 9, all D/HH), we allocated them a score of 69. This limited the capacity of
2 these standard scores to reflect variability in raw scores for these lower scoring participants. The
3 mean receptive standard score (TROG standard score + BPVS standard score/2) for the D/HH group
4 was around 1SD below the standardisation mean of 100, while the mean standard score for the NH
5 group was very close to the standardisation mean (Table 3).

6 *Effects of UNHS*

7 Compared to birth during a period without UNHS, birth during a period with UNHS was not
8 associated with significantly higher expressive and receptive language z scores (see Table 4; Figure
9 2). Effect sizes were all in the direction of favouring the UNHS group and were of small size.

10 The additional interaction term reflecting the interaction between the presence of a CI and
11 the effects of UNHS vs. no UNHS on overall language was not significant ($p = .22$).

12 *Effects of early confirmation*

13 Compared to confirmation of PCHL at a later age, confirmation of PCHL at ≤ 9 months of
14 age was not associated with significantly higher receptive and expressive language z scores for the
15 whole sample (see Table 4; Figure 2). Effect sizes for all three language outcome variables were in
16 the direction of favouring the early confirmed group and were of small size.

17 The additional interaction term reflecting the interaction between the presence of a CI and
18 the effects of early vs. late confirmation of PCHL on overall language was significant ($p = .03$)
19 suggesting that age at confirmation may be differentially affecting the language outcomes of those
20 D/HH participants without CIs (N = 48), compared to those with CIs (N = 12), therefore results were
21 also examined separately for the CI vs no CI groups.

22 For the D/HH participants without CIs (N = 48), confirmation of PCHL at ≤ 9 months was
23 associated with significantly higher receptive, but not expressive, language scores after adjustment

1 for the effects of severity of hearing loss, maternal education level, non-verbal ability and the
2 presence of English as an additional language in the home (see Table 4). Effect sizes for all three
3 language outcome variables were in the direction of favouring the early confirmed group and were
4 of medium size.

5 For the D/HH participants with CIs (N = 12), numbers were too small to carry out parallel
6 regression analyses but descriptive statistics comprising unadjusted means (SDs) and mean
7 differences are reported (Table 4) and indicate lower language scores in all three domains for the
8 early confirmed participants.

9 *Language development from childhood to adolescence*

10 For all D/HH participants who provided receptive language data at both time points (N = 59),
11 receptive language z score at T1 was entered at Step 1 of the hierarchical linear regression analysis
12 predicting receptive language z score at T2. Severity of hearing loss, non-verbal ability, maternal
13 education level and English as a main language at home were entered at Step 2 and finally, exposure
14 to UNHS (in model 1) or age at confirmation (in model 2) was entered as a dichotomous predictor
15 variable at Step 3. In both models, adding group membership (UNHS vs. no UNHS in Model 1, early
16 vs. late confirmed in Model 2) at Step 3 did not predict significant additional unique variance in T2
17 Receptive Language outcomes (see Table 5).

18 Parallel regression analyses were run predicting the two T2 expressive language outcomes
19 (Information and MLU) for all D/HH participants who provided expressive language data at both time
20 points (N = 54), with the equivalent expressive language score from T1 entered at Step 1. The
21 regression models for expressive language accounted for much lower proportions of the variance in
22 T2 expressive language than was the case for receptive language because the relationship between
23 T1 and T2 expressive language scores was much weaker than that between T1 and T2 receptive
24 language scores. Exposure to UNHS entered at Step 3 of the model did not account for significant

1 unique variance in either of the T2 expressive language outcomes. Age at confirmation entered at
2 Step 3 accounted for significant unique variance in Expressive Information score but not in
3 Expressive MLU (see Table 5).

4 Running the same set of analyses for the D/HH participants without CIs who had provided
5 language data at both time points (Receptive N = 48; Expressive N = 44) produced the same pattern
6 of significant effects of age at confirmation as in the whole sample, though in all cases the
7 percentage of unique variance explained by age at confirmation was higher than it was for the whole
8 D/HH group; Age at confirmation entered at Step 3 in the model predicted significant unique
9 variance in T2 Expressive Information (16%; $F(1, 37) = 8.42, p = .01$) but not T2 Expressive MLU (5%;
10 $F(1, 37) = 2.38, p = .13$) scores or T2 Receptive scores (2%; $F(1, 41) = 3.75, p = .06$). The number of
11 D/HH participants with CIs who had provided language data at both time points (Receptive N = 11;
12 Expressive N = 10) was too small to run these hierarchical multiple linear regression analyses for that
13 subgroup.

14 Discussion

15 This follow-up study in a cohort of spoken language-using D/HH teenagers found no significant
16 benefits of UNHS or confirmation of PCHL at ≤ 9 months of age on receptive or expressive language
17 outcomes in adolescence. For all outcomes, UNHS and early confirmation were associated with
18 higher language scores but effect sizes were of small size (range of Cohen's d s 0.22 to 0.43) and the
19 differences between groups were not significant. The lack of significant effects of UNHS and early
20 confirmation on teenage language outcomes within the whole sample differs from earlier findings
21 with this cohort in middle childhood (Kennedy et al., 2006) when UNHS exposure was associated
22 with significant benefits to receptive language and early confirmation with significant benefits to
23 both receptive and expressive language. Sample attrition over the approximately eight years
24 between these two assessment time points, coupled with inclusion only of those participants who
25 were spoken language users, reduced the sample size for the current phase of this study.

