ORIGINAL ARTICLE

Language Ability after Early Detection of Permanent Childhood Hearing Impairment

Colin R. Kennedy, M.B., B.S., M.D., Donna C. McCann, Ph.D., Michael J. Campbell, Ph.D., Catherine M. Law, M.B., B.S., M.D., Mark Mullee, Ph.D., Stavros Petrou, Ph.D., Peter Watkin, M.B., B.S., M.Sc., Sarah Worsfold, B.Soc.Sci., Ho Ming Yuen, M.Sc., and Jim Stevenson, Ph.D.

ABSTRACT

BACKGROUND

Children with bilateral permanent hearing impairment often have impaired language and speech abilities. However, the effects of universal newborn screening for permanent bilateral childhood hearing impairment and the effects of confirmation of hearing impairment by nine months of age on subsequent verbal abilities are uncertain.

METHODS

We studied 120 children with bilateral permanent hearing impairment identified from a large birth cohort in southern England, at a mean of 7.9 years of age. Of the 120 children, 61 were born during periods with universal newborn screening and 57 had hearing impairment that was confirmed by nine months of age. The primary outcomes were language as compared with nonverbal ability and speech expressed as z scores (the number of standard deviations by which the score differed from the mean score among 63 age-matched children with normal hearing), adjusted for the severity of the hearing impairment and for maternal education.

RESULTS

Confirmation of hearing impairment by nine months of age was associated with higher adjusted mean z scores for language as compared with nonverbal ability (adjusted mean difference for receptive language, 0.82; 95 percent confidence interval, 0.31 to 1.33; and adjusted mean difference for expressive language, 0.70; 95 percent confidence interval, 0.13 to 1.26). Birth during periods with universal newborn screening was also associated with higher adjusted z scores for receptive language as compared with nonverbal ability (adjusted mean difference, 0.60; 95 percent confidence interval, 0.07 to 1.13), although the z scores for expressive language as compared with nonverbal ability were not significantly higher. Speech scores did not differ significantly between those who were exposed to newborn screening or early confirmation and those who were not.

CONCLUSIONS

Early detection of childhood hearing impairment was associated with higher scores for language but not for speech in midchildhood.

From the Departments of Child Health (C.R.K., D.C.M., S.W.), Public Health Sciences and Medical Statistics (M.M., H.M.Y.), and Psychology (J.S.), University of Southampton, Southampton; the Institute of Primary Care and General Practice, University of Sheffield, Sheffield (M.J.C.); the Institute of Child Health, University College London, London (C.M.L.); the National Perinatal Epidemiology Unit, University of Oxford, Oxford (S.P.); and the Department of Audiology, Whipps Cross University Hospital, London (P.W.) — all in the United Kingdom. Address reprint requests to Dr. Kennedy at Mailpoint 21, Child Health, Southampton General Hospital, Southampton SO16 6YD, United Kingdom, or at crk1@soton.ac.uk.

N Engl J Med 2006;354:2131-41.

Bilateral permanent childhood hearing impairment that is moderate, severe, or profound affects 1 in 750 children and is present at birth in more than 80 percent of affected children.[1-3] Such impairments are associated with impaired language acquisition, learning, and speech development.[2-5]

Currently, screening for bilateral permanent childhood hearing impairment, with the use of transiently evoked otoacoustic emissions and automated measurement of auditory brain-stem responses, is recommended for all infants before the age of three months in the United States,[6] the United Kingdom,[7] and Europe.[8] The value of these recommendations is supported by studies showing that enrollment in an intervention program by nine months of age, as compared with later intervention, is associated with improvements in the verbal ability quotient by as much as 19 points[9] (equivalent to 0.5 to 0.6 SD) and that birth during periods in which universal hearing screening of newborns was in place is associated with a similar benefit.[10] The U.S. Preventive Services Task Force, however, has rated the quality of evidence linking early treatment or birth during periods with universal newborn hearing screening with improved language function as fair or poor.[3]

In a previous controlled trial in the Wessex region of southern England, we showed that universal newborn screening increased the rate of early referral (i.e., before six months of age) for audiologic assessment of babies with bilateral permanent childhood hearing impairment, defined as a hearing loss of at least 40 dB hearing level (HL), on two assessments at least 12 months apart.[11,12] In the present study, we assessed speech and oral language abilities in a sample of children with bilateral permanent childhood hearing impairment, including children enrolled in the earlier trial, and the relationship of these measures to the timing of confirmation of hearing impairment (by nine months of age or later) and to the availability of universal screening of newborns.

METHODS

The study sample included all children with bilateral permanent childhood hearing impairment of at least 40 dB HL identified from a cohort of 157,000 children born in eight districts of southern England. We did not include children with a known postnatal cause of bilateral permanent childhood hearing impairment (e.g., bacterial meningitis). The children in the sample were born between 1993 and 1996 in four districts in the Wessex region or between 1992 and 1997 in two pairs of adjacent districts in the Greater London region.

