Reading and communication skills after universal newborn screening for permanent childhood hearing impairment

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ABSTRACT

Background: Birth in periods with universal newborn screening (UNS) for permanent childhood hearing impairment (PCHI) and early confirmation of PCHI have been associated with superior subsequent language ability in children with PCHI. However their effects on reading and communication skills have not been addressed in a population-based study.

Methods: In a follow-up study of a large birth cohort in southern England, we measured reading by direct assessment and communication skills by parent report in 120 children with bilateral moderate, severe or profound PCHI aged 5.4–11.7 years, of whom 61 had been born in periods with UNS, and in a comparison group of 63 children with normal hearing.

Results: Compared with birth during periods without UNS, birth during periods with UNS was associated with better reading scores (inter-group difference 0.39 SDs, 95% CI 0.02 to 0.76, $p = 0.042$) and communication skills scores (difference 0.51 SDs, 95% CI 0.06 to 0.95, $p = 0.026$). Compared with later confirmation, confirmation of PCHI by age 9 months was also associated with better reading (difference 0.51 SDs, 95% CI 0.15 to 0.87, $p = 0.006$) and communication skills (difference 0.56 SDs, 95% CI 0.12 to 1.00, $p = 0.013$). In the children with PCHI, reading, communication and language ability were highly correlated ($r = 0.62–0.84$, $p<0.001$).

Conclusion: Birth during periods with UNS and early confirmation of PCHI predict better reading and communication abilities at primary school age. These benefits represent functional gains of sufficient magnitude to be important in children with PCHI.

Previously we reported that universal newborn screening (UNS) and early detection of permanent childhood hearing impairment (PCHI) were associated with higher scores for language at primary school age.[1] A recent systematic review by the US Preventative Services Task Force (USPSTF)[2] assessed that study as providing good-quality evidence of benefit to language development and was accompanied by a USPSTF recommendation to screen for hearing loss in all newborn infants.[3] In comparison with their hearing peers, children with PCHI also have poorer academic outcomes, particularly reading attainment with an average reading age of 9 years when aged 17 years.[4 5] In this report, we extend our previously reported findings on language acquisition to examine the impact of early detection of PCHI on applied practical skills further along the causal pathway, such as reading and communication skills.

What is already known on this topic

- Bilateral permanent childhood hearing impairment (PCHI) >40 dB hearing level impairs language and school achievement.
- Universal newborn screening and confirmation of PCHI by age 9 months improves language at primary school age.

What this study adds

- Universal newborn screening and early confirmation of permanent childhood hearing impairment improve reading ability at primary school age and adaptive behaviour with respect to communication skills.
- The best estimate of the size of these benefits is around 0.5 SD.

METHODS

Communication and reading skills were assessed at the same home visit and in the same children with bilateral PCHI ⩾40 dB hearing level, identified in a 1992–1997 birth cohort in eight districts of southern England, whose language abilities we reported previously.[1] In addition to the assessments previously reported, "basic reading" (BR) and "reading comprehension" (RC) scores were derived on the Weschler Objective Reading Dimensions[6] (WORD). The communication domain of the Vineland Adaptive Behaviour Scales (VABS)[7] was also completed, using information provided by the principal caregiver, usually the mother. Reading and communication scores provided by a group of normally hearing children, born in the same hospitals and of similar age at assessment, were used to derive z scores (including z aggregate reading = zBR + zRC) for the children with PCHI. Consistent with our previous trial of UNS,[8 9] early confirmation of PCHI was pre-specified as confirmation by 9 completed months of age.

Multiple linear regression (using Stata version 8[10]) was used to examine the effects of birth in periods with UNS and of early confirmation of PCHI on reading and communication scores and to adjust for other explanatory variables, namely severity of hearing impairment, non-verbal ability and maternal education. The South and West UK Multi-centre Research Ethics Committee approved this study (MREC/99/6/77) and participating principal caregivers provided written informed consent.

RESULTS

Of 168 identified children with bilateral PCHI in the cohort of 156 733 births, 120 were enrolled and assessed at a mean (range) age of 7.9 (5.4–11.7) years as were 63 children with normal hearing.[1] The characteristics of the families and children born in periods with and without UNS were similar except that the mothers of babies born in periods without UNS tended, presumably by chance, to have completed education to a higher level (table 1). The median age of confirmation of PCHI was 10 months (interquartile range 2–25) with a skewed distribution (mean 17.5 months). The children in the normally hearing comparison group were selected to be English speaking. They had fewer health problems and were more likely to have parents in "higher" occupations than the children with PCHI but were in other respects similar to them (table 1).