1 Additionally, both the screened and unscreened and the early and late confirmed groups of
2 teenagers showed very high within-group variation in their language outcomes. This high within
3 group variability shown by the D/HH teenagers, in combination with attrition of the sample over
4 time, may have limited the power of the study to detect significant effects of UNHS and early
5 confirmation. The question of benefit of UNHS and early confirmation to spoken language skills
6 should therefore be further examined in other population-based cohorts (e.g. Korver et al., 2010;
7 Wake et al., 2016) when they reach adolescence. [Individual participant meta-analysis combining
8 data across studies is likely to be valuable and should also be considered.](#)

9 Subgroup analysis indicated a differential benefit of early confirmation for those in the D/HH
10 sample with and without CIs. Caution must be taken in interpreting the results of subgroup analyses,
11 and particularly in this case where numbers of participants with CIs were small, however these
12 results indicate that early confirmation was not bringing the same benefits to the language
13 outcomes of those D/HH teenagers in the study with CIs as it did to those without. For the
14 participants in the D/HH sample who did not have a CI (this subgroup comprised 80% of the overall
15 sample), the effects of UNHS and confirmation of PCHL at ≤ 9 months of age were larger (range of
16 Cohen's *ds* 0.46 to 0.78) than they were for the whole sample and confirmation of PCHL at ≤ 9
17 months of age was associated with significant benefits to receptive language. By contrast, benefits of
18 confirmation of PCHL at ≤ 9 months of age were not apparent in the small group of participants
19 with CIs. This is consistent with the suggestion of Fitzpatrick et al. (2007) that the age at which these
20 children access effective intervention (i.e. the age they receive their CI) is likely to have more impact
21 on their language outcomes than the age at which their PCHL is identified; with the observation that
22 age at implantation predicts early language outcomes for D/HH children with CIs (Ching et al., 2013);
23 and with our own findings regarding literacy outcomes in this cohort (Pimperton et al., 2016). The
24 participants in this study were born at a time when age at implantation was typically much later than

1 it is in the present day even following early confirmation of PCHL: only one participant in this study
2 received a cochlear implant before the age of three years.

3 It is important to recognise however that age at implantation is unlikely to be the only
4 variable influencing language outcomes for the participants with CIs: other factors related to pre-
5 implant auditory experience are known to be important explanatory variables for variation in
6 language outcomes for children with CIs (Boons et al. 2012; Geers et al. 2008; Niparko et al. 2010). A
7 recent large-scale longitudinal study of children with CIs suggested a pattern of decreasing influence
8 of age at implantation on language outcomes as children move through middle childhood and
9 discussed other factors, such as the amount of time the CI is worn each day, that may drive variation
10 in language outcomes (Dunn et al. 2014). The sample of participants with CIs in the present study
11 was small but actually showed a trend in the opposite direction to the D/HH participants without CIs,
12 with the late confirmed group showing superior language skills to the early confirmed. One possible
13 explanation for this is that some of the participants with CIs in the late confirmed group may have
14 had an early, undocumented, progressive loss. This would have meant that, firstly, they may have
15 screened negative on UNHS and been more likely to be late confirmed, and secondly, that they
16 would have had some time with additional residual hearing prior to implantation, a factor previously
17 shown to predict better language outcomes post-implantation (Geers et al. 2008; Niparko et al.
18 2010).

19 As discussed above, within the subgroup of D/HH participants without CIs, early confirmation
20 was associated with significant benefits to receptive but not expressive language outcomes in
21 adolescence. A lack of sensitivity of the expressive language measure to the aspects of language that
22 are particularly vulnerable in D/HH children (e.g. inflectional morphology (Tomblin et al., 2015)) may
23 have contributed to this pattern of findings; on the expressive language measure used in this study,
24 MLU was calculated in words not in morphemes which would make it insensitive to deficits in
25 inflectional morphology, whereas one of the receptive language measures explicitly assesses

1 elements of inflectional morphological knowledge. In keeping with this potential differential
2 sensitivity of the receptive and expressive measures, the receptive language measures indicated
3 significant deficits for the D/HH group relative to the NH group while the expressive language
4 measure did not, a finding further considered at the end of the discussion. Rescoring the narrative
5 output from the ERRNI to more closely align with the constructs measured by the TROG may have
6 increased the sensitivity of this measure to differences in expressive morphology and syntax
7 between the early and late confirmed participants, as we found to be the case with a rescoring of
8 the Bus Story Narratives produced in the earlier phase of the current study when the participants
9 were aged 6-10 years (Worsfold et al., 2010).

10 As with the influence of age at implantation of language outcomes of D/HH teenagers with CIs, it
11 is important to emphasise that UNHS and early confirmation of PCHL is likely to be just one of a
12 multitude of variables that influence spoken language outcomes for D/HH teenagers without CIs. A
13 recent large-scale, longitudinal study of children [with mild to severe hearing loss](#) identified variables
14 associated with individual differences in their language outcomes ([Moeller & Tomblin., 2015](#)). These
15 variables related to access to language input and included variability in the quality of hearing aid
16 fitting, consistency of use of hearing aids, and characteristics of caregiver language input. It may be
17 the case that these variables associated with individual differences in language outcomes in D/HH
18 pre-schoolers have cumulative effects by the teenage years.

