The impact of universal newborn hearing screening on long-term literacy outcomes: a prospective cohort study

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ABSTRACT

Objective To determine whether the benefits of universal newborn hearing screening (UNHS) seen at age 8 years persist through the second decade.

Design Prospective cohort study of a population sample of children with permanent childhood hearing impairment (PCHI) followed up for 17 years since birth in periods with (or without) UNHS.

Setting Birth cohort of 100 000 in southern England.

Participants 114 teenagers aged 13–19 years, 76 with PCHI and 38 with normal hearing. All had previously their reading assessed aged 6–10 years.

Interventions Birth in periods with and without UNHS; confirmation of PCHI before and after age 9 months.

Main outcome measure Reading comprehension ability. Regression modelling took account of severity of hearing loss, non-verbal ability, maternal education and main language.

Results Confirmation of PCHI by age 9 months was associated with significantly higher mean z-scores for reading comprehension (adjusted mean difference 1.17, 95% CI 0.36 to 1.97) although birth during periods with UNHS was not (adjusted mean difference 0.15, 95% CI −0.75 to 1.06). The gap between the reading comprehension z-scores of teenagers with early compared with late confirmed PCHI had widened at an adjusted mean rate of 0.06 per year (95% CI −0.02 to 0.13) during the 9.2-year mean interval since the previous assessment.

Conclusions The benefit to reading comprehension of confirmation of PCHI by age 9 months increases during the teenage years. This strengthens the case for UNHS programmes that lead to early confirmation of permanent hearing loss.

Trial registration number ISRCTN03307358.

INTRODUCTION

Bilateral permanent childhood hearing impairment (PCHI) of moderate, severe or profound severity is the most common sensory disability affecting 1 in 750 children and is present at birth in more than 80% of affected children.¹ PCHI of this degree has a detrimental impact on aspects of oral language development²−⁴ and impacts significantly on skills that depend on language ability, such as reading and writing.⁵⁻⁶ Identification of PCHI in early childhood enables affected children to receive early intervention to optimise their language access during a ‘sensitive period’ for language development.⁷ More than half of babies born with PCHI do not have prospectively identifiable risk factors so that only universal newborn hearing screening (UNHS) programmes can identify the majority of those affected. UNHS, when first introduced in the UK, more than doubled the proportion confirmed by 9 months to three-quarters of all cases of bilateral PCHI ≥40 dB.⁸⁻¹⁰ We have previously reported that children with PCHI from that birth cohort had significant benefits to language and reading at age 6–10 years associated with birth in periods with UNHS and with confirmation of PCHI by age 9 months.¹¹⁻¹³

Systematic reviews have been increasingly supportive of UNHS¹⁴⁻¹⁶ and both the UK National Screening Committee and the US Preventative Services Task Force have recommended in favour of it.¹⁷⁻¹⁹ During the calendar year of 2009, an estimated 5073 cases of PCHI were detected by UNHS in the USA, accounting for 43% of all detected cases of the 29 medical conditions for which newborn screening is recommended.²⁰ Both the US Preventative Services Task Force¹⁵ ¹⁷ and a 2009 WHO report on UNHS²¹ have,
however, drawn attention to the evidence gap regarding benefits beyond primary school age and benefits to functional outcomes. This study consequently aimed to provide novel evidence regarding the effects of UNHS and early confirmation of PCHI on functional outcomes in the teenage years. We report findings regarding the abilities of teenagers with PCHI at age 17 years whom we previously assessed at age 8 years. Reading is a skill that is dependent on underlying language ability that relates very closely to educational and employment outcomes, and as such is a key functional outcome. Reading comprehension was therefore prespecified as the primary outcome in this study.

**PATIENTS AND METHODS**

The children in this prospective follow-up study, 120 children with bilateral PCHI ≥40 decibels hearing level (dB HL) (not known to be postnatally acquired) and a comparison group of 63 normally hearing children, were drawn from a birth cohort of 157,000 children born in eight districts of southern England (see online supplementary appendix 1), of whom about half were born in periods with UNHS. We previously reported a number of details relating to this population in infancy and first decade, including the UNHS programmes for PCHI to which they were exposed; the service provision by district and regional audiology and by other services for confirmation and management of their PCHI; and the language and reading abilities of the children at 6–10 years. Nine years after their previous language and reading assessments at 6–10 years, 76 (63%) teenagers with PCHI and 38 (60%) of the normally hearing comparison group have now participated in the study we report here (figure 1). We estimate that 49% of all oral language users with PCHI from the birth cohort had their reading assessed at age 17.1 years (see online supplementary appendix 1).