Reading (WORD) and communication skills (VABS)

Scores for WORD and for VABS were available in 102 and 98 of 120 (85% and 82%), respectively, of the children with PCHI, including 16 and 11 children who used British Sign Language (BSL). The unadjusted scores for these measures, standardised for age, were significantly higher in the normal hearing comparison group than in the children with PCHI (table 2). Comparing the hearing-impaired group with the normally hearing group, we found larger deficits in language comprehension than in communication and reading (table 2). Among children with PCHI, outcome scores were converted to z scores. There was a consistent trend to higher unadjusted scores in those who were born during periods with UNS compared with those born in periods without UNS or whose PCHI was confirmed by 9 months of age compared with those whose PCHI was confirmed later.

Effect of birth in periods with UNS

After adjustment for severity of PCHI, mother's educational level and the child's non-verbal ability, group mean z scores for WORD aggregate reading and for the VABS communication

Table 1 Characteristics of hearing-impaired and normally hearing children

	Hearing-impaired UNS		Normally hearing
	UNS	No UNS	
	n = 61 (%)	n = 59 (%)	n = 63 (%)
Mean (range) of age at assessment	7.5 (5.5–10.0)	8.3 (5.4–11.7)	8.1 (6.2–9.8)
Female	26 (43)	27 (46)	26 (41)
Degree of hearing loss			
Moderate	35 (57)	30 (51)	na
Severe	16 (26)	13 (22)	na
Profound	10 (16)	16 (27)	na
Other disabilities			
Cerebral palsy	2 (3)	3 (5)	0
Visual disability	5 (8)	8 (14)	0
Learning disability	3 (5)	5 (8)	0
Of chromosomal/syndromic origin	13 (21)	10 (17)	1 (2)
Other	16 (26)	18 (31)	
None	41 (67)	36 (61)	62 (98)
Mode of communication			
Oral ± sign language	50 (82)	47 (79)	na
Sign language only	6 (10)	10 (17)	na
Non-verbal	5 (8)	2 (3)	na
Non-verbal ability‡			
Above average	18 (30)	20 (34)	28 (44)
Average	8 (13)	13 (22)	17 (27)
Below average	29 (48)	20 (34)	18 (29)
Mother's education*			
No qualifications or <5 O levels†	27 (44)	16 (27)	25 (40)
⩾5 O levels or some A levels†	30 (49)	32 (54)	25 (40)
University degree and above	4 (7)	10 (17)	13 (21)
Occupation of head of household*			
Never worked/unemployed	10 (16)	8 (14)	2 (3)
Lower occupations	10 (16)	8 (14)	14 (22)
Intermediate occupations	20 (33)	16 (27)	12 (19)
Higher occupations	23 (38)	26 (44)	35 (56)
English first language at home			
Yes	54 (88)	45 (76)	60 (95)

*Classified as per 2001 UK census; one missing value.
†O levels (now replaced by general certificates of education) are usually taken at age 16 years; A levels (now replaced by "A2s") are taken 2 years later as qualifications for entry to higher education.
‡Ravens Coloured Progressive Matrices percentile scores for age; 12 missing values comprised of eight and four from children with early- and late-confirmed permanent childhood hearing impairments, respectively.
na, not applicable.
UNS, universal newborn screening.

Table 2 Unadjusted standardised scores for reading, communication and language in children with and without permanent childhood hearing impairment (PCHI) at primary school age