19 When examining the effects of UNHS and early confirmation on relative language gain from
20 middle childhood to adolescence, exposure to UNHS did not account for significant unique variance
21 in language scores at 13-19 years (T2) beyond that accounted for by existing language scores at 6-10
22 years (T1). Early confirmation of PCHL accounted for significant unique variance in T2 Expressive
23 Information score after adjusting for T1 Expressive Information score but not for T2 Expressive MLU
24 or Receptive scores after adjusting for corresponding T1 scores. The same pattern of significant
25 effects of early confirmation was evidenced in the subgroup of participants without CIs; early

1 confirmation of PCHL predicted significant unique variance in T2 Expressive Information but not in
2 Expressive MLU or Receptive. These findings suggest that the D/HH teenagers who had their hearing
3 loss confirmed early had made greater relative progress in one element of their expressive language
4 skills over the years subsequent to middle childhood, raising the possibility that earlier exposure to
5 language leading to better language skills in middle childhood may bring lasting benefits to later
6 language development. The relationship between expressive language z scores in middle childhood
7 with those in adolescence was much weaker than was the case for receptive language and
8 consequently the longitudinal expressive language models were a less good fit to the data; indeed
9 for expressive information, T1 scores did not account for any variance in T2 scores. This much
10 greater stability in receptive language skills may have contributed to the lack of a significant effect of
11 early confirmation on relative growth in receptive language skills from T1 to T2. The lack of stability
12 for expressive language may also indicate that the T1 and T2 expressive language measures are not
13 necessarily measuring the same sets of skills at both time points, a possibility considered further at
14 the end of the discussion.

15 Where there were significant benefits detected in this study, those were of early confirmation,
16 not of exposure to UNHS. Not all D/HH children who were exposed to UNHS in this sample were
17 confirmed early and some of those who were confirmed later *were* exposed to UNHS: 70% of the
18 early confirmed participants and 36% of the late confirmed participants in this study had been
19 exposed to UNHS at birth. UNHS is designed to take effect by allowing early confirmation of PCHL
20 and consequently early intervention to optimise the child's early communicative environment. If a
21 child is exposed to a UNHS programme but not screened, or is screened but not early confirmed,
22 then they are unable to access early intervention and the intended benefits of UNHS cannot be
23 realised. This emphasises the importance of ensuring that effective pathways are in place to follow-
24 up children picked up by UNHS, confirm the presence of PCHL, and initiate intervention within the
25 shortest possible timeframe (Kasai et al. 2012; Moeller et al. 2013). [The models that we constructed](#)
26 [took account of maternal education, English as first language and non-verbal ability but it is still](#)

1 possible that the relatively larger benefits to language associated with early confirmation of PCHL,
2 compared to those associated with birth in periods of UNHS, could have been contributed to by
3 residual confounding between other drivers, such as family engagement and efficacy, of both earlier
4 confirmation and superior language outcomes.

5 In contrast to the previous phase of this study when the D/HH children aged 6-10 years showed
6 significant deficits relative to the NH group in both receptive and expressive language (Kennedy et al.
7 2006), the D/HH teenagers showed significant deficits relative to the NH group in receptive, but not
8 expressive, language skills. There was some evidence of selective loss from the study of NH
9 participants whose mothers had lower educational qualifications at the earlier assessment time
10 point. However, when looking at a directly comparable sample (i.e. only those D/HH and NH
11 participants who provided receptive and expressive language data at both time points), the pattern
12 of apparently resolved deficits on the expressive language task in the face of persistent deficits on
13 the receptive language task for the D/HH group was still clear suggesting it cannot be attributed to
14 changes in the study sample between the two assessment time points. Additionally, when examining
15 standard scores for the D/HH group, which provide an indication of how they are performing relative
16 to the large hearing samples on which the language tests were standardised, the D/HH showed
17 standard scores that were near or above the mean for expressive language and 1SD below the mean
18 for receptive language, suggesting that the pattern of D/HH language performance was not a
19 function of the NH comparison group included in this study.

20 The question remains then as to why the expressive language deficit of the D/HH
21 participants is no longer evident while their receptive language deficit has remained consistent from
22 the primary to the secondary school years. One possibility relates to the tasks used to assess
23 receptive and expressive language. While the receptive language tasks were the same at both
24 assessment time points, the expressive language task used at 6-10 years (the Bus Story; designed for
25 3-8 year olds) differed from that used at 13-19 years (the ERRNI; designed for use from 4 years of

1 age to adulthood) because the ERRNI is a more age-appropriate assessment for a teenage sample.
2 The ERRNI was nevertheless selected as a measure that was as comparable as possible to the Bus
3 Story: both tests involve the participant viewing a series of pictures that tell a story and producing a
4 narrative based on the pictures. They differ, however, in that the Bus Story test administrator gives
5 the children a model spoken narrative whereas in the ERRNI they must produce their own narrative
6 solely based on the pictures. Skills related to the reception and retention of the model story in the
7 Bus Story assessment may therefore have given the hearing children an advantage at the earlier
8 assessment time point which was not the case with the ERRNI at the second assessment time point.
9 Indeed, this differential dependence of the receptive and expressive language tasks on auditory
10 access may have been a contributing factor to the discrepancy we observed at the present time
11 point in terms of D/HH deficits relative to the NH group on these tasks. This would be in addition to
12 the factor discussed earlier regarding the differential sensitivity of the receptive and expressive
13 language measures to the aspects of language that are most challenging for D/HH individuals (e.g.
14 inflectional morphology).