The four districts in the Wessex subgroup had provided the birth cohort for the Wessex trial, in which a program of universal newborn screening was or was not in place in each pair of districts for birth cohorts born in alternate four- or six-month periods.[11,12] The Greater London subgroup consisted of children born in the only two districts in the United Kingdom that provided universal newborn screening for permanent childhood hearing impairment in the early 1990s (Whipps Cross and Hillingdon[13,14]) and in two other districts, one adjacent to Whipps Cross and one adjacent to Hillingdon.[15] Other than variation in providing the newborn screening, protocols for the identification and confirmation of bilateral permanent childhood hearing impairment, previously reported,[11-15] were similar at all sites.

Follow-up of this birth cohort included audiologic screening at school entry and information both from multiple sources within the participating regions and from primary care teams in other regions. We obtained details of the detection and management of all cases of bilateral permanent childhood hearing impairment from pediatric audiologists, family practitioners, and other involved professionals and by review of the case records of the audiology service in each district. The severity of hearing loss was categorized from recent audiologic records as moderate (40 to 69 dB HL), severe (70 to 94 dB HL), or profound (≥95 dB HL) (Table 1) according to four-frequency averaging of the pure-tone thresholds from 500 to 2000 Hz (or, if pure-tone thresholds were unavailable, sound fields and electrophysiologic-test results).

We prespecified the definition of early confirmation of permanent childhood hearing impairment as confirmation by nine completed months of age. This was consistent with the definition in our previous trial of universal newborn screening[11] and with the U.S. Preventive Services Task Force benchmark for diagnosing or treating infants before 10 months of age.[3]

Two researchers unaware of the child's early hearing or audiologic history evaluated the child during a home visit. One researcher interviewed the principal caregiver (usually the mother), which included completion of the speech scale of the

Table 1. Characteristics of Children with Hearing Impairment, Children with Normal Hearing, and Their Families.

Characteristic	Hearing Impairment (N=120)		Normal Hearing* (N=63)
	Age at Confirmation ≤9 Months (N=57)	Age at Confirmation >9 Months (N=63)	
Female sex — no. (%)	23 (40)	30 (48)	26 (41)
English first language at home — no. (%)	50 (88)	49 (78)	60 (95)
Nonverbal ability — z score			
Median	−0.80	−0.44	−0.02
Interquartile range†	−1.38 to −0.03	−1.12 to 0.30	−0.68 to 0.66
Age at assessment — yr			
Mean	7.54	8.18	8.13
Range	5.42 to 10.00	5.92 to 11.67	6.25 to 9.75
Degree of hearing loss — no. (%)			
Moderate	32 (56)	33 (52)	NA
Severe	12 (21)	17 (27)	NA
Profound	13 (23)	13 (21)	NA
Other disabilities — no. (%)			
Cerebral palsy	3 (5)	2 (3)	0
Visual disability	2 (4)	3 (5)	0
Learning disability	4 (7)	6 (10)	0
Of chromosomal or syndromic origin	11 (19)	3 (5)	1 (2)
None	42 (74)	55 (87)	62 (98)
Mother's education — no. (%)‡			
No qualifications or <5 O-level examinations	22 (39)	21 (33)	25 (40)
≥5 O-level examinations or some A-level examinations	30 (53)	32 (51)	25 (40)
≥University degree	5 (9)	9 (14)	13 (21)
Occupation of head of household — no. (%)§			
Never worked or unemployed	5 (9)	13 (21)	2 (3)
Lower occupations	8 (14)	10 (16)	14 (22)
Intermediate occupations	20 (35)	17 (27)	12 (19)
Higher occupations	24 (42)	23 (37)	35 (56)

* NA denotes not applicable.

† Age-adjusted z scores are listed for Raven's Progressive Matrices total score. The z scores are the number of standard deviations by which the age-adjusted score differed from the mean score in children with normal hearing. Values are missing for eight children who had early confirmation (by nine months of age) of permanent hearing impairment and for four children who had late confirmation.

‡ O-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 two years later as qualifications for entry to higher education.

§ Data are from the Office for National Statistics, London.[16] Lower occupations include semiroutine and routine occupations; intermediate include small employers, own-account workers, and lower supervisory and technical occupations; and higher include higher managerial and professional occupations.

Children's Communication Checklist.[17] Simultaneously, the child was assessed by the other researcher in a separate space on the following: the Test for Reception of Grammar,[18] the British Picture Vocabulary Scale[19] (receptive language); the Renfrew Bus Story Test[20] (expressive language);

and Raven's Progressive Matrices Test (nonverbal abilities).[21] For all these measures, a higher score indicates better function. Normal receptive language is the ability to understand communication through gestures, facial expressions, and words, whereas expressive language is the ability to express needs with the use of gestures, vocalization, facial expressions, and words.

Other characteristics of the child and family, including maternal education according to the 2001 census in the United Kingdom, were also documented (Table 1). The mean age at assessment of language and speech was 7.9 years (range, 5.4 to 11.7).

Our study was approved by the South and West Multicenter Research Ethics Committee, United Kingdom. Principal caregivers provided written informed consent.