**Figure 1** Numbers of teenagers with permanent childhood hearing impairment who were eligible for the study and assessed for reading ability at primary school and teenage. Greyed out section of the figure indicates the previous study at age 6–10 years. dB HL, decibels hearing level.
Procedure
Each participant was assessed by a trained researcher, unaware of their audiological history, using the York Assessment of Reading for Comprehension Secondary Edition,32 a standardised reading test that provides measures of accuracy, comprehension and summarisation skill (see online supplementary appendix 1). A 20 min timed version13 of Raven’s Standard Progressive Matrices Plus31 was used as a measure of non-verbal ability. The preplanned primary outcome of our study was reading comprehension score after adjustment in a multiple linear regression for severity of hearing loss, non-verbal ability and maternal education, which were recognised as potential confounders of the primary outcome.11 Adjusted reading accuracy and reading summarisation ability z-scores were preplanned secondary outcomes.

Severity of hearing impairment was categorised from the most recent audiological evaluation at audiology and cochlear implant clinics as moderate (40–69 dB HL), severe (70–94 dB HL) or profound (≥95 dB HL) according to four-frequency averaging of the pure-tone thresholds at 0.5, 1, 2 and 4 kHz. Maternal education was classified according to the 2001 census in the UK.

This study was approved by the Southampton and SW Hampshire Research Ethics Committee. Written informed consent for participation in the study was obtained from principal caregivers and from the teenage participants themselves.

Analysis strategies
The primary outcome (reading comprehension) and the analysis strategy were prespecified and the statistical analysis plan was written before examination of the data. The target sample size of 96 with half of the sample born in periods with (or without) UNHS, or, in a parallel set of analyses, exposed to early (or late) confirmation of PCHI, was sufficient to have 90% power to detect a standardised difference in the primary outcome of at least 0.67 SDs at a 5% significance level (two sided) using a univariate model. Where statistically significant inter-group differences were found, subgroup analysis was then undertaken in those who had and had not received cochlear implants. The method of adjusting reading z-scores appropriately to look at change in reading ability over time comparing current scores with those previously obtained at aged 6–10 years is described in online supplementary appendix 1.

RESULTS
The 114 participating teenagers were similar to the 183 who had previously participated in the study of reading and language at 7.9 years with regard to sex, non-verbal ability and maternal educational level at the time of the previous study (table 1). The 76 participants with PCHI (figure 1) were similar to the 120

<table>
<thead>
<tr>
<th>Characteristic</th>
<th>Children with bilateral PCHI</th>
<th>Normally hearing children</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Whole sample (n=120)</td>
<td>Teenage sample participating in present study (n=76)</td>
</tr>
<tr>
<td>Mean age (SD) (range) in years</td>
<td>7.9 (1.3)</td>
<td>7.9 (1.1)</td>
</tr>
<tr>
<td>At primary school assessment</td>
<td>5.4 to 11.7</td>
<td>5.8 to 10.7</td>
</tr>
<tr>
<td>Female sex n (%)</td>
<td>53 (44)</td>
<td>37 (49)</td>
</tr>
<tr>
<td>Severity of hearing loss n (%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Moderate</td>
<td>62 (52)</td>
<td>38 (50)</td>
</tr>
<tr>
<td>Severe</td>
<td>29 (24)</td>
<td>16 (21)</td>
</tr>
<tr>
<td>Profound</td>
<td>29 (24)</td>
<td>22 (29)</td>
</tr>
<tr>
<td>Born in periods with UNHS n (%)</td>
<td>61 (51)</td>
<td>37 (49)</td>
</tr>
<tr>
<td>PCHI confirmed ≤9 months n (%)</td>
<td>57 (48)</td>
<td>35 (46)</td>
</tr>
<tr>
<td>English as main language at home n (%)</td>
<td>99 (83)</td>
<td>67 (88)</td>
</tr>
<tr>
<td>Maternal education n (%)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>No qualifications or &lt;5 O-levels†</td>
<td>42 (36)</td>
<td>24 (32)</td>
</tr>
<tr>
<td>≥5 O-levels or some A-levels†</td>
<td>62 (52)</td>
<td>40 (53)</td>
</tr>
<tr>
<td>University or higher degree</td>
<td>14 (12)</td>
<td>12 (16)</td>
</tr>
</tbody>
</table>