15 It is also possible that some of the D/HH participants have learnt, as they have got older, to
16 use compensatory language strategies which can be successfully deployed on the expressive
17 narrative task, but not on the receptive language tasks where there is simply a right or wrong
18 answer. One strategy, for example, might be to produce a lengthy response to the request for a
19 narrative which would be more likely to cover the key information points from the story and hence
20 increase the information score. Similarly, the mean length of utterance score does not reflect *quality*
21 of expressive language as it measures only the length of the utterances. Two participants could score
22 identically on mean length of utterance but the complexity and variety of the language used in their
23 utterances could be different (e.g. listing items within an utterance would increase the length of the
24 utterance but not necessarily the complexity of the language used). Again, a strategy focused on
25 producing a high volume of language is likely to inflate MLU scores.

1 The longitudinal design and population-based sample are strengths of this cohort study.
2 However, the duration of the study, in which children have been followed up over many years,
3 inevitably led to attrition of the study sample. The reduced sample of D/HH teenagers that provided
4 spoken language data at the present assessment time point was similar to those that did not in
5 terms of many key demographic characteristics though there was some evidence of selective
6 attrition of those participants who did not have English as a first language in the home, so caution
7 should be exercised when generalising these results to that population. The teenagers who provided
8 spoken language data in this phase of the study showed higher T1 receptive language scores than
9 those who did not. However, it is important to note that because this phase of the study collected
10 spoken language data only from spoken language users, this meant that sign language users who
11 were retained in this phase of the study were counted as non-participants for this examination of
12 spoken language outcomes despite some having provided receptive language data as children. This
13 inflated the T1 receptive language difference between the participants and non-participants because
14 these teenagers were more likely to have had low receptive language scores at T1; comparison of
15 the overall retained and non-retained samples for this phase of the study which included these sign
16 language users did not show higher receptive language skills in those who were retained.

17 Ideally we would have been able to include both the spoken and sign language users within the
18 same language analyses but the lack of directly comparable standardised tests for speech and sign
19 language users means it is difficult to make comparisons between the language skills of these two
20 groups. As mentioned previously, the inclusion of only those participants who used spoken language
21 reduced the sample size for these spoken language analyses; our work on reading comprehension
22 outcomes in this cohort as teenagers (Pimperton et al., 2016) did include both speech and sign
23 language users and found significant benefits of early confirmation to reading comprehension at the
24 whole group level. The results in this paper also do not address outcomes for those D/HH teenagers
25 who have significant additional disabilities that preclude them from completing the language

1 assessments. The impact of screening and early confirmation of PCHL on language outcomes for
2 these individuals remains unquantified.

3 In summary, significant benefits of UNHS exposure on teenage spoken language outcomes
4 were not demonstrated within the context of this study. Long-term significant benefits of early
5 confirmation of PCHL to spoken language outcomes were only detectable for those D/HH teenagers
6 who did not have CIs within this cohort and were not present for all language outcomes. High within-
7 group variability, a sample size reduced by attrition, and a lack of sensitivity of some measures may
8 have limited the power of this study to detect significant effects of early confirmation and of UNHS
9 exposure; further examination of the impact of UNHS on spoken language outcomes when larger
10 cohorts reach adolescence, [including individual participant meta-analysis combining data across](#)
11 [studies](#), would be valuable.