For the purpose of comparisons within the group of children with hearing impairment in this report, we used norms[22] obtained from a group of 63 English-speaking children with normal hearing, matched for place of birth and age at assessment with our group of 120 children with hearing impairment. The group mean score and standard-deviation scores in children with normal hearing were used to derive z scores for the children with hearing impairment, equal to the number of standard deviations of the distribution of scores in children with normal hearing by which their age-adjusted score differed from the mean score in children with normal hearing. We also calculated aggregate scores as follows: the z score for receptive language was equal to half the sum of the z score for the Test for Reception of Grammar and the z score for the British Picture Vocabulary Scale; and the z score for expressive language equal to half the sum of the z score for sentence information and the z score for five longest sentences. We calculated difference scores as follows: the z score for a deficit of receptive (or expressive) language as compared with nonverbal skills was equal to the difference between the z score for receptive (or expressive) language and the z score for nonverbal ability.

We assessed the associations between exposure to universal newborn screening (i.e., birth during periods when universal newborn screening was in place), or to confirmation by nine months of age, and age-adjusted individual and aggregate language and speech scores with the use of a two-sample t-test. The preplanned primary outcomes of our study were language and speech scores and differences between language and nonverbal scores at primary-school age after adjustment in a multiple linear regression (Stata software,[23] version 8) for severity of hearing impairment, maternal education, and (except in the case of difference scores) nonverbal ability, which were recognized as potential confounders of the primary outcomes. Normality and homogeneity of the residual variance were examined for all measures to ensure that the regression model was appropriate. All reported P values are two-sided. Adjustment was not made for multiple testing.

On the basis of an expected overall sample size of 154 children with bilateral permanent childhood hearing impairment (determined according to the expected rates in the general population), we anticipated a statistical power of 80 percent to detect a difference of 0.5 SD in verbal ability between the two groups, with a two-sided P value of 0.05. We also performed a subgroup analysis of children who were enrolled in the Wessex controlled trial of universal newborn screening.[11,12] The subgroup was expected to be balanced with respect to both known and unknown confounding factors, although that trial was not powered for the end points of speech and language.

RESULTS

Seventy-seven infants with bilateral permanent childhood hearing impairment (i.e., ≥40 dB HL) were identified among the 68,714 infants born during periods in which universal newborn screening was in place, and 91 infants were identified among the 88,019 infants born during periods without universal screening. These numbers are equivalent to an overall prevalence in our sample of 107 per 100,000, which is close to the expected population prevalence of 112 per 100,000.[1] Estimates of the completeness of ascertainment in our study sample exceeded 95 percent for both the London and Wessex subgroups.[12,13,15] Of the 168 cases identified, the principal caregivers of 120 children gave consent for participation in the study (Fig. 1). Of these 120 children, 61 were born during periods with universal newborn screening and 59 during periods without the screening. Participants were similar to nonparticipants with respect to age, sex, and severity of hearing loss. Hearing impairment was confirmed in 72 infants (60 percent) by one year of age, 90 children (75 percent)