*The ‘whole sample’ was a population-based sample of children with PCHI and a normally hearing comparison group that participated 9 years earlier in a study of language and reading at primary school age.
†0-level examinations (now replaced by general certificates of education) are usually taken at 16 years of age; A-level examinations (now replaced by A2s) are taken 2 years later as qualifications for entry to higher education.
NA, not applicable; PCHI, permanent childhood hearing impairment (see Patients and methods section for detailed definition of degree of PCHI); UNHS, universal newborn hearing screening.
who had previously participated with regard to severity of PCHI, exposure to UNHS and confirmation of PCHI prior to nine completed months from birth (table 1). These characteristics were also similar between those who had their PCHI confirmed by age 9 months (n=35) and those who had it confirmed later (n=41) (table 2) and between those who were born in periods with UNHS (n=37) and those who were not (n=39) (data not shown). The early and late confirmed PCHI groups were similar with respect to the percentages affected by cerebral palsy, visual disability or learning disability (table 2). These groups were also similar in that the aetiology of deafness was identified in about half and was genetic in 34%–43% (table 2). The proportion of teenagers in whom English was not the main language at home was higher in the late (12%) than the early (3%) confirmed group and adjustment for this variable was therefore included in the regression model in addition to the three prespecified variables (see Patients and methods section).

The early and late confirmed groups showed mean reading comprehension z-scores that were 0.63 and 1.74 SDs, respectively, below the mean reading z-score in the normally hearing comparison group (table 3). The teenagers who had their hearing impairment confirmed by nine completed months of age had significantly higher adjusted mean z-scores than the later confirmed teenagers for both reading comprehension (1.17 SD) and reading summarisation (0.96 SD) (table 3). These effect sizes were larger in the 78% (51/65) who had not received cochlear implants (adjusted inter-group differences 1.29, 95% CI 0.52 to 2.07, p=0.002; 1.00, 95% CI 0.30 to 1.70, p=0.006, respectively). Adjusted inter-group z-score differences on the three reading outcome measures between all teenage participants who were or were not born in periods with UNHS at birth were smaller (0.09 to 0.22) and not statistically significant (table 3).

Change in the estimates of effect sizes and p values of early confirmation and of screening was minimal when they were modelled together rather than separately, suggesting that these effects were working independently (see online supplementary table e1). Adding the effect of early confirmation into the regression model after screening was included made a significant additional contribution to model fit (likelihood ratio test
### Table 3: Reading z-scores for children with bilateral PCHI by age of confirmation of PCHI and by birth in periods with and without UNHS

<table>
<thead>
<tr>
<th>Measure</th>
<th>Number of observations</th>
<th>Mean z-score (SD)</th>
<th>PCHI confirmed at ≤9 months</th>
<th>PCHI confirmed at &gt;9 months</th>
<th>p Value</th>
<th>Adjusted* mean difference (95% CI)</th>
<th>p Value</th>
</tr>
</thead>
<tbody>
<tr>
<td>YARC reading comprehension</td>
<td>28</td>
<td>–0.63 (1.63)</td>
<td>1.11 (0.33 to 1.89)</td>
<td>0.006</td>
<td>1.17 (0.36 to 1.97)</td>
<td>0.005</td>
<td></td>
</tr>
<tr>
<td>YARC reading summarisation</td>
<td>28</td>
<td>–0.56 (1.37)</td>
<td>0.80 (0.09 to 1.51)</td>
<td>0.03</td>
<td>0.96 (0.24 to 1.68)</td>
<td>0.01</td>
<td></td>
</tr>
<tr>
<td>YARC reading accuracy</td>
<td>27</td>
<td>–1.12 (1.69)</td>
<td>0.59 (–0.22 to 1.40)</td>
<td>0.15</td>
<td>0.68 (–0.09 to 1.46)</td>
<td>0.08</td>
<td></td>
</tr>
</tbody>
</table>

*Adjusted for severity of PCHI, maternal education level, non-verbal ability and English as main language at home.

### DISCUSSION

The study of teenagers with PCHI who were involved in trials of UNHS is the first to describe the effects of UNHS and early confirmation of PCHI on outcomes beyond the primary school age (see Patients and methods). The recalculation of reading comprehension scores obtained 3 years earlier from these individuals showed that those at age 13–19 years whose PCHI had been confirmed at an early age had main-group differences in reading comprehension that were more than 1 SD of the prespecified primary outcome variable. This was associated with significantly better performance on reading comprehension at age 19 years. Confirmation of PCHI by 9 months of age was associated with an increased annual rate of change of reading comprehension scores by 0.06 per year, 95% CI 0.02 to 0.13, p=0.04. This inter-group difference was larger in annual rate of change of reading comprehension, the adjusted mean annual rate of change in the late confirmed group, the unadjusted mean annual rate of change in the late confirmed group, the unadjusted mean annual rate of change in the late confirmed group, and the adjusted mean annual rate of change in the late confirmed group (mean inter-group rate difference 0.06 per year, 95% CI 0.02 to 0.13, p=0.04). This difference was accounted for by exposure to UNHS."
hearing peers and is likely to impact on their life chances through educational achievement and employment.\textsuperscript{24}

Non-verbal ability was very similar in the early and late confirmed groups and adjustment for it was included in the regression model. This suggests that the deficit in reading scores in the late confirmed participants did not result from a general cognitive deficit but rather from the specific impact of delayed access to optimal language input early in life on language-related abilities. The early and late confirmed groups did not show different proportions of genetic and non-genetic aetiologies of deafness nor of disabilities additional to deafness that might account for the observed differences in reading z-scores.