12

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1 References

- 2 Bishop, D. V. M. (2003). Test for Reception of Grammar Version 2. London: Pearson Assessment.
- 3 Bishop, D. V. M. (2004). Expression, Reception and Recall of Narrative Instrument. London: Harcourt
4 Assessment.
- 5 Boons, T., Brokx, J. P. L., Dhooge, I., et al. (2012). Predictors of Spoken Language Development
6 Following Pediatric Cochlear Implantation. *Ear and Hearing, 33*, 627-639.
- 7 Ching, T. Y., Dillon, H., Marnane, V., Hou, S., Day, J., Seeto, M., . . . Yeh, A. (2013). Outcomes of early-
8 and late-identified children at 3 years of age: findings from a prospective population-based
9 study. *Ear and Hearing, 34*, 535-552.
- 10 Davis, A., Bamford, J., Wilson, I., et al. (1997). A critical review of the role of neonatal hearing
11 screening in the detection of congenital hearing impairment. *Health Technol Assess, 1*, i-iv,
12 1-176.
- 13 Dunn, C. C., Walker, E. A., Oleson, J., et al. (2014). Longitudinal speech perception and language
14 performance in pediatric cochlear implant users: the effect of age at implantation. *Ear Hear,*
15 *35*, 148-160.
- 16 Dunn, L. M., Dunn, D. M., National Foundation for Educational Research. (2009). The British Picture
17 Vocabulary Scale Third Edition. In. London: GL Assessment.
- 18 Eisenberg, L. S. (2007). Current state of knowledge: Speech recognition and production in children
19 with hearing impairment. *Ear and Hearing, 28*, 766-772.
- 20 Fitzpatrick, E., Durieux-Smith, A., Eriks-Brophy, A., et al. (2007). The impact of newborn hearing
21 screening on communication development. *Journal of Medical Screening, 14*, 123-131.
- 22 Geers, A., Tobey, E., Moog, J., et al. (2008). Long-term outcomes of cochlear implantation in the
23 preschool years: From elementary grades to high school. *International Journal of Audiology,*
24 *47*, S21-S30.
- 25 Hamel, R., Schmittman, V. D. (2006). The 20-minute version as a predictor of the Raven Advanced
26 Progressive Matrices Test. *Educational and Psychological Measurement, 66*, 1039-1046.
- 27 Howell, R. R., Terry, S., Tait, V. E., et al. (2012). CDC Grand Rounds: Newborn Screening and
28 improved Outcomes. *Morbidity and Mortality Weekly Report, 61*, 390-393.
- 29 Kasai, N., Fukushima, K., Omori, K., et al. (2012). Effects of Early Identification and Intervention on
30 Language Development in Japanese Children With Prelingual Severe to Profound Hearing
31 Impairment. *Annals of Otolaryngology and Laryngology, 121*, 16-20.
- 32 Kennedy, C. R., Kimm, L., Dees, D. C., et al. (1998). Controlled trial of universal neonatal screening for
33 early identification of permanent childhood hearing impairment. *Lancet, 352*, 1957-1964.
- 34 Kennedy, C. R., McCann, D., Campbell, M. J., et al. (2005). Universal newborn screening for
35 permanent childhood hearing impairment: an 8-year follow-up of a controlled trial. *Lancet,*
36 *366*, 660-662.
- 37 Kennedy, C. R., McCann, D. C., Campbell, M. J., et al. (2006). Language ability after early detection of
38 permanent childhood hearing impairment. *New England Journal of Medicine, 354*, 2131-
39 2141.
- 40 Korver, A. M., Konings, S., Dekker, F. W., et al. (2010). Newborn hearing screening vs later hearing
41 screening and developmental outcomes in children with permanent childhood hearing
42 impairment. *JAMA, 304*, 1701-1708.
- 43 Luckner, J. L., Cooke, C. (2010). A summary of the vocabulary research with students who are deaf or
44 hard of hearing. *American Annals of the Deaf, 155*, 38-67.
- 45 Lyness, C. R., Woll, B., Campbell, R., et al. (2013). How does visual language affect crossmodal
46 plasticity and cochlear implant success? *Neuroscience and Biobehavioral Reviews, 37*, 2621-
47 2630.
- 48 Meinzen-Derr, J., Wiley, S., Choo, D. I. (2011). Impact of early intervention on expressive and
49 receptive language development among young children with permanent hearing loss. *Am*
50 *Ann Deaf, 155*, 580-591.

1 Moeller, M. P., Carr, G., Seaver, L., et al. (2013). Best practices in family-centered early intervention
2 for children who are deaf or hard of hearing: An international consensus statement. *Journal*
3 *of Deaf Studies and Deaf Education*, 18, 429-445.

4 Moeller, M. P., & Tomblin, J. B. (2015). Epilogue: Conclusions and Implications for Research and
5 Practice. *Ear and Hearing*, 36, 92S-98S.

6 Moeller, M. P., Tomblin, J. B., Yoshinaga-Itano, C., et al. (2007). Current state of knowledge:
7 Language and literacy of children with hearing impairment. *Ear and Hearing*, 28, 740-753.

8 Nelson, H. D., Bougatsos, C., Nygren, P. (2008). Universal newborn hearing screening: Systematic
9 review to update the 2001 US Preventive Services Task Force Recommendation. *Pediatrics*,
10 122, E266-E276.

11 Niparko, J. K., Tobey, E. A., Thal, D. J., et al. (2010). Spoken Language Development in Children
12 Following Cochlear Implantation. *Jama-Journal of the American Medical Association*, 303,
13 1498-1506.

14 Pimperton, H., Blythe, H., Kreppner, J., et al. (2016). The impact of universal newborn hearing
15 screening on long-term literacy outcomes: A prospective cohort study. *Arch Dis Child*, 101, 9-
16 15.

17 Pimperton, H., Kennedy, C. R. (2012). The impact of early identification of permanent childhood
18 hearing impairment on speech and language outcomes. *Archives of Disease in Childhood*, 97,
19 648-653.

20 Renfrew, C. (1995). *Bus Story Manual: A Test of Narrative Speech* (3rd Edition). Oxford, England:
21 Renfrew/Winslow.

22 Styles, I., Raven, M., Raven, J. C. (1998). *Standard Progressive Matrices - Plus Version*. London:
23 Pearson.

24 Thomas, M. S. C., Johnson, M. H. (2008). New advances in understanding sensitive periods in brain
25 development. *Current Directions in Psychological Science*, 17, 1-5.

26 Thompson, D. C., McPhillips, H., Davis, R. L., et al. (2001). Universal newborn hearing screening -
27 Summary of evidence. *Jama-Journal of the American Medical Association*, 286, 2000-2010.