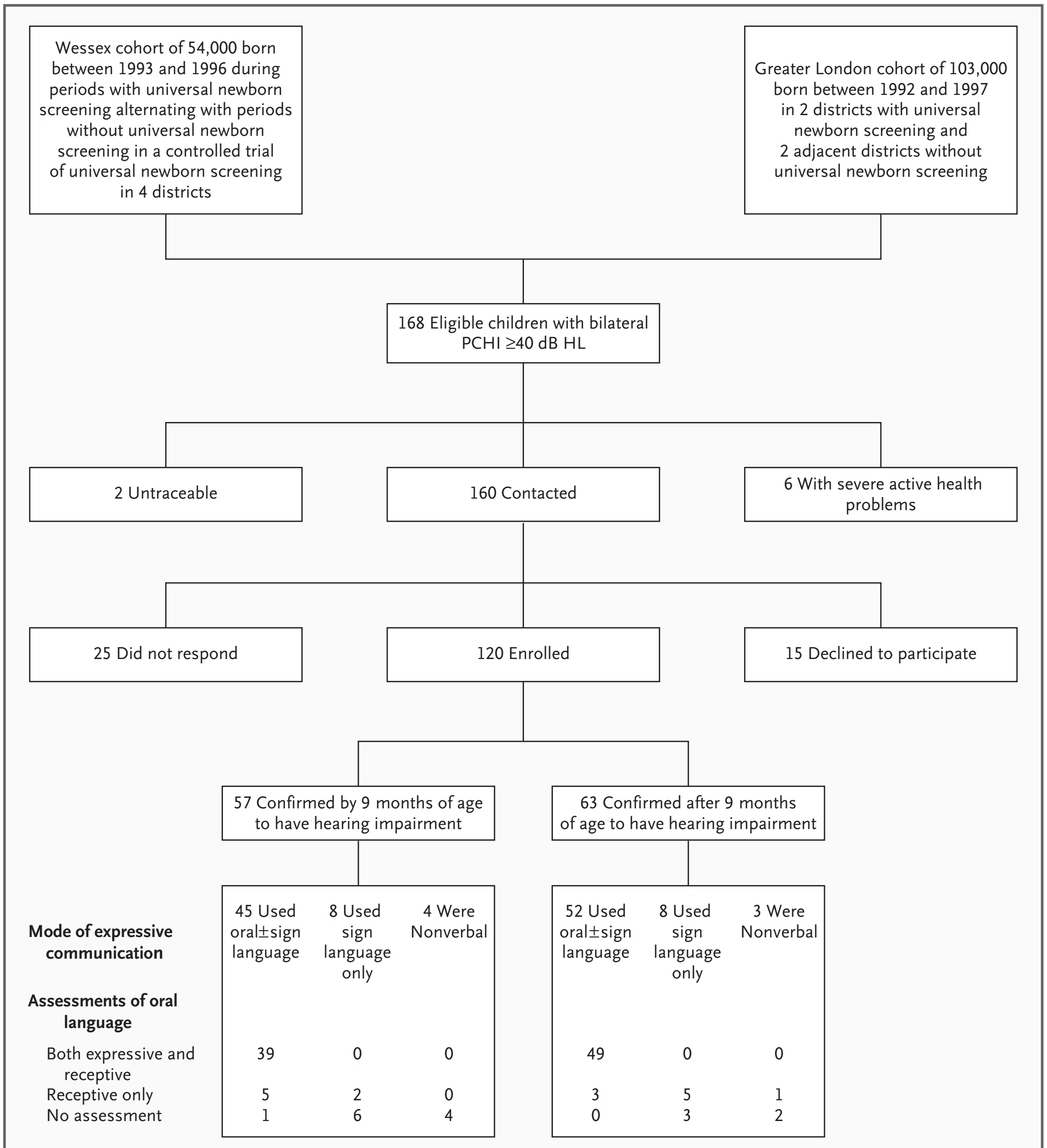

Figure 1. Numbers of Children with Permanent Childhood Hearing Impairment (PCHI) Who Were Eligible for the Study and Assessed for Oral Language Ability.

by two years of age, 106 children (88 percent) by four years of age, and in all 120 children (100 percent) by six years of age.

Remedial therapy for hearing impairment was provided to all participants, since it is a public service available to all deaf preschool children in the United Kingdom. All the children with hearing impairment in this study had received advice in their homes from a teacher of the deaf and hard of hearing (87 percent within three months after confirmation of impairment), and all had been offered audiology services, including high-quality commercial hearing aids fitted according to published national quality standards. Hearing aids were always in place during the assessments reported here. Five participants born during periods with universal newborn screening had cochlear implants, as did 11 children born during periods without the screening. Confirmation of hearing loss occurred at a median of 10 months of age (interquartile range, 3 to 25), enrollment in a management program at 13 months of age (interquartile range, 8 to 32), and fitting with a hearing aid at 15 months of age (interquartile range, 10 to 40). The median ages were similar in the Wessex and Greater London subgroups.

Baseline characteristics, including the severity of hearing impairment, were similar between the 61 children who were exposed to universal newborn screening and the 59 who were not (data not shown), and between 57 children whose impairment was confirmed by nine months of age and 63 whose impairment was confirmed later (Table 1). Confirmation of impairment by nine months of age was significantly more common among children exposed to universal newborn screening (41 of 61 [67 percent]) than among children not exposed to such screening (16 of 59 [27 percent]) (absolute difference, 40 percent; 95 percent confidence interval, 24 to 56 percent; $P<0.001$).

Children whose impairment was confirmed by nine months of age had significantly higher adjusted mean aggregate scores for receptive language than did children whose hearing impairment was confirmed later (difference in mean z scores, 0.76; 95 percent confidence interval, 0.26 to 1.27) and for expressive language ability (difference in mean z scores, 0.50; 95 percent confidence interval, <0.01 to 1.01). Furthermore, their difference scores (i.e., the z score for language minus the z score for nonverbal ability) showed smaller deficits in receptive and expressive language relative to nonverbal ability (Fig. 2A and Table 2). Children who were exposed to universal newborn screening also had higher adjusted mean aggregate scores for receptive language than those who were not exposed (difference in mean z scores, 0.56; 95 percent confidence interval, 0.03 to 1.08), and their difference scores showed a smaller deficit in receptive language relative to nonverbal ability (difference in mean z scores, 0.60; 95 percent confidence interval, 0.07 to 1.13) but no significant difference in expressive language ability (Fig. 2B and Table 3).

Because expressive language scores were not available for all children, we performed a post hoc analysis in the 88 children for whom both receptive and expressive oral language scores were available. We found that the effect size for the relationship between early confirmation of hearing impairment and receptive scores (0.73; 95 percent confidence interval, 0.19 to 1.27) was very similar to that between early confirmation and expressive scores among the same children (Table 2). There were no significant differences in measures of speech between those whose hearing impairment was confirmed by nine months of age and those whose impairment was confirmed after nine months of age or between those exposed to universal newborn screening and those not exposed to such screening (Tables 2 and 3).

Associations between early confirmation of hearing impairment or exposure to universal newborn screening and later language abilities were similar in the Wessex and Greater London subgroups (data not shown). In the Wessex subgroup, for whom the chance of confounding factors should have been reduced because the exposure to newborn screening occurred in a controlled trial, the unadjusted difference between receptive language and nonverbal scores in children born during periods with universal newborn screening as compared with those born during periods without newborn screening was 1.00 (95 percent confidence interval, 0.001 to 2.00; $P=0.05$) (see Table 2 in the Supplementary Appendix, available with the full text of this article at www.nejm.org). In addition, associations between early confirmation of impairment or newborn screening and higher language scores in this cohort were similar to, or higher than, those observed in the whole sample (Tables 2 and 3, and the Supplementary Appendix).