Factors other than age at confirmation of PCHI appeared to determine reading outcomes for that minority of participants who had received cochlear implants\textsuperscript{16} although this subgroup analysis was not preplanned and should be treated with caution. A greater dependency of teenage reading ability of the implanted subgroup on age at implantation than on age at confirmation may explain this difference but studies of larger numbers of cochlear implant users are needed to determine this.

The effects of early confirmation were seen in those born in periods with and without UNHS and the effect of UNHS appears to be dependent on the increase in rates of early confirmation of PCHI to which it leads. The same NHS district and regional audiology teams delivered, in almost all cases, the care of both screened and unscreened and of both early and late confirmed populations in this study\textsuperscript{11} and the different outcomes between these groups are likely to reflect the effect of UNHS and of early confirmation rather than any differences in the services to which they were exposed. A 2013 birth cohort in the UK would, nevertheless, be likely to show a much stronger relationship between birth in periods with UNHS and reading outcomes. Effective postscreening audiology and other services for those screening positive for PCHI in the newborn period, which were largely absent in the period from 1992 to 1997 for the population described in this report, are now in place\textsuperscript{18} 19 and therefore screening positive on UNHS in the UK would be more likely to lead to confirmation of PCHI by age 9 months.

The annual attrition rate (ie, 3% over 17 years since UNHS or 4% over the 9 years since assessment at primary school) among children with PCHI eligible for the present study is low for a teenage population with a chronic medical condition but limited the power of the study to examine change in reading comprehension between the primary school and teenage assessments. In spite of this limitation in power, the inter-group differences on the prespecified primary outcome of reading comprehension were large enough to be both statistically significant and clinically important.

CONCLUSIONS
As the Millenium Development Goals project approaches its 2015 target, UNESCO, UNICEF, the World Bank and WHO are increasingly considering early child development, in which infant hearing is a critical component, as a key determinant of subsequent health\textsuperscript{21 37} and this report is therefore timely. Confirmation of PCHI by nine completed months of age was associated with significantly better performance on reading comprehension, the prespecified primary outcome variable, and the effect size of this benefit of early confirmation of PCHI had increased from moderate to large between assessments at the ages of 8 and 17 years. This strengthens the case for national governments to fund UNHS programmes that increase the rates of early confirmation of PCHI in the many developed and developing countries where UNHS for PCHI is currently under discussion but not yet adopted as national policy.\textsuperscript{18–40}

Acknowledgements We thank the research assistants Eleanore Coulthard, Joanne Pickersgill, Lisa Shipway and Zahra Taghzadeh; the audiologists Margaret Baldwin, Alysson Bumby, Adrian Dighe, Harpreet Nijar, David Reed, Joy Roberts, Sue Robinson, Salim Suleman, Rosbin Syed and Huw Thomas; and the other medical and educational professionals who supported this study. We thank particularly the participating teenagers and their families.

Contributors HP oversaw the conduct and analysis of the study, drafted the initial manuscript and approved the final manuscript. JK, MM, JS and SW assisted in the design and supervision of the study, assisted with manuscript preparation and approved the final manuscript. JLP assisted in the design and supervision of the study, supervised the statistical analysis, assisted with manuscript preparation and approved the final manuscript. HB and ET assisted in the supervision of the study and approved the final manuscript. HMY undertook the statistical analysis and approved the final manuscript. CRX designed and supervised the study, assisted in manuscript preparation and approved the final manuscript.

Funding This work was funded by The Wellcome Trust (Grant number 089251/Z/09/Z) which had no role in the design, conduct of the study, collection analysis and interpretation of the data, preparation, review or approval of the manuscript, or the decision to submit the manuscript for publication.

Competing interests None.

Ethics approval Southampton and SW Hampshire Research Ethics Committee.

Provenance and peer review Not commissioned; externally peer reviewed.

Data sharing statement The authors are willing to share all unpublished data from the study with bona fide researchers. The database can be made available to them through discussion with the corresponding author and the Wellcome Trust.

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