28 Tomblin, J.B., Harrison, M., Ambrose, S.E., Walker, E.A., Oleson, J.J., & Moeller, M.P. (2015).
29 Language outcomes in young children with mild to severe hearing loss. *Ear and Hearing*, 36,
30 76S-91S.

31 Tucker, S.M., & Bhattacharya, J. (1992). Screening of hearing impairment in the newborn using the
32 auditory response cradle. *Archives of Disease in Childhood*, 67, 911-919.

33 Wake, M., Ching, T.Y.C., Wirth, K., et al. (2016). Population outcomes of three approaches to
34 detection of congenital hearing loss. *Pediatrics*, 137, e20151722.

35 Watkin, P.M., & Baldwin, M. (1999). Confirmation of deafness in infancy. *Archives of Disease in*
36 *Childhood*, 81, 380-389.

37 Whitehouse, A.J.O., Line, E.A., Watt, H.J., & Bishop, D.V.M. (2009). Qualitative aspects of
38 developmental language impairment relate to language and literacy outcome in adulthood.
39 *International Journal of Language and Communication Disorders*, 44, 489-510.

40 Worsfold, S., Mahon, M., Yuen, H. M., et al. (2010). Narrative skills following early confirmation of
41 permanent childhood hearing impairment. *Dev Med Child Neurol*, 52, 922-928.

42 Yoshinaga-Itano, C., Coulter, D., Thomson, V. (2000). The Colorado Newborn Hearing Screening
43 Project: effects on speech and language development for children with hearing loss. *J*
44 *Perinatol*, 20, S132-137.

Figure Legends

Figure 1. Flowchart of progression of participants through the trial. The greyed section relates to the previous phase of this research study at 6-10 years old (T1) while the section below that relates to the current phase at 13-19 years old (T2).

Figure 2. Mean age-adjusted receptive and expressive language z scores for the early- (filled circles) and late- (unfilled circles) confirmed and the UNHS (filled circles) and no UNHS (unfilled circles) D/HH participants. Error bars show 95% confidence intervals around the mean.

Table 1: Demographic characteristics at T1 (6-10 years assessment time-point) for participants who provided spoken language data at T1 and who either did or did not provide spoken language data in the current study of language outcomes in teenagers (T2)

		D/HH Participants		Participants with normal hearing	
		T2 Language Data (<i>n</i> =59)*	No T2 Language Data (<i>n</i> =42)	T2 Language Data (<i>n</i> =38)	No T2 Language Data (<i>n</i> =25)
Mean age (years) at T1 assessment (SD)		7.88 (1.09)	8.03 (1.58)	8.02 (1.08)	8.30 (0.99)
[range]		[5.75 to 10.67]	[5.50 to 11.67]	[6.25 to 9.75]	[6.42 to 9.67]
Female sex <i>n</i> (%)		29 (49)	16 (38)	13 (34)	13 (52)
Severity of hearing loss <i>n</i> (%)	Moderate	31 (53)	26 (62)		
	Severe	13 (22)	10 (24)	n/a	n/a
	Profound	15 (25)	6 (14)		
Cochlear Implant <i>n</i> (%)		9 (15)	3 (7)	n/a	n/a
Born in periods with UNHS <i>n</i> (%)		31 (52)	21 (50)	n/a	n/a
PCHL confirmed \leq9 months <i>n</i> (%)		26 (44)	19 (45)	n/a	n/a

English as main language at home <i>n</i> (%)		52 (88)	30 (71)	36 (95)	24 (96)
Maternal education <i>n</i> (%)	No quals or <5 O-levels or equiv^a	17 (29)	18 (43)	11 (29)	14 (56)
	≥5 O-levels or any A-levels or equiv^a	34 (58)	21 (50)	16 (42)	9 (36)
	University or higher degree or equiv	8 (14)	3 (7)	11 (29)	2 (8)
Mean (SD) age-adjusted receptive language z score at primary school assessment		-1.78 (1.44)	-2.41 (1.78)	0.19 (0.83)	-0.29 (0.84)

*One participant provided T2 language data but not T1.

PCHL=Permanent childhood hearing loss. UNHS=Universal newborn hearing screening. n/a=not applicable, quals=qualifications. equiv=equivalent.

^aO-level examinations (now replaced by general certificates of education) are usually taken at 16 years of age; five or more O levels was a benchmark for access to some further education courses; A-level examinations (now replaced by A2s) are taken two years later as qualifications for entry to higher education.

Table 2: Characteristics of participating teenagers who provided spoken language data