DISCUSSION

We observed significantly higher scores for language, but not for speech, in midchildhood among a population-based sample of children with bilateral permanent hearing impairment who were exposed to universal newborn screening or who had confirmation of hearing impairment by nine months of age than among those who were not exposed to newborn screening or whose impairment was confirmed after nine months of age. In the case of children whose hearing impairment was confirmed by nine months of age, this difference was equivalent to an increase of 10 to 12 points in the verbal as compared with the nonverbal intelligence quotient.

The estimated size of the benefit of newborn screening and of early confirmation of impairment in this sample may be conservative. This birth cohort was the first in the United Kingdom in which newborn screening was applied, and systems to ensure short intervals between positive results on newborn screening and confirmation of hearing impairment, now standard in the United Kingdom[24] and the United States,[25,26] were still evolving. Intervals between confirmation and the fitting of hearing aids were also longer than is the current standard of care in the United Kingdom.[13,24] Delays in confirmation and intervention might have decreased the benefit to language that was associated with early detection.

We excluded children whose hearing impairment was of known postnatal cause, but a minority of cases may have been postnatal in onset. Children who lost hearing after infancy would be expected to benefit less from early confirmation than children with an unchanging congenital hearing impairment. On the basis of the Wessex cohort, the maximum proportion of children with progressive hearing loss during childhood (derived by adding the proportion with negative results on newborn screening to the proportion with a positive result and in which a subsequent increase in the severity of hearing impairment was documented) was 23 percent[12]; a lower percentage was derived for the Greater London subgroup. A sensitivity analysis (not shown) based on the estimate that impairment worsens after birth in 23 percent of children suggests that the true benefit to language acquisition for those with congenital and unchanging hearing impairment would have been larger by a factor of 1.05 to 1.30 than the benefit that we reported for the whole sample.

In contrast to the higher language scores observed among children whose hearing impairment was confirmed early, measures of speech did not differ significantly between groups. Speech was assessed on the basis of parental or professional report, rather than by direct measurement, and may reflect a lack of sensitivity to relevant aspects of speech. Objective analysis of audiotaped samples of speech, recorded for the assessments of language, is currently being performed to further evaluate speech in these children.

Data collected in the Greater London subgroup were observational and cannot prove causality. However, conclusions from these data are strengthened by the similarity of results between the Greater London and Wessex subgroups, since exposure or lack of exposure to newborn screening and the resulting variation in early confirmation was quasi-experimental in design in the latter subgroup. The trial among the Wessex subgroup was not statistically powered to detect differences in language and speech in midchildhood.

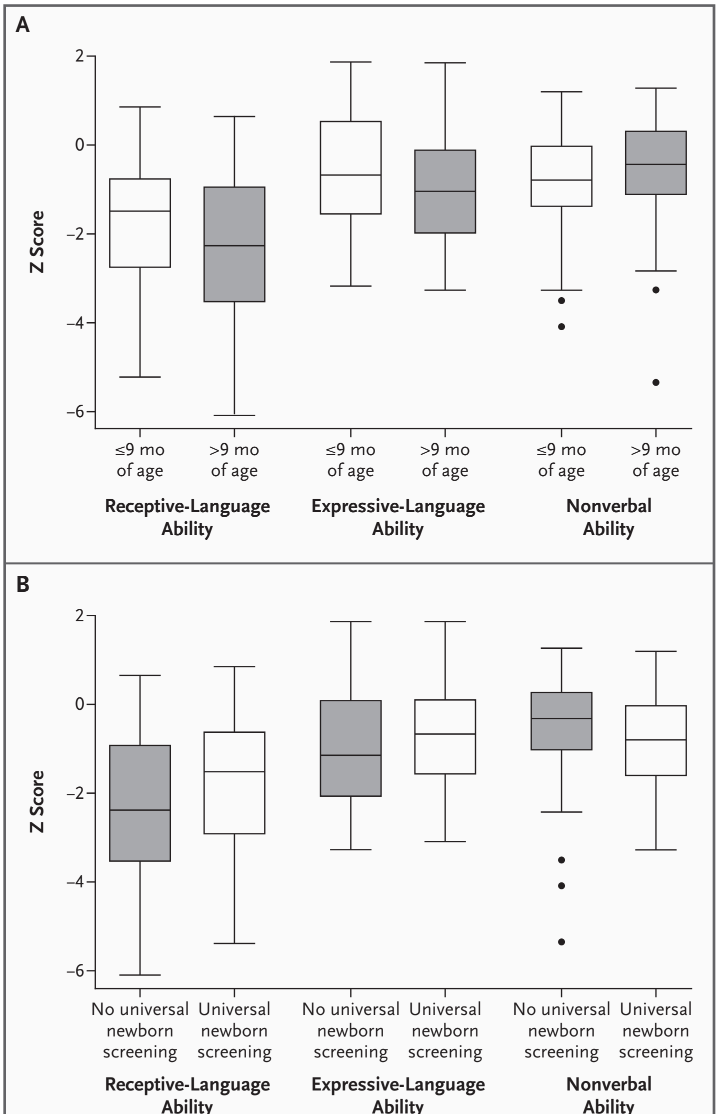

Figure 2. Box-and-Whisker Plots of Age-Adjusted Ability Scores, Relative to Those of Children with Normal Hearing, According to Age at Confirmation of Hearing Impairment (Panel A) and Exposure to Universal Newborn Screening (Panel B) in 104 Children.