Measure	Numbers of children: Normally hearing	Numbers of children: Hearing impaired	Mean score (SD): Normally hearing (a)	Mean score (SD): Hearing impaired (b)	Mean difference (95% CI) (a−b)	p Value	Difference expressed as SDs of score in (a) (a−b/SD of a)
Reading							
WORD basic reading	63	102	102.5 (15.5)	93.7 (17.7)	8.7 (3.4 to 14.1)	0.001	0.56
WORD reading comprehension	63	98	99.2 (15.2)	87.5 (16.5)	11.7 (6.6 to 16.8)	<0.0001	0.77
Communication							
VABS communication	63	98	108.4 (19.0)	93.9 (30.0)	14.5 (7.1 to 21.9)	0.0001	0.76
Language							
Receptive (TROG)	63	98	99.2 (15.3)	80.4 (15.0)	18.8 (14 to 23.6)	<0.0001	1.23
Receptive (BPVS)	63	101	103.5 (12.6)	79.0 (19.7)	24.6 (19.7 to 29.5)	<0.0001	1.95
Expressive (Bus Story information score)	63	87	36.6 (9.7)	26.7 (12.1)	9.9 (6.4 to 13.5)	<0.0001	1.02
Expressive (Bus Story − average 5 longest sentences)	63	87	11.9 (2.3)	10.3 (3.4)	1.6 (0.7 to 2.5)	0.0007	0.69

Mean scores are standardised for age at assessment but not otherwise adjusted.
BPVS, British picture vocabulary scale; TROG, test for assessment of grammar; VABS, Vineland Adaptive Behaviour Scales; WORD, Weschler Objective Reading Dimensions.

scale for those children born in periods with UNS were significantly higher than for children born in periods without UNS by 0.39 and 0.51, respectively (table 3). These adjusted scores were positively correlated with previously reported[1] receptive and expressive language z scores (r = 0.62–0.84, p<0.001).

Effect of early detection of PCHI

Compared with scores in those whose PCHI was confirmed later, adjusted group mean scores were also significantly higher in those whose PCHI was confirmed by 9 months by 0.51 on WORD aggregate reading and 0.56 on the VABS communication scale (Table 4). Compared to the size of effect of birth in periods with UNS and of early detection of PCHI observed in all children with PCHI, the effect observed in children that used BSL was similar with respect to reading scores and larger with respect to communication scores, but this was based on few observations.

Mediating role of language

Adding the aggregate language z scores to the regression models substantially reduced the differences between the group mean z scores, whether comparing children born in periods with and without UNS (to 0.16, 95% CI −0.10 to 0.42, p = 0.24 for reading and 0.31, 95% CI −0.08 to 0.70, p = 0.11 for communication) or children whose PCHI was detected early and late (to 0.09, 95% CI −0.17 to 0.36, p = 0.49 for reading and 0.19, 95% CI −0.20 to 0.59, p = 0.33 for communication), suggesting that the benefits to reading and communication were partially mediated by greater language ability.

Analysis in the Wessex subgroup

In the Wessex subgroup, whose exposure to UNS (or not) was allocated in a controlled trial, the chance of unknown confounding should have been reduced. In this subgroup, the best estimate of the size of the effect of birth in periods with UNS on WORD aggregate reading scores and VABS communication scale scores (table 5) was greater than that observed in the study population as a whole. This was also true with early confirmation of the PCHI (cf. tables 3 and 4 with tables 5 and 6). In the case of aggregate reading scores, these inter-group differences remained statistically significant (p = 0.04 and 0.004) in spite of reduced numbers of observations (tables 5 and 6).

DISCUSSION

This population-based sample of children showed a significant association between both birth in periods with UNS and also early detection of PCHI on the one hand and higher mean scores at primary school age for reading and communication (by 0.4–0.6 SDs) on the other. These outcomes demonstrate gains in

Table 3 Whole study sample: effect of birth in periods with universal newborn screening (UNS) on reading and communication z scores in children with permanent childhood hearing impairment (PCHI) at primary school age

Z scores‡	Born in periods with/without UNS for PCHI — No: With UNS	No: Without UNS	Group mean* z score‡ (SD): With UNS (a)	Group mean* z score‡ (SD): Without UNS (b)	Unadjusted* mean difference (95% CI) (a−b)	p Value	Adjusted† mean difference (95% CI)	p Value
WORD reading ability								
Basic reading (x)	51	51	−0.64 (1.19)	−0.97 (1.26)	0.31 (−0.17 to 0.79)	0.21	0.40 (−0.00 to 0.80)	0.051
Reading comprehension (y)	50	48	−0.82 (1.12)	−1.04 (1.24)	0.22 (−0.25 to 0.69)	0.35	0.38 (−0.02 to 0.77)	0.062
Aggregate reading (x + y)/2	51	51	−0.74 (1.12)	−1.06 (1.25)	0.30 (−0.17 to 0.77)	0.20	0.39 (0.02 to 0.76)	0.042
VABS communication scale	50	48	−0.21 (1.13)	−0.64 (1.39)	0.43 (−0.08 to 0.94)	0.096	0.51 (0.06 to 0.95)	0.026