		D/HH Participants (<i>n</i> =60)		Participants with normal hearing (<i>n</i> =38)
		Confirmation of PCHL at ≤9 months (<i>n</i> =27)	Confirmation of PCHL at >9months (<i>n</i> =33)	
Mean (SD) age at assessment in years		16.85 (1.55)	17.32 (1.36)	16.3 (1.2)
Female sex <i>n</i> (%)		13 (48)	17 (52)	13 (34)
Born in period with UNHS <i>n</i> (%)		19 (70)	12 (36)	n/a
Severity <i>n</i> (%)	Moderate+	13 (48)	15 (45)	n/a
	Severe	6 (22)	10 (30)	
	Profound	8 (30)	8 (24)	
Hearing device used <i>n</i> (%)	Cochlear implant/s	6 (22)	6 (18)	n/a
	Hearing aid/s	19 (70)	27 (82)	
	No hearing device	2 (7)*	0 (0)	
Mean (SD) non-verbal ability z-score ^a		-0.28 (0.88)	-0.23 (0.83)	0 (1)
Aetiology <i>n</i> (%)	Syndromic	4 (15)	2 (6)	n/a
	Other hereditary	6 (22)	10 (30)	
	Known non-genetic risk ^b	1 (4)	2 (6)	
	Not known	16 (59)	19 (58)	
English as main language at home <i>n</i> (%)		27 (100)	29 (88)	36 (95)
Maternal education <i>n</i> (%)	No quals/<5 O-levels or equiv ^c	8 (30)	5 (15)	6 (16)
	≥5 O-levels/A-levels or equiv ^c	12 (44)	20 (61)	14 (37)
	Univ/higher degree or equiv	7 (26)	8 (24)	18 (47)

PCHL=Permanent childhood hearing loss. UNHS=Universal newborn hearing screening. n/a=not applicable. quals=qualifications. Equiv=equivalent. Univ= university

^a: Age-adjusted z-scores are listed for Ravens Progressive Matrices total score. The z-scores are the number of standard deviations of the scores in normally hearing children by which the age-adjusted score differed from the mean score in the normally hearing children.

^b: Prematurity or cerebral palsy

^c: O-level examinations (now replaced by General Certificate of Secondary Education) were usually taken at 16 years of age; five or more O levels was a benchmark for access to some further education courses; A-level examinations (now replaced by A2s) were taken two years later as qualifications for entry to higher education.

+ : Six participants (two with confirmation of PCHL at ≤ 9 months, 4 with confirmation of PCHL > 9 months) classified with PCHL of moderate severity at T1 had shown improvements by the current study such that their better ear hearing thresholds now fell between 30dB and 40dB.

*: Both with moderate PCHL who were not current hearing aid users

Table 3. Group mean receptive and expressive language z-scores and standard scores for D/HH teenagers and teenagers with normal hearing.

	D/HH Mean (SD)	NH Mean (SD)	Unadjusted Mean Difference (95% CI)	(p)	Adjusted¹ Mean Difference (95% CI)	(p)	d
Receptive	<i>N = 60</i>	<i>N = 38</i>					
Composite z score	-2.02 (2.46)	0 (0.84)	-2.02 (-2.84 to -1.19)	<.001	-1.71 (-2.51 to -0.92)	<.001	0.89
<i>Composite SS*</i>	<i>86.73 (13.51)</i>	<i>99.67 (7.90)</i>					
Expressive	<i>N = 59</i>	<i>N = 38</i>					
Information z score	-0.18 (1.13)	0 (0.92)	-0.18 (-0.62, 0.25)	.40	-0.23 (-0.70, 0.24)	.33	0.20
<i>Information SS</i>	<i>108.41 (14.47)</i>	<i>110.92 (11.85)</i>					
MLU z score	-0.18 (0.91)	0 (1.00)	-0.18 (-0.57, 0.22)	.38	0.03 (-0.44, 0.38)	.90	0.03
<i>MLU SS</i>	<i>97.08 (11.69)</i>	<i>100.18 (12.22)</i>					

D/HH = deaf or hard of hearing; NH = normal hearing; SS = Standard score

¹ Adjusted regression models were adjusted for mother's education, age-adjusted total Raven's Progressive Matrices scores, English as the main language at home.

*N = 9 D/HH participants had raw scores below the basal standard score of 70 on the BPVS and were allocated a standard score of 69, thus limiting the capacity of these standard scores to reflect the variability in raw scores of these lower-performing participants.

Table 4: Receptive and expressive language z-scores for D/HH teenagers by birth in periods with and without universal newborn hearing screening and by age of confirmation of hearing loss.

All D/HH participants:

	UNHS	No UNHS	Unadjusted	(p)	Adjusted¹	(p)	d
	Mean (SD)	Mean (SD)	Mean Difference (95% CI)		Mean Difference (95% CI)		
Receptive	<i>N = 31</i>	<i>N = 29</i>					
Composite z score	-1.79 (2.52)	-2.26 (2.42)	0.47 (-0.80 to 1.75)	.46	0.68 (-0.56 to 1.93)	.28	.28
<i>Composite SS*</i>	<i>88.16 (13.65)</i>	<i>85.21 (13.42)</i>					
Expressive	<i>N = 30</i>	<i>N = 29</i>					
Information z score	-0.11 (1.03)	-0.26 (1.23)	0.15 (-0.44 to 0.74)	.62	0.40 (-0.26 to 1.05)	.23	.32
<i>Information SS</i>	<i>109.20 (12.90)</i>	<i>107.59 (16.12)</i>					
MLU z score	0.02 (1.01)	-0.38 (0.77)	0.39 (-0.07 to 0.86)	.10	0.45 (-0.08 to 0.98)	.10	.43
MLU SS	<i>99.13 (13.09)</i>	<i>94.97 (9.81)</i>					
	Early Confirmed	Late Confirmed					
Receptive	<i>N = 27</i>	<i>N = 33</i>					
Composite z score	-1.60 (2.50)	-2.36 (2.42)	0.76 (-0.51 to 2.04)	.24	0.95 (-0.22 to 2.11)	.11	.42
<i>Composite SS*</i>	<i>88.54 (14.95)</i>	<i>85.26 (12.24)</i>					