The z scores represent the number of standard deviations by which the score differed from the mean score. Values are expressed as the median (horizontal line in each box), with the quartiles (top and bottom of the box) and range (I bar), excluding out-of-range observations (i.e., >1.5 times the interquartile range beyond the quartile), shown as solid circles.

The severity of hearing impairment measured at the time of speech and language evaluation, rather than at the time of confirmation of impairment, was used in the regression models. This approach

Table 2. Timing of Confirmation of Permanent Childhood Hearing Impairment and Language and Speech Scores.*

Measure	Age at Confirmation of Hearing Impairment		Mean z Score (SD) for Age at Confirmation†		Unadjusted Mean Difference (95% CI)‡	P Value	Adjusted Mean Difference (95% CI)§	P Value
	≤9 months	>9 months	≤9 months	>9 months				
	number							
Receptive language								
Test for Reception of Grammar	44	54	−1.46 (1.50)	−2.25 (1.91)	0.78 (0.08 to 1.48)	0.03	0.90 (0.32 to 1.47)	0.003
British Picture Vocabulary Scale	45	56	−1.86 (1.40)	−2.36 (1.65)	0.50 (−0.11 to 1.11)	0.11	0.64 (0.13 to 1.16)	0.02
Aggregate score	45	56	−1.76 (1.47)	−2.38 (1.72)	0.61 (−0.02 to 1.24)	0.06	0.76 (0.26 to 1.27)	0.004
Aggregate minus nonverbal	45	56	−0.82 (1.23)	−1.68 (1.44)	0.86 (0.32 to 1.40)	0.002	0.82 (0.31 to 1.33)	0.002
Expressive language								
Renfrew Bus Story Test: sentence information	39	48	−0.73 (1.32)	−1.23 (1.15)	0.50 (−0.02 to 1.03)	0.06	0.54 (0.05 to 1.04)	0.03
Renfrew Bus Story Test: 5 longest sentences	39	48	−0.46 (1.48)	−0.87 (1.48)	0.42 (−0.22 to 1.05)	0.20	0.47 (−0.14 to 1.08)	0.13
Aggregate score	39	48	−0.59 (1.31)	−1.07 (1.21)	0.46 (−0.08 to 1.00)	0.09	0.50 (<0.01 to 1.01)	0.05
Aggregate minus nonverbal	39	48	0.14 (1.29)	−0.50 (1.34)	0.64 (0.08 to 1.21)	0.03	0.70 (0.13 to 1.26)	0.02
Speech								
Children's Communication Checklist, speech scale	44	51	−1.15 (1.42)	−1.33 (1.54)	0.18 (−0.43 to 0.79)	0.56	0.29 (−0.28 to 0.87)	0.32

* CI denotes confidence interval.
† Mean scores are adjusted for age at assessment. The group mean score and SD scores in children with normal hearing were used to derive z scores for the children with permanent childhood hearing impairment, equal to the number of SDs by which their age-adjusted score differed from the mean score in children with normal hearing. Negative group mean z scores indicate that scores were lower than those seen in the comparison group of children with normal hearing.
‡ The unadjusted mean difference was calculated by subtracting the mean z score for children whose hearing impairment was confirmed after nine months of age from the mean z score for those whose impairment was confirmed by nine months of age. The differences were affected by rounding.
§ This category shows the difference between mean scores adjusted (in a linear regression model) for severity of permanent childhood hearing impairment, maternal education, and except in the case of mean difference scores, age-adjusted total Raven's Progressive Matrices scores.

Table 3. Exposure to Universal Newborn Screening and Language and Speech in Hearing-Impaired Children.*

Measure	No Universal Newborn Screening	Universal Newborn Screening	Mean z Score (SD)†		Unadjusted Mean Difference (95% CI)‡	P Value	Adjusted Mean Difference (95% CI)§	P Value
			No Universal Newborn Screening	Universal Newborn Screening				
	number							
Receptive language								
Test for Reception of Grammar	46	52	−2.10 (1.75)	−1.71 (1.78)	0.39 (−0.32 to 1.11)	0.27	0.59 (−0.01 to 1.19)	0.05
British Picture Vocabulary Scale	49	52	−2.34 (1.58)	−1.94 (1.52)	0.41 (−0.20 to 1.02)	0.19	0.47 (−0.05 to 1.00)	0.08
Aggregate score	49	52	−2.32 (1.61)	−1.89 (1.65)	0.45 (−0.18 to 1.08)	0.16	0.56 (0.03 to 1.08)	0.04
Aggregate minus nonverbal	49	52	−1.67 (1.29)	−0.94 (1.45)	0.73 (0.19 to 1.28)	0.01	0.60 (0.07 to 1.13)	0.03
Expressive language								
Renfrew Bus Story Test: sentence information	41	46	−1.13 (1.18)	−0.90 (1.31)	0.24 (−0.30 to 0.77)	0.38	0.27 (−0.23 to 0.78)	0.29
Renfrew Bus Story Test: 5 longest sentences	41	46	−0.81 (1.61)	−0.58 (1.38)	0.22 (−0.41 to 0.86)	0.49	0.32 (−0.30 to 0.94)	0.30
Aggregate score	41	46	−0.99 (1.33)	−0.74 (1.23)	0.23 (−0.32 to 0.78)	0.40	0.30 (−0.22 to 0.81)	0.25
Aggregate minus nonverbal	41	46	−0.44 (1.35)	−0.02 (1.34)	0.43 (−0.15 to 1.00)	0.14	0.39 (−0.19 to 0.98)	0.18
Speech								
Children's Communication Checklist, speech scale	47	50	−1.30 (1.47)	−1.20 (1.50)	0.10 (−0.51 to 0.71)	0.75	0.12 (−0.46 to 0.71)	0.68