*Adjusted only for age at assessment.
†Adjusted, in a linear regression model, for severity of PCHI, maternal educational qualifications and Ravens Coloured Progressive Matrices (non-verbal ability) score.
‡Negative scores indicate that scores were lower than those seen in the normally hearing comparison group.[1]
VABS, Vineland Adaptive Behaviour Scales; WORD, Weschler Objective Reading Dimensions.

Table 4 Whole study sample: effect of early confirmation of permanent childhood hearing impairment (PCHI) on reading and communication z scores in children with PCHI at primary school age

	Age at confirmation of PCHI							
	No		Group mean* z score‡ (SD)		Unadjusted* mean difference (95% CI)	p Value	Adjusted† mean difference (95% CI)	p Value
	⩽9 months	>9 months	⩽9 months	>9 months				
WORD reading ability								
Basic reading (x)	45	57	−0.54 (1.18)	−1.02 (1.24)	0.45 (−0.03 to 0.93)	0.064	0.55 (0.17 to 0.93)	0.005
Reading comprehension (y)	43	55	−0.71 (1.18)	−1.09 (1.16)	0.38 (−0.10 to 0.85)	0.12	0.48 (0.09 to 0.86)	0.016
Aggregate (x + y)/2	45	57	−0.65 (1.14)	−1.10 (1.21)	0.43 (−0.04 to 0.89)	0.07	0.51 (0.15 to 0.87)	0.006
VABS communication scale	44	54	−0.13 (1.16)	−0.65 (1.33)	0.53 (0.02 to 1.03)	0.042	0.56 (0.12 to 1.00)	0.013

*Adjusted only for age at assessment.
†Adjusted, in a linear regression model, for severity of PCHI, maternal educational qualifications and Ravens Coloured Progressive Matrices (non-verbal ability) score.
‡Negative scores indicate that scores were lower than those seen in the normally hearing comparison group.[1]
VABS, Vineland Adaptive Behaviour Scales; WORD, Weschler Objective Reading Dimensions.

skills that are important in daily living and of sufficient magnitude to be of functional significance.

This study may have underestimated the size of the benefit of UNS for reading and communication skills for children with PCHI from birth because this birth cohort was the first in the UK to which UNS was applied so that intervals between screening positive, confirmation of PCHI and fitting of hearing aids were longer than is the current standard of care.[11 12] The data obtained in the Wessex subgroup were experimental since their exposure to UNS was the intervention in a controlled trial,[8 9] suggesting a cause-and-effect relationship between UNS and higher subsequent reading and communication scores.

The only previous population-based study to examine the effect of age at first diagnosis on reading in children with PCHI of primary school found no significant benefit.[13] The setting of that study differed from the present report in that the newborn screening for PCHI had been undertaken in high-risk but not in standard-risk newborns. Other limitations of that study were the inclusion of children with only mild PCHI (21% of the sample), an ascertainment rate of 89 of 132 eligible children (67%), and a relatively low rate of early confirmation of PCHI with only 11 (12%) cases with PCHI detected by age 6 months.

We have previously reported that confirmation of hearing impairment prior to age 9 months was associated with higher scores on language tests at age 8 years.[1] We chose language as the outcome measure because it can be precisely estimated by direct assessment and is on the causal pathway from early intervention to better functional outcomes. For both theoretical and practical reasons, however, those functional outcomes cannot be inferred from the finding of higher language scores, as explained below.

VABS communication domain scores assess what the individual understands, says, reads and writes as reflected in behaviours as they occur in an individual's environment rather than assessing responses to standardised auditory stimuli in a direct testing situation (ie, language testing). VABS communication scores are higher in children with auditory impairments than in those with visual impairments whereas the reverse is true for language tests.[7] Communication requires linguistic skills and the ability to decode a message that may also require auditory (as in spoken language), visual (as in gesture, lip movement and reading) or tactile (as in Braille) skills. Compared with that of normally hearing children, the communication of children with hearing impairment depends to a greater extent on gesture, on awareness of social cues and on other skills that compensate for their reduced auditory acuity. These skills are assessed by communication scores but not by standard language tests that are standardised on normally hearing children.