Expressive	<i>N = 26</i>	<i>N = 33</i>					
Information z score	0.004 (1.17)	-0.33 (1.08)	0.34 (-0.25 to 0.93)	.26	0.43 (-0.20, 1.05)	.18	.35
<i>Information SS</i>	<i>110.35 (15.51)</i>	<i>106.88 (13.64)</i>					
MLU z score	0.01 (0.80)	-0.32 (0.98)	0.34 (-0.14 to 0.81)	.16	0.22 (-0.30, 0.74)	.40	.22
<i>MLU SS</i>	<i>99.08 (10.08)</i>	<i>95.52 (12.75)</i>					

¹ Adjusted regression models were adjusted for mother's education, severity of permanent childhood hearing loss, age-adjusted total Raven's Progressive Matrices scores, English as the main language at home.

*N = 9 D/HH participants had raw scores below the basal standard score of 70 on the BPVS and were allocated a standard score of 69, thus limiting the capacity of these standard scores to reflect the variability in raw scores of these lower-performing participants.

D/HH stratified by CI vs. no CI:

No CI	Early Confirmed	Late Confirmed	Unadjusted	(p)	Adjusted¹	(p)	d
	Mean (SD)	Mean (SD)	Mean Difference (95% CI)		Mean Difference (95% CI)		
Receptive	<i>N = 21</i>	<i>N = 27</i>					
Composite z score	-1.28 (2.39)	-2.48 (2.51)	1.21 (-0.23 to 2.65)	.098	1.55 (0.38 to 2.71)	.01	.78
<i>Composite SS</i>	<i>91.10 (13.33)</i>	<i>84.59 (13.01)</i>					
Expressive	<i>N = 20</i>	<i>N = 27</i>					
Information z score	0.10 (1.08)	-0.45 (1.01)	0.55 (-0.07 to 1.17)	.08	0.57 (-0.06 to 1.21)	.08	.52

<i>Information SS</i>	<i>112.18 (13.55)</i>	<i>105.57 (13.09)</i>					
MLU z score	0.11 (0.83)	-0.50 (0.89)	0.62 (0.10 to 1.13)	.02	0.47 (-0.07 to 1.01)	.09	.51
<i>MLU SS</i>	<i>100.40 (10.20)</i>	<i>93.11 (11.61)</i>					
CI							
Receptive	<i>N = 6</i>	<i>N = 6</i>					
Composite z score	-2.73 (2.75)	-1.80 (2.07)	-0.92 (-4.05 to 2.20)				
<i>Composite SS</i>	<i>79.58 (18.09)</i>	<i>88.25 (8.10)</i>					
Expressive	<i>N = 6</i>	<i>N = 6</i>					
Information z score	-0.32 (1.52)	0.18 (1.33)	-0.50 (-2.34, 1.33)				
<i>Information SS</i>	<i>104.25 (21.17)</i>	<i>112.75 (15.75)</i>					
MLU z score	-0.31 (0.61)	0.48 (1.03)	-0.79 (-1.89, 0.30)				
<i>MLU SS</i>	<i>94.67 (9.07)</i>	<i>106.33 (12.96)</i>					

¹ Adjusted regression models were adjusted for mother's education, severity of permanent childhood hearing loss, age-adjusted total Raven's Progressive Matrices scores, English as the main language at home.

Table 5: Hierarchical multiple linear regression analyses predicting language scores at T2 for the D/HH participants*

		R ²	R ² Change	F change	P
Predicting T2 Receptive Language (N = 59)					
<i>Step 1</i>	T1* Receptive Language	.62	.62	94.56	<.001
<i>Step 2</i>	Non-verbal Ability; Maternal Education; Severity of PCHI; English as Main Language	.71	.08	3.84	.008
<i>Step 3</i>					
Model 1	UNHS vs. no UNHS	.72	.01	2.68	.11
Model 2	Early vs. Late Confirmation	.71	.01	1.01	.32
Predicting T2 Expressive Information (N = 54)					
<i>Step 1</i>	T1* Expressive Information	.00	.00	.02	.89
<i>Step 2</i>	Non-verbal Ability; Maternal Education; Severity of PCHI; English as Main Language	.14	.14	1.99	.11
<i>Step 3</i>					
Model 1	UNHS vs. no UNHS	.19	.05	2.61	.11
Model 2	Early vs. Late Confirmation	.22	.08	4.78	.03
Predicting T2 Expressive MLU (N = 54)					
<i>Step 1</i>	T1* Expressive MLU	.09	.09	5.11	.03
<i>Step 2</i>	Non-verbal Ability; Maternal Education; Severity of PCHI; English as Main Language	.13	.04	0.62	.65
<i>Step 3</i>					
Model 1	UNHS vs. no UNHS	.19	.06	3.33	.07
Model 2	Early vs. Late Confirmation	.15	.01	0.65	.42

*T1 refers to the time of the first language assessment undertaken at 6-10 years in a previous phase of this study. T2 refers to the current phase of this study when language assessment was undertaken at 13-19 years.