* CI denotes confidence interval.
† Mean scores are adjusted for age at assessment. The group mean score and SD scores in children with normal hearing were used to derive z scores for the children with permanent childhood hearing impairment, equal to the number of SDs by which their age-adjusted score differed from the mean score in children with normal hearing. Negative group mean z scores indicate that scores were lower than those seen in the comparison group of children with normal hearing.
‡ The unadjusted mean difference was calculated by subtracting the mean z score for children born in periods without universal newborn screening from the mean z score for those born in periods with universal newborn screening. The differences were affected by rounding.
§ This category shows the difference between mean scores adjusted (in a linear regression model) for the severity of permanent childhood hearing impairment, maternal education, and except in the case of mean difference scores, age-adjusted total Raven's Progressive Matrices scores.

minimizes the chance of bias due to possible differences in the estimates of the severity of impairment between groups with early confirmation of impairment and those with late confirmation among children in whom severity increased with age. This adjustment did not in any case materially alter our estimate of the size of the benefits of early confirmation, since the groups with early confirmation and those with late confirmation were similar in their distribution of severity of hearing impairment.

Our data extend findings from previous studies of the relationship between early identification of hearing impairment and later outcomes. Adjusted mean vocabulary scores of children with hearing impairment, assessed at the age of 5 years, were higher in children enrolled before 11 months of age in an early intervention program in Nebraska than in those enrolled at 11 to 23 months of age (by 0.69 SD) or at 24 to 35 months of age (by 0.99 SD).[27] Similar findings were reported from a study in Washington State comparing children enrolled before 24 months of age with those enrolled later.[28] Both these studies lacked clear criteria for inclusion, selected as participants only those who adhered to the early intervention programs, and used unblinded assessment of children whose language ability may have been greater than that of children whose hearing impairment was identified early but who were not available for follow-up.[3] In contrast to these studies, a population-based Australian study reported no benefit of early diagnosis before 6 or 12 months of age on speech, language, and other outcomes at 7 to 8 years of age among children with congenital hearing impairment.[29] All three studies excluded children with developmental disabilities, and none included sufficient children enrolled before 12 months of age to estimate reliably the benefit of early intervention in that age group.

In the largest previous study,[9] the language abilities of 150 children enrolled in the Colorado Home Intervention Program, assessed at 13 to 36 months by parental report, were 0.5 to 0.6 SD higher (scaled according to subsequently published norms[30]) in those in whom hearing impairment was identified by 6 months of age and who enrolled after a mean of 3 months more than in those whose impairment was identified later. In a smaller case–control study, these investigators used similar assessment methods and reported a benefit of 0.5 to 0.8 SD to language and speech associated with birth in hospitals providing universal newborn screening, as compared with those not providing it.[10] However, these Colorado studies did not adjust for differences between the baseline characteristics of the groups, were subject to possible selection bias, and relied on unblinded assessments of parental reports of language abilities.[3] The greater benefits reported, as compared with those reported in the present study, may have arisen from these potential sources of bias or from the differing ages at assessment.

Universal newborn screening and early confirmation of permanent childhood hearing impairment had clinically important benefits to the language abilities of children at primary-school age in this population-based study. Other data from this cohort suggest that such screening may be cost-effective.[31] Longer follow-up is needed to establish whether these children have higher academic achievement and continue to show superior language skills at high-school age.

Supported by the Wellcome Trust (061839).

No potential conflict of interest relevant to this article was reported.

We are indebted to the children, families, school staff, specialist teachers of the hearing impaired, speech and language pathologists, and audiology professionals in the local area teams for their help and assistance; to Margaret Baldwin, Joy Bhattacharya, Alyson Bumby, Irene Curtis, Carol Hunter, David Reed, Scott Richards, Peter Savundra, Huw Thomas, Tim Williamson, Jan Nanor, Helen Davis, Shirley Golden, Eleanor Lutman, Kristen Paul, and Helen Ryder; to Julie Brinton for her advice on assessments of speech and language; and to Sue Robinson for her advice on audiology.

REFERENCES

1. Davis A, Wood S. The epidemiology of childhood hearing impairment: factors relevant to planning of services. Br J Audiol 1992;26:77-90.

2. Davis A, Bamford J, Wilson I, Ramkalawan T, Forshaw M, Wright S. A critical review of the role of neonatal hearing screening in the detection of congenital hearing impairment. Health Technol Assess 1997;1:i-iv, 1-176.