Reading requires visual–perceptual skills to access a cognitive representation of linguistic information whereas in the case of comprehension and expression of spoken language this access is dependent on the ability to process an auditory input and to generate a verbal utterance. Reading is thus less dependent than oral language on auditory acuity which is reduced in hearing impairment. The existence of a relationship between the development of reading and language skills is well established in normally hearing children[14] but poorly understood in hearing-impaired children. In the present study, the greater deficits in language than in reading relative to those of the normally hearing comparison group suggest that the language scores underestimated communicating ability in the hearing-impaired children. This limitation on the use of language scores as a

Table 5 Wessex-controlled trial subgroup: effect of birth in a period with universal newborn screening (UNS) on reading and communication z scores in children with permanent childhood hearing impairment (PCHI) at primary school age

	Born in periods with/without UNS							
	No		Group mean* z score‡ (SD)					
Z scores‡	With UNS	Without UNS	With UNS (a)	Without UNS (b)	Unadjusted* mean difference (95% CI) (a−b)	p Value	Adjusted† mean difference (95% CI)	p Value
WORD aggregate reading	21	20	−0.64 (1.30)	−1.15 (1.14)	0.51 (−0.26 to 1.29)	0.19	0.58 (0.026 to 1.13)	0.04
VABS communication scale	21	19	−0.42 (−1.30)	−0.74 (1.16)	0.32 (−0.47 to 1.11)	0.41	0.52 (−0.21 to 1.24)	0.16

*Adjusted only for age at assessment.
†Adjusted, in a linear regression model, for severity of PCHI, maternal educational qualifications and Ravens Coloured Progressive Matrices (non-verbal ability) score.
‡Negative scores indicate that scores were lower than those seen in the normally hearing comparison group.[1]
VABS, Vineland Adaptive Behaviour Scales; WORD, Weschler Objective Reading Dimensions.

Table 6 Wessex-controlled trial subgroup: effect of early confirmation of permanent childhood hearing impairment (PCHI) on reading and communication z scores in children with PCHI at primary school age

	Age at confirmation of PCHI							
	No		Group mean* z score (SD)		Unadjusted* mean difference (95% CI)	p Value	Adjusted† mean difference (95% CI)	p Value
	⩽9 months	>9 months	⩽9 months	>9 months				
WORD aggregate reading	15	26	−0.62 (1.41)	−1.04 (1.13)	0.43 (−1.24 to 0.38)	0.29	−0.77 (−1.29 to −0.25)	0.004
VABS communication scale	15	25	−0.23 (1.46)	−0.78 (1.05)	−0.54 (−1.35 to 0.26)	0.18	−0.71 (−1.43 to 0.005)	0.052

*Adjusted only for age at assessment.
†Adjusted, in a linear regression model, for severity of PCHI, maternal educational qualifications and Ravens Coloured Progressive Matrices (non-verbal ability) score.
VABS, Vineland Adaptive Behaviour Scales; WORD, Weschler Objective Reading Dimensions.

measure of the benefit of UNS is circumvented by showing the benefits to reading and communication reported here.

The processes and mechanisms that underlie the development of reading ability in these children are not clear and may differ from those known to play a strong predictive role in the early development of reading skills in normally hearing children. These include language skills such as vocabulary and phonological awareness (the ability to segment and blend the sounds of a language eg, identifying the rhyme that is common to "hat", "bat" and "cat").[14–19] Reading for 7–8-year-old children with a severe to profound PCHI, for example, was reported to be predicted by expressive vocabulary and speech reading (ie, the child's ability to attend to the shape of the lips and mouth in understanding oral speech) but not by phonological awareness.[19] It is, however, possible that UNS followed by early confirmation of PCHI may allow children to follow a path to literacy more similar to that of hearing children.

Our observation of higher scores for reading and communication in the setting of UNS provides evidence that provision of UNS and confirmation of PCHI by 9 months can be expected to lead not only to higher scores on formal tests of language ability, but also to practical benefits to the reading and communication skills of these children, including the subgroup that uses BSL. Further assessment of the participating children after longer follow-up is planned to establish whether these children show improvement at high school age in language, reading, social communication, and school attainment.

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