3. Thompson DC, McPhillips H, Davis RL, Lieu TL, Homer CJ, Helfand M. Universal newborn hearing screening: summary of evidence. JAMA 2001;286:2000-10.

4. Wake M, Hughes EK, Poulakis Z, Collins C, Rickards FW. Outcomes of children with mild to profound congenital hearing loss at 7 to 8 years: a population study. Ear Hear 2004;25:1-8.

5. Allen TE. Patterns of academic achievement among hearing impaired students: 1974 and 1983. In: Schildroth AN, Karchmer AM, eds. Deaf children in America. San Diego, Calif.: College-Hill Press, 1986: 161-206.

6. Early identification of hearing impairment in infants and young children. Bethesda, Md.: National Institutes of Health, 1993. (Accessed March 13, 2006, at http://www.ncbi.nlm.nih.gov/books/bv.fcgi?rid=hstat4.chapter.11796.)

7. Quality standards in paediatric audiology. Vol. 1: guidelines for the early iden-

tification of hearing impairment. London: National Deaf Children's Society, 1994: 1-10.
8. Lutman ME, Grandori F. Screening for neonatal defects: European consensus statement. Eur J Pediatr 1999;158:95-6.
9. Yoshinaga-Itano C, Sedey AL, Coulter DK, Mehl AL. Language of early- and later-identified children with hearing loss. Pediatrics 1998;102:1161-71.
10. Yoshinaga-Itano C, Coulter D, Thomson V. Developmental outcomes of children with hearing loss born in Colorado hospitals with and without universal newborn screening programs. Semin Neonatol 2001;6:521-9.
11. Wessex Universal Hearing Screening Trial Group. Controlled trial of universal neonatal screening for early identification of permanent childhood hearing impairment. Lancet 1998;352:1957-64.
12. Kennedy C, McCann DC, Campbell MJ, Kimm L, Thornton R. Universal newborn screening for permanent childhood hearing impairment: an 8-year follow-up of a controlled trial. Lancet 2005;366:660-2.
13. Watkin PM, Baldwin M. Confirmation of deafness in infancy. Arch Dis Child 1999;81:380-9.
14. Tucker SM, Bhattacharya J. Screening of hearing impairment in the newborn using the auditory response cradle. Arch Dis Child 1992;67:911-9.
15. Watkin W, Hasan J, Baldwin M, Ahmed M. Neonatal hearing screening: have we taken the right road? Results from a 10-year targeted screen longitudinally followed up in a single district. Audiol Med 2005;3:175-84.
16. Office for National Statistics. Standard occupational classification. London: Her Majesty's Stationery Office, 2000.
17. Bishop DV. Development of the Children's Communication Checklist (CCC): a method for assessing qualitative aspects of communicative impairment in children. J Child Psychol Psychiatry 1998;39: 879-91.
18. Bishop DVM. Test for reception of grammar. Manchester, United Kingdom: Age and Cognitive Performance Research Centre, University of Manchester, 1983.
19. Dunn LM, Whetton C, Burley J. British Picture Vocabulary Scale. 2nd ed. Windsor, United Kingdom: NFER-Nelson, 1997.
20. Renfrew C. Renfrew Bus Story Manual: a test of narrative speech. 3rd ed. Oxford, England: Renfrew/Winslow, 1995.
21. Raven C, Raven JC, Court JH. Manual for Raven's Progressive Matrices and Vocabulary Scales. Oxford, England: Oxford Psychologists Press, 1998.
22. Stevenson J, McCann D, Watkin P, Worsfold S, Kennedy C. The relationship between language/speech development and behaviour problems in children with hearing loss. J Speech Lang Hear Res (in press).
23. Stata statistical software, release 8.0. College Station, Tex.: Stata Corporation, 2003.
24. Baldwin M. Reducing the age of audiological certainty in babies identified by neonatal screening. (Ph.D. thesis. Manchester, United Kingdom: University of Manchester, 2005.)
25. Harrison M, Roush J, Wallace J. Trends in age of identification and intervention in infants with hearing loss. Ear Hear 2003;24:89-95.
26. Dalzell L, Orlando M, MacDonald M, et al. The New York State universal newborn hearing screening demonstration project: ages of hearing loss identification, hearing aid fitting, and enrollment in early intervention. Ear Hear 2000;21: 118-30.
27. Moeller M. Early intervention and language development in children who are deaf and hard of hearing. Pediatrics 2000; 106:e43.
28. Calderon R, Naidu S. Further support for the benefits of early identification and intervention for children with hearing loss. In: Yoshinaga-Itano C, Sedley AL, eds. Language, speech and social-emotional development of children who are deaf or hard of hearing. Washington, D.C.: Alexander Graham Bell Association, 2000:53-84.
29. Wake M, Poulakis Z, Hughes EK, Carey-Sargeant C, Rickards FW. Hearing impairment: a population study of age at diagnosis, severity, and language outcomes at 7-8 years. Arch Dis Child 2005; 90:238-44.
30. Byrne JM, Backman JE, Bawden H, Minnesota Child Development Inventory: a normative study. Can Psychol 1995;36: 115-30.
31. Schroeder L, Petrou S, McCann D, et al. The economic costs of congenital bilateral permanent childhood hearing impairment. Pediatrics 2006;117:1101-12.